

Late-presenting developmental dysplasia of the hip in Jordanian males

A retrospective hospital based study

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ABSTRACT

الأهداف: وصف نمط خلع الورك التطوري متأخر الظهور عند الأطفال الأردنيين الذكور. وملاحظة عوامل الخطورة والموجودات المترابطة في هذه المجموعة من المرضى.

الطريقة: بلغ عدد المرضى الذكور الذين تمت معابنتهم في عيادة عظام الأطفال لفحص خلع الورك التطوري 1145 طفلاً تمت مراجعتهم بشكل استعادي. أجريت هذه الدراسة في شعبة جراحة العظام، الجامعة الأردنية في الفترة ما بين مارس 2011 وأكتوبر 2014. تم تقييم كل البيانات من الملف الطبي وكذلك قياسات الصور الشعاعية.

النتائج: من مجموع 1145 طفلاً تم فحصهم، 43 طفلاً شخصوا بخلع الورك التطوري متأخر الظهور وعدد مفاصل الورك المصابة 70 مفصلاً من مجموع 86 مفصلاً. الطفل المولود أولاً لديه اختطار زائد للتعرض للخلع 41.9% حالات التوليد القيصرية ترتبط بشكل ملحوظ مع زيادة خطر خلع الورك ($p=0.004$)، بينما ترتبط الولادة الطبيعية مع زيادة خلل التنسج الورك ($p=0.004$) من المرضى الذين يعانون من خلع الورك التطوري لم يكن لديهم أي عوامل خطر 44.2%. الحالات ثنائية الجانب هي أكثر شيوعاً من الحالات أحادية الجانب (26 [60.5%] مقابل 17 [39.5%]). محدودية تبعيد الورك كانت موجودة ثابتة في كل المفاصل المخلوعة ($p<0.001$) الحالات المرافقة والمرتبطة مثل حنف القدم و الصعر الخلقى لم تتم مشاهدتهم في هذه الدراسة.

الخاتمة: الولادة القيصرية عامل اختطار مهم لخلع الورك التطوري، بينما الولادة المهبلية الطبيعية مرتبطة بشكل مهم مع خلل التنسج الحقي. حالات خلل التنسج الورك ثنائية الجانب أكثر شيوعاً من أحادية الجانب حنف القدم والصعر الخلقى لم تتم مشاهدتهم في هذه الدراسة.

Objectives: To describe the pattern of developmental dysplasia of the hip (DDH) in late presenting Jordanian male patients and identify the risk factors and associated findings.

Methods: This is a retrospective study of 1145 male patients who attended the Pediatric Orthopedic Clinic for a DDH check up. This study was carried out in the Orthopedic Section, Special Surgery Department, Faculty of Medicine, The University of Jordan, Amman, Jordan between March 2011 and October 2014. Data was collected from medical records, and x-ray measurements were evaluated.

Results: Of the 1145 male patients, 43 (3.75%) with 70 involved hips were diagnosed with late-presenting DDH. Being a first-born baby resulted in 41.9% increased risk for DDH. Cesarean delivery was significantly associated with an increased risk of hip dislocation ($p=0.004$) while normal delivery was significantly associated with acetabular dysplasia ($p=0.004$). No predictable risk factors were found in 44.2% patients with DDH. Bilateral cases were more common than unilateral cases: (26 [60.5%] versus 17 [39.5%]). Limited abduction was a constant finding in all dislocated hips ($p<0.001$). Associated conditions, such as club foot and congenital muscular torticollis were not observed.

Conclusion: Cesarean section is a significant risk for dislocated hips while normal delivery is significantly associated with acetabular dysplasia. Bilateral DDH is more common than the unilateral. Club foot and torticollis were not observed in this series.

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Developmental dysplasia of the hip (DDH) is a widespread term first used by Klisic¹ in 1989, which indicates an extensive variety of vibrant abnormalities present in the immature hip joint in children. Approximately, one in every 1,000 child is delivered with a dislocated hip joint, while every 10 in 1,000 children are delivered with subluxation or dysplasia of the hip joint. The DDH represents a wide spectrum of anatomical abnormalities, which range from slight hip instability to frank dislocation of the hip joint, as well as the mal-development of the acetabulum. These abnormalities are not always apparent clinically at birth, so high index of suspicion should be raised when there are positive risk factors. Early diagnosis and treatment are crucial for optimal outcome and the key to stay out of trouble in dealing with these patients. Developmental dysplasia of the hips is usually developmental and can occur during the first year of a child's life. Developmental dysplasia of the hip is an evolving process and the clinical examination shows different signs based on the spectrum of the DDH and the age of the patient. In accordance with the clinical practice guideline of the "American Academy of Pediatrics", no considerable sign was observed, which indicates the pathognomonic for hip dislocation.² Therefore, the clinical examination is neither 100% sensitive, nor specific to diagnose DDH.³ Developmental dysplasia of the hip is more common in female patients, and has been well studied and analyzed regarding its incidence, presentation, and the associated risk factors.⁴ First-born infants have a higher incidence of DDH^{4,5} as well as those born with breech presentation.⁴ Since the neonatal hips are primarily a cartilaginous structure, it is difficult to observe with standard radiographic techniques, therefore, nowadays ultrasonograms are very popular. It is currently considered a safe tool for young infants and can shed light on pathological changes that cannot be seen using the standard radiological techniques. Only few studies addressed the issue of late-presenting DDH in male children. Kosar et al⁶ had studied this spectrum in male infants and showed no statistical significance presented between risk factors and DDH in male cases. Jordanians are an ethnically distinct and homogenous population that has not been studied before. In spite of the improvement in screening methods, late diagnosis

of DDH occurs and is of great concern for the family and the treating physician. In this study, we investigated the risk factors, the clinical, and radiological findings associated with DDH in late-presenting Jordanian male children.

Methods. At present in Jordan, no screening program was set up to diagnose DDH; however, there is an increased awareness on DDH among families and physicians (Personal communication with Dr. Jamal Qunnash, head of Orthopedic Surgery in the Ministry of Health Hospitals, Dr. Firas Ibrahim, head of Pediatric Orthopedic Surgery in the Royal Medical Services Hospitals, and Dr. Khaldoon Bashaireh, head of Orthopedic Surgery at Jordan University of Sciences and Technology). We reviewed the data of 1145 male children who visited our pediatric orthopedic clinic at the Orthopedic Section, Special Surgery Department, Faculty of Medicine, The University of Jordan, Amman, Jordan, between March 2011 and October 2014 for a developmental hip dysplasia check up. Ethical approval of the study was obtained from the Research Committee, University of Jordan, and we performed our study in accordance to the principles of the Helsinki Declaration.

To diagnose the late presentation of DDH in our study, the cut-off age was ≥ 3 months at the time of diagnosis. Patients with evident neuromuscular or syndromic disorders, children younger than 3 months at the time of diagnosis, and patients whose medical charts were incomplete were all excluded from the study. One thousand and one hundred forty-five patients who met the criteria were assessed at the time of presentation for associated risk factors including rank, family history of DDH, type and mode of delivery, which were all recorded in their medical charts. The clinical examinations focused on the presence of congenital muscular torticollis (CMT), limited hip abduction, and foot deformities. In our hospital, a standard radiological evaluation by means of a hip and pelvis x-ray was taken, while the child remains in the mid abduction and mid internal rotation.⁷ All clinical and radiological data were recorded in the patient's medical charts and then collected after permission. The patient's hip joints are then classified according to Tönnis measurements of the acetabular angles into 4 grades⁸ (Table 1).

A MEDLINE search was carried out to retrieve literature in English, searching for DDH, male, and late presenting DDH.

Statistical analysis. The Statistical Package for Social Sciences (SPSS Inc., Chicago, IL, USA) Windows

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Table 1 - Acetabular index angles according to Tönnis.

Age (months)	Grade 1 (normal)	Grade 2 (mildly pathological)	Grade 3 (moderately pathological)	Grade 4 (extremely pathological)
3-4	<30°	≥30° - <35°	≥35° - <40°	≥40°
5-24	<25°	≥25° - <30°	≥30° - <35°	≥35°

Table 2 - Clinical variables associated with developmental dysplasia of the hip (DDH).

Finding	P-value	OR	95% CI
Dislocated hips in C/S versus in AD in normal delivery	0.004	9.844	1.85-52.39
Bilateral versus unilateral DDH	0.55		
Right- versus left-sidedness	1.00		
Limited hip abduction in dislocated hips	<0.001	OR:4.0	1.5-10.66

P value of ≤0.05 is statistically significant, C/S - cesarean section, AD - acetabular dysplasia, DDH - developmental dysplasia of the hip, OR - odds ratio, 95%CI - 95% Confidential interval

Version 10.01 was used for statistical analysis. Moreover, the contingency tables, Chi-squared, and Fisher's Exact test were used when appropriate to evaluate the disparities among the variables in the clinical findings and associated risk factors. However, the 2-tail statistical significance remained constant at the *p*-value of ≤0.05.

Results. Forty-three of the 1145 (3.75%) children screened had late-presenting DDH. There were 70 involved hips out of the 86 hip joints. The mean age at diagnosis was 4.98 months (range 3-18). The incidence of DDH was found to be the highest in the first-born male child (18/43 [41.9%]) and then decreased with subsequent deliveries. Sixteen of the 43 children (37.2%) had a positive family history of DDH, but no statistically significant factor for DDH.

Table 2 summarizes the results of the statistical analysis. Seven of 9 (78%) dislocated hips were associated with cesarean section (CS) delivery versus 2 (22%) associated with normal delivery, a statistically significant difference. Four of the 9 (44%) dislocated hips presented as a right sided disorder, whereas 5 (56%) were diagnosed as a left sided disorder, a statistically insignificant difference. Four infants were breech-presented and were delivered by CS. A significant association was found between normal delivery and the acetabular dysplasia as compared with the CS, which was statistically significant. Twenty-six (60.5%) children had bilateral involvement while 17 (39.5%) had unilateral involvement. Of the unilateral cases, 9 (20.9%) had left-sided DDH while 8 (18.6%) had right-sided DDH. No statistically significant difference was found in the relative frequency of right versus left sided DDH or unilateral versus bilateral DDH.

The only significant physical finding found in the patients with DDH was limited abduction, which

Table 3 - Number of hips and percentage of involvement according to the Tönnis classification (N=86).

Grades	Description	Number of hips (%)
Grade 1	Normal	16 (18.6)
Grade 2	Mildly pathological	33 (38.4)
Grade 3	Moderately pathological	19 (22.1)
Grade 4	Extremely pathological	18 (20.9)
Total		86 (100)

was only observed in children with frankly dislocated hips (Table 2). None of the assessed patients presented with any kind of feet deformities or torticollis. In late presenting patients, radiographic evaluation was performed using a plain radiograph. According to Tönnis classification of DDH,⁹ the average acetabular index at the age of 3 months in males was considered to be $23.4 \pm 4.5^\circ$ with an upper limit of normal of $<30^\circ$. The acetabular index is considered as the most reliable radiographic measure to evaluate the developmental of DDH in this group. Table 3 shows the 86 hips in 43 children classified according to the Tönnis classification.

Discussion. Developmental dysplasia of hip or DDH is considered as a combination of abnormalities that affects the immature hip joint in children. In this disorder, the femoral head and acetabulum are neither aligned nor grow normally.¹⁰ In spite of increased awareness of DDH in the society, late diagnosed cases of DDH trouble both parents and treating physicians. Three months is the cut off age we used in our study to diagnose the late presentation of DDH, similar to Woodacre et al,¹¹ although different criteria was used in the literature for definition of late presenting DDH. The incidence of late diagnosed DDH in this group of male patients was 3.75%, which was higher than that

observed by Kosar et al⁶ in males diagnosed before the age of 14 weeks. The mean age at presentation in this study was 4.98 months (range 3-18). First-born babies have a high risk of DDH among their siblings (41.9%), which is higher than that reported in the literature. The risk is mostly related to the tight uterine musculature of the primigravida and its crowding effect on the fetal development. In this study, 37.2% of children had a positive family history of DDH, which is also higher than that reported in the literature for DDH in general¹² and it supports the data on familial and genetic etiology of DDH.¹³ Approximately 32.6% of the DDH male children are delivered by CS with 7 out of 9 dislocated hips belonging to this entity, which supports the association of DDH and CS as published in the literature.¹⁴ Approximately 9.3% of the male children with DDH were indicated to be delivered with breech presentation, which is significantly lower as compared with the reports from previous study,¹⁴ which attribute to the mixed gender of population.⁷ Normal delivery is not a risk factor for DDH, however, in this study, a significant association was found between normal delivery and acetabular dysplasia in comparison with the association between CS and acetabular dysplasia; this may increase our awareness to the concept of 'primary acetabular dysplasia' as a key factor for the development of hip dysplasia,¹⁵ where the genetic predisposition may significantly contribute to its development and raise the suspicion of DDH occurrence in societies with high incidence of DDH. Moreover, it was also found that the left sided hip is more affected by DDH (almost 60% of the individuals).⁵ However, in our study, we found that 26 individuals, approximately 60.5% of the children involved in the study had bilateral DDH, which is more widespread as compared with the unilateral DDH in such case studies.⁵ Moreover, 17 children or 39.5% of cases were found to be unilateral and the side of involvement showed nearly equal distribution between left (9 hips) and right side (8 hips), which supports previous report of similar population,⁷ whereas in contrast to what is published in the literature,¹⁴ this may be explained by mechanical factors such as swaddling, a habit still practiced in our society. Approximately 44.2% of children with developmental hip dysplasia, mainly subclinical acetabular dysplasia did not demonstrate any risk factors. Kosar et al⁶ found 0.9% of infants without risk factors developed DDH. This may increase our awareness regarding the significance of screening programs with high prevalence of hip dysplasia. Kim et al¹⁶ analyzed the relationship

between congenital muscular torticollis (CMT) and DDH in infants. Their results showed the coexistence rate as 14.9%. Contrary to this, none of our patients at the time of diagnosis was found to have CMT. We did not find any patient at the time of evaluation to have clubfoot deformity (CTEV), keeping in mind that there is a negligible involvement between CTEV and DDH in children. The same relationship was also supported by the study of Chou et al.¹⁷ According to Tönnis classification of DDH, based on the principle of the acetabular index angle, boys aged 3 months, the acetabular index was $23.4 \pm 4.5^\circ$ and the upper limit of normal was $<30^\circ$ (Table 1). In our study, 38.4% of the involved hips showed type 2 acetabular dysplasia (Table 3). Haidukewych & Sink¹⁸ observed that children who were born and swaddled were at a considerably high risk in developing DDH in their initial years of life. It was also concluded by further studies¹⁹ that a child who had tightly swaddled legs with knees and hips is prone to suffer from developmental hip dysplasia during the first year of their life. Since swaddling has become more popular, it is more noteworthy for parents to swaddle the newborns safely and take appropriate measures where the lower limbs should not be tightly wrapped straight down and pressed together, but allowing the lower limb to flex and abduct at the level of the hip joint, which allows it to naturally develop.

Study limitation. The limitation of this study is the small number of subjects who participated. Furthermore, our study was retrospective and clinical and radiological information was not readily available on all patients. Future research should focus on male infants with DDH since there is an increased incidence of DDH in male children that has not been recognized before. Early detection of DDH is then likely to decrease the burden and the cost of treatment of this disease when detected at later stage.

In conclusion, all male patients with frankly dislocated hips represented positive risk factors; however, a significant proportion of our male patients represented no risk factors, but had a subclinical acetabular dysplasia, which can lead to the future onset of osteoarthritis. Therefore, a well organized screening program should be recommended and established to diagnose these cases in order to decrease the burden of late diagnosis and treatment of DDH cases. In our series, CS is a significant risk for dislocated hips especially when associated with breech presentation, while normal delivery was significantly associated with acetabular dysplasia. The bilateral hip dysplasia is more common than the unilateral ones.

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Clinical Practice Guidelines

Clinical Practice Guidelines must include a short abstract. There should be an Introduction section addressing the objective in producing the guideline, what the guideline is about and who will benefit from the guideline. It should describe the population, conditions, health care setting and clinical management/diagnostic test. Authors should adequately describe the methods used to collect and analyze evidence, recommendations and validation. If it is adapted, authors should include the source, how, and why it is adapted? The guidelines should include not more than 50 references, 2-4 illustrations/tables, and an algorithm.