Implementation of preventive treatments for hip disorders in cerebral palsy: a preliminary study in Granada

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ABSTRACT

Background: Hip subluxation and dislocation are one of the most frequent musculoskeletal disorders observed in patients with cerebral palsy (CP).This study pretends to demonstrate the implementation percentage of preventive treatments for hip displacement in the population of Granada affected by cerebral palsy, as well as their results. **Methods:** 35 subjects with CP (age range 8-20 years) participated in a retrospective observational study. Preventive treatments for hip disorders were analyzed, as well as the degree of coxofemoral joint decoaptation. Data concerning preventive treatments using botulinum toxin injections (BTA), orthoses and soft tissue release surgery (STRS), was gathered. Coxofemoral morphology was measured through X-rays using Reimer's Migration Index (RMI). **Results:** Total percentage of hip dislocation/subluxation was 31.4%. For subjects belonging to levels IV and V according to Gross Motor Function Classification System (GMFCS), the implementation percentage of the studied preventive treatments was less than 50%. Furthermore, for all these levels, the percentage of subjects that received BTA and STRS and didn't present RMI > 30%, was above 60%. **Conclusions:** In our population belonging to levels IV and V of GMFCS, the implementation percentages of BTA and STRS were low. The limited application of these preventive treatments results in a great number of hip disorders.

Key words: Hip dislocation, cerebral palsy, prevention and control, botulinum toxin, surgical procedures.

RESUMEN

Antecedentes: *La subluxación y dislocación de la cadera son uno de los trastornos musculoesqueléticos más frecuentes observados en los pacientes con parálisis cerebral (PC). Este estudio pretende mostrar el porcentaje de aplicación de tratamientos preventivos para el desplazamiento de cadera en la población de Granada afectada por parálisis cerebral, así como sus resultados.* ***Métodos:*** *35 sujetos con parálisis cerebral (rango de edad 8-20 años) participaron en un estudio observacional retrospectivo. Se analizaron los tratamientos preventivos para los trastornos de la cadera, así como el grado de decoaptación de la articulación coxofemoral. Se recolectaron los datos relativos a los tratamientos preventivos que utilizan inyecciones de toxina botulínica (ITB), ortesis y cirugía de liberación de tejidos blandos (CLTB). La morfología coxofemoral se midió a través de rayos X utilizando el índice de migración de Reimers (IMR).* ***Resultados:*** *El porcentaje total de luxación/subluxación de cadera fue de 31.4%. Para los sujetos que pertenecen a los niveles IV y V de acuerdo con el Sistema de Clasificación de la Función Motora Gruesa (SCFMG), el porcentaje de aplicación de los tratamientos preventivos estudiados fue de menos de 50%. Además, para todos estos niveles, el porcentaje de sujetos que recibieron ITB y CLTB y que no presentaron IMR > 30%, fue superior al 60%.* ***Conclusiones:*** *En nuestra población perteneciente a los niveles IV y V de SCFMG, los porcentajes de ejecución de ITB y CLTB eran bajos. La aplicación limitada de estos tratamientos preventivos resulta en un gran número de trastornos de cadera.*

Palabras clave: *Dislocación de cadera, parálisis cerebral, prevención y control, toxina botulínica, procedimientos quirúrgicos.*

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Abbreviations:
CP = Cerebral palsy.
BTX = Botulinum toxin.
STRS = Soft tissue release surgery.
RMI = Reimer's Migration Index.
CBS = Corrective bone surgery.
MS = Modeling seat.

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INTRODUCTION

Hip subluxation and dislocation are one of the most frequent musculoskeletal disorders observed in patient with cerebral palsy (CP)[1]. Its incidence rate is directly related to the gravity of CP; therefore, higher rates are detected in subjects with severe mobility impairments[2,3].

One of the most important consequences of these disorders is the presence of pain, which has a profound effect in the quality of life of the affected subjects[4].

These orthopedic disorders manifest themselves during infancy[5]; moreover, recent studies indicate that there is a higher risk of onset between 2-7 years[6].

Therefore, it is considered that the presence of coxofemoral subluxation and dislocation, in this population, is a result of biomechanical causes secondary to the neurological disorders associated with CP. Furthermore, several studies relate hip dislocation to asymmetrical activity of periarticular muscles[7], particularly hypertonia of adductor and flexor muscles[8].

Thus, there is a group of muscles that act like hip stabilizers (coaptators) (pyramidal, external obturator, and gluteus minimus and medium), there is another group that because of their vertical orientation, they tend to rather dislocate de hip (adductors)[9].

In this sense, computer models show the negative role of adductor muscles, even during positions used to reduce hip dysplasia[10].

Therefore, many preventive treatments target these muscle groups, trying to balance the different forces that act upon the coxofemoral joint as much as possible.

For this reason, the use of botulinum toxin type A (BTX) is a common measure to improve care and comfort of people suffering of CP[11]; and although it helps to postpone hip surgery, it only does so for a brief period of time, and without improving orthopedic prognosis[12].

Other authors emphasize the importance of orthotics as preventive treatment, particularly the use of modeling seats[13].

Nevertheless, the use of these techniques (orthotics and BTX) in combination with physical therapy, while they are able to postpone articular displacement, they are not effective by themselves to prevent its onset[8]; therefore, these measures are left as general management options.

As a result, most studies focus on complementing and organizing the line of approach to prevent hip orthopedic disorders in CP, beginning with BTX, continuing with soft tissue release surgery (STRS), and ultimately with corrective bone surgery (CBS) when all the above measures have failed[14]. Other authors propose protocols that start directly with surgery, beginning with soft tissue release and ending with corrective bone surgery[15]. In either case, It's important to take into account the age at which the subject will undergo these surgeries to program these procedures[6,16].

Nevertheless, regarding STRS, there is no consensus among the authors about timing and indication criteria to perform these procedures, about which specific muscles have to be considered for treatment or the post-surgical management that come afterwards[17]. Also, paradoxically, subjects with major motor disabilities (levels IV and V according to GMFCS) show the worst results[18].

Accordingly, we found there are great variety of preventive protocols that deal with both subluxations and dislocations; therefore, professionals dedicated to this field of work don't have enough evidence to make right therapeutic decisions among different treatments, nor which is the best time to use them[19].

This study derives from the need to evaluate the present situation of our field about orthopedic hip disorders associated with the CP population, as well as the preventive treatments to avoid them.

Accordingly, we propose the following objectives:

- To show the implementation percentage of different preventive treatments for hip disorders used in subjects with CP of a certain population in the city of Granada.
- To compare the results of the preventive measures used in this population with those obtained from other populations, where there were used standard protocols.

METHODS

Study design and participants

This study followed the protocols established by the centers «Fundación Purísima Concepción» and «San Rafael», both belonging to the city of Granada (Spain). These protocols included informed consent using a Child Caregiver Consent Form and the approval of Hermanas Hospitalarias' Ethics Committee for Clinical Trials. Data was collected during May-June 2014, following the steps of the application protocol by keeping subjects' personal information confidential at all times.

The present study is descriptive, observational and transversal by design. A sample was selected from the total population of students enrolled at «Purísima Concepción» and «San Rafael» Special Education Centers in the city of Granada, Spain.

The inclusion criteria include having an infant cerebral palsy (ICP) diagnosis and age older than seven years. Exclusion criteria consisted of not having a history that included surgical procedures, BTX injections, orthotics, and annual X-ray control studies.

A consecutive sampling took place during the data acquisition period in order to select subjects who fitted the inclusion

criteria. From a potential sample of 40 subjects, four children had incomplete radiographic monitoring, and one of them didn't have a history of BTX injections; therefore, the final sample came down to thirty five subjects with CP, ages 8-20 years old, which attended to the physical therapy unit of their respective centers on regular basis.

Variables

Independent variables were established based on the different preventive treatments for hip disorders, which were conducted[6,11-16]; while the dependent variable was determined by the condition and alignment of the coxofemoral joint.

Physical therapy was considered a constant, since every subject received therapy twice a week, for 30 minutes each. Every session consisted of articular management that included bilateral muscle stretching of both adductors and hip flexors.

Hence, the variables were established as:

- Botulinum toxin type A injection (BTX).
- Soft tissue release surgery (STRS).
- Coxofemoral joint alignment.

To objectify the condition of the coxofemoral joint, a radiological exam was conducted measuring the Reimer's Migration Index (RMI)[20], which is considered the most accurate method to monitor hip stability in the CP population[21]. Based on the findings of Hägglund et al[21], who proposed that 33% of RMI as threshold for a more thorough observation, and Robin et al[22] which categorized grades IV and V with a RMI > 30% as having hip subluxation or dislocation, radiographic findings were divided into two groups:

- Coxofemoral joint with RMI greater or equal to 30% (RMI ≥ 30%).
- Coxofemoral joint with RMI less than 30% (RMI < 30%).

To obtain information about BTX injections for bilateral hip adductors, there were taken into account at least 2 annual injections for Adductor Magnus and Gracilis muscles, previous to presentation of a migration index greater or equal to 30%[23]. For all cases, the declared dosage administered was 50-100 units of Dysport® per injection point, and 25-50 units of Botox® per injection point.

Based on the classification proposed by Elkamil et al[24], surgical procedures were divided into two categories:

- *Soft Tissue Release Surgery (STRS):* involves stretching or myotendinous release, previous to development of a RMI ≥ 30%.
- *Corrective Bone Surgery (CBS):* as soon as hip dislocation is diagnosed (RMI≥ 30%), in addition to soft tissue release, other corrective surgeries are performed, including: varus osteotomy of the proximal femur, pelvic osteotomy and/or femoral head resection. Since all this procedures are not preventive but corrective measures, they were not taken into account as part of the data.

Statistical analysis

Data analysis was conducted using SPSS 20.0 software for Windows operating system. First, in order to make a descriptive analysis it was determined frequencies and percentages. In order to compare the relationship between preventive treatments for coxofemoral disorders and migration indexes, there were used contingency tables and Pearson's Chi Square Test (χ^2).

RESULTS

35 subjects with CP participated in this study, with ages between 8-20 years (M = 12.83 ± 3.58 years), had their hips examined. 11 cases (31.4%) presented coxofemoral disorders; and 7 (20%) out of these cases were bilateral. *Table 1* shows type and topographic distribution of CP, GMFCS level, and RMI ≥ 30%, in relation to gender.

Figure 1 shows a migration index equal or greater than 30% regarding GMFCS levels. Worth mentioning is that 10% (n = 1) of subjects with right hips with RMI > 30% belong to level III, 30% (n = 3) to level IV, and 60% (n = 6) to level V according to GMFCS. Regarding the percentages for each GMFCS level, 75% (n = 3) of subjects with right hips with RMI > 30%, were categorized as level IV, 33.3% were in levels III (n = 1) and V (n = 6), and 0% in levels I (n = 0) and II (n = 0).

In the other hand, 12.5% (n = 1) of subjects with left hips with RMI > 30%, belong to level III, 25% (n = 2) to IV and 62.5% (n = 5) belonged to level V according to GMFCS. Regarding each GMFCS level, 50% of subjects with left hips with RMI ≥ 30% were categorized as level IV, 33.3% were in level III, and 27.8% were in level V, and 0% were in levels I and II.

Frequencies of interventions previous appearance of RMI ≥ 30% according to gross motor function (GMFCS) are shown on *figure 2*. Surgical procedures performed previous appearance of RMI greater or equal to 30% consisted of two percutaneous musculotendinous enlargement through bilateral microtenotomy of adductors and rectus anterior muscles, one bilateral adductor tenotomy, and one left adductor and psoas tenotomy.

The outcomes of treatments performed in subjects with different GMFCS levels are shown on *table 2*. Regarding BTX treatments previous to appearance of RMI greater or equal to 30%, it was proved that for GMFCS levels IV and V, 66.7%

Table 1. Population distribution by gender, types of cerebral palsy (CP) based on motor function and topographic distribution, level of gross motor function classification system (GMFCS), y Migration Index equal or greater than 30% (RMI ≥ 30%).

			Gender		
			Female	Male	Total
Type of cerebral palsy (based on motor function)	Spastic	Count	12	13	25
		% type	48%	52%	100%
	Dyskinesia	Count	2	4	6
		% type	33.3%	66.6%	100%
	Ataxic	Count	1	1	2
		% of type	50%	50%	100%
	Mixed	Count	1	1	2
		% type	50%	50%	100%
Type of cerebral palsy (based on topographic distribution)	Quadriplegia	Count	10	9	19
		% Distribution	52.6%	47.4%	100%
	Diplegia	Count	5	8	13
		% Distribution	38.5%	61.5%	100%
	Hemiplegia	Count	1	2	3
		% Distribution	33.3%	66.6%	100%
Level of gross motor function classification system	I	Count	1	1	2
		% GMFCS	50%	50%	100%
	II	Count	3	5	8
		% GMFCS	37.5%	62.5%	100%
	III	Count	2	1	3
		% GMFCS	66.6%	33.3%	100%
	IV	Count	1	3	4
		% GMFCS	25%	75%	100%
	V	Count	9	9	18
		% GMFCS	50%	50%	100%
Migration index equal or greater than 30% for right hip		Count	6	4	10
		% RMI ≥ 30%	60%	40%	100%
Migration index equal or greater than 30% for left hip		Count	6	2	8
		% RMI ≥ 30%	75%	25%	100%
Migration index equal or greater than 30% for right hip presented before seven years		Count	4	4	8
		% RMI ≥ 30%	50%	50%	100%
Migration index equal or greater than 30% for left hip presented before seven years		Count	5	2	7
		% RMI ≥ 30%	71.4%	28.6%	100%

(n = 6) of right hips and 71.4% (n = 5) of left hips with RMI ≥ 30% had not received BTA as preventive treatment. In this sense, 62.5% (n = 5) of subjects who received BTA as preventive measure did not developed RMI ≥ 30% at their right hips; and 81.1% (n = 9) subjects did not developed RMI ≥ 30% at their left hips. Statistically significant differences were found for GMFCS levels I-III (p = 0.015), where 91.7% (n = 11) of subjects did not received BTX injections nor developed RMI ≥ 30% in both hips.

The association between migration index and the use of modeling seats is presented at *table 3*, where GMFCS levels IV and V are established for right hips with RMI ≥

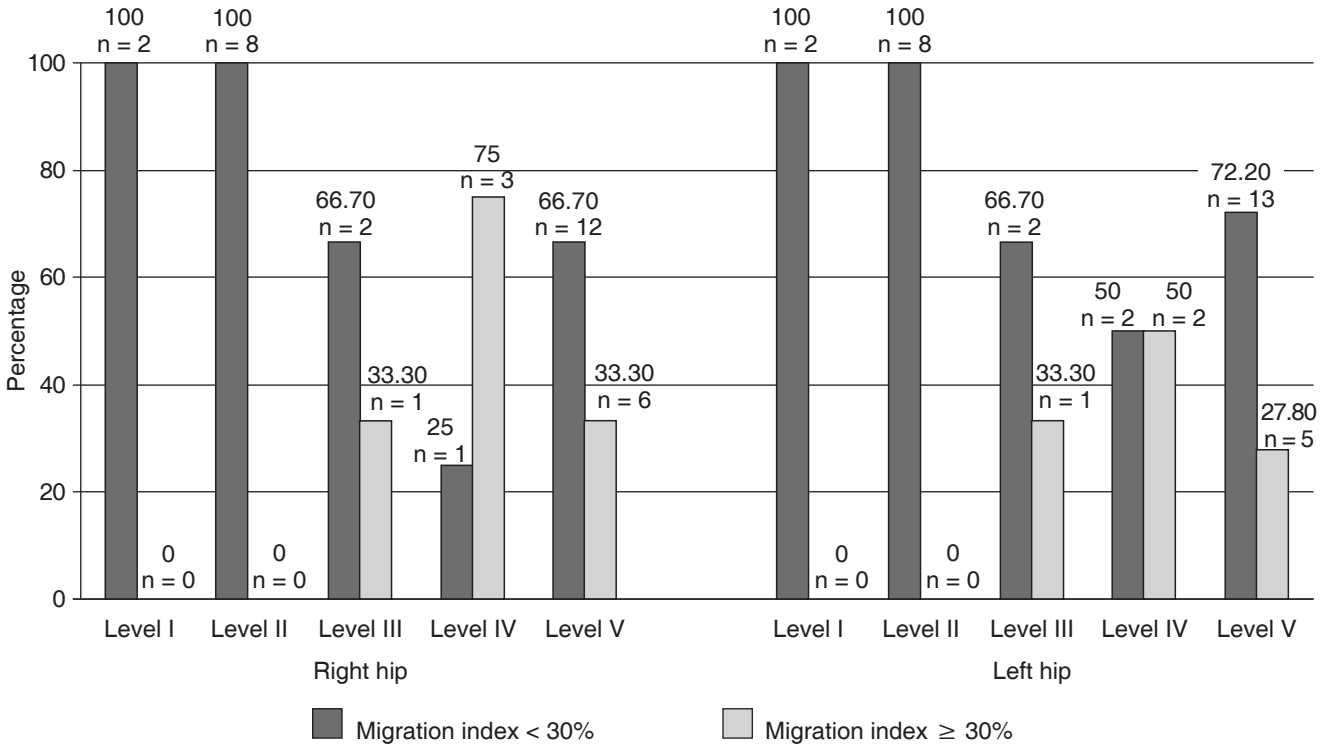
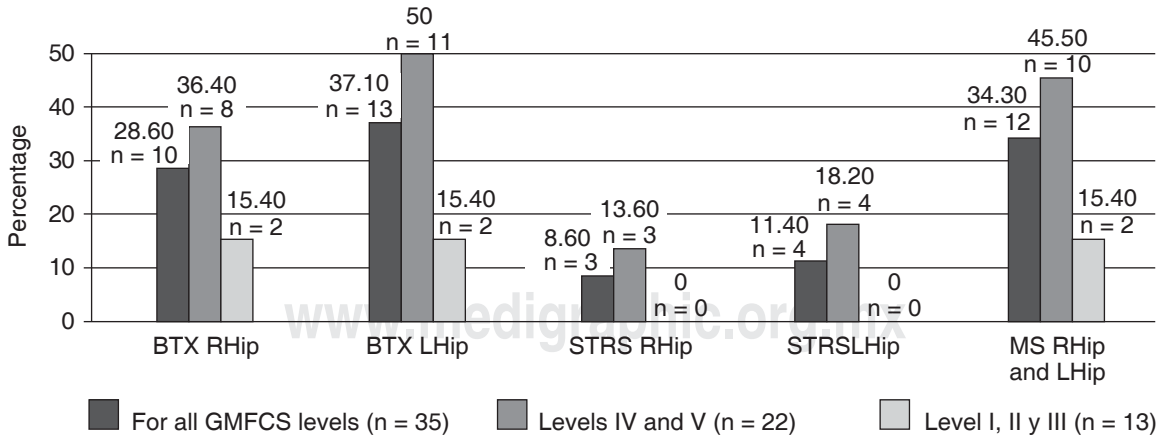


Figure 1. Migration Index according to level of gross motor function classification system (GMFCS).



BTX = Botulimum toxin type A; STRS = Soft tissue release surgery; MS = Modeling seat; RHip = For right hip; LHip = For left hip.

Figure 2. Frequencies and percentages of preventive treatments for hip disorders according to each level of gross motor function classification system (GMFCS).

30%. For this group, the outcomes between cases that used modeling seats as preventive treatment (55.6%, n = 5) and those who did not (44.4%, n = 4) are very similar. The same was observed for left hips with RMI ≥ 30%, where 42.9% (n = 3) of cases used modeling seats and 57.1% (n = 4) did not. For GMFCS levels I-III there were found statistically significant differences (p = 0.015), where 91.7% (n = 11) of subjects did not use orthosis nor developed RMI ≥ 30% in both hips.

A comparative analysis between migration indexes greater or equal to 30% and soft tissue release surgery performed previous appearance of such index, determined that for GMFCS levels IV and V, 100% (n = 3) of right coxofemoral joints that underwent soft tissue release surgery did not developed RMI ≥ 30%; the same ensued for 75% (n = 3) of left hips. For levels I-III, no subjects (n = 0.0%) underwent STRS *(Table 4)*.

DISCUSSION

In the present study, the percentages of hip dislocation and subluxation (31.4%) were found to be consistent with studies conducted in larger populations[2]. It was found that greater percentages of joints with RMI ≥ 30% belong to levels IV and V, concurring with articles that associated GMFCS levels to joint codition[2]. Also, in correspondence to previous studies[6], most disorders occurred before age 7.

However, our findings show a large variety of treatments and protocols used for subjects with cerebral palsy in our population, with a total of 31.4% of hip dislocation/subluxations; whereas studies where a combined protocol had been applied, the percentage of disorders which didn't required bone surgery was 6.32%[25]. These findings are quite a concern for subjects who belong to groups IV and V according to GMFCS, who, based on our outcomes, have percentages of RMI > 30% between 33.3% and 75% for right hips, and between 27.8% and 50% for left hips. Therefore, although data suggests that the appearance of hip disorders in our population (where still there is little consensus about protocols) do not differ much from prevalence studies[2]. Nevertheless, they vary from articles than deal with the effectiveness of supervised assistance programs for the hip, based on standard protocols seen in other countries with larger populations[25].

Table 2. Relationship between a preventive treatment using botulinum toxin type A (BTA) and the presentation of a migration index equal or greater than 30% (RMI ≥ 30%) according levels of gross motor function classification system (GMFCS).

			Preventive treatment for right hip using BTA		Preventive treatment for left hip using BTA	
Levels IV and V of GMFCS (n = 22)			No	Yes	No	Yes
Migration index equal or greater than 30%	No	Count	8	5	6	9
		% RMI ≥ 30%	61.5%	38.5%	40%	60%
		% BTA	57.1%	62.5%	54.5%	81.8%
	Yes	Count	6	3	5	2
		% RMI ≥ 30%	66.7%	33.3%	71.4%	28.6%
		% BTA	42.9%	37.5%	45.5%	18.2%
			p = 0.806		**p = 0.170**	
Levels I, II and III of GMFCS (n = 13)						
Migration index equal or greater than 30%	No	Count	11	1	11	1
		% RMI ≥ 30%	91.7%	8.3%	91.7%	8.3%
		% TBA	100%	50%	100%	50%
	Yes	Count	0	1	0	1
		% RMI ≥ 30%	0%	100%	0%	100%
		% TBA	0%	50%	0%	50%
			p = 0.015*		**p = 0.015***	

Among our population it was observed high levels of articular maintenance in subjects belonging to GMFCS levels IV and V, who received BTX as preventive treatment, especially for the left coxofemoral joint. These findings, although they are consistent with studies that support BTX as beneficial[11,26,27], they differ from those that give little evidence to this course of treatment[28] or bear limitations for orthopedic prognosis[12]. Despite some authors' indication that the use of modeling seats has good results[13], or are valuable for rapid evolutions[29], our findings don't show too many differences between subjects who used these orthoses, in comparison with those who didn't use them as preventive treatment. Still, 70% of subjects who belonged to GMFCS levels IV and V and used modeling seats didn't manifest RMI ≥ 30% for their left hips. These somewhat confusing results could be related with lack of consensus and subjectivity during the construction of modeling seats, since the degree of abduction required for a correct coxofemoral mechanomorphosis is not established nor protocolled among subjects with muscle tone disorders.

In the other hand, regarding STRS, our results are well within limits settled by studies which give these procedures high rates of success[30]. In that regard, there was only one case for left hip and none for the right hip which developed a RMI ≥ 30%, even though the proportion of subjects that underwent these interventions was rather small.

In summary, it is important to emphasize the low percentages of implementation of these preventive treatments in our population (36.4-50% for BTX, 13.6-18.2% for STRS, and 45.5% for modeling seats), especially in groups with a higher risk of developing hip dislocation (levels IV and V).

Study limitations

A limitation of the present study is the small sample size due to its preliminary nature. Although the study aims to check implementation treatments in a particular place, multiple large studies are needed to have a global vision.

Another limitation of the study is the inability to objectively evaluate the effectiveness of the use of orthosis by not following the established protocols during its design; hence for future projections of this study, in order to assess this issue, it is necessary to unify the criteria to subsequently evaluate its effectiveness as preventive treatment for hip disorders.

Table 3. Relationship between a preventive treatments using modeling seat and the presentation of a migration index equal or greater than 30% (RMI ≥ 30%) according to levels of gross motor function classification system (GMFCS).

			Preventive treatment for right hip using modeling seat		Preventive treatment for left hip using modeling seat	
Levels IV and V of GMFCS (n = 22)			No	Yes	No	Yes
Migration Index equal or greater than 30%	No	Count	8	5	8	7
		% RMI ≥ 30%	61.5%	38.5%	53.3%	46.7%
		% for Modeling seat	66.7%	50%	66.7%	70%
	Yes	Count	4	5	4	3
		% RMI ≥ 30%	44.4%	55.6%	57.1%	42.9%
		% for Modeling seat	33.3%	50%	33.3%	30%
			p = 0.429		**p = 0.867**	
Levels I, II and III of GMFCS (n = 13)						
Migration Index equal or greater than 30%	No	Count	11	1	11	1
		% RMI ≥ 30%	91.7%	8.3%	91.7%	8.3%
		% for Modeling seat	100%	50%	100%	50%
	Yes	Count	0	1	0	1
		% RMI ≥ 30%	0%	100%	0%	100%
		% for Modeling seat	0%	50%	0%	50%
			p = 0.015*		**p = 0.015***	

Table 4. Relationship between a preventive treatment using soft tissue release surgery (STRS) and the presentation of a migration index equal or greater than 30% (RMI ≥ 30%) according to levels of gross motor function classification system (GMFCS).

			Preventive treatment for right hip using soft tissue release surgery		Preventive treatment for left hip using soft tissue release surgery	
Levels IV and V of GMFCS (n = 22)			No	Yes	No	Yes
Migration index equal or greater than 30%	No	Count	10	3	12	3
		% RMI ≥ 30%	76.9%	23.1%	80%	20%
		% STRS	52.6%	100%	66.7%	75%
	Yes	Count	9	0	6	1
		% RMI ≥ 30%	100%	0%	85.7%	14.3%
		% STRS	47.4%	0%	33.3%	25%
			p = 0.121		**p = 0.746**	
Levels I, II and III of GMFCS (n = 13)						
Migration index equal or greater than 30%	No	Count	12	0	12	0
		% RMI ≥ 30%	100%	0%	100%	0%
		% STRS	92.3%	0%	92.3%	0%
	Yes	Count	1	0	1	0
		% RMI ≥ 30%	100%	0%	100%	0%
		% STRS	7.7%	0%	7.7%	0%
			No statistical was calculated since no subject in this group underwent soft tissue release surgery			

CONCLUSIONS

The following conclusions were drawn from the present study:

- The implementation percentages of the studied preventive treatments (BTX, orthotics, and STRS) for hip subluxation and dislocation, in this population, were less than 50%.
- The outcomes of these low implementation percentages in this population; especially among groups with the greatest disability (levels IV and V according to GMFCS) were less successful than those obtained from other countries with larger populations, which followed standard protocols.

Ethical approval: This study followed the protocols established by the centers «Fundación Purísima Concepción» and «San Rafael», both belonging to the city of Granada (Spain). These protocols included informed consent using a Child Caregiver Consent Form and the approval of Hermanas Hospitalarias' Ethics Committee for Clinical Trials.

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