

## What Risk Factors and Characteristics Are Associated With Late-presenting Dislocations of the Hip in Infants?

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### Abstract

**Background** Most infants with developmental dysplasia of the hip (DDH) are diagnosed within the first 3 months of life. However, late-presenting DDH (defined as a diagnosis after 3 months of age) does occur and often results in more

complex treatment and increased long-term complications. Specific risk factors involved in late-presenting DDH are poorly understood, and clearly defining an associated set of factors will aid in screening, detection, and prevention of this condition.

**Questions/purposes** Using a multicenter database of patients with DDH, we sought to determine whether there were differences in (1) risk factors or (2) the nature of the dislocation (laterality and joint laxity) when comparing patients with early versus late presentation.

**Methods** A retrospective review of prospectively collected data from a multicenter database of patients with dislocated hips was conducted from 2010 to 2014. Baseline demographics for fetal presentation (cephalic/breech), birth presentation (vaginal/cesarean), birth weight, maternal age, maternal parity, gestational age, family history, and swaddling history of patients were compared among nine different sites for patients who were enrolled at age younger than 3 months and those enrolled between 3 and 18 months of age. A total of 392 patients were enrolled at baseline between 0 and 18 months of age with at least one dislocated hip. Of that group, 259 patients were younger

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Each author certifies that his or her institution approved the human protocol for this investigation, that all investigations were conducted in conformity with ethical principles of research, and that informed consent for participation in the study was obtained.

Included patients were recruited from all listed sites as well as Centro Médico ABC Santa Fe, Mexico City, Mexico; Hospital for Sick Children, Toronto, Ontario, Canada; and Southampton General Hospital, Southampton, UK. Data analysis and review were carried out at BC Children's Hospital, Vancouver, British Columbia, Canada.

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than 3 months of age and 133 were 3 to 18 months of age. The proportion of patients with DDH who were enrolled and followed at the nine participating centers was 98%.

**Results** A univariate/multivariate analysis was performed comparing key baseline demographics between early- and late-presenting patients. After controlling for relevant confounding variables, two variables were identified as risk factors for late-presenting DDH as compared with early-presenting: cephalic presentation at birth and swaddling history. Late-presenting patients were more likely to have had a cephalic presentation than early-presenting patients (88% [117 of 133] versus 65% [169 of 259]; odds ratio [OR], 5.366; 95% confidence interval [CI], 2.44–11.78;  $p < 0.001$ ). Additionally, late-presenting patients were more likely to have had a history of swaddling (40% [53 of 133] versus 25% [64 of 259]; OR, 2.053; 95% CI, 1.22–3.45;  $p = 0.0016$ ). No difference was seen for sex ( $p = 0.63$ ), birth presentation ( $p = 0.088$ ), birth weight ( $p = 0.90$ ), maternal age ( $p = 0.39$ ), maternal parity ( $p = 0.54$ ), gestational age ( $p = 0.42$ ), or family history ( $p = 0.11$ ) between the two groups. Late presenters were more likely to present with an irreducible dislocation than early presenters (56% [82 of 147 hips] versus 19% [63 of 333 hips]; OR, 5.407; 95% CI, 3.532–8.275;  $p < 0.001$ ) and were less likely to have a bilateral dislocation (11% [14 of 133] versus 28% [73 of 259]; OR, 0.300; 95% CI, 0.162–0.555;  $p = 0.002$ ).

**Conclusions** Those presenting with DDH after 3 months of age have fewer of the traditional risk factors for DDH (such as breech birth), which may explain the reason for a missed diagnosis at a younger age. In addition, swaddling history was more common in late-presenting infants. A high index of suspicion for DDH should be maintained for all infants, not just those with traditional risk factors for DDH. Further investigation is required to determine if swaddling is a risk factor for the development of hip dislocations in older infants. More rigorous examination into traditional screening methods should also be performed to determine whether current screening is sufficient and whether late-presenting dislocations are present early and missed or whether they develop over time.

**Level of Evidence** Level III, retrospective study.

## Introduction

Developmental dysplasia of the hip (DDH) is a common disorder in otherwise healthy infants. Severity can range from mild instability to a completely dislocated hip. In Western society, hip instability can be detected in 1% to 3% of newborns with 10 of 1000 requiring treatment and approximately one to two children per thousand presenting

with a complete hip dislocation at birth [4, 6, 13, 28]. If left untreated, mild dysplasia can lead to early hip arthritis [38], and children left with an untreated dislocated hip can be at risk of lifelong hip disabilities [12]. In some children, hip dislocations may not be detected until a limp is evident when a child has reached walking age. Nonsurgical management of hip dislocation is more likely to fail in the older infant and therefore often requires more intrusive treatment like prolonged immobilization or surgery in comparison to treatment of those who have a confirmed DDH diagnosis at a younger age [2, 14, 18, 20, 39].

Screening procedures are the cornerstone for efficiently and effectively detecting and diagnosing DDH in infants and are critical to prevention or reduction of late-presenting cases. Clinical examination is a universally standard practice for DDH screening in all newborns. Many European countries have also adopted universal ultrasound screening to confirm clinical findings [5, 16, 33, 34, 37]. In North America, however, selective ultrasound screening is used only for infants with defined risk factors such as breech presentation, family history, or a clinical history of hip instability [25, 29, 30, 34, 35]. Screening methods are controversial, and late presentations still occur with both clinical and ultrasound screening programs, although the reported incidence is variable [19, 21, 22, 31, 32]. To prevent late presentation, we must better define the factors involved to more accurately identify, and therefore monitor, patients potentially at risk. Previous retrospective studies of late-presenting diagnoses of DDH report an incidence of less than 1% to 3% out of all afflicted infant hips with dysplasia; however, most do not report specifically on frankly dislocated hips and instead include the full spectrum of hip dysplasia [3, 15, 21, 29, 41]. Frank dislocations represent the most severe form of DDH, require the most drastic treatment measures, and tend to have more long-term complications. By including the spectrum of DDH, from mild to severe dysplasia and dislocations, important risk factors for the more severe dislocation may be concealed. There are few large, multicenter studies that have prospectively followed infants and young children with dislocated hips [10, 11]. Multicenter studies enable the capture of a range of presenting populations, diagnoses, screening methods, treatment strategies, and outcomes. Diverse populations may also reveal new risk factors or delineate risk factors by diagnosis, geography, or patient demographics.

We, therefore, asked in a large cohort of patients with dislocations of the hip whether there were differences in (1) risk factors or (2) the nature of the dislocation (laterality and joint laxity) when comparing patients with early versus late presentation.

## Materials and Methods

### Study Design

A retrospective review of prospectively collected data was conducted from a multicentered study containing nine sites from North America, Europe, and Australia. Data were collected on frankly dislocated hips at rest from patients enrolled in the study at contributing centers from 2010 to 2014. Collected data were entered and managed using the Vanderbilt Research Electronic Database Capture (RED-Cap) and then reviewed to examine potential risk factors for late-presenting DDH in the study cohort.

### Inclusion and Exclusion Criteria

For the purposes of this study, only fully dislocated hips were considered for analysis, and late diagnosis was defined as a diagnosis of dislocation made after 3 months of age. Infants 18 months of age or younger at baseline who had at least one frankly dislocated hip were included in this analysis. Included patients were divided into two age groups: younger than 3 months and those 3 months to 18 months of age. All included hips were verified as dislocated by ultrasound (percent coverage of the femoral head less than 35%) or radiography (IHDI grade of 3 or above [26]) before treatment.

Patients with an unconfirmed diagnosis of DDH as well as existing congenital hip abnormalities and developmental syndromes were excluded from the study. Other milder forms of DDH such as subluxatable or dysplastic hips as well as all infants who had received previous treatment for DDH were also excluded.

### Study Population

A total of 392 patients were enrolled at baseline between 0 and 18 months of age. All patients presented with at least one frankly dislocated hip. At baseline, patients were divided into two groups according to age: younger than 3 months of age and those between 3 months and 18 months of age inclusive. Of 392 patients, 259 (66%) were younger than 3 months of age (333 dislocated hips) and 133 (34%) were 3 to 18 months of age (147 dislocated hips). Within the late-presenting group, there were 48 patients between 3 and 6 months of age, 47 between 6 and 12 months, and 38 between 12 and 18 months with 51 of 133 being of potential walking age. The overall percentage of female patients (81% [319 of 392]) was much larger than that of males (19% [73 of 392]).

### Variables and Measures Assessed

The database was queried (EKS) for baseline demographics previously explored for an association with DDH: sex [1], fetal presentation (cephalic/breech) [3, 29], birth presentation (vaginal/cesarean) [3, 29], birth weight [3], family history [1], parent-reported swaddling history [24], maternal age [3], maternal parity [3], and gestational age [3]. These variables were determined for patients who were enrolled at age younger than 3 months and those enrolled between 3 and 18 months of age. The nature of the diagnosed dislocation (reducible or irreducible) and laterality were also noted. These variables were determined by consultation with patient charts, clinic nurses, and surgeons and analyzed to determine if there were any predictive factors for patients that presented with DDH at or after 3 months of age. Type and frequency of swaddling were not available for this analysis.

### Statistical Analysis

An exploratory univariate analysis was performed (EKS) on extracted demographic information to determine if there was an association between baseline demographics and the age of the participant at baseline. Variables considered were sex, fetal presentation, birth presentation, family history, swaddling, maternal parity, maternal age, gestational age, and birth weight. This was followed by a more definitive logistic regression analysis (in consultation with a statistician) using two models: one with all predictors entered and one with backward stepwise regression resulting in a final model with two significant predictors. The unit of analysis was the patient.

## Results

### Differences in Risk Factors Between Early- and Late-presenting Dislocations

Following logistic regression analysis, two variables were identified as risk factors for late-presenting dislocations in comparison to early-presenting dislocations: fetal presentation and swaddling.

The late-presenting group was more likely to have had a cephalic presentation at birth than the early-presenting group (88% [117 of 133] versus 65% [169 of 259]; odds ratio [OR], 5.366; 95% confidence interval [CI], 2.44–11.78;  $p < 0.001$ ; Table 1). This finding is consistent with the fact that infants with cephalic presentation are not monitored as closely and therefore may be missed early [29].

**Table 1.** Categorical and measurement variables for early-presenting and late-presenting patients enrolled at baseline with at least one dislocated hip

| Presentation at diagnosis              | Early-presenting < 3 months<br>(%; numerator/denominator) | Late-presenting 3–18 months<br>(%; numerator/denominator) | OR<br>[95% CI]              | p value |
|--|---|---|-----------------------------|---------|
| <b>Categorical variable comparison</b> |   |   |                             |         |
| Sex (female)                           | 81% (209/259)   | 83% (110/133)   | 1.1 [0.7–2.0]               | 0.63    |
| Sex (male)                             | 19% (50/259)  | 17% (23/133)  |                             |         |
| Fetal presentation (cephalic)          | 65% (169/259)   | 88% (117/133)   | 5.4 [2.4–11.8]              | <0.001  |
| Fetal presentation (breech)            | 32%* (82/259)   | 9%* (12/133)  |                             |         |
| Birth presentation (vaginal)           | 49% (127/259)   | 58% (77/133)  | 0.7 [0.5–1.1]               | 0.095   |
| Birth presentation (cesarean)          | 51%* (131/259)  | 41%* (55/133)   |                             |         |
| Swaddling history (yes)                | 25% (64/259)  | 40% (53/133)  | 2.1 [1.2–3.5]               | 0.002   |
| Swaddling history (no)                 | 69%* (180/259)  | 54%* (72/133)   |                             |         |
| Family history (yes)                   | 34% (87/259)  | 26% (34/133)  | 0.7 [0.4–1.1]               | 0.11    |
| Family history (no)                    | 63%* (164/259)  | 71%* (94/133)   |                             |         |
| Presentation at diagnosis              | Early-presenting <3<br>months Mean (SD)                   | Late-presenting 3–18<br>months Mean (SD)                  | Mean difference<br>[95% CI] | p value |
| <b>Measurement variable comparison</b> |   |   |                             |         |
| Birth weight (kg)                      | 3.3 (0.7)   | 3.3 (0.7)   | 0 [–0.1 to 0.2]             | 0.88    |
| Maternal age (average years)           | 30.2 (5.9)  | 29.6 (5.6)  | –0.6 [–1.8 to 0.7]          | 0.39    |
| Maternal parity (average)              | 1.4 (0.9)   | 1.5 (1.0)   | 0.06 [–0.1 to 0.2]          | 0.13    |
| Gestational age (average weeks)        | 39.3 (1.5)  | 39.1 (2.2)  | –0.2 [–0.6 to 0.2]          | 0.42    |

\* Paired proportions that do not add to 100% are subject to having “unknown” values; OR = odds ratio; CI = confidence interval.

There was also a distinct difference seen in the prevalence of swaddling between the early- and late-presenting patients. The late-presenting group was more likely to have a history of swaddling than the early-presenting group (40% [53 of 133] versus 25% [64 of 259]; OR, 2.053; 95% CI, 1.22–3.45;  $p = 0.0016$ ). Other explored variables—sex, birth presentation, birth weight, family history, maternal age, maternal parity, and gestational age—were not found to be risk factors for late-presenting dislocations (Table 1).

#### Differences in the Nature of the Dislocation at Diagnosis Between Early-presenting and Late-presenting Patients

In our study, all patients presented either with one (unilateral) or both (bilateral) hips dislocated. Overall, 26% (101 of 392) of patients had a unilateral right dislocation, 52% (204 of 392) had a unilateral left dislocation, and 22% (87 of 392) had bilateral dislocations. The late-presenting group was less likely than the early-presenting group to have bilateral dislocations (11% [14 of 133] versus 28% [73 of 259]; OR, 0.300; 95% CI, 0.162–0.555;  $p = 0.002$ ; Table 2). Irrespective of unilateral or bilateral status, no difference was seen in the overall incidence of left-affected and right-affected hips between the early- and late-presenting patients.

Dislocated hips can be reducible or irreducible, indicating whether the femoral head can be reduced back into the acetabulum. The late-presenting group was more likely than the early-presenting group to have an irreducible dislocation (56% [82 of 147 hips] versus 19% [63 of 333 hips]; OR, 5.407; 95% CI, 3.532–8.275;  $p < 0.001$ ).

#### Discussion

Current research has focused on identifying potential risk factors for DDH at birth to determine which infants need more thorough clinical and radiographic screening [1]. Careful screening and early detection can prevent the potential complications and more difficult treatment often required when DDH is diagnosed in the later stages of infancy [2, 14, 18, 20, 39]. According to the newly published guidelines on DDH from the American Academy of Orthopaedic Surgeons, breech fetal presentation, family history, and a history of clinical hip instability are significant risk factors for DDH [1]. Insufficient evidence exists to support the inclusion of other previously described risk factors such as foot abnormalities [7], oligohydramnios [8], and torticollis [9]. What remains to be determined is whether a subset of these or other risk factors are specific indicators for late-presenting DDH. Identification of such factors would allow for more careful and selective

**Table 2.** Ultrasonographically or radiographically confirmed diagnosis of laterality and laxity at baseline

| Presentation at diagnosis   | Early-presenting < 3 months<br>(%; numerator/denominator) | Late-presenting 3–18 months<br>(%; numerator/denominator) | OR [95% CI]      | p value |
|---|---|---|------------------|---------|
| Laterality: OR of bilateral dislocation in late- versus early-presenting patients |   |   |                  |         |
| Unilateral  | 72% (186/259)   | 89% (119/133)   | 0.30 [0.16–0.56] | 0.002   |
| Bilateral   | 28% (73/259)  | 11% (14/133)  |                  |         |
| Laxity: OR of irreducible dislocation in late- versus early-presenting patients   |   |   |                  |         |
| Reducible   | 81% (270/333)   | 44% (65/147)  | 5.41 [3.53–8.28] | <0.001  |
| Irreducible   | 19% (63/333)  | 56% (82/147)  |                  |         |

OR = odds ratio; CI = confidence interval.

screening of infants at particular risk to prevent late detection and diagnosis. This study sought to identify risk factors for and differences in the nature of late-presenting dislocations as compared with early-presenting dislocations. We identified cephalic presentation and history of swaddling to be risk factors specifically for late-presenting dislocations. Additionally, late presenters were less likely to have a bilateral dislocation but more likely to have an irreducible dislocation than early presenters in this patient population.

There are limitations to this study. First, a 34% incidence of late presentation within this patient cohort represents an alarmingly higher rate in comparison to previous studies [3, 21, 29]. However, direct comparison may be difficult as a result of differences in late-presenting definition and the degree of dysplasia considered. Additionally, although all sites within this study are associated with birthing hospitals, they also act as tertiary referral centers for infants born at outside sites. These external referrals may be contributing to the high incidence of late-presenting patients in this study cohort. Despite this, the study was focused on identifying risk factors and characteristics of late-presenting dislocations rather than quantifying incidence of late presenters within the entire population. Furthermore, with a 98% recruitment rate, we were capturing the vast majority of eligible patients presenting to study centers. Second, we are unable to comment on certain risk factors identified by previous late-presenting publications because we do not collect data points on place of birth (rural/urban) and length of hospital stay [29]. Additional risk factors to explore in future studies also include maternal socioeconomic status, country of birth, and ethnicity. The lack of these data does not, however, impact the legitimacy of the risk factors and dislocation characteristics we did identify, because these are independent demographics. Third, although we identified swaddling as a potential risk factor specific to late-presenting dislocations, there are important limitations to consider before drawing potential causative conclusions from this finding. The swaddling incidence was determined

by the parents reporting whether their child had been swaddled before diagnosis and the type, frequency, and duration of swaddling were not available for analysis. There are different methods used to swaddle, some of which constrict hip movement and others that allow for free movement. Consequently, the method of swaddling may have a large impact on the ultimate hip outcome. Frequency and length of swaddling are also important to be taken into account. Infants in the older group have had more opportunity to be swaddled, which may suggest a causative relationship with DDH when none actually exists. However, the evident discrepancy in swaddling prevalence between the late- and early-presenting groups warrants further investigation. Finally, this study included only frankly dislocated hips rather than the entirety of the DDH spectrum. This may have influenced the identification of risk factors, because the more mild forms of DDH such as subluxatable or dysplastic hips may manifest in patients with different demographic or characteristic risk factors. However, we would argue that, because dislocations represent the most severe form of DDH, identifying risk factors specific to this patient cohort is of great importance. Future studies will aim to compare risk factors between more mild forms of DDH such as dysplasia or subluxation with the frank dislocation cohort.

In the current study, we identified cephalic presentation at birth and history of swaddling as risk factors for late-presenting hip dislocations in infants. Little is known about the epidemiology of late-presenting DDH; however, it is apparent that DDH is more difficult to treat in an older infant or child [2, 14, 18–20, 39]. The long-term impact of late-diagnosed hip dislocations in infancy is not yet understood; therefore, a more complete understanding of the risk factors involved in these specific presentations would aid in earlier detection and appropriate treatment and management. Few studies have looked at late-presenting clinical indicators and outcomes during infancy. Those that have are retrospective in nature and involve only a small number of children ( $n = 26, 27,$  and  $67$ ) [3, 21, 29]. Within these existing studies, only one [3] focused

solely on frankly dislocated hips, whereas the others instead have included the entire spectrum of hip dysplasia ranging from complete dislocations to a dysplastic hip. In contrast, our study contained a larger late-presenting patient cohort with frankly dislocated hips, was conducted from a prospectively maintained database, and represented patient populations from nine major centers across three continents. The previous studies correlated late-presenting DDH with female sex, rural birth setting, lower birth weight, and a shorter hospital stay [29]. Being born breech or by cesarean was found to be protective of late-presenting DDH, suggesting that these infants' hips were more carefully monitored during early development [29]. Consistent with this last finding, we identified cephalic presentation as a risk factor in the late-presenting group, suggesting that not only is this a risk factor for milder forms of late-presenting dysplasia, but also for the more severe dislocations. In contrast to these earlier findings, however, this study did not determine sex, birth weight, or birth method to be risk factors in late-presenting patients. We also identified a history of swaddling to be more prevalent in the late-presenting group. Swaddling has been proposed as a potential causative factor in hip dysplasia because it can prevent proper hip abduction [24, 40]; however, this had not been definitively shown. The correlation of late-presenting dislocations to swaddling warrants further investigation, including more rigorous delineation of type and frequency in a prospective manner to more completely understand the degree of risk associated with this factor.

No difference was found between the two groups for maternal factors such as maternal age at birth, maternal parity, and gestational age. Prior studies investigating these maternal factors also did not find a correlation between them and those that present with DDH after 3 months of age [3, 27]. These findings suggest that not only do maternal factors not impact mild forms of late-presenting dysplasia, but also the more severe form of dislocations.

We also examined differences in the nature of dislocation at initial presentation and found late-presenting patients were less likely to have bilateral dislocations at diagnosis and more likely to have an irreducible dislocation. No difference was seen in the ratio of affected left versus right hips between the two groups. Our finding that late-presenting patients were less likely to have bilateral dislocation is somewhat surprising, because it has been suggested that symmetrical dislocations may be more difficult to detect. Indeed, Haasbeek et al. found older patients were more likely to be bilateral than younger [15]. This discrepancy may have arisen as a result of the Haasbeek study defining late presenting as 20 months of age or older. Finally, the finding that irreducible dislocations are more common in late-presenting patients is a result of the

potential for increased development of muscles and tendons surrounding the hip. Additionally, an irreducible hip can be more difficult to detect by conventional physical examination methods. Irreducible hips are thought to be more difficult to treat by conservative measures [2, 17, 23, 36], emphasizing the importance of early detection and management to avoid unnecessary complications. To our knowledge, no studies have reported on differences in dislocation reducibility between patients with early- and late-presenting DDH and, although not a predictive factor, provides further justification for identifying predictive factors to enable earlier diagnosis for this patient cohort.

Overall, we identified cephalic presentation and history of swaddling as risk factors for late-presenting dislocations of the hip in infants. Other factors such as birth weight and birth method were not found to differ between the two groups. Additionally, at diagnosis, these late-presenting patients were less likely than early presenters to have bilateral dislocation and more likely to have an irreducible dislocation. This is important because improving early detection methods for these patients will decrease complications of treatment and potential long-term impact on patient health and quality of life. The findings suggest that clinicians should monitor patients who have a history of swaddling and ask more information of the families about type and frequency of swaddling. Given that cephalic presentation was the most prominent risk factor, an outstanding question that will be crucial to answer is whether these late-presenting hips are simply going undetected by current screening methods because they are not being closely monitored or whether instability is being developed over time in a previously normal hip. The study also suggests that bilateral dislocation is less frequent than initially thought in late-presenting patients, and with increased irreducible dislocations, we can expect many patients to be Ortolani negative on clinical examination. Future studies might further explore a correlative or causative link between swaddling and late presentation by prospectively collecting data on swaddling method, frequency, and duration. Additionally, studies may endeavor to define other potential risk factors involving patient demographics to improve our current screening and detection methods. If we can accrue a number of risk factors associated with late-presenting dislocations, while also armed with the knowledge of how a case typically presents (ie, unilateral and irreducible), we can develop a better screening algorithm to reduce the number of late-presenting cases.

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