

Is There an Association Between the Familial Form of Generalized Joint Hypermobility and Developmental Dysplasia of the Hip in Children?

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Abstract

Background and Objectives: This study aimed to examine the association between the familial generalized joint hypermobility (GJH) and developmental dysplasia of the hip (DDH) in children. **Materials and Methods:** All infants and children included in this case (with DDH)–control study were examined for features of GJH by the revised Beighton's Test Scale (BTS), provided that they had no teratogenic or pathological causes of DDH or GJH. **Results:** Two hundred and eight participants were included in this study: 101 (48.6%) cases and 107 (51.4%) controls; age mean 16.57 [standard deviation (SD) 18.1] months; 61.1% (127) females and 38.9% (81) males. The odds ratio was 2.1 (95% confidence interval 1.197–3.679). The cases had a higher value of BTS score (mean = 4.7, SD = 3.015) when compared with the controls (mean = 3.42, SD = 3.448). The difference was statistically significant, but the correlation coefficient (r) between the study groups and BTS scores was weak ($r = 0.194$, $P = 0.005$). The correlation gained slight improvement by justification of the age to obtain homogeneity between the groups ($r = 0.276$, $P < 0.001$), but deteriorated and lost its significance when the sex was controlled as a confounder ($r = 0.121$, $P = 0.082$). There was a gradual increase in the mean BTS score from grade 1 to grade 4 of DDH but without a statistically significant value ($r = 0.132$, $P = 0.128$). **Conclusion:** The familial GJH has a weak positive correlation with DDH but cannot be considered as a possible risk factor for it.

Keywords: Association, developmental dysplasia of the hip, generalized joint hypermobility

INTRODUCTION

Developmental dysplasia of the hip (DDH) has a wide spectrum of abnormalities ranging from mild acetabular dysplasia to irreducible dislocation.^[1] The exact etiology is unknown but suspected to be multifactorial. It may be genetic, hormonal, or mechanical whether intrauterine (positioning and packaging abnormalities) or postnatal factors.^[2-4]

The pathological changes involve both bony parts (especially acetabular dysplasia) and surrounding soft tissue elements (especially capsule-ligament structures). Most of the studies outweigh the acetabular dysplasia to be the primary changes in the pathogenesis of DDH.^[2,5-8]

The capsule-ligament structures in DDH undergo stretching and laxity. This laxity is claimed to occur from the abnormal mechanical loading on the joint, but

genetics (as in some families), hormonal (as in females), and some connective tissue disorders (as in the teratogenic form of DDH) are also incriminated.^[1,3]

Ligament laxity may cause joint laxity or joint hypermobility (which may be localized or generalized). The generalized joint hypermobility (GJH) may be inherited either in a familial form which is usually asymptomatic and present in healthy persons or is part of certain chromosomal or connective tissue disorders.

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Also, it may be acquired from widespread inflammatory or endocrinal disorders.^[9-13]

It may be noticed that patients who have the typical type of DDH may also have clinical features of GJH in their extremities. In the literature, it is not clear whether excessive ligament laxity due to the familial GJH has a role as a risk factor (etiology) of DDH or not.

Aim of the study

This study aimed to examine the association between the familial GJH and DDH in children.

Objectives

1. How many patients with DDH also have features of GJH (proportion and percentage)?
2. How many persons (especially children) who had features of GJH have also DDH (proportion and percentage)?
3. Is there a correlation between the presence of GJH features and the occurrence of DDH?
4. Could the GJH be a risk factor for DDH?

Hypothesis

- *Null hypothesis (Ho):* There is no correlation between DDH and GJH.
- *Alternative hypothesis (Ha):* There is a correlation between DDH and GJH.

MATERIALS AND METHODS

Approvals

The approvals to perform this study were gained from the Institutional Scientific Committee and the Local Health Authority/Committee of Ethics for medical researches.

Study type, source population, and sittings

The study was an unmatched case-control observational type. The children who attained three medical places (Early Detection of Childhood Disability Center, Heevi Pediatric Teaching Hospital, and the Emergency Teaching Hospital) in Duhok city/Iraq during the period from October 1, 2020 to January 30, 2021 were chosen as a source population for this study. The children attained these medical health centers for different complaints including musculoskeletal problems. The sampling was done in the outpatient clinics of these centers.

Participants, sampling, and eligibility

To achieve a sort of random selection of the participants, the sampling was done every other day and every other patient from the waiting list.

The inclusion criteria were infants and children whose ages ranged from 6 months to 12 years (infants less than

6 months might have the residual effect of the maternal hormone on their ligaments)^[14] of both sex and Kurdish race only. Exclusion criteria were as follows:

- Children with syndromic disorders which may cause the teratogenic type of DDH or non-familial cause of GJH (such as arthrogryposis, Ehlers–Danlos syndrome, Marfan's syndrome, Down's syndrome, and osteogenesis imperfecta);
- Children with the acquired cause of GJH (such as juvenile rheumatoid arthritis, hypothyroidism, etc.);
- Children with neuromuscular disorders (such as meningomyelocele, cerebral palsy);
- Children with a history of pyogenic arthritis of their hip during the neonatal period;
- Children with possible confounders such as the first baby in the family; a history of breech presentation; a history of oligohydramnios; a history of swaddling.

The eligible participants were checked for evidence of DDH and GJH. Those who had clinical and radiological evidence of DDH were considered as cases and those without this evidence were considered as controls. All the included children (of both groups) were examined for features of GJH. In this study, the presence of GJH features was considered as an exposure (possible risk factor) and the presence of DDH as the outcome (the disease).

Data sources, variables, and measurements

After taking permission from the parents with a signed concept, the data of each patient were reported in a special worksheet paper with a code.

The data included age, gender, race, evidence of DDH, side of DDH, grade of DDH (according to the Tönnis Classification—four grades),^[15] and finally the score of GJH by using the revised Beighton's Test Scale (BTS) for joints hypermobility in children (which evaluates five parts of the body bilaterally and the total score was recorded between 0 and 10, a score of 5 and more was considered to have GJH).^[16] This revised form of BTS was suggested by Romeo *et al.* for the younger preschool children (less than 5 years). It is a mixture between the classical BTS and that described by Carter *et al.*^[14,17] It replaces putting the palms flat on the ground (a step which may be difficult to be performed by the toddlers and impossible by the infants) by dorsiflexion of ankles. It was found to be suitable for the present study because most of the participants suspected to be included would be young children (infants and toddlers).

Sample size calculation

The assumed sample size was calculated by using the G*Power 3.1.9.7 computer software program. A minimum of 138 participants (69 for each group) was required to have a power of 95% chance of detecting the association between GJH and DDH and a two-sided 95% level of confidence (0.05 level of significance) to avoid the type 1 alpha error, with a probable

hypothetical correlation of about 0.3. However, during the sampling process, the number of cases and controls was increased to overcome the justification for homogeneity of other characteristics such as age. This increase was aimed also to diminish and minimize the random error and sampling error and to narrow the margins of error during estimation of the confidence interval (CI) bounds.

Statistical methods

The collected data were transferred to a computer by using the Statistical Package for Social Sciences software program (IBM Corp., Released 2015, IBM Corp., IBM SPSS Statistics for Windows, Version 23.0, Armonk, NY, USA) for further statistical analysis with a 2×2 contingency table. The following estimations and analysis were done:

- To know how many cases of DDH also had features of GJH, the proportion and the percentage were calculated by dividing the number of DDH cases who had GJH by the total number of DDH cases, and then the result was multiplied by 100.
- To know how many participants with GJH also had DDH, the proportion and percentage were calculated by dividing the number of participants with GJH and had DDH by the total number of participants with GJH, and then the result was multiplied by 100.
- To examine the association between DDH and GJH, the following statistical analytic tests were conducted:
 1. First, to know whether there was a relationship between them or not, the odds ratio (OR) was estimated by dividing the odds of cases with GJH by the odds of controls. The relationship was considered to be present when the OR was more or less than 1 and considered absent when it was equal to 1. The CI was calculated at a 95% level of confidence.
 2. Secondly, to determine the direction and strength of the relationship (correlation) between them, a correlation coefficient (CC) was calculated according to the type of data:
 - The Phi CC was used when the data of both the exposure and outcome were of nominal binary dichotomous (two levels) type.
 - Goodman and Kruskal lambda CC or Pearson's χ^2 tests were used when the data of both exposure and outcome were nominal but not dichotomous (more than two levels).
 - The point-biserial correlation was done by using Pearson's CC if one variable was nominal and the other was continuous numerical scale type.
 - The rank-biserial correlation was done by using Spearman's CC when one variable was of the ordinal type and the other one was of the nominal type.
 - Kendall's tau_b CC was used when one variable was ordinal and the other one was of continuous numerical scale type.

- Partial correlation was done to control the effect of confounder (gender).

The value of the CC (r) was used to interpret the direction of the correlation (positive or negative values) and the strength of the correlation according to the recommended guidelines of Cohen (less than 0.3 has a weak relationship, between 0.3 and 0.5 has a moderate relationship, and more than 0.5 has a strong relationship).^[18]

3. To control the effect of the confounders (especially for gender), the partial correlation was used.
4. For further confirmation of the confounder control and to make a possible prediction for the occurrence of DDH from the BTS score, the binary logistic regression was conducted.
5. An independent t -test was used to compare the means of variables when they were of numerical scale type.
6. The result of any statistical analysis test was considered statistically significant, and the null hypothesis was rejected when the P -value (P) was less than 0.05.

Bias, chance (random error), and confounders

To prevent or minimize an incorrect estimation of the association between GJH and DDH, the following points were taken into consideration to prevent bias during the conduction of the study: the selection criteria (inclusion criteria) of participants' characteristics for both groups (cases and controls) were unified especially for age; the same method for measurements and classifications was used for all participants (of both groups) for diagnosis and grading DDH and GJH to prevent misclassification bias.

To minimize the probability of detection of the association by chance, the following points were taken into consideration: the sampling of the participants (especially for controls) was done randomly according to the presentation of the participants to these centers, which was beyond the control of the authors; the sampling was done for children who presented to these medical centers for different complaints (not for musculoskeletal problems only especially during sampling of controls); the sample size was increased beyond the estimated one to minimize the random error (i.e., detection of the finding by chance); the confidence level was set at 95% of detection, and the P -value was set at 0.05.

The following points were taken into consideration to control the effect of confounders: the eligibility of the participants was restricted—for those with possible confounder effect—in the exclusion criteria; the statistical analysis included partial correlation and logistic regression to control the effect of gender as a confounder.

RESULTS

Two hundred and eight participants were included in this study: 101 (48.6%) were children who had DDH

and they were considered as cases, whereas the rest 107 (51.4%) were considered as controls because they did not have DDH. Their age ranged from 6 months to 11 years with a mean of 16.57 and a standard deviation (SD) of 18.1 months. The participants were mainly females 61.1% (127) vs. 38.9% (81) males. The details of differences between the cases and controls were summarized in Table 1.

The percentage of the cases (had DDH) with features of GJH was 50.5% among whole cases. The percentage of those with GJH features among the cases was 59.3% and among the controls was 32.7%. The OR was 2.1 with 95% CI bounds (1.197–3.679) [Table 2].

The study groups had a statistically significant correlation with the GJH features (Phi CC = 0.180, $P = 0.009$). The cases had a higher value of BTS score (mean = 4.7, SD = 3.015) compared with the controls (mean = 3.42, SD = 3.448). The difference was statistically significant when their means were compared by the independent t -test with equal variance assumed (t -value = 2.849, $P = 0.005$). The point-biserial correlation between the study groups and the BTS score also had a statistically significant value ($r = 0.194$, $P = 0.005$) [Figure 1].

The age variable had several extreme values (outliers) which caused a high SD. After justification of the data regarding the age variable (by deleting the outliers) to obtain homogeneity of the groups, there were a mild rise in the OR (2.7, 95% CI 1.46–5.01), Phi CC between study groups and GJH features ($r = 0.236$, $P = 0.001$), and between study groups and BTS scores ($r = 0.276$, $P < 0.001$).

When partial correlation was performed to control the effect of sex as a confounder, the correlation of the study groups with GJH features was diminished and became non-statistically significant ($r = 0.108$, $P = 0.123$). The same thing occurred for its correlation with the BTS score ($r = 0.121$, $P = 0.082$).

Logistic regression was conducted to predict the log odds of DDH from the BTS score. There was a statistically significant rise in the likelihood of DDH

log odds by 1.129 (95% CI 1.036–1.229) when the BTS score increased by one point. This prediction became non-significant when the sex variable holds constant (controlled as a confounder). The likelihood of DDH log odds became 1.083 and the CI bounds became 1 (95% CI 0.990–1.185) with the increase of the BTS score by one point [Table 3].

The cases (those who had DDH) were 101. Twenty-four (23.8%) of them were males and the other 77 (76.2%) were females. The mean age was 12.19 (SD 6.2) months. The type of DDH according to the side involved was as follows: right-sided 41 (40.6%), left-sided 16 (15.8%), and bilateral 44 (43.6%). The severity (grades) of DDH was as follows: Grade 1 20 (19.8%), Grade 2 75 (74.3%), Grade 3 5 (5%), and Grade 4 1 (1%). The cases that had features of GJH were 51 (50.5%) and those who did not have these features were 50 (49.5%). The mean of BTS score for GJH features among the participants of this group was 4.7 (SD 3.015). There were no statistically significant results from the correlation tests, which had been conducted between the different variables of this group [Table 4]. There was a gradual increase in the mean BTS score from grade 1 to grade 4 of DDH but without a statistically significant value ($r = 0.132$, $P = 0.128$) [Figure 2].

DISCUSSION

DDH and ligamental laxity

The shoulder and the patellofemoral joints have a preliminary deficiency of their bony restrained function. Their primary stabilizers are the capsule-ligamental structure and the surrounding muscle tone. These joints sustained recurrent instability when the ligaments become lax or lost their integrity. Contrary to them, the bony arrangement is an important stabilizer for the hip. Acetabular dysplasia is thought to be the primary pathology in DDH. Capsulo-ligamental stretching and laxity with subsequent femoral head displacement are supposed to be secondary changes.^[6,8] It might be unusual to find a hip with a typical type of DDH (in a healthy child) with a normal acetabulum unless the familial form of GJH is to be blamed for that. Hence, it was interesting to examine the relationship between the presence of GJH

Table 1: Differences between both study groups

Participants characteristic	Study groups		Total
	Cases (have DDH*) $n^{\dagger} = 101$	Controls (do not have DDH) $n = 107$	
Gender (frequency)			
Female	77	50	127
Male	24	57	81
Age in months [mean (SD [‡])]	12 (6)	21 (24)	16.57 (18.1)

*DDH = developmental dysplasia of the hip

[†] n = number

[‡]SD = standard deviation

Table 2: Study groups with GJH features

		Study groups		Total
		Cases (have DDH [†]), n [‡] (%)	Controls (do not have DDH), n (%)	
GJH* features	With	51 (59.3%)	35 (32.7%)	86 (100%)
	Without	50 (41%)	72 (59%)	122
Total		101	107	208

*GJH = generalized joint hypermobility

[†]DDH = developmental dysplasia of the hip

[‡]n = number

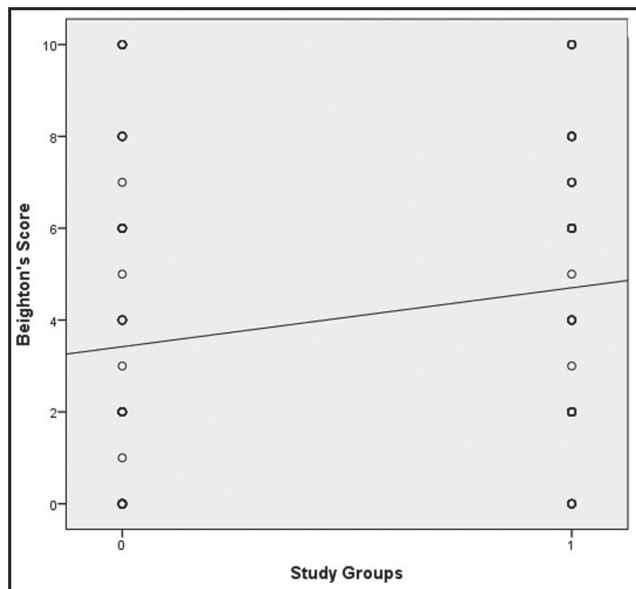


Figure 1: Scatterplot with a fit line at total (line of trend) for point-biserial correlation examination of Pearson's CC between the study groups and the BTS scores (0 = controls, 1=cases)

features and the occurrence of the typical type of DDH, excluding list-wise the pathological causes of GJH.

Association

The percentage of those with GJH features among cases (59.3%) and controls (32.7%) was more than that reported by Carter and Wilkinson,^[14] but less than that reported by Wynne-Davies,^[19,20] Muldoon *et al.*,^[21] and Santore *et al.*^[22] However, the former study calculated only proportions and percentages without any statistical inferential analysis to test a hypothesis, and the latter two articles included adult participants only.

In this study, the odds of DDH with GJH were 2.1 times the odds of those without GJH features. In other words, there were greater (2.1 times) odds of the DDH happening in those with GJH features vs. those without. This result was considered statistically significant because the CI did not include the value of 1 within its bounds (95% CI = 1.197–3.679). The presence of association was confirmed and the role of chance was excluded. So, it could be concluded that with repeated sampling the possibility of detecting DDH in those with GJH features would be between 1.2

and 3.7 (average 2.1) times greater than those without these features in about 95% of the samples.

Correlation

The results of the present study showed a weak positive but statistically significant correlation between the study groups and the presence of GHJ features ($r = 0.18$, $P = 0.009$). After justification of the data to obtain homogeneity between the study groups, the new results showed a minimal rise of the OR and CC, but still within the weak zone of correlation according to the Cohen-recommended guidelines. The authors sympathized to keep the data as such without justification because the extreme values (outliers) of the samples—especially for the age—were still within the inclusion criteria, which had been settled before conducting the sampling procedure.

There was a gradual increase in the scores of joint hypermobility (BTS), and there was a slope in the fit line of the trend when moving from the group of controls to cases [Figure 1]. That is to say, the cases with DDH had a higher degree of GJH features than the controls. The difference was mild and the correlation was weak but was statistically significant. These results encouraged the rejection of the null hypothesis and made the alternative one more favorable.

Risk factor

Wynne-Davies and Dunn differentiated two types of DDH: the first group had GJH (from hormonal, genetic, or constitutional factors) and manifests as hip instability at birth and the second group is characterized by dysplasia of the acetabulum without any significant ligamentary laxity.^[19,23] As mentioned before, acetabular dysplasia is now thought to be the primary pathology in DDH, and the capsulo-ligamentary stretching and laxity with subsequent femoral head displacement are supposed to be secondary changes.^[6,8]

It is unusual in DDH cases to find a displaced hip with the normal acetabulum. For example, in Graf's evaluation for hips, all types which have an increase in the beta angle also have diminished alpha angle (indicating that the acetabulum is dysplastic) apart from what is called the “elastic whipping” phenomenon.^[24] But, the acetabular growth and maturation were not finished yet by the end of the first year of life.

Table 3: Variables entered in the regression equation

Variable(s) entered		B*	SE†	Wald‡	df§	Sig.**	Exp(B)††	95% CI†† for Exp(B)	
								Lower	Upper
Step 0	Constant	-0.058	0.139	0.173	1	0.677	0.944		
Step 1	BTS score	0.121	0.044	7.693	1	0.006	1.129	1.036	1.229
Step 2	Constant	-0.547	0.226	5.873	1	0.015	0.578		
	BTS score§§	0.080	0.046	3.034	1	0.082	1.083	0.990	1.189
	Sex (1)	-1.167	0.313	13.942	1	0.000	0.311	0.169	0.574
	Constant	0.056	0.280	0.040	1	0.841	1.058		

*B = values for logistic regression equation (in log odds units)

†SE = standard error associated with the coefficients

‡Wald = Wald χ^2 value

§df = the degree of freedom for each test of the coefficients

**Sig. = two-tailed P-value

††Exp(B) = odds ratios for the predictors (exponentiation of the coefficients)

‡‡CI = confidence interval

§§BTS: Beighton's Test Scale

Table 4: Correlation tests between the different variables of the group of cases

Variables		Correlation coefficient		P-value
Variable 1 (type)	Variable 2 (type)	Type	Value	
DDH* sides (nominal)	GJH† groups (binary dichotomous)	Goodman and Kruskal lambda coefficient	0.022	0.108
		Pearson χ^2 test	4.733	0.094
DDH sides (nominal)	BTS's score‡ (scale)	Point-biserial correlation by Pearson's CC	0.176	0.078
DDH grades (ordinal)	GJH groups (binary dichotomous)	Rank-biserial correlation by Spearman's CC	-0.174	0.082
DDH grades (ordinal)	BTS score (scale)	Kendall's tau_b	0.132	0.128

*DDH = developmental dysplasia of the hip

†GJH = generalized joint hypermobility

‡BTS = Beighton's Test Scale

When the walking commences, the shearing force between the femoral head and the acetabulum might cause micro-instability and acetabular maturation defects if the capsule-ligament structures are not tight enough. Muldoon *et al.*^[21] and Santore *et al.*^[22] found that hypermobility was an important factor for hip dysplasia and pain in adults. Devitt *et al.*^[25] found a significant negative correlation between the presence of GJH and the thickness of the hip joint capsule. They proposed that thinning of the capsule in those with GJH may weaken the static stabilizer of the hip joint and initiate micro-instability. The same scenario may occur during fetal intrauterine life. Here, the ligament laxity would be the primary defect, and the acetabular changes would be the secondary one.

Although up to date the magnitude of ligament laxity (not joint laxity) was not been measured objectively, the ligament laxity from pathological causes (such as in Ehlers-Danlos syndrome) may not be equivocal to that of familial GJH. The present study looked for an association between the familial GJH and typical DDH and excluded the teratogenic and possible pathological risk factors for both DDH and GJH.

The results of the present study showed that the correlation between familial GJH and BTS score with DDH diminished

and became statistically not significant when the effect of the sex as a confounder was controlled. It could be concluded that sex masked the real association. So, the familial GJH could not be incriminated (according to the results of the present study) to disturb the stability of the hip joint. The results of the mentioned studies (Muldoon *et al.*, Santore *et al.*, and Devitt *et al.*) probably did not come from cases with the familial GJH. So, it can be concluded that familial GJH could not be considered as a possible risk factor for secondary acetabular dysplasia (DDH) and hip pain in adults. The field now became far away from the discussion about the possibility of causality in this association (cause-effect relationship).

BTS scores and DDH grades

When examining the data of the cases only, the degree of BTS score was gradually increased when the grade of DDH increased. This could be noticed easily from the positive value of the CC ($r = 0.132$) and the slope of the fit line of the trend in the scatterplot diagram [Figure 2]. However, this correlation was considered weak and it did not assume the significance level ($P = 0.128$). So, it could not be accepted that the raise of the BTS score had a role in raising or determining the grade of DDH. Therefore, the authors of this study were not encouraged to do the

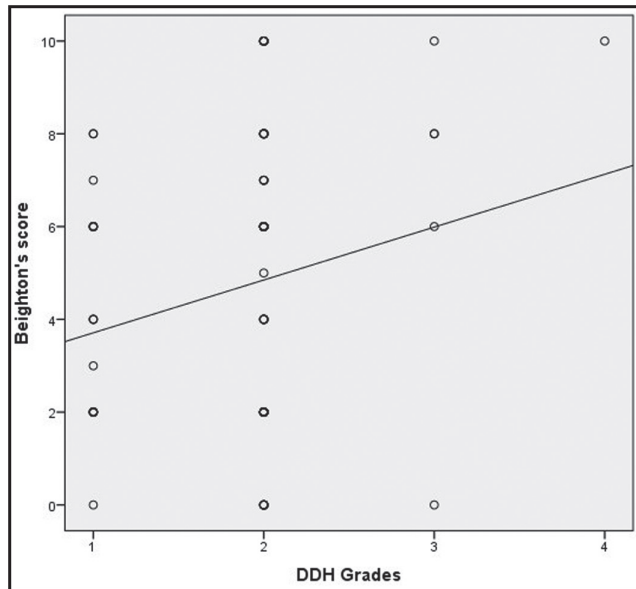


Figure 2: Scatterplot with a fit line at total (line of trend) for the grades (severity) of DDH and BTS score

regression analysis to predict the raise of DDH grade from the raise in the score of the BTS score.

It can be noticed that the values of all the CCs which had been counted between the different variables in this study were low and considered weak according to Cohen's recommended guidelines.

CONCLUSION

The familial GJH has a positive weak correlation with DDH but cannot be considered as a possible risk factor for it.

Limitations

The sampling of the participants was from the children who attained the outpatient clinics of some medical centers (hospital-based source). None of the participants (especially controls) was chosen from schools or public places (general population-based source) because the study was conducted during the crisis of Covid-19.

For comparative purposes, the authors of the present study did not find satisfying recent articles or studies discussing the association between the familial forms of GJH and DDH during literature reviews.

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Conflicts of interest

There are no conflicts of interest.

REFERENCES

- Gargan M, Blom A, Jones SA, Behman A, Kelley S. The hip. In: Blom A, Warwick D, Whitehouse M, editors. Apley & Solomon's

- System of Orthopaedics and Trauma. 10th ed. Boca Raton, FL: CRC Press, Taylor & Francis Group; 2018. p. 531-6.
- Wright J, James K. Developmental dysplasia of the hip. In: Aresti N, Ramachandran M, Paterson M, Barry M, editors. Paediatric Orthopaedics in Clinical Practice. London: Springer; 2016. p. 69-90.
- Herring JA. Developmental dysplasia of the hip. In: Herring JA, editor. Tachdjian's Pediatric Orthopaedics: From the Texas Scottish Rite Hospital for Children. 5th ed. Philadelphia, PA: Elsevier Saunders; 2014. p. 483-535.
- de Hundt M, Vlemmix F, Bais JM, Hutton EK, de Groot CJ, Mol BW, *et al.* Risk factors for developmental dysplasia of the hip: A meta-analysis. *Eur J Obstet Gynecol Reprod Biol* 2012;165:8-17.
- Kelly DM. Congenital and developmental abnormalities of the hip and pelvis. In: Canale ST, Beaty JH, Azar FM, editors. Campbell's Operative Orthopaedics. 13th ed. Philadelphia, PA: Elsevier; 2017. p. 1118-54.
- Shapiro F. Pediatric Orthopaedic Deformities, Vol. 2. Cham: Springer Nature Switzerland; 2019. p. 1-182.
- Storer SK, Skaggs DL. Developmental dysplasia of the hip. *Am Fam Physician* 2006;74:1310-6.
- Weinstein SL, Mubarak SJ, Wenger DR. Fundamental concepts of developmental dysplasia of the hip. *Instr Course Lect* 2014;63:299-305.
- Scheper MC, de Vries JE, Juul-Kristensen B, Nollet F, Engelbert RH. The functional consequences of generalized joint hypermobility: A cross-sectional study. *BMC Musculoskelet Disord* 2014;15:243.
- Engelbert RH, Bank RA, Sakkers RJ, Helden PJ, Beemer FA, Uiterwaal CS. Pediatric generalized joint hypermobility with and without musculoskeletal complaints: A localized or systemic disorder? *Pediatrics* 2003;111:e248-54.
- What is hypermobility spectrum disorders (HSD)? [The Ehlers-Danlos Society]. 2021. Available from: <https://www.ehlers-danlos.com/what-is-hsd/>. Accessed January 18, 2021.
- Castori M, Tinkle B, Levy H, Grahame R, Malfait F, Hakim A. A framework for the classification of joint hypermobility and related conditions. *Am J Med Genet C Semin Med Genet* 2017;175:148-57.
- Tofts LJ, Elliott EJ, Munns C, Pacey V, Silience DO. The differential diagnosis of children with joint hypermobility: A review of the literature. *Pediatr Rheumatol Online J* 2009;7:1.
- Carter C, Wilkinson J. Persistent joint laxity and congenital dislocation of the hip. *J Bone Joint Surg Br* 1964;46:40-5.
- Tonnis D. Congenital Dysplasia and Dislocation of the Hip in Children and Adults. Berlin: Springer-Verlag; 1987. p. 100-42.
- Romeo DM, Lucibello S, Musto E, Brogna C, Ferrantini G, Velli C, *et al.* Assessing joint hypermobility in preschool-aged children. *J Pediatr* 2016;176:162-6.
- Beighton P, Solomon L, Soskolne CL. Articular mobility in an African population. *Ann Rheum Dis* 1973;32:413-8.
- Cohen J. Statistical Power Analysis for the Behavioral Sciences. 2nd ed. New York: Routledge; 1988. p. 109-43.
- Wynne-Davies R. Acetabular dysplasia and familial joint laxity: Two etiological factors in congenital dislocation of the hip. A review of 589 patients and their families. *J Bone Joint Surg Br* 1970;52:704-16.
- Wynne-Davies R. Familial joint laxity. *Proc R Soc Med* 1971;64:689-90.
- Muldoon M, Gosey G, Healey R, Santore R. Hypermobility: A key factor in hip dysplasia. A prospective evaluation of 266 patients. *J Hip Preserv Surg* 2016;3(Suppl. 1):hnnw030.034.
- Santore RF, Gosey GM, Muldoon MP, Long AA, Healey RM. Hypermobility assessment in 1,004 adult patients presenting with hip pain: Correlation with diagnoses and demographics. *J Bone Joint Surg Am* 2020;102:27-33.
- Hefti F. Pediatric Orthopedics in Practice. 2nd ed. Berlin: Springer; 2015. p. 211-34.
- Graf R. Hip sonography: Background; technique and common mistakes; results; debate and politics; challenges. *Hip Int* 2017;27: 215-9.
- Devitt BM, Smith BN, Stapf R, Tacey M, O'Donnell JM. Generalized joint hypermobility is predictive of hip capsular thickness. *Orthop J Sports Med* 2017;5:2325967117701882.