Predictors of Hip Dysplasia at 4 Years in Children with Perinatal Risk Factors

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**Background:** While perinatal risk factors are widely used to help identify those at risk for developmental dysplasia of the hip (DDH) within the first 6 to 8 weeks of life, limited data exist about their association with radiographic evidence of dysplasia in childhood. The purpose of this study was to determine which perinatal risk factors are associated with acetabular dysplasia in children who are ≥2 years of age.

**Methods:** Pelvic radiographs were made in 1,053 children (mean age, 4.4 years [range, 2 to 7 years]) who had been assessed prospectively as having at least 1 of 9 widely accepted perinatal risk factors for DDH. Two radiologists who were blinded to patient risk factors, history, and age determined the acetabular index (AI). The primary outcome was defined as an AI >2 standard deviations from the Tönnis reference values (“severe” dysplasia). The secondary outcome was an AI of >20° at ≥2 years of age. The association between risk factors and outcomes was assessed using logistic regression. The effect of treatment in infancy was adjusted for in 37 hips.

**Results:** Twenty-seven participants (3%) showed “severe” hip dysplasia; 3 of these had received treatment for DDH in infancy. Girls were more likely to experience this outcome (odds ratio [OR] = 2.59; 95% confidence interval [CI] = 1.04 to 6.46; p = 0.04); no other examined risk factors were significant. The secondary outcome appeared in 146 participants (14%), 12 of whom had received treatment in infancy. We observed the following predictors for this outcome: female sex (OR = 1.77; 95% CI = 1.21 to 2.59; p = 0.003), breech presentation (OR = 1.74; 95% CI = 1.08 to 2.79; p = 0.02), and being a firstborn child, which had a protective effect (OR = 0.67; 95% CI = 0.46 to 0.96; p = 0.03).

**Conclusions:** We identified a substantial number of cases that will require at least radiographic surveillance for mild and severe hip dysplasia; 92% had no prior diagnosis of DDH. Those who had been born breech were affected by this outcome even if ultrasonography of the hip had been normal at 6 to 8 weeks, suggesting a benefit from additional radiographic testing.

**Level of Evidence:** Prognostic **Level III.** See Instructions for Authors for a complete description of levels of evidence.

Perinatal risk factors are widely used to help identify those at risk for developmental dysplasia of the hip (DDH) within the first 6 to 8 weeks of life, but little is known about their association with radiographic evidence of dysplasia in childhood. A meta-analysis showed that the mean follow-up of infants with perinatal risk factors was 6 months, but this is probably insufficient time to make robust inferences about radiographic evidence of dysplasia.

While at-risk infants with normal ultrasound results and clinical screening will not routinely receive hip follow-up, some of these infants may develop radiographic evidence of hip dysplasia later in childhood. A longitudinal study of skeletally mature patients showed no differences in radiographic evidence of hip dysplasia between those who had perinatal risk factors and those who did not. Recent studies have observed that infants with breech presentation may develop radiographic evidence of hip dysplasia at an age ranging from 4 to 13 months, even when there have been normal clinical and ultrasound results at 6 to 8 weeks. Other studies have concluded that radiographic monitoring is unnecessary if the 6...
to 8-week ultrasound results are normal in infants with a perinatal risk factor or with a family history of DDH. The data on perinatal risk factors and radiographic evidence of hip dysplasia remain conflicting. Most of the literature is based on risk factors that have been ascertained retrospectively, and other deficiencies include knowledge of outcomes, relatively small sample sizes, and a lack of multivariate analysis, with the potential that the true effect of risk factors was overestimated. We sought to prospectively assess perinatal risk factors in consecutive newborns, who were followed by radiographs at a minimum age of 2 years. We aimed to determine how often radiographic evidence of acetabular dysplasia occurs in such at-risk children, and how perinatal risk factors, patient characteristics, and radiographic evidence of dysplasia are associated.

Materials and Methods

The institutional review board approved this case-control study. Informed consent was obtained for all study participants. Children who were eligible for this study were those from a previously assembled cohort of newborns (2010 to 2013) who had been examined at a median age of 1 day (interquartile range [IQR], 0 to 1 day) for the presence of the following perinatal risk factors for DDH: family history of DDH in a first-degree relative, breech presentation (frank, incomplete, or complete), oligohydramnios (ultrasound-based diagnosis at 18 to 20 weeks gestation with an amniotic fluid index of ≤5), torticollis, and foot deformities (i.e., metatarsus adductus, calcaneovalgus followed at least once with a physiotherapist to ensure improvement, or structural clubfeet rotated 15°). Positive Ortolani or Barlow signs, asymmetrical hip abduction of ≥20°, and leg-length discrepancies were recorded. Senior residents, overseen by attending neonatologists and pediatric orthopaedic surgeons, undertook the examinations. Foot deformities were confirmed by 1 physiotherapist with respect to severity as well as the need for ongoing clinical review. Assessment of 13,210 consecutive newborns identified 2,271 newborns with ≥1 risk factor. Of these, 2,191 (96%) were recruited. Birth weight, parity, twin pregnancy, and mode of delivery were recorded. All of the infants underwent standardized hip ultrasonography at a mean age of 8 weeks, which was performed by a dedicated sonography team; splinting according to standardized diagnostic criteria was required in 77 infants.

We invited children in this cohort to attend a study appointment in a dedicated nurse-run research clinic that was held from 2015 to 2016, with oversight by the senior author (A.R.) and the senior radiologist (M.A.H.-C.). Parents/caregivers were contacted up to 4 times, initially with a letter containing an information leaflet, followed by up to 3 subsequent telephone calls that were made on evenings and weekends. We contacted the family physicians of nonresponders. Database searches of our hospital detected no additional cases of late-presenting DDH or surgery among nonresponders.

At study appointments, the research nurse asked participants and their parents or caregivers if any problems existed with the child’s hips. A supine anteroposterior pelvic radiograph that was centered on the hips with the feet internally rotated 15° was made. A digital imaging system (FCR XG 5000; Fujiﬁlm) was used with age-dependent exposure parameters (60 to 80 kV, 4 to 40 mAs), with a focus-to-film distance of 150 cm. We ensured optimal image quality, including adequate pelvic rotation. As in other studies, the Hilgenreiner acetabular index (AI) was used as a measure of acetabular dysplasia. This index is valid and reliable, and its cutoff values have been used previously to determine cases in DDH research.

The primary outcome was defined as an AI >2 standard deviations (SDs) above age and sex-based reference values, an accepted measure of dysplasia. The secondary outcome included the presence, in all patients who were >2 years of age, of an AI of >20° in at least 1 hip. This definition is in keeping with the Tönnis definition of “light” dysplasia (AI between 1 and 2 SDs above normal values). Tönnis emphasized the importance of identifying such hips because 20% of these hips deteriorate with age.

In our study, 2 musculoskeletal radiologists who were blinded to patient risk factors, history, and age measured the radiographs electronically (Centricity; GE Medical Systems). We held training sessions with a pediatric orthopaedic surgeon (A.R.) and the radiologists (M.A.H.-C. and T.H.) to ensure consistent measurement methods. We practiced measurements on a set of representative radiographs and agreed-upon landmarks, measuring the AI in consensus. The radiologists subsequently reviewed a random set of 41 radiographs, and their interrater reliability was excellent (intraclass correlation coefficients were 0.90 to 0.96 for the right and left hips, respectively). We also derived limits-of-agreement plots to compare the radiologists’ ratings of the AI; they measured all outliers in consensus to improve measurement consistency. With interrater reliability established, 1 radiologist (T.H.) evaluated all of the radiographs in

![Flow diagram demonstrating sample selection.](image-url)
the study, having first confirmed that his intrarater reliability was excellent (intraclass correlation coefficient, 0.94).

**Statistical Analysis**

In estimating the sample size, we considered that 10 cases of DDH per examined regression coefficient were required\(^23\) to estimate coefficients with adequate precision. When the data did not satisfy this rule, penalized logistic regression\(^24\) was used to avoid overfitting. In univariate logistic regression models for the primary and secondary outcomes, we determined an odds ratio (OR) as a measure of association with the following candidate predictors: female sex, family history of a first-degree relative affected with DDH, firstborn child, twin pregnancy, birth weight, breech presentation, mode of delivery, presence of foot deformity, and abnormal hip examination. Other risk factors occurred too infrequently to allow meaningful inclusion in the analysis. Variables with a p value of <0.5 were entered into a multivariate logistic regression model for the primary outcome. We adjusted all of the models based on the fact that 37 patients were treated for DDH. In secondary analyses, we used mixed effects models with hips as the unit nested within the patient. Because the results remained unchanged, the results from the fixed effects models were reported. The amount of missing data was small and it included several variables: parity in 31 instances, mode of delivery in 17 instances, and twin pregnancy in 17 instances. Thus, we reported all of the regression coefficients with no imputations. All of the hypothesis testing was 2-sided. Analyses were performed with STATA statistical software (version 11; StataCorp).

**Results**

Of the 2,191 subjects who were invited to participate, 1,053 (48%) attended the appointments and were included in the study (Fig. 1). No participant reported any hip-related
TABLE II: Association of Perinatal Risk Factors and Radiographic Evidence of Acetabular Dysplasia at a Mean Age of 4.4 Years*

| Risk Factor                      | Odds Ratio (95% Confidence Interval) | P Value |
|----------------------------------|--------------------------------------|---------|
| Univariate analyses              |                                      |         |
| Female sex                       | 3.26 (1.31-8.15)                     | 0.01    |
| First-born child                 | 0.88 (0.41-1.89)                     | 0.74    |
| Twin pregnancy                   | 0.48 (0.11-2.05)                     | 0.32    |
| Vaginal delivery                 | 1.18 (0.55-2.54)                     | 0.67    |
| First-degree family history      | 1.90 (0.64-5.62)                     | 0.25    |
| Breech presentation              | 1.07 (0.48-2.42)                     | 0.86    |
| Abnormal hip examination         | 2.60 (0.76-8.96)                     | 0.13    |
| Birth weight, per kg             | 0.74 (0.38-1.42)                     | 0.36    |
| Multivariate analysis†           |                                      |         |
| Female sex                       | 2.59 (1.04-6.46)                     | 0.04    |
| Twin pregnancy                   | 0.49 (0.11-2.21)                     | 0.35    |
| First-degree family history      | 1.53 (0.32-7.43)                     | 0.59    |
| Abnormal hip examination         | 1.00 (0.97-1.03)                     | 1       |
| Birth weight in kg               | 0.66 (0.30-1.43)                     | 0.29    |
| Treatment                        | 1.09 (0.11-10.65)                    | 0.94    |

*Acetabular index > 2 standard deviations above normative values. Among 27 patients with this outcome, 3 (11%) had been treated in early infancy. †Adjusted for any perinatal risk factors associated (Table II).

Discussion

The current study supports a recent observation that those who are born breech can develop radiographic evidence of hip dysplasia even if postnatal ultrasonography and clinical examinations are normal at 6 to 8 weeks. In our study, breech presentations showed a nearly twofold increased odds for an AI of >20° at ≥3 years of age. Following 131 breech presentations, Imrie et al. reported radiographic evidence of hip dysplasia warranting treatment at 4 to 6 months in 29% of cases. Other studies investigating breech presentations have reported 10% to 20% rates of radiographic evidence of hip dysplasia at 6 months of age, and 7% at 13 months. This poses the question as to whether those who are born breech should always receive radiographic follow-up and, if so, at what age? Perhaps disease modulation in this group of newborns is such that ultrasonography cannot provide accurate identification of dysplasia. Our study suggests that a radiograph at the age of 4 to 5 years should be considered to identify cases with clinically relevant acetabular dysplasia. Because of the study design, we cannot comment as to whether there is a benefit to having earlier radiographs; however, it should be noted that many providers would prefer to identify acetabular dysplasia at an earlier age in order to allow bracing treatment and/or early surgical intervention in the form of an infantile pelvic osteotomy.

Another noteworthy finding of this study is the frequency (13.9%) with which abnormal hips (i.e., mild or light symptoms. The mean age at the study visit (and SD) was 4.4 ± 0.78 years (range, 2.0 to 6.6 years), and 52.4% of the participants were girls. Patients who were lost to follow-up did not differ in terms of the distribution of perinatal risk factors (Table I). Of those included, 37 had been treated for DDH in the postnatal period, predominantly with a harness (see Appendix 1). The distribution of perinatal risk factors was similar in those who were treated and in those who were not treated (p > 0.05) (see Appendix 2).

Severe dysplasia was found in 27 participants (2.6%) with a mean age of 4.1 ± 0.6 years (range, 3.2 to 5.4 years). Of those, 3 (11.1%) had received treatment for DDH in infancy. While girls were more likely to show this outcome (OR = 2.59; 95% confidence interval [CI] = 1.04 to 6.46; p = 0.04), no other perinatal risk factors were associated (Table II).

An AI of >20° occurred in 146 participants (13.9%) with a mean age of 4.2 ± 0.6 years (range, 3.0 to 6.0 years). Of these, 12 (8.2%) had been treated in infancy. The median AI in these patients was 22° (IQR, 21° to 23°). We observed the following predictors for this outcome: female sex (OR = 1.77; 95% CI = 1.21 to 2.59; p = 0.003), breech presentation (OR = 1.74; 95% CI = 1.08 to 2.79; p = 0.02), and being a firstborn child, which had a protective effect (OR = 0.67; 95% CI = 0.46 to 0.96; p = 0.03) (Table III).

Bilateral involvement was present in 1 of 27 children with severe acetabular dysplasia and in 32 of the additional 119 children with the secondary outcome (“light” dysplasia).

TABLE III: Association of Perinatal Risk Factors and Acetabular Index of >20° at ≥3 Years of Age*

| Risk Factor                      | Odds Ratio (95% Confidence Interval) | P Value |
|----------------------------------|--------------------------------------|---------|
| Univariate analyses              |                                      |         |
| Female sex                       | 1.90 (1.31-2.74)                     | 0.001   |
| First-born child                 | 0.65 (0.46-0.93)                     | 0.02    |
| Twin pregnancy                   | 1.18 (0.68-1.83)                     | 0.67    |
| Vaginal delivery                 | 1.27 (0.90-1.81)                     | 0.18    |
| First-degree family history      | 1.05 (0.57-1.95)                     | 0.87    |
| Breech presentation              | 1.30 (0.90-1.88)                     | 0.15    |
| Abnormal hip examination         | 2.30 (1.19-4.43)                     | 0.01    |
| Foot deformity                   | 0.89 (0.31-2.56)                     | 0.82    |
| Birth weight in kg               | 0.84 (0.63-1.14)                     | 0.26    |
| Multivariate analysis†           |                                      |         |
| Female sex                       | 1.77 (1.21-2.59)                     | 0.003   |
| First-born child                 | 0.67 (0.46-0.96)                     | 0.03    |
| First-degree family history      | 1.31 (0.81-2.13)                     | 0.26    |
| Breech presentation              | 1.74 (1.08-2.79)                     | 0.02    |
| Abnormal hip examination         | 1.38 (0.36-2.97)                     | 0.94    |
| Birth weight in kg               | 0.85 (0.61-1.17)                     | 0.31    |
| Treatment                        | 1.95 (0.65-5.89)                     | 0.24    |

*Among 146 patients with this outcome, 12 (8%) had been treated in early infancy. †Adjusted for any treatment received in infancy.
Predictors of Hip Dysplasia at 4 Years in Children with Perinatal Risk Factors

Dysplasia (or hip dysplasia) were encountered in this sample of 1,053 children. The majority (92%) had no prior diagnosis of DDH; all had undergone ultrasonography at 6 to 8 weeks. Even if study participants with a prior DDH diagnosis had been excluded from the analysis, the proportion of hips with dysplasia would have remained at 12.8% in this sample. The mean AI in abnormal hips was 22° (secondary outcome), with some hips showing indices as high as 30°. This is similar to the study by Brusalis et al., who reported a mean AI of 25° at a mean age of 6 months. In line with other studies⁹,20,26, we believe that it is important to detect such hips as they will benefit from radiographic surveillance or treatment. According to Tönnis³⁰, 20% of hips within this range of AI will not improve with age.

In our study, girls had 2.5 times greater odds for the primary outcome and nearly twofold greater odds for the secondary outcome. This is similar to the relative risk of 2.5 that was reported in a meta-analysis of perinatal risk factors, but in which the outcome was DDH at a mean age of 6 months¹. A health registry study reported an OR of 3.9 for girls, with the outcome measured within 1 year postpartum²⁷. Female predominance is widely known for DDH that is diagnosed postnatally (98%) and also for hip dysplasia that is diagnosed in adolescents (88%)²⁰. In comparison, 52% of the present sample were girls, as were 66% of those with the secondary outcome.

We were unable to identify any other predictors. For the perinatal period, a family history of DDH is a widely accepted risk factor; however, studies have reported no association between this variable and DDH postnatally²⁹,³⁰ and at 12 months¹. Being a firstborn showed a protective association with the secondary outcome (and had no association with the primary outcome). The proportion of firstborn children was similar among study participants (53.4%) and nonresponders (58.6%), and firstborns were not more likely to have had prior treatment for DDH. A protective effect previously has been reported for firstborns²⁹. It is important to note that parity order previously has been identified as a potential risk factor in the perinatal period, whereas our sample involved much older children.

The strengths of our study include the prospective collection of predictors in consecutive newborns without knowledge of outcomes, a comparatively long follow-up period, and the collection of outcomes with a high degree of precision. Because hip function in infants and young children is not a reliable indicator of long-term hip function⁵, we chose radiographic outcomes. We also screened each participant for the presence of any hip-related symptoms, and there were none. Use of the Tönnis classification allowed us to compare our results with the literature⁵,⁷,⁹,¹⁷,₂⁸,₃₁ and also provided a guide for clinical practice. For example, clinicians can now decide if they wish to encourage the parents of affected children to return for a radiograph at the age of 4.5 years (or an earlier age) based on perinatal risk factors.

We acknowledge the potential limitations of this study. Participation was moderate, with 48.1% of the original cohort attending the research clinic; this proportion is very similar to comparable longitudinal studies that have included perinatal risk factors for DDH²⁴. Those who were lost to follow-up did not differ in their baseline characteristics—we assume that nonparticipation occurred at random. Because none of the participants reported any hip-related symptoms and because acetabular dysplasia as observed in this study was typically clinically “silent,” there is no reason to assume that participation was biased by disease severity. The outcome numbers that we reported do not represent prevalence estimates. We employed threshold values as described by Tönnis for classifying dysplasia. Because, to our knowledge, these threshold values have never been studied prospectively to skeletal maturity, their ultimate relevance in determining lasting pathology remains somewhat uncertain—some hips could improve spontaneously. However, the cutoff values that we used have been used widely in other DDH research to define outcomes. Thus, our approach allowed for comparison with other literature. The AI is commonly used to assess acetabular development, but the variability in its measurement is of concern¹⁸. We performed several steps to mitigate this risk: we ensured adequate observer reliability and adhered to set protocols for image acquisition and evaluation. We utilized digital radiographs and measurements, which have been shown to maximize reproducibility⁷, with a reported interobserver variance of only 0° to 1°. Prior treatment for DDH had been employed in 3% of the sample. We addressed this by adjusting the analyses, and we reported the duration and method of treatment for individuals (see Appendix 1). Crudely, treated participants showed higher proportions of outcomes (8% versus 2% for the primary outcome, and 32% versus 13% for the secondary outcome). In 22 participants with prior treatment, neither outcome occurred. Finally, we were unable to subclassify the variable “breech presentation.” However, this variable’s effect was large in the multivariate analysis, which is in keeping with other studies. We were unable to examine the role of the subtypes of breech presentation; however, the definition used here is in line with most of the previous literature¹. Female sex alone was not regarded as an inclusion criterion when the inception cohort was established; thus, we were unable to estimate the true effect of female sex in the absence of other risk factors for DDH.

While the association between female sex and breech presentation with childhood hip dysplasia has been well known, our study clarifies this observation further: our results are based on predictors and outcomes that were collected with a high degree of precision, as well as adjusted risk ratios. Perinatal risk factors were of limited value for the outcomes that we studied. Given that they had been derived from newborns for use in the perinatal period, this may be of limited surprise. What our study adds is evidence that acetabular dysplasia is frequent in preschool-aged children who have an identified perinatal risk factor. It further supports recent evidence advocating radiographic monitoring of
those born in a breech presentation. The fact that acetabular dysplasia was frequently observed in this sample poses the question of whether these hips represent the very early beginnings of a distinct form of hip disease (late-onset acetabular dysplasia) that is diagnosed in adolescence because of the onset of pain, or whether these hips represent a “late” form of the dysplasia as reported to occur with breech presentations. Because of the study design, we cannot make inferences about how the dysplastic hips that were identified in this sample should be best managed; however, because residual dysplasia can produce substantial problems at skeletal maturity, in our opinion, continued radiographic monitoring of these cases is necessary. Additional research is needed to determine the importance of dysplastic hips in the long term, especially with regard to the need for osteotomy, and the benefits of hip radiography, regardless of ultrasound screening that is performed in those with breech presentations.

Appendix

Supporting material provided by the authors is posted with the online version of this article as a data supplement at jbjs.org (http://links.lww.com/JBJSOA/A254).

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