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# Chapter 7

## Breech presentation and developmental dysplasia of the hip: a systematic review and meta-analysis

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*Submitted*

# Abstract

## BACKGROUND

Breech presentation is a known risk factor for developmental dysplasia of the hip (DDH). Whether this increased risk of DDH is similar for all breech presenting fetuses is not clear. The aim of this study was therefore to assess the association between different aspects of breech presentation and its relation with DDH by performing a systematic review and meta-analysis. In this review we focussed on three different aspects of breech presentation: premature breech presentation, breech presentation in multiple gestation pregnancies and breech presentation with successful external cephalic version (ECV).

## METHODS

We searched Medline, EMBASE and The Cochrane Library from conception until January 2015 for articles on the relation between breech presentation and DDH. We selected articles that reported on the incidence of DDH in neonates that were born after different types of breech presentation. These associations were expressed as common odds ratios with a 95% confidence interval.

## RESULTS

Our search identified 61 articles, of whom six were eligible for this review (five cohort studies and one case control study), reporting on 38,604 neonates. One cohort study found the risk of DDH in premature breech neonates (2.3%) to be comparable to breech neonates born at term (1.8%), (OR 1.27, 95% CI 0.25-6.4). The incidence of DDH was significantly lower in neonates born in breech presentation from multiple gestation pregnancies compared to neonates born in breech presentation from singleton pregnancies (OR 0.08 (95% CI 0.01-0.57)). No significant difference was found in the incidence of DDH, for neonates born in breech presentation from multiple gestation pregnancies compared to neonates born in cephalic presentation from singleton pregnancies (OR 0.30 (95% CI 0.06-1.5)). No significant difference was found in the incidence of DDH between neonates born in cephalic presentation after successful ECV and neonates with persistent breech presentation (OR 0.81 (95% CI 0.08-8.7)).

## CONCLUSIONS

This study gives more insight in the relation between DDH and breech presentation. DDH is confirmed to be associated with breech presentation, but this impact seems to be influenced by different factors such as multiple gestations. This knowledge can be used in improving existing and developing new screening programmes for DDH.

# Introduction

Developmental dysplasia of the hip (DDH) represents a spectrum of anatomical abnormalities in the shape, size and orientation of the femoral head, acetabulum or both. The definition of DDH is not well established and DDH covers a wide range of severity, ranging from minor dysplasia to irreducible dislocation. The precise aetiology of DDH is unknown, but genetic and environmental factors may act as internal or external influences.<sup>1</sup> From all environmental factors breech presentation seems to form the greatest risk for DDH. The relation between breech presentation and DDH is described in numerous cohort and case-control studies. In addition, there are two meta-analyses that quantify this relation. The first meta-analysis, that included 15 studies (632,359 children), showed that children born in breech presentation are significantly at risk for DDH (OR 3.75 (95% CI 2.25-6.24)).<sup>2</sup> The second meta-analysis assessing 22 studies (1,494,387 children) found an even stronger association (OR 5.7 (95% CI 4.4-7.4)).<sup>3</sup> Screening for DDH in the Netherlands is part of the programme for child health surveillance and takes place in publicly financed centres for the health surveillance and care for all neonates and children. The current Dutch screening strategy for DDH includes physical examination of all neonates at four weeks of age and additional ultrasound screening for DDH at three months in case of risk factors. Risk factors include a positive family history of DDH, congenital postural or foot deformities and breech presentation. Within this screening strategy, breech presentation is defined as breech presentation in the last trimester of pregnancy or breech delivery.

Whether all neonates born in breech presentation are at increased risk for DDH is not clear, as current meta-analyses do not specifically report on, premature neonates born in breech presentation, neonates born in breech from multiple gestations, or neonates born after successful external cephalic version (ECV). The aim of this study was therefore to assess the association between different aspects of breech presentation and its relation with DDH by performing a systematic review and meta-analysis.

## Materials and methods

### SEARCH STRATEGY

We performed a computerized search in MEDLINE, Embase and the current Cochrane databases from conception until January 2015, to identify articles reporting on the relation between breech presentation and DDH. We focused on three different aspects of breech presentation: premature breech presentation, breech presentation in multiple gestation pregnancies and breech presentation with successful ECV. We used the following keywords

“breech”, “breech presentation”, “developmental dysplasia of the hip”, “congenital hip dysplasia”, “congenital hip dislocation”, “hip dislocation”, “hip dysplasia”. References from identified publications were manually searched for additional relevant articles. The complete electronic search is available from the first author. Reference Manager 12 was used to manage the results of all searches.

## STUDY SELECTION

Studies were selected in a two-stage process. First the electronic searches were screened for eligible studies by title and abstract, after which all identified articles were retrieved in full. Case-control, cohort and cross-sectional studies were all eligible for inclusion if they reported on the relation between breech presentation and DDH, in case of premature birth, multiple gestations or after successful ECV. A two-by-two table had to be available from the articles, either directly or retractable from the information provided by the articles. Methodological quality of included studies was determined by using an adapted version of the QUADAS tool.<sup>4</sup> The study had a representative selection of patients when the neonates were consecutively selected and clear in- and exclusion criteria of the study population were provided. The description of the test was classified as adequate when it was clearly stated how the diagnosis DDH was made, either by clinical examination or ultrasound. The description of the diagnosis was representative if a clear ultrasonographic description of the diagnosis DDH was given or the endpoint was neonates treated for DDH. Study withdrawal was representative if a flow diagram was presented or withdrawal was clearly described in the methods.

## STATISTICAL ANALYSIS

From the data of each study, two-by-two tables were constructed. Odds ratios (OR) and their 95% confidence intervals (CI) were calculated. The  $I^2$  test was used to assess homogeneity using an  $I^2$  value of 30% as a threshold. A common OR with a 95% CI was calculated for each exposure by means of the Mantel-Haenszel method. If homogeneity was rejected, a random effects model was used to calculate the common OR. Review Manager 5 was used to construct forest plots and visualize the data.

# Results

The initial computerized search detected 231 studies, of which 61 were retrieved for complete assessment after reading title and abstract. Of these 61 studies, six studies were included in this review, reporting on 38,604 neonates. Figure 1 summarizes the process of literature identification and selection. Of the included studies, five were cohort studies and one was a case-control study. The number of neonates included in the studies ranged from 291 to 26,236. Four studies defined DDH by the number of neonates treated for the condition,<sup>5,6,9,10</sup> one study defined DDH as Graf type  $\geq$  IIa ( $\alpha < 55^\circ$ ),<sup>7</sup> one study defined DDH as Graf type  $\geq$  IIc.<sup>8</sup> Characteristics and quality of the included studies are outlined in Table 1 and 2.

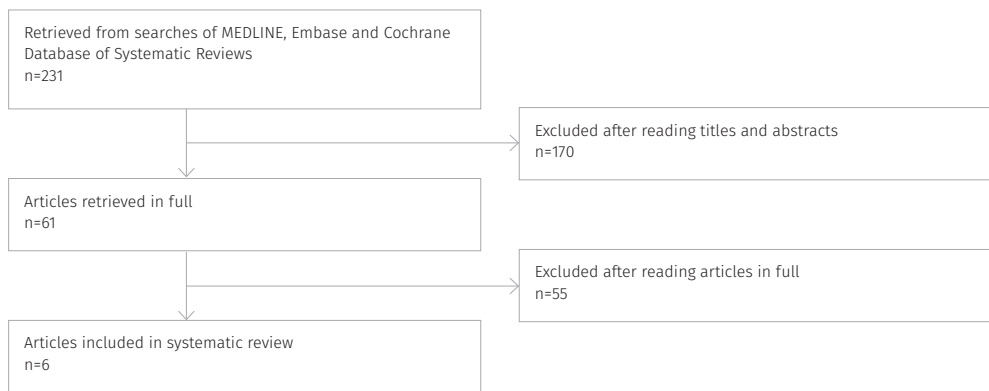


Figure 1 - Flow diagram of literature search

**Table 1 - Characteristics and study quality of the included studies**

Study	Study design	Included neonates	Prevalence DDH n (%)	Reference test	Definition of cases
Quan 2013	Retrospective cohort	Preterm breech n=129 Term breech n=163	3 (2.3) 3 (1.8)	Clinical examination Ultrasound	Treated for DDH
Ruhmann 2000	Retrospective cohort	Single gestation n=4379 Breech n=165 Multiple gestation n=97 Breech n=8	267 (6.1) 28 (17.8) 2 (2.2) 0 (0)	Ultrasound	≥ IIa ( $\alpha < 55^\circ$ )
De Pellegrin 2010	Case-control	Single gestation n=322 Breech n=48 Multiple gestation n=210 Breech n=92	(3.4) (8.3) (0) (0)	Ultrasound	≥ IIc
Barr 2013	Retrospective cohort	Single gestation n=25246 Breech n=not reported Multiple gestation n=990 Breech n=136	58 (0.2) 3 (0.3) 0 (0)	Clinical examination Ultrasound	Treated for DDH
Andersson 2000	Cohort	Cephalic presentation n=6571 Successful ECV n=62 Breech no ECV n=120 Unsuccessful ECV n=75	9 (0.1) 2 (3.2) 1 (0.8) 1 (1.3)	Clinical examination Ultrasound	Treated for DDH
Lambeek 2012	Cohort	Successful ECV n=177 Unsuccessful ECV n=321	5 (2.8) 30 (9.3)	Clinical examination Ultrasound	Treated for DDH

ECV; external cephalic version, DDH; developmental dysplasia of the hip

**Table 2 - Study quality**

Study	Representative selection of patients	Representative description of test	Representative description of diagnosis	Reporting study withdrawal
Quan	Yes	Yes	Yes	Yes
Ruhmann	No	Yes	Yes	No
De Pellegrin	Yes	Yes	Yes	Yes
Barr	No	Yes	Yes	No
Andersson	Yes	Yes	Yes	Yes
Lambeek	Yes	Yes	Yes	Yes

## PREMATURITY AND BREECH PRESENTATION

We found one retrospective cohort study reporting on the incidence of DDH in premature neonates born in breech presentation.<sup>5</sup> This study, examined the incidence of neonates treated for DDH in a retrospective cohort of 291 breech born neonates. There was no significant difference in the incidence of DDH in the group of preterm compared to the group of term breech neonates OR 1.27 (95% CI 0.25-6.4). There were three out of 129 (2.3%) neonates born in preterm breech, that were treated for DDH and three out of 163 (1.8%) term breech neonates. Preterm delivery was defined as a gestational age below 37 weeks. Of the three preterm neonates treated for DDH, two were born at a gestational age of 36 weeks and one at a gestational age of 29 weeks.

## MULTIPLE GESTATION AND BREECH PRESENTATION

Three studies investigated the incidence of DDH in breech born neonates from multiple gestation pregnancies, two cohort studies and one case control study.<sup>6-8</sup>

Pooling the data was possible for the comparison of the incidence of DDH in breech presenting neonates from multiple gestations to breech presenting neonates from singleton gestations in two studies.<sup>7,8</sup> Meta-analysis of these two studies found a significantly lower incidence of DDH in neonates born in breech presentation from multiple gestation pregnancies compared to neonates born in breech presentation from single gestation pregnancies OR 0.08 (95% CI 0.01-0.57, I<sup>2</sup> 16%), Figure 2. No significant difference was found in the incidence of DDH for neonates born in breech presentation from multiple gestation pregnancies compared to neonates born in cephalic presentation from singleton pregnancies in three studies,<sup>6-8</sup> OR 0.30 (95% CI 0.06-1.5, I<sup>2</sup> 20%), Figure 3.

The incidence of breech presentation in multiple gestations ranged from 8% to 44%, from the data reported in these studies it was not possible to determine whether the neonates born from multiple gestation pregnancies in breech presentation were the first or second twin. Second, the studies did not report which percentage of neonates were born preterm. Overall, in none of the studies, neonates born from multiple gestation pregnancies in breech presentation were diagnosed with DDH.<sup>6-8</sup>



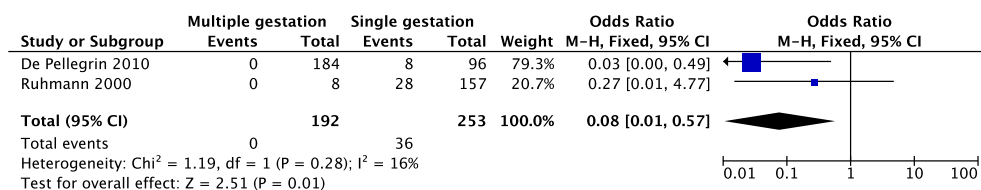


Figure 2 - Breech presentation in multiple gestation versus breech presentation in single gestation

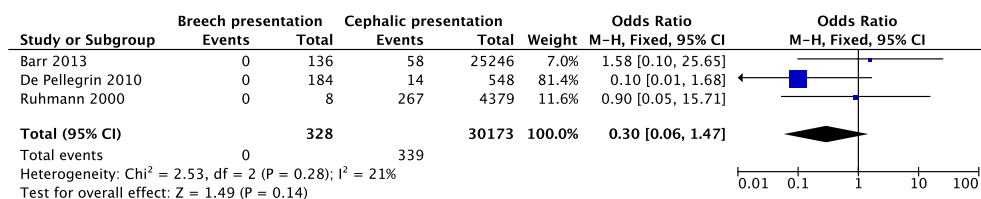


Figure 3 - Breech presentation in multiple gestation versus cephalic presentation in singleton gestation

## SUCCESSFUL ECV AND BREECH PRESENTATION

Two cohort studies reported on the incidence of DDH after successful ECV.<sup>9,10</sup> Meta-analysis of these two studies found no significant difference in the incidence of DDH in neonates born in cephalic presentation after successful ECV and persistent breech presentation (pooled OR 0.81, 95% CI 0.08-8.8,  $I^2$  79%), Figure 4. The first study was a cohort study on 6,571 women, of which 257 had a fetus in breech presentation after 36 weeks of gestation.<sup>9</sup> Of the women with a fetus in breech presentation 137 (53%) had an ECV attempt, of which 62 (45%) were successful. Of the neonates born in breech presentation, there were two out of 195 (1%) treated for DDH, compared to two neonates out of 62 (3.2%) born in cephalic presentation after successful ECV (OR 3.2 (95% CI 0.44-23.3)). Also, nine neonates (0.1%) with cephalic presentation were treated for DDH. There was a significant difference in the incidence of neonates treated for DDH, between neonates delivered in cephalic presentation after successful ECV (3.2%), or naturally cephalic presentation (0.1%) (OR 4.7 (95% CI, 1.1-16.0)). The second study was a cohort of 498 women with a fetus in breech position after 34 weeks of gestation, without a positive family history of DDH or known congenital abnormalities of the fetus.<sup>10</sup> All women underwent an ECV attempt, after which there were 321 (64%) women with a fetus in breech presentation and 177 (36%) women with a fetus in cephalic presentation. There were significantly more neonates after breech delivery that required treatment for DDH 30 from 321 (9.3%) compared to five from 177 (2.8%) of neonates born in cephalic presentation after successful ECV (OR 0.29 (95% CI 0.09-0.95)). This study did not incorporate a control group of spontaneously cephalic presenting fetuses. Therefore, we were not able to calculate a pooled OR from both studies to compare the incidence of DDH in neonates after successful ECV to neonates with a spontaneous cephalic presentation.

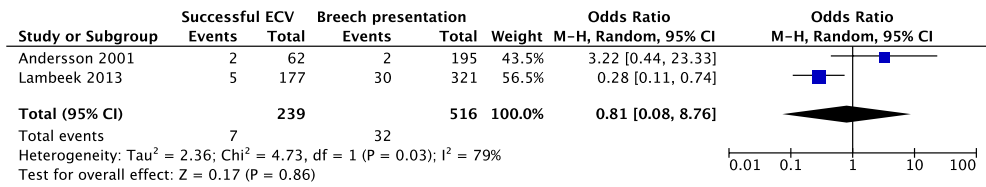


Figure 4 - Successful external cephalic version versus persistent breech presentation

## Discussion

This is, to our knowledge, the first review that explored the influence of different aspects of breech presentation on the risk for DDH. There seems to be little evidence about different aspect of breech presentation and the association with DDH. We found only one study that reported on the incidence of DDH in premature neonates, three studies that reported on the incidence of DDH in breech presentation in multiple gestation pregnancies and only two studies that examined the incidence of DDH in neonates born in cephalic presentation after successful ECV. Premature neonates born in breech presentation have a similar risk of DDH compared to neonates born in breech presentation at term. Neonates born in breech presentation from multiple gestation pregnancies have a decreased risk of DDH compared to neonates born in breech presentation from singleton pregnancies and a similar risk of DDH compared to neonates born in cephalic presentation in singleton pregnancies. Neonates born after successful ECV have a similar risk of DDH compared to neonates in born in breech presentation.

We conducted this review with a comprehensive search strategy and made a concerted effort to find all the evidence, nevertheless there are some limitations. First, a limitation of this study, and all other studies on DDH, is the fact that a clear definition of the condition is not well established in the international literature. In this review the definition of DDH ranged from an ultrasonographic diagnosis, Graf type  $\geq$ IIa, to neonates treated for the condition. Absence of a clear definition implies difficulty in comparing studies concerning DDH, therefore we reported for all the included studies which definition was utilized. Second, we were only able to perform univariable analyses, since we were dependent on the data provided in the articles and did not have individual patient data from the original studies. Not all factors could be analysed for interaction, for instance a positive family history of DDH or gender. Third, there is substantial statistical uncertainty due to the small number of neonates included in the studies, for instance in the study by Ruhmann et al. there were only eight neonates from multiple gestation pregnancies in breech presentation.

It is thought that the mechanical strengths that are placed on the neonatal hip by its position

in utero causes the increased risk of DDH in breech presentation, especially during the last weeks of gestation when the breech will be engaged in the maternal pelvis. In case of preterm delivery the fetus has a shorter period of exposure to these mechanical influences and it can be expected that premature neonates in breech presentation have a lower incidence of DDH. A lower incidence of DDH is also found in premature neonates independent of their position in the womb.<sup>11-13</sup> We found only one small retrospective cohort that examined the relation between preterm neonates born in breech position and DDH.<sup>5</sup> This study found that neonates born preterm in breech position have a similar incidence of DDH compared to term neonates born in breech position. In the group of neonates born preterm there were two neonates born at a gestational age of 36 weeks and one at a gestational age of 29 weeks. Breech delivery in borderline prematurity might well be exposing neonates to a similar risk for DDH, but this study does not provide the evidence for an increased risk in extreme premature neonates, <32 weeks of gestation. It would be interesting to further examine the risk of DDH in premature and extreme premature neonates born in breech presentation, especially since the incidence of breech presentation increases with a decrease in gestational age.<sup>11</sup> Longitudinal studies that examine whether the duration of breech presentation or the moment in gestation that a fetus is in breech presentation is of influence on the incidence of DDH are necessary.

Neonates from multiple gestation pregnancies born in breech presentation do not seem to have an increased risk of DDH, in contrary to breech presentation in singleton pregnancies. With multiple gestation pregnancies there is a higher probability of breech presentation, limitation of fetal mobility and increased intrauterine pressure. This would suggest that neonates born from multiple gestation pregnancies are at increased risk of DDH. In three studies on multiple gestation pregnancies, this increased risk of DDH in neonates born in breech presentation was not found. Although, there were some limitations to the design of these studies, none of the studies reported at what gestational age the neonates were born or whether they were the first or second twin. Since it can be expected that more neonates from multiple gestations were born premature and prematurity might decrease the chance of DDH this could have influenced the results. Nevertheless, in none of the studies DDH was diagnosed in neonates born in breech presentation from multiple gestation pregnancies.<sup>6-8</sup> An explanation for this finding might be the type of breech presentation in multiple gestation pregnancies, with more often complete breech presentation and the leg position more resembling that of a cephalic presenting fetus.

There are two cohort studies, with conflicting results concerning the risk of DDH after successful ECV.<sup>9,10</sup> The first study found an increased risk for DDH in neonates after successful ECV compared to persistent breech presentation, although not significant. And an increased risk for DDH after successful ECV, compared to primary cephalic presenting fetuses. While the second study found a lower incidence of DDH in neonates after successful ECV compared

to persistent breech presentation. Without a control group of primary cephalic presenting fetuses, it is not clear whether the risk of DDH after successful ECV is still increased compared to fetuses with a spontaneous cephalic presentation. Pooled data from both studies showed that there was no significant difference in the incidence of DDH between neonates in persistent breech presentation and neonates born in cephalic presentation after successful ECV. Additional studies, with a primary cephalic control group, seem necessary to clarify the exact relation between successful ECV and the risk of DDH. One theory, for a persistent increased risk after successful ECV, could be that DDH might be one of the reasons for breech presentation, in which malfunction of the hip and as a consequence different leg function causes these fetuses to present in breech position. Which was also seen in an observational study, where differences in fetal leg position between cephalic and breech presenting fetuses could already be observed early in the third trimester.<sup>15</sup>

The precise aetiology of DDH is unknown, but in case of breech presentation it is thought that mechanical influences are responsible for this increased risk. With this review we found evidence to support this mechanical theory, with less DDH in breech presenting neonates from multiple gestation pregnancies and a decrease in the incidence of DDH after successful ECV. Nevertheless, it might well be possible that breech presentation is a reflection of an already existing DDH, especially in singleton term pregnancies, which in turn decreases the chance of successful ECV.

## Conclusion

The evidence from this review can be helpful in optimizing existing screening programmes for DDH in which breech presentation is considered a risk factor for selective ultrasound screening. We showed that there is no evidence to screen neonates born in breech presentation from multiple gestation pregnancies. Premature neonates born in breech presentation are entitled to selective screening, but further research is necessary to determine if screening is also justified in extreme premature neonates born in breech presentation. Children after successful ECV born in cephalic presentation are still considered to have an increased risk for DDH, and therefore should receive ultrasound screening.

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