Breech Presentation in Twins as a Risk Factor for Developmental Dysplasia of the Hip

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Introduction: Identifying risk factors associated with developmental dysplasia of the hip (DDH) is essential for early diagnosis and treatment. Breech presentation is a major DDH risk factor, possibly because of crowding of the fetus within the uterus. In multifetal pregnancy, fetuses are generally smaller than singletons, which may obscure the effect of breech presentation on fetal hips. Only a few studies have investigated the occurrence of DDH in multifetal pregnancies. In this study, we aimed to evaluate whether the breech presentation is a major risk factor of DDH in twin pregnancies.

Methods: This retrospective study included 491 consecutive live births (after 23+0 weeks gestation) delivered through cesarean section with at least 1 baby with noncephalic presentation in single or twin pregnancies from April 2013 to October 2018. We analyzed the incidence of DDH and its associated factors, including sex, breech, and multifetal pregnancy, with a generalized linear mixed model.

Results: The incidence of DDH was 12.5% in singleton with breech presentation, 9.8% in twin-breech presentation, and 0.7% in twin-cephalic presentation. Multivariate analysis showed that singleton-breech presentation (P=0.003), twin-breech presentation (P=0.003), and female sex (P=0.008) were independent risk factors for DDH.

Conclusion: Breech presentation is an independent risk factor for DDH in twin pregnancies, although twin pregnancy itself is not an independent risk factor for DDH.

Key Words: breech presentation, developmental dysplasia of the hip, multifetal pregnancy, twin pregnancy

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Developmental dysplasia of the hip (DDH) refers to a wide spectrum of pathologic disorders of the hip and is mainly caused by the displacement of the femoral head from the acetabulum. The incidence of DDH is 1% to 7% in newborns.1–4 Since early diagnosis and treatment improves the prognosis of DDH, various studies have focused on evaluating its risk factors and screening strategies.5 Breech presentation, family history, and female sex are well-known risk factors for DDH.3,6,7

Breech presentation is a major DDH risk factor, possibly because of crowding of the fetus within a limited intrauterine space, limiting fetal movement that forces their hips into adduction.8 In recent years, multifetal pregnancies, including twins and triplets, have increased because of the growing rates of assisted reproductive technology and advanced maternal age.9 Several studies investigated multifetal pregnancy as a DDH risk factor because of the assumption that the intrauterine environment in multifetal pregnancies is likely to be cramped. Conversely, it has been speculated that babies in multifetal pregnancies tend to be cramped. However, unlike singletons, the presentation combinations during delivery in multifetal pregnancies vary, making it difficult to assess the breech presentation as an independent risk factor of DDH.

Therefore, in this study, we aimed to evaluate whether the breech presentation was a major risk factor of DDH, even in twin pregnancies.
METHODS

Study Design

This retrospective study was approved by the institutional review board of our hospital, a tertiary referral center for multifetal pregnancies and DDH (B-1905/544-103). The need for informed consent was waived because of the retrospective nature of the study.

Several consensus-building sessions comprised of pediatric, obstetric, and orthopaedic departments were held in April 2013. Since then, it is recommended that every singleton-breech and multifetal pregnancies with breech or T-lie and their twin counterpart delivered in our hospital be referred for pediatric orthopaedic surgeon for assessment of DDH before discharge either in the nursery or in the neonatal intensive care unit. An additional sonographic screening for DDH 6 weeks after birth has been recommended.

We searched for patients based on the International Classification of Diseases codes for breech (O32.1) or multifetal pregnancies (O30.042) from April 2013 to October 2018 through the clinical data warehouse in our hospital [Healthcare Information and Management Systems Society (HIMSS), stage 7]. A total of 1256 patients were screened (Fig. 1).

Inclusion criteria were (1) consecutive live births through cesarean section, (2) more than 23 weeks in gestational age, (3) singletons in the breech presentation or twin pregnancies with at least 1 fetus in the breech presentation, and (4) those who have undergone a pediatric orthopaedic consultation for DDH. The exclusion criterion was (1) twin pregnancies in the transverse-lie presentation.

A consensus-building session was held among the authors (E.J.O., J.Y.P., J.J.M., and M.S.P.), and definitions for each variable in this study were set. DDH was defined as the sonographic diagnosis of type Graf IIb or a more severe type in either of the hips. An oligohydramnios diagnosis was made when a single deep pocket of amniotic fluid below 2 cm in antenatal ultrasound was noted during the second and third trimesters.14,15

After consensus building, 2 obstetricians (E.J.O. and J.Y.P.) with 3 and 10 years of experience, respectively, reviewed the electronic medical records for obstetric and neonatal information (parity, fetal position, mode of delivery, gestational age at delivery, birth weight, and the presence of oligohydramnios). A review of medical records containing orthopaedic information, including a diagnosis of DDH and a sonographic finding of the hip, was performed by 2 orthopaedic surgeons (J.J.M. and M.S.P.) with 3 and 19 years of experience, respectively. The incidence of DDH and the potential influencing factors were analyzed using a generalized linear mixed model (GLMM).

Statistical Analyses

The Kolmogorov-Smirnov test was used to verify the normal distribution of continuous variables. Descriptive variables were presented using medians, range, and frequencies. Continuous variables of the independent group were compared using a Mann-Whitney U test and proportions were compared using the Fisher exact test.

The factors that influence the incidence of DDH were assessed using a GLMM with sex, maternal age, gestational age at delivery, body weight, presence of oligohydramnios, and presentation as the fixed effects and fetuses with the same mother as the random effect. The GLMM estimated data with correlations or nonconstant variability and where the response was not normally

FIGURE 1. A flow diagram of the study population. DDH indicates developmental dysplasia of the hip.
distributed. The covariance structure was assumed as the variance component. The restricted maximum likelihood estimation was used to estimate parameters for the GLMM. The models were accepted as valid for estimation of responses using the Akaike Information Criterion and the Bayesian Information Criterion. A smaller Akaike Information Criterion or Bayesian Information Criterion value was preferred regarding model selection.

All statistical analyses were performed using the SAS statistical package, version 9.4 software (SAS Institute, Cary, NC) and R (version 3.5.1) (R Foundation for Statistical Computing, Vienna, Austria; ISBN 3-900051-07-0, URL http://www.r-project.org) with the stats package. All statistical tests were 2-tailed. Confidence intervals were considered significant when they did not include the value 0, and \( P \)-values of < 0.05 were considered statistically significant.

## RESULTS

After implementing the inclusion and exclusion criteria, 491 cases were included in this study (Fig. 1). Among them, 152 were singleton-breech, 204 were twin-breech, and 135 were twin-cephalic pregnancies (Table 1). The incidence of DDH diagnosis was 12.5% (19/152) in singleton-breech, 0.7% (1/135) in twin-cephalic, and 9.1% (20/204) in twin-breech pregnancies.

In cephalic-first and breech-second twins, 10.2% (9/88) of the breech fetuses developed DDH. In breech-first and cephalic-second twins, 8.5% (4/47) of the breech fetuses developed DDH. In twins with both fetuses in the breech position, the incidence of DDH was 10.3% (3/29) for both fetuses (Fig. 2).

A GLMM showed that singleton-breech presentation \( (P = 0.003) \), twin-breech presentation \( (P = 0.003) \), and female sex \( (P = 0.008) \) were independent risk factors for DDH. In our analysis, female neonates had a 6.6% higher risk of DDH compared with male patients; singleton neonates with a breech presentation had a 10.2% higher risk of DDH compared with twin-cephalic babies, and twin-breech babies had an 8.6% higher risk of DDH compared with twin-cephalic babies (Table 2).

## DISCUSSION

In this study, DDH was present in 12.5%, 9.1%, and 0.7% of singleton-breech, twin-breech, and twin-cephalic deliveries, respectively. This study showed that breech presentation was an independent risk factor for the diagnosis of DDH. The incidence of DDH did not differ among babies born through singleton-breech and twin-breech deliveries, but twin-cephalic births showed a significantly lower incidence of DDH compared with the other 2 groups. As a result, even in twin pregnancies, breech presentation is the strongest risk factor for the diagnosis of DDH.

Breech presentation is a well-known risk factor for DDH. A meta-analysis on various factors for DDH development reported that the breech presentation showed a strong relationship to DDH, with a pooled odds ratio of 5.7 (95% confidence interval: 4.4-7.4).\(^7\) The results of our study were consistent with those of previous studies that breech presentation is a risk factor of DDH in singleton pregnancy. Furthermore, our results show that breech presentation is an independent risk factor of DDH even in twin pregnancies.

Breech presentation as a risk factor for DDH is often explained by prolonged hip joint malposition in a cramped space.\(^10,16\) As the fetus number increases, the space inside the uterus becomes more crowded. A few studies suggested that multifetal pregnancies may protect against DDH, owing to a younger gestational age and lower birth weight compared with singletons.\(^11\) However, our study results showed that multifetal pregnancy was not a risk factor, nor was it a protective factor in the development of DDH.

### TABLE 1. Demographic Characteristics of the Study Population

|                  | Singleton-breech (n = 152) | Twin-breech (n = 204) | Twin-cephalic (n = 135) |
|------------------|---------------------------|----------------------|------------------------|
| Maternal age (y) | 34.1 ± 4.4                | 33.7 ± 3.3           | 33.9 ± 3.4             |
| Sex (female/male)| 77/75                     | 109/95               | 67/68                  |
| Gestational age at delivery (wk) | 36.6 ± 2.9 | 35.9 ± 1.9 | 35.9 ± 1.9 |
| Birth weight (g) | 2684.0 ± 685.7           | 2324.8 ± 457.4       | 2349.0 ± 395.0         |
| Oligohydramnios  | 11.2% (17/152)            | 2.9% (6/204)         | 0.0% (1/135)           |
| DDH              | 12.5% (19/152)            | 9.8% (20/204)        | 0.7% (1/135)           |

DDH indicates developmental dysplasia of the hip.

### FIGURE 2.

Diagrams showing the positions of twins and the incidence of developmental dysplasia of the hip: (A) cephalic-breech, (B) breech-cephalic, and (C) breech-breech (incidences are presented as percentage and number of cases in parenthesis). Only twins with both fetuses meeting the inclusion criteria were included in these diagrams.
The strengths of this study are the relatively large twin pregnancy sample size in a single medical center and the analysis of the various presentation combinations in twins. We considered all combinations, including cephalic-breech, breech-cephalic, and breech-breech presentations, although, in this study, the order of fetuses was not found to be a risk factor for DDH.

This study has limitations that should be addressed. First, the study was retrospective in nature. However, a GLMM was adopted to overcome this inherent limitation. Furthermore, this study does not compare the frank position and the complete position of breech presentation separately. Further research is needed to determine the positive effects of the flexed knee position during a breech presentation on DDH. Third, the study only included twins in the analysis of DDH in multifetal pregnancies. Owing to the rarity of triplet births, triplets were excluded from the analysis. An additional study on the DDH risk for triplets is warranted. Lastly, owing to its rarity, twins with a transverse-lie presentation were excluded from the statistical analysis. Therefore, whether transverse-lie is an independent risk factor of DDH warrants further study.

CONCLUSION

Breech presentation is a well-known risk factor for the diagnosis of DDH. Breech presentation is an independent risk factor for DDH in twin pregnancies, although twin pregnancy itself is not an independent risk factor for DDH. Extensive and careful screening of DDH is recommended in twin-breech babies.

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