Original Article

Universal in-house neonatal hips ultrasonography screening in the United Arab Emirates

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ABSTRACT

الأهداف: التحقق مما إذا كانت عوامل خطر الإصابة بخلل التنسج النمائي للورك (DDH) تختلف مع الأخذ في الاعتبار الأصل الجغرافي أو الصورة السريرية أو وجود عوامل الخطر في مجموعة متجانسة من المواليد في مستشفى واحد في الإمارات العربية المتحدة.

المنهجية: تم إجراء التصوير بالموجات فوق الصوتية الشاملة لمفصل الفخذ عند الولدان باستخدام طريقة غراف (Graf) في قسم الولادة. كان متوسط عمر الأطفال حديثي الولادة 3 أيام. تم تشكيل مجموعتين متجانستين: مجموعة دول مجلس التعاون الخليجي، والتي تكونت من 169 طفلاً (47.7%)، والمجموعة غير الخليجية من 185 طفلاً(52.3%).

النتائج: كانت نسبة حدوث خلل التنسج النمائي للورك (DDH) هي 1.7%. وكانت أعلى بين مواليد منطقة دول مجلس التعاون الخليجي (2.9%) وأعلى بكثير بين الفتيات من هذه المنطقة (6.3%). بالإضافة إلى ذلك، ارتبطت نتائج الورك غير الطبيعية وعرض المقعد بشكل كبير بخلل التنسج النمائي للورك. كانت نسبة حدوث إصابة بالوركين غير الناضجين 9% (النوع IIa) وكانت مماثلة بغض النظر عن الأصل أو الجنس.

الخلاصة: حديثي الولادة من منطقة دول مجلس التعاون الخليجي، والفتيات على وجه الخصوص، لديهم نسبة أعلى من خطر الإصابة بخلل التنسج النمائي للورك. تسلط هذه النتائج الضوء على الحاجة الملحة لإنشاء برنامج وطني لفحص الموجات فوق الصوتية لخلل التنسج الوركي التنموي.

Objectives: To investigate if the incidence of developmental dysplasia of the hip (DDH) differs considering the geographical origin, clinical picture, or presence of risk factors in homogenous cohort of neonates born in Mediclinic Al Jowhara hospital, Al Ain, United Arab of Emirates (UAE).

Methods: Universal ultrasonography hips screening of the neonates in the maternity ward of Mediclinic Al Jowhara hospital, Al Ain, UAE, was carried out using the Graf method. The average age of the neonates was 3 days. Two groups were formed for comparison: I) the Gulf Cooperation Council (GCC) group (n=169, 47.7%), and II) the non-GCC group (n=185, 52.3%).

Results: The incidence of DDH was 1.7%. It was higher among neonates from the GCC region (2.9%) and significantly higher among girls from this region (6.3%). The incidence of immature hips (type IIa) was 9% and was similar regardless of origin or gender.

Conclusion: Neonates from the GCC region, girls in particular, have a higher incidence of DDH. These results highlight the emergency to establish a national ultrasonography DDH screening program.

Keywords: universal DDH screening, Graf technique, neonate, United Arab Emirates

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Developmental dysplasia of the hip (DDH) encompasses various forms of underdevelopment in the immature joint, from mild acetabular dysplasia to complete dislocation, and is defined as a dynamic condition "potentially capable, as the baby develops, of getting better or getting worse".[1] For more than a century now, the orthopedic community has struggled to diagnose DDH as early as possible, which would give the baby a chance for swift and less radical treatment.[2,3]

All efforts should be made to distinguish the physiological variances of acetabular development and hips laxity in neonates from true DDH.[4,5] Proper imaging of neonatal hips was lacking until Professor Graf introduced hip ultrasonography in the late 1970s.[6] The technique has since been refined, and it is currently capable of clearly presenting all non-ossified elements of the neonatal hip. Graf ultrasonography technique (GUS) should be carried out by a certified examiner

and comprises a battery of clear instructions to classify the level of hip maturity at birth.[6]

Most of the lax neonatal hips stabilize spontaneously, in case the full hip mobility is provided. Any restriction in free leg movements after delivery affects proper development of hips, of which, tight-leg swaddling (TLS) is a major one.[2,7]

The ultrasonography became an integral part of DDH screening programs in many developed countries by the end of 20th century. Depending mainly on the health systems' resources and politics, it was either carried out on all babies (universal ultrasonography screening [UUS]) or carried out based on the presence of risk factors (selective ultrasonography screening [SUS]).[6-9] Major risk factors are positive family history and breech presentation. Besides these, any reasons for restricted intrauterine mobility or clinical findings that indicate it at birth could be considered a risk.[8]

The United Arab Emirates (UAE) is a part of the Gulf Cooperation Council (GCC) region and has a shared tradition of TLS as well as a high incidence of consanguinity.[9,10] It puts infants of the GCC region under a higher risk for DDH. Consanguinity narrows the genetic pool in each generation and enables clinical expression without positive family history. Tight-leg swaddling reduces proper development of acetabulum during the period of the fastest growth.

Almost all studies of DDH in the GCC region originate from the Kingdom of Saudi Arabia (KSA), beginning in the late 1980s. The intention of these studies was to design the most appropriate local screening program for early detection of DDH and implement it in the national health system (a process that is still ongoing).[9,11] The UAE lacks official guidance for ultrasonography screening of DDH, and only one retrospective study has been published to date.[12,13]

The Mediclinic Al Jowhara Hospital, located in the city of Al Ain, Abu Dhabi, UAE, along its border with Oman, offers medical services to the patients of neighboring countries as well as to a sizeable expat community. Hence, this center presents an ideal case for a study with groups of comparable sizes based on demography.

The purpose of the present study was to investigate whether DDH incidence differs in relation to geographical origin, gender, and the presence of risk factors in a homogenous cohort of neonates. An additional goal was to inform the caretakers regarding the harmful effects of TLS and the benefits of early GUS.

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Methods. The study was carried out as a prospective clinical and ultrasonography assessment of the hips of all healthy newborns delivered at Mediclinic Al Jowhara Hospital in Al Ain, UAE, from December 2019 to June 2020. The research was approved by the hospital's ethical committee (MCME.CR.83.MJOW.2019).

An informed consent form was signed by the caretakers of all the babies. The babies' nationality, gender, and risk factors (such as breech presentation, positive family history, primiparity, body weight over 4 kg, twin pregnancy, and oligohydramnios) were noted and recorded. Deformities of the musculoskeletal system and abnormal hip findings (AHF), including Barlow test, Ortolani test, leg-length discrepancy, and limited-hip abduction (LHA), were also noted. The harmful effects of TLS were explained, a wide blanket-folding technique was demonstrated to the parents and the instructional leaflet was given (**Figure 1**).

A total of 391 deliveries of healhy newborns were recorded at the center during the study period. Graf ultrasonography technique was carried out on 326 (83%) babies during workdays in a maternity ward in the presence of parents or relatives. Outpatient appointments were scheduled for 65 babies who were discharged on weekends and thus missed GUS while in the hospital. Only 28 (43%) babies out of the 65 were brought to the outpatient clinic. This resulted in a cohort that consisted of 354 neonates (175 girls and 179 boys). The average age at GUS was 3 days (**Figure 2**).

The cohort consisted of 34 different nationalities. A total of 148 (42%) babies were Emiratis, while 21 were from other countries of the GCC region (Oman, Yemen, KSA, and Bahrain). Together, they formed the GCC group (47.7%). The rest of the newborns constituted the diverse non-GCC group (n=185). It consisted of 29 nationalities grouped regionally. The incidence of the regions is presented in **Figure 3**.

An examination cradle was padded with lateral supports for the babies' standard positioning. The classification system developed by Graf was used to differentiate mature hips (type Ia)- the mature group, from immature ones (type IIa)- the immature group, and dysplastic hips (types IIc, D, III, and IV)-the DDH group.

The incidence of dysplastic and immature hips was assessed in the whole cohort and in the GCC- and non-GCC groups in relation to the risk factors.

Figure 1 - Instructional leaflet indicating proper and improper legs wrapping techniques.

All babies with dysplastic hips were treated using Pavlik harnesses. The harnesses were applied the same day of the DDH diagnosis.

For babies with immature hips, double diapering and a 6-week follow-up were advised and parents were instructed on how to carry the baby and allow their legs free movement while keeping them covered. If the hip maturation was not achieved even at 6 weeks, a new appointment was given for 3 months.

Statistical analysis. Chi-square test was used to compare the proportions between the 2 groups and to study the associations. Fisher's exact probability test was used for the small cell numbers. The data were analyzed using the Statistical Package for the Social Sciences, 24.0 software (IBM Corp., Armonk, NY, USA).

Results. The incidence of DDH was 1.7% for the cohort (Table 1). It was 0.6% for boys, which was significantly less compared to the 2.9% for girls. The incidence of DDH was higher in the GCC group (2.9%) than in the non-GCC group (0.5%). Girls from the GCC group had a significantly higher incidence of DDH (6.3%), as no case of DDH was diagnosed among girls in the non-GCC group.

Immature hips were present in 32 (9%) babies, of which 27 babies had unilateral immature hips (22 left and 5 right), while 5 babies had immature hips on both sides (total 37 hips). Occurrence of hip immaturity was similar regardless the babies' nationality (Table 1). The incidence of immature hips was significantly higher among girls (12%), and the GCC-region girls (15.2%; Table 1).

A total of 6 neonates (5 girls and one boy), had dysplastic hips at birth, constituting the DDH group. Of the 6, 5 were from the GCC countries (4 Emiratis and one Saudi). The details are presented in Table 2.

The presence of risk factors was assessed in relation to the level of hip development (Table 3). Most of the newborns in the DDH group were girls (83%). The incidence of breech presentation was higher in the DDH group (16.6%) when compared to the mature group. It was even higher among girls with DDH (20%), with the highest being recorded for girls with DDH from the GCC region (25%), although it was not statistically significant. Overall, one third (33.3%) of the babies

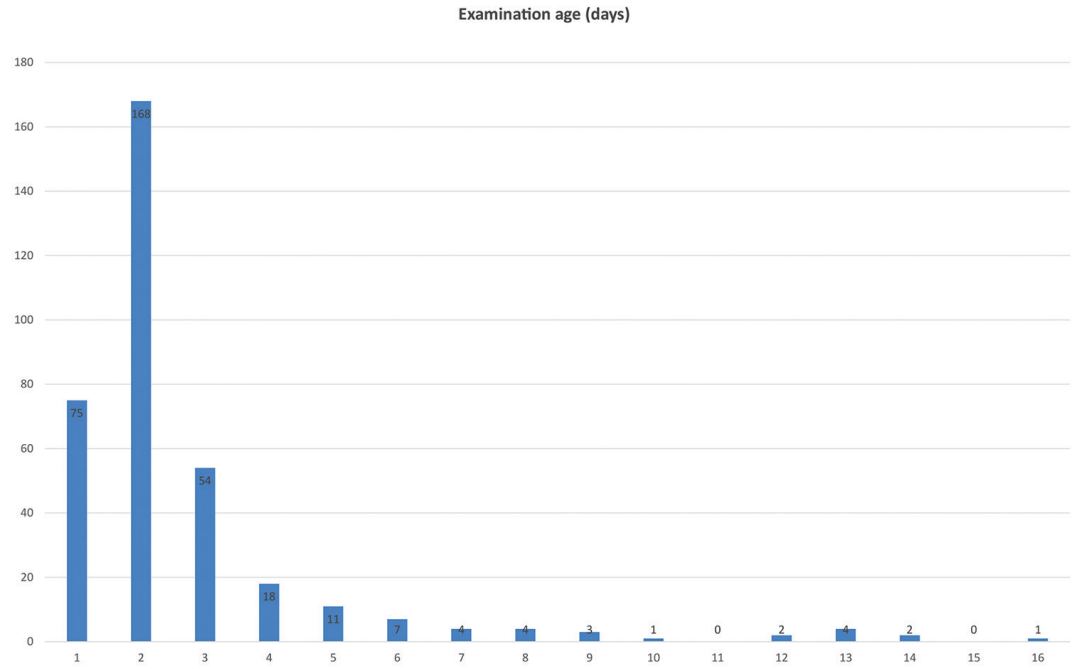

Figure 2 - Age distribution of the neonates on the day of examination.

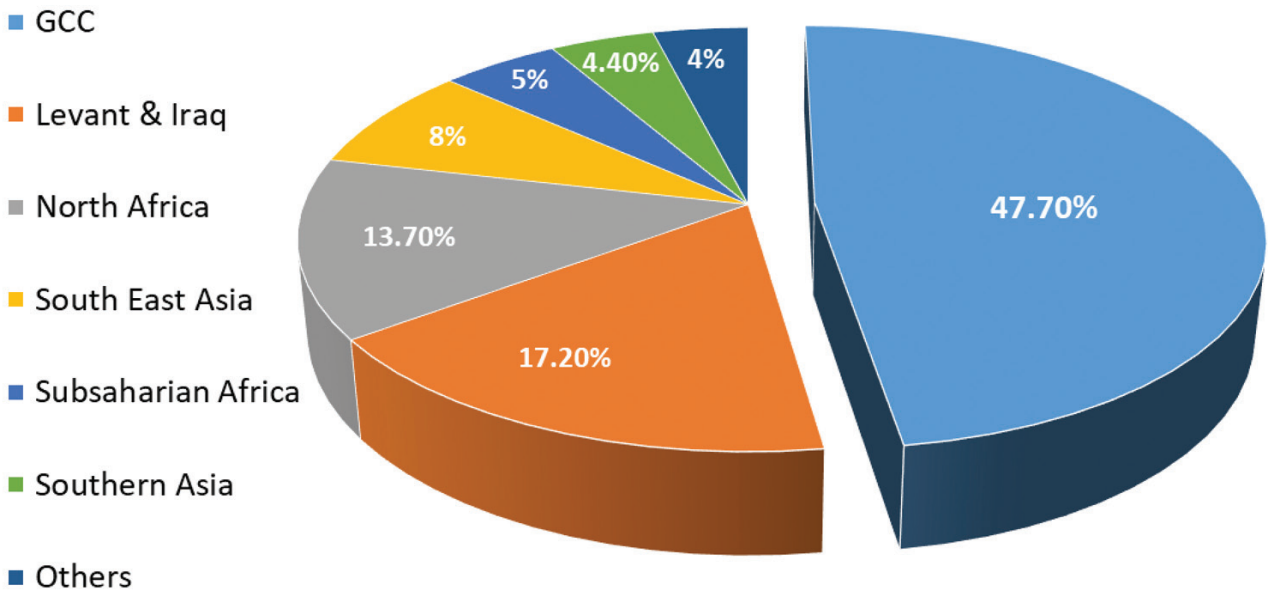

Figure 3 - Demographic distribution. GCC: Gulf Cooperation Council

with dysplastic hips had AHF, which is significantly more compared to the mature group (1.6%).

Positive family history was noted almost to a similar extent in the immature group (15.6%) and among girls from the GCC region (16.7%), which was significantly higher compared to the mature group. Primiparity was noted more frequently in the immature group and significantly more often among girls from the

non-GCC regions (55.5%). Limited-hip abduction was found significantly more often in the immature group (18.8%) than in the mature group (2.5%). There was no significant difference in the incidence of AHF in the DDH and the immature groups (Table 3).

On one hand, those with dysplastic hips were treated with Pavlik harnesses. The parents and family members of the neonates in this group were very cooperative. On the other hand, those in the immature group were only advised to apply double diapers, keep the baby's legs free of movements and come for follow-up. Overall no-show rate on follow-up was 43.7%.

Discussion. The incidence of DDH observed in this study is consistent with the results of similar prospective studies that used GUS in universal neonatal screening (1.3-5.7%).[14-16] Only one prospective study that used GUS on 276 referred infants (1-6 months old, 72.5% girls) was published in the GCC region.[17] Reported DDH incidence was 6.5%, which corresponds with the incidence of DDH among girls from the GCC region in the present study.

Female gender bears a high risk for DDH, yet it is not regularly listed among the risk factors in the SUS guidance. However, a risk model including the female gender was proposed recently.[18] The reasons behind the gender risk could be related to increased ligament laxity in females due to hormonal influence.[2] Laxity by itself is not the main cause of DDH, and it usually diminishes in the first months of life. During this period, proper contact between the femoral head and acetabulum contributes to hip maturation. The tradition of TLS restricts adequate acetabular growth. Though it has been eradicated widely, it still has a stronghold in the GCC region.[9]

Despite the genes responsible for DDH having been identified, a need for established clinical intervention has not been advised.[19] However, the whole picture changes when consanguinity is taken into account. In the KSA, 20 years ago, almost half of the DDH patients were born of consanguineous parents. More than half the marriages in the GCC region take place among relatives.[10] Consequently, the genetic pool gets tighter, and each generation is under a higher risk for DDH. Al Ain, in particular, is the city with the highest rate of consanguinity in the UAE (54%), and high incidence of positive family history of DDH among locals was expected.[10] However, although the DDH group in this study consisted mainly of girls from the GCC region, this risk factor was missing. A possible explanation for this could be that the study sample was too small or that the narrowing of genetic pool finally led to DDH in the

Table 1 - Incidence of different types of hip maturity in the whole cohort and according to the gender and origin.

Variables	Mature group	DDH group	Immature group
Cohort	316 (89.3)	6 (1.7)	32 (9.0)
Female gender	149 (85.1)	5 (2.9)*	21 (12.0)§,‡
Male gender	167 (93.3)	1 (0.6)*	11 (6.1)§
GCC	149 (87.6)	5 (2.9)**	16 (9.5)
Non-GCC	167 (90.9)	1 (0.5)**	16 (8.6)
GCC female	62 (78.5)	5 (6.3)†	12 (15.2)‡
Non-GCC female	86 (90.6)	0 (0.0)†	9 (9.3)

Values are presented as numbers and precentages (%). *P-value of 0.034, **p-value of 0.076, †p-value of 0.01, ‡p-value of 0.0465, and §p-value of 0.483. DDH: developmental dysplasia of the hip, GCC: Gulf Cooperation Council

Table 2 - Developmental dysplasia of the hip group.

Nationality	Side	Risk factors	Clinical findings	Other deformities	Graf type
UAE	R	Nil	Normal	Nil	IIc
Morocco	R	Nil	Normal	Nil	IIc
UAE	L	Nil	Normal	Nil	D
UAE	R	Nil	Limited abd. bil.	Nil	D
KSA	L	Nil	Barlow and Ortolani	Nil	III
UAE	L	Primiparity and breech	Normal	Nil	III

UAE: United Arab Emarites, KSA: Kingdom of Saudi Arabia, R: right, L: left, Nil: none, abd. bil.: abduction bilateral

Table 3 - Incidence of risk factors considering hip maturity, female gender, and origin.

Risk factors	Female	Family hx.	Breech	Primiparity	AHF
Cohort	175 (49.4)	10 (2.8)	25 (7.1)	71 (20.9)	13 (3.6)
Mature hips	149 (47.1)[*†]	5 (1.6)[‡§]	23 (7.3)[**]	63 (19.9)[††§§]	5 (1.6)[§§]
DDH group	5 (83.3)[*]	0 (0.0)	1 (16.6)	1 (16.6)	2 (33.3)[§§***]
DDH girls	-	0 (0.0)	1 (20.0)	1 (20.0)	2 (40.0)
DDH GCC girls	-	0 (0.0)	1 (25.0)[**]	1 (25.0)	2 (50.0)
DDH non-GCC girls	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)
Immature group	21 (65.6)[†]	5 (15.6)[§]	1 (3.1)	10 (31.2)	6 (18.7)[§§***]
Immature girls	-	2 (9.5)	1 (4.8)	7 (33.3)[††]	3 (14.3)
Immature GCC girls	-	3 (16.7)[‡§]	0 (0.0)	3 (16.7)	3 (16.7)
Immature non-GCC girls	-	0 (0.0)[§]	1 (11.1)	5 (55.5)[§§]	1 (11.1)

Values are presented as numbers and precentages (%). [*]*P*-value of 0.078, [†]*p*-value of 0.04, [‡]*p*-value of 0.000, [§]*p*-value of 0.004, [§]*p*-value of 0.197, [**]*p*-value of 0.182, [††]*p*-value of 0.14, [§§]*p*-value of 0.009, [§§]*p*-value of 0.01, and [***]*p*-value of 0.42. hx.: history, AHF: abnormal hip findings, DDH: developmental dysplasia of the hip, GCC: Gulf Cooperation Council

last generation. On the other hand, in the immature group and in the subgroup of GCC girls with immature hips, the rate of positive family history was found to be significant.

The latest National Health Service guidlines define a high risk for DDH if there is a breech presentation at birth or anytime at or after the 36th week of gestation, irrespective of presentation at birth or mode of delivery.[20] In this study, the information regarding the breech presentation in the last 4 weeks of pregnancy was collected by interviewing the mothers, so it is possible that some data of their obstetric history might be missing. For the babies with breech presentation at birth, supportive medical documentation was available, and they were all delivered by cesarean section. The incidence of breech presentation in the DDH group and the subgroup of girls from the GCC region matches the results obtained previously from the region.[11]

Clinical examination of the newborns' hips requires skilled hands.[2] In this study, AHF was significantly associated with both hip dysplasia and physiological immaturity. The clinical examination was carried out only once, just before the GUS. There was no need for repetitive maneuvers, as suggested in studies dealing with clinical or selective ultrasonography screening, because all structures could be analyzed precisely by the ultrasonography.[2,21,22] On the other hand, during the neonatal period, clinical tests have a limited sensitivity.[23] Even without AHF, the combination of female gender and major risk factors was strong enough for the recently proposed model of SUS.[18] Given the above-mentioned data, the role of clinical examination of neonatal hips in DDH screening should be reconsidered and, by introducing UUS, could even be skipped.

Incidence of immature newborn hips in this study was 9%, consistent with the results of Tonnis et al.[23] Though sufficient development happens in the first 3 months in the majority of immature hips, the condition could deteriorate; therefore, close GUS surveillance is needed.[24] In the present study, full maturation was found in all immature hips that completed follow-up until 3 months after birth. However, it is unclear how many dysplastic hips remained undiagnosed among the substantial number of no-shows in the immature group and how many of them were subjected to TLS once out of hospital. The high no-show rate is a strong indicator of a low social awareness of DDH in the UAE.

Study limitations. The given size of the cohort led to small numbers of neonates in the subgroups and, potentially, could result in an insufficient statistical outcome in the risk factors analysis.

In conclusion, the deep-rooted tradition of TLS and high consanguinity rate combined with low social awareness put all babies from the GCC region, and baby girls in particular, under a high risk for DDH, justifying the need for a legally established, preferably universal, ultrasonography DDH screening program in the UAE. Meanwhile, in-house screening is currently minimizing the number of babies who are not brought for consultation, and caretakers are being informed regarding the harmful effects of TLS.

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