

Associations among the Degree of Hip Adductor Spasticity, the Level of Gross Motor Function Classification System (GMFCS) and the Migration Percentage in Children with Cerebral Palsy

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ABSTRACT

Objectives : This study was aimed at detecting the occurrence of hip dislocation in children with cerebral palsy (CP) by seeking the association among the degree of hip adductor spasticity, level of Gross Motor Function Classification System (GMFCS) and Migration Percentage (MP).

Methods : It was a cross sectional study with inclusion criterias were children with cerebral palsy, both male and female aged 2-10 years old, and parent's approval by signing informed consent. Exclusion criterias were comorbidities of other neuromotor impairments, such as spina bifida; other neuromuscular or musculoskeletal diseases, etc; uncooperative; and presenting severe diseases at the day of assessment. Measurement methods included the Modified Tardieu Scale (MTS) R2, R1 and R2-R1 components to measure hip adductor spasticity; Gross Motor Function Classification System (GMFCS) protocol to assess gross motor ability; and Anterior-Posterior (AP) pelvic plain radiograph to calculate the Migration Percentage (MP) value.

Results : At the initial phase, 31 children were included, however only 57 legs were analyzed for hip adductor spasticity and MP. There were no correlation between the degree of hip adductor spasticity and MP both the R2 with MP ($r = -0.060$; $p = 0.658$), and R1 with MP ($r = -0.136$; $p = 0.314$). Moreover, there was insignificant difference between level of GMFCS and MP ($p = 0.831$).

Conclusion : This study indicates nill correlation between the degree of hip adductor spasticity and the MP, and insignificant difference between the level of GMFCS and MP to detect the occurence of hip dislocation in children with cerebral palsy.

Keywords : Modified Tardieu Scale, R2, R1, level of GMFCS, AP pelvic plain radiograph, MP value.

INTRODUCTION

Cerebral palsy (CP) is the most common cause of physical disability in the developing countries. A non-ambulant child with CP is prone to further joint contractures and postural deformity that are often progressive. Early detection of postural deformities particularly hip subluxation or dislocation is necessary.

CP is defined as a group of movement and posture developmental disorders, causing activities limitation. It is due to nonprogressive disturbances during the period of fetal or infant brain development in the first 2 or 3 years of life.^{1,2} It is the most common cause of physical disability in the developing country.³ The prevalence of CP in Indonesia is estimated around 1-5 per 1000 live births.⁴

Brain injury can occur during prenatal, perinatal and postnatal periods and leads to disorganization and developmental delay of neurological mechanism of postural control or balance and movement.⁵⁻⁷ Based on muscle tone abnormalities, CP can be classified as spastic (75%), dyskinetic, hypotonic and mix. Body parts involvement in CP is classified as diplegia (80%), quadriplegia, triplegia and hemiplegia.^{1,6} Another classification is determined by the Gross Motor Function Classification System (GMFCS) in which the gross motor function disorder is assessed.^{1,8,9}

A non-ambulant CP child is vulnerable to joint contractures and postural deformity that are often progressive and subsequently will impair the quality of life.^{10,11}

Hip displacement in children with CP is usually attributed to spasticity and contracture of the hip adductors and flexors as well as the medial

hamstrings.^{7,12-14} According to the Consensus Statement on Hip Surveillance for Children with Cerebral Palsy: Australian Standards of Care (2008), spasticity can be measured using the Modified Tardieu Scale (MTS).¹⁵ Spasticity is a motor disorder characterized by velocity-dependent increase in muscle tone will exaggerated tendon jerks resulting from hyperexcitability of stretch reflex.¹⁶ The increase in muscle tone causes loss of trunk balance dan restricted active movement in the extremities. It causes abnormal gait and posture.^{17,18}

Hip displacement is thought to be common in children with CP and may progress from silent subluxation to painful dislocation when left untreated.^{12-14,19,20} Around 15-20% of children with CP has risk of hip dislocation²⁰ and 60% affect those who have not been able to walk at the age of five.^{19,21} CP children at the age of 30 months are suggested to make an AP pelvic plain radiograph on standard position.²²

There are two aspects of abnormality in spastic hip, which are extremely high force of hip joint reaction and incorrect force vector that create an abnormal force on the developing acetabulum. The posterosuperior dislocation pattern is caused when the hip is positioned in adduction and flexion. The direct causes of this hip positioning are abnormal pattern of muscle length and contraction force. The primary muscle that causes adduction and flexion is the adductor longus; however, the gracilis and the medial hamstrings, and then the adductor brevis are affected subsequently.²³

The most important qualification of a normal hip joint is the adequate coverage of the femoral head, which can be assessed by the migration percentage.^{13,24} The risk of hip displacement varies between 0% in the sub-type of pure ataxia

to 79% in spastic tetraplegia, and 0% in the GMFCS level I to 64% in GMFCS level V.^{20,25}

The aim of this study was to seek any correlations among the degree of hip adductor spasticity, level of GMFCS and Migration Percentage (MP) to detect the occurrence of hip dislocation in children with CP. They were hypothesized to be correlated.

METHODS

The study design was a cross-sectional study. The measurement of the independent variable (degree of spasticity and GMFCS) and the dependent variable (MP) were performed at the same time. This study was approved by ethics of the Research Ethics Committee of the Faculty of Medicine, University of Indonesia; and granted permission by the Cipto Mangunkusumo National Hospital (RSCM). Sampling was done by consecutive sampling of CP children who met the inclusion and exclusion criteria. The inclusion criteria were male and female children with CP, aged two to ten years, and parent’s approval by signing the informed consent. Exclusion criteria were other neuromotoric diseases (spina bifida, neuromuscular or musculoskeletal disease, etc.), not cooperative

during the study period, and had have severe disease on the day of examination.

Calculation of the sample using a type I error (α) was set at 5% and type II error (β) by 10% with $Z\alpha= 1.96$ and $Z\beta= 1.28$, the correlation coefficient is 0.5 in order to get the sample size is 30. Statistical analysis using a non-parametric test for normality Kolmogorov-Smirnov. Correlation analysis R2 with MP, R1 with MP using Spearman correlation test. The relationship between GMFCS and MP were analyzed with the Kruskal-Wallis test considering the data distribution is not normal

RESULTS

Characteristics of Respondents

One out of 31 children was excluded due to detected with bilateral hip dislocation. Many of the respondents were male. The youngest age of respondents was 24 months old and the eldest was 108 months (55.4 + 22.23 months). Kolmogorov-Smirnov test result showed normal distribution ($p = 0.261$). Majority of respondents had spastic quadriplegic CP whom had prenatal risk factor (Table 1).

Table 1. Demographic and Clinical Characteristics

Variable	N (30)	% (100)
Gender		
▪ Male	18	60.0
▪ Female	12	40.0
Age		
▪ 2-5 years old	23	76.7
▪ >5 years old	7	23.3
Classification of CP		
▪ Spastic hemiplegic	3	10.0
▪ Spastic diplegic	7	23.3
▪ Spastic quadriplegic	20	66.7
Risk factor		
▪ Prenatal	15	50.0
▪ Perinatal	2	6.7
▪ Postnatal	13	43.3

There were 3 children with unilateral hip dislocation, thus the affected limb were eliminated in the study. This created a total of 57 legs were examined for their spasticity and

MP analysis. The measurement of spasticity degree (°) adopted the Modified Tardieu Scale (MTS) and showed that the average of R2 was greater than R1 (Table 2).

Table 2. Characteristic of Spasticity by Modified Tardieu Scale (MTS)

Variable (N=57)	Mean (SD)
▪ R2	34.6 (12.15)
▪ R1	22.3 (11.50)
▪ R2-R1	11.9 (6.86)

The assessment of gross motor skills using the GMFCS showed that there were no level 1 and 2 of GMFCS. The percentages (%) of GMFCS

in sequent were level 3 and 4 by 14%, and level 5 by 71.9% (Table 3).

Table 3. Characteristics of Gross Motor Skill by GMFCS

Variable	N (57)	% (100)
GMFCS		
▪ Level 1	0	0
▪ Level 2	0	0
▪ Level 3	8	14.0
▪ Level 4	8	14.0
▪ Level 5	41	71.9

The AP pelvic X-ray examinations were performed on the 57 limbs. Most of them

(56.1%) were the third MP classification, which was the hip dysplasia (Table 4).

Table 4. Characteristics of Hip Displacement by MP

Variable	Mean (SD)	N (57)	% (100)
MP (%)	29.9 (18.25)		
Classification			
▪ 1 (MP < 10%, normal)		4	7.0
▪ 2 (MP 10% £ 15%, almost normal)		3	5.3
▪ 3 (MP > 15% £ 30%, dysplasia)		32	56.1
▪ 4 (MP > 30% < 100%, subluxation)		18	31.6

The distribution of data by the Kolmogorov-Smirnov test revealed that the R2 and R1 variables were normally distributed (p = 0.004, p = 0.117, respectively), and also the R2-R1

variables were normally distributed (p = 0.068). However, the MP were not normally distributed (p = 0.032).

The Relationships between Variables

The correlation between hip adductor muscle spasticity and MP

Spearman correlation analysis showed no

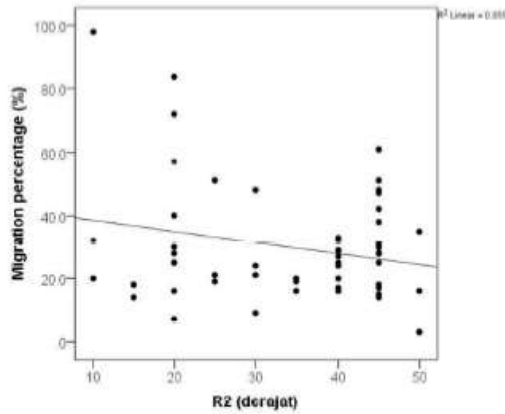


Figure 1. Correlation between R2 MTS of Hip Adductor Muscle and MP

correlation between the R2 and MP ($r = -0.060$; $r^2 = 0.055$; $p = 0.658$). Similarly, among the R1 and MP ($r = -0.136$; $r^2 = 0.059$; $p = 0.314$) (Figure 1 & 2).

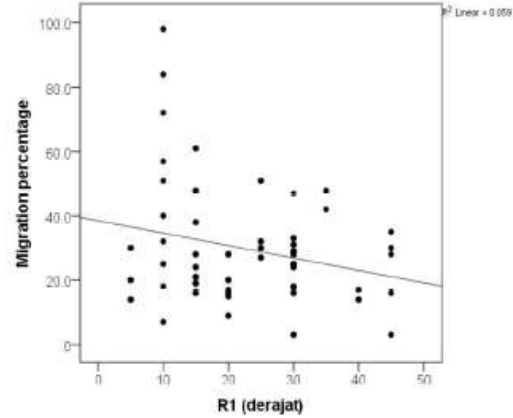


Figure 2. Correlation between R1 MTS of Hip Adductor Muscle and MP

The relationship between GMFCS level and MP
 There were no significant differences between the mean of MP among different GMFCS levels which were analyzed by the Kruskal-Wallis test, while considering the MP distribution data

were not normal (Table 5). This study found that there was no relationship between the level of the femoral head migration and the patient's ability to ambulate.

Table 5. The Relationship between GMFCS Level and MP

GMFCS	Migration Percentage (%)
	Mean (SD)
1 (n=0)	0
2 (n=0)	0
3 (n = 8)	24 (15.0)
4 (n = 8)	35 (26.2)
5 (n = 41)	30 (17.2)
<i>p value*</i>	0.831

* *Kruskal-Wallis test*

DISCUSSION

This study found that most of the respondents were male (60%) which is consequent with the studies by Anggoro SC (2011)²⁶, Gorter

et al. (2009)²⁷, Wu et al. (2010)²⁸, Mutlu et al. (2008)²⁹, and Chounti et al (2013)³⁰.

To avoid bias, aged 2 to 10 years old were set as the inclusion criteria because of the instructions

of GMFCS. There is no evident distinction between level I and II in children below two.^{8,9} The literatures said that the earliest pelvic subluxation can occur at the age of 2 to 10 years old as the upper baseline limit at the puberty age, when there is a growth spurt.

Table 1 shows that the age group of two to five years old represents majority of respondents. Terjesen (2006) in his research on the development of hip joint in children with CP who had not been operated found that increasing MP value in spastic quadriplegic CP at aged below 5 years old was greater compared to other children (13% and 7%, respectively).¹³

It is found that most of respondents have spastic quadriplegic type of CP (Table 1) which is parallel to the studies of Kim et al. (2012)³¹, Cooke et al. (1989)³², Gorter et al. (2009)²⁷, Terjesen (2006)¹³, and Dobson et al. (2002).³³

A total of 57 limbs out of 30 respondents with spastic CP (Table 2) is analysed for their spasticity using MTS method on R2 and R1 components. Similar method had been used by Boyd and Graham (1999) and Boyd et al. (2000) to examine the degree of spasticity. The R2, R1, and R2-R1 components were analysed to determine the possibility of changes due to Botulinum toxin A injection into the spastic muscles of lower extremity.^{34,35}

In regards of gross motor function, this study discovers the presence of level III-IV of GMFCS (Table 3) whilst nil of level I and II. These are consistent with Hagglund et al. (2007)²⁵ and Kim et al. (2012)³¹ findings. The absence of level I and II of GMFCS in this study may be suggested by the fact that these children have good ambulation capacity thus they attend no therapy sessions. Since this study was held in

RSCM, a tertiary referral hospital, hence only worse conditions require treatment here.

Table 4 shows the highest MP value falls on the pelvic dysplasia group ($MP > 15\% \leq 30\%$) with a mean value of 29.9% (18.25%). It is almost the same value obtained by Parrot et al. (2002) in which among 20 pelvic x-rays of bilateral CP children showed the mean MP of right pelvic was 25.1% (13.9%) whereas the left pelvic was 24.95% (11.3%).³⁶ In addition, Hodgkinson et al. (2001) indicated that 52.7% of respondents with MP value below 33% was in the group of without pelvic pain disorders, and 22% in the group with pelvic pain disorders.³⁷

The Relationship between Variables

The correlation between hip adductor muscle spasticity and MP

The Spearman correlation test results show no correlation between R2 and MP ($r = -0.060$; $r^2 = 0.055$; $p = 0.658$); and between R1 and MP ($r = -0.136$; $r^2 = 0.059$; $p = 0.314$). These indicate that the degree of spasticity as measured by R2 and R1 do not have any association with the severity of joint migration as visualized on plain radiography examination. In other words, level of severity does not indicated by hip adductor muscle spasticity regardless the high value of MP (or subluxation), .

Various studies have described hip subluxation or dislocation in children with spastic quadriplegic CP is affected by various factors, such as neuromuscular imbalance, therapeutic benefit and many more. According to Spiegel et al. (2006), hip dislocation is caused by neuromuscular imbalance on the growth and hip joint development. Soft tissues abnormalities in relation with hip dislocation risk show manifestation in hip muscle strength imbalance

between the flexor and adductor that are stronger than the extensor and abductor muscles.

The occurrence of dynamic imbalance will result in myostatic contracture in adduction position with or without the presence of hip flexion position. As a result, it will disrupt patient's growth that cause progressive changes in the femur and acetabulum. The forms of bone abnormalities consist of femoral torsion with or without coxa valga, and abnormal femoral head or acetabulum. Hip contracture in flexion and adduction position will shift the center of hip rotation from the femoral head to the lesser trochanter and gradually to the proximal part of the femur will shift up and out.³⁸

According to Yalcin S et al. (2010) in the HELP Guide to Cerebral Palsy, it is written that the progression of pelvic instability is associated with the combination of muscle imbalance, persistent primitive reflexes, incorrect posture, and no weight bearing exercise to stimulate bone tissue formation. The spastic hip adductor and iliopsoas muscles result in contractures in hip adduction and flexion position. Hamstring muscle spasticity also contribute to muscle imbalance. Excessive muscle tension gives constant pressure against the pelvic that will cause femur and acetabulum deformation. Pelvic abnormalities occur in the form of femur anteversion and coxa valga.¹⁷

Additionally, Bleck EE (1987) concludes that hip dislocation is associated with a persistent infantile hip-flexion contracture and infantile excess anteverted femur (coxa valga). Constant iliopsoas muscle spasticity will shorten the muscles and result in contracture. When spastic iliopsoas muscles are at the time of contracting, they will hit the hip joint capsule medially and lead to femoral head displacement laterally.

Spastic hip adductor muscle is considered as the major force that causes deformity. A medial hamstring muscle contracture contributes to hip dislocation because it will cause the femoral head to deviate out of the acetabulum when the knee is extended and the pelvis is adducted.¹⁸

The following studies also illustrate the importance of physical therapy to prevent hip dislocation. Hagglund et al. (2007) demonstrates the need of non-operative therapy as a prevention program as shown in large amounts of the studied pelvic that received no operative interventions, but they showed decrease in the AI and MP values.²⁵ Supporting this, Yalcin et al. (2010) and Spiegel et al. (2006) reviews show that non-operative therapy is able to prevent or delay the progression of deformity by maintaining the range of motion and decrease spasticity.^{17,38} Physiotherapy is a fundamental part in the management of spasticity because muscle overactivity causes the shortening of muscles and ultimately increases the sensitivity of muscle spindles.

The suggested various treatments include positioning, stretching exercises, neurofacilitation techniques, casting and bracing inhibitif.¹⁷ Two case reports by Picciolini et al. (2009) show that children with clinical CP with supporting radiographic result of hip displacement who are not responsive to pharmacological and operative interventions, are indeed improved with postural management. The given programs at the time of study were physiotherapy focuses on neurodevelopment activities twice a week, and postural program that requires patient to sit five hours per day using a special tool (siege moule). These showed decrease of MP value during follow-up session.³⁹ Furthermore, a retrospective study by Pountney et al. (2002) concludes

that postural management plays an important role in preventing hip dislocation. Postural management, such as lying, sitting and standing positions are significantly maintain the integrity of the pelvic.⁴⁰ In this current study, respondents who underwent therapy in RSCM received positioning, stretching, neurofacilitation therapies and equipped with a special chair for home use. The mean duration of therapy 13.8 ± 15.78 months; minimum length of therapy was 0 months and maximum 70 months.

Researchers did not get any study of spasticity measurement with MTS on R2, R1, and R2-R1 components on the single hip adductor muscle while assessing hip dislocation. This study adopted Gorter et al. (2009) research in which the spasticity measurements of the hip adductor muscle, gastrocnemius muscle and the hamstring muscles (the third this value is calculated as the total score of spasticity) were involved to see the relationship between spasticity and GMFM-66 that is used to assess gross motor function.²⁷

Above discussion has conclude no correlation between the hip adductor muscle spasticity and MP value in this study because the cause of hip dislocation is multifactorial. In addition, the MP only assesses femoral head displacement to the lateral, known as biomechanics of hip dislocation that carries out in a complicated series.

The relationship between GMFCS level and MP
This study showed insignificant differences between the mean MP among all level groups of GMFCS. Hagglund et al. (2007) found a direct correlation between the incidence of hip subluxation with the GMFCS level.²⁰ Soo et al. (2006) conducted a cohort study on 323 CP children for about 2 years found the incidence of hip subluxation for about 35%

and has shown a linear relationship with the GMFCS level.¹⁴ Nonetheless, Beckung et al. (2007) demonstrates that higher level of GMFCS produces lower score of Gross Motoric Function Measure (GMFM).⁴¹ Kim and Park (2011), in their research found a significant direct relationship between spasticity and gross motor function, including muscle strength and gross motor function.⁴²

As previous discussion on the relationship of hip adductor muscle spasticity and MP, the researchers considered that the results in this study showed no significant differences between the mean MP and the level group of GMFCS which is closely associated with the duration of therapy received by the respondent.

Limitations of this study include inappropriate study time when most of respondents were on the treatment period, hence the measurement results might be biased; and target of spasticity examination was performed only on the hip adductor muscle spasticity alone whilst the cause of hip dislocation is multifactorial. Moreover, This study had not discuss about the type of therapy received by each respondent.

CONCLUSION

This study suggests that there is no correlation between the degree of hip adductor muscle spasticity with Migration Percentage value in the detection of hip dislocation. There is no significant difference in the level of Gross Motor Function Classification System with Migration Percentage value in the detection of hip dislocation.

It is advisable to build an integrated team among the Pediatrics, Physical Medicine

and Rehabilitation (PMR), Radiology and Orthopaedic departments. The PMR management team needs to perform pelvic anteroposterior radiograph series and makes immediate referral to the orthopaedic department as required in order to delay the onset of hip dislocation. Further research is needed to seek other factors that influence the MP value. Moreover, a thorough evaluation on the effectiveness of treatments in determining the degree of spasticity, GMFCS classification and value of the MP by involving new patients.

Researchers recommend the use of a more convenient goniometer in the future because of the difficulty faced by researchers in holding the tool while moving the limb and maintaining the extension position whereas this position is hardly accomplished due to spastic hip and knee flexor muscles when performing measurement of hip adductor muscle spasticity using the Modified Tardieu Scale on R2 and R1 components.

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