GELİŞİMSEL KALÇA DİSPLAZİSİ HASTALARINDA TANI VE TEDAVİDE GECİKME NEDENLERİNİİN ARAŞTIRILMASI: TEK MERKEZLİ ÇALIŞMA

An Investigation of the Causes of Delay in the Diagnosis and Treatment of Patients with Developmental Hip Dysplasia: A Single-Centre Study

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ÖZET

Amaç: Gelişimsel kalça displazisi (GKD) önemli bir halk sağlığı problemidir. Tanı ve tedavide gecikme artmış tedavi maliyeti ve sakatlığa yol açabilmektedir. Bu bağlamda çalışmamızın amacı kliniğimize başvuran GKD'li hastalarda tanı ve tedavide gecikme nedenlerinin sunulmasıdır.

Gereç ve Yöntem: Ocak 2017 - Ocak 2020 yılları arasında 3-12 aylık arasındaki kliniğimize başvuran ve GKD tanısı konan 44 hasta çalışmaya alındı. Hastaların yaş, cinsiyet, kaçıncı doğum olduğu, aile öyküsü, kundaklama öyküsü, tanıda gecikme sebep ve süreleri, uygulanan eski tedavi şekilleri ile sosyodemografik özellikleri retrospektif olarak toplanarak analiz edildi.

Bulgular: Çalışmaya alınan 44 hastanın 4 (%9,1) ü erkek, 40 (%90,9) ı kız çocuktu. Ortalama yaş 5,44 ay (mean 3,5-12) idi. 19 hasta (%43,2) ailelerin birinci çocukları idi. 11 hasta da (%25) pozitif aile öyküsü, 22 (%50) hasta anamnezinde kundaklama uygulaması vardı.

Çalışmadaki GKD'li hastalardan; 6 hastanın (%13,6) normal raporlanmış kalça USG nedeniyle, 8 hasta (%18,2) sosyal nedenlerle ihmal, 2 hasta (%4,5) yeterli bilgilendirilmeme, 28 hasta (%63,6) çoklu ara bezi tedavisi nedenleri ile geç başvuru yaptıkları tespit edildi. Hastalarda ortalama 2,34 ay (aralık: 0,5-9) doğru tanı ve tedaviye ulaşmada gecikme tespit edildi. Gecikme süreleri ile gecikme nedenleri arasında istatiksel olarak anlamlı bir farklılık bulunmazken, (p=0,538), gecikme süresi ile yapılan tedavi şekli arasında anlamlı bir farklılık tespit edildi (p=0,006).

Sonuç: GKD gecikmiş tanı ve tedavinin en önemli ayağını yanlış kalça usg değerlendirmesi ve kalça usg sonucunda uygun tedavi protokolunun izlenmemesi oluşturmakta idi. 3 aydan büyük GKD'li çocuklarda Çoklu ara bezi kullanımının devam ettirilmesi tedavi yaklaşımını olumsuz etkilemektedir. Doktorların ve ailelerin bilgilendirilmesi için çalışmaların artırılması önem arz etmektedir.

Anahtar kelimeler: *Gelişimsel kalça displazisi; tedavi gecikmesi, tanı gecikmesi, kundaklama, çoklu ara bezi kullanımı, kalça ultrasonografisi.*

ABSTRACT

Aim: Developmental dysplasia of the hip (DDH) is an important public health problem. A delayed diagnosis and treatment may lead to increased treatment costs and disability. The purpose of our study is to present the causes of delay in the diagnosis and treatment of DDH patients who applied to our clinic.

Material and Method: Forty-four patients who were 3-12 months old, were admitted to our clinic between January 2017 and January 2020 and diagnosed with DDH were included in the study. The age, gender, birth order, familial history, history of swaddling, reason for and the time to delayed diagnosis, previous treatment methods and sociodemographic characteristics of the patients were analyzed retrospectively.

Results: Of the 44 patients included in the study, four (9.1%) were boys and 40 (90.9%) were girls. The average age was 5.44 months (mean: 3.5-12). Nineteen patients (43.2%) were the first children in their families. Eleven patients (25%) had a positive family history, and 22 (50%) had a history of swaddling.

Of the patients with DDH in the study; the reason for late presentation was a hip USG report with normal results in six patients (13.6%), neglect due to social reasons in eight patients (18.2%), provision of insufficient information in two patients (4.5%), and treatment with multiple diapers in 28 patients (63.6%). While there was no statistically significant difference between the delay time and cause of delay (p=0.538), a significant difference was found between the delay time and the treatment method (p=0.006).

Conclusion: The most important factor for delayed diagnosis and treatment of DDH was the wrongful USG evaluation of the hip and the failure to follow the appropriate treatment protocol based on the hip USG results. The continued use of multiple diapers in DDH patients older than 3 months has an adverse effect on the treatment approach. It is important to increase the studies to inform physicians and families.

Keywords: *Delayed diagnosis; delayed treatment; developmental hip dysplasia; hip ultrasonography; multiple diaper use; swaddling.*

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INTRODUCTION

Developmental dysplasia of the hip (DDH) is one of the most important orthopedic pathologies seen during childhood (1). Although its frequency has been reported as 1:1000 in the literature, the rate is estimated to be as high as 5 to 15:1000 in our country. If left untreated, the disease will be one of the causes of disability for tens of thousands of newborns in our country (2).

The success of treatment is related with early diagnosis. Clinical examination and hip ultrasonography (USG) are the most important diagnostic methods in the first trimester (3,4). The aim is the treatment of patients who have an alpha angle level of 59 and less in the hip USG performed at the 3rd month after birth with dynamic bandages or static orthoses. Achieving acetabular development through concentric hip reduction is aimed in the treatment of DDH (5).
Early diagnosis and treatment allow the disease to recover without sequelae, however, serious sequelae may be observed in patients who are late for treatment (6). Serious studies conducted within the last 10 years in our country undoubtedly promise positive results for the early diagnosis and treatment of this disease (2). However, it is known that setbacks in the diagnosis and treatment of the disease will irreversibly affect the future of the individual and also cause serious damage to the country's economy (7).

Setbacks in the diagnosis stage reduce the chance of early treatment of the patients. The aim of the study is to investigate the causes of delay in the diagnosis and treatment of patients who were diagnosed with DDH and were older than three months. We believe that identifying these causes and taking precautions can make positive contributions in the course of the disease.

MATERIAL AND METHOD

Permission was obtained from Cumhuriyet University Faculty of Medicine ethics committee dated 18.03.2020 numbered 2020-03/04. Seventy-five pediatric patients who were diagnosed with DDH, were aged between 3 months and 12 months, and applied to our clinic between January 2017 and January 2020 were examined. Within the scope of the study, teratogenic hips, patients whose files could not be accessed, and control patients whose treatment had been initiated in another center were excluded from the study. Finally, the study was continued with 44 patients.

The gender, age, familial history, history of swaddling, birth order, hip USG results, reason for delayed diagnosis, time of presentation of the patients and the treatment methods performed were noted.

The reasons for delay were grouped under the following headings: normal results in the USG report, social reasons, provision of insufficient information, and suggesting the use of multiple or large diapers. When estimating the time to delayed treatment, the period after the third month was taken into account. All patients were included in the appropriate treatment program according to age at the time of presentation and to the acetabular angle values on the USGs or roentgenograms of the hip.

The practices were evaluated using the SPSS 23.0 software package. The analysis of the data set has been examined under two main titles. First, descriptive statistics of the variables (minimum, maximum, average, standard deviation) were examined. In the second stage, the difference analyses of the variables were performed. In order to select the appropriate test for analyzing the differences, first, the normality test of the numerical variables was carried out using the Kolmogorov-Smirnov Z test. The results of the normality test showed that none of the variables showed normal distribution. When analyzing the differences, the Mann-Whitney U test was used to compare the variables with two categories. At the same time, the chi-square test was used to examine the categorical variables. The significance level was set at 95% in the tests to be performed.

RESULTS

Of the 44 patients included in the study, four (9.1%) were boys and 40 (90.9%) were girls. The average age of the patients was 5.44 months (range: 3.5-12). Nineteen patients (43.2%) were the first children in their families. Eleven patients (25%) had a positive

family history, and 22 (50%) had a history of swaddling (Table 1).

Of the patients included in the study, six (13.6%) stated that they did not think an examination was necessary due to the normal results in their USG report of the hip, eight (18.2%) said they were neglectful due to social reasons, two (4.5%) said they did not show up as they had been given insufficient information, and 28 patients (63.6%) said they thought using multiple diapers was sufficient for treatment, thus their presentations were late.

The patients had an average delay of 2.34 months (range: 0.5-9). While there was no statistically significant difference between the delay time and cause of delay (p=0.538), a significant difference was found between the delay time and the treatment method (p=0.006) (Table 2).

In evaluation of the recommendations for patients to use multiple diapers, it was found that 17 of the 28 patients were directed by orthopedists, 15 by pediatricians and two by radiologists.

Table 1. Demographics of the patients.

Gender, n (percentage)	Male	4 (9,1%)
	Female	40 (90,9%)
Positive family history, n (percentage)	Yes	11 (25,0%)
	No	33 (75,0%)
History of swaddling, n (percentage)	Yes	22 (50,0%)
	No	22 (50,0%)
First born child, n (percentage)	Yes	19 (43,2%)
	No	25 (56,8%)
Reason for delay, n (percentage)	Normal USG results	6 (13,6%)
	Use of multiple diapers	28 (63,6%)
	Social reasons	8 (18,2%)
	Lack of information	2 (4,5%)

Table 2. The reason for delay and its effects on the treatment.

		Minimum	Maximum	Mean	Standard Deviation	p
Reason for delay	Normal USG	0,50	9,00	3,00	3,36	0,538
	Use of multiple diapers	0,50	9,00	2,27	1,72	
	Social reasons	0,50	3,00	1,69	0,96	
	Lack of information	2,00	6,00	4,00	2,83	
Treatment	Pavlik bandage	0,50	2,00	1,19	0,48	0,006[a]
	Abduction orthosis	1,00	3,00	1,92	0,92	
	Pelvipedal plaster	0,50	9,00	3,04	2,28	

a: The difference between the Pavlic bandage and pelvipedal plaster groups was calculated (p=0.003). p<0.05 was considered statistically significant.

DISCUSSION

Although DDH has been intensely screened in recent years, it still carries a risk of serious disability for our country. In this disease, where early diagnosis and treatment is of great importance, we believe that determining the causes of delay in diagnosis and treatment and elaborating the studies in this context

will provide serious benefits for the prevention of the disease. Our study stands out in this regard.

Several risk factors have been identified for the development of DDH. In children with these risk factors, the prevalence of the disease appears to be significantly increased (8,9). Identifying the predisposing factors and following these patients closely is important for the risky group. It is known that 25-90% of the children in the studies are included in a risk group. This high rate of distribution led researchers to the belief that these patients have not been followed properly. This is as an important knowledge also for the patients in our study, highlighting the possibility of overlooking or misdirecting the diagnosis even when under doctor control.

Mechanical factors after birth affect the development of DDH. Although there are many reasons for the high prevalence in our country, it is clear that the swaddling culture, which is one of the preventable risk factors, undoubtedly contributes to the increase of these rates (10). Experimental studies have shown that immobilization with the knee and the hip in extension causes hip dislocation (11,12). With swaddling, children are immobilized with their knees in extension, and their hips in extension and adduction. It has been articulated for years that forceful hip extensions increase the risk of DDH (11). In countries where this practice had been terminated, serious decreases have been reported in the incidence of DDH (12,13). Unfortunately, in rural areas of our country, swaddling is still a followed practice in newborn children. The fact that 50% of the children in the study had been swaddled confirms this statement. Although there are publications in the past literature showing that public education provides serious benefits for DDH, the reduction in this education have been shown to lead to an increase in DDH cases (14). The harms caused by swaddling and similar practices should be explained to mothers through education (15). These trainings should be repeated frequently. Otherwise, it is obvious that the cultural lifestyle will continue to dominate the scientific facts.

Ultrasonography (Graf) of the hip is used as an effective method in DDH scans. The Graf technique, which is a static method, evaluates the acetabulum using coronal images in the lateral plane (16). Graf advocated that the Graf technique is a very reliable technique when the reference points are identified correctly and that providing trainings by authorized trainers is important in getting accurate measurements (17). Although there are studies reporting that USG of the hip gives more effective results when compared to clinical examination in screening programs (18), Rosenberg et al. reported a small number of patients with a clinically unstable hip despite having a large series of patients with a normal USG of the hip (4). Although the evaluation of the Graf method is subjective, standardization is of much importance. This can be achieved with experience and training programs on hip USG (19). In our study, the reason for delay in the diagnosis of six patients was due to the interpretation of the hip USGs as normal. On the X-ray of the patients, we determined that the AC index was between 36-42 degrees and the alpha angle level in the repeat USGs of the hip was between 44-50. In the light of these values, we believe that the 'normal' hip USG examinations may have been due to a technical error. For accurate measurements, the necessity of physicians' participation in training programs on hip USG is of importance.

In addition to performing a technically appropriate USG of the hip, Graf emphasized the necessity of proper treatment planning according to alpha beta angle (17). At the time of admission of 30 patients in our study to our clinic, the families knew that the hip USG data were insufficient. However, the appropriate treatment protocol was not applied. If the hip USG performed with the Graf method is not assessed with the correct technique and if the appropriate treatment protocol is not given to patients, it has been observed to cause serious setbacks in the treatment. In our study, 13 patients were treated with Pavlik harness, six patients with abduction orthosis and 25 patients with reduction and pelvipedal plastering. It was observed that the treatment protocol applied to the patients changed as the delay times increased. This situation affects the recovery time negatively.

Using a wide or double diaper, especially in the

neonatal period, causes slight flexion and abduction in the hips and decreases the incidence of DDH (13). There are countries that have adopted and utilized the large diaper application as a national policy and have realized serious decreases in the frequency of DDH (12,20). The main purpose of these applications is to keep the hip in mild flexion and abduction during the three-month development period. This practice has been suggested as a preventive measure in the literature (8). Although Pavlik harness and abduction orthoses are frequently used in children older than three months, depending on the growth deficiency of the hip, closed or open reduction and pelvipedal plastering are performed in severe dislocation cases (8). The use of large or multiple diapers is not acceptable in treatment. As seen in the study, persistence or failure of multiple or large diaper application in children older than 3 months have led to serious failure and delay of treatment in children. It is important that the diapers used in infants are long enough to be tied around the belly and do not press against the groin (21). During the examination of the hip, it was observed that the application of multiple diapers caused bulging over the inguinal region and limited the hip flexion (Figure 1). According to the data from our study, it was observed that there was an average delay of 2.27 months in children due to the use of multiple diapers. In addition, the treatment approach followed varied with the increase in the delay in the treatment period, initiating the implementation of more radical applications. One patient in our study underwent final treatment with Salter pelvic osteotomy. It is thought that a prolonged delay period and a prolonged treatment may cause both psychosocial and economic losses.

Although there was no statistically difference between the causes of delay and delay times, it was seen that the lowest average delay time was due to social reasons. However, an important point is that other causes of delay were due to medical errors and drawbacks, and that they raised concerns regarding the professional knowledge and skills required for the early diagnosis and treatment of DDH. Informing the healthcare staff, and especially working in coordination with pediatricians, gynecologists, orthopedists, family physicians specialists and nurses will be beneficial (21). The fact that the 17 patients in the study were incompetently assessed by orthopedic specialists who have received serious training on DDH is also thought-provoking and saddening. The pediatricians have an important role in identifying the risky groups and detecting the positive examination findings. In addition, they should have sufficient information about the treatment process of the disease. The delay of treatment with the recommendation by pediatricians about using plenty of diapers in 15 patients reveals the severity of the condition.

Although the delay in the time of starting the treatment affects the treatment cost, the lack of a cost analysis constitutes the limitation of the study. In addition, no comparison could be made between the groups due to the limited number of patients.

Figure 1. Practice of multiple diapers in a 4.5-month-old girl. A) The appearance of the bulging diaper over the right inguinal region. B) Restriction of movement due to multiple diapers, while the hip is flexed (arrow).

CONCLUSION

Informative training on DDH should be carried out on a continuous basis for both the general public and physicians. Our study showed that there is still more to be done about the diagnosis and treatment algorithms of DDH, both among families and physicians.

The family should be provided serious trainings about swaddling practice. Public awareness should also be increased.

Courses should be organized and participation of the physicians should be ensured in order to perform hip

USG with the right technique and to determine the appropriate treatment protocol.
Post-specialization trainings should be supported and information gaps should be eliminated.

Although the causes of diagnosis and treatment delay in the study are discussed, the main point to emphasize is that efforts toward preventing the disease should be increased.

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