

# Hip subluxation in Italian cerebral palsy children and its determinants: a retrospective cohort study

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The study's aim was two-fold: to describe the trend of hip subluxation in the largest sample of Italian nonambulatory cerebral palsy (CP) children ever published; to investigate its determinants. This single-centre retrospective cohort study included patients with spastic or dyskinetic CP, Gross Motor Function Classification System (GMFCS) level IV or V, age 0–18 years, having been referred to our unit before March 2020. The hip subluxation was measured by means of the migration percentage (MP). Other data were gathered such as sex, CP subtype, GMFCS level, presence of drug-resistant epilepsy, age, use of walkers with weight relief or standing devices, previous botulinum injection or hip surgery, oral or intrathecal baclofen and hip pain. Multiple linear stepwise regression was performed and descriptive statistics are provided. Spastic CP had MP maximum increase in early ages, with GMFCS level V values persistently higher than level IV. The dyskinetic subtype showed a slower increase of the MP, with GMFCS level IV presenting similar or higher values, compared to level V. Age, CP severity and spastic subtype are the main determinants. The stepwise multiple regression analysis demonstrated that weight relief walking and standing assistive devices, combined with botulinum contributed to reduce the MP progression. Dyskinetic CP showed overall lower MP values and a more variable behaviour relative to age and GMFCS

level, compared to the spastic subtype. Standing and walking assistive devices, with partial or total weight relief, combined with individually targeted botulinum injections, should be considered in the management of bilateral nonambulatory CP patients, to prevent hip subluxation or its recurrence after surgery. *International Journal of Rehabilitation Research* 45: 319–328 Copyright © 2022 The Author(s). Published by Wolters Kluwer Health, Inc.

*International Journal of Rehabilitation Research* 2022, 45:319–328

**Keywords:** assistive devices, botulinum toxins, cerebral palsy, hip dislocation, migration percentage, orthopaedics, postural management, rehabilitation, standing devices

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Received 4 May 2022 Accepted 21 July 2022

## Introduction

Children affected by cerebral palsy (CP) have an increased risk of progressive hip subluxation. Subluxation is measured by means of the migration percentage (MP), which is considered the gold standard for assessing it [1]. Complete luxation corresponds to a 100% MP; subluxation or displacement is mostly defined as an MP of between 30 and 99% [2]. The incidence of the subluxation is related to CP severity, which is defined according to the Gross Motor Function Classification System (GMFCS). More severe nonambulatory bilateral patients (GMFCS IV and V) are known to be the most vulnerable to hip subluxation [3–4].

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Screening programmes are strongly recommended [5–7] to intercept hip luxation at the initial stages and to implement treatment [8]. Interventions include surgery, postural management and spasticity treatment. A multimodal approach is recommended in CP surveillance programmes, even though evidence for conservative treatments is poor.

Older studies investigated conservative approaches, such as standing or sitting devices [9–12] or botulinum combined with abduction orthoses [13], but the results were inconclusive, given the small sample sizes, low level of evidence and no significant improvement in terms of MP.

Subsequently, the literature focused on the efficacy of surgery, which significantly reduces the MP [14–16]. Bone surgery appeared more effective than soft tissue surgery [14,17], and an early approach was recommended [18]. Nevertheless, recent studies have demonstrated

that surgery itself has a risk of recurrence, which is higher in children with GMFCS level IV-V and in surgery at a younger age [16,19,20]. Considering this, interest is aroused again in the deepening role and type of conservative preventive approaches, to stabilize the MP and prevent relapse after surgery, or to reduce the hip subluxation slope and delay surgery.

Italy does not yet have a national surveillance programme. However, as a tertiary referral centre, we perform regular clinical and radiographic monitoring of our patients and perform surgery or provide spasticity treatments and daily postural management, based on individual needs.

A retrospective cohort study was conducted, involving CP children at GMFCS level IV-V who referred to our centre. The objective of our study was two-fold: to describe the trend of hip subluxation, by means of the MP and the MP progression, in the largest sample of Italian non-ambulatory CP children ever published; to investigate its determinants.

## Methods

### Participants

Inclusion criteria were: bilateral spastic or dyskinetic CP, according to surveillance of CP in Europe (SCPE) classification; GMFCS level IV or V; age 0–18 years; having attended the Children Rehabilitation Unit at the S. Maria Nuova Hospital in Reggio Emilia, before March 2020; having at least one pelvic radiography with MP reported or measurable; informed consent of the parents for minors; informed consent of the adult patient if he/she was able to independently subscribe it or of the support administrator for vulnerable adults. The study was approved by the Area Vasta Emilia Nord Ethics Board on 21 April 2020 (200/2020/OSS\*/AUSLRE). Only spastic or dyskinetic CP were included because they are the most exposed to hip luxation, conversely, the ataxic subtype is rarely affected, as previously described [3,21].

We acquired the following information from the patients' charts.

### Clinical and functional assessment

Sex; CP subtype, spastic or dyskinetic, according to SCPE; GMFCS level; drug-resistant epilepsy; age corresponding to the date of the acquired radiography; botulinum (BoNT-A) injections in hip muscles in previous 6 months; soft tissue or bone surgery, relative to the hip, in previous 12 months; lumbar scoliosis; hip pain; use of assistive devices for standing or walking; oral or intrathecal baclofen. Lumbar scoliosis and hip pain were recorded as dichotomous variables (present or absent), based on what was reported on the clinical chart. In local clinical practice, hip pain was enquired either as directly observed during the clinical assessment or with specific

questions to patients and caregivers, about the symptom and its indirect signs (i.e. sleep disturbances, difficulties in transferring or daily life assistance activities).

### Radiographic evaluation

MP and pelvic tilt measures for right and left hip for each acquired radiography. Both were calculated on anterior-posterior radiography of the pelvis, acquired in a supine position with the legs parallel, avoiding either pelvic rotation or anteversion [22–23]. MP measures of the examined sample were either acquired from the clinical chart or calculated by one data extractor, whenever the radiographic image was available. Either the physiatrists having filled in the clinical charts or the data extractor were experts in MP assessment. The pelvic tilt was measured as the angle between the line tangent to the superior edge of the iliac crest and one horizontal line. It was reported in terms of degrees. The value was considered positive on the higher side and negative on the lower one.

### Statistical analysis

Descriptive statistics are presented for baseline demographic clinical characteristics for the subjects' group.

To enquire possible correlations between the MP and nonfixed features, such as age or interventions (surgery, botulinum, devices, etc), the data were analyzed at the level of the individual hips. Descriptive statistics are provided for the entire hip sample. Data relative to hips having undergone palliative surgery were excluded from the analysis.

For the MP progression analysis, only hips having at least two radiographic measures acquired in consecutive years were selected. The difference between the MP measured on the consecutive radiographies was calculated for each individual hip, to express the MP progression per year, as in Terjesen *et al.*[3] The MP progression might be positive in case of increase of the MP (i.e. hip displacement worsening), negative in case of decrease (i.e. improvement of the hip). Two mixed stepwise multiple regression analyzes were computed, either with MP or with MP progression as the dependent variables, and the acquired clinical data as independent variables. Finally, the best multiple linear regression models were selected, and factors having an independent significant role, in reducing or increasing the MP, and the MP progression were identified.

Statistical analyses were performed using STATA software version 14 (StataCorp. 2015. Stata Statistical Software: Release 14. College Station, Texas: StataCorp LP.).

## Results

As represented in Fig. 1, a total of 1208 quadriplegic CP patients attended our Unit between January 2008 and

February 2020, according to our records. However, we were able to examine charts of only 530 of these patients, because those ones archived before 2015 were not available for consultation. A total of 504 patients gave their informed consent to participate in the present study. Demographic and clinical data of the subjects at the first visit are represented in Table 1.

A total of 375 patients had more than one radiography acquired, with a mean of 3.37 (SD, 2.49) radiographies each. The mean length of follow-up was 6 years (0.1–17 years; SD, 4). Table 2 represents the interventions and clinical data recorded during the entire follow-up. The participants used several types of assistive devices, according to individualized needs. Walking devices ensured total or partial weight support. The standing devices were vertical, prone or supine, with or without weight relief, depending on the severity of the CP. They were both used in usual life settings, involving children in playful and social activities, according to compliance and contextual barriers. Among the overall sample, 96 subjects (19%) incurred in hip dislocation (MP 91–100), of whom 80 (15.9%) were GMFCS V and 8.9% experienced hip pain at least once (6.3% were GMFCS V). Twenty-one patients, with at least one luxated hip, complained of hip pain and it was recurrent (just 3 were GMFCS IV). They corresponded to 4.2% of the 504

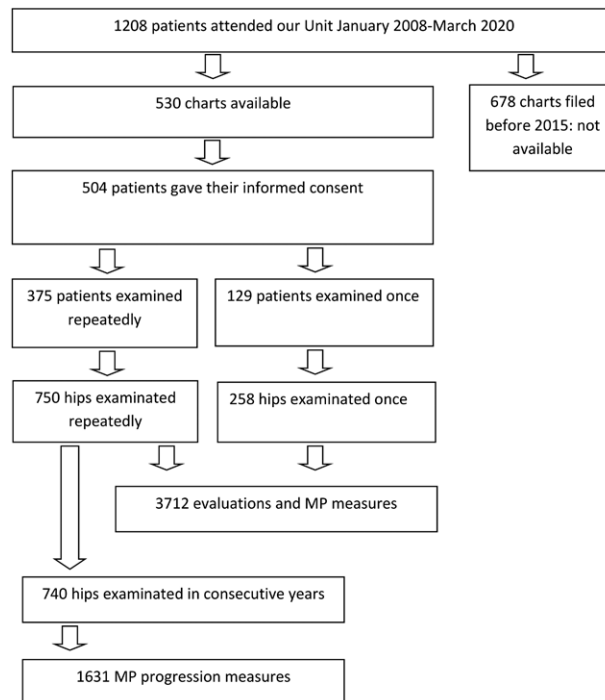
recruited children and just 39% of the subgroup with a dislocated hip.

As in previous studies [9,13], to assess correlations between possible determinants and the MP, the data were analysed at the level of the individual hips. Then 1008 hips were studied. Provided that 750 hips were examined almost twice, a total of 3712 MP measurements were included for the MP analysis (see Table 3). Among them, 740 hips had at least two radiographic measures acquired in consecutive years, then a total of 1631 MP measurements were acquired for the MP progression analysis.

### Migration percentage

The mean values of MP per year of age are reported in Fig. 2, separate charts are provided for hips in spastic and dyskinetic CP, which did not undergo any surgery (Fig. 2a,b), after soft tissue surgery (Fig. 2c,d) and after bone surgery (Fig. 2e,f). The overall trend in dyskinetic CP was less severe, compared to spastic CP, with complete luxation observed in only four subjects and unilaterally. At a glance, soft tissue surgery did not appear to change the MP trend neither in spastic (Fig. 2c) nor in dyskinetic (Fig. 2d) patients, even though the latter ones were a few. Conversely, bone surgery reduced the MP values in both groups (Fig. 2e,f). After femoral

**Fig. 1**



Flow chart representing the recruitment process.

**Table 1 Characteristics of the subjects at the first visit, considering the total sample and stratified by GMFCS level and CP subtype**

Characteristics	Total	GMFCS IV		GMFCS V	
		Dyskinetic	Spastic	Dyskinetic	Spastic
	504	18	184	54	248
Age <sup>a</sup>	5.2±3.6(0–17)	4.2±3.2(0–11)	5.1±3.6(0–17)	5.1±4.0(1–17)	5.0±3.6 (0–15)
Male <sup>b</sup>	295 (58.5)	12 (66.7)	109 (59.2)	30 (55.6)	144 (58.1)
Female <sup>b</sup>	209 (41.5)	6 (33.3)	75 (40.8)	24 (44.4)	104 (41.9)
Drug-resistant epilepsy <sup>b</sup>	40 (8.0)	0	1 (0.5)	3 (5.5)	36 (14.5)
MP (%) <sup>a,c</sup>	23.2±26.0 (0–100)	12.0±16.3 (0–66)	19.2±20.0 (0–100)	13.8±19.9 (0–100)	29.0±29.4 (0–100)
Walking device <sup>b</sup>	162 (32.1)	12 (66.7)	98 (53.3)	21 (38.9)	31 (12.5)
Standing device <sup>b</sup>	216 (42.8)	6 (33.3)	98 (53.3)	26 (48.1)	86 (34.7)
Oral Baclofen <sup>b</sup>	81 (16.1)	1 (5.5)	16 (8.7)	6 (11.1)	58 (23.4)
ITB <sup>b</sup>	5 (1.0)	0	0	1 (1.8)	4 (1.6)
Hip pain <sup>b</sup>	10 (2.0)	0	3 (1.6)	1 (1.8)	6 (2.4)
Lumbar scoliosis <sup>b</sup>	36 (7.1)	0	4 (2.2)	3 (5.5)	29 (11.7)

GMFCS, Gross Motor Function Classification System; ITB, intrathecal baclofen; MP, migration percentage.

<sup>a</sup>Mean ± SD (range).

<sup>b</sup>n (%).

<sup>c</sup>Mean MP including both right and left hips.

**Table 2 Interventions and clinical aspects recorded at least once during the entire follow-up, considering the total sample and stratified by GMFCS level and CP subtype**

Characteristics	Total	GMFCS IV		GMFCS V	
		Dyskinetic	Spastic	Dyskinetic	Spastic
Subjects	504	18	184	54	248
BTX-A injections <sup>a,b</sup>	100 (19.8)	4 (22.2)	44 (23.9)	10 (18.5)	42 (16.9)
Soft tissue surgery <sup>a,c</sup>	145 (29.0)	1 (0.5)	57 (31.0)	7 (13.0)	80 (32.2)
Bone surgery <sup>a</sup>	22 (4.7)	1 (0.5)	9 (4.9)	2 (3.7)	10 (4.0)
Femoral or pelvi-femoral osteotomy <sup>a,d</sup>	16 (3.2)	1 (0.5)	8 (4.3)	1 (0.5)	6 (2.4)
TMH-PF <sup>a,d</sup>	5 (1.0)	0	1 (0.5)	1 (0.5)	3 (1.2)
Oral baclofen <sup>a</sup>	174 (34.5)	3 (1.7)	42 (22.8)	15 (27.8)	114 (46.0)
ITB <sup>a</sup>	37 (7.3)	1 (0.5)	5 (2.7)	6 (11.1)	25 (10.1)
Hip pain <sup>a</sup>	45 (8.9)	1 (0.5)	12 (6.5)	5 (9.2)	27 (10.9)
Lumbar scoliosis <sup>a</sup>	36 (7.1)	0	4 (2.2)	3 (5.5)	29 (11.7)
Pelvic tilt <sup>a</sup>	56 (11.1)	1 (0.5)	17 (9.2)	6 (11.1)	32 (12.9)

GMFCS, Gross Motor Function Classification System; ITB, intrathecal baclofen; TMH-PF, temporary medial hemiepiphyodesis of proximal femur.

<sup>a</sup>n (%).

<sup>b</sup>Single or repeated injections involving muscles around the hip, such as adductors, gracilis, hamstrings, psoas or rectus femoris.

<sup>c</sup>Surgical release of one or more of the following muscles, according to individual features: adductors, gracilis, hamstrings, psoas, rectus femoris.

<sup>d</sup>Uni or bilateral procedure.

or combined osteotomy, a significant reduction of the MP was observed in all cases, and it was maintained at the follow-up (1–7 years). Regarding temporary medial hemiepiphyodesis of the proximal femur (TMH-PF), progression of subluxation was observed in just one hip of a spastic CP child at GMFCS IV. In the other cases, the MP remained stable or decreased at a 3-year follow-up.

Table 4 shows the estimated relationships between the MP, as the dependent variable, and different independent variables, stratified by GMFCS levels IV and V.

### Migration percentage progression

The MP progression per year is represented in Fig. 3 comparing GMFCS IV and V subgroups, for spastic (Fig. 3a) and dyskinetic (Fig. 3b) CP. To explore the spontaneous trend only hips that did not undergo any surgery were included in the charts. According to our results in spastic CP (Fig. 3a), the mean MP progression

per year was high during the first years of life in both groups, reaching its maximum value at age 2 years (11.6% in GMFCS V and 12% in GMFCS IV). MP progression then decreased with age, until around puberty, when it increased again (after age 12 years in GMFCS V and after age 16 years in GMFCS IV). Negative values of the mean MP progression per year were found at 10–11 and 17–18 years in GMFCS V, and at 11–15 years in GMFCS IV, indicative of lower values of hip displacement. Conversely, dyskinetic CP (Fig. 3b) presented a more variable behaviour, with large SD: GMFCS V reached the maximum values of the MP progression at age 3–4 years, whereas GMFCS IV at age 7 years. These values never exceeded 10%, demonstrating an overall MP progression lower than in the spastic subtype. Finally, negative values were noticed also among dyskinetic GMFCS IV at age 13–15 years.

Table 4 shows the best multiple linear regression model for MP progression, as the dependent variable.

**Table 3 Characteristics of the total hip sample and stratified by GMFCS level**

Characteristics	Total	GMFCS IV	GMFCS V	P value <sup>d</sup>
	3712	1482	2230	
Age <sup>a,b</sup>	7.7±4.1 (0–18)	7.79±4.1 (0–18)	7.6±4.1 (0–18)	0.042
Male <sup>c</sup>	2132 (57.4)	882 (59.5)	1250 (56.0)	0.037
Female <sup>c</sup>	1580 (42.6)	600 (49.5)	980 (44.0)	
Dyskinetic CP <sup>c</sup>	458 (12.3)	122 (8.2)	336 (15.1)	<0.001
Spastic CP <sup>c</sup>	3254 (87.6)	1360 (91.8)	1894 (84.9)	
Drug-resistant epilepsy <sup>c</sup>	298 (8.0)	10 (0.7)	288 (12.9)	<0.001
MP (%) <sup>a</sup>	31.2±28.6 (0–100)	22.8±20.8 (0–100)	36.7±31.6 (0–100)	<0.001
MP progression (%) <sup>a</sup>	2.6±12.6 (–100 to 100)	1.4±10.6 (–100 to 71)	3.3±13.7 (–100 to 100)	<0.001
BoNT-A injection in the previous 6 months <sup>c</sup>	274 (7.4)	132 (8.9)	142 (6.4)	0.004
Soft tissue surgery in the previous 12 months <sup>c</sup>	343 (9.2)	143 (9.65)	200 (8.9)	0.483
Bone surgery in the previous 12 months <sup>c</sup>	41 (1.1)	20 (1.3)	21 (0.9)	0.244
Walking device <sup>c</sup>	1374 (37.0)	992 (66.9)	382 (17.1)	<0.001
Standing device <sup>c</sup>	1788 (48.2)	882 (59.5)	906 (40.6)	<0.001
Oral Baclofen <sup>c</sup>	938 (25.3)	222 (14.9)	719 (32.1)	<0.001
ITB <sup>c</sup>	218 (5.8)	40 (2.7)	178 (7.9)	<0.001
Hip pain <sup>c</sup>	130 (3.5)	34 (2.3)	96 (4.3)	0.001
Lumbar scoliosis <sup>c</sup>	482 (12.9)	108 (7.3)	374 (16.7)	<0.001

CP, cerebral palsy; GMFCS, Gross Motor Function Classification System; ITB, intrathecal baclofen.

<sup>a</sup>Mean ± SD (range).

<sup>b</sup>Age at the time of the acquired radiography, corresponding to the MP measurement.

<sup>c</sup>n (%).

<sup>d</sup>To compare groups, unpaired Student's *t*-test was used for continuous variable and Pearson's chi-square test for categorical variables.

## Discussion

To our knowledge, this is the largest sample of Italian quadriplegic CP patients with hip subluxation that has been described.

The results of the present study are consistent with previous studies, with hip displacement worsening particularly in the first years of life and then again around puberty [3,7]. Nonetheless, slight differences emerged between spastic and dyskinetic subtypes, being the overall MP values lower in the latter ones. Moreover, a predominance of higher MP values was observed in GMFCS V spastic CP. Whereas in the dyskinetic subtype, GMFCS IV and V showed a closer trend, overcoming each other depending on the age. In general, the MP tended to increase with age, whereas the MP progression mostly decreased with age, because the 'worsening rate' reduced. Still, a risk of deterioration persisted in the latter years, with MP progression per year increasing again in puberty. These findings confirm that surveillance in both subgroups of patients must start early and be especially vigilant in the first years of life, and it must be continued even after skeletal maturity, as Wynter *et al.* [5] suggest.

While GMFCS level was confirmed to be the strongest determinant of hip luxation [4,24–26], based on our regression analysis, other factors appeared to influence the MP. The first was age, which related negatively to the MP progression, confirming the considerable increase of the MP at younger ages. Second, drug-resistant epilepsy correlated significantly with higher MP values as recently evidenced [26]. Nonetheless, this may be explained by the strong association between this feature and more severe GMFCS level, considering that just one GMFCS IV patient presented drug-resistant epilepsy. Okuno *et al.* [27] specifically enquired about determinants of hip

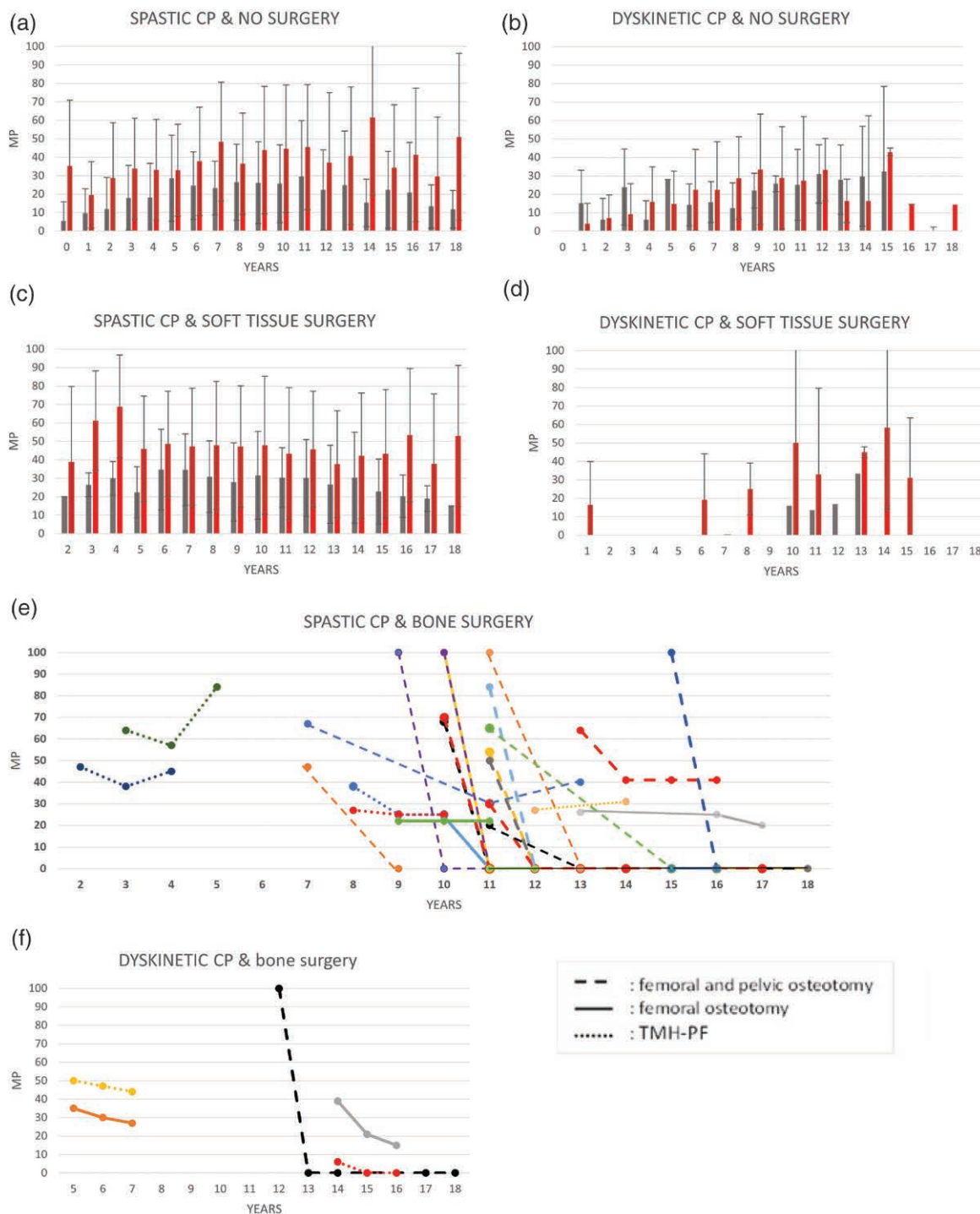
displacement in dyskinetic CP. Beyond the GMFCS level, the primary brain lesion location resulted in an important predicting factor. In particular, globus pallidus lesions were significantly associated with hip displacement (MP >30%). MRI characteristics were not included in the present study analysis, for comparison.

The spastic subtype proved to be another determinant of higher MP values, as previously observed [3]. This indicates that spasticity is an aetiological factor, consistent with the findings by Cho *et al.* [28], who demonstrated a strong association between MP and adductor muscle spasticity. However, according to our results, spasticity treatment alone (BoNT-A, oral or intrathecal baclofen) did not have a significant role in reducing neither the MP nor the MP progression. Recent reviews [11,17] conclude that the evidence is insufficient to support or refute the use of botulinum to prevent hip luxation. Nonetheless, we found a contribution of botulinum combined with standing devices to reducing the MP progression in the GMFCS V group. This may be interpreted as a short-term favourable effect of the combined treatment, and it is consistent with the findings relative to BoNT-A by Love *et al.* [29] and Hagglund *et al.* [21].

Higher rates of hip luxation were encountered in the present sample compared to other studies, mostly based on CP registries [18,20,21,30]. This may be due to the lack of an Italian hip surveillance programme, with patients being addressed to our unit as a tertiary referral centre with advanced displacement. It may also be related to a more cautious approach to nonambulatory severe patients, with reduced rates of reconstructive surgery, compared to previous studies [18,20,21,30].

Nonetheless, the prevalence of hip pain in our sample was lower than what was previously published [31–33],

Fig. 2



MP per year in the examined sample. GMFCS IV and V mean MP per year of age in (a) spastic and (b) dyskinetic CP, that did not undergo any surgery; GMFCS IV and V mean MP per year of age in (c) spastic and (d) dyskinetic CP after soft tissue surgery; individual MP at the year of age, at which the radiography was acquired, just before and after bone surgery in spastic (e) and dyskinetic (f) CP: each hip is represented by individually coloured dots and line, and the three types of line identify the three surgical approaches. CP, cerebral palsy; GMFCS, Gross Motor Function Classification System; MP, migration percentage.

**Table 4** Parameter estimates of the best multiple linear regression models for the MP and for the MP progression, on the overall sample and stratified by GMFCS level

Dependent variable	Independent variables	Coefficient	Std. Err.	P value	95% confidence interval
MP	Total	11.6	1.0	<0.001	9.5–13.7
	Age	0.7	0.1	<0.001	0.5–0.9
	Walkingdevice	−3.0	1.1	0.007	−5.2 to −0.8
	Standingdevice	−5.4	0.9	<0.001	−7.3 to −3.6
	Softtissuesurgery	3.8	1.5	0.034	0.2–6.3
	Bonesurgery	−20.1	4.2	<0.001	−28.5 to −11.7
	SpasticCP	12.8	1.3	<0.001	10.1–15.5
	Drug-resistantepilepsy	5.1	1.6	0.002	1.8–8.4
	GMFCSlevelIV				
	Age	0.3	0.1	0.022	0.04–0.5
	Standingdevice	−2.3	1.0	0.030	−4.4 to −0.2
	Softtissuesurgery	3.3	1.8	0.069	−0.2 to 6.8
	Bonesurgery	−11.9	4.6	0.010	−21.1 to −2.8
	SpasticCP	7.5	1.9	<0.001	3.7–11.3
	GMFCSlevelV				
	Age	0.9	0.1	<0.001	0.6–1.2
	Walkingdevice	−4.9	1.9	0.009	−8.7 to −1.2
	Standingdevice	−6.5	1.4	<0.001	−9.3 to −3.8
Softtissuesurgery	3.1	2.2	0.158	−1.2 to 7.6	
Bonesurgery	−28.1	6.6	<0.001	−41.2 to −15.0	
SpasticCP	13.9	1.8	<0.001	10.2–17.6	
Drug-resistantepilepsy	5.7	1.9	0.003	1.9–9.5	
MP progression	Total				
	Age	−0.1	0.04	0.043	−0.2 to −0.03
	Softtissuesurgery	−3.9	0.6	<0.001	−5.2 to −2.5
	Bonesurgery	−31.3	1.9	<0.001	−34.9 to −27.5
	GMFCSlevelIV				
	Age	−0.14	0.06	0.017	−0.2 to −0.03
	Softtissuesurgery	−3.8	0.87	<0.001	−5.65 to −2.12
	Bonesurgery	−31.4	2.26	<0.001	−35.8 to −26.9
	GMFCSlevelV				
	Age	−0.04	0.06	0.474	−0.18–0.08
	Standing###BoNT-A	−4.22	2.31	0.070	−8.79 to 0.33
	Softtissuesurgery	−3.97	0.99	<0.001	−5.9 to −2.01
Bonesurgery	−31.0	2.93	<0.001	−36.0 to −25.2	

BoNT-A, botulinum; CP, cerebral palsy; GMFCS, Gross Motor Function Classification System.

still similar to that reported by Poirot *et al.* [34] in a longitudinal national CP cohort study in France. In our experience, only a part of chronic dislocations progresses to become painful. This topic is being processed in a separate article. The prevalence of scoliosis in our sample is consistent with data reported by Hagglund *et al.* [35], based on a prospective register study in Sweden: at 10 years of age about 10% of the subject at GMFCS IV, and 30% at GMFCS V showed scoliosis; the incidence increased to 45% in GMFCS IV and 80% in GMFCS V, at 20 years of age. Differently from Bertonecelli *et al.* [26], lumbar scoliosis did not result in an independent determinant of MP increase. This discrepancy may be due to the examined population: the present sample included only the most severe nonambulatory CP subjects, whereas the previous study [26] involved any level of GMFCS. One possible interpretation is that trunk asymmetry may have a more relevant role in contributing to pelvic tilt, than to hip displacement, in walking patients. Conversely, other factors might overcome scoliosis as a determinant in non-ambulatory subjects.

No significant correlation was found between pelvic tilt and MP in our sample. This aspect is controversial in the literature, still, a moderate correlation was recently reported by Hagglund *et al.* [1] The pelvic tilt may be

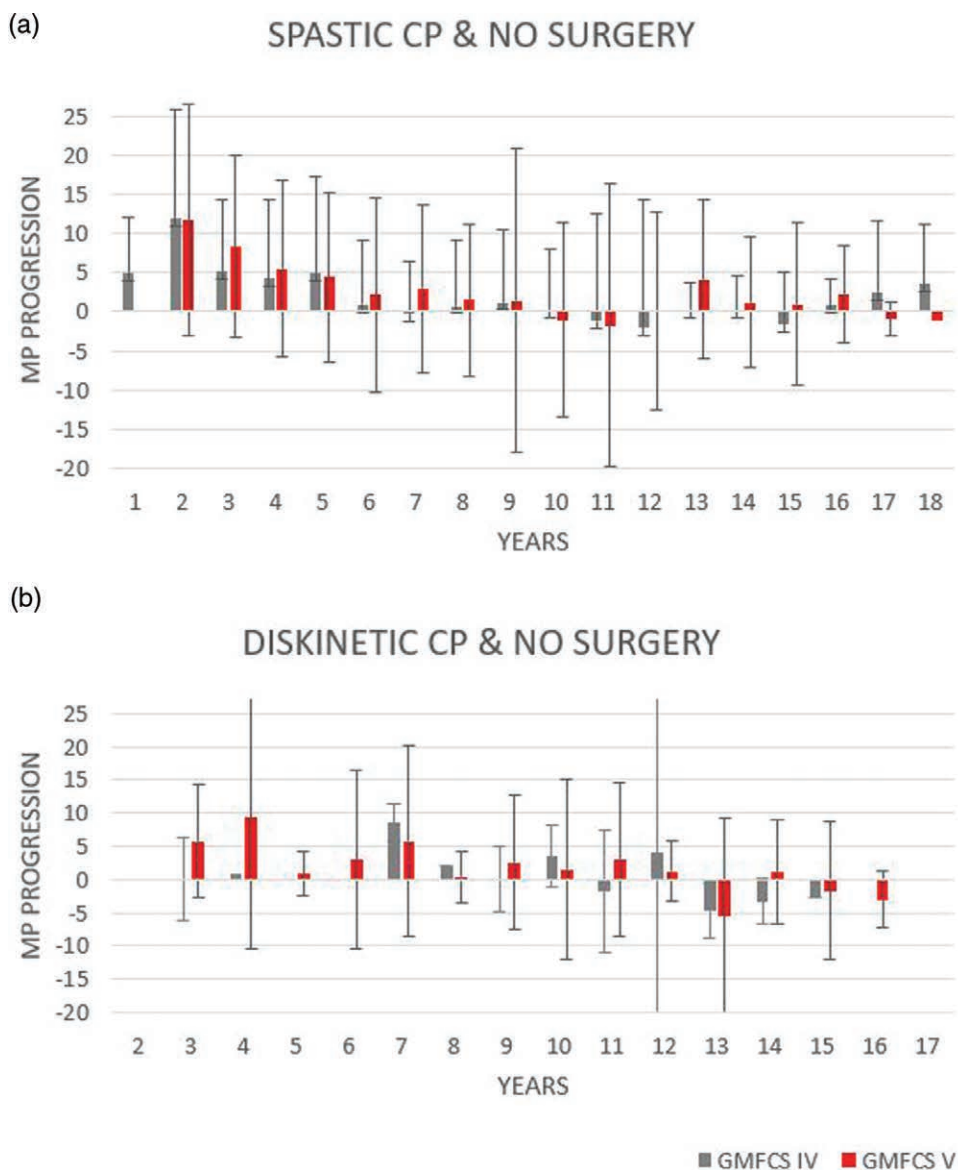
irreversible (i.e. in the case of lumbar scoliosis) or reducible, and it depends on the position of the child on the examination table when the radiography is acquired. An explicit recommendation to reduce it was included in the instructions given for the radiographic examination. A specific study should be conducted to enquire about this topic, considering factors that may influence it. In general hip pain, scoliosis and pelvic tilt recurred in spastic subjects more than in dyskinetic ones.

Femoral and combined osteotomies were confirmed to significantly reduce MP values in both groups, as demonstrated by many studies [16,17,20,28]. Nonetheless, not all the luxated hips, in our population, were addressed to reconstructive surgery. This was a choice of either the caregivers or the medical team, mostly based on the clinical severity of the child.

Encouraging results emerged regarding TMH-PF. The data are too exiguous to draw any conclusion, but they are in line with the literature. In fact, three retrospective studies [36–38] reported evidence in favour of the guided growth of the proximal femur, based on a 3–5-year follow-up.

Soft tissue surgery appeared to induce an increase of the MP, but this may be attributed to the fact that it was

Fig. 3



MP progression per year in GMFCS IV and V, among spastic (a) and dyskinetic (b) CP. CP, cerebral palsy; GMFCS, Gross Motor Function Classification System; MP, migration percentage.

administered not only to prevent hip luxation but sometimes to improve hip range of movement, in favour of perineal hygiene and positioning in severe patients with advanced displacement, for whom reconstructive surgery was ruled out. No reduction of the MP was expected in these cases, and the statistical impact of soft tissue surgery was therefore weaker. Nonetheless, soft tissue surgery showed a significant protective influence on MP progression. This may be interpreted as a positive short-term effect, which was lost in the MP trend over time. Recent studies have in fact reported a long-term rate of reoperation or re-subluxation (i.e. MP over 50%) of soft tissue surgery ranging between 40 and 77% [15-17].

The most interesting result is the statistically significant correlation between using standing and walking devices, without weight-bearing, and MP decreasing. As described above, the walking devices ensured total or partial weight support, to free up propulsive strategies otherwise impeded in quadriplegic patients. Children were positioned over the walking devices by the caregivers, and they used them, for exercise, playing and socializing context. It may be inferred a positive role of active motion over the hips, even without weight-bearing. Also, the standing devices might ensure partial weight relief, by keeping a slightly prone or supine position, and, whenever needed, by providing pelvic support. In this case,



the extended position of the hips might have played a protective role. Regular daily use was recommended, particularly at least 1 h per day for standing devices [10]. Nevertheless, a limitation of our analysis was missing precise information about how much time was dedicated to these activities. The NICE guidelines [7] generally recommend postural management strategies to prevent secondary deformities, without specifying type and time of use, and their role in preventing hip luxation is not yet clear. Besides, the evidence focuses on weight-bearing methods. A systematic review by Gmelig Meyling *et al.* [12] concluded that the evidence is too limited to formulate recommendations for standing programmes. Only two of the selected studies reported statistically significant positive results [39,40] but Macias-Merlo *et al.* [39] treated only GMFCS III patients, whereas Martinsson *et al.* [40] included just 10 GMFCS IV-V patients. Dalen *et al.* [41] reported a negative effect on hip migration, which was attributed to spasticity and low compliance to the standing shell. More recently, Martinson *et al.* [42] presented evidence in favour of abducted standing, based on a retrospective study with nonambulatory subjects, from the CPUP register. Authors demonstrated that practicing standing with 15°–30° of abduction for each leg, for 10 h/week, enhanced the long-term results after soft tissue surgery and reduced the MP, compared to standing without abduction (0°–10°). These findings are extremely interesting. Nonetheless, as Meyling evidenced [43] possible adverse effects and tolerance of such an abducted position should be enquired and addressed. In our sample, targeted botulinum injections were used to reduce pathological patterns and improve compliance with the device. Moreover, weight relief itself probably helped reduce spasticity.

Our findings increase evidence in favour of individually targeted conservative approaches, such as standing and walking devices with weight relief, combined with botulinum injections. Nonetheless, based on the previous study [44], it has to be pointed out that any conservative approach is unlikely to be effective for MP values  $\geq 50\%$ , beyond which increasing hip displacement will probably incur unless addressing surgery.

### Study limitations

The results of this study should be interpreted with caution as they derive from a retrospective design. Prospective studies, preferably RCT, must be conducted to confirm the positive role in preventing hip luxation, of standing and walking devices.

The sample was not recruited from a surveillance programme of the general CP population in Italy but from a single site. Being a tertiary referral centre, patients referred to this Unit may have presented a more severe or complicated clinical situation, than that of the general quadriplegic CP population.

Relative to BoNT-A treatment, no specification about the injected hip muscles was enquired.

Due to the heterogeneity of the sample at subsequent visits, it was not possible to conduct an adequate statistical analysis, to compare the MP progression values of dyskinetic and spastic subtypes.

### Conclusion

The results of the present study confirm age, spastic subtype and CP severity as the strongest determinants of hip displacement. Dyskinetic CP showed overall lower MP values and a more variable behaviour, relative to age and GMFCS level, compared to the spastic subtype. Hip pain, scoliosis and pelvic tilt were more rarely reported in dyskinetic than in spastic CP.

Moreover, the results suggest a protective role of standing and walking devices with weight relief, combined with botulinum injections, for bilateral nonambulatory CP children and MP  $< 50\%$ .

Implementing an Italian CP hip surveillance program is advisable, to early intercept and manage hip displacement.

### Acknowledgements

The dataset generated and analyzed is available upon reasonable request from the corresponding author.

### Conflicts of interest

There are no conflicts of interest.

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