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Prevalence of Developmental Dysplasia of the Hip in a Maternity Hospital in São Paulo, Brazil*

Prevalência de displasia do desenvolvimento do quadril em uma maternidade de São Paulo, Brasil

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Abstract

Objective To evaluate the prevalence of developmental dysplasia of the hip (DDH), that is, hips classified as Graf type-IIc or higher, among a sample of the population of newborns aged from 0 to 3 days of life, and to correlate the findings with the main risk factors described in the literature.

Methods An observational, cross-sectional, prospective study on a sample of newborns at a Maternity Hospital School in the city of São Paulo, Brazil, to assess the prevalence of DDH diagnosed by the Graf method and verify its correlation with the risk factors.

Results A total of 678 newborns underwent hip ultrasound (1,356 hips). The prevalence of DDH was of 5.46%. The logistic regression analysis showed odds ratios (ORs) with statistical significance for the following parameters: white ethnicity (OR = 2.561; 95% confidence interval [95%CI]: 1.07 to 6.11); multiparity (OR = 3.50; 95%CI: 1.62 to 7.38), female gender (OR = 4.95; 95%CI: 1.86 to 13.13); and breech presentation (OR = 2.03; 95%CI: 1,01 to 4.11).

Conclusion The prevalence of DDH in the sample was of 5.45% using ultrasound as a diagnostic method. This result is different from that of studies that assessed prevalence exclusively through physical examination (Ortolani maneuver). The main risk factors associated with a higher risk of developing DDH were newborns of the female gender, with breech presentation, firstborns, and of white ethnicity.

Keywords
- hip dislocation, congenital
- ultrasonography
- diagnosis
- newborn

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Resumo

Objetivo Avaliar a prevalência de displasia de desenvolvimento de quadril (DDQ), ou seja, quadris de tipo de Graf IIc ou maior, em amostra da população de recém-nascidos de 0 a 3 dias de vida, e correlacionar os achados com os principais fatores de risco descritos na literatura.

Métodos Estudo observacional, transversal e prospectivo em amostra de recém-nascidos, em um hospital maternidade em São Paulo, para avaliar a prevalência da DDQ diagnosticada pelo método de Graf e verificar sua correlação com os fatores de risco.

Resultados Um total de 678 recém-nascidos foram submetidos a ultrassonografia de quadril (1.356 quadris). A prevalência de DDQ (quadris tipo IIc, D, IIIa, IIIb, e IV de Graf) foi de 5,46%. A análise de regressão logística mostrou razões de chances (RCs) com significância estatística para os parâmetros etnia branca (RC = 2.561; intervalo de confiança de 95% [IC95%]: 1,07 a 6,11), multiparidade (RC = 3,50; IC95%: 1,62 a 7,38), sexo feminino (RC = 4,95; IC95%: 1,86 a 13,13), e apresentação pélvica (RC = 2,03; IC95%: 1,01 a 4,11).

Conclusão A prevalência de DDQ na amostra foi de 5,45% usando a ultrassonografia como método de diagnóstico. Este resultado é diferente do dos estudos que avaliam a prevalência exclusivamente do exame físico (manobra de Ortolani). Os principais fatores de risco associados ao maior risco de DDQ foram recém-nascidos do sexo feminino, com apresentação pélvica, primogênitos, e de etnia branca.

Palavras-chave

- luxação congênita de quadril
- ultrassonografia
- diagnóstico
- recém-nascido

Introduction

There is a lack of studies and evidence available to guide the clinical practice in the treatment of developmental dysplasia of the hip (DDH). Among the limitations observed, there is no consensus regarding the classification and diagnosis of DDH, and a wide variability in decision-making, almost always guided by studies with small samples.[1] Most of the existing studies on DDH are retrospective, have small samples (considering the number of hips and not individuals), and do not correct the results when cases of bilateral DDH are included.[1] Moreover, many studies do not include the entire spectrum of DDH.[1]

In the study published by Guarniero et al.[2] (1988) based on a meta-analysis and multiple logistic regression protocols, the estimated prevalence of this condition in Brazil was close to 1.1%, and this data was obtained through the positivity of the Ortolani maneuver. To improve the evidence available, it is necessary to use a standardized framework for reports and diagnoses, with consistency, and to conduct prospective studies with a sound methodological design.[1]

Acetabular dysplasia cannot be excluded by a normal physical examination, and ultrasound can be an important tool for the early diagnosis and treatment of the disease. Articular instability and hip dislocation can be diagnosed by physical examination using the Barlow and Ortolani maneuvers respectively.[3]

The present study aims to evaluate the prevalence of DDH in newborns aged between 0 and 3 days of life in a public maternity hospital and to correlate the findings with the main risk factors described in the literature.

Materials and Methods

The present study was approved by the Ethics Committee of both institutions involved, under numbers 1554 and 2016. The present manuscript was written following the guidelines of the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) for the communication of observational studies.[4]

The present observational, transversal, prospective study was conducted in a high-risk public maternity hospital in the city of São Paulo, Brazil, in which an average of 7,900 deliveries are performed per year. We performed the exams in the nursery because this would be the only time when we would have a minimally-acceptable sample to assess the prevalence among the population of live births, even though spontaneous improvement regarding the classification has been described in the literature, specifically in the hips that do not show instability on the dynamic examination. The collection was performed randomly when the main researcher and her team attended the hospital, from March to September 2018. The parents or legal guardians were informed about the study conditions through the Free and Informed Consent Form presented before the ultrasonography examination.

The population of the present epidemiological study consisted of roomed-innewborns aged between 0 and 3 days of life for the early detection of the disease. Each participant was eligible only once, and random sampling was performed. To calculate the sample size required, we estimated the number of 500 cases to obtain an incidence of up to 5.5% with an estimated error rate of up to 2%. A 95% confidence interval (95%CI) was established. Since, after the

collection, the covariate and risk factor assessments would be performed using multivariate analysis, with possible data loss, we estimated an increase of 30% in the size of the sample. Thus, the required sample size for the present study was estimated as at least 650 cases.

The inclusion criterion was roomed-in newborns in the maternity hospital on the dates when the research team attended it.

The exclusion criteria were:

1) Extreme preterm birth: due to the need for oxygen and heating in the incubator, and to avoid excessive heat loss and manipulation of the at-risk newborn, as it is necessary to use cold gel and change the newborn's position to perform the ultrasonography, which increases the risk of intracranial hemorrhage;
2) Breathing difficulty: due to the need for oxygen and monitoring, as it is necessary to manipulate the newborn and change his or her position to perform the ultrasonography;
3) Pathologies and/or conditions that required intensive care: to avoid manipulating the newborn, as it is necessary to change his or her position to perform the ultrasonography;
4) Congenital anomalies, genetic diseases: because patients with genetic diseases and/or congenital disorders have a known higher prevalence of DDH, which could be assessed as a selection bias and increase prevalence, not translating the average dysplasia in the general population;
5) High-risk pregnancy: due to the need to monitor the newborn;
6) Parents or legal guardians who did not sign the consent form because they did not agree to participate in the study; and
7) Hips classified as type-IIa according to the Graf Method because of the immaturity that could normalize in a few weeks.

Upon acceptance to participate in the study, a hip ultrasonography examination was performed on the patients who had already undergone a clinical examination by a neonatologist, with the Ortolani and Barlow maneuvers.[5] Finally, a standardized questionnaire on the subject of the study, made by the authors, was applied to record the risk factors for DDH.

Ultrasonography Examination

The method used was developed by Graf[6] in 1980; it is considered the reference method, and it is accepted by healthcare systems in several countries. All examinations in the present study were performed strictly following the Graf method.

A pediatric sonographer with more than 20 years of experience in pediatric hip ultrasonography, who was blinded to the clinical examination results, performed the ultrasound examinations.

Positioning

The newborn is positioned in lateral decubitus, opposite to the hip to be examined, with the leg slightly flexed and adducted, and the foot in slight internal rotation (►**Fig. 1**), using a linear transducer, in the coronal plane of the hip, thus establishing a standard plane in which it is possible to evaluate the acetabular morphology and the degree of femoral head coverage.

Fig. 1 Correct position of the newborn during the exam: lateral decubitus, with the lower limbs flexed and adducted. Credits: art by Vinicius Mustafa.

In the classification of the Graf method, type I is normal, with an α angle $> 60°$. Type IIa hips represent immaturity, with an α angle between 50° and 59°, and a β angle $> 55°$. Hips with an α angle $\leq 49°$ are defined as presenting pathological development, and are classified as types IIc, IId, IIIa, or IIIb. Type-IV hips on the Graf method are not measurable. Developmental dysplasia of the hip was defined by Graf when the ultrasonography classification reveals a pattern $\geq$ IIb; type-IIa hips are classified as immature, but follow-up and treatment are suggested when they persist after 30 days of life.[7]

Statistical Analysis

The data were inserted in an Excel (Microsoft Corp., Redmond, WA, United States) spreadsheet. For the statistical analysis, the data were exported to the Statistical Package for the Social Sciences (IBM SPSS Statistics for Macintosh, IBM Corp., Armonk, NY, United States) software, version 24.0. The descriptive statistics of the categorical data are presented according to their absolute and relative frequencies. The continuous data are presented according to their means and respective standard deviations (SDs). The prevalence was calculated by the ratio between the number of newborns with at least one hip with a classification $\geq$ IIc according to the Graf method and the number of newborns assessed.

For the analysis of the risk factors, binomial logistic regression was performed to verify the effects of gender (male and female), ethnicity (white and others), parity (one delivery or multiparity), and the intrauterine presentation of the fetus at the time of delivery (breech or others) to identify the increased risk of developing DDH. For the regression analysis, we used independent variables with biological plausibility that presented an association on the univariate analysis with acceptance in the $p \leq 0.1$ model. To be accepted

as statistically significant, in the risk factor prediction equation, the significance value should be $p < 0.05$.

Results

There were 3,970 deliveries between April and September 2018, of which 733 newborns were available for examination. However, during that period, 28 newborns were premature, 16 presented transient respiratory distress, and 3 had aspirated meconium, and they were excluded from the study due to the need for intensive care. Eight newborns whose parents or legal guardians did not accept to participate in the study were also excluded. Approximately 8% of the newborns were excluded from the sample according to the stipulated criteria (►**Fig. 2**) There were no complications during the examination performed for the research.

A total of 678 newborns (1,356 hips) were included in the present study. The prevalence of DDH was of 5.46% (37 newborns). In total, 24 newborns had unilateral dysplasia: 3, on the right hip (8.1%), and 21, on the left hip (56.7%); and 13 (31,5%) newborns had bilateral dysplasia. The sample characteristics are described in ►**Table 1**. Among the newborns with DDH examined, the prevalence was assessed in relation to each type of hip using the Graf method (►**Table 2**).

Among the risk factors assessed, the following presented statistical significance on the univariate analysis:

- Female sex (relative risk [RR] = 5.78; 95%CI: 2.28 to 14.67; $p = 0.0002$);
- Breech presentation (RR = 2.94; 95%CI: 1.58 to 5.48; $p = 0.0007$);
- One delivery (RR = 3.36; 95%CI: 1.65 to 6.83; $p = 0.0008$); and
- White ethnicity (RR = 3.69; 95%CI: 1.65 to 8.3; $p = 0.001$).

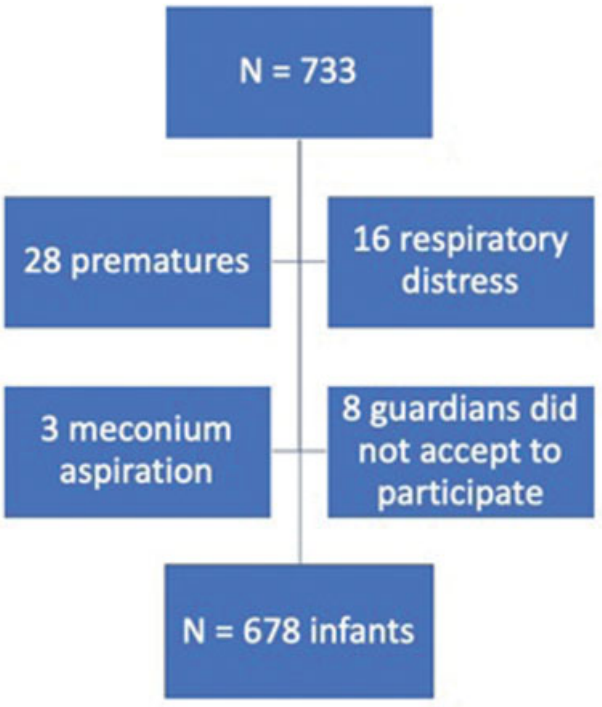

Fig. 2 Flowchart of the sample selection.

Table 1 Characteristics of the studied population in relation to the risk factors

Characteristic	Children evaluated ($N = 678$) – n (%)	Newborns with developmental dysplasia of the hip ($N = 37$; 5,4%) – n (%)
Gender		
- Female	356 (52.5%)	32 (86.5%)
- Male	322 (47.5%)	5 (13.5%)
Ethnicity		
- White	364 (53.7%)	30 (81%)
- Black	177 (26%)	0
- Brown	137 (20.3%)	7 (19%)
Type of delivery		
- Vaginal	449 (66%)	22 (59%)
- Cesarean	229 (34%)	15 (41%)
Presentation		
- Pelvic	179 (26.4%)	19 (51%)
- Cephalic	474 (69.9%)	17 (46%)
- Transversal	25 (3.7%)	1 (3%)
Parity		
- One delivery	302 (44.5%)	27 (73%)
- Multiparous	376 (55.4%)	10 (27%)
Twinning	4 (0.6%)	0
Family history	19 (2.8%)	1 (2.7%)
Orthopedic pathologies	0	0
Genetic syndromes	0	0
Positive Ortolani maneuver	4 (0.6%)	2 (5.4%)

Table 2 Prevalence in relation to each type of hip using the Graf method ($N = 1,356$)

Hip Classification	Right hip – n (%)	Left hip – n (%)
Ia	594 (43.6%)	554 (40.6%)
Ib	8 (0.6%)	23 (2.3%)
IIa	60 (4.4%)	67 (4.9%)
IIc	1 (0.07%)	3 (0.22%)
IId	9 (0.66%)	23 (1.65%)
IIIa	4 (0.3%)	4 (0.3%)
IV	2 (0.14%)	4 (0.3%)

The risk factors that were not statistically significant were:

- Cesarean section (RR = 1.33; 95%CI: 0.70 to 2.52; $p = 0.37$);
- Family history of DDH (RR = 0.96; 95%CI: 0.14 to 6.66; $p = 0.97$); and
- Twins (RR = 1.80; 95%CI: 0.12 to 25.4; $p = 0.66$).

Regarding family history, there may be information bias due to the difficulty in collecting data regarding the families, because most of the parents or legal guardians felt insecure about providing information regarding family history. Twins, also referred to as a risk factor, did not present statistical significance. It is necessary to consider a possible selection bias due to prematurity and indication for hospitalization, which was considered an exclusion factor in the present study.

The sensitivity of the Ortolani maneuver for hips with dysplasia (Graf type-IIc or higher) was only of 5.41% (95%CI: 0.66 to 18.2%), with a specificity of 99.7% (95%CI: 98.8 to 99.9%). For the diagnosis of dislocated hip (Graf type-IV), the sensitivity of the Ortolani maneuver was of 50% (95%CI: 6.76% to 93.24%), with a specificity of 99.7% (95%CI: 98.8% to 99.9%). The positive predictive value was of 50% (95%CI: 15.55 to 84.5%), and the negative predictive value was of 99.7% (95%CI: 99.2% to 99.9%).

The binomial logistic regression verified the effects of gender, ethnicity, parity, and fetal presentation at the time of delivery on the increased risk of developing DDH. The logistic regression model was statistically significant: $\chi^2(3) = 44.553$; $p < 0.001$. The model proved to be able to explain 18.4% (Nagelkerke R^2) of variation in the risk of developing DDH, and correctly classified 94.5% of the cases. All of the four predictive variables were statistically significant: gender, ethnicity, parity, and breech presentation. Being white, female, firstborn, and having breech presentation increased the risk of developing DDH, as shown in ►**Table 3**.

The logistic regression analysis showed an increased odds ratio (OR) for the white ethnicity (OR = 2.561; 95%CI: 1.07 to 6.11); multiparity (OR = 3.50; 95%CI: 1.62 to 7.38); the female gender (OR = 4.95; 95%CI: 1.86 to 13.13); and breech presentation (OR = 2.03; 95%CI: 1.01 to 4.11).

Discussion

The literature review did not reveal another prospective study estimating the prevalence of DDH in the city of São Paulo.

Developmental dysplasia of the hip can manifest in three ways: by joint instability, which can be diagnosed by the Barlow maneuver; by a hip dislocation, which can be diagnosed by the Ortolani maneuver; and by acetabular dysplasia, which cannot be excluded after a normal physical examination and requires the help of ultrasound for the diagnosis. Acetabular dysplasia is one of the most common forms of the disease, represented by the Graf method as types IIa, IIb, IIc, and D; and this information can be confirmed in the present study. As the late diagnosis of DDH can cause serious problems in adulthood, the use of tools such as the ultrasound for identification is justified, especially in a more recurring fashion.[3]

Guarniero et al.[2] evaluated the prevalence of DDH in their prospective/retrospective study conducted in the city of São Paulo through physical examination using the positivity of the Ortolani maneuver for its statistical calculation, finding a result of 1.1%. A comparison cannot be made between the present study and the aforementioned one, because the methodologies are different, and we understand that newborns with acetabulbar dysplasia who do not have a dislocated hip may have the disease, and the Ortolani maneuver may be negative.

Barbosa and Albernaz[8] were the only authors who tried to estimate the prevalence of DDH in Brazil, in a retrospective study performed in the School of Medicine at Universidade Católica de Pelotas. The authors reported many potential selection biases because the analysis was performed with a small number of cases identified through the patients' medical records.

In the meta-analysis by Ortiz-Neira et al.,[9] the prevalence of DDH was of 1,9% (20,196 cases of DDH among 1,065,867 patients). In 2019, Zhao et al.[10] found a prevalence of 174.9/1,000 in Tibet in a study with design similar to that of the present study, conducting echographic and clinical evaluations in newborns for DDH over 1 year, in 10 districts of different altitudes. The prevalence of DDH showed a significant correlation with high altitudes.[10]

"The incidence per 1000 live births ranges from 0.06 in Africans in Africa to 76.1 in Native Americans. There is a significant variability in incidence within each racial group by geographic location."[11] The role of acetabular dysplasia and adult hip osteoarthritis is complex.[11] Archaeological studies demonstrate that the epidemiology of DDH may be changing.[11]

Table 3 Binomial logistic regression

95% confidence interval for EXP(B)									
		B	SE	Wald	df	Sig.	Exp(B)	Lower	Upper
Step 1	White ethnicity	0.940	0.443	4.499	1	0.034	2.561	1.074	6.107
	Parity	1.240	0.387	10.247	1	0.001	3.455	1.617	7.381
	Gender	1.599	0.498	10.293	1	0.001	4.946	1.863	13.134
	Breech presentation	0.709	0.359	3.895	1	0.048	2.032	1.005	4.109
	Constant	−5.617	0.620	82.012	1	0.000	0.004		

In 2010, Pollet et al.[12] found a prevalence of 6.6/1,000 live births in a province in Canada, while other studies described an average incidence of 1 to 2/1,000 based only on clinical screening. A study[13] conducted in the United Kingdom (UK) and Ireland found an overall incidence of 6.7/1,000 in Ireland based on clinical screening with late presentation. In the UK, 0.34/1,000 infants had late DDH (after 3 months) based on an ultrasonography screening performed in a program involving 107,440 newborns.[13] In Norway, Engesæter et al.[14] found late presentation (after 1 month) in 0.32/1,000 newborns based on neonatal ultrasonography screening of a large group of newborns with risk factors and/or clinical findings. Güler et al.[15] reported a prevalence of 9.9% in Turkey.

In the present study, the main risk factors found were: female gender (RR = 5.78; $p = 0.0002$), breech presentation (RR = 2.94; $p = 0.0007$), firstborn (RR = 3.36; $p = 0.0008$), and white ethnicity (RR = 3.69; $p = 0.0015$), which were strongly associated with an increased risk of developing DDH. Black ethnicity is a protective factor; there were no black patients with DDH in the present study. There were only two pairs of twins eligible for examination in the first three days of life, who did not present DDH; thus this did not present a considerable statistical value.

Even though the characteristics family history and being a twin were shown to be significant risk factors in another study,[16] in the present study, they were not adequately assessed due to the selection bias, a limitation that mainly involves the exclusion factors, such as newborns in intensive care, prematurity, respiratory distress, hypoglycemia, or any clinical alteration that requires specific care.[16] There were only two pairs of twins eligible for examination in the first three days of life, who did not present DDH; thus did not present a considerable statistical value. We could not accurately assess the family history because most parents or legal guardians did not know about the existence of DDH cases in their families, which may have been underestimated in the resent study. Other limitations are the fact that the ultrasounds were performed by a single pediatric sonographer, and there was no follow-up of the development of the patients after the ultrasound. We also emphasize that the spresent tudy was performed in a hospital in the city of São Paulo, and we have reported the prevalence of the disease in this establishment.

Early diagnosis is considered essential for an effective treatment and a good prognosis.[17] Considering that there are cases of mild dysplasia, which, according to studies, resolve spontaneously in $\sim$ 90% of the cases,[18] an ultrasonography assessment of all newborns who do not present risk factors or clinical signs at 21 days of life could reveal cases of immaturity that would benefit from conservative treatment. We understand that there is a wide discussion about early treatment[19] in cases of immaturity of the hips. However, we know that the psychological, family and socioeconomic impact seems to be superior if we compare a treatment performed with a brace of Pavlik to surgical treatment with plaster-cast immobilization. Carlile et al.[20] suggest that newborns with important risk factors or positive clinical signs should undergo the examination until the sixth week of life.

Regarding DDH screening, it is recommended that morphology and stability be verified by ultrasonography, in addition to the clinical examination, which, similar to what happens in other countries, could be implemented in Brazilian institutions with easy access to a sonographer with experience to perform this exam. The physical exam does not always detect early acetabular dislocation, the acetabular index, and subluxation. The late diagnosis of dislocation is not an evidence that the physical test was not properly conducted. An increased acetabular index may provide the femoral head to run laterally out of the acetabulum and develop into a postponed dislocation. The late diagnosis of subluxated or dislocated hip is not necessarily associated with the neonatal subluxable or dislocatable hip.[3]

There is a high rate of false-negative clinical examinations, even when performed by experienced physicians.[21] To assess whether the result of the present study reflects the prevalence of the disease among the population of the São Paulo metropolitan area, further studies conducted in other centers and with larger samples are required.

Conclusion

The present study showed that the prevalence of ultrasound exams showing DDH in the sample was of 5.45%. The main risk factors associated with a higher risk of developing DDH were female gender, breech presentation, firstborn, and white ethnicity.

Author Contributions

GM: development of the instruments for data collection, data collection, initial analyses, conceptualization and study design, writing of the initial manuscript, and writing and revision of the final manuscript.
AC: review of the literature, and writing and revision of the manuscript.
EC: anályis and preparation of the data for the statistical analysis, and revision of the manuscript.
NV: contribution regarding the sections involving the clinical aspects of pediatric orthopedics and contribuition regarding the sections involving musculoskeletal radiology, and writing and revision of the manuscript.
MLD: writing and revision of the manuscript.
WI: conceptualization and study design, coordination and supervision of the data collection, writing of the initial manuscript, critical revision of the manuscript regarding important intellectual content, and writing and revision of the final manuscript.

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Conflict of Interests

The authors have no conflict of interests to declare.

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