Is foot deformity associated with developmental dysplasia of the hip?
RESULTS AFTER EXAMINATION OF 60,844 NEWBORNS

Aims
To assess if congenital foot deformity is a risk factor for developmental dysplasia of the hip (DDH).

Methods
Between 1996 and 2012, 60,844 children were born in Sør-Trøndelag county in Norway. In this cohort study, children with risk factors for DDH were examined using ultrasound. The risk factors evaluated were clinical hip instability, breech delivery, a family history of DDH, a foot deformity, and some syndromes. As the aim of the study was to examine the risk for DDH and foot deformity in the general population, children with syndromes were excluded. The information has been prospectively registered and retrospectively analyzed.

Results
Overall, 494 children (0.8%) had DDH, and 1,132 (1.9%) a foot deformity. Of the children with a foot deformity, 49 (4.3%) also demonstrated DDH. There was a statistically significant increased association between DDH and foot deformity ($p < 0.001$). The risk of DDH was highest for talipes calcaneovalgus (6.1%) and club foot (3.5%), whereas metatarsus adductus (1.5%) had a marginal increased risk of DDH.

Conclusion
Compared with the general population, children with a congenital foot deformity had a significantly increased risk for DDH and therefore we regard foot deformity as a true risk factor for DDH.

Introduction
Developmental dysplasia of the hip (DDH) is a well-described condition in newborns. Different screening programmes have been introduced to detect hips in need of treatment, and to minimize the number of children with late-diagnosed DDH. In 2019, Broadhurst et al. reported that in England there had been no reduction in the incidence of children with late-diagnosed DDH over the last 35 years. This therefore suggests that there is a need to improve the screening of children with risk factors for DDH.

Ultrasound is a method frequently used in diagnosing DDH, either through universal or selective screening among children at risk of DDH. However, there is no consensus concerning risk factors for DDH. Clinical hip instability, a family history of DDH, or breech deliveries have been reported to be the most important risk factors for DDH. However, foot deformity has not generally been identified as a true risk factor for DDH.

Paton et al. reported a slight increased risk of DDH among children with foot deformities compared to the general population in Blackburn, UK. In later studies the same group found that there was no increased risk of DDH for clubfoot or any other foot deformity, concluding that foot deformities should not be considered a risk factor for DDH. Furthermore de Hundt et al., in a meta-analysis, recorded no significant relationship between foot deformities and DDH. In another meta-analysis, Ibrahim et al. found a comparable pooled prevalence of DDH among those with congenital talipes equinovarus (CTEV) and the general population. Therefore, they too did not recommend ultrasound screening of newborns with CTEV.

Others have reported an increased risk of DDH in children with CTEV, concluding that first-born
females, CTEV, and breech position were all associated with an increased risk of DDH. These studies reported that the relative risk of DDH in children with CTEV was 10.34, which is markedly increased. Zhao et al\textsuperscript{17} also compared CTEV and DDH and recorded that out of 184 patients with CTEV, five demonstrated DDH (2.7\%). Based on these findings of an increased incidence of DDH among children with CTEV compared with the general population, they recommended ultrasound examination of the hip in this group with foot deformities.

In previous reports from our hospital we have been unable to conclude whether a foot deformity should be considered a risk factor for DDH.\textsuperscript{3,4} In this current study we have increased the number of children in the survey in order to further examine the relationship between foot deformity and DDH.

**Methods**

Between 1996 and 2012, 60,844 children were born in Sør-Trøndelag county in Norway. All the children underwent clinical hip screening by a paediatrician on the first day after birth. A total of 27 children with syndromes, who were examined using ultrasound, were excluded from the study; the syndromes included Downs, Kippel-Feil, Merkel-Gruber, Di George, and arthrogryposis. The remaining 60,817 children were all included in the review. The clinical examination by the paediatrician involved both the Ortholani\textsuperscript{18} and Barlow\textsuperscript{19} tests, the degree of abduction of the mother’s child number, gestation week, the paediatrician’s clinical findings, and the ultrasound results.

Because it has been shown in a randomized trial that there was no significant increased risk for DDH when comparing a group undergoing selective screening with a group undergoing universal screening,\textsuperscript{20}we have elected to perform selective ultrasound hip screening. Based on the clinical findings and risk factors for DDH, 8,709 (14\%) children were examined using ultrasound of the hips.

The reasons for ultrasound examination were as follows: pathological clinical findings from the paediatrician’s examination, a family history of DDH, breech presentation including breech position and caesarean section, and foot deformity.

The ultrasound examination was performed by either one of three orthopaedic surgeons and one paediatrician, all who had a special interest in hip disorders in children. We used a Siemens Antares (Siemens AG, Munich, Germany) with an 8 mHz to 16 mHz linear probe in most of the examinations, and since 2007 we have used a GE Logic 7 (GE Healthcare, Milwaukee, Wisconsin, USA) with the same type of probe. The ultrasound transducer was placed on the lateral side of the hip, parallel to the long axis of the body. The femoral head coverage was calculated as a percentage expression of the acetabular coverage of the femoral head, where femoral head coverage above 50\% is considered to be normal.\textsuperscript{4} A dynamic examination was performed to evaluate the stability of the hip. If uncertain ultrasound findings were obtained after the first examination, treatment for DDH was not started, but the child was re-examined two to three weeks later. If treatment for DDH was started after the first or second examination, children were then registered as in need for treatment for DDH. In addition to risk factors, we also registered data such as birth weight and body length, the mother’s child number, gestation week, the paediatrician’s clinical findings, and the ultrasound results.

Foot deformities were classified into four categories: idiopathic CTEV; congenital metatarsus adductus (MA); congenital talipes calcaneovalgus (CTCV); and a non-specific foot deformity, which were soft and flexible feet that did not fit into one of the other three categories.

Since registration started in 1996, all data were recorded on paper sheets, and later results of all ultrasound examinations were digitized into a local quality registry for children with DDH. The results from this registry were compared with data from the birth records of our hospital to ensure that all foot deformities were registered. In total, 230 children (2.6\%) examined by ultrasound had missing information regarding foot deformity and 54 children (0.6\%) regarding DDH.

**Ethical approval.** The data has been prospectively registered. This cohort study was approved by the Regional Committee for Medical Research Ethics in Western Norway (project number 2013/1508).

**Table I.** The observed and expected count of children with and without developmental dysplasia of the hip (DDH), and with and without foot deformity.

| Foot deformity, n | DDH | No | Yes |
|------------------|-----|----|-----|
|                  |     |    |     |
| Overall          | 60,817 | 494 | 0.8 |
| Family history   | 4,162 | 131 | 3.1 |
| Breech position  | 2,548 | 120 | 4.7 |
| Foot deformity   | 1132 | 49  | 4.3 |
| CTEV             | 170  | 6   | 3.5 |
| MA               | 136  | 2   | 1.5 |
| CTCV             | 642  | 39  | 6.1 |
| Uncategorized foot deformities | 184 | 2 | 1.1 |

| Factor                         | Total, n | Children with DDH, n | Risk for DDH, % |
|-------------------------------|----------|----------------------|-----------------|
| Overall                       | 60,817   | 494                  | 0.8             |
| Family history                | 4,162    | 131                  | 3.1             |
| Breech position               | 2,548    | 120                  | 4.7             |
| Foot deformity                | 1132     | 49                   | 4.3             |
| CTEV                          | 170      | 6                    | 3.5             |
| MA                            | 136      | 2                    | 1.5             |
| CTCV                          | 642      | 39                   | 6.1             |
| Uncategorized foot deformities| 184      | 2                    | 1.1             |

CTCV, congenital talipes calcaneovalgus; CTEV, congenital talipes equinovalgus; MA, metatarsus adductus.
Table III. Results of weight, length, and gestational age of children with and without developmental dysplasia of the hip (DDH) foot deformity. The total represents the selection of 8,709 children examined due to risk factors for DDH.

| Variable                  | Total               | Yes | No   | Yes | No   |
|---------------------------|---------------------|-----|------|-----|------|
| Mean weight, g (SD; 95% CI)| 3,430 (638; 3,420 to 3,440) | 3,666 (600; 3,634 to 3,687) | 3,426 | 3,457 | 3,440 |
| Mean body length, cm (SD; 95% CI) | 49.8 (2.7; 49.7 to 49.9) | 50.4 (2.1; 50.3 to 50.5) | 49.7 (2.7; 49.7 to 49.8) | 49.6 (2.3; 49.5 to 49.7) | 49.8 (2.8; 49.7 to 49.8) |
| Mean gestational age, wks (SD; 95% CI) | 39.1 (3.1; 39.1 to 39.2) | 40 (1.1; 40.0 to 40.0) | 39.1 (3.2; 39.0 to 39.1) | 39.6 (6.7; 39.3 to 39.9) | 39.1 (2.1; 39.0 to 39.1) |

CI, confidence interval.

Table IV. Results of weight, body length, and gestational age of children with subgroups of foot deformity, and with and without developmental dysplasia of the hip (DDH).

| Variable                  | CTEV | CTEV and DDH | MA | MA and DDH | CTCV | CTCV and DDH |
|---------------------------|------|--------------|----|------------|------|--------------|
| Mean weight, g (SD; 95% CI) | 3,318 (635; 1,548 to 5,020) | 3,566 (642; 2,630 to 4,440) | 3,448 (526; 2,160 to 4,995) | 3,223 (N/A; 3,215 to 3,230) | 3,429 (500; 2,085 to 5,050) | 3,651 (533; 2,430 to 4,900) |
| Mean body length, cm (SD; 95% CI) | 48.9 (2.7; 42.0 to 56.0) | 49 (2.7; 44.0 to 51.0) | 49.5 (2.1; 43.0 to 55.0) | 48.5 (6.6; 48.0 to 49.0) | 49.6 (2.3; 32.0 to 56.0) | 50.3 (1.9; 46.0 to 53.0) |
| Mean gestational age, wks (SD; 95% CI) | 40 (2.1; 38.0 to 42.1) | 40 (1.2; 39.2 to 40.8) | 39.3 (1.4; 39.1 to 39.5) | 39.5 (0.6; 38.6 to 40.4) | 39.6 (1.3; 39.5 to 39.7) | 39.9 (1.1; 39.7 to 40.2) |

*Only two cases in this group.

CI, confidence interval; CTC, congenital talipes calcaneovalgus; CTEV, congenital talipes equinovarus; MA, metatarsus adductus; NA, not applicable.

for Medical and Health Research Ethics on 6 December 2016 (2016/1151).

Statistical analysis. Statistical calculations were performed using SPSS v. 23 (IBM, Armonk, New York, USA). Results regarding weight, height, and gestational age are presented as mean with 95% confidence intervals (CIs) and SD. The chi-squared test was used to assess the association between DDH and foot deformity.

Results

Between 1996 and 2012, 60,844 children were born in Sør-Trøndelag county, and after exclusion of those with severe syndromes as has been listed, 8,709 children (14.3%) were examined using ultrasound because of risk factors of DDH and these comprise the main study group (Figure 1).

The risk factors in this group leading to ultrasound examination were: a family history of DDH, 4,162 (48%); breech position, 2,548 (29%); abnormal clinical findings, 1,617 (19%); and foot deformity, 1,132 (13%).

The results regarding children with and without DDH in terms of foot deformity are presented in Table I. A total of 494 children had DDH and 1,132 children had a foot deformity. There was a statistically significant association between DDH and foot deformity (p < 0.001) with an increased number of foot deformity amongst those with DDH.

Among 1,619 children with abnormal clinical findings, 399 (24.6%) had DDH. The results concerning other risk factors and DDH are presented in Table II.

In total, 494 children were treated for DDH, giving an overall incidence of DDH of 8.1/1,000. Of the 1,132 newborns with a foot deformity, 184 newborns had a non-categorized foot deformity. The remaining 948 newborns were categorized into three groups: CTEV, MA, and CTCV.

In the foot deformity group, six out of 170 newborns with CTEV had DDH (3.5%), two out of 136 with MA had DDH (1.5%), and 39 of the 642 with CTCV had DDH (6.1%). In the non-specified foot deformity group, two of 184 newborns (1.1%) had DDH. Most of the children had a single foot deformity, but 11 children had two different foot deformities (CTEV, MA, or CTCV). There were no additional risk factors for DDH among newborns with CTEV, MA, or in the uncategorized group. Among those with CTCV, six of the 39 treated for DDH had additional risk factors, five with a positive family history, and one with breech delivery.

Among the 8,709 children who were examined with ultrasound, further data was recorded including weight, body length, and gestation age (Table III).

The mean birth weight and body length were 3,430 g (SD 637.8) and 49.8 cm (SD 2.7), respectively, which are close to the mean values for all newborns in our country (3,475 g and 50 cm). Children with DDH were heavier, longer, and of higher gestational age than those without DDH, although these differences were small. Children with foot deformity were heavier, shorter, and older than children without a foot deformity, but again the differences between the means were small. Children with foot deformity and DDH (except for those with CTEV) had a higher mean birth weight, a higher mean body length, and a higher gestational age compared with those without DDH (Table IV). Children with CTEV or CTCV and simultaneous DDH had a higher mean weight, body length, and age. Children with MA had a lower mean weight and body length than those with MA and simultaneous DDH.

Discussion

The aim of our study was to investigate whether foot deformity was associated with DDH and we report a significant positive association between the two. CTEV and CTCV represented the majority of children who had an increased risk of DDH, whereas MA resulted in only a slightly increased risk of DDH compared with the general population.
We found an overall mean birth weight of 3,430 g, a body length of 49.8 cm, and the overall mean gestational age was 39.1 weeks (SD 3.1). Compared with all children examined using ultrasound, children with a foot deformity (except CTEV) and DDH had a small increase in weight and gestational age with a small decrease in body length, although these differences are most likely to be without clinical significance.

Paton et al.15 in a prospective longitudinal observational study, found no children with DDH among newborns with CTEV, and concluded that CTEV should not be considered as a significant or “true” risk factor. This is in contrast with our findings where there was an increased risk of DDH both in the CTEV group and in the CTCV group, where six out of 170 children (3.5%) with CTEV had DDH and 39 out of 642 children (6.1%) with CTCV had DDH. For MA, there were 136 children (1.5%), and for uncategorized foot deformities, two out of 184 children (1.1%) had DDH, demonstrating a marginal increase in risk of DDH compared with the overall risk of DDH of 0.8%. Our findings are in accordance with those of Perry et al.,15 who recorded that almost 6% of children with DDH had CTEV. Our results are also similar to those reported by Zhao et al.,17 who found that five children out of 184 with DDH had CTEV; they concluded that CTEV was a risk factor for DDH, and recommended ultrasound examination of the hips in these children.

Paton et al.15 examined the relationship between different types of foot deformity and DDH as defined by ultrasound. In their study they classified foot deformities as postural talipes equinovarus (postural TEV, a CTEV-like position but soft and clearly flexible), CTEV, CTCV, and MA. They found that the overall risk of ultrasound-defined dysplasia or instability was 1.27 in postural TEV, 1.86 in CTEV, 1.52 in CTCV, and 1.25 in MA. However, when they examined only the Graf21 type IV instability (irreducible dislocation), the overall risk was 1.436 in postural TEV, 1.154 in CTCV, 1.25 in MA, and none in CTEV. These authors concluded that routine screening for postural TEV and CTEV was not necessary, but that newborns with CTCV and possibly also with MA should be offered an ultrasound of the hip. These results however are in contrast with ours, where we found a significantly increased risk of CTEV and a minor increase in risk in the MA group (p < 0.001).

Perry et al.15 examined 119 newborns with CTEV with hip ultrasound. They found that a total of nine hips in seven newborns were classified as Graf type IIB or worse and were in need of treatment. The treatment frequency was 5.9% (1:17) in CTEV newborns, which was 45 times more than in the general population (1.3/1,000). They concluded that CTEV was an important risk factor for DDH and should be included in a selective ultrasound hip-screening programme.

In our study population, however, children with CTCV had the highest risk of DDH (6.1%), and even discounting the six children with additional risk factors, there was still a markedly increased risk of DDH among those with CTCV. In those children with CTEV, our results show that the risk of DDH was 3.5%, which is a little below the increased risk of 5.9% found by Perry et al.15

The overall incidence for DDH at our hospital was 0.8%, for MA the incidence was 1.5%, and for the uncategorized foot deformities the incidence was 1.1%, raising the question whether MA and uncategorized foot deformities do in fact require any special attention, since the risk of DDH was minimal. However, we have reached the conclusion to continue to offer ultrasound examination for all types of foot deformities.

Among the syndromes associated with DDH or foot deformity, Szöke et al.12 found that 26 out of 95 patients with amyoplasia (the most common form of arthrogryposis) had hip dislocations. In our study, 36 children with arthrogryposis were detected during pregnancy, and 32 women decided to end their pregnancy, leaving four children who were born and were excluded from the study.

There are limitations to our study including missing data on weight, body length, and gestational age of children not examined by ultrasound, and this has prevented more detailed statistical modelling. Also, the number of children with combined DDH and foot deformity was too low for further statistical calculations. However, the strength of the study is the large number of children involved. Also that the examination and registration were performed by three orthopaedic surgeons and one paediatrician, all with experience and a special interest in hip disorders in children. On the other hand hardly any of the overall data were missing, and pathological findings were double checked in the patients records.

In conclusion, in this study we have found that foot deformity is a significant risk factor for DDH, of which CTEV and CTCV were the foot deformities with the largest increased risk of DDH. On the basis of these findings we will continue to regard congenital foot deformity as true risk factors for DDH.

Take home message
- Children with a foot deformity has a significant increased risk for developmental dysplasia of the hip.
- Based on this study we recommend ultrasonographic examination of the hip in newborns with foot deformity.

Twitter
Follow Ø. Håberg @oyvindhhaberg

References
1. Broadhurst CR, Rhodes AML, Harper P, et al. What is the incidence of late detection of developmental dysplasia of the hip in England? Bone Joint J. 2019;101-B(3):281–287.
2. Bache CE, Clegg J, Herron M. Risk factors for developmental dysplasia of the hip: ultrasonographic findings in the neonatal period. J Pediatr Orthop B. 2002;11(3):212–219.
3. Cyvin KB. Congenital dislocation of the hip joint. Acta paediatrica Scandinavica Supplement. 1977;263:1–67.
4. Helen KJ, Terjesen T, Tegnander A, et al. Ultrasound screening for hip dysplasia in newborns. J Pediatr Orthop. 1994;14(5):687–673.
5. Jones DA. Importance of the clicking hip in screening for congenital dislocation of the hip. Lancet. 1989;1(8738):599–601.
6. Ortiz-Neira CL, Paolucci EO, Donnon T. A meta-analysis of common risk factors associated with the diagnosis of developmental dysplasia of the hip in newborns. Eur J Radiol. 2012;81(3):e344–e351.
7. Shorter D, Hong T, Osborn DA. Cochrane review: screening programmes for developmental dysplasia of the hip in newborn infants. Evid Based Child Health. 2013;8(1):11–54.
8. Paton RW. Screening in developmental dysplasia of the hip (DDH). Surgeon. 2017;15(6):290–296.
9. Paton RW, Choudry QA, Jugdey R, Hughes S. Is congenital talipes equinovarus a risk factor for pathological dysplasia of the hip? A 21-year prospective, longitudinal observational study. Bone Joint J. 2014;96-B(11):1553–1555.
10. Talbot CL, Paton RW. Screening of selected risk factors in developmental dysplasia of the hip: an observational study. Arch Dis Child. 2013;98(9):692–696.
11. Paton RW, Hinduja K, Thomas CD. The significance of at-risk factors in ultrasound surveillance of developmental dysplasia of the hip. A ten-year prospective study. J Bone Joint Surg Br. 2005;87(9):1264–1266.

12. Paton RW, Choudry Q. Neonatal foot deformities and their relationship to developmental dysplasia of the hip: an 11-year prospective, longitudinal observational study. J Bone Joint Surg Br. 2009;91(5):655–658.

13. de Houdt M, Vlemmix F, Bais JM, et al. Risk factors for developmental dysplasia of the hip: a meta-analysis. Eur J Obstet Gynecol Reprod Biol. 2012;165(1):8–17.

14. Ibrahim T, Riaz M, Hegazy A. The prevalence of developmental dysplasia of the hip in idiopathic clubfoot: a systematic review and meta-analysis. Int Orthop. 2015;39(7):1371–1378.

15. Perry DC, Tawfiq SM, Roche A, et al. The association between clubfoot and developmental dysplasia of the hip. J Bone Joint Surg Br. 2010;92(11):1586–1588.

16. Pollet V, Percy V, Prior HJ. Relative risk and incidence for developmental dysplasia of the hip. J Pediatr. 2017;181:202–207.

17. Zhao D, Rao W, Zhao L, et al. Is it worthwhile to screen the hip in infants born with clubfeet? Int Orthop. 2013;37(12):2415–2420.

18. Ortolani M. Congenital hip dysplasia in the light of early and very early diagnosis. Clin Orthop Relat Res. 1976(119):6–10.

19. Barlow TG. Treatment of congenital dislocation of the hip in the newborn. Proc R Soc Med. 1963;56(9):804–806.

20. Holen KJ, Tegnander A, Bredland T, et al. Universal or selective screening of the neonatal hip using ultrasound? A prospective, randomised trial of 15,529 newborn infants. J Bone Joint Surg Br. 2002;84(6):886–890.

21. Graf R. Fundamentals of sonographic diagnosis of infant hip dysplasia. J Pediatr Orthop. 1984;4(6):735–740.

22. Szöke G, Staheli LT, Jaffe K, Hall JG. Medial-approach open reduction of hip dislocation in amnioplasia-type arthrogryposis. J Pediatr Orthop. 1996;16(1):127–130.

Author information:
Ø. Håberg, MD, Orthopedic Surgeon, PhD Candidate
Ø. B. Lian, MD, PhD, Orthopedic Surgeon, Associate Professor

Department of Orthopaedic Surgery, Kristiansund Hospital, Kristiansund, Norway; Department of Neuromedicine and Movement Science, Norwegian University of Science and Technology, Trondheim, Norway.

O. A. Foss, MD, PhD, Orthopedic Surgeon, Professor
K. J. Holen, MD, PhD, Orthopedic Surgeon, Associate Professor

Department of Neuromedicine and Movement Science, Norwegian University of Science and Technology, Trondheim, Norway; Department of Orthopaedic Surgery, Trondheim University Hospital, Trondheim, Norway.

Author contributions:
Ø. Håberg: Designed the study, Wrote the manuscript.
O.A. Foss: Supervised the study, Performed the statistical analysis, Wrote the manuscript.
Ø.B. Lian: Supervised the study, Wrote the manuscript.
K.J. Holen: Designed the study, Wrote the manuscript.

Funding statement:
No benefits in any form have been received or will be received from a commercial party related directly or indirectly to the subject of this article.

Acknowledgements:
Special thanks to Dr. Helge Rønningen and Dr. Torarin Lamvik, who performed some of the ultrasound examinations. A very special thanks to Dr. Ole Jacob Johansen, the pediatrician who performed most of the clinical examinations and also some ultrasound examinations, and to Myrtle Slettvåg Hoel for her plotting work in establishing our Local Registry for Children with DDH.

Ethical review statement:
This cohort study was approved by the Regional Committee for Medical and Health Research Ethics on 6 December 2016 (2016/1151).

Open access statement:
This is an open-access article distributed under the terms of the Creative Commons Attribution Non-Commercial No Derivatives (CC BY-NC-ND 4.0) licence, which permits the copying and redistribution of the work only, and provided the original author and source are credited. See https://creativecommons.org/licenses/by-nc-nd/4.0/

This article was primary edited by S. P. F. Hughes.