Impact of Multiple Factors on the Incidence of Developmental Dysplasia of the Hip

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Abstract

Introduction: Developmental dysplasia of the hip (DDH) is one of the most common musculoskeletal conditions in children. Not treated DDH leads to disability, gait abnormalities, limb shortening and chronic pain. Our study aims to determine the impact of multiple risk factors on the occurrence of DDH and develop an interactive risk assessment tool.

Methods: We conducted a retrospective cohort study in the Outpatient Clinic for Children of University Hospital. The Graf classification system was used for ultrasonographic universal screening. In total, 4881 infants met the eligibility criteria (n = 9762 hip joints). Hypothesis testing was performed with χ^2 test and logistic regression.

Results: The incidence of DDH was 4.57%. We have proven risk factors of DDH: female gender (OR=7.11), breech position (OR=3.65), Caesarean section (OR=1.43), positive family history in parent (OR=1.92) or sibling (OR=3.84). Preterm delivery decreased the risk (OR=0.17). Logistic regression was used to construct the interactive risk calculator.

Conclusion: The DDH risk calculator was built but needs external validation in prospective study before being used in a clinical setting. We confirmed well-known DDH risk factors in the studied population. Our results support the recent hypothesis that preterm infants (37 < week) have lower rate of DDH.

Level of Evidence:

Retrospective cohort study: Level III

Introduction

Background

Developmental dysplasia of the hip (DDH) is one of the most common musculoskeletal disorders in children. DDH is an abnormal growth of the hip joint and surrounding tissues. It refers to a heterogeneous spectrum of abnormalities that range from mild acetabular defect to subluxation or complete dislocation of the femoral head. There is no universal definition of DDH, and the term is imprecisely described in the literature [1]. Undetected and not treated DDH can lead to severe disability, gait abnormalities, limb shortening, reduced range of motion in affected joints and chronic pain. Still, it is the leading cause of osteoarthritis and the main indication for total hip replacement in young adults [2]. The pathogenesis of DDH is still unclear. However, the literature identifies several risk factors, such as female sex, left side, breech position, family history of DDH, and first-born births [3]. The other studied risk factors are oligohydramnios, macrosomia, multiple pregnancy (MuP), hyperlaxity, torticollis, clubfoot, and metatarsus varus. Recently, vitamin D level alterations in DDH patients were demonstrated [4, 5]. Many genes, loci, and polymorphisms are also being investigated in DDH, but the evidence is limited [6, 7].

The diagnosis of DDH in newborns is based on clinical and ultrasound examination. The most commonly used ultrasound methods are Graf*s, Harcke*s and Terjesen`s [8]. Radiographs are useful only from the 3rd – 4th month of life and are the preferred method of evaluating and monitoring DDH after 6 months. The physical examination should include the test for leg length discrepancy (Galeazzi test), stability examination (Barlow's and Ortolani tests), assessment for asymmetric thigh or gluteal creases, and detection of limited abduction. Universal clinical screening of newborns is recommended by The American Academy of Pediatrics (AAP), the Orthopaedic Society of North America (POSNA), American Academy of Orthopaedic Surgeons (AAOS), and Canadian DDH Task Force [9]. However, even in experienced hands, physical examination findings in DDH can be subtle [10]. Dogruel et al. reported limited physical examination specificity (13.68%) compared to ultrasound findings (Graf). Most ultrasonography-diagnosed dysplastic hips were normal in clinical examination (71.63%) [11].

Currently, there are two primary approaches to DDH ultrasound screening – universal and selective. Ultrasound as a universal screening method for early diagnosis was introduced in several European countries such as Austria (1991), Switzerland (1995), Germany (1996) and the Czech Republic [12]. As the first among Asian countries, Mongolia (2017) recently launched universal ultrasound screening for newborns [13]. Implementing ultrasound-based universal screening led to a significant reduction of hip surgery due to DDH in children [14]. A second screening plan established in the United States and England is based on newborn clinical examination. Selective ultrasonography screening is recommended only in infants with positive or suspicious findings on physical examination. According to the AAP, selective ultrasound could also be useful in children with risk factors such as breech presentation, positive family history, hip instability, or history of lower extremity swaddling [9, 15]. Two randomised trials on universal vs. selective screening were conducted; however, the results were inconclusive, and the need for universal screening is still under debate in many countries [16, 17]. According to national arthroplasty registers, selective screening has not reduced the number of procedures performed due to early-onset OA in patients with DDH [18]. In universal screening countries, the first ultrasound is usually recommended before 6 weeks of life. It is recommended in the first week of life in cases with risk factors or positive physical examination [8]. There is no official universal screening program or guidelines for the DDH screening and control visits timeframes in Poland. However, ultrasound examination is done in most children. The frequency of DDH depends on ethnicity, race, age of the population studied, diagnostic criteria, and screening method (physical examination, plain radiographs, ultrasound technique). The incidence of DDH can range from 0.1% in Africans to 7.6% in Native Americans [19]. The current incidence of DDH and risk factors on the first visit in 6 weeks of life in Poland's outpatient clinics remains unknown. What is more, little is known about the cumulative effect of the beforementioned risk factors on DDH incidence in the population.

Objectives

This study aimed to determine the incidence of DDH in newborns in Poland with the Graf method during the first visit to an outpatient clinic. We also wanted to assess the occurrence of potential risk factors for DDH in the studied population. In our study, we present a linear regression model for DDH and a

dedicated risk calculator. In the discussion section, we confront the study results with other findings in the field of DDH.

Methods

The study design is a retrospective cohort study. The study setting was the Outpatient Clinic for Children of Orthopaedics and Traumatology Department of Medical University of Warsaw, Poland. The data was collected for all patients who attended the Outpatient Clinic for the universal ultrasonographic hip infant screening from January 2013 to December 2018. The study size was determined from the study type and included all who met eligibility criteria for participation. The screening method used in every patient was the Graf classification system (I-IV). Certified medical assistants performed the data collection in paper and electronic form. The physical examination included the hip-oriented orthopedic examination and general examination of the newborns. The consent for accessing and retrieving the medical data from the Outpatient Clinic archive was obtained from the head of the Orthopaedics and Traumatology Department of the Medical University of Warsaw, Poland (PM). The Institutional Review Board of the Medical University of Warsaw approved the study protocol on 10 June 2019 (AKBE/227/2019). Due to retrospective nature of the study informed consent was waived by ethics committee of the Institutional Review Board of the Medical University of Warsaw. All methods were performed in accordance with the relevant guidelines and regulations. For this report, we used STROBE Statement for observational studies.

Participants

The examined eligibility included all patients who attended the Outpatient Clinic for the ultrasonographic hip infant screening from January 2013 to December 2018. The newborns who attended the first visit for hip ultrasound in another secondary facility were excluded from the study. The population represents all socioeconomic groups, but the ethnicity is relatively homogenous.

Variables

According to Graf's classification, the diagnostic criteria of DDH were Graf type IIa (-), IIb, IIc, D, III, and IV images. Type IIa (+) hips were monitored and treated only in the absence of signs of sufficient maturation (IIa (-) or IIb) [18]. In our clinic first ultrasound examination is recommended at 6 weeks of life. In case of a positive physical examination upon birth or risk factors, ultrasound is recommended upon first weeks of life. The second control visit is also recommended for healthy children at 12 weeks. The record of the orthopedic examination of the hips included maximum abduction angle value for each hip joint, Ortolani test, Barlow test and Galeazzi test. The asymmetry of abduction was defined as the difference of 20^0 or more. Articular noises in physical examination such as “clicks”, or “creaks” were not classified as pathological findings [18]. The record included the name, national identification code, age, and the date of the visit. The information on potential risk factors such as female sex, abnormal presentation, high birth weight, term of birth, MuP, mode of delivery, diabetes, positive family history and coexisting medical conditions in children were also collected.

Data sources

The physical examination was conducted by an experienced orthopedic surgeon (PW, PG, WW, RW and GT) who also performed the ultrasound and the α and β angles measurements. Ultrasound device operating with a 7-10 MHz linear transducer and holding cradle was used. The diagnostic criteria of DDH were according to Graf's classification [20]. The analysis included the first control visit in the clinic in type Ia, type Ib Graf hips. In patients with IIa (+) upon the first visit, the analysis included the subsequent visits to assess if the treatment was implemented in patients with insufficient hip maturation on next visits.

Bias

Our study is limited to children who do not have obvious dislocations diagnosed upon birth. This group of newborns could be directed to dedicated centers specialized in children's orthopedic surgery. This fact can modify the study results and possibly lower the DDH rate in our cohort - especially Graf type III and IV with hip instability in physical examination. It is also worth mentioning that the accuracy of the examination, especially regarding specificity, is closely related to the examiner's skills. Graf method must be performed in strict compliance with the author's instructions [18]. Only certified orthopedic surgeons performed the ultrasound examination (PW, PG, WW, RW and GT).

Quantitative variables

We decided to set a cutoff value for limited hip abduction as the difference of 20^0 or more between both hips. We do not analyze the bilateral limitation of abduction. According to Jari et al., bilateral limitation of hip abduction is not a useful indicator of DDH; however unilateral limitation of 20^0 or more abduction is a specific and sensitive sign of DDH [21].

We adopted the American College of Obstetricians and Gynecologists (ACOG) definition for fetal macrosomia as birthweight over 4.000 g irrespective of gestational age [22]. Preterm delivery was defined as birth before 37 weeks of gestation. We used the ACOG definition of post-term pregnancy. It is defined as a pregnancy extended to or beyond 42 weeks of gestation [23].

Statistical methods

From the data, 2 x 2 tables were constructed. Odds ratios (ORs) and their 95% confidence intervals (CIs) were calculated from these tables. Hypothesis Testing was performed using a 2 x 2 contingency table with χ^2 Pearson Test. Yates's correction was used to prevent the overestimation of statistical significance for small data samples. Cases with missing values were removed from each 2 x 2 table. For each variable the complete cases number is given in Table 1. The subgroup analysis was also performed with the χ^2 Pearson Test. Binomial logistic regression was performed to ascertain the effects of risk factors on the likelihood of DDH. A stepwise hierarchical model was used. We used the Statistica 13.3 analytics software by TIBCO.

Table 1
Risk factors of developmental dysplasia of the hip (DDH)

Variable	No	Yes	All	χ^2	OR (CI 95%)
Female Gender					
Healthy	2413	2247	4660	p=0.000	7.11
DDH	29 (1.19%)	192 (7.87%)	221	(Pearson)	(4.79-10.55)
All	2442	2439	4881		
Delivery presentation - Breech					
Healthy	3552	189	3741	p=0.000	3.65
DDH	144 (3.90%)	28 (12.90%)	172	(Pearson)	(2.38-5.62)
All	3696	217	3913		
Cesarean section					
Healthy	2407	1560	3967	p=0.016	1.43
DDH	97 (3.87%)	90 (5.45%)	187	(Pearson)	(1.07-1.92)
All	2504	1650	4154		
Delivery < 37 week					
Healthy	3288	259	3547	p=0.003	0.17
DDH	148 (4.50%)	2 (0.77%)	150	(Yates)	(0.04-0.70)
All	3436	261	3697		
Abnormal findings in physical examination					
Healthy	4415	11	4426	p=0.001	25.76
DDH	187 (4.07%)	12 (52.17%)	199	(Pearson)	(11.21-59.13)
All	4602	23	4625		
Positive family history of DDH - parent					
Healthy	3781	285	3781	p=0.004	1.92
DDH	166 (4.21%)	24 (7.77%)	166	(Pearson)	(1.23- 2.99)
All	3947	309	3947		
Positive family history of DDH - sibling					
Healthy	3990	58	4048	p=0.001	3.84
				(Pearson)	(1.93-7.64)

OR – odds ratio; *p* – significance; 95% *CI* – 95% confidence interval

Variable	No	Yes	All	χ^2	OR (CI 95%)
DDH	179 (4.29%)	10 (14.71%)	189		
All	4169	68	4237		
OR – odds ratio; *p* – significance; 95% *CI* – 95% confidence interval					

Results

Participants

Among 4891 infants who underwent hip ultrasonography in the Outpatient Clinic from 01 January 2013 to 31 December 2018, examined for eligibility 4881 met the criteria (n = 9762 hips). Ten initially screened infants excluded from the study were continuing care started in an external facility.

Outcome & descriptive data

Females constituted 48.23% of participants while males 51,73%. Mean delivery was at 38.90 ± 2.00 weeks and mean birth weight was 3369.59 ± 575.58 g. The first visit mean time from birth was 7.98 weeks (median 8 weeks minimum 1 week maximum 31 weeks). The number of participants with available data for each variable is shown in Table 1.

Main results

The incidence of DDH in the studied group that needed treatment was 4.57%. Bilateral DDH occurred in 100 out of 218 DDH cases (45.87%). The distribution of Graf hip types is presented in Table 2. The treatment methods used are listed in Table 3. The unadjusted estimates for odds ratio (OR) of each statistically significant DDH risk factor and their precision - confidence interval (CI 95%) are included in Table 1. The category boundaries for continuous variables are discussed in the methods section. The proven risk factors in study group where: female gender (OR = 7.11; CI 95% 4.79-10.55), breech position (OR = 3.65; CI 95% 2.38-5.62), caesarean section (OR = 1.43; CI 95% 1.07-1.92), positive family history of DDH in at least one parent (OR = 1.92; CI 95% 1.23- 2.99), and positive family history of DDH in at least one sibling (OR = 3.84; CI 95% 1.93-7.64). Preterm delivery (37 < week) decreased the risk of DDH (OR= 0.17 CI 95% 0.04-0.70). Abnormal findings in the physical examination were highly associated with DDH (OR = 25.76; CI 95% 11.21-59.13). However, abnormalities in the clinical examination were infrequent and occurred in 0.47% of all patients, possibly due to strict criteria (methods section). In our study, 96,83% of DDH patients had at least one confirmed risk factor (female gender, cesarean section, breech position, family history of DDH). Only 7 patients were free from any significant risk factors (3.17%).

Table 2
Hip type, according to Graf in study participants

Hip type according to Graf	Right hip	Left hip
Type I	4615 (94.70%)	4632 (95.07%)
A	2501 (51.32%)	2510 (51,52%)
B	2114 (43.38%)	2122 (43.55%)
Type II	239 (4.90%)	220 (4.52%)
A	206 (4.23%)	180 (3.69%)
B	10 (0.21%)	11 (0.22%)
C	23 (0.47%)	29 (0.60%)
Type D	1 (0.02%)	0
Type III	16 (0.32%)	19 (0.39%)
Type IV	2 (0.04%)	1 (0.02%)

Table 3
Treatment modality used in study participants with DDH

Treatment method	Number of patients (%)
Treatment not necessary	4652 (95.43%)
Tübinger orthosis	163 (3.34%)
Padded abduction diapers	47 (0.96%)
Frejka pillow	5 (0.10%)
Cast	2 (0.04%)
Koszla abduction brace	2 (0.04%)
Pavlik Harness	1 (0.02%)
Hospitalization	2 (0.04%)
Parents refused treatment	1 (0.02%)

Other analyzed

Cesarean section (n = 1490) was a significant risk factor of DDH, and it is associated with a high rate (13.5%; n = 196) of the breech position OR = 28.24 (CI 15.71-50.80; p<0.001). While in vaginal deliveries, the breech position was reported in only 0.53% (n=12). Breech position in infants delivered with Cesarean section was a significant risk factor of DDH (OR = 3.59 CI 2.21-5.86; p<0.001). After excluding breech

delivered infants, cesarean section was not a significant risk factor of DDH (OR=1.12 CI 0.81-1.55; p>0.05).

Logistic regression model

The regression algorithm reached its final solution in 9 steps. The final logistic regression model was statistically significant, $\chi2(5) = 169.81$, $p < .001$. Hosmer and Lemeshow's goodness of fit test was statistically insignificant indicating that the model is not a poor fit. Of the all-predictor variables only five were statistically significant: gender, breech position, delivery 37 < week, positive family history of DDH in at least one sibling, abnormal findings in the physical examination (as shown in Table 4). Females were more likely to develop DDH. The presence of the breech position, positive family history of DDH in at least one sibling, abnormal findings in the physical examination caused the DDH to be more likely, whereas delivery 37 < week lowered the likelihood of DDH. The model explained 17% (Nagelkerke R2) of the variance and correctly classified 95.8% of cases. Sensitivity was 0.06%, specificity was 99.8%. DDH risk score is calculated based on OR of the before mentioned factors. The risk calculator is attached in a supplementary file, and it is free to use. The result can be obtained by selecting “yes” or “no” in the checkbox.

Table 4
Model for predicting group belonging (DDH or no DDH) resulting from regression analysis

	Estimate	*SE*	*OR*	z	Wald Test	*p*	95% *CI*	
							LL	UL
Intercept	-4.82	0.26	0.008	-18.78	359.74	<.001	-5.32	-4.32
Gender (Female)	2.02	0.26	7.56	7.76	60.26	<.001	1.51	2.53
Breech position (Yes)	1.51	0.27	4.55	5.53	30.57	<.001	0.98	2.05
Delivery 37 < week (Yes)	-2.08	0.76	0.12	-2.74	7.49	.003	-3.57	-0.59
Positive family history in sibling (Yes)	1.23	0.46	3.44	2.67	7.14	<.001	0.33	2.14
Abnormal physical examination (Yes)	2.97	0.54	19.53	5.54	30.74	<.001	1.92	4.02

SE – standard error; *OR* – odds ratio; *z* – *z* score; *p* – significance; 95% *CI* – 95% confidence interval; *LL* and *UL* – lower and upper 95% confidence interval limit

Discussion

Key results

The overall aim of the study was to assess the incidence of DDH in the Polish population and investigate whether the risk factors described in the literature for DDH are also reflected in this group of patients. The

occurrence of DDH during the first screening visit is high (4,57%). In our work, we confirmed the already known risk factors of DDH (Table 1). We also discuss some other risk factors that have appeared in the literature. Logistic regression was used to construct the risk calculator, which can be used as a clinical decision tool in the future.

Interpretation

In Poland, before implementing ultrasonography, the DDH rate was relatively high 6.80% and the dislocation was reported in 1.06% of the population [24]. It can be speculated that the high DDH rate was due to different diagnosis methods at the time, and only suspected infants had undergone the diagnostic process. Using the ultrasound screening, DDH was diagnosed in 5.60% of the newborns in the first week of life (Łódź, Poland) [25]. In our study, the occurrence of DDH in the university hospital in the capital city (Warsaw, Poland) during the first screening visit was lower – 4.57% than in the beforementioned study. This difference is probably due to the hip maturation curve and visit timing 1 week vs. median of 8 weeks. The DDH diagnosis can depend on the timing of the examination and the method used. As a child grows older, the hip joint matures, which can be observed with both ultrasound (α angle) and radiographs (Acetabular Index, Acetabular Depth Ratio) [20, 26].

Our work confirmed some of the already known risk factors such as female gender, breech position, cesarean section, abnormal findings in the physical examination, and positive family history of DDH. However, according to the literature, there was not even one risk factor in up to 73 - 95% of DDH cases. Also, most children with risk factors do not develop DDH, and the disease can be observed only in 1 - 10% of cases [27–30]. In contrast to these results in our study, 96.83% of DDH had at least one statistically significant risk factor.

The female gender is considered one of the most important risk factors of DDH [31]. We can find 2.4 -9.2 (OR) statistics in the available literature when comparing females to males [27]. This strong relationship was also confirmed in our study 7.11 (OR). This phenomenon is still under investigation, and the mechanism of this connection remains unclear. Various theories explain it - the most common is the gender-dependent influence of hormones, particularly relaxin, on hip joint development. Relaxin is a polypeptide hormone produced by the corpus luteum, endometrium, decidua, and placenta [32]. It has an inhibitory effect on uterine muscle contractions and relaxing on the pubic symphysis during labor. The role of relaxin, present in the blood serum, ultimately stimulates collagen turnover by increasing the secretion of matrix metalloproteinases (MMPs), collagenase and a plasminogen activator [27]. Yamasato et al. confirmed a higher expression of relaxin receptors in the placenta of the female fetus [33]. Another study by Dragooet et al. revealed that the female sex is also associated with higher relaxin receptor expression in the anterior cruciate ligament [34]. The recent reports from Ayanoget et al. confirm the association of DDH and the number of relaxin receptors in the ligament of the femoral head. However, the study does not report the difference in receptor expression depending on infant gender [35]. Although the function of relaxin is already known, there is no evidence in the literature that this mechanism of hip joint laxity is exclusively responsible for the higher frequency of DDH in females. What is more, some studies have demonstrated opposite results – lower level of the hormone in umbilical blood and higher risk of

DDH [36] Given the above, some researchers have theorized that other maternal factors, such as poorer preparation of a mother's delivery canal, may also play a role, but this does not fully explain the significant prevalence of DDH in females [37].

There is also a whole group of so-called mechanical factors, in which there is pressure on the hip joint during pregnancy by the uterus walls or by the delivery canal tract at birth. One of the mentioned factors is the abnormal position and presentation of the fetus. Many authors have already described breech positioning as a risk factor. Andersen et al. advocated that neonate in a breech-presenting position in the fetal state have exerted a significant stretching force on the hip joint capsule, thus causing hip instability [38]. According to Dezateux et al. complete breech vaginal delivery (3% of all births) links with a 17- fold increased risk of DDH (OR = 17.15; CI 95% 2.79 - 22.99), while breech presentation resolved by Caesarean section relates to ten times increased risk (OR = 10.03; CI 95% 8.58 -11.72) [31]. These findings were confirmed in metanalysis (35,139 infants) by Panagiotopoulou et al. [39]. Therefore, breech positioning is probably an important risk factor of DDH during pregnancy and birth when significant forces are applied to the hip joint [40]. Our results appear to be consistent with the existing literature. We recorded 217 (5.55%) babies in the breech position, of whom 28 developed DDH, which is over 12.90% (OR = 3.65; CI 95% 2.38 - 5.62).

All authors describe abnormalities on physical examination as a risk factor - which was also unquestionably demonstrated in our study. We considered the following as abnormalities: positive Ortolani/Barlow test and/or hip joint abduction asymmetry 20 degrees or more and abduction of the joint less than 45 degrees, which is consistent with the available evidence. Some patients do not present any abnormalities on clinical examination, but an ultrasound examination reveals dysplasia. In our study abnormalities on physical examination predisposed to DDH diagnosis with OR = 25.76 (CI 95% 11.21-59.13)

Available scientific knowledge indicates that positive family history is one of the most important risk factors of DDH [27]. According to the consensus of the Committee on Quality Improvement, Subcommittee on Developmental Dysplasia of the Hip, a risk of DDH was defined as 6% in cases of healthy parents and recognized DDH in siblings, 12% in cases of confirmed DDH of the mother/father and 36% in cases where DDH was recognized in one parent and a brother/sister. The correlation was also confirmed in our findings [41]. Authors state that higher risk can also be observed if the disease was recognized in a first-degree cousin of the child. In those cases, the risk is specified at a level of 1.7% [28]. Until recently, it was believed that individual genes are responsible for malfunctions in the physiology of connective tissue or proteins of the joint capsule. A hypothesis of a two-gene system in DDH inheritance has also arisen in literature. However, new, more progressive methods for studying molecular biology have led to further findings. We are now familiar with more than a dozen DDH-associated genes and their locations. For Caucasians, these include IL-6 and TGF-β1 genes mutations, for Asian populations: COL3A1, DKK1, HOXB9, HOXB9, HOXD9, WISP3. Non-location-specific genes mutations were also found: COL1A1, CX3CR1, GDF-5 and PAPPA2. At present, research is managing to identify individual genes in specific populations - unfortunately, at this time, we do not know the individual genes that may be

involved in DDH for the world population [42]. At present, researchers are attempting to determine the exact mechanisms by which these genes may be involved in the development of DDH. No differences in a higher frequency of DDH were found in genetic disorders like Ehlers-Danlos or Marfan's syndrome [43].

Some studies investigate the mode of delivery as potential risk factor of DDH, but the results are inconclusive. In metanalysis by L. Ortiz-Neira et al. two studies indicated that vaginal delivery could be a risk factor of DDH, and one study suggested the opposite. The results of the fourth study were inconclusive. The metanalysis of the beforementioned studies resulted in non-significant influence [44]. In our study caesarean section was associated with high DDH rate (OR = 1.43; CI 95% 1.07-1.92; p=0.016). However, cesarean section is related to a very high rate of breech position (13.5%; n = 196), which is a significant risk factor of DDH. The mode of delivery is most likely not a risk factor itself. After excluding breech delivered infants, cesarean section was not a significant risk factor of DDH (OR=1.12 CI 0.81-1.55; p>0.05).

We also examined MuP as a potential risk factor in our publication. Although the results of our study do not indicate a direct correlation, there is some evidence in the literature that the relationship could be significant for the female gender. According to Dezateux et al., special attention should be paid to MuP, especially when the children's gender is female; congenital joint hypermobility was observed in 70% of those cases [31].

Some authors consider the presence of congenital diseases as one of the DDH risk factors. Congenital Muscular Torticollis (CMT) can be associated with an increased risk of DDH to a level of 17%. Significant differences in correlations were also observed by gender - a fivefold increase of hip joint dysplasia was observed in male newborns with coexisting CMT compared to female newborns with CMT [27]. Some publications indicate Congenital Foot Deformities: as a possible risk factor for DDH. It has been shown that Talipes Calcaneovalgus or Metatarsus Adductus may be associated with an elevated risk (at the level of 4-6% and 4%, respectively). However, no connection between Talipes Equinovarus (TEV) and DDH was revealed in available data [28]. This relationship was not confirmed among our patient group.

In our study preterm delivery (37 < week) decreased risk of DDH (OR= 0.17 CI 95% 0.04-0.70). The theory explaining this phenomenon is shorter exposure to maternal hormones and lack of mechanical problems with intrauterine leg movement. Similar results were obtained in the study by Lange et al. [45] and data from the Swedish Medical Birth Register [46].

The literature indicates that most ultrasonography-diagnosed dysplastic hips are normal in clinical examination (71.63%) [11]. Similarly, only 12 out of 221 treated for DDH had a positive physical examination in our study (5,43%). Probably due to strict criteria of 20^{o} of abduction angle difference vs. contralateral side and characteristics of the cohort – first clinical screening with physical examination upon birth. Neonates with positive Ortolani and Barlow signs are directed directly to dedicated wards after the birth.

Other risk factors, sometimes raised in scientific discussions, appear to be statistically insignificant in most studies. We examined some of them (i.e., Apgar Score <10, oligohydramnios, fetal macrosomia, parity, post-term pregnancy). Our results are consistent with the worldwide results (included in supplementary file).

Sahin F et al. highlighted that the negative predictive value of DDH risk factors combined with physical examination is high and calculation of patient's risk could be used as a decision tool for ultrasound screening [47]. Similarly, Woodacre et al. proposed to modify the UK screening program by calculating risk for each child [48]. Roposch et al. proposed the first DDH risk calculator based on analysis of selectively screened patients in the British population. Female gender, family history, physical examination and birthweight were taken into consideration. The model demonstrated excellent discrimination and calibration of observed and predicted risk [49]. Our model was built based on universal ultrasound screening and includes risk factors from a logistic regression significant for the Polish population.

Limitations

The study is retrospective, so it could influence the data collection quality and increase the missing value rate. Furthermore, the results do not reflect the situation for the entire country. Thus, additional well-designed multicenter prospective studies on this subject are required.

Generalisability

The study results indicate that the DDH rate in Poland is high. There is a need to establish an official guideline for DDH infant screening. The proposed risk model with a high specificity of 99.8% and good negative predictive value can contribute to the discussion concerning selective vs. universal ultrasound hip screening in the population. The DDH risk calculator could be used as a decision tool for selective screening in the future. However, setting the optimal threshold for ultrasound screening will be challenging.

Declarations

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Competing Interests Statement

We report no conflict of interest.

Data availability statement

The datasets analysed during the current study are available from the corresponding author on reasonable request.

Author contributions

Conceived and designed the analysis – ŁP, PŁ

Collected the data – KR, AJ, PK, IT, PW

Contributed data or analysis tools - ŁP

Performed the analysis – ŁP

Wrote the manuscript - ŁP, AS

All authors reviewed the manuscript

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