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Assessing the risk of asymptomatic dysplasia in parents of children with developmental hip dysplasia

Tacettin Ayanoglu [a, *], Muhammet Baybars Ataoglu [b], Nil Tokgöz [c], Elif Ersöz [c], Hakan Atalar [b], Sacit Turanlı [b]

[a] Department of Orthopaedics and Traumatology, Yozgat City Hospital, Yozgat, Turkey
[b] Department of Orthopedics and Traumatology, Medical Faculty of Gazi University, Ankara, Turkey
[c] Department of Radiology, Medical Faculty of Gazi University, Ankara, Turkey

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ABSTRACT

Objective: The aim of this study was to investigate whether being the parents of children with developmental hip dysplasia (DDH) is a risk factor for asymptomatic dysplasia.
Methods: Asymptomatic parents of children who were diagnosed with DDH were assessed for presence of dysplasia by examining their anteroposterior pelvis radiographs at the neutral position. Eighty-six hips of 43 participants were included in the study group and 98 hips of 49 participants were included in the control group. Presence of hip dysplasia over the anteroposterior pelvis radiographs was analyzed for Wiberg's angle, acetabular index of the weight-bearing zone (the Tönnis angle), acetabular depth/width index, femoral head coverage ratio (FHCR) and femoral neck/shaft angle.
Results: The mean acetabular depth/width ratio was 44.3% in the study group and 53.5% in the control group. And, the mean FHCR was 80% in the study group and 82% in the control group. There was a statistically significant difference between the two groups in terms of mean acetabular depth/width ratio ($p < 0.05$) and FHCR ($p < 0.05$). In addition, 21 participants in the study group and 2 in the control group had a pathological acetabular depth/width ratio. And, the number of participants with a pathological FHCR was 22 in the study group and 13 in the control group. A statistically significant difference was found between the two groups regarding the number of pathological measurements of acetabular depth/width ratio ($p < 0.05$) and FHCR ($p < 0.05$).
Conclusion: Having a parent with DDH is a definitive risk factor for the development of hip dysplasia in childhood. In addition, being a parent of a child with DDH is a risk factor for asymptomatic dysplasia. These parents should be screened by roentgenogram.
Level of Evidence: Level III, Diagnostic Study.
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Introduction

Developmental dysplasia of the hip (DDH) is a debilitating condition characterized by incomplete formation of the acetabulum and femoral head that occurs in different shapes at different ages. The most commonly accepted reason for the etiology, is the excessively loose joint capsule and the failure of ensuring the continuity of the femoral head within the acetabular cavity.[1] There is not a single cause of DDH, but different predisposing factors have been described. These factors are: ligament laxity, intrauterine causes, prenatal and postnatal position, genetic causes, racial difference and environmental factors.[2] Numerous studies have been conducted on hereditary transition of DDH.[3,4] In genetic studies on DDH, it has been shown that the presence of DDH in one of the family members increases the risk five times for other family members.[3] It can be said that when these hereditary factors are taken into consideration, parents of children with DDH also constitute a risky population.

In ideal treatment of DDH, early diagnosis can be made in presence of concentric reduction without causing epiphysis

* Corresponding author. Department of Orthopedics and Traumatology, Yozgat City Hospital, 66100, Yozgat, Turkey. Tel.: +90 3542190010, +90 5312331970.
E-mail address: drtacettin@gmail.com (T. Ayanoglu).
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destruction and continuation of this reduction. If DDH is detected in the first years of life, the treatment can be often done easily and the development of the affected hip may exhibit normal results.[5,6] However, DDH can progress silently until around 40–50 years of age and can be diagnosed when it shows symptoms such as pain, limitation of movement. In a radiology study on 2152 trauma patients, acetabular dysplasia was found as 9.3%.[7]

A newborn should be examined physically and radiologically in more detailed way because of this familial transition. However, in our daily practice, the asymptomatic parent of a child with hip dysplasia is not screened physically and radiologically in terms of asymptomatic hip dysplasia. We realized that this reverse transition was not studied in the literature and wanted to initiate awareness toward this risky population.

What would be the benefit of this study? The current treatment approach for symptomatic elderly patients is often arthroplasty. On the other hand, arthroplasty is an invasive procedure for patients and it is economically very demanding in some countries. Early diagnosis of acetabular dysplasia may not be cured. However, in case dysplasia is detected in the asymptomatic phase, measures by lifestyle changes, weight control, exercise programs may be taken to delay the being symptomatic or reduce the need for surgical treatment in the future. Thus, the aim of this study was to investigate whether being the parents of children with DDH is a risk factor for asymptomatic dysplasia.

Patients and methods

The study protocol was approved by the Ethics Committee of Clinical Investigations (2/10/2014). This study was conducted in accordance with the principles of the Declaration of Helsinki.

The inclusion criteria were having a child diagnosed with Stage 2B, 3 or 4 DDH by ultrasonography, being between the ages of 20 and 40 or not having symptoms such as hip pain or joint motion limitation. Patients with a history of trauma or steroid use, DDH, metabolic bone disease, Perthes disease and symptoms such as hip joint pain or decreased joint motion were excluded.

Asymptomatic parents of the children who were diagnosed with DDH were assessed for presence of dysplasia between the years 2015 and 2017 by examining the anteroposterior pelvis radiographs at the neutral position. Eighty-six hips of 43 participants were included in the study group and 98 hips of 49 participants were included in the control group. The control group consisted of patients who presented with soft tissue trauma, lumbago and trauma. Anteroposterior pelvis radiographs of all participants from both groups were taken at the neutral position.

Presence of hip dysplasia over the anteroposterior pelvis radiographs was analyzed for Wiberg' s angle, acetabular index of the weight-bearing zone (the Tönnis angle), acetabular depth/width index, femoral head coverage ratio (FHCR) and femoral neck/shaft angle (FNSA). The measurements were made by two radiologists, and the averages of the measurements were taken into account.

For Wiberg's angle measurement, the line passing through both femur heads is plotted first. Then, the angle between the vertical line passing through the center of the femoral head and the line connecting the outer edge of the acetabulum to the center of the femoral head was measured as the Wiberg angle (Fig. 1). A value of lower than 20° has been associated with acetabular dysplasia.[8]

For the measurement of the acetabular index of the weight-bearing zone (the Tönnis angle), the angle between the line passing through the medial and lateral of the weight-bearing surface of the acetabulum and the transverse line passing through the medial of the weight-bearing surface was measured (Fig. 2). A value greater than 13° has been associated with acetabular dysplasia.[9]

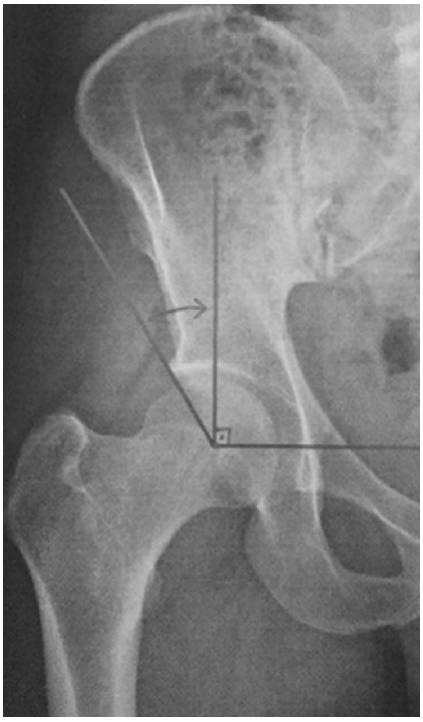

Fig. 1. Calculation of the Wiberg's angle.

For acetabular depth/width index measurement, a line was drawn connecting the superolateral and inferomedial edge of the acetabulum (width). Then, another line perpendicular to the first line was drawn (depth). Depth/Width*100 formula was used, and values greater than 38° were considered normal (Fig. 3).[10]

Three vertical lines must be drawn for FHCR. The line passing through the medial edge of the weight-bearing surface of the

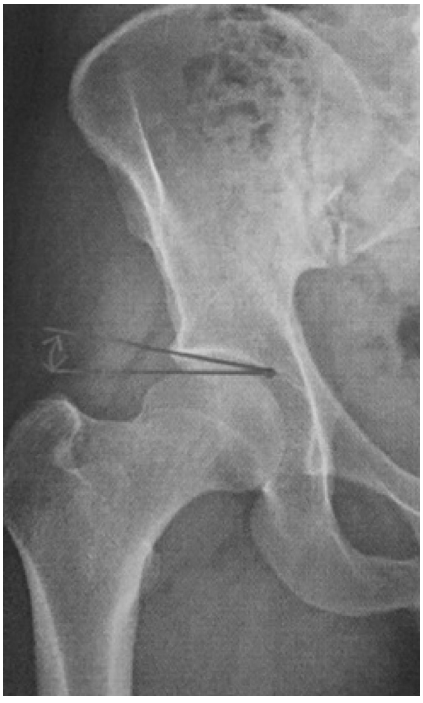

Fig. 2. Calculation of the acetabular index (the Tönnis angle).

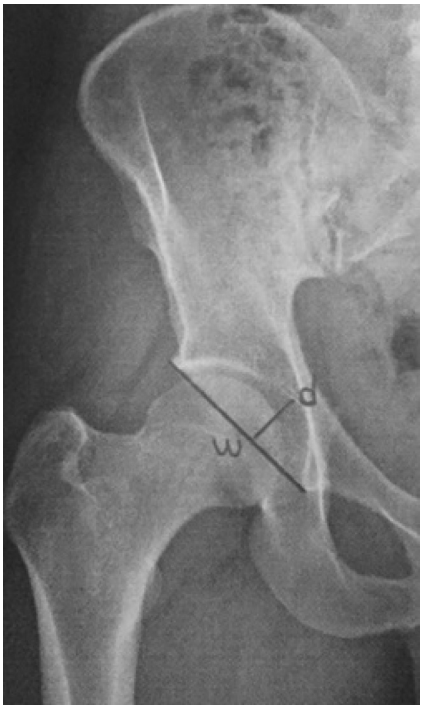

Fig. 3. Calculation of the acetabular depth/width ratio.

acetabulum (1), the line passing through the lateral edge of the acetabulum (2), and the line passing through the lateral edge of the femoral head (3) (Fig. 4). The FHCR is then calculated by dividing the distance between the medial and lateral edge of the acetabulum to the distance between the medial edge of acetabulum and the lateral edge of femoral head, multiplied by 100. Values higher than 75% are considered normal.[11]

The FNSA measurement is calculated by measuring the angle between the line parallel to the femoral head-neck and the line parallel to the femur shaft (Fig. 5). Values between 120° and 135° are considered normal.[10]

Statistical analyses were performed using the SPSS v.21.0 software. The Mann–Whitney U and Wilcoxon signed-rank tests were used for mean values. The chi-square test was used to assess participants with pathological results. P values less than 0.05 were considered significant.

Results

The mean age of children at the DDH diagnosis was 2,2 (range, 2–3) months. 17 of these children were stage 2b, three children were stage 3 and two children were stage 4.

The mean age of 22 male participants in the study group was 34.6 (range, 25–39) years, while the mean age of 21 female participants was 33.1 years (range, 24–36) years.

In control group, the mean age of 25 male participants was 34.1 (range, 24–37) years, while the mean age of 24 female participants was 33.7 years (range, 25–36) years.

The mean acetabular depth/width ratio was 44.3% in the study group and 53.5% in the control group. There was a statistically significant difference between the two groups ($p = 0.00$) (Table 1). Twenty-one participants in the study group and two in the control group had a pathological acetabular depth/width ratio and a statistically significant difference was found between the two groups ($p = 0.00$) (Table 2).

The mean FHCR was 80% in the study group and 82% in the control group, with a statistically significant difference between the two groups ($p = 0.04$) (Table 1). The number of participants with a pathological FHCR was 22 in the study group and 13 in the control group. A statistically significant difference was also found between these groups ($p = 0.03$) (Table 2).

The mean Wiberg's angle was 34.2° in the study group and 33.8° in the control group. No statistically significant difference was found between the two groups ($p = 0.25$) (Table 1). Five

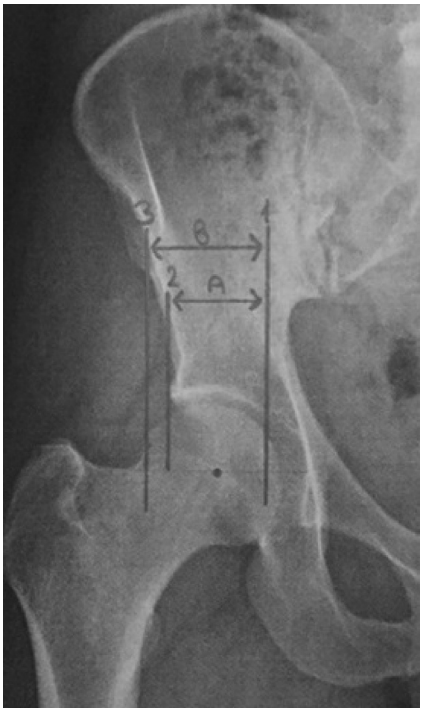

Fig. 4. Calculation of the femoral head coverage ratio.

Fig. 5. Calculation of the femoral neck-shaft angle.

Table 1
Distribution of the mean radiological measurements between groups. (Statistically significant p values are written in bold).

	Study group (n = 86)	Control group (n = 98)	p
Wiberg's angle	34.2	33.8	0.25
Acetabular index (°)	6.8	7.6	0.06
Acetabular depth/width ratio (%)	44.3	53.5	**0.00**
FHCR (%)	80.0	82.0	**0.04**
FNSA	140.8	134.5	0.85

FHCR: femoral head coverage ratio, FNSA: femoral neck-shaft angle.

Table 2
Distribution of the number of participants with pathological measurements between groups. (Statistically significant p values are written in bold).

	Study group (n = 86)	Control group (n = 98)	p
Wiberg's angle	5	3	0.32
Acetabular index	3	5	0.21
Acetabular depth/width ratio	21	2	**0.00**
FHCR	22	13	**0.03**
FNSA	35	33	0.31

FHCR: femoral head coverage ratio, FNSA: femoral neck-shaft angle.

participants in the study group and three in the control group had pathological Wiberg's angle. No statistically significant difference between the two groups was detected (p = 0.32) (Table 2).

The mean acetabular index was 6.8° in the study group and 7.6° in the control group, with no statistically significant difference between the groups (p = 0.06) (Table 1). Three participants in the study group and five in the control group had pathological acetabular indices. Again, no statistically significant difference was detected between the two groups (p = 0.21) (Table 2).

The mean FNSA was calculated as 140.8° in the study group and 134.5° in the control group. There was no statistically significant difference between the two groups (p = 0.85) (Table 1). Thirty-five participants in the study group and 33 in the control group had a pathological FNSA, with no statistically significant difference being detected between the two groups (p = 0.31) (Table 2).

Discussion

Developmental dysplasia of the hip covers a wide range of anomalies ranging from a shallow acetabulum to a pseudo-acetabulum. The natural course of DDH is well described in the literature. It may show a silent progress up to around 40–50 years of age, and osteoarthritis can be diagnosed with the onset of symptoms such as pain and limitation of joint motion. For the first time in 1882, Kronlain organized the family tree for DDH cases and demonstrated the anomalies in different generations.[2,12,13] Subsequent genetic studies carried out in families with DDH have shown that the incidence was high.[13] In 1964 Carter and Wilkinson and in 1970 Wynne–Davis reported that heredity is passed through two distinct gene systems. . Several genes have been implicated in individuals with DDH in familial studies.[14–16] If one of the family members has DDH, the risk for other family members is increased five-folds.[3] Today, the presence of DDH in a family is considered a definite risk factor for the development of DDH in children.

Taking familial transmission into consideration, having a children with DDH may be a risk for asymptomatic dysplasia for their parents.[4] We realized that this reverse transition was not studied in the literature. The results of our study also support this idea. The most important finding of this study is that the parents who have children with DDH but no complaints yet carry a risk for asymptomatic dysplasia. In our study, the FHCR and acetabulum depth/width ratios showed statistically significant differences between the study and control groups. There is a single publication that supports our results. In Japan, a genetic study that investigated the mothers of children with DDH who did not have a known disease and genetic transmission was detected in 9% of the mothers.[17] We have shown this relationship radiologically as distinct from this genetic study. Thus, we think that parents should also be considered a risky population for dysplasia and screened just like their children.

Traditionally, the diagnosis of hip dysplasia is made by radiological evaluation. In this context, the Wiberg's angle, acetabular index of the weight-bearing zone, acetabular depth/width rate and FHCR are used as radiological measurements in the evaluation of acetabular dysplasia in adults. In addition, FNSA is used to obtain information about the femur head-neck-shaft relationship.[10] These measurements were also included in our study. In a study evaluating the same population with a large case series, the average Wiberg angle and Tönnis angle values were measured. On pelvic AP radiographs, mean Wiberg angle was measured as 31.0 ± 6.2° for right hips and 32.1 ± 6.6° for left hips. Mean Tönnis angle was 6.6 ± 4.1° for right hips and 6.1 ± 4.2° for left hips. In our mean values, both the study group and the control group were consistent with these values. However, the parameters that were significant in our study were not evaluated in that study.[7] Repeated surveys were taken in suboptimal patient position to avoid wrongful measurements. Besides, the measurements were made by two different radiologists due to the challenges in detecting the anatomic landmarks.

In conclusion, having a parent with DDH is considered to be a definitive risk factor for the development of hip dysplasia in childhood. Our study shows that the presence of asymptomatic dysplasia is more common in parents of children with DDH than the normal population. We suggest screening of the parents by direct roentgenogram because late diagnosis of hip dysplasia requires more complicated treatment. Finally, we also believe that this study will initiate awareness toward this risky population.

Conflicts of interest

The authors declare no conflicts of interest.

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