J Membrane Biol (2013) 246:115–119
DOI 10.1007/s00232-012-9509-4

Are Consanguineous Marriage and Swaddling the Risk Factors of Developmental Dysplasia of the Hip?

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Received: 11 September 2012 / Accepted: 10 October 2012 / Published online: 19 October 2012

Abstract The purpose of this study was to investigate prospectively the effects of swaddling and consanguineous marriage on developmental dysplasia of the hip and associated risk factors. We screened by ultrasound 265 infants using the Graf method. The Pediatrics Department referred all newborn infants with suspected instability or a recognized risk factor to the orthopedic clinic. Risk factors of developmental dysplasia of the hip were searched and noted in these patients. Swaddling and consanguineous marriage were also determined and noted. We observed 164 of 265 infants (61.9 %) who had been swaddled and that 64 of 265 infants' parents were in a consanguineous marriage (24.2 %). In the statistical analysis that was conducted for swaddling and consanguineous marriage, highly significant differences were found. Our study showed that the rate of developmental dysplasia of the hip is very high, 11.7 %, in our region, eastern Turkey. Also, we commonly see improper swaddling and consanguineous marriage in our region, which affects many infants.

Keywords Hip dislocation · Congenital · Risk factor · Swaddling · Consanguineous marriage

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Introduction

Developmental dysplasia of hip (DDH) refers to aberrance in the volume, shape, direction or organization of the femoral head, acetabulum or both. Acetabular dysplasia is characterized by an immature, shallow acetabulum and can result in subluxation or dislocation of the femoral head. In a subluxed hip the femoral head is displaced from its normal place but still makes contact with a part of the acetabulum. With a dislocated hip, there is no relationship between the articular surface of the femoral head and the acetabulum (Storer and Skaggs 2006). In the spectrum of DDH, severe dysplasia or irreducible dislocation with early degenerative changes may require total hip replacement in early adulthood (Engesaeter et al. 2003). DDH can be treated nonsurgically if it is determined early. Newborn screening with ultrasound, which has been practiced at the first month after birth, is intended to decrease the need for surgery and to prevent degenerative joint disease, pain and mobility limitations (Lehmann et al. 2000).

There is a current trend toward swaddling to help reduce crying and support uninterrupted sleep in neonates. There

is a paucity of experimental evidence on the developmental effects of this frequent practice, despite frequent speculation as to the risks and benefits of swaddling (Bener et al. 2007). The incidence of consanguineous marriage (CM) has decreased considerably over the past decades worldwide. On the other hand, CM is still very common in certain places, especially in countries of the Middle East, northern Africa and southern Asia (Mahan and Kasser 2008).

The purpose of this study was to investigate prospectively the effects of swaddling and CM on DDH and associated risk factors and to detect DDH ultrasonographically in infants who participated in a screening program for this disease.

Patients and Methods

This prospective study was performed between January 2010 and September 2011 at the Medical School of Yuzuncu Yil University in Turkey. We informed 834 mothers who gave birth in our Gynecology Department about DDH and then advised them to come to the Pediatric Department for an examination when the infant was 4 weeks old. Of the 834 infants 265 were brought for the ultrasound scan by their parents. The Pediatrics Department referred all newborn infants with suspected instability or a recognized risk factor to the Orthopedic Clinic. Risk factors of DDH were searched and noted in these patients (gender, family history, breech delivery, oligohydramnios and skeletal deformities). Swaddling and CM were also determined and noted.

Ultrasonographic assessment of the hips and physical examinations were performed by the same pediatric orthopedic surgeon. The physician examined all the hips clinically using the Barlow and Ortolani tests and ultrasonographically by Graf's static morphological methods (Graf et al. 1993).

Ultrasonographic Evaluations

Ultrasonography was performed with a 7.5 MHz linear probe (LOGIQ C2; GE Healthcare, Beijing, China). Hip ultrasonography was performed on all of the infants. All ultrasonographic examinations were performed according to the technique described by Graf (1980). Type II, III and IV hips were considered to be DDH.

Graf Classification

The Graf classification of infant hips is based on the depth and shape of the acetabulum as seen on coronal ultrasonograms (Graf 1980) (Fig. 1).

Type I: Normal, characterized by a well-formed acetabular cup with the femoral head beneath the acetabular roof

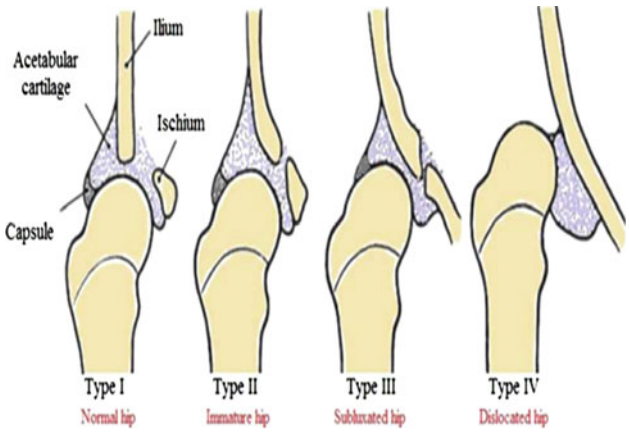

Fig. 1 Graf classification of infant hips

Type II: Immature in infants less than 3 months of age and mildly dysplastic in infants older than 3 months, characterized by a shallow acetabulum with a rounded rim

Type III: Subluxated, characterized by a very shallow acetabulum with some displacement of the femoral head

Type IV: Dislocated, characterized by a flat acetabular cup and loss of contact with the femoral head

Statistical Analysis

The data were computerized and analyzed with the SAS V9.2 (SAS Institute, Cary, NC) statistical package, using Proc Freq and Proc logistic. Statistical analyses were performed with chi-squared and multiple logistic regression analyses. Descriptive summaries were obtained for all variables of interest, using frequency tabulations for categorical variables. The chi-squared analysis was used to evaluate the association of gender, family history, breech delivery, oligohydramnios, skeletal anomaly, swaddling and CM with DDH. The effect of each of the covariates (as fixed effects) on the binary outcome of DDH was determined with a logistic regression model. Odds ratios (ORs) and corresponding 95 % confidence intervals (CIs) are reported. $p < 0.05$ was considered significant.

Results

This study was performed on 265 infants, and DDH was observed in 31 (11.7 %) of them. Of these cases, 11 (35.5 %) were on the left side, 9 (29 %) were on the right side and 11 (35.5 %) were bilateral (Fig. 2). The distributions of risk factors are shown in Fig. 3.

Gender

There were 128 male (48.3 %) and 137 female (51.7 %) infants. The rate of occurrence of DDH was statistically

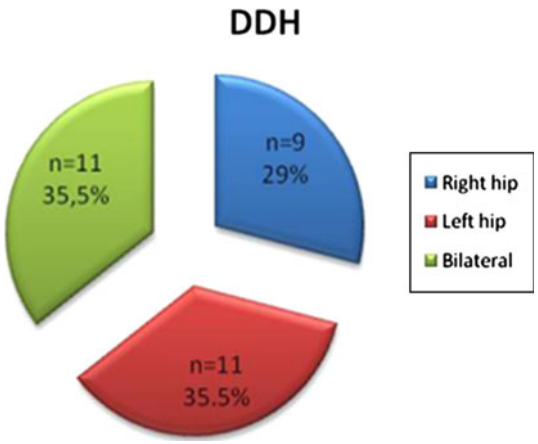

Fig. 2 Distribution of developmental DDH localization

significant between girls and boys (OR = 0.427, 95 % CI 0.185–0.986, $p < 0.05$).

Family History

There were 33 of 265 infants with a positive family history (12.5 %). This factor considerably increased the risk compared with infants with no family history of DDH (OR = 3.131, 95 % CI 1.190–8.237, $p < 0.05$).

Breech Delivery

There were 23 of 265 breech presentations (8.7 %) in our study. No statistically significant differences were found between breech delivery and nonbreech delivery (OR = 0.577, 95 % CI 0.118–2.834, $p > 0.05$).

Oligohydramnios

Sixteen of 265 cases had oligohydramnios (6 %). Statistical analysis for oligohydramnios associated with DDH showed no significant difference (OR = 0.315, 95 % CI 0.036–2.777, $p > 0.05$).

Skeletal Anomalies

There were 22 of 265 infants with skeletal anomalies (8.3 %). No statistically significant differences were found in infants with skeletal anomalies and DDH (OR = 1.844, 95 % CI 0.535–6.364, $p > 0.05$).

Swaddling

We observed 164 infants (61.9 %) on whom swaddling was used. The risk was highly significant for swaddling (OR = 2.652, 95 % CI 1.004–7.005, $p < 0.05$).

Consanguineous Marriage

We found 64 of 265 infants were of parents with a CM (24.2 %). Highly significant differences were found on statistical analysis for CM (OR = 2.613, 95 % CI 1.146–5.959, $p < 0.05$).

Discussion

Risk Factors for DDH

Female gender has been proposed as a risk factor for DDH (Herring 2002). We also observed this as 67.7 % of females in our study had DDH. We noted that the DDH rate among girls was similar that reported in the literature. This association may be attributed to potentiation by endogenous estrogens produced by the female infant and the transiently increased ligamentous laxity in the perinatal period caused by high levels of circulating maternal hormones. Female susceptibility to relaxin hormone and

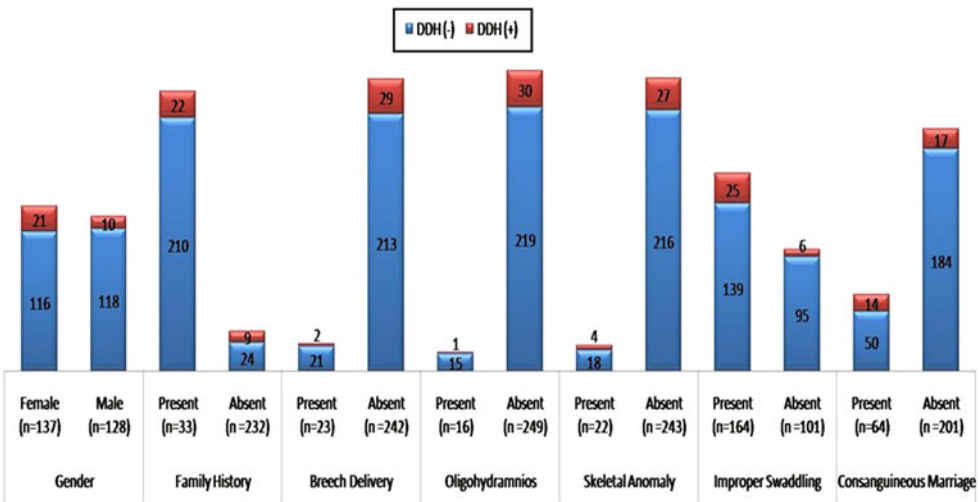

Fig. 3 Distribution of risk factors

possible relaxin hormone receptor sensitivity have been discussed in the literature (Grill et al. 1988; MacLennan and MacLennan 1997).

Family history has been reported previously as a risk factor for DDH (MacLennan and MacLennan 1997). Our findings support the relevance of familial or genetic factors in DDH. There was a family history of DDH in 29 % of children, which is compatible with the incidence in the literature of 10–28 % (Herring 2002; Grill et al. 1988).

Breech delivery has been described as a risk factor for DDH in terms of the intrauterine position concerned, in which lower extremity movement is restricted and knee joint extension can strain the hamstring and thereby increase the possibility of DDH (Palmen 1984). In our study, no statistically significant association was found with breech delivery and DDH.

Oligohydramnios as a risk factor has increased in recent years. However, we did not find any statistically significant differences in the study. Oligohydramnios is not a risk factor unless it is associated with syndromes such as arthrogryposis (Paton et al. 2005), but this needs further investigation.

Skeletal anomalies have been studied as a risk factor for DDH (Noordin et al. 2010). However, in our study, no statistically significant association was observed between skeletal anomalies and DDH.

Swaddling and Consanguineous Marriage

Swaddling is recognized to be a very old technique; it helps to reduce crying and support uninterrupted sleep in neonates (Franco et al. 2005). Even if swaddling has been recognized to be a risk factor for DDH (Kutlu et al. 1992), there are limited data in the literature. We found a statistically highly significant association with DDH. Improper swaddling fixes the hips in an extended and adducted position. The extension and adduction may cause hip dislocation during the neonatal period (Ishida 1977) as revealed experimentally in animal hips (Wang et al. 2012). For several cultures in which improper swaddling has been particularly common, a higher ratio of DDH has been observed, including in Turkey (Kutlu et al. 1992), Japan (Ishida 1977), Saudi Arabia (Kremli et al. 2003) and the Navajo nation (Pratt et al. 1982). Allowing even tightly swaddled infants to still have this flexion and abduction in their hips would allow for safe development of the hips (Ishida 1977) (Fig. 4). Families who improperly swaddle their infant should recognize the possible risk to the hips, and techniques that allow the hips to be abducted and flexed should be underlined. According to the American Academy of Pediatrics clinical practice guidelines, if dysplasia is found, swaddling should be avoided (American Academy of Pediatrics 2000).

There is no data about the relationship between CM and DDH in the scientific literature. We found a statistically significant association between CM and DDH. The incidence of CM has decreased considerably over the past decades worldwide. Though CM is observed at a rate of 1 % in developed Western countries, is extremely high in Eastern and Middle Eastern countries and isolated communities (Koc 2008). The CM rate was reported to be

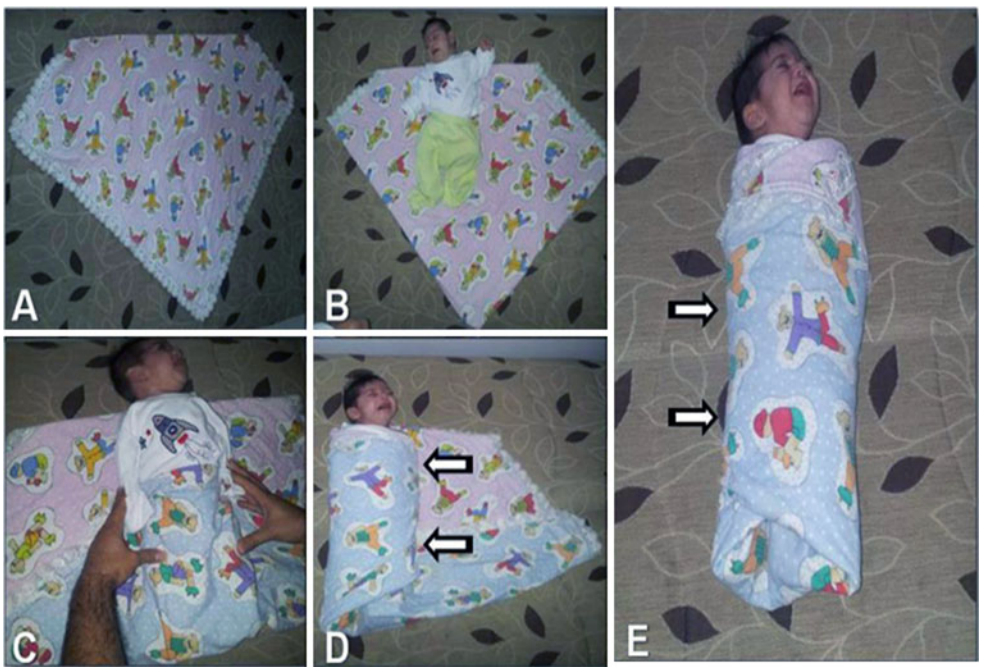

Fig. 4 Stages of improper swaddling

0.56 % in England (Hajnal 1963), 0.36 % in the Netherlands (Van Straaten 1986), 54.3 % in Kuwait (Ai-Awadi et al. 1985), 12.9 % in India (Bittles 2002) and 34.4 % in Turkey (Akbayram et al. 2009). The CM rate was 24.2 % in our study population. CM is associated with a higher rate of genetic disorders in the offspring. Also, the frequency of congenital malformations is approximately 2.5 times higher in the offspring of first cousins than in the offspring of unrelated parents (Akrami et al. 2009).

DDH is a frequent childhood disorder, affecting about 20 per 1,000 at birth (Campion and Benson 2007). We found in this study population that the incidence of DDH was 117 per 1,000 at birth. The fact that a remarkably high incidence of DDH was found in the Van population compared with other European rates may be related to the traditional swaddling and CM that are extensively practiced.

Conclusions

In this study, the role of risk factors in the diagnosis of DDH was analyzed. The incidence of DDH appears to be significantly higher in the presence of improper swaddling and CM. This is the first study to examine the effect of CM on DDH and needs to be supported by further study.

Conflict of interest The authors declare that they have no conflict of interest

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