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Vaginal breech delivery and the risk for perinatal death and cerebral palsy.

Registry-based cohort study in Norway.

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ABSTRACT:

Objective: To study if vaginal breech delivery is associated with increased risk for stillbirth, neonatal mortality (NNM) or cerebral palsy (CP) in Norway where vaginal delivery accounts for 1/3 of all breech deliveries.

Design: Cohort study using information from the national Medical Birth- and Cerebral Palsy Registers.

Setting: Births in Norway 1999-2009.

Participants: 520 047 term born singletons without congenital malformations.

Main outcome measures: Stillbirth, NNM and CP. Outcomes of breech birth were compared with cephalic birth, and related to actual and planned mode of delivery.

Results: Fetuses in breech (N=16 700) had increased risk for stillbirth (OR:1.8; 95%CI:3.7 to 6.9), NNM (OR:1.9; 95%CI:1.1 to 3.2) and CP (OR:1.3; 95%CI:0.9 to 2.1).Vaginal delivery was planned for 7917 of the fetuses, while 5561 actually delivered vaginally. The latter group had a threefold increased risk for NNM compared to vaginal cephalic delivery, but not different from those born by caesarean delivery in breech. Regarding planned mode, the excess risk for NNM was higher for vaginal than for caesarean delivery. Vaginal breech delivery was associated with a 30% increased risk for CP while there was a 70% excess risk for CP among children delivered by caesarean.

Conclusion: Breech delivery was associated with a substantial excess relative risk for stillbirth and NNM, and a 30% excess risk for CP. However, the absolute risks for these outcomes are low. The overall increased risk for stillbirth and the finding that the relative risk for NNM and CP was largely independent of mode of delivery suggest that some fetuses in breech have an antenatal acquired inherent risk for NNM per se, and/or for severe perinatal

complications. Thus, we conclude that vaginal delivery may be offered to women with a fetus in breech, provided competent obstetric care and strict criteria for selection to vaginal delivery.

ARTICLE SUMMARY:

Strengths and limitations of this study:

- More then 500 000 births included in the study.
- Prospectively recording of the data in the two registers.
- Restriction of the analyses to singletons at term without congenital malformation.
- The number of infants with adverse outcomes in breech were low.
- Register based data has limited ability to address explanatory factors.

INTRODUCTION:

Mode of delivery of a fetus in breech position is a controversial issue.[1] The Term Breech Trial (TBT)[2] reported lower perinatal mortality and morbidity of fetuses in breech position following planned caesarean delivery compared with planned vaginal delivery. The study had great impact, changing clinical practice in a number of countries.[3-6] However, the conclusion of the TBT were criticized by several experts.[7-9] In Norway, the Norwegian Board of Health invited a group of national experts to review the evidence underlying these recommendations. The expert group reviewed the literature published between 1980 – 2001. Taking into account the much lower perinatal mortality in Norway than that reported in the TBT, they concluded that vaginally breech delivery would still be safe, provided careful selection of mothers, qualified clinicians, and adequate fetal assessment.[10] Therefore, approximately 1/3 of fetuses in breech position in Norway are still delivered vaginally.[6] In a prospective study in France and Belgium, Goffinet et al compared vaginal delivery with planned caesarean delivery in breech. They concluded, in line with the Norwegian recommendations, that vaginal delivery is a safe option when strict selection criteria are followed.[11] The controversies of mode of delivery have also been reflected in studies of the long term outcome of infants born in breech position. Several studies reported that infants born in breech had increased risk for cerebral palsy (CP).[12-15]Although it was unclear whether mode of delivery affected this increased risk,[16-18] it has been suggested that planned caesarean delivery may prevent some cases of CP.[12 13]

The aim of this study was therefore to explore if singletons without congenital malformations born vaginally at term have higher risk for stillbirth, neonatal mortality (NNM) and CP if they present in breech than in cephalic position.

METHODS:

In this population based study, perinatal data from all children born in Norway during 1999-2009 were retrieved from the Medical Birth Registry of Norway (MBRN), and combined with information recorded in the Cerebral Palsy Register of Norway (CPRN). The 11-digit personal identification number unique for every Norwegian citizen was used to link information from the two registers. The MBRN records demographic variables, as well as information on maternal health before and during pregnancy, interventions and complications during delivery and neonatal outomes. Registration in this register has been compulsory since 1967 ensuring prospective recording of this information at birth.[19]

The Cerebral Palsy Register of Norway (CPRN) is an informed consent based national quality register established in 2006, and aims to record detailed information on all children with CP born in Norway since 1996.[20] Information is reported at diagnosis, at 5 years and at 15-17 years of age. Neuropediatric habilitation centres in Norway provide summary and detailed data about the children. A validation study indicated that 80% of children with CP in Norway born 1999-2009 have detailed information in the CPRN.[21]

We excluded children born preterm (before week 37), multiple births, children with congenital malformations, children in transverse lie and those with lacking information on mode of delivery (Figure 1).

Study variables:

The predefined main outcome measures were stillbirth, NNM and CP. Stillbirth and NNM were defined according to the WHO.[22] Cerebral palsy was diagnosed and confirmed at five years of age according to the definition and classification proposed by the Surveillance of

Cerebral Palsy in Europe.[23] Paediatricians at the Neurohabilitation centers in Norway completed the information of each child on a standardized form.

Information on maternal age, parity, gestational age, mode of delivery, the child's sex, birth weight and Apgar scores was collected from the MBRN. Newborns with a birth weight below -2 standard deviations of the population mean weight[24] for gestational age, adjusted for sex were defined as small-for-gestational age (SGA).

Analytic approach:

First, we assessed the risks for stillbirth, NNM and CP for children born in breech compared to cephalic position, independent of mode of delivery.

Second, we explored whether actual mode of delivery (vaginal or caesarean) influenced the risks of these outcomes.

The Norwegian Society for Gynecology and Obstetrics suggests that if the gestational age is at least 34 weeks, estimated birth weight is between 2000 and 4000 grams and no maternal and fetal contraindications for vaginal delivery exists, a vaginal delivery can be recommended. An essential premise of this recommendation is that the obstetric department is capable to perform immediate caesarean delivery and that trained paediatric personnel are available. Thus, some of the planned vaginal breech deliveries will be converted to a caesarean delivery during the birth process. The analysis of actual mode of delivery will not evaluate these recommendation of vaginal births correctly, since the caesarean group will be a

mixture of both planned and emergency caesarean delivery, and the vaginal group will comprise only those not changed to a caesarean delivery during birth.

Third, we therefore repeated the analyses, but now explored the association between planned mode of delivery at admission to the obstetric department, and outcome. We divided cephalic and breech births into the two categories originally planned vaginal and caesarean deliveries.

However, the MBRN does not record specifically planned vaginal births, but do record the variable "planned caesarean delivery". Unfortunately, the latter variable has a large proportion of missing values (N= 349 881), and we therefore utilized information from several variables in the MBRN to complete the dichotomization. Births were categorized as planned caesarean delivery, if this was indicated in the similarly named variable, or if "initiation of delivery" was recorded as elective. Births that did not satisfy these criteria were categorized as planned vaginal delivery. Stillbirth, NNM and CP were then assessed related to the four exposure groups: cephalic position and planned vaginal births (reference group), cephalic position and planned caesarean delivery, breech position and planned vaginal births, and breech position and planned caesarean delivery.

Finally, we explored if the risk for stillbirth, NNM or CP differed between children born by vaginal delivery and caesarean delivery or between planned vaginal delivery and planned caesarean delivery within the group of children who were born in breech.

Statistical analyses:

IBM SPSS software for Windows version 22 was used for data analyses. Differences in proportions between groups were analyzed using the chi-square test and prevalence rates with 95% confidence intervals (CI) were calculated according to Newcombe and Altman.[25] In the estimates of the prevalence of NNM, stillbirths were excluded and in the estimates of the prevalence of CP, stillbirths and children with post neonatal CP were excluded. We used logistic regression to estimate odd ratios (OR) with 95% confidence intervals (CI) for adverse outcome of children in breech position at birth, using cephalic presentation as the reference. Moreover we adjusted for potential confounders including maternal age, parity, gestational age, child sex and SGA status based on *a priori* knowledge and directed acyclic graphs methodology.[26]

Patient involvement:

No patients were involved in setting the research question or the outcome measures, nor were they involved in the design and implementation of the study. There are no plans to involve patients in dissemination.

RESULTS:

A total of 650 968 children were born in Norway during the study period. The study population of singleton children born at a gestational age of at least 37 weeks in either cephalic or breech position, and with no congenital anomalies comprised 520 047 children (Figure 1). A total of 841 (2 per 1000) of these were stillborn. Of the liveborn, 239 (0.5 per 1000) died in the neonatal period, and 552 children were diagnosed with CP. Of the latter, 32

had a post-neonatal cause of their CP, resulting in 520 with congenital CP (1 per 1000 liveborn).

Among the 520 047 included children, 16 700 (3%) were in breech and 503 347 (97%) in cephalic position (Table 1). More mothers in the breech group were nullipara, and a higher proportion of their infants were females, were born SGA and had low Apgar scores (Table 1). The mean gestational age of children born in breech was 39.1 weeks compared with 39.7 weeks for children born in cephalic. Of the 16 700 women with a fetus in breech 7917 (47%) were planned for vaginal delivery while 5561 (33%) actually delivered vaginally. The corresponding figures for planned caesarean delivery was 8783 (53%) while 11 139 (67%) actually delivered by caesarean. For women with fetuses in cephalic position, 94 % were planned to vaginal delivery while 90% delivered vaginally; 6% were planned to caesarean delivery and 10% delivered by caesarean.

Children born in breech had increased risk for stillbirth, NNM and CP compared with children born in cephalic position (Table 2). Sixty-eight of the stillborn children (seven in breech and 61 in cephalic position) died during delivery. When these children were excluded from the analyses, the risk for stillbirth was essentially unchanged (OR: 1.6; CI: 1.2 to 2.3).

According to actual mode of delivery, children born vaginally in breech had higher OR for stillbirth compared with children born vaginally in cephalic position (Table 3). Regardless of mode of delivery, children in breech had a nearly three-fold increased OR for NNM, while the OR for CP was 1.7 (CI: 1.0 to 2.3) if the child was delivered by caesarean delivery (Table 3).

As expected, children in cephalic position had higher prevalence of stillbirth, NNM and CP if they were delivered by caesarean delivery compared with vaginal delivery, reflecting that caesarean delivery in this group is mainly done in high risk births (Table 3).

According to planned mode of delivery, the prevalence of stillbirth and NNM was increased in breech vaginal delivery compared with cephalic vaginal delivery (Table 4). Whereas the OR for NNM was 2.4 (CI: 1.5 to 3.4) for planned vaginal delivery, the OR for NNM among those planned to be delivered by caesarean was 1.6 (CI: 0.7 to 3.7). The higher prevalence of CP, regardless of mode of delivery, did not differ significantly from cephalic vaginal delivery (Table 4). Among children born in the cephalic position, the prevalence of NNM was higher among those born by caesarean, than among children born by vaginal delivery.

Analyses restricted to the 16 700 children in breech position, showed that the prevalence of stillbirth was significantly higher in vaginal delivery compared with caesarean both for planned as well as for actual mode of delivery (Table 5). The risk for NNM was not increased among infants actually born by vaginal delivery compared with caesarean delivery, while there was a 50% increased risk (OR: 1.5; CI: 0.5 to 4.3) for NNM in the group where vaginal delivery was planned (Table 5). The risk for CP was not increased for children born by vaginal delivery compared with caesarean delivery regardless of actual or planned mode of delivery (Table 5).

Multivariable analyses adjusting for gestational age, parity, maternal age, sex and SGA did not substantially affect any of the associations described above (data not shown).

DISCUSSION:

In this national cohort study we found an excess risk for stillbirth and NNM for term singletons without congenital malformations born in breech. We also found a higher prevalence of CP among children in breech, although not statistically significantly different from children in cephalic position. However, the absolute risks for NNM and CP were low, ranging between 0.5 and 1.7 per 1000 liveborn.

The overall high proportion of stillbirths in the breech group, and the fact that the risks for NNM and CP were largely independent of mode of delivery may suggest that some fetuses in breech have an antenatal acquried vulnerablity for NNM and CP.

Strengths and limitations:

Strengths of the present study are the large number of births and the prospectively recording of the data in the two registers. Nonetheless, among children in breech position the number of children with the adverse outcomes NNM and CP were low, limiting the statistical power of the study.

We restricted the analyses to singletons born at term and without congenital malformations, limiting the possibility of confounding by these factors. Multivariable analyses suggested that maternal age, parity, the child`s sex, gestational age and SGA did not confound the associations between breech position and adverse outcome.

Analysis of the association between mode of delivery and adverse outcome after breech delivery is challenging. Selection to vaginal delivery is recommended on strict criteria and is

therefore expected to identify pregnancies with low risk for adverse outcome compared to those selected for caesarean delivery. Furthermore, some of the planned vaginal deliveries will be converted to an emergency caesarean delivery intrapartum, increasing the risk for adverse outcome in the caesarean group. A comparison of adverse outcome between vaginal and caesarean deliveries would therefore be expected to favor the vaginal delivered group. While this was the case for children born in cephalic position (table 3 and table 4), the ORs for NNM were similar or even higher in the vaginal compared with the caesarean delivery group for children born in breech. Thus, caution is needed in the interpretation of the lack of difference between vaginal and caesarean delivery.

We categorized, not only according to actual mode of delivery, but also according to planned mode of delivery. Although we cannot fully rule out errors in this classification, the risk for NNM and CP was higher for actual than for planned caesarean delivery, as would be expected if the classification was correct. Thus, we consider it unlikely that misclassification explains the main results.

Vaginal delivery is the preferred mode of delivery of a dead fetus. Thus, reverse causality is the probable explanation of the finding of a high risk for stillbirth among children in breech position born by vaginal delivery.

Finally, the use of register based data has limited ability to address explanatory factors, as suggested by Goffinet et al.[11] In their prospective study of breech deliveries, they found that 33 (26%) of 129 cases with severe neonatal complications had nonlethal major or minor malformations that sometimes explained the neonatal complications.[11] We cannot rule out that

some undiagnosed or unrecorded malformations, may have contributed to the higher proportions of stillbirths and NNM among children born in breech in our study.

Comparison with other studies:

Our findings regarding excess risk for stillbirth[27] and NNM associated with breech presentation are consistent with earlier findings[28-30] and an excess risk for NNM was also reported in recent studies including children born after the Term breech trial (TBT)[2] in Denmark[4] and in Norway.[6]

We found a slightly higher risk for NNM in planned vaginal than in planned caesarean delivery and this could be considered to be consistent with the results of the TBT. On the other hand, the overall interpretation of our findings is that the risk for NNM was largely independent of mode of delivery, and this interpretation is not consistent with the results of the TBT. First, the different designs of the two studies may explain the different findings. The TBT was a randomized controlled trial considered to be the gold standard, while our study is an observational study. Nonetheless, the much lower perinatal mortality in Norway compared with the TBT may also explain some of the diverging results in the two studies. Moreover, to be eligible to participate in the TBT, women had to have a singleton live fetus at term (≥ 37 week`s gestation) in breech without any known lethal fetal congenital anomaly. Women were excluded if there was evidence of fetopelvic disproportion, or if the fetus was judged to be clinically large or to have an estimated fetal weight of 4000 g or more, hyperextension of the fetal head or other fetal anomaly or condition that might cause a mechanical problem at delivery. Women with contraindication for labor or vaginal delivery such as placenta praevia

were also excluded.[2] These criteria are similar to the criteria for vaginal breech delivery recommended by the Norwegian Society for Gynecology and Obstetrics. However, in the TBT a lower proportion of women (57%) selected for vaginal delivery actually delivered vaginally compared with our study population where 70% of those selected actually had a vaginal delivery. One may therefore speculate that the probability for adverse outcome in the planned vaginal group in the TBT was higher than in our study, since a larger proportion of mothers in the TBT needed acute caesarean delivery. Instead, antenatal acquired vulnerability may have played a larger role in our population.

We are not aware of studies addressing the association between breech presentation at birth and CP in populations born after the TBT.[2] A follow-up study of 923 children included in that trial did not have the statistical power to address this severe, neurodevelopmental outcome.[31] In studies done before the TBT some authors found that fetuses in breech had increased risk for CP,[12-15] whereas others did not find an increased risk.[16 32-34] Two studies, including one from our own group, also found some evidence that the risk was associated with vaginal delivery.[12 15] The lower risk for CP in this, compared with our previous Norwegian study [12] could be explained by the larger sample size, better quality of the data in the MBRN [19] and better ascertainment of cases in the CPRN in the present study.[21] Nonetheless, it is also possible that changes in the delivery of breech births in Norway including an increasing proportion of fetuses born by planned caesarean delivery,[6] may have improved outcome, and may reflect better selection of mothers for vaginal delivery.

Interpretation:

The overall higher risk for stillbirth and the excess risk for NNM and - to a lesser extent for CP – independent of mode of delivery among children born in breech suggest that fetuses with antenatal acquired risk factors for these outcomes, are more likely to present in breech than in cephalic position at birth. This explanation may be consistent with the nearly identical OR for NNM among children born by vaginal and by caesarean delivery, and with the higher proportion of infants born SGA in breech than in cephalic position. However, the risk for NNM was slightly higher for vaginal than for caesarean delivery of fetuses in breech. This could suggest that fetuses in breech with or without antenatal acquired risk factors are more likely to experience complications during birth if they are born by vaginally, as one might have expected a particular low risk for adverse outcome in the group of women selected for vaginal delivery. Such complications may lead to acute intrapartum hypoxemia in the most severe cases leading to death in the neonatal period or to CP. Thus, a combination of antenatal acquired risk factors for neonatal death and CP with increased vulnerability to the birth process is probably the most likely explanation of our findings.

Regarding CP, antenatal factors are considered to be involved in 90% of the cases with CP,[35] and one might have expected a higher than the 30% increased risk for CP in breech births. This finding could however, suggest that antenatal factors increasing the risk for stillbirth and NNM are different or at least not completely overlapping with antenatal risk factors involved in the causal pathway leading to CP. This interpretation may explain that in contrast to the risk for NNM, the risk for CP was slightly higher for planned caesarean than for planned vaginal breech delivery.

Implications:

Taking into consideration the very low absolute risk for NNM and CP, the increasing evidence for acute and long term maternal complications[36] and for later health problems among children following caesarean delivery[37 38] our results suggest that vaginal delivery in selected cases may be an option for women with a fetus in breech position. This option requires that strict criteria are followed including access to competent obstetric care. In addition, a secondary advantage of having a certain volume of vaginal breech deliveries is that obstetricians retain their competence for unexpected vaginal breech deliveries. In the discussion with the pregnant mother and her partner regarding choice of delivery mode of a fetus in breech, the relative risk for NNM and CP should be explained and related to the very low absolute risk. Moreover, it may be appropriate to emphasize that adverse outcome probably to a large degree is caused by antenatal acquired insults and that there are potential advantages of vaginal birth over caesarean delivery for long term health of the child and the mother. Regarding obstetric care, awareness of the excess risk for fetal death should be emphasized, and studies are warranted to optimize antenatal follow up of mothers with a fetus in breech.

Caution is needed if results of observational studies are included in the development of clinical guidelines, and more studies are needed to support our results. On the other hand, a new RCT in our part of the world is unrealistic as it would require the participation of 20 000 women with a fetus in breech in order to document a difference in NNM between mothers selected for planned vaginal and planned caesarean delivery.[10]

Nonetheless, the high prevalence of neonatal mortality among planned vaginal deliveries compared with planned caesarean delivery is of concern and warrants further studies, including perinatal audits and prospective studies as suggested by Goffinet et al.[11]

CONCLUSION

Breech delivery was associated with a substantial excess relative risk for stillbirth and NNM, and a 30% excess risk for CP. The excess risk of stillbirth associated with vaginal breech delivery is most likely explained by reverse causality. The overall increased risk for stillbirth and the finding that the relative risk for NNM and CP was largely independent of mode of delivery suggest that some fetuses in breech have an antenatal acquired inherent risk for these adverse outcomes. Nonetheless, our findings of a 50% increased risk for adverse outcome in planned vaginal compared with planned caesarean delivery suggest that intrapartum complications during vaginal breech delivery also may play a role in the excess risk for NNM. The potential risk related to vaginal birth should be weighted against potential unfavorable outcomes related to caesarean delivery. Taking the very low absolute risk for NNM and CP into account, and that the majority of adverse events may be related to antenatal acquired vulnerability, we conclude that vaginal delivery may be offered to women with a fetus in breech, provided competent obstetric care and strict criteria for selection to vaginal delivery.

What is already known about this topic?

- Breech delivery is associated with excess risk for stillbirth, neonatal mortality and cerebral palsy.
- Planned caesarean delivery has been recommended for all breech deliveries in order to reduce the risk for neonatal mortality and morbidity.
- Whether planned caesarean delivery reduces the risk for cerebral palsy is not known.

What this study adds?

- The increased risk for adverse outcome of breech births in a population with low perinatal mortality was largely independent of mode of delivery.
- The risk for neonatal mortality was slightly higher in planned vaginal than in planned caesarean breech delivery.
- Fetuses in breech have an antenatal acquired vulnerability for neonatal mortality and cerebral palsy and probably also for perinatal complications during vaginal delivery.
- Taking into consideration the low absolute risk for these complications, vaginal breech delivery may be offered to women with a fetus in breech position, provided strict selection criteria.

Competing interest: All authors have completed the ICMJE uniform disclosure form at www.icmj.org/cio_disclosure.pdf (available on request from the corresponding author) and declare: no financial support from any organization for the submitted work; no financial relationship with any companies that might have an interest in the submitted work in the previous three years; and have no non-financial interests or relationships that may be relevant to the submitted work.

Contributors: SB analyzed the data, contributed to study design and data interpretation and wrote the first draft of the manuscript. GA was principally responsible for the data from the CPRN and revised the manuscript. MM contributed to the data interpretation and revised the manuscript. PR contributed to the research hypotheses, study design and revised the manuscript. SH contribute to the data interpretation and revised the manuscript. DM contributed to study design, the data analyses, data interpretation and revised the manuscript. TV proposed the research questions, and contributed in the interpretation of the data and the revision of the manuscript. He is the guarantor of the study and accepts full responsibility for the work and the conduct of the study. All authors approved the final version of the submitted manuscript**.**

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Ethical approval: The study was approved by the Regional Ethical Committee for Medical Research in Mid-Norway (ref 2011/754).

Data sharing: The protocol is available on request from the corresponding author at: sbjellmo@hotmail.com

Transparency declaration: The lead author affirms that this manuscript is an honest, accurate, and transparent account of the study being reported; that no important aspects of the study have been omitted; and that any discrepancies from the study as planned have been explained.

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Figure 1: Flow chart of the study population.

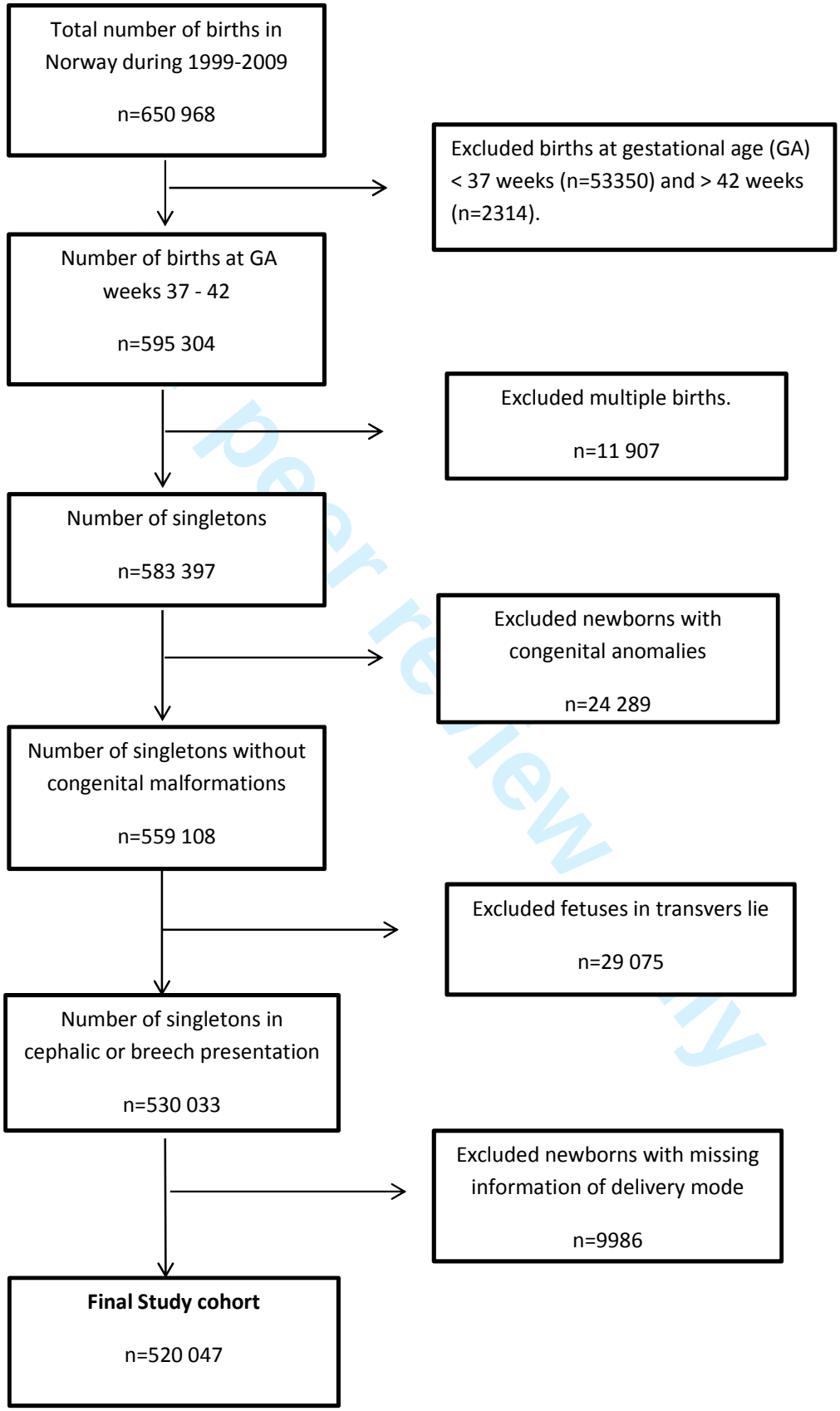

Table 1: Maternal and infants characteristics in pregnancies where the child was born in breech or in cephalic position.

	Breech position		Cephalic position	
	N	(%)	N	(%)
Number of births:	16 700	(100)	503 347	(100)
Maternal age[a]				
≤ 19 y	290	(2)	11 889	(2)
20-34 y	13 412	(80)	409 401	(81)
≥ 35 y	2998	(18)	82 031	(17)
Parity				
Nullipara	9280	(56)	199 822	(40)
Primipara	4599	(27)	184 068	(36)
>1 para	2822	(17)	119 457	(24)
Sex[b]				
Male	7540	(45)	257 128	(51)
Female	9160	(55)	246 216	(49)
Small for gestational age[c]	424	(2.5)	7130	(1.4)
Apgar score at 5 min[d]				
0-3	93	(0.6)	1477	(0.3)
4-6	427	(2.4)	7913	(1.7)
7-10	16 139	(97)	492 858	(98)

[a]Information on maternal age was missing in 26 children in cephalic.
[b]Information on Sex was missing in 1 child in cephalic.
[c]Information on Small for gestational age was missing in 12 children in breech and 361 in cephalic.
[d]Information on Apgar at 5 min was missing in 41 children in breech and 1099 in cephalic.

Table 2. ***All births:*** Prevalence and odds ratios (OR) with 95% confidence intervals (CI) for stillbirth, neonatal mortality and cerebral palsy among singletons born at term, without congenital anomalies in cephalic and breech positions

	Number of infants with adverse outcome	Total number infants*	Prevalence per 1000	OR (CI)
Stillbirths				
Cephalic	794	503 347	1.6 (1.5 to 1.7)	1.0 (Reference)
Breech	47	16 700	2.8 (2.1 to 3.7)	1.8 (1.3 to 2.4)
Neonatal mortality				
Cephalic	225	502 553	0.5 (0.4 to 0.5)	1.0 (Reference)
Breech	14	16 653	0.8 (0.5 to 1.4)	1.9 (1.1 to 3.2)
Cerebral palsy*				
Cephalic	498	502 524	1.0 (0.9 to 1.1)	1.0 (Reference)
Breech	22	16 650	1.3 (0.9 to 2.0)	1.3 (0.9 to 2.1)

*Removed stillbirths from the analyses of NNM and CP.
** Removed post neonatal CP in the analyses of CP as outcome.

Table 3. ***Actual mode of delivery:*** Prevalence and odds ratios (OR) with 95% confidence intervals (CI) for stillbirths, neonatal mortality and cerebral palsy among singletons born at term, without congenital anomalies according to actual mode of delivery.

	Number of infants with adverse outcome	Total number infants*	Prevalence per 1000	OR (CI)
Stillbirths				
Cephalic				
Vaginal delivery	697	451 761	1.5 (1.4 to 1.7)	1.0 (Reference)
Cesarean delivery	97	51 586	1.9 (1.5 to 2.3)	1.2 (1.0 to 1.5)
Breech:				
Vaginal delivery	43	5561	7.7 (5.8 to 10.4)	5.0 (3.7 to 6.9)
Cesarean delivery	4	11 139	0.4 (0.1 to 0.9)	0.2 (0.1 to 0.6)
Neonatal mortality (NNM)				
Cephalic				
Vaginal delivery	137	451 064	0.3 (0.4 to 2.1)	1.0 (Reference)
Cesarean delivery	88	51 489	1.7 (1.4 to 2.1)	5.6 (4.3 to 7.4)
Breech:				
Vaginal delivery	5	5518	0.9 (0.4 to 2.1)	3.0 (1.2 to 7.3)
Cesarean delivery	9	11 135	0.8 (0.4 to 1.5)	2.7 (1.4 to 5.2)
Cerebral palsy (CP)**				
Cephalic				
Vaginal delivery	388	451 042	0.9 (0.8 to 1.0)	1.0 (Reference)
Cesarean delivery	110	51 482	2.1 (1.8 to 2.6)	2.5 (2.0 to 3.1)
Breech:				
Vaginal delivery	6	5517	1.1 (0.5 to 2.4)	1.3 (0.6 to 2.8)
Cesarean delivery	16	11 133	1.4 (0.9 to 2.3)	1.7 (1.0 to 2.8)

*Removed stillbirths from the analyses of NNM and CP.
** Removed post neonatal CP in the analyses of CP as outcome.

Table 4. ***Planned mode of delivery:*** Prevalence and odds ratios (OR) with 95% confidence intervals (CI) for stillbirths, neonatal mortality and cerebral palsy among singletons born at term, without congenital anomalies according to planned mode of delivery.

	Number of infants with adverse outcome	Total number infants*	Prevalence per 1000	OR (CI)
Stillbirths				
Cephalic				
Planned vaginal delivery	753	474 976	1.6 (1.5 to 1.7)	1.0 (Reference)
Planned caesarean delivery	41	28 371	1.5 (1.1 to 2.0)	0.9 (0.7 to 1.2)
Breech:				
Planned vaginal delivery	44	7917	5.6 (4.1 to 7.5)	3.5 (2.6 to 4.8)
Planned caesarean delivery	3	8783	0.3 (0.1 to 1.0)	0.2 (0.1 to 0.7)
Neonatal mortality(NNM)				
Cephalic				
Planned vaginal delivery	198	474 223	0.4 (0.4 to 0.5)	1.0 (Reference)
Planned caesarean delivery	27	28 330	1.0 (0.7 to 1.4)	2.3 (1.5 to 3.4)
Breech:				
Planned vaginal delivery	8	7873	1.0 (0.5 to 2.0)	2.4 (1.2 to 4.9)
Planned caesarean delivery	6	8780	0.7 (0.3 to 1.5)	1.6 (0.7 to 3.7)
Cerebral palsy(CP)*				
Cephalic				
Planned vaginal delivery	453	474 198	1.0 (0.9 to 1.1)	1.0 (Reference)
Planned caesarean delivery	45	28 326	1.6 (1.2 to 2.1)	1.7 (1.2 to 2.3)
Breech:				
Planned vaginal delivery	10	7872	1.3 (0.7 to 2.3)	1.3 (0.7 to 2.5)
Planned caesarean delivery	12	8778	1.4 (0.8 to 2.4)	1.4 (0.8 to 2.5)

*Removed stillbirths from the analyses of NNM and CP.
** Removed post neonatal CP in the analyses of CP as outcome.

Table 5. ***Restricted to breech deliveries:*** Prevalence and odds ratios with 95% confidence intervals for stillbirths, neonatal mortality and cerebral palsy among singletons in breech position born at term, without congenital anomalies according to actual and planned mode of delivery.

	Number of infants with adverse outcome	Total number infants*	Prevalence per 1000	OR (CI)
Stillbirths				
Actual mode of delivery				
Caesarean delivery	4	11 139	0.4 (0.1 to 0.9)	1.0 (Reference)
Vaginal delivery	43	5561	7.7 (5.8 to 10.4)	21.7 (7.8 to 60.5)
Planned mode of delivery				
Caesarean delivery	3	8783	0.3 (0.1 to 1.0)	1.0 (Reference)
Vaginal delivery	44	7917	5.6 (4.1 to 7.5)	16.4 (5.1 to 52.7)
Neonatal mortality (NNM)*				
Actual mode of delivery				
Caesarean delivery	9	11 135	0.8 (0.4 to 1.5)	1.0 (Reference)
Vaginal delivery	5	5518	0.9 (0.4 to 2.1)	1.1 (0.4 to 3.3)
Planned mode of delivery				
Caesarean delivery	6	8780	0.7 (0.3 to 1.5)	1.0 (Reference)
Vaginal delivery	8	7873	1.0 (0.5 to 2.0)	1.5 (0.5 to 4.3)
Cerebral palsy(CP)*				
Actual mode of delivery				
Caesarean delivery	16	11 133	1.4 (0.9 to 2.3)	1.0 (Reference)
Vaginal delivery	6	5517	1.1 (0.5 to 2.4)	0.8 (0.3 to 1.9)
Planned mode of delivery				
Caesarean delivery	12	8778	1.4 (0.8 to 2.4)	1.0 (Reference)
Vaginal delivery	10	7872	1.3 (0.7 to 2.3)	0.9 (0.4 to 2.2)

*Removed stillbirths from the analyses of NNM and CP.
** Removed post neonatal CP in the analyses of CP as outcome.

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Is vaginal breech delivery associated with higher risk for perinatal death and cerebral palsy compared with vaginal cephalic birth? Registry-based cohort study in Norway.

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Is vaginal breech delivery associated with higher risk for perinatal death and cerebral palsy compared with vaginal cephalic birth?
Registry-based cohort study in Norway.

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ABSTRACT:

Objective: To study if vaginal breech delivery is associated with increased risk for neonatal mortality (NNM) or cerebral palsy (CP) in Norway where vaginal delivery accounts for 1/3 of all breech deliveries.

Design: Cohort study using information from the national Medical Birth- and Cerebral Palsy Registers.

Setting: Births in Norway 1999-2009.

Participants: 520 047 term born singletons without congenital malformations.

Main outcome measures: NNM, CP and a composite outcome of these and death during birth.

Results: Compared with cephalic births, breech births had substantially increased risk for NNM but not for CP. Vaginal delivery was planned for 7917 of 16700 fetuses in breech, while 5561 actually delivered vaginally. Among these, NNM was 0.9 per 1000 compared with 0.3 per 1000 in vaginal cephalic delivery, and 0.8 per 1000 in those actually born by caesarean delivery (CD) in breech. Compared with planned cephalic delivery, planned vaginal delivery was associated with excess risk for NNM (OR: 2.4; 95%CI:1.2 to 4.9), while the OR associated with planned breech CD was 1.6 (95%CI: 0.7 to 3.7). These risks were attenuated when NNM was substituted by the composite outcome. Vaginal breech delivery was not associated with excess risk for CP compared with vaginal cephalic delivery.

Conclusion: Vaginal breech delivery, regardless of whether planned or actual, and actual breech CD were associated with excess risk for NNM compared with vaginal cephalic delivery, but not with CP. The risk for NNM and CP in planned breech CD did not differ significantly from planned vaginal cephalic delivery. However, the absolute risk for these

outcomes was low and taking into consideration potential long-term adverse consequences of CD for the child and later deliveries we therefore conclude that vaginal breech delivery may be recommended, provided competent obstetric care and strict criteria for selection to vaginal delivery.

ARTICLE SUMMARY:

Strengths and limitations of this study:

- More than 500 000 births included in the study.
- Prospectively recording of the data in the two registers.
- Restriction of the analyses to singletons at term without congenital malformation.
- The number of infants with adverse outcomes in breech were low.
- Register based data has limited ability to address explanatory factors.

INTRODUCTION:

Mode of delivery of a fetus in breech position is a controversial issue.[1] The Term Breech Trial (TBT)[2] reported lower perinatal mortality and morbidity of fetuses in breech position following planned caesarean delivery compared with planned vaginal delivery. The study had great impact, changing clinical practice in a number of countries.[3-6] However, the conclusion of the TBT were criticized by several experts.[7-9] In Norway, the Norwegian Board of Health invited a group of national experts to review the evidence underlying these recommendations. The expert group reviewed the literature published between 1980 – 2001. Taking into account the much lower perinatal mortality in Norway than that reported in the TBT, they concluded that vaginally breech delivery would still be safe, provided careful selection of mothers, qualified clinicians, and adequate fetal assessment.[10] Therefore, approximately 1/3 of fetuses in breech position in Norway are still delivered vaginally.[6] In a prospective study in France and Belgium, Goffinet et al compared vaginal delivery with planned caesarean delivery in breech. They concluded, in line with the Norwegian recommendations, that vaginal delivery is a safe option when strict selection criteria are followed.[11] The controversies of mode of delivery have also been reflected in studies of the long-term outcome of infants born in breech position. Several studies reported that infants born in breech had increased risk for cerebral palsy (CP).[12-15]Although it was unclear whether mode of delivery affected this increased risk,[16-18] it has been suggested that planned caesarean delivery may prevent some cases of CP[12 13].

In the vast majority of previous studies on adverse outcome of vaginal breech delivery, the comparison group has been caesarean breech delivery. However, the main results of these studies do not take into account the risk for complications of caesarean delivery in later pregnancies both for the mother and for the child. There is also an increased awareness of later health problems in children born by caesarean suggested in recent reports[19]. Therefore, to

assess if vaginal breech delivery as currently practiced in Norway can be characterized as safe, the appropriate comparison group of breech deliveries should be vaginal cephalic birth; which is the natural way of giving birth. In line with this, a recent systematic review and meta-analysis of breech deliveries recommended that comparative studies of vaginal breech with vaginal cephalic deliveries should be undertaken[20].

The aim of this study was therefore to explore if singletons without congenital malformations born vaginally at term have higher risk for stillbirth, neonatal mortality (NNM) and CP if they are born in breech position compared to cephalic position.

METHODS:

In this population based study, perinatal data from all children born in Norway during 1999-2009 were retrieved from the Medical Birth Registry of Norway (MBRN), and combined with information recorded in the Cerebral Palsy Register of Norway (CPRN). The 11-digit personal identification number unique for every Norwegian citizen was used to link information from the two registers. The MBRN records demographic variables, as well as information on maternal health before and during pregnancy, interventions and complications during delivery and neonatal outomes. Registration in this register has been compulsory since 1967 ensuring prospective recording of this information at birth.[21]

The Cerebral Palsy Register of Norway (CPRN) is an informed consent based national quality register established in 2006, and aims to record detailed information on all children with CP born in Norway since 1996.[22] Information is reported at diagnosis, at 5 years and at 15-17 years of age. Neuropediatric habilitation centres in Norway provide summary and detailed

data about the children. A validation study indicated that 80% of children with CP in Norway born 1999-2009 have detailed information in the CPRN.[23]

We excluded children born preterm (before week 37), multiple births, children with congenital malformations, children in transverse lie and those with lacking information on mode of delivery (Figure 1).

Study variables:

The predefined main outcome measures were stillbirth, NNM and CP. Stillbirth and NNM were defined according to the WHO.[24] Stillbirth was further divided into those who were dead before birth (antepartum) and during birth (intrapartum). Cerebral palsy was diagnosed and confirmed at five years of age according to the definition and classification proposed by the Surveillance of Cerebral Palsy in Europe.[25] Paediatricians at the Neurohabilitation centers in Norway completed the information of each child on a standardized form. Since fetuses who dies antepartum usually are delivered vaginally, stillbirth was not included in the analyses of risk associated with mode of delivery. However, since intrapartum death, NNM and CP may share the same causes we, as a secondary outcome, also calculated a composite adverse outcome variable comprising the sum of intrapartum death, NNM and CP.

Information on maternal age, parity, gestational age, mode of delivery, the child's sex, birth weight and Apgar scores was collected from the MBRN. Newborns with a birth weight below -2 standard deviations of the population mean weight[26] for gestational age, adjusted for sex were defined as small-for-gestational age (SGA).

Analytic approach:

First, we assessed the risks for stillbirth, NNM, CP and the composite outcome for children born in breech compared to cephalic position, independent of mode of delivery.

Second, we explored whether the risks for NNM, CP and the composite outcome were increased in children who had been born vaginally or by caesarean delivery (i.e. "actual mode of delivery") compared with those who had been born vaginally in cephalic position.

According to the Norwegian Society for Gynecology and Obstetrics vaginal breech delivery can be recommended if gestational age is at least 34 weeks, estimated birth weight is between 2000 and 4000 grams and no maternal and fetal contraindications for vaginal delivery exists. An essential premise of this recommendation is that the obstetric department is capable to perform immediate caesarean delivery and that trained paediatric personnel are available. Thus, some of the planned vaginal breech deliveries will be converted to a caesarean delivery during the birth process. The analysis of actual mode of delivery will therefore not evaluate these recommendation of vaginal births correctly, since the caesarean group will be a mixture of both planned and emergency caesarean delivery, and the vaginal group will comprise only those not changed to a caesarean delivery during birth.

Third, we therefore repeated the analyses, but now we compared the outcome of planned mode of breech delivery at admission to the obstetric department with planned vaginal cephalic delivery. We divided cephalic and breech births into the two categories originally planned vaginal and caesarean deliveries, based upon the initial handling of the birth, using information on how the birth started (spontaneous, induced, or by caesarean delivery) and

how caesarean delivery was recorded (as elective, emergency or planned). Births that did not satisfy these criteria were categorized as planned vaginal delivery.

The three outcomes, NNM, CP and the composite adverse outcome variable were then assessed related to the four exposure groups: cephalic position and planned vaginal births (reference group), cephalic position and planned caesarean delivery, breech position and planned vaginal births, and breech position and planned caesarean delivery.

Finally, we explored if the risk for NNM, CP and the composite outcome differed between children born by vaginal delivery and caesarean delivery or between planned vaginal delivery and planned caesarean delivery within the group of children who were born in breech.

Statistical analyses:

IBM SPSS software for Windows version 22 was used for data analyses. Differences in proportions between groups were analyzed using the chi-square test and prevalence rates with 95% confidence intervals (CI) were calculated according to Newcombe and Altman.[27] In the estimates of the prevalence of NNM, stillbirths were excluded and in the estimates of the prevalence of CP, stillbirths and children with post-neonatal CP were excluded. We used logistic regression to estimate odd ratios (OR) with 95% confidence intervals (CI) for adverse outcome of children in breech position at birth, using cephalic presentation as the reference. Moreover we explored the roles of potential confounders including maternal age, parity, gestational age, child sex and SGA status in multivariable logistic regression analyses, based on *a priori* knowledge and directed acyclic graphs methodology.[28]

Patient involvement:

No patients were involved in setting the research question or the outcome measures, nor were they involved in the design and implementation of the study. There are no plans to involve patients in dissemination.

RESULTS:

A total of 650 968 children were born in Norway during the study period. The study population of singleton children born at a gestational age of at least 37 weeks in either cephalic or breech position, and with no congenital anomalies comprised 520 047 children (Figure 1). A total of 841 (2 per 1000) of these were stillborn. Of the liveborn, 239 (0.5 per 1000) died in the neonatal period, and 552 children were diagnosed with CP. Of the latter, 32 had a post-neonatal cause of their CP, resulting in 520 with congenital CP (1 per 1000 liveborn).

Among the 520 047 included children, 16 700 (3%) were in breech and 503 347 (97%) in cephalic position (Figure 1). More mothers in the breech group were nullipara, and higher proportions of their infants were females, were born SGA and had low Apgar scores (Table 1). The mean gestational age of children born in breech was 39.1 weeks compared with 39.7 weeks for children born in cephalic position. Of the 16 700 women with a fetus in breech 7917 (47%) were planned for vaginal delivery while 5561 (33%) actually delivered vaginally. The corresponding figures for planned caesarean delivery was 8783 (53%) while 11 139 (67%) actually delivered by caesarean. For women with fetuses in cephalic position, 94 %

were planned to vaginal delivery while 90% delivered vaginally; 6% were planned to caesarean delivery and 10% delivered by caesarean.

Table 1: Maternal and infants characteristics in pregnancies where the child was born in breech or in cephalic position.

	Breech position		Cephalic position	
Number of births:	N 16 700	(%) (100)	N 503 347	(%) (100)
Maternal age[a]				
≤ 19 y	290	(2)	11 889	(2)
20-34 y	13 412	(80)	409 401	(81)
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Sex[b]				
Male	7540	(45)	257 128	(51)
Female	9160	(55)	246 216	(49)
Small for gestational age[c]	424	(2.5)	7130	(1.4)
Apgar score at 5 min[d]				
0-3	93	(0.6)	1477	(0.3)
4-6	427	(2.4)	7913	(1.7)
7-10	16 139	(97)	492 858	(98)

[a]Information on maternal age was missing in 26 children in cephalic.
[b]Information on Sex was missing in 1 child in cephalic.
[c]Information on Small for gestational age was missing in 12 children in breech and 361 in cephalic.
[d]Information on Apgar at 5 min was missing in 41 children in breech and 1099 in cephalic.

Children born in breech had increased risk for stillbirth, NNM, CP and the composite outcome compared with children born in cephalic position (Table 2). Sixty-eight of the stillborn children (Seven in breech and sixty-one in cephalic position) died during delivery.

Table 2. ***All births:*** Prevalence and unadjusted odds ratios (OR) with 95% confidence intervals (CI) for various adverse outcomes among singletons born at term, without congenital anomalies in cephalic and breech positions.

	Number of infants with adverse outcome	Total number of infants*	Prevalence per 1000 (CI)	OR (CI)
Stillbirths				
Cephalic	794	503 347	1.6 (1.5 to 1.7)	1.0 (Reference)
Breech	47	16 700	2.8 (2.1 to 3.7)	1.8 (1.3 to 2.4)
Stillbirth antepartum				
Cephalic	733	503 286	1.5 (1.4 to 1.6)	1.0 (Reference)
Breech	40	16 693	2.4 (1.8 to 3.3)	1.6 (1.2 to 2.3)
Stillbirth intrapartum				
Cephalic	61	502 614	0.1 (0.1 to 0.2)	1.0 (Reference)
Breech	7	16 600	0.4 (0.2 to 0.9)	3.5 (1.6-7.6)
Neonatal mortality (NNM)*				
Cephalic	225	502 553	0.5 (0.4 to 0.5)	1.0 (Reference)
Breech	14	16 653	0.8 (0.5 to 1.4)	1.9 (1.1 to 3.2)
Cerebral palsy (CP)**				
Cephalic	498	502 524	1.0 (0.9 to 1.1)	1.0 (Reference)
Breech	22	16 650	1.3 (0.9 to 2.0)	1.3 (0.9 to 2.1)
Composite outcome**				
Cephalic	784	502 585	1.6 (1.5 to 1.7)	1.0 (Reference)
Breech	43	16 657	2.6 (1.9 to 3.5)	1.7 (1.2 to 2.3)

*Removed stillbirths from the denominator in the analyses of NNM and CP.
** Removed post-neonatal CP from the denominator in the analyses of CP as outcome.
*** Comprising intrapartum death, NNM and CP.

According to actual mode of delivery, children in breech, regardless of whether they were born vaginally or by caesarean delivery, had a nearly three-fold increased OR for NNM compared with children born vaginally in cephalic, while the OR for CP was 1.7 (CI: 1.0 to 2.3) if the child was delivered by caesarean delivery (Table 3). As expected, children delivered by caesarean in cephalic position had higher prevalence of NNM, CP and the composite outcome compared with vaginal cephalic delivery, reflecting that caesarean delivery in this group is mainly done in high-risk births (Table 3).

Table 3. ***Actual mode of delivery:*** Prevalence and unadjusted odds ratios (OR) with 95% confidence intervals (CI) for various adverse outcomes among singletons born at term, without congenital anomalies according to actual mode of delivery.

	Number of infants with adverse outcome	Total number of infants*	Prevalence per 1000 (CI)	OR (CI)
Neonatal mortality (NNM)*				
Cephalic				
Vaginal delivery	137	451 064	0.3 (0.3 to 0.4)	1.0 (Reference)
Cesarean delivery	88	51 489	1.7 (1.4 to 2.1)	5.6 (4.3 to 7.4)
Breech:				
Vaginal delivery	5	5518	0.9 (0.4 to 2.1)	3.0 (1.2 to 7.3)
Cesarean delivery	9	11 135	0.8 (0.4 to 1.5)	2.7 (1.4 to 5.2)
Cerebral palsy (CP) **				
Cephalic				
Vaginal delivery	388	451 042	0.9 (0.8 to 1.0)	1.0 (Reference)
Cesarean delivery	110	51 482	2.1 (1.8 to 2.6)	2.5 (2.0 to 3.1)
Breech:				
Vaginal delivery	6	5517	1.1 (0.5 to 2.4)	1.3 (0.6 to 2.8)
Cesarean delivery	16	11 133	1.4 (0.9 to 2.3)	1.7 (1.0 to 2.8)
Composite outcome***				
Cephalic				
Vaginal delivery	553	451 070	1.2 (1.1 to 1.3)	1.0 (Reference)
Cesarean delivery	231	51 515	4.5 (3.9 to 5.1)	3.7 (3.1 to 4.3)
Breech:				
Vaginal delivery	17	5523	3.1 (1.9 to 4.9)	2.5 (1.5 to 4.1)
Cesarean delivery	26	11 134	2.3 (1.6 to 3.4)	1.9 (1.3 to 2.8)

*Removed stillbirths from the denominator in the analyses of NNM and CP.
** Removed post-neonatal CP from the denominator in the analyses of CP as outcome.
***Comprising intrapartum death, NNM and CP.

According to planned mode of delivery, the OR for NNM was 2.4 (CI: 1.5 to 3.4) for vaginal breech delivery compared with vaginal cephalic delivery whereas the OR for NNM among those planned to be delivered by caesarean was 1.6 (CI: 0.7 to 3.7) compared with planned vaginal cephalic delivery (Table 4). The prevalence of CP in planned vaginal breech and in planned caesarean breech delivery did not differ significantly from planned vaginal cephalic delivery (Table 4). Among children born in the cephalic position, the prevalence of NNM was higher among those born by caesarean, than among children born by vaginal delivery.

Table 4. ***Planned mode of delivery:*** Prevalence and unadjusted odds ratios (OR) with 95% confidence intervals (CI) for various adverse outcomes among singletons born at term, without congenital anomalies according to planned mode of delivery.

	Number of infants with adverse outcome	Total number of infants*	Prevalence per 1000 (CI)	OR (CI)
Neonatal mortality (NNM)*				
Cephalic				
Planned vaginal delivery	198	474 223	0.4 (0.4 to 0.5)	1.0 (Reference)
Planned caesarean delivery	27	28 330	1.0 (0.7 to 1.4)	2.3 (1.5 to 3.4)
Breech:				
Planned vaginal delivery	8	7873	1.0 (0.5 to 2.0)	2.4 (1.2 to 4.9)
Planned caesarean delivery	6	8780	0.7 (0.3 to 1.5)	1.6 (0.7 to 3.7)
Cerebral palsy (CP)				
Cephalic				
Planned vaginal delivery	453	474 198	1.0 (0.9 to 1.1)	1.0 (Reference)
Planned caesarean delivery	45	28 326	1.6 (1.2 to 2.1)	1.7 (1.2 to 2.3)
Breech:				
Planned vaginal delivery	10	7872	1.3 (0.7 to 2.3)	1.3 (0.7 to 2.5)
Planned caesarean delivery	12	8778	1.4 (0.8 to 2.4)	1.4 (0.8 to 2.5)
Composite outcome*				
Cephalic				
Planned vaginal delivery	705	474 252	1.5 (1.4 to 1.6)	1.0 (Reference)
Planned caesarean delivery	79	28 333	2.8 (2.2 to 3.5)	1.9 (1.5 to 2.4)
Breech:				
Planned vaginal delivery	24	7878	3.0 (2.1 to 4.5)	2.1 (1.4 to 3.1)
Planned caesarean delivery	19	8779	2.2 (1.4 to 3.4)	1.5 (0.9 to 2.3)

*Removed stillbirths from the denominator in the analyses of NNM and CP.
** Removed post-neonatal CP from the denominator in the analyses of CP as outcome.
***Comprising intrapartum death, NNM and CP.

In analyses restricted to the 16 700 children in breech position, the risk for NNM was not increased among infants actually born by vaginal delivery compared with caesarean delivery, while the OR for NNM in the group where vaginal delivery was planned was 1.5 (CI: 0.5 to 4.3) compared with planned caesarean delivery (Table 5). The risk for CP was not increased for children born by vaginal delivery compared with caesarean delivery regardless of actual or planned mode of delivery (Table 5).

Table 5. ***Restricted to breech deliveries:*** Prevalence and unadjusted odds ratios with 95% confidence intervals for various adverse outcomes among singletons in breech position born at term, without congenital anomalies according to actual and planned mode of delivery.

	Number of infants with adverse outcome	Total number of infants*	Prevalence per 1000 (CI)	OR (CI)
Neonatal mortality (NNM)*				
Actual mode of delivery				
Caesarean delivery	9	11 135	0.8 (0.4 to 1.5)	1.0 (Reference)
Vaginal delivery	5	5518	0.9 (0.4 to 2.1)	1.1 (0.4 to 3.3)
Planned mode of delivery				
Caesarean delivery	6	8780	0.7 (0.3 to 1.5)	1.0 (Reference)
Vaginal delivery	8	7873	1.0 (0.5 to 2.0)	1.5 (0.5 to 4.3)
Cerebral palsy (CP) **				
Actual mode of delivery				
Caesarean delivery	16	11 133	1.4 (0.9 to 2.3)	1.0 (Reference)
Vaginal delivery	6	5517	1.1 (0.5 to 2.4)	0.8 (0.3 to 1.9)
Planned mode of delivery				
Caesarean delivery	12	8778	1.4 (0.8 to 2.4)	1.0 (Reference)
Vaginal delivery	10	7872	1.3 (0.7 to 2.3)	0.9 (0.4 to 2.2)
Composite outcome**				
Actual mode of delivery				
Caesarean delivery	26	11 134	2.3 (1.6 to 3.4)	1.0 (Reference)
Vaginal delivery	17	5523	3.1 (1.9 to 4.9)	1.3 (0.7 to 2.4)
Planned mode of delivery				
Caesarean delivery	19	8779	2.2 (1.4 to 3.4)	1.0 (Reference)
Vaginal delivery	24	7878	3.0 (2.1 to 4.5)	1.4 (0.8 to 2.6)

*Removed stillbirths from the denominator in the analyses of NNM and CP.
** Removed post-neonatal CP from the denominator in the analyses of CP as outcome.
***Comprising intrapartum death, NNM and CP.

Multivariable analyses adjusting for gestational age, parity, maternal age, sex and SGA did not substantially affect any of the associations described above (data not shown).

DISCUSSION:

In this national cohort study of term singeltons without congenital malformations we found that vaginal breech delivery, regardless of whether it was planned or not, was associated with an excess risk for NNM and with a composite outcome of intrapartum death, NNM and CP, compared with cephalic vaginal delivery. However, also children who had actually been delivered by caesarean had excess risk for NNM and the composite outcome compared with

those who were actually born vaginally in the cephalic position, whereas a 60% increased risk for NNM, and a 50% increased risk for the composite outcome among those born in breech by planned caesarean delivery did not reach statistical significance, compared with planned vaginal cephalic delivery. A slightly higher prevalence of CP among children in breech, was not statistically significantly different from children born in cephalic position, regardless of mode of delivery. The risk for the composite outcome of intrapartum death , NNM and CP associated with the breech position was attenuated compared with the risk for NNM

Regardless of mode of delivery, the absolute risks for the adverse outcomes of breech births were low, ranging between 0.7 and 1.0 per 1000 liveborn for NNM, and it may be noteworthy that the prevalence rates for all adverse outcomes associated with vaginal breech delivery and with caesarean breech delivery were of similar magnitude.

Strengths and limitations:

Strengths of the present study are the large number of births and the prospectively recording of the data in the two registers. Nonetheless, among children in breech position, the number of children with the adverse outcomes NNM and CP were low, and the results of the analyses restricted to children in breech should therefore be interpreted with caution.

We restricted the analyses to singletons born at term and without congenital malformations, limiting the possibility of confounding by these factors. Multivariable analyses suggested that maternal age, parity, the child`s sex, gestational age and SGA did not confound the associations between breech position and adverse outcome.

Analysis of the association between mode of delivery and adverse outcome after breech delivery is challenging. Selection to vaginal delivery is recommended on strict criteria and is therefore expected to identify pregnancies with low risk for adverse outcome compared to those selected for caesarean delivery. Furthermore, some of the planned vaginal deliveries will be converted to an emergency caesarean delivery intrapartum, increasing the risk for adverse outcome in the caesarean group. A comparison of adverse outcome between vaginal and caesarean deliveries would therefore be expected to favor the vaginal delivered group. While this was the case for children born in cephalic position (table 3 and table 4), the ORs for NNM were similar or even higher in the vaginal compared with the caesarean delivery group for children born in breech. Thus, caution is needed in the interpretation of the lack of difference between vaginal and caesarean delivery.

We categorized not only according to actual mode of delivery, but also according to planned mode of delivery, which is essential in order to evaluate the national recommendations. Although the risk for NNM and the composite outcome was higher for actual than for planned caesarean delivery, as would be expected if the classification was correct, we cannot rule out some errors in this classification. Since the forms of the MBRN are completed immediately after birth, it is possible that some deliveries originally planned as caesarean delivery, may have been misclassified as emergency caesarean delivery. The latter is expected to be associated with higher risk for adverse outcome, and such misclassification would therefore erroneously reduce the risk associated with planned caesarean delivery. This misclassification would be expected to have most impact on the risk associated with planned breech caesarean delivery. A similar misclassification is also possible for planned vaginal cephalic deliveries, but in this case, the effect is negligible considering the large number of vaginal cephalic births, and the low proportion of cephalic caesarean delivery. Thus, the lack of statistical

significance of adverse outcome between planned breech caesarean delivery and planned vaginal cephalic delivery should be interpreted with caution.

Finally, the use of register-based data has limited ability to address explanatory factors, as suggested by Goffinet et al.[11] In their prospective study of breech deliveries, they found that 33 (26%) of 129 cases with severe neonatal complications had nonlethal major or minor malformations that sometimes explained the neonatal complications.[11] We cannot rule out that some undiagnosed or unrecorded malformations, may have contributed to the higher proportions of children with adverse outcomes among those born in breech than among those born in cephalic position in our study.

Comparison with other studies:

Our findings regarding excess risk for stillbirth[29] and NNM associated with breech presentation are consistent with earlier findings[30-32] and an excess risk for NNM was also reported in recent studies including children born after the TBT[2] in Denmark[4] and in Norway.[6]

We found a slightly higher risk for NNM in planned vaginal than in planned caesarean delivery and this could be considered to be consistent with the results of the TBT. On the other hand, the overall interpretation of our findings is that the risk for NNM was largely independent of mode of delivery, and this interpretation is not consistent with the results of the TBT. First, the different designs of the two studies may explain the different findings. The TBT was a randomized controlled trial considered to be the gold standard, while our study is

an observational study. Nonetheless, the much lower perinatal mortality in Norway compared with the TBT may also explain some of the diverging results in the two studies. Moreover, to be eligible to participate in the TBT, women had to have a singleton live fetus at term (≥ 37 week`s gestation) in breech without any known lethal fetal congenital anomaly. Women were excluded if there was evidence of fetopelvic disproportion, or if the fetus was judged to be clinically large or to have an estimated fetal weight of 4000 g or more, hyperextension of the fetal head or other fetal anomaly or condition that might cause a mechanical problem at delivery. Women with contraindication for labor or vaginal delivery such as placenta praevia were also excluded.[2] These criteria are similar to the criteria for vaginal breech delivery recommended by the Norwegian Society for Gynecology and Obstetrics. However, in the TBT a higher proportion of women (43%) selected for vaginal delivery needed caesarean delivery compared with our study population where only 30% of those selected for planned vaginal delivery needed caesarean delivery. One may therefore speculate that the probability for adverse outcome in the planned vaginal group in the TBT was higher than in our study. Instead antenatal acquired vulnerability may have played a larger role in our population.

The lack of excess risk for CP associated with breech position at birth is consistent with four studies published before the TBT, but inconsistent with four other studies. We are not aware of studies addressing the association between breech presentation at birth and CP in populations born after the TBT.[2] A follow-up study of 923 children included in that trial did not have the statistical power to address this severe, neurodevelopmental outcome.[33] Two earlier studies, including one from our own group, also found some evidence that the risk was associated with vaginal delivery.[12 15] The lower risk for CP in the present study, compared with our previous Norwegian study [12] could be explained by the larger sample size, better

quality of the data in the MBRN [21] and better ascertainment of cases in the CPRN in the present study.[23] Nonetheless, it is also possible that changes in the delivery of breech births in Norway including an increasing proportion of fetuses born by planned caesarean delivery,[6] may have improved outcome, and may reflect better selection of mothers for vaginal delivery.

Interpretation:

The overall higher risk for stillbirth and the higher proportion of infants born SGA among children born in breech than in cephalic position may suggest that fetuses with antenatal acquired risk factors for adverse outcomes are more likely to present in breech than in cephalic position at birth. On the other hand, the slightly higher ORs for NNM and for the composite outcome among children born vaginally than by caesarean delivery both when restricted to the breech group as well as when compared with vaginal cephalic delivery may suggest that fetuses in breech are more likely to experience complications during birth if they are born vaginally than if delivered by caesarean. This interpretation may be further supported by the fact that women selected for vaginal breech delivery would be expected to have a particular low risk for complications during birth. Thus, a combination of antenatal acquired risk factors for neonatal death with increased vulnerability to the birth process is probably the most likely explanation of our findings.

Regarding CP, antenatal factors are considered to be involved in 90% of the cases with CP,[34] and one might have expected an excess risk for CP in breech births, similar to that of NNM. The much lower risk for CP among children in breech, not statistically different from cephalic position could therefore suggest that antenatal factors increasing the risk for NNM are different or at least not completely overlapping with antenatal risk factors involved in the causal pathway leading to CP.

Implications:

Taking into consideration the very low absolute risk for NNM and CP, the increasing evidence for acute and long term maternal complications[35] and for later health problems among children following caesarean delivery,[36 37] our results suggest that vaginal delivery in selected cases may be an option for women with a fetus in breech position. This option requires that strict criteria are followed including access to competent obstetric care. In addition, a secondary advantage of having a certain volume of vaginal breech deliveries is that obstetricians retain their competence for unexpected vaginal breech deliveries. In the discussion with the pregnant mother and her partner regarding choice of delivery mode of a fetus in breech, the relative risk for NNM should be explained but related to the very low absolute risk. Moreover, it may be appropriate to emphasize that adverse outcome probably to a large degree is caused by antenatal acquired insults and that there are potential advantages of vaginal birth over caesarean delivery for long term health of the child and the mother. Regarding obstetric care, awareness of the excess risk for fetal death should be emphasized, and studies are warranted to optimize antenatal follow up of mothers with a fetus in breech.

Caution is needed if results of observational studies are included in the development of clinical guidelines, and more studies are needed to support our results. On the other hand, a new RCT in our part of the world is unrealistic as it would require the participation of 20 000 women with a fetus in breech in order to document a difference in NNM between mothers selected for planned vaginal and planned caesarean delivery.[10]

Nonetheless, the higher risk of neonatal mortality among planned vaginal deliveries than for planned caesarean delivery compared with cephalic delivery warrants further studies, including perinatal audits and prospective studies as suggested by Goffinet et al.[11]

CONCLUSION

Vaginal breech delivery, regardless of whether planned or actual and actual caesarean breech delivery were associated with excess risk for NNM and for a composite outcome of intrapartum death, NNM and CP, compared with vaginal cephalic delivery. The risk for adverse outcome in planned caesarean breech delivery did not differ significantly from planned vaginal cephalic delivery. However, the absolute risk for these outcomes was low. Taking into consideration potential long-term adverse consequences of caesarean delivery for the child, the mother and for later deliveries we therefore conclude that vaginal delivery may be offered to women with a fetus in breech, provided competent obstetric care and strict criteria for selection to vaginal delivery. Our findings did not support the notion that some cases of CP may be prevented if all fetuses in breech are delivered by caesarean delivery.

Competing interest: All authors have completed the ICMJE uniform disclosure form at www.icmj.org/cio_disclosure.pdf (available on request from the corresponding author) and declare: no financial support from any organization for the submitted work; no financial relationship with any companies that might have an interest in the submitted work in the previous three years; and have no non-financial interests or relationships that may be relevant to the submitted work.

Contributors: SB analyzed the data, contributed to study design and data interpretation and wrote the first draft of the manuscript. GA was principally responsible for the data from the CPRN and revised the manuscript. MM contributed to the data interpretation and revised the manuscript. PR contributed to the research hypotheses, study design and revised the manuscript. SH contribute to the data interpretation and revised the manuscript. DM contributed to study design, the data analyses, data interpretation and revised the manuscript. TV proposed the research questions, and contributed in the interpretation of the data and the revision of the manuscript. He is the guarantor of the study and accepts full responsibility for the work and the conduct of the study. All authors approved the final version of the submitted manuscript**.**

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Ethical approval: The study was approved by the Regional Ethical Committee for Medical Research in Mid-Norway (ref 2011/754).

Data sharing: The protocol is available on request from the corresponding author at: sbjellmo@hotmail.com

Transparency declaration: The lead author affirms that this manuscript is an honest, accurate, and transparent account of the study being reported; that no important aspects of the study have been omitted; and that any discrepancies from the study as planned have been explained.

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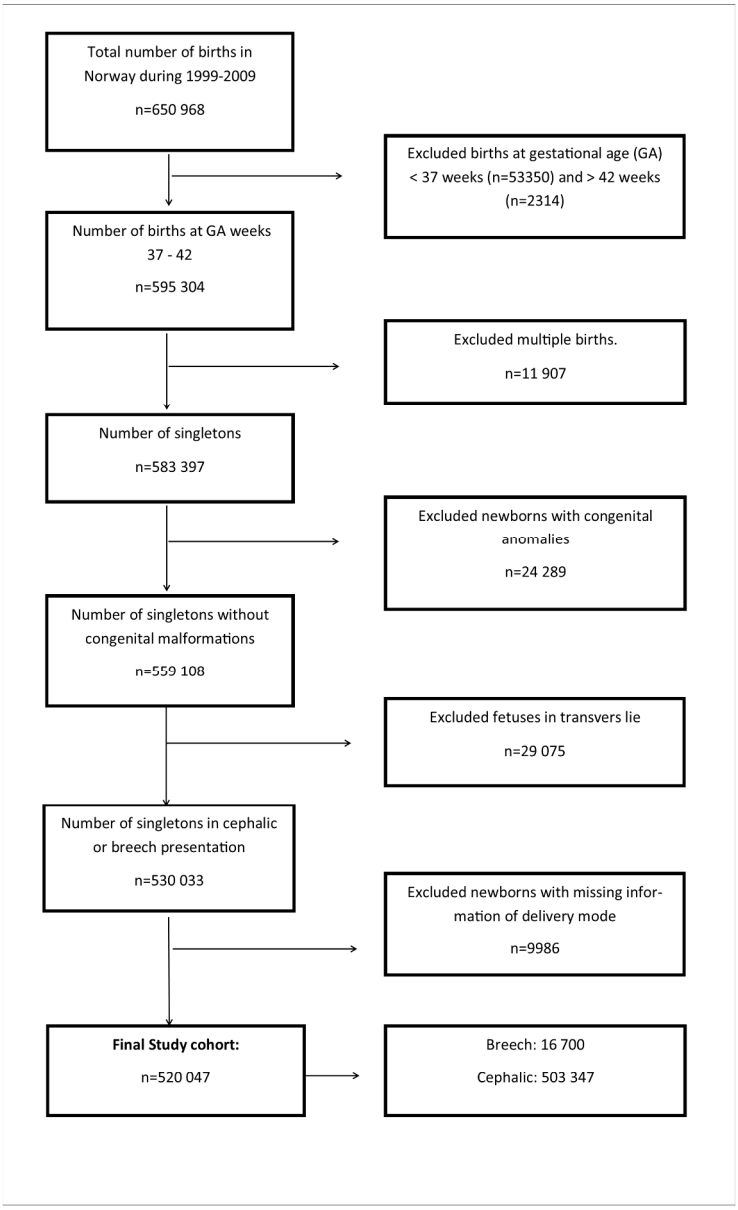

Flow chart of the study population.
Figure 1
159x260mm (300 x 300 DPI)

STROBE Statement—checklist of items that should be included in reports of observational studies

	Item No.	Recommendation	Page No.	Relevant text from manuscript
Title and abstract	1	(*a*) Indicate the study's design with a commonly used term in the title or the abstract	1	Yes
		(*b*) Provide in the abstract an informative and balanced summary of what was done and what was found	2-3	Yes
Introduction				
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	4-5	Introduction
Objectives	3	State specific objectives, including any prespecified hypotheses	5	The aim of this study was therefore to explore if singletons without congenital malformations born vaginally at term have higher risk for stillbirth, neonatal mortality (NNM) and CP if they are born in breech position compared to cephalic position.
Methods				
Study design	4	Present key elements of study design early in the paper	5	In this population based study, perinatal data from all children born in Norway during 1999-2009 were retrieved from the Medical Birth Registry of Norway (MBRN), and combined with information recorded in the Cerebral Palsy Register of Norway (CPRN).
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection	5	Data from the MBRN and CPRN (1999-2009)
Participants	6	(*a*) *Cohort study*—Give the eligibility criteria, and the sources and methods of selection of participants. Describe methods of follow-up *Case-control study*—Give the eligibility criteria, and the sources and methods of case ascertainment and control selection. Give the rationale for the choice of cases and controls *Cross-sectional study*—Give the eligibility criteria, and the sources and methods of selection of	5	Cohort-study with data from the MBRN and CPRN.

		participants		
		(*b*) *Cohort study*—For matched studies, give matching criteria and number of exposed and unexposed *Case-control study*—For matched studies, give matching criteria and the number of controls per case		
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	6	The predefined main outcome measures were stillbirth, NNM and CP. Stillbirth and NNM were defined according to the WHO
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group	*7-8*	*Analytic approach*
Bias	9	Describe any efforts to address potential sources of bias	7-8	Compared both actual and planned mode of delivery.
Study size	10	Explain how the study size was arrived at	6	We excluded children born preterm (before week 37), multiple births, children with congenital malformations, children in transverse lie and those with lacking information on mode of delivery (Figure 1).

Continued on next page

Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why		
Statistical methods	12	(*a*) Describe all statistical methods, including those used to control for confounding	8	IBM SPSS software for Windows version 22 was used for data analyses. Differences in proportions between groups were analyzed using the chi-square test and prevalence rates with 95% confidence intervals (CI) were calculated according to Newcombe and Altman. In the estimates of the prevalence of NNM, stillbirths were excluded and in the estimates of the prevalence of CP, stillbirths and children with post neonatal CP were excluded. We used logistic regression to estimate odd ratios (OR) with 95% confidence intervals (CI) for adverse outcome of children in breech position at birth, using cephalic presentation as the reference. Moreover we explored the roles of potential confounders including maternal age, parity, gestational age, child sex and SGA status in multivariable logistic regression analyses based on *a priori* knowledge and directed acyclic graphs methodology.
		(*b*) Describe any methods used to examine subgroups and interactions		
		(*c*) Explain how missing data were addressed	Table 1 (10)	Information on missing.
		(*d*) *Cohort study*—If applicable, explain how loss to follow-up was addressed *Case-control study*—If applicable, explain how matching of cases and controls was addressed *Cross-sectional study*—If applicable, describe analytical methods taking account of sampling strategy		
		(*e*) Describe any sensitivity analyses		

Results				
Participants	13*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed	9	A total of 650 968 children were born in Norway during the study period. The study population of singleton children born at a gestational age of at least 37 weeks in either cephalic or breech position, and with no congenital malformation comprised 520 047 children.
		(b) Give reasons for non-participation at each stage		
		(c) Consider use of a flow diagram	Figure 1	Yes
Descriptive data	14*	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders	9	Among the 520 047 included children, 16 700 (3%) were in breech and 503 347 (97%) in cephalic position (Table 1). More mothers in the breech group were nullipara, and a higher proportion of their infants were females, were born SGA and had low Apgar scores (Table 1). The mean gestational age of children born in breech was 39.1 weeks compared with 39.7 weeks for children born in cephalic.
		(b) Indicate number of participants with missing data for each variable of interest		
		(c) *Cohort study*—Summarise follow-up time (eg, average and total amount)		
Outcome data	15*	*Cohort study*—Report numbers of outcome events or summary measures over time	*10-14*	*Among all, actual delivery and planned delivery.*
		Case-control study—Report numbers in each exposure category, or summary measures of exposure		
		Cross-sectional study—Report numbers of outcome events or summary measures		

Main results	16	(*a*) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included	10-14 Table 2-5	See tables and text.
		(*b*) Report category boundaries when continuous variables were categorized		
		(*c*) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period		

Continued on next page

Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses	14	Multivariable analyses adjusting for gestational age, parity, maternal age, sex and SGA did not substantially affect any of the associations described above (data not shown).
Discussion				
Key results	18	Summarise key results with reference to study objectives	14	In this national cohort study of term singletons without congenital malformations we found that breech vaginal delivery, regardless of whether it was planned or not, was associated with an excess risk for NNM and with a composite outcome of intrapartum death, NNM and CP, compared with cephalic vaginal delivery.
Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias	15-17	Strength and limitations
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence	19	Interpretation
Generalisability	21	Discuss the generalisability (external validity) of the study results	19-21	Interpretation and implications.
Other information				
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	22	Supported by a grant from The Liaison Committee between the Central Norway Regional Health Authority (RHA) and the Norwegian University of Science and Technology (NTNU).

*Give information separately for cases and controls in case-control studies and, if applicable, for exposed and unexposed groups in cohort and cross-sectional studies.

Note: An Explanation and Elaboration article discusses each checklist item and gives methodological background and published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely available on the Web sites of PLoS Medicine at http://www.plosmedicine.org/, Annals of Internal Medicine at http://www.annals.org/, and Epidemiology at http://www.epidem.com/). Information on the STROBE Initiative is available at www.strobe-statement.org.

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Is vaginal breech delivery associated with higher risk for perinatal death and cerebral palsy compared with vaginal cephalic birth? Registry-based cohort study in Norway.

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Is vaginal breech delivery associated with higher risk for perinatal death and cerebral palsy compared with vaginal cephalic birth?
Registry-based cohort study in Norway.

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ABSTRACT:

Objective: To study if vaginal breech delivery is associated with increased risk for neonatal mortality (NNM) or cerebral palsy (CP) in Norway where vaginal delivery accounts for 1/3 of all breech deliveries.

Design: Cohort study using information from the national Medical Birth- and Cerebral Palsy Registers.

Setting: Births in Norway 1999-2009.

Participants: 520 047 term born singletons without congenital malformations.

Main outcome measures: NNM, CP and a composite outcome of these and death during birth.

Results: Compared with cephalic births, breech births had substantially increased risk for NNM but not for CP. Vaginal delivery was planned for 7917 of 16700 fetuses in breech, while 5561 actually delivered vaginally. Among these, NNM was 0.9 per 1000 compared with 0.3 per 1000 in vaginal cephalic delivery, and 0.8 per 1000 in those actually born by caesarean delivery (CD) in breech. Compared with planned cephalic delivery, planned vaginal delivery was associated with excess risk for NNM (OR: 2.4; 95%CI:1.2 to 4.9), while the OR associated with planned breech CD was 1.6 (95%CI: 0.7 to 3.7). These risks were attenuated when NNM was substituted by the composite outcome. Vaginal breech delivery was not associated with excess risk for CP compared with vaginal cephalic delivery.

Conclusion: Vaginal breech delivery, regardless of whether planned or actual, and actual breech CD were associated with excess risk for NNM compared with vaginal cephalic delivery, but not with CP. The risk for NNM and CP in planned breech CD did not differ significantly from planned vaginal cephalic delivery. However, the absolute risk for these

outcomes was low and taking into consideration potential long-term adverse consequences of CD for the child and later deliveries we therefore conclude that vaginal breech delivery may be recommended, provided competent obstetric care and strict criteria for selection to vaginal delivery.

ARTICLE SUMMARY:

Strengths and limitations of this study:

- More than 500 000 births included in the study.
- Prospectively recording of the data in the two registers.
- Restriction of the analyses to singletons at term without congenital malformation.
- The number of infants with adverse outcomes in breech were low.
- Register based data has limited ability to address explanatory factors.

INTRODUCTION:

Mode of delivery of a fetus in breech position is a controversial issue.[1] The Term Breech Trial (TBT)[2] reported lower perinatal mortality and morbidity of fetuses in breech position following planned caesarean delivery compared with planned vaginal delivery. The study had great impact, changing clinical practice in a number of countries.[3-6] However, the conclusion of the TBT were criticized by several experts.[7-9] In Norway, the Norwegian Board of Health invited a group of national experts to review the evidence underlying these recommendations. The expert group reviewed the literature published between 1980 – 2001. Taking into account the much lower perinatal mortality in Norway than that reported in the TBT, they concluded that vaginally breech delivery would still be safe, provided careful selection of mothers, qualified clinicians, and adequate fetal assessment.[10] Therefore, approximately 1/3 of fetuses in breech position in Norway are still delivered vaginally.[6] In a prospective study in France and Belgium, Goffinet et al compared vaginal delivery with planned caesarean delivery in breech. They concluded, in line with the Norwegian recommendations, that vaginal delivery is a safe option when strict selection criteria are followed.[11] The controversies of mode of delivery have also been reflected in studies of the long-term outcome of infants born in breech position. Several studies reported that infants born in breech had increased risk for cerebral palsy (CP).[12-15]Although it was unclear whether mode of delivery affected this increased risk,[16-18] it has been suggested that planned caesarean delivery may prevent some cases of CP[12 13].

In the vast majority of previous studies on adverse outcome of vaginal breech delivery, the comparison group has been caesarean breech delivery. However, the main results of these studies do not take into account the risk for complications of caesarean delivery in later pregnancies both for the mother and for the child. There is also an increased awareness of later health problems in children born by caesarean suggested in recent reports[19]. Therefore, to

assess if vaginal breech delivery as currently practiced in Norway can be characterized as safe, the appropriate comparison group of breech deliveries should be vaginal cephalic birth; which is the natural way of giving birth. In line with this, a recent systematic review and meta-analysis of breech deliveries recommended that comparative studies of vaginal breech with vaginal cephalic deliveries should be undertaken[20].

The aim of this study was therefore to explore if singletons without congenital malformations born vaginally at term have higher risk for stillbirth, neonatal mortality (NNM) and CP if they are born in breech position compared to cephalic position.

METHODS:

In this population based study, perinatal data from all children born in Norway during 1999-2009 were retrieved from the Medical Birth Registry of Norway (MBRN), and combined with information recorded in the Cerebral Palsy Register of Norway (CPRN). The 11-digit personal identification number unique for every Norwegian citizen was used to link information from the two registers. The MBRN records demographic variables, as well as information on maternal health before and during pregnancy, interventions and complications during delivery and neonatal outomes. Registration in this register has been compulsory since 1967 ensuring prospective recording of this information at birth.[21]

The Cerebral Palsy Register of Norway (CPRN) is an informed consent based national quality register established in 2006, and aims to record detailed information on all children with CP born in Norway since 1996.[22] Information is reported at diagnosis, at 5 years and at 15-17 years of age. Neuropediatric habilitation centres in Norway provide summary and detailed

data about the children. A validation study indicated that 80% of children with CP in Norway born 1999-2009 have detailed information in the CPRN.[23]

We excluded children born preterm (before week 37), multiple births, children with congenital malformations, children in transverse lie and those with lacking information on mode of delivery (Figure 1).

Study variables:

The predefined main outcome measures were stillbirth, NNM and CP. Stillbirth and NNM were defined according to the WHO.[24] Stillbirth was further divided into those who were dead before birth (antepartum) and during birth (intrapartum). Cerebral palsy was diagnosed and confirmed at five years of age according to the definition and classification proposed by the Surveillance of Cerebral Palsy in Europe.[25] Paediatricians at the Neurohabilitation centers in Norway completed the information of each child on a standardized form. Since fetuses who dies antepartum usually are delivered vaginally, stillbirth was not included in the analyses of risk associated with mode of delivery. However, since intrapartum death, NNM and CP may share the same causes we, as a secondary outcome, also calculated a composite adverse outcome variable comprising the sum of intrapartum death, NNM and CP.

Information on maternal age, parity, gestational age, mode of delivery, the child's sex, birth weight and Apgar scores was collected from the MBRN. Newborns with a birth weight below -2 standard deviations of the population mean weight[26] for gestational age, adjusted for sex were defined as small-for-gestational age (SGA).

Analytic approach:

First, we assessed the risks for stillbirth, NNM, CP and the composite outcome for children born in breech compared to cephalic position, independent of mode of delivery.

Second, we explored the risks for NNM, CP and the composite outcome according to "actual mode of delivery" by comparing vaginal or caesarean breech delivery with vaginal cephalic delivery.

According to the Norwegian Society for Gynecology and Obstetrics vaginal breech delivery can be recommended if gestational age is at least 34 weeks, estimated birth weight is between 2000 and 4000 grams and no maternal and fetal contraindications for vaginal delivery exists. An essential premise of this recommendation is that the obstetric department is capable to perform immediate caesarean delivery and that trained paediatric personnel are available. Thus, some of the planned vaginal breech deliveries will be converted to a caesarean delivery during the birth process. The analysis of actual mode of delivery will therefore not evaluate these recommendation of vaginal births correctly, since the caesarean group will be a mixture of both planned and emergency caesarean delivery, and the vaginal group will comprise only those not changed to a caesarean delivery during birth.

Third, we therefore repeated the analyses, but now we compared the outcome of planned mode of breech delivery at admission to the obstetric department with planned vaginal cephalic delivery. We divided cephalic and breech births into the two categories originally planned vaginal and caesarean deliveries, based upon the initial handling of the birth, using information on how the birth started (spontaneous, induced, or by caesarean delivery) and

how caesarean delivery was recorded (as elective, emergency or planned). Births that did not satisfy these criteria were categorized as planned vaginal delivery.

The three outcomes, NNM, CP and the composite adverse outcome variable were then assessed related to the four exposure groups: cephalic position and planned vaginal births (reference group), cephalic position and planned caesarean delivery, breech position and planned vaginal births, and breech position and planned caesarean delivery.

Finally, we explored if the risk for NNM, CP and the composite outcome differed between children born by vaginal delivery and caesarean delivery or between planned vaginal delivery and planned caesarean delivery within the group of children who were born in breech.

Statistical analyses:

IBM SPSS software for Windows version 22 was used for data analyses. Differences in proportions between groups were analyzed using the chi-square test and prevalence rates with 95% confidence intervals (CI) were calculated according to Newcombe and Altman.[27] In the estimates of the prevalence of NNM, stillbirths were excluded and in the estimates of the prevalence of CP, stillbirths and children with post-neonatal CP were excluded. We used logistic regression to estimate odd ratios (OR) with 95% confidence intervals (CI) for adverse outcome of children in breech position at birth, using cephalic presentation as the reference. Moreover we explored the roles of potential confounders including maternal age, parity, gestational age, child sex and SGA status in multivariable logistic regression analyses, based on *a priori* knowledge and directed acyclic graphs methodology.[28]

Patient involvement:

No patients were involved in setting the research question or the outcome measures, nor were they involved in the design and implementation of the study. There are no plans to involve patients in dissemination.

RESULTS:

A total of 650 968 children were born in Norway during the study period. The study population of singleton children born at a gestational age of at least 37 weeks in either cephalic or breech position, and with no congenital anomalies comprised 520 047 children (Figure 1). A total of 841 (2 per 1000) of these were stillborn. Of the liveborn, 239 (0.5 per 1000) died in the neonatal period, and 552 children were diagnosed with CP. Of the latter, 32 had a post-neonatal cause of their CP, resulting in 520 with congenital CP (1 per 1000 liveborn).

Among the 520 047 included children, 16 700 (3%) were in breech and 503 347 (97%) in cephalic position (Figure 1). More mothers in the breech group were nullipara, and higher proportions of their infants were females, were born SGA and had low Apgar scores (Table 1). The mean gestational age of children born in breech was 39.1 weeks compared with 39.7 weeks for children born in cephalic position. Of the 16 700 women with a fetus in breech 7917 (47%) were planned for vaginal delivery while 5561 (33%) actually delivered vaginally. The corresponding figures for planned caesarean delivery was 8783 (53%) while 11 139 (67%) actually delivered by caesarean. For women with fetuses in cephalic position, 94 %

were planned to vaginal delivery while 90% delivered vaginally; 6% were planned to caesarean delivery and 10% delivered by caesarean.

Table 1: Maternal and infants characteristics in pregnancies where the child was born in breech or in cephalic position.

	Breech position		Cephalic position	
Number of births:	N 16 700	(%) (100)	N 503 347	(%) (100)
Maternal age[a]				
≤ 19 y	290	(2)	11 889	(2)
20-34 y	13 412	(80)	409 401	(81)
≥ 35 y	2998	(18)	82 031	(17)
Parity				
Nullipara	9280	(56)	199 822	(40)
Primipara	4599	(27)	184 068	(36)
>1 para	2822	(17)	119 457	(24)
Sex[b]				
Male	7540	(45)	257 128	(51)
Female	9160	(55)	246 216	(49)
Small for gestational age[c]	424	(2.5)	7130	(1.4)
Apgar score at 5 min[d]				
0-3	93	(0.6)	1477	(0.3)
4-6	427	(2.4)	7913	(1.7)
7-10	16 139	(97)	492 858	(98)

[a]Information on maternal age was missing in 26 children in cephalic.
[b]Information on Sex was missing in 1 child in cephalic.
[c]Information on Small for gestational age was missing in 12 children in breech and 361 in cephalic.
[d]Information on Apgar at 5 min was missing in 41 children in breech and 1099 in cephalic.

Children born in breech had increased risk for stillbirth, NNM, CP and the composite outcome compared with children born in cephalic position (Table 2). Sixty-eight of the stillborn children (Seven in breech and sixty-one in cephalic position) died during delivery.

Table 2. ***All births:*** Prevalence and unadjusted odds ratios (OR) with 95% confidence intervals (CI) for various adverse outcomes among singletons born at term, without congenital anomalies in cephalic and breech positions.

	Number of infants with adverse outcome	Total number of infants*	Prevalence per 1000 (CI)	OR (CI)
Stillbirths				
Cephalic	794	503 347	1.6 (1.5 to 1.7)	1.0 (Reference)
Breech	47	16 700	2.8 (2.1 to 3.7)	1.8 (1.3 to 2.4)
Stillbirth antepartum				
Cephalic	733	503 286	1.5 (1.4 to 1.6)	1.0 (Reference)
Breech	40	16 693	2.4 (1.8 to 3.3)	1.6 (1.2 to 2.3)
Stillbirth intrapartum				
Cephalic	61	502 614	0.1 (0.1 to 0.2)	1.0 (Reference)
Breech	7	16 600	0.4 (0.2 to 0.9)	3.5 (1.6-7.6)
Neonatal mortality (NNM)*				
Cephalic	225	502 553	0.5 (0.4 to 0.5)	1.0 (Reference)
Breech	14	16 653	0.8 (0.5 to 1.4)	1.9 (1.1 to 3.2)
Cerebral palsy (CP)**				
Cephalic	498	502 524	1.0 (0.9 to 1.1)	1.0 (Reference)
Breech	22	16 650	1.3 (0.9 to 2.0)	1.3 (0.9 to 2.1)
Composite outcome**				
Cephalic	784	502 585	1.6 (1.5 to 1.7)	1.0 (Reference)
Breech	43	16 657	2.6 (1.9 to 3.5)	1.7 (1.2 to 2.3)

*Removed stillbirths from the denominator in the analyses of NNM and CP.
** Removed post-neonatal CP from the denominator in the analyses of CP as outcome.
*** Comprising intrapartum death, NNM and CP.

According to actual mode of delivery, children in breech, regardless of whether they were born vaginally or by caesarean delivery, had a nearly three-fold increased OR for NNM compared with children born vaginally in cephalic, while the OR for CP was 1.7 (CI: 1.0 to 2.8) if the child was delivered by caesarean delivery (Table 3). As expected, children delivered by caesarean in cephalic position had higher prevalence of NNM, CP and the composite outcome compared with vaginal cephalic delivery, reflecting that caesarean delivery in this group is mainly done in high-risk births (Table 3).

Table 3. ***Actual mode of delivery:*** Prevalence and unadjusted odds ratios (OR) with 95% confidence intervals (CI) for various adverse outcomes among singletons born at term, without congenital anomalies according to actual mode of delivery.

	Number of infants with adverse outcome	Total number of infants*	Prevalence per 1000 (CI)	OR (CI)
Neonatal mortality (NNM)*				
Cephalic				
Vaginal delivery	137	451 064	0.3 (0.3 to 0.4)	1.0 (Reference)
Cesarean delivery	88	51 489	1.7 (1.4 to 2.1)	5.6 (4.3 to 7.4)
Breech:				
Vaginal delivery	5	5518	0.9 (0.4 to 2.1)	3.0 (1.2 to 7.3)
Cesarean delivery	9	11 135	0.8 (0.4 to 1.5)	2.7 (1.4 to 5.2)
Cerebral palsy (CP) **				
Cephalic				
Vaginal delivery	388	451 042	0.9 (0.8 to 1.0)	1.0 (Reference)
Cesarean delivery	110	51 482	2.1 (1.8 to 2.6)	2.5 (2.0 to 3.1)
Breech:				
Vaginal delivery	6	5517	1.1 (0.5 to 2.4)	1.3 (0.6 to 2.8)
Cesarean delivery	16	11 133	1.4 (0.9 to 2.3)	1.7 (1.0 to 2.8)
Composite outcome***				
Cephalic				
Vaginal delivery	553	451 070	1.2 (1.1 to 1.3)	1.0 (Reference)
Cesarean delivery	231	51 515	4.5 (3.9 to 5.1)	3.7 (3.1 to 4.3)
Breech:				
Vaginal delivery	17	5523	3.1 (1.9 to 4.9)	2.5 (1.5 to 4.1)
Cesarean delivery	26	11 134	2.3 (1.6 to 3.4)	1.9 (1.3 to 2.8)

*Removed stillbirths from the denominator in the analyses of NNM and CP.
** Removed post-neonatal CP from the denominator in the analyses of CP as outcome.
***Comprising intrapartum death, NNM and CP.

According to planned mode of delivery, vaginal breech delivery had an estimated 2.4 (CI: 1.2 to 4.9) times increased risk of NNM, and a 2.1 (CI: 1.4 to 3.1) times increased risk of the composite outcome. Planned caesarean breech delivery had an estimated 1.6 (CI: 0.7 to 3.7) increased risk of NNM and a 1.5 (CI: 0.9 to 2.3) times increased risk of the composite outcome, both compared to planned vaginal cephalic delivery (table 4). The prevalence of CP in planned vaginal breech and in planned caesarean breech delivery did not differ significantly from planned vaginal cephalic delivery (table 4). Among children born in the cephalic position, the prevalence of NNM, CP and the composite outcome was higher among those born by caesarean, than among those born by vaginal delivery.

Table 4. ***Planned mode of delivery:*** Prevalence and unadjusted odds ratios (OR) with 95% confidence intervals (CI) for various adverse outcomes among singletons born at term, without congenital anomalies according to planned mode of delivery.

	Number of infants with adverse outcome	Total number of infants*	Prevalence per 1000 (CI)	OR (CI)
Neonatal mortality (NNM)*				
Cephalic				
Planned vaginal delivery	198	474 223	0.4 (0.4 to 0.5)	1.0 (Reference)
Planned caesarean delivery	27	28 330	1.0 (0.7 to 1.4)	2.3 (1.5 to 3.4)
Breech:				
Planned vaginal delivery	8	7873	1.0 (0.5 to 2.0)	2.4 (1.2 to 4.9)
Planned caesarean delivery	6	8780	0.7 (0.3 to 1.5)	1.6 (0.7 to 3.7)
Cerebral palsy (CP)				
Cephalic				
Planned vaginal delivery	453	474 198	1.0 (0.9 to 1.1)	1.0 (Reference)
Planned caesarean delivery	45	28 326	1.6 (1.2 to 2.1)	1.7 (1.2 to 2.3)
Breech:				
Planned vaginal delivery	10	7872	1.3 (0.7 to 2.3)	1.3 (0.7 to 2.5)
Planned caesarean delivery	12	8778	1.4 (0.8 to 2.4)	1.4 (0.8 to 2.5)
Composite outcome*				
Cephalic				
Planned vaginal delivery	705	474 252	1.5 (1.4 to 1.6)	1.0 (Reference)
Planned caesarean delivery	79	28 333	2.8 (2.2 to 3.5)	1.9 (1.5 to 2.4)
Breech:				
Planned vaginal delivery	24	7878	3.0 (2.1 to 4.5)	2.1 (1.4 to 3.1)
Planned caesarean delivery	19	8779	2.2 (1.4 to 3.4)	1.5 (0.9 to 2.3)

*Removed stillbirths from the denominator in the analyses of NNM and CP.
** Removed post-neonatal CP from the denominator in the analyses of CP as outcome.
***Comprising intrapartum death, NNM and CP.

In analyses restricted to the 16 700 children in breech position, the risk for NNM was not increased among infants actually born by vaginal delivery compared with caesarean delivery, while the OR for NNM in the group where vaginal delivery was planned was 1.5 (CI: 0.5 to 4.3) compared with planned caesarean delivery (Table 5). The risk for CP was not increased for children born by vaginal delivery compared with caesarean delivery regardless of actual or planned mode of delivery (Table 5).

Table 5. ***Restricted to breech deliveries:*** Prevalence and unadjusted odds ratios with 95% confidence intervals for various adverse outcomes among singletons in breech position born at term, without congenital anomalies according to actual and planned mode of delivery.

	Number of infants with adverse outcome	Total number of infants*	Prevalence per 1000 (CI)	OR (CI)
Neonatal mortality (NNM)*				
Actual mode of delivery				
Caesarean delivery	9	11 135	0.8 (0.4 to 1.5)	1.0 (Reference)
Vaginal delivery	5	5518	0.9 (0.4 to 2.1)	1.1 (0.4 to 3.3)
Planned mode of delivery				
Caesarean delivery	6	8780	0.7 (0.3 to 1.5)	1.0 (Reference)
Vaginal delivery	8	7873	1.0 (0.5 to 2.0)	1.5 (0.5 to 4.3)
Cerebral palsy (CP) **				
Actual mode of delivery				
Caesarean delivery	16	11 133	1.4 (0.9 to 2.3)	1.0 (Reference)
Vaginal delivery	6	5517	1.1 (0.5 to 2.4)	0.8 (0.3 to 1.9)
Planned mode of delivery				
Caesarean delivery	12	8778	1.4 (0.8 to 2.4)	1.0 (Reference)
Vaginal delivery	10	7872	1.3 (0.7 to 2.3)	0.9 (0.4 to 2.2)
Composite outcome**				
Actual mode of delivery				
Caesarean delivery	26	11 134	2.3 (1.6 to 3.4)	1.0 (Reference)
Vaginal delivery	17	5523	3.1 (1.9 to 4.9)	1.3 (0.7 to 2.4)
Planned mode of delivery				
Caesarean delivery	19	8779	2.2 (1.4 to 3.4)	1.0 (Reference)
Vaginal delivery	24	7878	3.0 (2.1 to 4.5)	1.4 (0.8 to 2.6)

*Removed stillbirths from the denominator in the analyses of NNM and CP.
** Removed post-neonatal CP from the denominator in the analyses of CP as outcome.
***Comprising intrapartum death, NNM and CP.

Multivariable analyses adjusting for gestational age, parity, maternal age, sex and SGA did not substantially affect any of the associations described above (data not shown).

DISCUSSION:

In this national cohort study of term singeltons without congenital malformations we found that vaginal breech delivery, regardless of whether it was planned or not, was associated with an excess risk for NNM and with a composite outcome of intrapartum death, NNM and CP, compared with cephalic vaginal delivery. However, also children who had actually been delivered by caesarean had excess risk for NNM and the composite outcome compared with those who were actually born vaginally in the cephalic position, whereas a 60% increased risk

for NNM, and a 50% increased risk for the composite outcome among those born in breech by planned caesarean delivery did not reach statistical significance, compared with planned vaginal cephalic delivery. A slightly higher prevalence of CP among children in breech, was not statistically significantly different from children born in cephalic position, regardless of mode of delivery. The risk for the composite outcome of intrapartum death, NNM and CP associated with the breech position was attenuated compared with the risk for NNM

Regardless of mode of delivery, the absolute risks for the adverse outcomes of breech births were low, ranging between 0.7 and 1.0 per 1000 liveborn for NNM, and it may be noteworthy that the prevalence rates for all adverse outcomes associated with vaginal breech delivery and with caesarean breech delivery were of similar magnitude.

Strengths and limitations:

Strengths of the present study are the large number of births and the prospectively recording of the data in the two registers. Nonetheless, among children in breech position, the number of children with the adverse outcomes NNM and CP were low, and the results of the analyses restricted to children in breech should therefore be interpreted with caution.

We restricted the analyses to singletons born at term and without congenital malformations, limiting the possibility of confounding by these factors. Multivariable analyses suggested that maternal age, parity, the child`s sex, gestational age and SGA did not confound the associations between breech position and adverse outcome.

Analysis of the association between mode of delivery and adverse outcome after breech delivery is challenging. Selection to vaginal delivery is recommended on strict criteria and is therefore expected to identify pregnancies with low risk for adverse outcome compared to those selected for caesarean delivery. Furthermore, some of the planned vaginal deliveries will be converted to an emergency caesarean delivery intrapartum, increasing the risk for adverse outcome in the caesarean group. A comparison of adverse outcome between vaginal and caesarean deliveries would therefore be expected to favor the vaginal delivered group. While this was the case for children born in cephalic position (table 3 and table 4), the ORs for NNM were similar or even higher in the vaginal compared with the caesarean delivery group for children born in breech. Thus, caution is needed in the interpretation of the lack of difference between vaginal and caesarean delivery.

We categorized not only according to actual mode of delivery, but also according to planned mode of delivery, which is essential in order to evaluate the national recommendations. Although the risk for NNM and the composite outcome was higher for actual than for planned caesarean delivery, as would be expected if the classification was correct, we cannot rule out some errors in this classification. Since the forms of the MBRN are completed immediately after birth, it is possible that some deliveries originally planned as caesarean delivery, may have been misclassified as emergency caesarean delivery. The latter is expected to be associated with higher risk for adverse outcome, and such misclassification would therefore erroneously reduce the risk associated with planned caesarean delivery. This misclassification would be expected to have most impact on the risk associated with planned breech caesarean delivery. A similar misclassification is also possible for planned vaginal cephalic deliveries, but in this case, the effect is negligible considering the large number of vaginal cephalic births, and the low proportion of cephalic caesarean delivery. Thus, the lack of statistical

significance of adverse outcome between planned breech caesarean delivery and planned vaginal cephalic delivery should be interpreted with caution.

Finally, the use of register-based data has limited ability to address explanatory factors, as suggested by Goffinet et al.[11] In their prospective study of breech deliveries, they found that 33 (26%) of 129 cases with severe neonatal complications had nonlethal major or minor malformations that sometimes explained the neonatal complications.[11] We cannot rule out that some undiagnosed or unrecorded malformations, may have contributed to the higher proportions of children with adverse outcomes among those born in breech than among those born in cephalic position in our study.

Comparison with other studies:

Our findings regarding excess risk for stillbirth[29] and NNM associated with breech presentation are consistent with earlier findings[30-32] and an excess risk for NNM was also reported in recent studies including children born after the TBT[2] in Denmark[4] and in Norway.[6]

We found a slightly higher risk for NNM in planned vaginal than in planned caesarean delivery and this could be considered to be consistent with the results of the TBT. On the other hand, the overall interpretation of our findings is that the risk for NNM was largely independent of mode of delivery, and this interpretation is not consistent with the results of the TBT. First, the different designs of the two studies may explain the different findings. The TBT was a randomized controlled trial considered to be the gold standard, while our study is

an observational study. Nonetheless, the much lower perinatal mortality in Norway compared with the TBT may also explain some of the diverging results in the two studies. Moreover, to be eligible to participate in the TBT, women had to have a singleton live fetus at term (≥ 37 week`s gestation) in breech without any known lethal fetal congenital anomaly. Women were excluded if there was evidence of fetopelvic disproportion, or if the fetus was judged to be clinically large or to have an estimated fetal weight of 4000 g or more, hyperextension of the fetal head or other fetal anomaly or condition that might cause a mechanical problem at delivery. Women with contraindication for labor or vaginal delivery such as placenta praevia were also excluded.[2] These criteria are similar to the criteria for vaginal breech delivery recommended by the Norwegian Society for Gynecology and Obstetrics. However, in the TBT a higher proportion of women (43%) selected for vaginal delivery needed caesarean delivery compared with our study population where only 30% of those selected for planned vaginal delivery needed caesarean delivery. One may therefore speculate that the probability for adverse outcome in the planned vaginal group in the TBT was higher than in our study. Instead antenatal acquired vulnerability may have played a larger role in our population.

The lack of excess risk for CP associated with breech position at birth is consistent with four studies published before the TBT, but inconsistent with four other studies. We are not aware of studies addressing the association between breech presentation at birth and CP in populations born after the TBT.[2] A follow-up study of 923 children included in that trial did not have the statistical power to address this severe, neurodevelopmental outcome.[33] Two earlier studies, including one from our own group, also found some evidence that the risk was associated with vaginal delivery.[12 15] The lower risk for CP in the present study, compared with our previous Norwegian study [12] could be explained by the larger sample size, better

quality of the data in the MBRN [21] and better ascertainment of cases in the CPRN in the present study.[23] Nonetheless, it is also possible that changes in the delivery of breech births in Norway including an increasing proportion of fetuses born by planned caesarean delivery,[6] may have improved outcome, and may reflect better selection of mothers for vaginal delivery.

Interpretation:

The overall higher risk for stillbirth and the higher proportion of infants born SGA among children born in breech than in cephalic position may suggest that fetuses with antenatal acquired risk factors for adverse outcomes are more likely to present in breech than in cephalic position at birth. On the other hand, the slightly higher ORs for NNM and for the composite outcome among children born vaginally than by caesarean delivery both when restricted to the breech group as well as when compared with vaginal cephalic delivery may suggest that fetuses in breech are more likely to experience complications during birth if they are born vaginally than if delivered by caesarean. This interpretation may be further supported by the fact that women selected for vaginal breech delivery would be expected to have a particular low risk for complications during birth. Thus, a combination of antenatal acquired risk factors for neonatal death with increased vulnerability to the birth process is probably the most likely explanation of our findings.

Regarding CP, antenatal factors are considered to be involved in 90% of the cases with CP,[34] and one might have expected an excess risk for CP in breech births, similar to that of NNM. The much lower risk for CP among children in breech, not statistically different from cephalic position could therefore suggest that antenatal factors increasing the risk for NNM are different or at least not completely overlapping with antenatal risk factors involved in the causal pathway leading to CP.

Implications:

Taking into consideration the very low absolute risk for NNM and CP, the increasing evidence for acute and long term maternal complications[35] and for later health problems among children following caesarean delivery,[36 37] our results suggest that vaginal delivery in selected cases may be an option for women with a fetus in breech position. This option requires that strict criteria are followed including access to competent obstetric care. In addition, a secondary advantage of having a certain volume of vaginal breech deliveries is that obstetricians retain their competence for unexpected vaginal breech deliveries. In the discussion with the pregnant mother and her partner regarding choice of delivery mode of a fetus in breech, the relative risk for NNM should be explained but related to the very low absolute risk. Moreover, it may be appropriate to emphasize that adverse outcome probably to a large degree is caused by antenatal acquired insults and that there are potential advantages of vaginal birth over caesarean delivery for long term health of the child and the mother. Regarding obstetric care, awareness of the excess risk for fetal death should be emphasized, and studies are warranted to optimize antenatal follow up of mothers with a fetus in breech.

Caution is needed if results of observational studies are included in the development of clinical guidelines, and more studies are needed to support our results. On the other hand, a new RCT in our part of the world is unrealistic as it would require the participation of 20 000 women with a fetus in breech in order to document a difference in NNM between mothers selected for planned vaginal and planned caesarean delivery.[10]

Nonetheless, the higher risk of neonatal mortality among planned vaginal deliveries than for planned caesarean delivery compared with cephalic delivery warrants further studies, including perinatal audits and prospective studies as suggested by Goffinet et al.[11]

CONCLUSION

Vaginal breech delivery, regardless of whether planned or actual and actual caesarean breech delivery were associated with excess risk for NNM and for a composite outcome of intrapartum death, NNM and CP, compared with vaginal cephalic delivery. The risk for adverse outcome in planned caesarean breech delivery did not differ significantly from planned vaginal cephalic delivery. However, the absolute risk for these outcomes was low. Taking into consideration potential long-term adverse consequences of caesarean delivery for the child, the mother and for later deliveries we therefore conclude that vaginal delivery may be offered to women with a fetus in breech, provided competent obstetric care and strict criteria for selection to vaginal delivery. Our findings did not support the notion that some cases of CP may be prevented if all fetuses in breech are delivered by caesarean delivery.

Competing interest: All authors have completed the ICMJE uniform disclosure form at www.icmj.org/cio_disclosure.pdf (available on request from the corresponding author) and declare: no financial support from any organization for the submitted work; no financial relationship with any companies that might have an interest in the submitted work in the previous three years; and have no non-financial interests or relationships that may be relevant to the submitted work.

Contributors: SB analyzed the data, contributed to study design and data interpretation and wrote the first draft of the manuscript. GA was principally responsible for the data from the CPRN and revised the manuscript. MM contributed to the data interpretation and revised the manuscript. PR contributed to the research hypotheses, study design and revised the manuscript. SH contribute to the data interpretation and revised the manuscript. DM contributed to study design, the data analyses, data interpretation and revised the manuscript. TV proposed the research questions, and contributed in the interpretation of the data and the revision of the manuscript. He is the guarantor of the study and accepts full responsibility for the work and the conduct of the study. All authors approved the final version of the submitted manuscript**.**

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Ethical approval: The study was approved by the Regional Ethical Committee for Medical Research in Mid-Norway (ref 2011/754).

Data sharing: The protocol is available on request from the corresponding author at: sbjellmo@hotmail.com

Transparency declaration: The lead author affirms that this manuscript is an honest, accurate, and transparent account of the study being reported; that no important aspects of the study have been omitted; and that any discrepancies from the study as planned have been explained.

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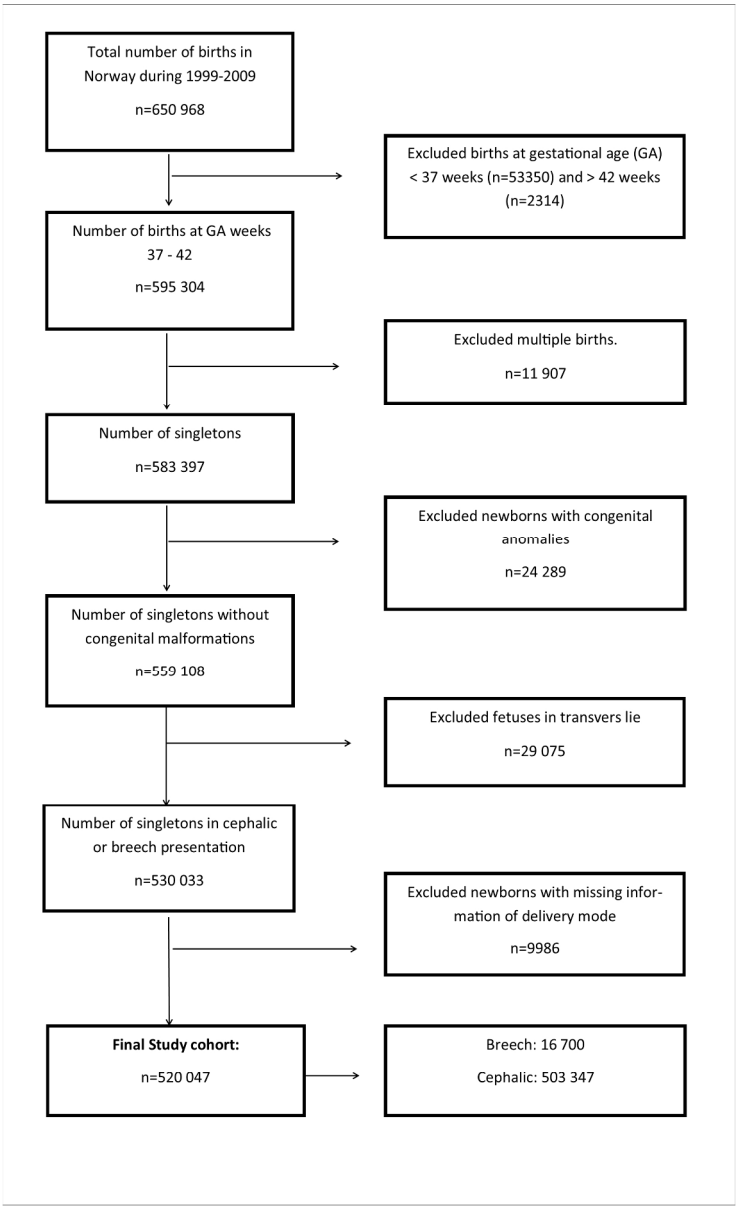

Flow chart of the study population.
Figure 1
159x260mm (300 x 300 DPI)

STROBE Statement—checklist of items that should be included in reports of observational studies

	Item No.	Recommendation	Page No.	Relevant text from manuscript
Title and abstract	1	(*a*) Indicate the study's design with a commonly used term in the title or the abstract	1	Yes
		(*b*) Provide in the abstract an informative and balanced summary of what was done and what was found	2-3	Yes
Introduction				
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	4-5	Introduction
Objectives	3	State specific objectives, including any prespecified hypotheses	5	The aim of this study was therefore to explore if singletons without congenital malformations born vaginally at term have higher risk for stillbirth, neonatal mortality (NNM) and CP if they are born in breech position compared to cephalic position.
Methods				
Study design	4	Present key elements of study design early in the paper	5	In this population based study, perinatal data from all children born in Norway during 1999-2009 were retrieved from the Medical Birth Registry of Norway (MBRN), and combined with information recorded in the Cerebral Palsy Register of Norway (CPRN).
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection	5	Data from the MBRN and CPRN (1999-2009)
Participants	6	(*a*) *Cohort study*—Give the eligibility criteria, and the sources and methods of selection of participants. Describe methods of follow-up *Case-control study*—Give the eligibility criteria, and the sources and methods of case ascertainment and control selection. Give the rationale for the choice of cases and controls *Cross-sectional study*—Give the eligibility criteria, and the sources and methods of selection of	5	Cohort-study with data from the MBRN and CPRN.

		participants		
		(*b*) *Cohort study*—For matched studies, give matching criteria and number of exposed and unexposed *Case-control study*—For matched studies, give matching criteria and the number of controls per case		
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	6	The predefined main outcome measures were stillbirth, NNM and CP. Stillbirth and NNM were defined according to the WHO
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group	*7-8*	*Analytic approach*
Bias	9	Describe any efforts to address potential sources of bias	7-8	Compared both actual and planned mode of delivery.
Study size	10	Explain how the study size was arrived at	6	We excluded children born preterm (before week 37), multiple births, children with congenital malformations, children in transverse lie and those with lacking information on mode of delivery (Figure 1).

Continued on next page

Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why		
Statistical methods	12	(*a*) Describe all statistical methods, including those used to control for confounding	8	IBM SPSS software for Windows version 22 was used for data analyses. Differences in proportions between groups were analyzed using the chi-square test and prevalence rates with 95% confidence intervals (CI) were calculated according to Newcombe and Altman. In the estimates of the prevalence of NNM, stillbirths were excluded and in the estimates of the prevalence of CP, stillbirths and children with post neonatal CP were excluded. We used logistic regression to estimate odd ratios (OR) with 95% confidence intervals (CI) for adverse outcome of children in breech position at birth, using cephalic presentation as the reference. Moreover we explored the roles of potential confounders including maternal age, parity, gestational age, child sex and SGA status in multivariable logistic regression analyses based on *a priori* knowledge and directed acyclic graphs methodology.
		(*b*) Describe any methods used to examine subgroups and interactions		
		(*c*) Explain how missing data were addressed	Table 1 (10)	Information on missing.
		(*d*) *Cohort study*—If applicable, explain how loss to follow-up was addressed *Case-control study*—If applicable, explain how matching of cases and controls was addressed *Cross-sectional study*—If applicable, describe analytical methods taking account of sampling strategy		
		(*e*) Describe any sensitivity analyses		

Results				
Participants	13*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed	9	A total of 650 968 children were born in Norway during the study period. The study population of singleton children born at a gestational age of at least 37 weeks in either cephalic or breech position, and with no congenital malformation comprised 520 047 children.
		(b) Give reasons for non-participation at each stage		
		(c) Consider use of a flow diagram	Figure 1	Yes
Descriptive data	14*	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders	9	Among the 520 047 included children, 16 700 (3%) were in breech and 503 347 (97%) in cephalic position (Table 1). More mothers in the breech group were nullipara, and a higher proportion of their infants were females, were born SGA and had low Apgar scores (Table 1). The mean gestational age of children born in breech was 39.1 weeks compared with 39.7 weeks for children born in cephalic.
		(b) Indicate number of participants with missing data for each variable of interest		
		(c) *Cohort study*—Summarise follow-up time (eg, average and total amount)		
Outcome data	15*	*Cohort study*—Report numbers of outcome events or summary measures over time	*10-14*	*Among all, actual delivery and planned delivery.*
		Case-control study—Report numbers in each exposure category, or summary measures of exposure		
		Cross-sectional study—Report numbers of outcome events or summary measures		

Main results	16	(*a*) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included	10-14 Table 2-5	See tables and text.
		(*b*) Report category boundaries when continuous variables were categorized		
		(*c*) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period		

Continued on next page

Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses	14	Multivariable analyses adjusting for gestational age, parity, maternal age, sex and SGA did not substantially affect any of the associations described above (data not shown).
Discussion				
Key results	18	Summarise key results with reference to study objectives	14	In this national cohort study of term singletons without congenital malformations we found that breech vaginal delivery, regardless of whether it was planned or not, was associated with an excess risk for NNM and with a composite outcome of intrapartum death, NNM and CP, compared with cephalic vaginal delivery.
Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias	15-17	Strength and limitations
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence	17-20	Comparison with other studies and Interpretation
Generalisability	21	Discuss the generalisability (external validity) of the study results	20	Taking into consideration the very low absolute risk for NNM and CP, the increasing evidence for acute and long term maternal complications[35] and for later health problems among children following caesarean delivery,[36 37] our results suggest that vaginal delivery in selected cases may be an option for women with a fetus in breech position. This option requires that strict criteria are followed including access to competent obstetric care. In addition, a secondary advantage of having a certain volume of vaginal breech deliveries is that obstetricians retain their competence for unexpected vaginal

				breech deliveries. In the discussion with the pregnant mother and her partner regarding choice of delivery mode of a fetus in breech, the relative risk for NNM should be explained but related to the very low absolute risk. Moreover, it may be appropriate to emphasize that adverse outcome probably to a large degree is caused by antenatal acquired insults and that there are potential advantages of vaginal birth over caesarean delivery for long term health of the child and the mother. Regarding obstetric care, awareness of the excess risk for fetal death should be emphasized, and studies are warranted to optimize antenatal follow up of mothers with a fetus in breech. Caution is needed if results of observational studies are included in the development of clinical guidelines, and more studies are needed to support our results.
Other information				
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	22	Supported by a grant from The Liaison Committee between the Central Norway Regional Health Authority (RHA) and the Norwegian University of Science and Technology (NTNU).

*Give information separately for cases and controls in case-control studies and, if applicable, for exposed and unexposed groups in cohort and cross-sectional studies.

Note: An Explanation and Elaboration article discusses each checklist item and gives methodological background and published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely available on the Web sites of PLoS Medicine at http://www.plosmedicine.org/, Annals of Internal Medicine at http://www.annals.org/, and Epidemiology at http://www.epidem.com/). Information on the STROBE Initiative is available at www.strobe-statement.org.