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Prevalence and predictive factors of hip displacement in children with cerebral palsy at Paediatric Institute, Kuala Lumpur Hospital

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Abstract

Objective: We aim to study the prevalence and predictive factors for hip displacement, in order to justify a hip surveillance programme for children with cerebral palsy (CP) in Malaysia. *Methods:* Children aged 2 to18 years old with CP were recruited from September 2013 till June 2014. The hip joint migration percentage (MP) and acetabular index (AI) were measured on all hip radiographs. The CP subtype was determined and gross motor function was classified according to the gross motor function classification system (GMFCS).

Results: Seventy-five children were recruited. Fifty-five percent of them had marked hip displacement with MP ⩾ 30% and 15% developed hip dislocation (MP=100%). Marked hip displacement occurred as early as age of 2 years and most hip dislocations were detected by age of 10 years. The risk of marked hip displacement was directly related to the GMFCS level, from none in GMFCS I to 75% in GMFCS V. There was a moderate positive correlation between the initial AI and initial MP.

Conclusions: One in every two children with CP was at risk of hip displacement, with GMFCS level and initial AI as significant predictive factors. We recommend a hip surveillance programme for Malaysian children with CP, based on the child's age and GMFCS level, with both MP and AI as indicators for hip surveillance.

Keywords: Cerebral Palsy, hip displacement, prevalence, predictive factors

INTRODUCTION

Hip displacement is a common and serious complication in children with severe cerebral palsy (CP). It may lead to pain and discomfort, functional impairment affecting the ability to sit, stand, walk and will also interfere with dressing and perineal care. Hip displacement has been shown to adversely affect the child's quality of life and increases caregiver burden.[1-2]

Early identification of these "at-risk" children is critical. Early preventive orthopaedic surgery and targeted botulinum toxin injection over hip adductors are effective modalities in enhancing hip stability, and thus prevent hips at risk from further displacement and eventually dislocation.[3-4] As a result, hip surveillance has now been adopted as standard of care in the management of children with CP in many developed countries. Hip surveillance is the process of identifying and monitoring the critical early indicators of progressive hip displacement, and triaging to orthopaedic services as part of the overall prevention of hip dislocation.

Migration percentage is an early indicator of hip displacement. It measures the single most important qualification of a hip joint, which is an adequate coverage of the femoral head by the acetabular roof. Cut-off values of 30 to 33% and 90 to 100% were used in the definitions, respectively for marked hip displacement and hip dislocation from most studies.[5-11] As it is easy to measure, is little influenced by the rotational position of the femur, and has good inter-observer agreement, it has been universally accepted as a good and reliable indicator of hip displacement.[12-15]

On the other hand, hip dysplasia in CP is the second most common deformity after ankle equinus and its incidence and severity is directly proportionate to the gross motor function classification system (GMFCS) level.[16-17] Early dysplasia results in failure of proper development of the proximal femur and the acetabulum, later on hip displacement and dislocation. Acetabular dysplasia can be assessed by acetabular index (AI). However, the recommendations to use AI as an indicator for hip surveillance vary in literatures.

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Cooke *et al*. found that AI was a single powerful radiographic predictor for hip displacement.[13] Other studies suggested AI should be considered as prognostic indicator only when the child approached 48 to 60 months of age, based on the fact that hip displacement preceded acetabular dysplasia.[3,9,18] Recently, Terjesen found AI was a significant prognostic factor for hip displacement in children less than 30 months of age as well as older children.[18]

The rate of hip displacement was found to be one-third in children with CP from two population studies in developed countries, and was not related to the movement disorder but was directly related to gross motor function.[3,5] It is perceived that the prevalence of hip displacement is similarly high in children with CP in Malaysia especially those with severe disabilities. However to our knowledge, there is no local published data on hip displacement in children with CP. The aim of this study is todetermine the prevalence and predictive factors of hip displacement among children with CP at Institute of Paediatrics, Kuala Lumpur Hospital, in order to justify a local hip surveillance programme.

METHODS

Participants and data collection

This was a cross-sectional study. All children aged 2 to 18 years old with CP, who attended paediatric neurology clinic, CP clinic or admitted to neurology ward were recruited over a period of 10 months, from September 2013 till June 2014. Children included in the study must have at least one hip radiograph of acceptable quality. We also included children who have undergone surgery for hip displacement, received botulinum toxin injections, using assistive devices and physiotherapy. Children with hip displacement due to causes other than CP, those with incomplete data or whose parents refused to participate in the study were excluded. Figure 1 illustrates the recruitment process.

Data was collected using a data collection form filled by the attending paediatric neurologist or clinical fellow during the clinic session. Some preliminary work had been undertaken with a small number of parents and children to pilot the instruments and evaluate the data collection form. The source of data was obtained from the children's case notes and specific information from the children or caregiver.

The CP subtype was determined by the attending paediatric neurologist or clinical fellow and gross motor function was classified according to the age-related GMFCS. GMFCS is a five-level ordinal scale in which level I corresponds to the highest and level V to the lowest function.[19] Children in GMFCS IV and V (non-ambulators) were defined as severe CP in this study. Children whom gross motor function could not be determined reliably were excluded from the study.

As the degree of hip displacement in CP is best determined and monitored by Reimers' migration percentage (MP), the MP was measured on all available hip radiographs.[12] The MP describes the percentage of the ossified femoral head displaced lateral to the acetabular margin on an anteroposterior hip radiograph (Figure 2A). All

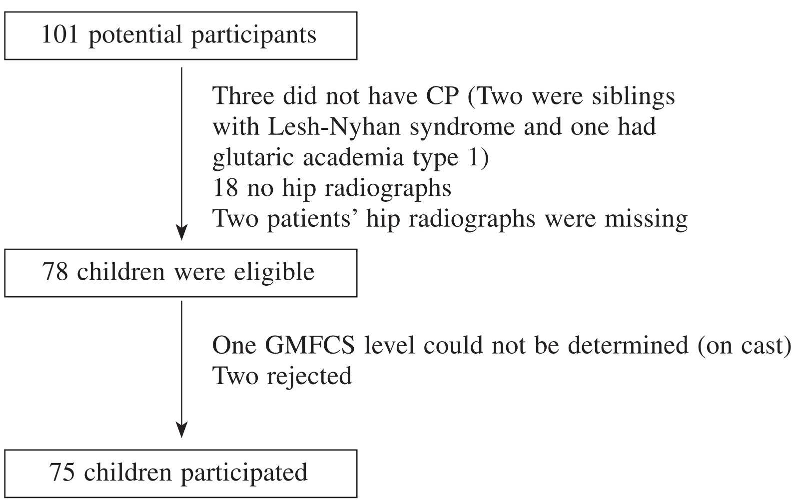

Figure 1. Recruitment process

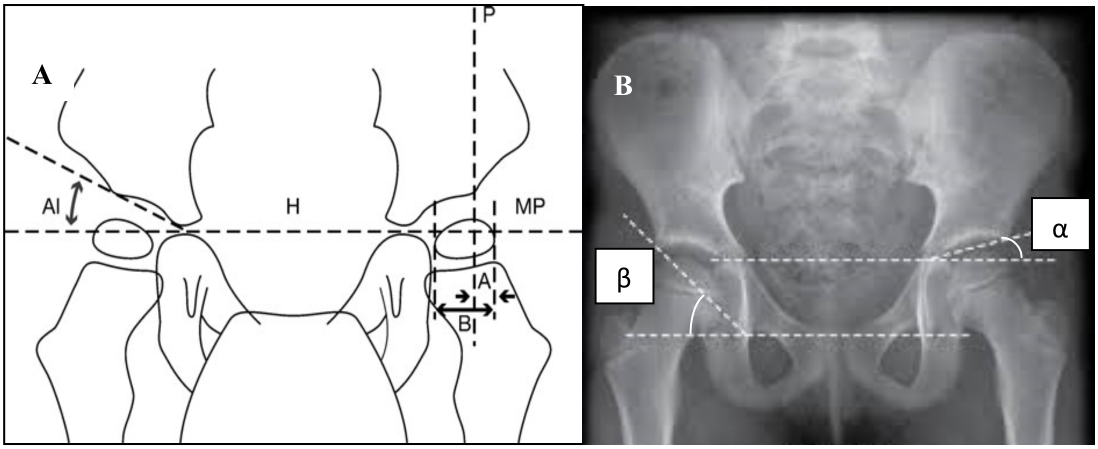

Figure 2. A) The Migration Percentage (MP) is obtained by identifying Hilgenreiner's line (H) and Perkin's line (P) and then measuring the proportion (%) of capital epiphysis that has migrated beyond Perkin's line laterally (A/B x 100). The acetabular index (AI) is the angle between the slope of the acetabulum and Hilgenreiner's line. (H, Hilgenreiner's horizontal line between triradiate cartilages, P, Perkin's line drawn perpendicular to the H-line at the lateral margin of the acetabulum).[11] B) Different ways of measuring the acetabular index in children (α) and in adult (β).[23]

measurements were performed by either one of the authors. Depending on their migration percentage, the hips were classified as normal (MP <10%), hip displacement (MP 10 to 29%), hip at risk with marked displacement (MP 30 to 99%), and dislocation (MP =100%).[3,19] In this study, children with hips showing MP⩾30% were analysed in relation to those with hips below these limits. The hip with higher MP determined the classification of the child.

The acetabular index (AI) of the worse hip was also measured (Figure 2A). As triradiate cartilage closure occurs at puberty (approximately 13 years of age in girls and 15 years in boys), and therefore inapparent in hip radiograph, the inferior margin of the pelvic tear drop is used instead to measure AI in teenagers and adults (figure 2B).[20-22] This shifts the horizontal line inferiorly and changes the value of the angle. In order to obtain a uniform and standardised measurement of AI, hip radiographs with inapparent triradiate cartilage due to its closure were excluded from AI measurement.

For the purpose of this study, acceptable hip surveillance is defined as at least one hip radiograph was done on first visit to paediatric neurology clinic or CP clinic for a new patient or at least two hip radiographs were done in two consecutive years for an old patient. The prevalence (period prevalence) of hip displacement was calculated as the number of children with MP⩾ 30% over the study period. The potential predictive factors for hip displacement were studied, i.e. child's age, gender, race, weight percentile, GMFCS level, type of cerebral palsy, age when first hip radiograph was done, whether hip surveillance was done, timing of hip surveillance (early or late), acetabular index, botulinum toxin injection, physiotherapy and the use of assistive devices. For the purpose of this study, late hip surveillance is defined as the first pelvic X-ray was done after the age of 30 months old, for patient presented before 24 months of age, or the first pelvic X-ray was not done on first visit to paediatric neurology or CP clinic if their age at presentation was older than 30 months old.

The study was approved by the Medical Research and Ethics Committee, Ministry of Health, Malaysia (NMRR-13-753-17312). Informed consents were obtained from all parents/legal guardians of children participating in the study. Verbal assents were obtained from children above seven years of age if they could comprehend.

Statistical analysis

Data was analysed using the Statistical Package for the Social Sciences (SPSS) version 20 software. To assess the statistical differences between groups, Student's *t*-tests and χ^2 test were used when two groups were compared and analysis of variance (ANOVA) was used when more than two groups were compared. Correlation between parameters was evaluated by Pearson's correlation coefficient (r). Differences were considered significant when the p-value was <0.05. Multivariate analysis using logistic regression model was used to look for predictive factors of hip displacement.

RESULTS

General characteristics and prevalence of hip displacement

A total of 75children with initial hip surveillance were recruited. Out of these, 41(55%) children had marked hip displacement with MP ⩾ 30% on first hip radiograph at the mean age of 5.8 years (SD 3.00 years). Hip dislocation occurred in 11 children at a mean age of 7.3 years (SD 3.26 years). The general characteristic of the participants is described in Table 1.

General characteristic of the 18 children without hip radiographs is described in Table 2. Most patients (88.9%) without hips radiographs were in GMFCS I-III. Hip radiographs were not ordered as clinically there were no signs of hip displacement.

Of these 75 children, only 37 (40%) had acceptable hip surveillance as defined in this study, i.e. at least one hip radiograph was done on first clinic visit for a new patient or at least two hip radiographs were done in two consecutive years for an existing patient.

Prevalence of hip displacement in relation to GMFCS level

The risk of hip displacement increased progressively with higher level of GMFCS as shown in Figure 3. The proportion of children with marked hip displacement (MP ⩾30%) increased from none in GMFCS I to 75% in GMFCS V (p=0.006). Twenty-two percent of 49 children (n=11) with the lowest level of function, GMFCS V, had hip dislocation but these phenomenon was not observed in children with GMFCS I to IV.

Prevalence of hip displacement in relation to cerebral palsy subtype

The proportion of children with various degree of hip displacement in relation to CP subtype is presented in Figure 4. More than 75% of children

Table 1: General characteristics of participants

Characteristics	Total, N= 75 (%)
Age (median, IQR, year)	7.7, 6.5
Sex	
Male	42 (56.0)
Female	33 (44.0)
Gross motor function classification system (GMFCS)	
Level I	1 (1.3)
Level II	6 (8.0)
Level III	7 (9.3)
Level IV	12 (16.0)
Level V	49 (65.3)
Type of cerebral palsy	
Spastic hemiplegia	4 (5.3)
Spastic diplegia	16 (21.3)
Spastic triplegia	7(9.3)
Spastic quadriplegia	21(28.0)
Mixed spastic dystonic quadriplegia	18 (24.0)
Dystonic quadriplegia	8 (10.7)
Choreoathetoid	1(1.3)
Age at first hip radiograph (mean±2SD, year)	6.0 ± 3.33
Age at the first hip radiograph showing MP ⩾ 30% (mean±2SD, year)	5.8 ± 3.00
Age at the first hip radiograph showing MP = 100% (mean±2SD, year)	7.3 ± 3.26
First migration percentage (mean±2SD, %)	36.9 ± 25.61
Acetabular index (mean±2SD,°)	23.2 ± 7.72

Table 2: General characteristics of children without hip radiographs

Characteristics	Total, N= 18 (%)
Age (mean, SD, year)	7.7 ± 4.42
Sex	
Male	12 (66.7)
Female	6 (33.3)
Gross motor function classification system (GMFCS)	
Level I	3 (16.7)
Level II	10 (55.6)
Level III	3 (16.7)
Level IV	0 (0.0)
Level V	2 (11.1)
Type of cerebral palsy	
Spastic hemiplegia	9 (50.0)
Spastic diplegia	4 (22.2)
Spastic triplegia	3 (16.7)
Spastic quadriplegia	2 (11.1)

with quadriplegic CP had marked hip displacement (MP ⩾30%). This was observed in children with spastic quadriplegia, dystonic quadriplegia and mixed spastic dystonic quadriplegia. All children with quadriplegic CP were non-ambulators with GMFCS IV and V (Table 3). Similarly, all children who developed hip dislocation were children with quadriplegic CP, ranging from 11% in mixed spastic dystonic quadriplegic CP to 33% in spastic quadriplegic CP. In contrast, children with choreoathetoid and spastic hemiplegic CP had the lowest risk of developing marked hip displacement whereas children with spastic diplegia and triplegia carried intermediate risk.

The only one child with choreoathetoid CP was in GMFCS V but his serial hip radiographs showed stable hips (MP <30%) throughout follow-up period (3 years 6 months). Four out of 7 (57%) children with spastic triplegia were non-ambulators, and 75% of them (n=3) had marked hip displacement. However, all spastic triplegic children (n=3) in GMFCS I to III did not develop marked hip displacement. For children with spastic hemiplegia and spastic diplegia, 78 to 100% of them in GMFCS I to III had stable hips (MP < 30%) but 43 to 50% of them in GMFCS IV to V had marked hip displacement.

Acetabular index and hip displacement

Of the 75 children with hip radiographs done, 63 children had their AI measured for the worse hip

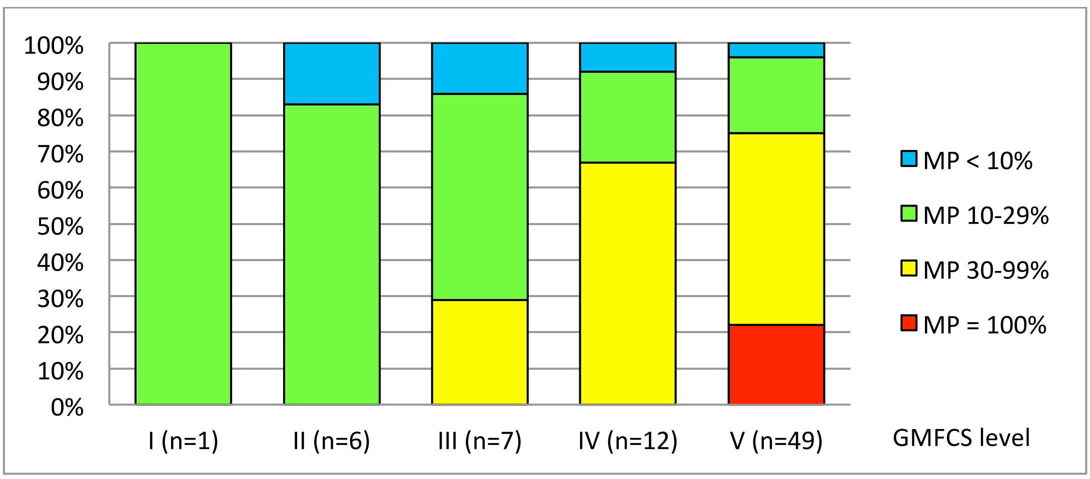

Figure 3. Proportion of children (%, n=75) with different severity of hip displacement in relation to their gross motor function classification system (GMFCS) level.

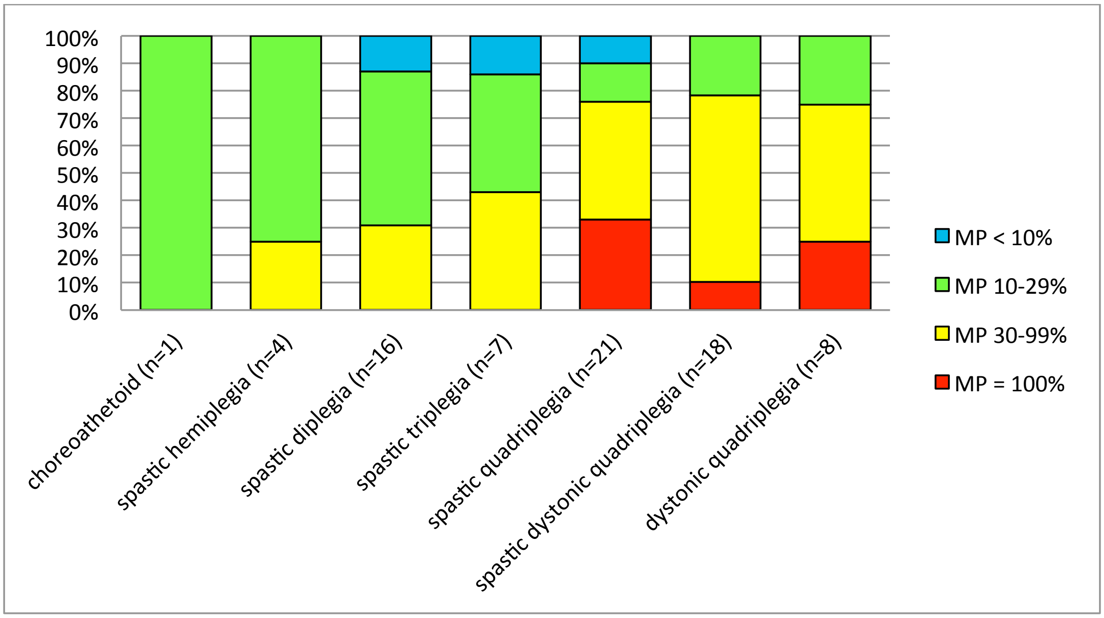

Figure 4. Proportion of children (%, n=75) with various degree of hip displacement in relation to cerebral palsy subtype

on initial radiographs. AI were not measured in 12 children due to: closure of the triradiate cartilage (7 children, mean age 13.5 SD 2.61 years), rotated hip radiographs in 2 children (Hilgenreiner's line could not be obtained reliably but MP could be measured as both hips were completely dislocated from these two radiographs) and failure to trace the initial radiographs in three children (MP were measured by one of the researcher in the past and documented clearly but radiographs were not traceable).

The mean AI for the worse hip was 23.2° (SD 7.72°) at the initial hip radiographs. Initial mean AI was highest in the children with hip dislocation (29.4°, SD 10.92°), followed by children with marked hip displacements (MP 30 to 99 %) and children with MP < 30%, with mean AI of 24.6° (SD 7.99°) and 20.1° (SD 4.83°) respectively. Mean initial AI was significantly different between these three groups of children (p=0.006). Using Bonferroni post-hoc test, mean initial AI was

Table 3: Children with various degree of hip displacement in relation to cerebral palsy subtype and gross motor function classification system (GMFCS) level

CP subtypes	GMFCS level				Total, N
	I-III (n, %)		IV-V (n, %)		
	MP < 30%	MP ⩾ 30%	MP < 30%	MP ⩾ 30%	
Spastic hemiplegia	2 (100)	0 (0)	1 (50)	1(50)	4
Spastic diplegia	7 (78)	2 (22)	4 (57)	3 (43)	16
Spastic triplegia	3 (100)	0 (0)	1(25)	3 (75)	7
Spastic quadriplegia	0 (0)	0 (0)	5 (24)	16 (76)	21
Mixed spastic dystonic quadriplegia	0 (0)	0 (0)	4 (22)	14 (78)	18
Dystonic quadriplegia	0 (0)	0 (0)	2 (25)	6 (75)	8
Choreo-athetoid	0 (0)	0 (0)	1(100)	0 (0)	1
Total, N	12	2	18	43	75

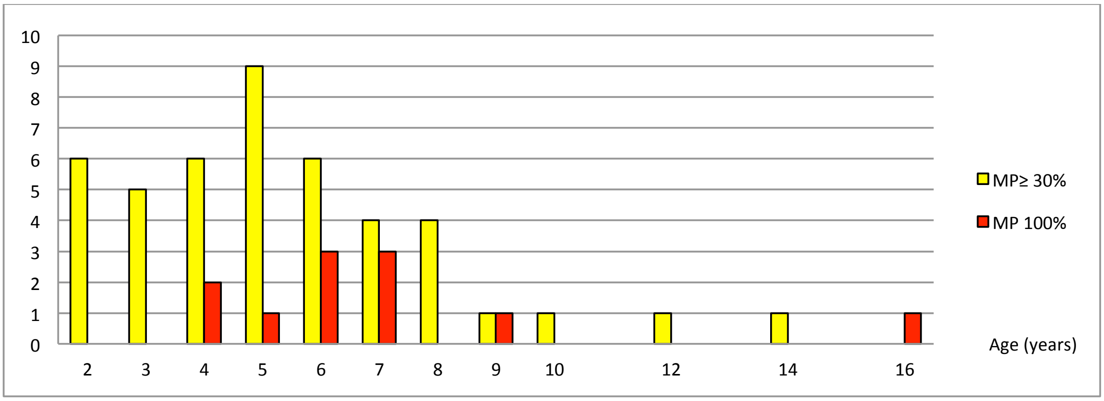

Figure 5. Number of children with severe cerebral palsy (Gross motor function classification system -GMFCS IV-V) related to age (years) at first registration of migration percentage (MP) ⩾ 30% and 100 % (n= 45)

significantly different between MP < 30% and hip dislocation group (p=0.010), but not between MP < 30% and MP 30 to 99% groups (p=0.071), and hip dislocation and MP 30 to 99% groups (p=0.346).

Predictive factors for hip displacement

Multivariate analysis using logistic regression model was performed to evaluate the variables that had the greatest influence on the MP. The variables studied were child's age, gender, race, weight percentile, GMFCS level, type of cerebral palsy, age when first hip radiograph was done, whether acceptable hip surveillance was done, timing of hip radiograph (early or late), acetabular index, botulinum toxin injection, physiotherapy and the use of assistive devices.

GMFCS level and AI were the only variables with significant influence. The risk of marked hip displacement was significantly higher in children with severe CP (GMFCS IV to V) as compared to children with GMFCS I to III (adjusted OR 9.31, 95% CI: 1.7-50.4 with p = 0.010). Higher AI similarly increased the risk of marked hip displacement with adjusted OR 1.16, 95% CI: 1.05-1.28 and p = 0.003.

Age at first detection of significant hip displacement among children with severe cerebral palsy

As 75% of children with severe CP developed marked hip displacement, we also looked into the age of presentation when this group of children first detected MP ⩾ 30% from hip radiograph. The age at first registration of MP ⩾ 30% or detected hip dislocation is presented in Figure 5. The mean age at the first radiograph showing marked hip displacement was 5.9 years with SD 3.04 years (range 2 to 16 years). Children with severe CP developed marked hip displacement as early as the age of 2 years. The incidence peaked at the age of 5 years then gradually reduced in number with fairly low incidence from 9 years onwards. When we looked into hip dislocation in children with severe CP, it occurred at a mean age of 7.3 years, with SD 3.26 years (range 4 to16 years). It was detected as early as 4 years old. No children with severe CP in this study developed hip dislocation after 10 years of age except one child whose hip was noted to have dislocated at the age of 16 years old.

DISCUSSION

Prevalence of hip displacement in Malaysia

The prevalence of hip displacement is estimated to occur in 25 to 60%, while complete hip dislocation is reported to be between10 and 15%.[3,13, 24-27] Our study yielded similar finding with period prevalence of hip displacement occurred in 55% and complete hip dislocation in 15%.

Predictive factors for hip displacement

Recent population-based studies showed the prevalence of hip displacement is directly related to the GMFCS levels.[3,5,6,28,29] This correlation was similarly seen in the present study (Figure 3). Children who are non-ambulators (GMFCS level IV-V) are at significantly higher risk of marked hip displacement as compared to children who are ambulating (GMFCS level I to III). In children with GMFCS IV to V, hip displacement was more frequently seen in spastic quadriplegic, dystonic quadriplegic and mixed spastic dystonic quadriplegic CP than in choreoathetoid CP, indicating that spasticity and dystonia are important aetiological factors.

Children with hemiplegia, diplegia and triplegia who had good gross motor function had very low incidence of marked hip displacement (Table 3). However, in children with poor gross motor function (GMFCS level IV to V), almost half of the children in same CP subtype developed marked hip displacement. GMFCS remains as a significant predictive factor when multivariate analysis was done using logistic regression model, but not for CP subtypes. This finding suggests GMFCS level is a better clinical determinant on risk of hip displacement as compared to CP subtypes. Furthermore, the description for the CP subtypes has not been universally agreed on, and it is sometimes difficult to determine CP subtypes at young age. In contrast, GMFCS has been proven to be a valid and reliable tool and has been reported to remain relatively stable over time, from as early as 2 years of age.[23,28-31]

In our study, mean initial AI for children with marked hip displacement and hip dislocation were 24.6° and 29.4° respectively. The recommended AI threshold value for intervention in the literature lies between 27° to 30°.[7,13,18,28,29,32,33] Cornell *et al*. found poor results in 13 of 15 hips with pre-operative AI ⩾ 27°[34] whereas Dobson *et al*. recommended preventive surgery in those with MP ⩾ 40% and AI ⩾ 27°.[7] The significant positive correlation between the AI and MP in this study is also consistent with other series.[8,18,35]

Our result suggests that children with AI ⩾ 25° and MP ⩾ 30% should be closely monitored. Appropriate preventive measures should be instituted timely before it progresses to hip dislocation. Since both MP and AI could be measured from the same anteroposterior pelvic radiograph, and AI has additional prognostic value in term of surgical intervention, AI should be considered as a supplementary indicator of hip displacement besides MP.

Age at first detection of significant hip displacement among children with severe cerebral palsy

The mean age of dislocation was 7.3 years in our study, which is consistent with other clinical series.[8,27,36] However, children with severe CP developed marked hip displacement as early as the age of 2 years, with the highest incidence at the age of 5 years.

The ultimate aim of hip surveillance programme is to prevent hip dislocation. Therefore, children with CP should be diagnosed as early as possible so that hip surveillance can be instituted as early as the age of 2 years, and necessary preventive measures can be done once hip displacement is detected.

Hip surveillance in Malaysia

Hip surveillance for children with CP has been adopted as standard of care in many developed countries.[11] A survey conducted in Europe in year 2012 showed 15 of 31 responders across Europe registered hip status in children with CP, and 20 reported guidelines for hip management which had been used for 1 to 23 years.[37]

We were not surprised with the reported 40% acceptable hip surveillance rate in our centre. At a glance, hip surveillance seems misleadingly simple and straightforward. It is common to think that it would be carried out by practitioners, since there is increased awareness among medical professionals taking care of children with CP and guidelines are easily accessible. Unfortunately, there is no evidence that current local practice gives adequate attention to hip surveillance as part of the comprehensive care bundle for children with CP. Many children were frequently not seen until their hips were dislocated. Some had never had their hip radiographs done and in others though it was done, early hip displacement was ignored and no proper follow up was given.

Unless hip surveillance programme is being adopted as standard of care, the surveillance might not be carried out by those who have first contact with the child, generally the general paediatricians and family medicine physicians. Cooperation and support from the caregivers are also very important in this surveillance programme. This can be achieved by increasing caregiver's awareness about the importance of hip surveillance, as early preventive treatments are available to prevent further displacement and dislocation.[3-4]

Recommendations

Our results provide first local epidemiological data that could serve as the foundation to propose a framework for local hip surveillance programme in children with CP. Based on the findings of this study, we would make the following recommendations:

(1) All children with CP should have regular hip radiographs done based on the GMFCS level,
    - GMFCS I to II: Hip radiograph if clinically suspicious of hip displacement, or evidence of pelvic obliquity or scoliosis. If MP not stable (MP ⩾ 30% or annual

rate of MP >10%), repeat six monthly until stability is achieved.
- GMFCS III: Initial hip radiograph at two years or at age of CP diagnosis whichever is the later. Continue six monthly surveillance until 5 years old. Then yearly surveillance until skeletal maturity if MP stable (MP <30% or annual rate of MP < 10%).
- GMFCS IV to V: Initial hip radiograph at two years or at age of CP diagnosis whichever is the later. Continue six monthly surveillance until 10 years old. Then yearly surveillance until skeletal maturity if MP stable (MP <30% or annual rate of MP < 10%).

(2) Acetabular index should serve as a supplementary predictive and prognostic indicators of hip displacement, with closer monitoring and early referral to orthopaedic surgeons if MP ⩾ 30% and AI ⩾ 25°.

The above recommendations are based on the data from this study, taking into consideration the limited resources in our local setting. It is meant to be more practical and easier to follow compared to the published recommendations from developed countries e.g. Australia.[11] It should serve as a preliminary guideline before more comprehensive local guideline can be established based on larger population-based study.

Limitations

Although this is a cross-sectional study of 75 children with CP aged 2 to 18 years of age, majority of our study population (81.3%) consists of children with severe CP. This phenomenon could be explained by the fact that our centre is a tertiary centre and we tend to see more children in severe spectrum referred by general paediatrician from district hospitals. Some children with less severe CP with GMFCS I and II might have been excluded from the study because no hip radiograph were done as the managing clinicians did not think there was likely any hip displacement. As this is a single centre study, it may not truly reflect the prevalence of hip displacement in children with CP in the wider population. Hence, a follow up population-based study should be conducted, ideally when National CP Registry is established in Malaysia.

In conclusion, marked hip displacement in CP occurs as early as 2 years of age and most hip dislocation occurs by 10 years of age. The risk of marked hip displacement is directly proportionate to GMFCS level and AI, with severe quadriplegic CP at highest risk. We propose that hip surveillance programme with radiographic examinations should be the standard of care for children with CP, with both MP and AI as indicators for hip surveillance and preventive intervention.

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DISCLOSURE

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Conflict of interest: None

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