CHILDERN’S ORTHOPAEDICS

Prediction of hip displacement in children with cerebral palsy

DEVELOPMENT OF THE CPUP HIP SCORE

M. Hermanson, G. Hägglund, J. Riad, E. Rodby-Bousquet, P. Wagner

From Lund University, Lund, Sweden

Hip displacement, defined in this study as a migration percentage (MP) of more than 40%, is a common, debilitating complication of cerebral palsy (CP). In this prospective study we analysed the risk of developing hip displacement within five years of the first pelvic radiograph.

All children with CP in southern and western Sweden are invited to register in the hip surveillance programme CPUP. Inclusion criteria for the two groups in this study were children from the CPUP database born between 1994 and 2009 with Gross Motor Function Classification System (GMFCS) III to V. Group 1 included children who developed hip displacement, group 2 included children who did not develop hip displacement over a minimum follow-up of five years. A total of 145 children were included with a mean age at their initial pelvic radiograph of 3.5 years (0.6 to 9.7).

The odds ratio for hip displacement was calculated for GMFCS-level, age and initial MP and head-shaft angle. A risk score was constructed with these variables using multiple logistic regression analysis. The predictive ability of the risk score was evaluated using the area under the receiver operating characteristics curve (AUC).

All variables had a significant effect on the risk of a MP > 40%. The discriminatory accuracy of the CPUP hip score is high (AUC = 0.87), indicating a high ability to differentiate between high- and low-risk individuals for hip displacement. The CPUP hip score may be useful in deciding on further follow-up and treatment in children with CP.

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Cerebral palsy (CP) is the most frequent cause of motor disability in children. Hip displacement is a painful and severe complication of CP which can be prevented with regular clinical and radiographic examination and early intervention as part of a surveillance programme. Migration percentage (MP) is commonly used to assess lateral displacement of the femoral head on the pelvic radiograph. Hips with MP > 40% are at a high risk of further displacement, indicating the need for surgical intervention. In hips with MP 33% to 40%, other parameters used in deciding further management include clinical signs, the type of CP and the child’s age.

Coxa valga is often seen in children with CP and can be measured as the neck-shaft angle (NSA). The femoral head is also often in valgus in relation to the femoral neck. These combined deformities can be measured as the head-shaft angle (HSA) (Fig. 1). HSA is less affected by hip rotation than the NSA; there is only a 5° measurement error for hip rotation of up to 45°. A previous study showed a correlation between increasing HSA and the risk of hip displacement (MP > 40%) over a follow-up period of five years.

In southern Sweden a surveillance programme was initiated for children with CP, known as CPUP, in 1994. The severity of CP is classified according to Gross Motor Function Classification System (GMFCS) levels I to V. Children at GMFCS V are the most affected. All children in the programme are examined according to a schedule based on GMFCS level and age which includes radiographs and physical examinations. Children with more severe gross motor limitations (GMFCS III to V) are examined annually until the age of eight years, and thereafter radiographic examinations are performed as indicated.

In the present study we developed a risk score, called the CPUP hip score, for children with CP at GMFCS III to V, as a tool to determine the individual risk for hip displacement within five years.

Patients and Methods
This prospective study was based on data from CPUP. This database includes all children...
HSA was measured by the angle intersecting two lines; one passing through the proximal mid diaphyseal line of the femoral shaft and a second perpendicular to the proximal femoral physis (Fig. 1). MP was measured as shown in Figure 1.

**Statistical analysis.** We analysed the risk of developing hip displacement within five years of the first radiographic examination, as defined by a MP > 40%. The odds ratio (OR) with 95% confidence intervals for hip displacement was calculated for GMFCS-level, age, and initial MP and HSA. A risk score was constructed with these variables using multiple logistic regression analysis. The discriminatory accuracy of the risk score was evaluated using the area under the receiver operating characteristics curve (AUC).

The AUC is the area under the sensitivity plotted against 1 - specificity. The AUC itself corresponds to the probability that a child who develops hip displacement within five years is predicted to be at a higher five-year risk than a child who does not develop hip displacement in the same time span. Because an AUC calculated on the same data used to estimate the risk score parameters may be overly optimistic, potential effects of over-fitting on the AUC was evaluated by means of ten-fold cross validation; a method used for investigating how well prediction results generalise to similar populations. In ten-fold cross validation, the study data are randomly divided into ten subsets, while the tenth is used for testing the accuracy of the predictions of the resulting risk score. The procedure is then repeated for each subset and the results are summarised to produce a single valid risk score. The procedure is then repeated for each subset and the results are summarised to produce a single valid risk score.

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A total of 51 children developed hip displacement during the study period, with a MP of either hip > 40%. Furthermore, five children in GMFCS III to V and primary MP < 40% were excluded; four moved out of the defined area and one child was deceased at follow-up. In total 11 children were excluded because of previous hip surgery.

We found that all variables (GMFCS-level, age, MP, HSA) had a significant effect on the risk of hip displacement (MP > 40%) within five years of the first radiographic examination (Table II). The risk of hip displacement decreased with age but increased with a higher level of GMFCS, a higher degree of HSA and a higher MP.

The equation for calculating the risk score (RS) was determined to be:

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RS = -14.1 + 0.71 \cdot \text{GMFCS}_{IV} + 2.48 \cdot \text{GMFCS}_{V} + 0.07 \cdot \text{HSA}_{\text{max}} + 0.09 \cdot \text{MP}_{\text{max}} - 0.5 \cdot \text{Age}
\]

GMFCS\(_{IV}\) is a dichotomous indicator variable which assumes the value 1 when the individual has GMFCS level IV, and 0 otherwise. GMFCS\(_{V}\) is the corresponding indicator variable for GMFCS V. HSA\(_{\text{max}}\) is the maximum HSA when measuring both hips. MP\(_{\text{max}}\) is the maximum MP when measuring both hips. Age is the age in years at the first radiographic examination. The risk score can be translated into the risk of hip displacement using Table I. For example, a child with GMFCS-level IV, HSA\(_{\text{max}}\) 16.5, MP\(_{\text{max}}\) 15 and an age of four will have a risk score of -2.49 and a risk of developing hip displacement (MP > 40%) within five years of 0% to 10%.

The sensitivity and specificity of the risk score is shown in Figure 2. The sensitivity is the proportion of children correctly predicted to have a displaced hip within five years, while the specificity is the proportion of children correctly predicted not to have a displaced hip within five years according to what risk level is chosen to indicate a high-risk individual (Fig. 2).

The predictive ability of the CPUP hip score is high, AUC = 0.87, indicating that it has a high ability to differentiate between high- and low-risk individuals. The cross validation AUC was 0.85 which lessens concerns of over-fitting and confirms the predictive ability of the CPUP hip score.

**Discussion**

To our knowledge, there are no studies published on scores that predict the development of hip displacement in children with CP.

The CPUP hip score AUC value of 0.87 demonstrates great accuracy in differentiating the risk for individuals of developing hip displacement within five years.

The hips were examined with AP radiographs, which is a limitation when measuring HSA. However, it has been shown that the measurement error for HSA is ≤ 5° for hip rotation of up to 45°, and that differences in the rotational position of the femur do not have an important effect on the measurement error of HSA. We considered the use of the Hilgenreiner epiphyseal angle (HEA) (the angle between the proximal femoral physis and Hilgenreiner’s line) as Craven, Pym and Boyd showed that HEA was significantly lower in displaced hips. In hips positioned in neutral position, the HEA is the complimentary angle to HSA. However, we felt the HSA would be more reliable as the HEA is sensitive to adduction or abduction of the femur.

The present study is based on a total population of children with GMFCS III to V in a defined area, which is a major strength and minimises the risk of selection bias.

When applying the CPUP hip score in clinical practice, a decision has to be made on which level merits intervention, that is, a limit for what constitutes a high-risk individual. If a lower and more liberal limit is chosen, a greater number of individuals will be predicted to develop hip displacement, which increases the sensitivity while decreasing the specificity. If a higher and more conservative limit is chosen, fewer individuals will be predicted to develop hip displacement and...
the sensitivity will decrease while the specificity will increase. This relationship between the risk limits and their respective sensitivity and specificity are shown in Figure 2.

It would be advisable to base the decision of risk limit on the nature of the treatment option being considered. For example, when considering an extensive treatment, one should limit the number of children inaccurately predicted to develop hip displacement. Consequently, one then chooses a high predicted risk of say 0.80, which, as seen in Figure 2, yields a sensitivity of 0.25 and a specificity of 0.95. This means that 25% of those who reach MP > 40% within five years are correctly predicted to do so, while 5% of those that do not are incorrectly predicted to develop MP > 40%.

With less extensive treatment options it may be important to capture as many of those that develop MP > 40% as possible. A broader risk limit of say 0.5, may then be used, resulting in correct predictions of hip displacement in 65% of the affected hips and incorrect predictions in 14%. In this way a suitable limit of predicted risk might be chosen according to the treatment option under consideration.

Operative treatment options of hip displacement are adductor-psoas release and varus-derotation osteotomy of the proximal femur. Acetabular dysplasia usually occurs at a later stage and might require a pelvic osteotomy. In a 20-year follow-up of the CPUP hip surveillance programme in Sweden, 31 of 91 children with hip displacement were successfully managed with just an adductor psoas-release. The remaining 60 children were treated with femoral osteotomy - in two cases in combination with pelvic osteotomy.9 The early identification of hips at risk of dislocation is crucial in hip surveillance. The CPUP hip score will improve the prediction of hips at risk, and provide the opportunity to treat more children with less extensive surgery.

In conclusion, children with CP at GMFCS III to V are at a high risk of hip displacement. We have developed the CPUP hip score as a risk score to determine which individuals are at high- or low-risk for hip displacement (MP > 40%). The predictive ability to differentiate between high- and low-risk individuals is high (AUC = 0.87) and the CPUP hip score may be a valuable clinical tool in decision making for future follow-up and treatment.

Author contributions:
M. Hermanson: collected the data, wrote the first draft, designed the study, revised the manuscript, and approved the final draft.
G. Hägglund: designed the study, revised the manuscript, and approved the final draft.
J. Riad: designed the study, revised the manuscript, and approved the final draft.
E. Rodby-Bousquet: designed the study, revised the manuscript, and approved the final draft.
P. Wagner: performed the statistical analyses, designed the study, revised the manuscript, and approved the final draft.

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