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Research Article

Incidence and Screening Practices of Developmental Hip Dysplasia in Preterm Breech Infants

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Abstract

Objective: To evaluate the incidence of Developmental Hip Dysplasia (DDH) in preterm breech infants at our institution and screening practices among providers that manage DDH in the preterm population.

Design: We reviewed charts of infants born in a single quaternary care urban medical center between 2012-2022. Infants born <37 weeks Gestation Age (GA) with breech presentation and screening ultrasonography done within the system were included. Infants with genetic syndromes were excluded. Data were extracted using Epic Slicer/Dicer and collected using RedCAP. A survey assessing provider screening practices concerning DDH in preterm infants was sent to pediatricians, neonatologists, and orthopedists.

Results: Two thousand seven hundred ninety six charts were reviewed, and 147 met the inclusion criteria. An overall incidence of DDH was found to be 6.1% (9/147), with 5.6% (1/18) in <32 weeks GA infants and 6.2% (8/129) in 32-37 weeks GA infants. Screening before 40 weeks of corrected GA was performed in 34.7% (51/147) of all infants and in 66.7% (6/9) of infants with DDH.

234 physicians responded to the survey. A large majority of each group believed this population should be screened. While most think both preterm and late preterm breech infants should be screened at 6 weeks corrected age, survey results showed discordance among current screening practices.

Conclusion: The incidence of DDH in preterm breech infants is lower than the reported incidence in the term breech population, and clinicians should consider this before ultrasound screening. Additionally, variation exists in provider practices for screening preterm breech infants suggesting a need for updated clinical guidelines.

Keywords: Neonatology; Developmental hip dysplasia; Breech; Prematurity; Outcomes

Introduction

Developmental Hip Dysplasia (DDH) is defined as the improper development of the hip joint and its two main bony components, the femoral head, which makes up "the ball" within the acetabulum, "the socket." Improper alignment within the joint results in incomplete contact between the femoral head and acetabulum, leading to a wide range of clinical impacts from minor hip instability to frank luxation. This condition was formerly known as congenital hip dysplasia. However, advances in our understanding of the etiology of hip dysplasia have changed this designation to a developmental pathology [1,2].

Hip dysplasia refers to the range of anatomical hip abnormalities that result from aberrations to this developmental process. Normal hip development begins early in the fetal period. By six weeks, areas of mesenchyme have formed an outline of the ilium, ischium, pubis, and femoral shaft [3]. By week 11, the femoral head is completely formed [4]. During subsequent fetal development, the acetabulum becomes progressively shallower. The acetabulum begins to deepen at birth, attaining a definitive shape around the end of the first decade of life [5].

Newborns are often born with physiologic laxity of the hip and immaturity of the hip joint within the first weeks or months of life. In many cases, this laxity will resolve. As many as 88% of infants born with hip instability will have a spontaneous resolution by the eighth week of life [6]. Left untreated, persistent hip instability can alter biomechanics, leading to functional disabilities, leg length discrepancies, and early osteoarthritis [7]. In young adults, DDH represents the leading cause of total hip replacement [8]. Therefore, screening and treating infants at the highest risk for DDH is critical.

The exact pathogenesis of DDH remains to be fully elucidated. Risk factors, including genetic, mechanical, and environmental considerations, likely confer risk. The most robust risk factors for DDH include female sex, being first-born, family history, tight swaddling, physical limitation in utero, postmaturity, and breech positioning [9]. Breech positioning in the third trimester is considered the most significant risk factor, with an Odds Ratio (OR) of 5.47 [10]. Breech positioning causes prolonged knee extension, resulting in sustained hamstring forces on the hip that impair proper development [11]. There is a growing body of evidence to suggest that breech positioning is a risk factor for DDH only in term infants, as breech preterm

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infants are a unique subpopulation of infants that have not sustained abnormal hip forces for a prolonged duration when compared to term infants [7,12-14]. Furthermore, the incidence of DDH in the breech term population has been estimated to be 12% to 24%, compared to 0.5% to 14% in the breech preterm population [7,13,15-17].

Official guidance on DDH screening in infants was published by the American Academy of Pediatrics in 2000 [18]. In these guidelines, it is recommended that breech infants, regardless of gestational age, be screened for DDH either by ultrasound at six weeks of age or radiograph at four months of age. These recommendations also suggest that DDH may be unrecognized in preterm infants. The discord between these guidelines and recent evidence regarding the risk factors for DDH in preterm infants may lead to discrepancies in provider practices around screening in the preterm population.

The aims of our study were thus twofold. First, we aimed to determine the incidence of DDH in preterm breech infants at our institution. Our second aim was to evaluate the screening practices among providers that manage DDH in the preterm population.

Materials and Methods

A retrospective chart review of all infants born <37 weeks Gestation Age (GA) at a single quaternary care urban medical center in the Northeastern United States between 2012 and 2022 was performed. Two initial patient lists were electronically generated using Epic's Slicer Dicer to obtain records of preterm infants with a recorded hip US at the institution. One was derived from the Neonatal Intensive Care Unit (NICU), where the standard practice is to admit newborns born less than 35 weeks Gestation Age (GA) or with a birth weight of <2 kg. The second patient list came from our well-baby nursery and was used to capture data on infants born between 35- and 37-weeks' gestation. Only preterm infants who were born having a breech presentation at birth and had ultrasound screening specifically for diagnosing DDH were included. Exclusion criteria included patients with genetic syndromes or musculoskeletal abnormalities. Infants transferred to local pediatricians for ultrasound screening who became lost to follow-up were also excluded.

Neonatal and maternal electronic medical chart records were reviewed, and data was securely collected using RedCAP. The following demographic variables of interest were collected: gender, gestational age, birth weight, age and corrected GA at the first hip ultrasound, ultrasound results, and family history. For DDH-positive patients only, the type of DDH and an indicated need for orthopedic follow-up were recorded. Information on obstetric details was collected, including maternal age, gravida, parity, mode of delivery, amniotic fluid, multiple gestations, and birth order. Study variables reflected the previously studied and widely accepted risk factors of DDH in the term breech infant population.

An anonymous online survey was created using the NYU Qualtrics system to fulfill our second objective. It was distributed to providers using national listservs, including pediatric orthopedists, general pediatricians, and neonatologists. Our survey allowed providers to identify their specialty and posed three questions regarding their clinical opinion on ultrasound screening of preterm breech infants. We aimed to investigate the consensus around DDH guidelines and ultrasound screening practices for our study population among the pediatric providers that order hip ultrasounds and those that treat infants with DDH.

Both the chart review and survey received approval from the Institutional Review Board. Survey participants provided written consent and confirmed their eligibility before submitting their responses.

Statistical analysis

The statistical significance of demographic and obstetric history data was evaluated and compared between DDH-positive and DDH-negative infants. The student's t-test and Mann-Whitney U test were used to analyze continuous variables, with medians and interquartile ranges computed to compare the characteristics of infants with and without DDH. The chi-square test was utilized for categorical variables, with each group's counts and frequencies (%) calculated. A p-value of <0.05 was considered to be statistically significant. Our statistical analysis was completed using Stata.

Results

Case study

During the study period, 2378 infants were admitted to the NICU, and 418 infants admitted to the well-baby nursery received hip US. Of these patients, 147 were included in our study for being preterm, breech-presenting, and having a hip US to diagnose DDH specifically. 9 out of 147 (6.1%) were positive for DDH. Infants in the study population were stratified into two age cohorts: very preterm (<32 weeks) and moderate to late preterm (32-37 weeks). There was no significant difference in the incidence of DDH between the very preterm (1/18 [5.6%]) and the moderate to late preterm (8/129 [6.2%])] groups.

The demographic characteristics between DDH-positive and DDH-negative infants are shown in Table 1. No significant differences were found in the categories of gender and family history, which are studied risk factors in the term breech infant population.

Maternal history and obstetric data between the DDH-positive and DDH-negative groups are outlined in Table 2. All infants positive for DDH were delivered by cesarean section. Infants positive for DDH were more likely to have abnormal amniotic fluid than infants without DDH (p=0.023). Oligohydramnios and polyhydramnios were equally present in the DDH positive group. Moreover, multiple gestations were not associated with DDH positivity.

Table 1: Demographic Information.

Variable	DDH+	DDH-	p-value
Gender			1
Male	4 (44.4%)[a]	63 (45.6%)	
Female	5 (55.6%)	75 (54.4%)	
Gestational age at birth (weeks)	35.2 [33.1, 36.2][b]	35.3 [33.5, 36.1]	0.992
Birth weight (g)	2060.7 (512.8)[c]	2238.3 (564.2)	0.359
Family history of DDH?			0.174
Yes	1 (11.1%)	2 (1.4%)	
Age at time of hip US (weeks)	8 (88.9%)	136 (98.6%)	
Age at time of hip US (weeks)	5 [2, 10][c]	6 [4, 9]	0.475
Corrected age at time of hip US (weeks)	39 [37, 43]	40 [39, 43]	0.204
Type of DDH (DDH+ only)			N/A
Unilateral	1 (11.1%)[d]		
Bilateral	8 (88.9%)		
Orthopedic follow-up required? (DDH+ only)			N/A
Yes	9 (100%)		
No	0 (0%)		

Categorical variables represented as [a,d]number (percentage); Continuous variables represented as [b]median [interquartile range] or [c]mean (standard deviation) depending on variable distribution

Table 2: Obstetric Information.

Variable	DDH+	DDH-	p-value
Maternal age (years)	36.1 (5.6)[a]	35.1 (4.7)	0.528
Maternal gravida	2 [1, 2][b]	2 [1, 3]	0.419
Maternal parity	1 [1, 1]	1 [1, 2]	0.047
Mode of delivery			1
C-section	9 (100%)[c]	127 (92%)	
Spontaneous vaginal delivery	0 (0%)	11 (8%)	
Amniotic Fluid			0.023
Normal	4 (50%)	107 (81.1%)	
Oligohydramnios	4 (50%)	11 (8.3%)	
Polyhydramnios	0 (0%)	9 (6.8%)	
Anhydramnios	0 (0%)	5 (3.8%)	
Multiple gestation			1
Yes	3 (42.9%)	63 (46%)	
No	4 (57.1%)	74 (54%)	
Birth order			0.238
A	0 (0%)	32 (52.5%)	
B	3 (100%)	29 (47.5%)	

Continuous variables represented as [a]mean (standard deviation) or [b]median [interquartile range] depending on variable distribution. Categorical variables represented as [c]number (percentage)

All nine infants positive for DDH received orthopedic follow-up and treatment at our institution. Five out of nine (55.6%) infants received hip US before 6 weeks of CGA (Table 3). Intervention history is included below (Table 3).

Study survey

Two hundred thirty four individuals responded to the survey, including 168 neonatologists, 46 pediatric orthopedists and 20 pediatricians. 90.6% of all survey respondents (212 of 234) believed that infants born breech and less than 37 weeks gestational age should be screened for DDH using ultrasound. A breakdown of responses by specialty is shown in Figure 1.

For the moderate to late preterm breech group, most survey respondents believed the appropriate screening time to be 6 weeks CA (Figure 2). The second most popular response was 6 weeks of age. Only 3.8% (9/234) of respondents believed that screening this preterm group for DDH is not necessary. For the early preterm group, most physicians believed the proper screening time to be 6 weeks corrected age (Figure 3). 20.5% (48/234) believed that it is not necessary to screen the early preterm group.

Discussion

Our findings support that the incidence of DDH in preterm breech infants is lower than the reported incidence in the term breech population. Breech infants born prematurely are not exposed to the same degree of sustained hip forces that would otherwise contribute to developing DDH in a term infant. When considering the population of infants born in the breech position, it is likely that prematurity is protective or otherwise mitigates the negative impact of

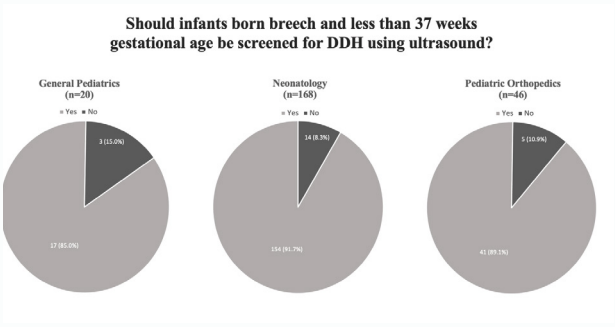

Figure 1: Survey responses "Should infants born breech and less than 37 weeks gestational age be screened for DDH using ultrasound?"

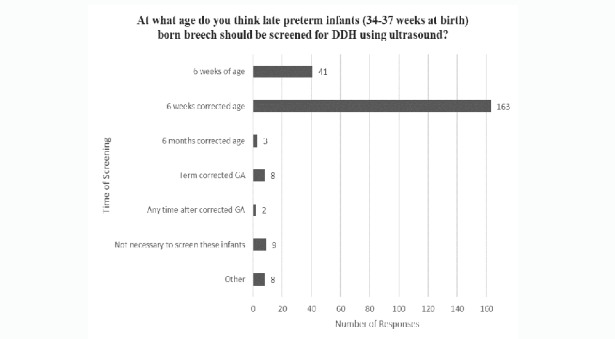

Figure 2: Survey responses "At what age do you think late preterm infants (34-37 weeks at birth) born breech should be screened for DDH using ultrasound?"

breech positioning on DDH.

The risk factors for DDH are not equivocally agreed upon. Certain risk factors, such as female sex, being first-born, family history, tight swaddling, physical limitations in utero, postmaturity, and breech positioning, are more widely accepted than others. A recent systematic review and meta-analysis of the risk factors found a strong association with breech positioning, family history, sex, and clicking hips on physical examination with DDH [19]. Interestingly, they found that multiple studies demonstrated low birth weight (<2500 gm) was associated with an OR of less than one. A second meta-analysis found that breech positioning, female sex, positive family history, and being first-born were associated with DDH [20]. More recently, investigators sought to analyze the available evidence for an association between prematurity and DDH [21]. After reviewing the available literature, they found that prematurity is not strongly associated with DDH and that early gestational age was associated with a higher incidence of mature hips than immature hips. The association between other suggested risk factors and DDH remains

Table 3: Intervention histories of DDH positive patients.

Patient	Age of Screening (weeks corrected GA)	Follow-Up Ultrasound	Age of Harness Placement (weeks corrected GA)	Failed Harness? *
1	36	No	36	No
2	37	No	37	No
3	37	No	37	Yes
4	38	No	38	No
5	39	No	39	No
6	39	Yes	45	Yes
7	43	Yes	-	-
8	44	No	44	No
9	47	No	47	No

* Infants that failed harness went on to receive surgical intervention for DDH

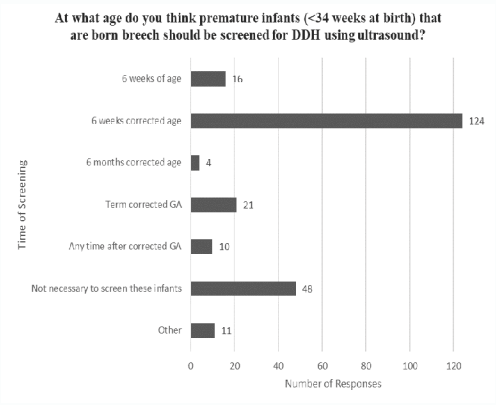

Figure 3: Survey responses "At what age do you think premature infants (<34 weeks at birth) that are born breech should be screened for DDH using ultrasound?"

unclear without overwhelming evidence or reviews. Risk factors are critical to understanding the potential pathophysiologic mechanisms by which hip dysplasia develops; they also enable clinicians to determine which infants are most likely to develop this condition and which infants are most likely to benefit from early screening and intervention.

The overall incidence of DDH in the preterm population remains controversial. An ideal incidence would reflect the population of infants with hips that, if left untreated, would develop dysplasia [22]. However, there are discrepancies in how pathologic neonatal hips are defined. For example, the reported incidence of DDH in ultrasound-screened infants ranges between 34.0 and 60.3 per 1000 [23]. This is substantially higher than the incidence in infants screened by Ortolani and Barlow physical maneuvers, which is between 1.6 to 28.5 per 1000, and considerably higher than that of persistent DDH in unscreened populations, estimated to be between 1.3 per 1000 [10,23]. Kokavek and Bailik developed a protocol to determine the 'real' incidence of DDH, defined above as the population of infants developing hip dysplasia if left untreated [24]. They found sonographic screening detected an incidence of 69.5 per 1000. However, only 21 hips remained abnormal and required treatment, indicating a true incidence of 4.8 per 1000 hips. This is aligned with the observation that a majority of neonatal hip instability is self-resolving. The overall incidence of DDH in our study was defined by ultrasonographic findings. We found that 9/147 preterm breech infants (the equivalent of 61.2 per 1000) were diagnosed with DDH, which is in range with the overall incidence of DDH in the population per ultrasound screening.

Another finding of our study was that screening before 40 weeks of Corrected GA (CGA) was performed in 34.7% of all infants and 66.7% of infants were ultimately diagnosed and treated for DDH. It has been demonstrated that preterm breech infants screened before 40 weeks GA has a nearly eight-fold higher risk of having abnormal US findings that spontaneously resolve compared to those screened at a corrected age of 44 weeks [5]. In this study [7], the authors proposed that this potentially contributed to the number of DDH cases observed secondary to hip immaturity rather than true hip pathology, which was 85% of all cases observed. It is not surprising that infants born prematurely would demonstrate immature hip development. Hip development continues throughout gestation and during the neonatal period, fully complete at the end of the first decade of life [5].

Additionally, recent evidence has shown that ultrasound screening on preterm infants before 38 weeks GA increases the detection rate of minor abnormal findings that resolve spontaneously in preterm infants [12]. In our study, detected cases of DDH were treated conventionally with a Pavlik harness and surgery if an infant failed to respond to the harness. It is impossible to know whether these infants would have had spontaneous resolution without appropriate treatment. However, it is interesting to note that many were screened and treated within this timeframe, which is known to increase the rate of falsely positive hip dysplasia findings.

Because many of the adverse effects of DDH are not apparent within the first year of life, screening is an essential tool to determine which infants are at risk. An ideal screening protocol avoids over- and under-screening, both associated with adverse consequences. The consequences of under-screening are straightforward. Missing a chance to intervene on early dysplastic hips increases the risk of long-term morbidity from this condition, including functional disability, hip pain, and accelerated osteoarthritis [7]. Whereas over-screening increases the risk of false positives, which lead to interventions that are not harmless. Abduction devices like the Pavlik harness treat DDH non-surgically as a first-line intervention. Complications from these devices include avascular necrosis of the femoral head, skin irritation, and femoral nerve palsy [25]. Controversy exists regarding what screening protocols should be utilized, with many countries opting for universal screening, others opting for no screening, and some choosing to selectively screen. The United States Preventative Services Task Force (USPSTF) last updated recommendations for DDH screening in 2006 [26]. In it, they describe an inability to assess the benefits and harms of screening for DDH but were concerned about the potential harms associated with treating infants identified by routine screening. The potential harms of routine screening are likely amplified in the preterm breech population compared to the term breech population due to the decreased incidence detected in our study and others.

Few investigations have studied the association between premature breech infants and DDH. A retrospective review in Australia sought to determine if preterm breech infants are at high risk for DDH [15]. This study found no significant differences between DDH incidence in term and preterm breech infants as determined by clinical examination at six weeks of corrected age. Their incidences were 1.8% in term and 2.3% in preterm breech infants. A retrospective review completed in 2016 in New York assessed DDH status in preterm breech infants [7]. They subdivided preterm breech infants into 32-37 weeks GA and <32 weeks GA. In the former population, the incidence was 9%, while in the latter, this was 2%. Another Australian group sought to determine if preterm breech infants have a similar risk of DDH compared to term breech infants [16]. This was a retrospective review with a clinical exam done in the neonatal period and an ultrasound screening done at six weeks of corrected age. They found that the incidence of DDH did not differ between any subgroups, which included a 23-27-week GA group, a 28-31-week GA group, a 32-36-week GA group, and a >37-week GA group. Most recently, a group in Pennsylvania completed a retrospective review on the incidence of DDH in preterm infants to determine if being breech is associated with DDH in this population [13]. They found the incidence of DDH in preterm breech infants born <35 weeks to be 0.47% and concluded that there is no association between breech presentation and DDH in preterm infants. Many groups have similarly demonstrated no increased risk associated with breech positioning in premature infants

[27-29]. Interestingly, the studies relating prematurity and breech presentation with DDH differ between those performed in the United States and those performed in Australia. Our results align with local research suggesting that the incidence of DDH is lower in preterm breech infants compared to the term breech population. According to clinicaltrails.gov, no ongoing trials evaluate DDH in the preterm population, regardless of breech status.

A novel component of our study was evaluating provider practices related to DDH screening in the breech preterm population. According to our survey results, there is ongoing discordance among provider practices. The majority from each group sampled believed that preterm breech infants should be screened. However, survey respondents varied regarding when they thought these infants should be screened. This variation highlights the need for updated clinical guidelines for evidence-based practices. The American Academy of Pediatrics published the most recent guidelines in 2000. In it, they recommend ultrasound screening at six weeks of age for infants born in breech presentation, but these guidelines do not comment on ultrasound screening specific to preterm infants. In fact, these guidelines suggest that DDH may go unnoticed in the preterm population [18]. Our understanding of DDH is rapidly evolving. There is continued investigation into the incidence of hip dysplasia, the risk factors associated with the condition, the most appropriate screening guidelines, and the appropriate treatment and surveillance options. With such a dynamic landscape, clinicians need access to clear guidelines with appropriate recommendations concerning the most current clinical evidence.

There are several limitations to our study. First, this study was conducted as a single-center retrospective cohort with a relatively small sample size, especially in the DDH-positive subgroup. Consequently, this restricts our statistical power and precision in approximating DDH incidence in our study population. Also, some patients did not receive ultrasound screening at our institution or were discharged and received follow-up care outside of the NYU Langone system. The medical records of these patients were not accessible for us to determine the results of their examinations or medical treatment. Finally, our digital survey is limited by selection bias as it did not reach all segments of the provider population. Despite this, our national listservs were able to reach a wide audience and demonstrate the variety of clinical opinions regarding our focus of study. Furthermore, our study adds to a small but growing body of evidence regarding DDH risk in the preterm breech population in North America.

Our results suggest that breech positioning is not a risk factor for DDH in preterm infants. We conclude that the incidence of DDH in the preterm breech population is lower than in the term breech population. Additionally, we conclude that significant variation exists in provider screening practices relating to DDH in the preterm breech population. Clinical guidelines should be updated to reflect these and recent findings, especially concerning the discordance between provider practices in this condition.

Conclusion

In conclusion, we have demonstrated that the incidence of DDH in preterm breech infants is lower than the reported incidence in term breech infants. Furthermore, we have demonstrated that variation exists in the practices regarding screening for this condition in the preterm breech population. Updated guidelines, both nationally and internationally, are needed to address these clinical updates and disparities in clinical care to properly care for this population of infants.

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