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REVIEW ARTICLE

The association between mode of delivery and developmental dysplasia of the hip in breech infants : a systematic review of 9 cohort studies

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Although developmental dysplasia of the hip (DDH) is a relatively common disorder, its aetiology remains elusive. The authors undertook a systematic review to determine whether there is an association between DDH and vaginal or caesarean delivery for singleton breech infants. The review focussed on cohort studies which provided risk estimates for DDH in breech-presenting infants, as a function of mode of delivery. Nine cohort studies with 35,139 infants were found. In the short-term, breech infants delivered through caesarean section had a significantly lower risk (13.5% less) for DDH : 5.95%, versus 6.88% (weighted values) in the vaginal delivery group (p = 0.008) {RR = 0.87 (95% CI 0.78-0.97)}. This might be mediated by the reduced stretch of the hip capsule, due to the absence of increased uterine pressure, which normally occurs in the active phase of labour. This pleads for the hypothesis that the mode of delivery is the critical factor promoting dislocation, not the breech presentation itself. Long-term data were not available, so that the overall effectiveness of caesarean section compared to vaginal delivery could not be established.

Keywords : developmental dysplasia of the hip ; DDH ; breech presentation ; caesarean section ; vaginal breech delivery.

INTRODUCTION

Developmental dysplasia of the hip (DDH) is a complex disorder which embraces a wide clinical spectrum of severity, from dislocated, dislocatable, or subluxable hips to stable or clicky hips with radiological or ultrasonographic evidence of acetabular dysplasia (*9,12,20*).

Prevalence of the clinical condition varies significantly among different racial groups, from 0.1 per 1000 among the Chinese in Hong Kong, to 1-2 per 1000 in England and Sweden, and 75 per 1000 in Greece and Italy (*24*). Variations may be due to differences in genetic or environmental factors or differences in clinical skills used in detection as well as definition of the condition.

The aetiology of DDH remains elusive with several theories being proposed, including inheritance, mechanical or environmental factors, hormone-induced joint laxity, and primary acetabular

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dysplasia (*5*). As to environmental factors, breech presentation, with a prevalence of 11% to 50% in DDH (*4,23,26,31*), has been considered the most important since the 19th century.

The obstetric management of breech deliveries has been greatly influenced by a number of trials which studied the perinatal mortality and morbidity of breech presentation infants based on the mode of delivery. A recent Cochrane systematic review indicated that planned caesarean section for singleton term breech presentation was associated with a decrease in perinatal or neonatal death and/ or neonatal morbidity. This was at the expense of increased maternal morbidity (*18*), but in many countries in Northern Europe and North America caesarean section has become the normal mode of breech delivery. However, no difference was observed in the composite outcome of death or neurodevelopmental delay at two years of age. Moreover, the effect of mode of delivery of breech infants on the incidence of DDH remains unclear. Some studies support the idea that the mode of delivery is the critical factor promoting dislocation (*6,22*), other studies suggest it is the breech presentation itself (*7,19*). So we need a more accurate estimate of the link between mode of delivery for breech infants and DDH, in view of the uncertainty of the long-term advantages of caesarean section for breech presentation for both mothers and infants. It was decided to perform a meta-analysis, in order to better estimate the effect of mode of delivery on DDH in singleton breech infants.

MATERIALS AND METHODS

Search strategy and selection criteria

The MEDLINE and EMBASE electronic databases were searched until March 2012, for studies reporting prevalence of DDH in breech-presenting neonates on the basis of clinical or ultrasonographic examination. Combinations of text words and thesaurus terms that included developmental dysplasia of the hip, developmental hip dysplasia, congenital dislocation of the hip, congenital hip dislocation, neonatal hip instability, developmental dislocation, developmental subluxation, and breech (MeSH : Medical Subject Headings) were used. The search was supplemented with the references of the selected studies and via correspondence with other researchers.

Two investigators (KB and NP) independently assessed the identified studies for eligibility. The inclusion criteria were : (1) randomized controlled trials or cohort, case-control, or cross sectional studies, (2) trials or studies carried out on singleton, breech-presenting infants, (3) stipulating the mode of delivery, (4) mentioning the effects of the mode of delivery, (5) relating the outcomes of interest to DDH, and (6) written in an international language (when necessary, colleagues fluent in the original language helped with translation). Exclusion criteria were : (1) letters, abstracts, systematic reviews, conference proceedings, (2) multiple births, (3) births with congenital malformations, (4) oligohydramnios, as well as (5) stillbirths. For the purposes of this study, DDH-affected infants were infants with at least one affected hip as diagnosed by clinical examination, ultrasonographic examination or a combination of both.

Quality assessment

Two investigators (KB and NP) independently assessed the quality of the included studies using a 6-point scoring system which was designed with reference to Moose (*28*), Qatso (*32*), and Strobe (*30*). One point was allocated for each of the following characteristics : (1) justifications for cohort, (2) use of appropriate inclusion and exclusion criteria, (3) systematic approach in DDH diagnosis, (4) adjustments for gender, family history, firstborn and miscarriages in earlier pregnancies, (5) assessment of outcome blinded for mode of delivery, and (6) long-term follow-up.

Statistical analysis

The authors concentrated on prevalence and total number of events for the association between mode of delivery and DDH in breech-presenting infants. A meta-analysis was performed on the studies which yielded sufficient information to allow the calculation of relative risk and 95% confidence interval (CI). RevMan 5.1.4 software was used for the analyses, and the Peto-modified Mantel-Haenszel method to combine the (log-transformed) risk ratios across studies. Weighted percentages of DDH risk for both modes of delivery derived from this meta-analysis for the risk ratio. The percentage of total variation in study estimates which is due to heterogeneity rather than sampling error was quantified with the I^2 statistic. The heterogeneity was tested with the χ^2 test.

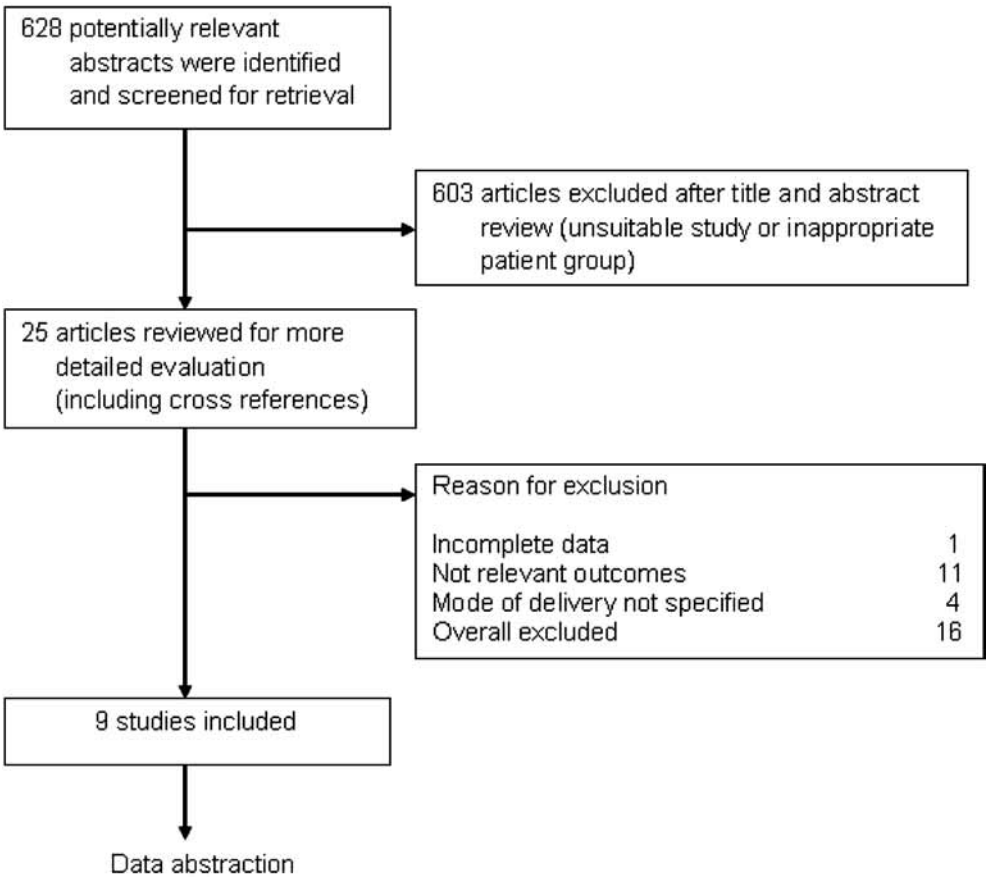

Fig. 1. — Flow of citations through the cross-sectional identification and retrieval of relevant papers indexed until March 2012

RESULTS

The search identified 628 papers (Fig. 1). Initial screening excluded 603 records. The remaining 25 full-text articles were re-checked, which further eliminated 16 studies, so that 9 studies remained. Agreement among the two reviewers (NP and KB) was excellent across all the screening phases. Discrepancies were resolved by discussion and the involvement of the third reviewer (WJH). The majority of articles were excluded because they did not report relevant outcomes or did not specify the mode of delivery for breech presenting infants. The 9 eligible reports were all cohort studies (*1,6,7,11,13, 17,22,29,33*). They included 35,139 infants in breech presentation, who were either delivered through the vaginal route or with a caesarean section. Most data were collected during the 1990s. A funnel plot of all included studies revealed no significant indication of publication bias (Fig. 2).

Although no study scored the highest level of quality (maximum 6 points), the overall level was adequate, with all studies scoring 3-4 points. Agreement between both reviewers (NP and KB) for the evaluation of the quality of the 9 included studies was high : 89%. None of the studies provided long-term follow-up or ensured blinding of the clinicians assessing DDH in breech-presenting infants. Of the 9 included studies, 8 reported an inverse association between caesarean section and DDH (Fig. 3). Only one retrospective cohort study by Clausen and Nielsen (*7*) revealed a higher incidence of DDH in the caesarean section group, but the difference did not reach statistical significance. Pooling the retrieved measures of association showed that caesarean section in singleton

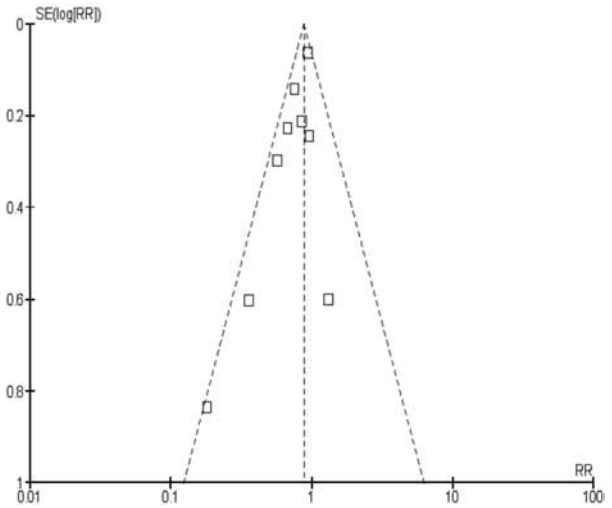

Fig. 2. — Funnel plot comparing the risk of DDH after Lower Segment Caesarean Section (LSCS) and vaginal delivery, in breech-presenting infants. The vertical-axis represents the standard error (SE) of the log-transformed risk ratio (RR) reflecting the study size and hence the precision of the estimate ; larger studies are at the top of the figure while smaller at the bottom. The horizontal axis represents the RR in a log scale measuring the treatment effect. The vertical dashed line represents the meta-analysis' combined RR (0.87) while the dashed lines – forming the cone – are the 95% prediction limits around the combined RR. If there is no indication of publication bias, estimates will cluster around the combined RR and the funnel plot will be roughly symmetrical as is the plot outlined here. If important heterogeneity is present, some study estimates would lie outside the cone formed by the 95% prediction limits.

breech-presenting infants led to a 13% decrease in the risk for DDH : 6.88% in the vaginal delivery group versus 5.95% in the caesarean section group (weighted percentages from meta-analysis). The rough, non-weighted, incidences were 4.50% and 4.98%. The Risk Ratio (RR) for the weighted risks was 0.87 (95% CI 0.78 to 0.97, p = 0.008). Note that similar evidence for a decreased risk after caesarean section was found when performing a meta-analysis on the odds ratios (results not shown). There was no evidence of heterogeneity in the studies included (I^2 = 33%).

DISCUSSION

Less DDH after caesarean section

Eight of the 9 studies included in this meta-analysis reported reduction in the risk of DDH associated with caesarean section, and this reduction reached statistically significant levels in two of them : Chan *et al* (*6*) and Demirci *et al* (*11*). Only Clausen and Nielsen (*7*) found a higher incidence of DDH among breech presenting infants delivered by caesarean section. The authors' meta-analysis of these 9 studies showed that Caesarean section for

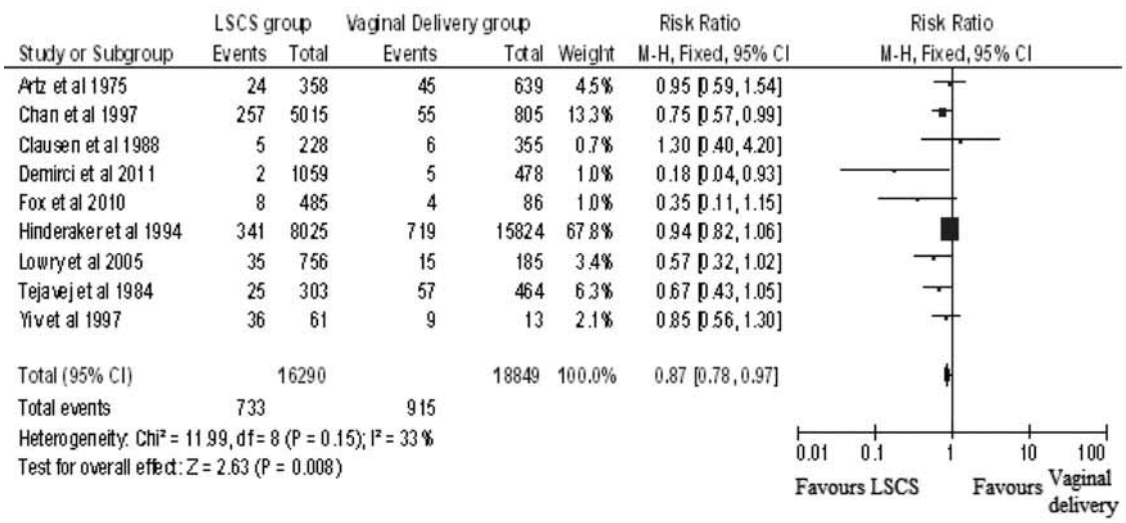

Fig. 3. — Risk Ratio (RR) for DDH in breech-presenting infants who were delivered with a Lower (uterine) Segment Caesarean Section (LSCS) compared with vaginal delivery. The combined RR 0.87 was calculated using the Peto-modified Mantel-Haenszel method : each included study was given a weight (Peto-modified Mantel-Haenszel weight) which reflected its size. The rough, non-weighted, incidences were 4.50% (733/16290) after caesarean section and 4.98% (915/18349) after vaginal delivery.

the delivery of breech-presenting infants was associated with a 13% reduction in the risk of hip dysplasia and instability, as compared with vaginal delivery. The difference (6.88% DDH in the vaginal delivery group versus 5.95% in the caesarean section group) was statistically significant ($p = 0.008$), as far as the short-term or immediate post-natal period was concerned. A long-term association could not be assessed, because of lack of data.

A possible explanation

The favourable effects of caesarean section on the incidence of DDH seem mainly mediated by the decreased stretch of the hip capsule due to the absence of increased uterine pressure which normally occurs in the active phase of labour (*31*). Further support of the above theory was offered by Moore's observation (*25*) that repeated and forceful neonatal hip examination may lead to hip dislocation by stretching the joint capsule.

Weaknesses

Controlled studies were not found. Beyond the caution needed in interpretation of data from observational studies, one must also consider other aspects associated with DDH. For instance, the high rate of spontaneous resolution of neonatal hip instability and dysplasia means that most children will not suffer impaired functional outcomes. Self-limited hip instability is a common finding in newborns (*14*). More than 80% of clinically unstable hips noted at birth have been shown to resolve spontaneously (*2*). However, the articles included in our analysis did not provide the information needed to evaluate the rate of spontaneous resolution and any long-term problems among children with newly diagnosed DDH. Eight of the 9 studies provided no follow-up, and only one study provided a 6 months follow-up. Moreover, the multiple screening and diagnostic modalities which have been used over the last decades and the variable anatomic abnormalities which come under the term of DDH make a precise definition of DDH controversial (*3*). Historically, DDH has been identified by clinical examination. Universal clinical screening was introduced in most European countries during the late 1950's and early 1960's, with radiographic follow-up in children with known risk factors (*8*). During the early 1990's ultrasound scan screening was introduced in order to reduce surgical treatment through early diagnosis and non-surgical treatment. Articles included in the current meta-analysis used different clinical examination procedures and criteria for the diagnosis of DDH. Also, they did not always provide a clear definition of DDH.

Strengths

First of all this study had a novel character : previous studies have also evaluated the effect of caesarean section on DDH, but this review was the first attempt to amalgamate existing studies. Secondly, publication and selection biases were minimized by including studies regardless of the language of publication. Thirdly, the large sample size (a total of 35,139 infants were studied) ensures increased precision in estimations and detection of even small differences.

Other advantages and disadvantages of caesarean section

The Term Breech Trial (*16*) and 4 large European population studies (*10,15,21,27*) all showed improved *neonatal* outcomes, other than DDH, after elective caesarean section. However, the absence of *long-term* follow-up in the current study, the known high rate of spontaneous resolution of neonatal hip instability, and the proven increased maternal morbidity associated with a policy of planned caesarean section make the net benefit of an elective caesarean section for all breech infants unclear. Randomized controlled trials, if ethically feasible, with long-term follow-up, would provide higher levels of evidence ; they might also test causality rather than just association.

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