Original Clinical Article

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Associations between risk factors and developmental dysplasia of the hip and ultrasonographic hip type: a retrospective case control study

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Abstract

Purpose We aimed to revisit the correlation between the previously defined risk factors and the occurrence of developmental dysplasia of the hip (DDH) and to assess the influence of these factors on the ultrasonographic type of hip dysplasia according to the Graf's classification in patients with DDH.

Methods Data of healthy infants (mean age 33 days) who had bilateral mature (normal) hips (Graf type I) were compared with the data of infants (mean age 105 days) who were treated by abduction brace due to unilateral or bilateral DDH (Graf type IIa- and worse hips).

Results Infants with at least one risk factor had a significantly higher rate of DDH than those with no risk factors ($p < 0.001$). Likewise, infants with more than one risk factor had a significantly higher rate of DDH than those with only one risk factor ($p = 0.008$). Family history, breech presentation and swaddling were found to be the three significant risk factors related to the development of DDH. Family history, swaddling and oligohydramnios were found to be the three significant risk factors correlated with a higher rate of unstable/decentred hip(s) (Graf types D/III/IV) in patients with DDH.

Conclusion The risk of DDH significantly increases in infants who have more than one risk factor for DDH. Positive family history and postnatal traditional swaddling are the two main factors both in the aetiology of DDH and in development of a more severe hip dysplasia in patients with DDH. Besides, breech presentation increases the risk of development of DDH and oligohydramnios leads to development of a more severe hip dysplasia in patients with DDH. By introducing these four variables as 'absolute risk factors for DDH' to the selective newborn hip screening programmes, the sensitivity and specificity of these programmes may be optimized and the risk of delayed diagnosis may be lessened.

Level of Evidence Level III prognostic study

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Keywords: developmental dysplasia of the hip; risk factors; hip ultrasonography; Graf method

Introduction

Multiple potential factors have been described in relation to developmental dysplasia of the hip (DDH). Positive family history, joint laxity, breech presentation, firstborn child, oligohydramnios, swaddling of the newborn, female gender, multiple pregnancy, ethnicity and coexistence of several orthopaedic conditions in infants such as torticollis and foot deformities have been considered as causative or coexisting conditions for DDH which have commonly been named as risk factors in the classical knowledge.[1-6]

Hip ultrasonography is currently the most widely used method in the definitive diagnosis of DDH in the first six months of life.[1,2,6-8] The risk of brace treatment failure in DDH increases in infants with sonographically dislocated hips.[9-12] Thus, the initial ultrasonographic type of hip dysplasia is an important determinant of the outcome in conservative treatment of DDH in infants.

The correlation between DDH and different risk factors has been assessed, and inconsistent conclusions have been reported for many years. Although, the influence of various causative factors on the occurrence of DDH has widely been accepted, it is still not possible to put precisely one or more of these factors forward as the main cause(s) of DDH due to heterogeneity of the patient data and methodologies in different studies. Besides, to our knowledge the correlation between the risk factors and the ultrasonographic type of hip pathology in patients

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with DDH has not been assessed yet, therefore, this is the very first study that aims to focus on this issue.

In this retrospective case-control study including univariate and multivariate analysis, we aimed to revisit the association between the previously defined risk factors and occurrence of DDH and to assess for the first time the correlation between the risk factors and the ultrasonographic type of hip dysplasia, which had been graded as stable/dysplastic and unstable/decentred, in patients with DDH.

Materials and methods

All procedures performed in this study were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. An institutional ethics board approval was obtained prior to the study. The data presented in this study was collected from the newborn hip screening programme of the Department of Orthopaedics and Traumatology at the Eskişehir Osmangazi University Hospital.

Residents in the Orthopaedics and Traumatology department obtained a detailed history for DDH from the infants' parents prior to the ultrasonographic hip examination by Graf's method.[7] Family history (first and second degree relatives), breech presentation at the third trimester, history of postnatal traditional swaddling (wrapping the newborn for a certain period of time with the hips in extension and adduction and knees in extension), foot deformities (including metatarsus adductus, idiopathic and postural club foot, pes calcaneovalgus), torticollis, multiple pregnancy, oligohydramnios (diagnosed by an obstetrician) and the firstborn baby were initially defined and recorded as 'risk factors for DDH'. Since the beginning of this newborn hip screening programme, firstborn baby and female gender have been gathered in one risk group named 'firstborn girl' to assess whether or not this coexistence has additive effects on the aetiology. So, the female data in the control and study groups was used to assess this variable. As the number of infants having foot deformities or torticollis or history of multiple pregnancy was very limited in the study group, we later decided to gather these three factors in one group named 'intrauterine packing' in order to obtain more reliable statistical conclusions. The clinical examination data of the infants was not included in the study as hip ultrasound was used to make to the definitive diagnosis as well as to avoid bias as different physicians performed the clinical examinations.

All ultrasonographic hip examinations were made by the senior authors (HÖ and NK) or under the strict supervision of them using the examination principles of Graf[7] with the infants lying in lateral decubitus position in a special cradle. The first senior author (HÖ), who was blinded to the history of the babies assessed all the magnified ultrasonographic image printouts, made the measurements by the same special hip ultrasound goniometer, and classified the hips according to the 'Graf's hip ultrasonography classification system'.[7]

The data of healthy consecutive infants with sonographically bilateral mature (normal) hips (Graf type I) who were under the age of 45 days (control group) were compared with that of consecutive infants who were treated by any kind of abduction brace due to unilateral or bilateral sonographically documented DDH (Graf type IIa- and worse hips) under the age of six months (study group). All the patients in the study group had no previous history of DDH treatment and the very first DDH diagnosis of these patients was made on the day of their hip ultrasound examinations. Infants over the age of 45 days were not included in the control group in order to exclude the immature hips (Graf type IIa), which may, without any intervention, usually complete the maturation process within the first three months of life but occasionally become true dysplastic after three months of age. Thus, all the infants in the control group had sonographically bilateral mature hips in the first six weeks of life. Patients with neuromuscular or teratologic hip dislocations were not included in the study group.

Sonographically pathological hips were initially classified as 'stable/dysplastic' (Graf types IIa-, IIb, IIc) and 'unstable/decentred' (Graf types D, III, IV) according to Graf's classification system.[7] Patients with unilateral or bilateral stable hips were included in the 'stable/dysplastic group'. Patients with unilateral or bilateral unstable hip(s) and patients having bilateral involvement with one side unstable and contralateral side stable were included in the 'unstable/decentred group'.

SPSS 22.0 (IBM Corporation, Armonk, New York) was used for data analysis. The correlation between the previously mentioned risk factors and development of unilateral or bilateral DDH; and the correlation between the risk factors and ultrasonographic type of hip dysplasia in patients with DDH were assessed. The chi-squared test was used to assess each risk factor independently (univariate analysis) and the backward stepwise regression analysis was used to assess the risk factors concomitantly (multivariate analysis). A p-value less than 0.05 was considered significant.

Results

The control group included 760 infants (377 girls, 383 boys) with a mean age of 33 days (SD 7; 4 to 45). The study group included 192 infants (154 girls, 38 boys) with a mean age of 105 days (SD 43; 21 to 186). The affected

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side was unilateral in 116 patients (41 right and 75 left) and bilateral in 76 in the study group. Stable and unstable groups included 141 and 51 infants, respectively.

Infants with at least one risk factor had significantly higher rate of DDH than those with no risk factors ($p < 0.001$) (Table 1). Likewise, infants with more than one risk factor had significantly higher rate of DDH than those with only one risk factor ($p = 0.008$) (Table 1).

Infants with positive family history or breech presentation or swaddling had a significantly higher rate of DDH than those without any such history (Table 2). Oligohydramnios or firstborn girl were not correlated with the development of DDH (Table 2). Infants with signs of intrauterine packing had a significantly lower rate of DDH than those without signs of intrauterine packing (Table 2). Thus, this factor was also considered not to be related to the development of DDH.

The multivariate analysis of the risk factors revealed that family history ($p < 0.001$; odds ratio (OR) 2.39; 95% confidence interval (CI) 1.47 to 3.89), breech presentation ($p = 0.015$; OR 1.90; 95% CI 1.13 to 3.18) and swaddling ($p < 0.001$; OR 2.87; 95% CI 1.75 to 4.69) were the three significant risk factors related to the development of DDH.

In the study group, the rates of family history, swaddling and oligohydramnios were higher in the unstable/decentred group compared with the stable/dysplastic group (Table 3). The multivariate analysis of the study group showed that family history ($p = 0.010$; OR 3.31; 95% CI 1.32 to 8.29), swaddling ($p = 0.015$; OR 3.16; 95% CI 1.25 to 7.98) and oligohydramnios ($p = 0.006$; OR 6.42; 95% CI 1.73 to 23.87) were the three significant risk factors correlated with a higher rate of unstable/decentred hip(s) in patients with DDH.

Discussion

There is a wide consensus on the fact that several risk factors are associated with DDH.[1-6] We initially aimed to revisit the maternal, foetal and postnatal causative factors and coexisting conditions for DDH both independently and concomitantly as well as to assess the effect of these

Table 1 Comparison of study and control groups concerning the presence or absence of risk factors for developmental dysplasia of the hip (OR, odds ratio; CI, confidence interval)

	Study group, n (%)	Control group, n (%)	p-value*/OR/95% CI
Infants without any risk factor	67 (35)	402 (53)	
Infants having at least one risk factor	125 (65)	358 (47)	p < 0.001/OR 2.10/95% CI 1.51 to 2.92
Infants having one risk factor	84 (67)	283 (79)	
Infants having more than one risk factor	41 (33)	75 (21)	p = 0.008/OR 1.84/95% CI 1.17 to 2.89

*Test used for p-values: Chi-square test

Table 2 Univariate analysis of the influence of risk factors on the development of DDH in the assessed population (OR, odds ratio; CI, confidence interval)

	Study group, n (%)	Control group, n (%)	p-value/OR/95% CI
Family history – Yes	30 (16)	54 (7)	
Family history – No	162 (84)	706 (93)	p < 0.001/OR 2.42/95% CI 1.50 to 3.91
Breech presentation – Yes	24 (13)	54 (7)	
Breech presentation – No	168 (87)	706 (93)	p = 0.015/OR 1.87/95% CI 1.12 to 3.11
Swaddling – Yes	30 (16)	47 (6)	
Swaddling – No	162 (84)	713 (94)	p < 0.001/OR 2.81/95% CI 1.72 to 4.59
Oligohydramnios – Yes	17 (9)	56 (7)	
Oligohydramnios – No	175 (91)	704 (93)	p = 0.489/OR 1.22/95% CI 0.69 to 2.16
Intrauterine packing– Yes	4 (2)	74 (10)	
Intrauterine packing – No	188 (98)	686 (90)	p = 0.001/OR 0.20/95% CI 0.07 to 0.55
Firstborn girl – Yes	67 (44)	164 (44)	
Firstborn girl – No	87 (56)	213 (56)	p = 0.999/OR 1.00/95% CI 0.68 to 1.46

Table 3 Univariate analysis of effects of the risk factors on the ultrasonographic type of hip dysplasia in patients with developmental dysplasia of the hip (OR, odds rato; CI, confidence interval)

	Unstable/decentred group, n (%)	Stable/dysplastic group, n (%)	p-value/OR/95% CI
Family history – Yes	14 (28)	16 (11)	
Family history – No	37 (72)	125 (89)	p = 0.007/OR 2.96/95% CI 1.32 to 6.62
Breech presentation – Yes	4 (8)	20 (14)	
Breech presentation – No	47 (92)	121 (86)	p = 0.241/OR 0.52/95% CI 0.17 to 1.59
Swaddling – Yes	14 (28)	16 (11)	
Swaddling – No	37 (72)	125 (89)	p = 0.007/OR 2.96/95% CI 1.32 to 6.62
Oligohydramnios – Yes	9 (18)	8 (6)	
Oligohydramnios – No	42 (82)	133 (94)	p = 0.018/OR 3.56/95% CI 1.29 to 9.82
Intrauterine packing– Yes	1 (2)	3 (2)	
Intrauterine packing – No	50 (98)	138 (98)	p = 1.0/OR 0.92/95% CI 0.09 to 9.05
Firstborn girl – Yes	18 (42)	49 (44)	
Firstborn girl – No	25 (58)	62 (56)	p = 0.798/OR 0.91/95% CI 0.45 to 1.86

factors on the ultrasonographic type of hip dysplasia in patients with DDH.

The main shortcoming of the present study is the lack of sample size analysis. Secondly, the limited number of patients in the unstable/decentred group may prevent us from drawing stronger statistical conclusions concerning this variable. Thirdly, the study group includes only four patients with a history of intrauterine packing (two torticollis and two metatarsus adductus) and the comparisons concerning this variable may be considered controversial. On the other hand, to our knowledge this is the very first study assessing the effect of several risk factors on the ultrasonographic type of hip dysplasia in infants with DDH. Besides, the control group was composed of healthy infants under the age of 45 days with bilateral mature hips and such an age restriction in the control group has never been taken into consideration in the previous studies. The aim of composing such a control group while planning the study has been to exclude the immature hips, which have a delayed maturation process and most likely become normal up to three months of age. It is well known that sonographically mature and immature hips have different natural histories. A sonographically mature hip at birth deteriorates in time only if the initial diagnosis is wrong or a neuromuscular disorder or a septic arthritis develops in the future.[7] However, 5% of sonographically immature hips can deteriorate over time.[13] So, it is better to include younger infants who are known to have bilateral mature hips at birth in the control group rather than to include the older infants who may initially have immature or even dysplastic hips, which then spontaneously resolve and become mature over time and are considered mature during ultrasound examination over three months of age.

Two meta-analysis articles have concluded that one or a combination of the following risk factors; breech presentation, female gender and family history are the three leading predisposing factors which are considerably correlated with the development of DDH.[14,15] Our findings have shown that the risk of developing DDH is two-times higher in infants with at least one risk factor, compared with infants with no risk factors. Likewise, among infants with at least one risk factor for DDH, the risk of developing DDH is two times higher in infants with more than one risk factor, compared with infants with only one risk factor. We can say that more attention should be paid to infants with any risk factor(s), especially to those with more than one predisposing factor in selective newborn hip screening programmes.

Genetically inherited abnormal hip development and/or joint laxity has commonly been accepted as one of the main causes of DDH.[1-6] It has been reported that there is a 12-times increased risk for developing DDH if a newborn has a first-degree relative with DDH.[16] In addition to that, infants with positive family history for DDH have been found to have a slightly higher rate of failure in brace treatment than those without family history for DDH.[12] We have not made a separate analysis of the correlation between a positive history of DDH in first-degree relatives and development of DDH in the present study. Besides, we have not assessed or recorded whether or not the babies have hypermobile joints. However, our results have revealed that infants with a positive history of DDH in their first- or second-degree relatives have about a 2.5-times higher risk of developing hip dysplasia requiring treatment. In addition, we have found that infants with a diagnosis of DDH accompanied by a positive family history for DDH have about a 3.5-times higher risk of developing sonographically unilateral or bilateral unstable/decentred hips. We can argue that acetabular dysplasia and/or joint laxity may be more severe in patients with a positive family history for DDH, and these genetically inherited severe anatomical alterations may explain why higher rates of impaired acetabular development and the lateral and upward displacement of the femoral head are observed in such patients.

Traditional swaddling can cause a mechanical stress on the hips that may be followed by lateral and upward displacement of the femoral head and disruption of the acetabular development. In a rat model, straight-leg swaddling has been shown to have harmful effects on infant hips, especially in those which have been swaddled at a younger age, and those which have undergone prolonged swaddling.[17] Limited number of clinical studies have emphasized the fact that traditional swaddling significantly increases the risk of DDH.[18,19] Besides, it has been reported that the success rate of Pavlik harness treatment is slightly lower in previously swaddled infants than in not previously swaddled infants.[10] Traditional swaddling of infants is still applied by some parents in the geographic area where the present study has been conducted, so this study presents a considerable data on this variable. Our results have shown that infants with a postnatal traditional swaddling history have about a three-times higher rate of DDH that requires treatment. Besides, previously swaddled patients with DDH have about a three-times higher risk of having sonographically unilateral or bilateral unstable/decentred hip(s). However, this study does not present detailed data concerning the initiation age and duration of swaddling in these infants. Swaddling may provide a safe environment for the infant to sleep, but it is important to allow ample room for the hips and knees in a swaddle to avoid DDH.[20] We believe that, in order to provide correct advice to the physicians, parents and caregivers on healthy swaddling practices, the positive effects of allowing lower limb movement and the negative effects of positioning the lower limbs together in a straight position should clearly be emphasized.

Breech presentation carries a considerable risk for DDH.[1-6] The risk of DDH is the highest in frank breech

presentation (one or both knees extended) and this is probably due to the pull of the hamstrings across the flexed hip.[2] Breech presentation may also be related to the increased risk of failure in brace treatment in infants.[12] It has also been reported that singleton breech infants delivered by a caesarian section have a significantly lower risk of DDH than those delivered vaginally.[21] Although we have initially not documented the type of breech presentation and the mode of delivery, we have observed that infants with breech presentation have about a two-times higher risk of DDH. However, breech presentation does not have any effect on the ultrasonographic type of hip dysplasia in infants with DDH.

A decrease in the volume of the amniotic fluid increases the pressure on the foetus, restricts foetal mobility and prevents the foetus from changing its position. Such a mechanical alteration may have an adverse effect on the intrauterine development of the hip.[4-6] Our findings have shown that history of oligohydramnios itself does not increase the risk of DDH. However, if a patient with DDH has a history of oligohydroamnios, then the risk of developing sonographically unilateral or bilateral decentred hip is about six times higher. We think that such an effect cannot directly be associated with oligohydramnios itself, but with some other genetic and/or mechanical predisposing factors as well.

Female gender and firstborn baby are the two known predisposing factors for DDH.[1-6] Our findings have shown that being the firstborn female baby is not a predisposing factor for DDH and also not a determinant of the type of hip dysplasia.

Postural or rigid foot deformity, torticollis and multiple pregnancy have been considered to correlate with the development of DDH.[1-6] However, our limited data about these variables in the study group prevents us to draw stronger conclusions.

We can conclude that several factors are associated with the development of DDH in the screened infant population of the present study and with the ultrasonographic type of hip pathology in infants with DDH, which has previously not been reported. The results of the present study may help to improve the selective newborn hip screening protocols. It is better to perform universal newborn hip screening to avoid late detection and to lessen the rate of surgery in DDH.[22] However, selective newborn hip screening is still used in many parts of the world. If an infant has at least one risk factor for DDH, then the risk of DDH significantly increases, and this increase is more evident in infants with more than one risk factor. So, infants with more than one risk factor may require the highest attention in selective newborn hip screening programmes. We can put forward family history and postnatal traditional swaddling as the two main causative factors for DDH and also two predisposing factors in development of a sonographically more severe hip dysplasia (Graf type D, III, IV hips) which has been known to correlate with an increased risk of failure in brace treatment in DDH. Besides, breech presentation increases the risk of development of DDH and oligohydramnios leads to development of a sonographically more severe hip dysplasia (Graf type D, III, IV hips) in patients with DDH. We think that the above-mentioned four causative factors can be introduced as 'absolute risk factors' to selective newborn hip screening programmes. To us, it is really indicated that the infants having at least one or more of these four significant risk factors should be referred to ultrasonographic hip examination to optimize the sensitivity and specificity and to lessen the risk of delayed diagnosis as much as possible in selective newborn hip screening programmes.

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COMPLIANCE WITH ETHICAL STANDARDS

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ETHICAL STATEMENT

Ethical approval: All procedures performed in this study were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. An institutional ethics board approval was obtained prior to the study.

Informed consent: Not required.

ICMJE CONFLICT OF INTEREST STATEMENT

All the authors declare that they have no conflict of interest.

AUTHOR CONTRIBUTIONS

HÖ: Designed the study, carried out the clinical works, contributed in analyzing the data, wrote the paper, provided critical feedback, gave final approval of the version to be submitted.

AA: Collected the data, contributed in analyzing the data, contributed in drafting the paper, provided critical feedback, gave final approval of the version to be submitted.

NK: Carried out the clinical works, contributed in analyzing the data, contributed in drafting the paper, provided critical feedback, gave final approval of the version to be submitted.

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