

Neonatal hip instability and risk of total hip replacement in young adulthood

Follow-up of 2,218,596 newborns from the Medical Birth Registry of Norway in the Norwegian Arthroplasty Register

Ingvild Ø Engesæter¹, Stein Atle Lie^{1,2}, Trude G Lehmann^{1,3}, Ove Furnes^{1,3},
Stein Emil Vollset⁴, and Lars B Engesæter^{1,3}

¹The Norwegian Arthroplasty Register, Department of Orthopaedic Surgery, Haukeland University Hospital, ²Department of Health, University Research Bergen, Unifob, Bergen, ³Department of Surgical Science, University of Bergen, ⁴Department of Public Health and Primary Health Care, University of Bergen, Bergen, Norway
Correspondence IØE: ingvild.engesater@student.uib.no
Submitted 07-08-10. Accepted 08-01-31

Background and purpose Dysplasia is probably the most common underlying condition in osteoarthritis of the hip, leading to total hip replacement (THR) in young adulthood. We investigated whether hip instability at birth predisposes to THR in young adulthood.

Methods Since 1967, all newborns in Norway have been screened for neonatal hip instability (NHI) and the results have been reported to the Medical Birth Registry of Norway (MBRN). In the period 1967–2004, 2,218,596 newborns were registered. From 1987 to 2004, 442 of these individuals had been reported to the Norwegian Arthroplasty Register (NAR) after undergoing total hip replacement (mean age 25 (12–36) years).

Results Neonatal hip instability was reported in 19,432 newborns (0.88%) in the MBRN; according to the NAR, they had a 2.6 (CI 1.4–4.8) times increased risk of THR in young adulthood compared to those without NHI. The absolute risk was low, however; only 57 (95% CI: 30–105) in 10⁵ for patients with NHI compared to 20 (95% CI: 18–22) in 10⁵ for those without registered hip pathology. Of the 442 patients with THR, 95 were operated because of osteoarthritis of the hip secondary to dysplasia, according to the surgeon's report. However, only 8 of these 95 patients had been reported to have hip instability at birth.

Interpretation Neonatal hip instability increases the risk of THR in young adulthood. Unexpectedly, only 8% of those who underwent THR due to dysplasia were

reported to have unstable hips at birth. Our results indicate that clinical testing for NHI is insufficient as a screening method for dysplastic hips that require THR in young adulthood. ■

Neonatal hip instability (NHI) occurs in about 1 per 100 live births (Rosendahl et al. 1996, Holen et al. 1999, Reikerås et al. 1999, Lehmann et al. 2000). The instability is believed to be due to acetabular dysplasia or laxity of the joint capsule. It is essential to diagnose the instability at birth, because the treatment is then easier and the prognosis is better (Weinstein et al. 2003).

Medical information about all newborns in Norway is recorded in the Medical Birth Registry of Norway (MBRN) (Irgens 2000). Data concerning both the birth and the child are recorded, including instability of the hips. All newborns in Norway are screened for instability of the hips at birth by clinical examination. From the beginning of the 1990s, those with instability have also been examined by ultrasonography. Unstable hips are usually treated with the Frejka pillow (Tegnander et al. 2001). This treatment is relatively simple and harmless; thus, far more children are treated than necessary (Weinstein et al. 2003). The general

understanding is that the clinical screening program will reveal most of the unstable hips at birth (Sahin et al. 2004).

Dysplasia of the hip has been assumed to be the most common condition underlying coxarthrosis in young adulthood (Harris 1986, Albinana et al. 2004, Jacobsen et al. 2005). The current treatment for severe coxarthrosis is total hip replacement (THR). THRs performed in Norway are reported to the Norwegian Arthroplasty Register (NAR) (Havelin et al. 2000).

By combining data from these two national registries, we investigated whether instability of the hips at birth (NHI) might predispose to coxarthrosis requiring THR in young adulthood.

Patients and methods

Our study is based on medical information from two national registers, the Medical Birth Registry of Norway and the Norwegian Arthroplasty Register.

Since January 1, 1967, the Medical Birth Registry of Norway (MBRN) has registered medical information on all newborns (Irgens 2000). Reporting to this register is compulsory, using a standardized form. Information about the child, the birth, and any congenital malformations, including instability of the hip, is included. Two standard forms have been used since the register was established: the first one was used until December 1, 1998, and the second one from that time to the present.

The various diagnoses on the standardized MBRN form are recorded according to the International Classification of Diseases (ICD). Until 1998, the MBRN used ICD 8, and from that time ICD 10 was used. Under the term neonatal hip instability (NHI), for 1967–1998 we included the ICD 8 codings “luxation of the hip” (755.6), “dysplasia of the hip” (755.7), and “positive Ortolani test” (778.5). For the ICD 10 coding, we included the entry for “dysplasia of the hip treated with Frejka pillow” and any specifications of neonatal diagnoses in the category congenital abnormalities of the hip (Q65.0–Q65.9). It is important to note that for patients diagnosed with unstable hips, the form does not record whether the left hip, the right hip, or both hips are affected. In addition, the proportion of newborns with hip instability reported to the MBRN is not

known, but a study on another congenital malformation (oral clefts) indicated that 94% of all cleft lip and palate (CLP) were reported, but the percentage varied with the severity of the malformation (Kubon et al. 2007). Further on, an instruction book for the registration form used from 1967–1998 stated that unstable hips (positive Ortolani test) was a congenital malformation to be recorded on the form (HelseDirektøren 1966), which also may have contributed to an accurate reporting.

Regarding unstable hips, the American Academy of Orthopaedic Surgeons (AAOS) and the Pediatric Orthopaedic Society of North America (POSNA) have recommended the use of the term developmental dysplasia or dislocation of the hip (DDH). DDH describes all abnormalities in the condition hip instability, including both congenital instability and developmental instability, and dysplasia. We have used the term neonatal hip instability (NHI) because we only had information on the clinical stability of the hip at birth and not on any abnormalities that developed during childhood. Otherwise, our term includes the same conditions as defined for DDH.

The Norwegian Arthroplasty Register (NAR) has been registering all THRs performed in Norway since September 15, 1987 (Havelin et al. 2000). A hip prosthesis operation is reported to the register by the surgeon, who completes a standard form. The form includes the identity of the patient, the date of the operation, and the reason for surgery as given by the surgeon (Havelin et al. 2000). Both primary operations and reoperations are recorded. Reporting to the NAR is not compulsory, but it has been estimated that at least 97% of joint replacements performed in Norway are reported (Espeshaug et al. 2006). From the start of the register in September 1987 until the end of December 2004, 106,716 THRs had been reported (Furnes et al. 2005). Of these, 633 THRs were performed on 442 patients born after January 1, 1967; thus, they were also registered in the MBRN.

All Norwegian residents have a unique national identity number. With this number we were able to combine information from the birth register and the arthroplasty register. Patients from NAR were included until the end of December 2004; the maximum age for those with THR would thus be 38 years of age (defined as young age).

Table 1. Number of cases with total hip replacement (THR) and neonatal hip instability (NHI), and relative risk

	THR		Relative risk (95% CI)
	No	Yes	
NHI			
No	2,198,733	431	1 [reference]
Yes	19,421	11	2.6 (1.4–4.8)
Sex			
Male	1,139,835	192	1 [reference]
Female	1,075,914	250	1.4 (1.1–1.6)
Year of birth			
After 1975	1,636,904	102	1 [reference]
In 1975 or earlier	581,250	340	0.87 (0.67–1.1)

Approval for the study was obtained from the Regional Committee for Medical and Health Research Ethics and the Data Inspectorate.

Statistics

The probability of undergoing total hip replacement was calculated using the Kaplan-Meier method, with age at the first registered prosthesis operation as endpoint. For all individuals without any prosthesis, age at the end of the study (December 31, 2004) was considered to be a censored observation. We used Cox regression analysis to calculate risk estimates for undergoing total hip replacement at a young age (adjusted for sex, NHI, and time period). The analyses were performed using the statistical program SPSS version 14.0, and also S-Plus 6.2 (Insightful Corp, Seattle, WA, USA).

Results

Epidemiology

2,218,596 newborns were registered in the MBRN between 1967 and 2004. Of these, 19,432 (0.88%) were reported to have unstable hips at birth (Table 1). The average number of reported NHIs per year in Norway is 511 (156–728) (SD 154). Mean incidence is 0.88% (0.25–1.33). The incidence was somewhat lower after 1997, which could be due partly to the introduction of a new registration form in December 1998, with new ICD codings. The overall incidence of NHI was 5 in 1,000 for males and 13 in 1,000 for females.

Table 2. Reason(s) for total hip replacement recorded in the Norwegian Arthroplasty Register (NAR) for patients < 38 years of age who were registered in both the NAR and the Medical Birth Registry of Norway

Diagnoses ^a	Individuals	Percentage (%)
Idiopathic coxarthrosis	16	3.5
Rheumatoid arthritis	83	18
Sequ. fractura colli femoris	27	5.9
Sequ. dysplasia	71	16
Sequ. dysplasia with dislocation	24	5.3
Sequ. Perthes/epiphysiolysis	83	18
Morbus Bechterew	20	4.4
Others	128	28
Missing diagnosis	3	0.7

^a More than one diagnosis is possible.

106,716 THR had been registered in the Arthroplasty Register as of December 31, 2004. Only 633 of these THR (both primary operations and revisions) had been performed in the 442 patients born after January 1, 1967, and were thus included in both the MBRN and NAR. 66 patients underwent revision, and 21 patients have had more than 1 revision. 1 patient had 8 revisions. 346 individuals (78%) underwent unilateral primary THR and 96 patients underwent bilateral primary THR (192 THRs). The average age for a person in NAR, with a primary THR and born after 1967, was 25 (12–36) years.

Sequelae after dysplasia with or without dislocation as sequelae was the most common reason for THR in these young adults (21%) (Table 2). In this group, 87% were women. In addition, Calvé-Legg-Perthes' disease/slipped capital femoral epiphysis (SCFE) and rheumatoid arthritis were two important groups of sequelae, constituting 18% each.

NHI and risk of THR

Of the 442 individuals registered in both the NAR and the MBRN, 2.5% (95% CI: 1.4–4.7) were born with NHI (Table 3) compared to 0.88% (95% CI: 0.87–0.90) in the whole MBRN. After adjustment for gender and year of birth, we found a 2.6-times (95% CI: 1.4–4.8) increased risk of THR for those reported to have NHI compared to those with stable hips at birth ($p = 0.002$) (Table 1). The absolute risks were, however, low: 57 (95% CI: 30–105) in 10⁵ (11 of 19,421) for children with NHI and

Table 3. Occurrence of neonatal hip instability (NHI) in the 442 individuals registered in the Norwegian Arthroplasty Register (NAR), according to sex, year of birth, and age at time of total hip replacement, and among the 2,218,596 individuals in the Medical Birth Registry of Norway (MBRN), according to sex and year of birth

	MBRN			NAR		
	Total	% Total	% NHI	Total	% Total	% NHI
Sex						
Men	1,140,027	51	0.5	192	43	2.1
Women	1,076,164	49	1.3	250	57	2.8
Unknown	1,811	0.1	0.4	0	0	0.0
Year of birth						
Before 1976	581,590	26	0.91	340	77	2.6
1976–1985	516,838	23	1.1	97	22	1.0
1986–1995	591,101	27	1.0	5	1.1	2.0
After 1995	529,062	24	0.48	0	0.0	0.0
Age at prosthesis						
< 25 years				183	41	2.7
25–29 years				134	30	2.2
30–34 years				97	22	3.1
≥ 35 years				28	6.3	0.0
Total	2,218,596	100	0.88	442	100	2.5

20 (95% CI: 18–22) in 10⁵ (431 of 2,198,733) for children without unstable hips at birth (Table 1). We found no statistically significant difference in the risk of prosthesis for children born before 1975 compared to those born later (Table 1).

Of the 442 patients reported to the NAR, only 11 of these had been diagnosed with unstable hips at birth. Of these 11 patients, 8 underwent THR due to dysplasia of the hip and 3 underwent THR for other reasons (femoral neck fracture, Calvé-Legg-Perthes' disease, and ankylosing spondylitis (Mb. Bechterew) as sequelae).

Discussion

The clinical importance of NHI has not yet been fully determined (Weinstein et al. 2003, Albinana et al. 2004); however, the general understanding is that instability at birth is associated with an increased risk of coxarthrosis later in life. In accordance with this, we have shown that neonatal hip instability at birth is associated with a 2.6-times increased risk of THR in young adulthood compared to those with stable hips at birth. However, only 8% of those who had undergone THR before the age of 38 due to dysplasia of the hip had been reported as having NHI. This suggests that

the screening of neonatal hip instability is not functioning properly.

The data from the Medical Birth Registry of Norway gave an incidence of neonatal hip instability of 0.88 per 100. This number agrees well with the figure of 1 in 100 reported by others (Rosendahl et al. 1996, Holen et al. 1999, Reikerås et al. 1999, Lehmann et al. 2000). The incidence of NHI between 1998 and 2004 was lower than that for 1967–1997. This may partly be explained by the introduction of a new standardized form from December 1998. The predominance of girls with NHI in our data (71%) is

also in accordance with the literature; for example, a figure of 71% was reported by Cyvin (1977) and 68% by Harris (1986). Furthermore, a relatively high incidence of THR due to osteoarthritis has been found in Norwegian women compared to other Nordic countries (Lohmander et al. 2006). This could indicate a genetic disposition for dysplasia of the hip in our country, although we have not performed any genetic analyses.

The incidence of unstable hips in the MBRN has not been validated, but as already stated, the incidence found is in good agreement with the literature. Furthermore, a study on completeness of registration of another congenital malformation (oral clefts) showed that 94% of cases of cleft lip and palate (CLP) were reported, but the percentage varied with the severity of the malformation (Kubon et al. 2007). Both of these findings support the view that the reporting of unstable hips to the MBRN is satisfactory.

The surgeon reports the reason for the operation to the registry. The accuracy of this diagnosis can be discussed, but in addition to the clinical examination and radiographs, these young patients should know if they have been treated for dysplasia of the hip in childhood. A study on the registered diagnoses in the Danish Hip Arthroplasty Register showed a high positive predictive value for hip

dysplasia (Pedersen et al. 2004). The registration process is the same in the Danish and the Norwegian Arthroplasty Registers, and it is justified to presume a similar accuracy in the diagnosis.

Newborns with unstable hips had a 2.6-times increased risk of THR compared to those with stable hips, until 38 years of follow-up. However, 92% of the patients who underwent THR due to dysplasia (87 of 95) had no reported instability of the hips at birth. This is a surprisingly high figure, as we would assume that a hip dysplasia requiring THR at a young age (before the age of 38 years) would be serious, and expect it to be diagnosable at birth. The basis of this reasoning is that the pathology is detectable by clinical examination at birth, which is what the clinical hip screening system is based on. Some explanations can be considered to explain our finding. Firstly, if we are to assume that all newborns diagnosed with NHI received treatment (i.e. with a Frejka pillow) and that the treatment was so effective that almost all developed normal hips, the patients with NHI and THR would be those for whom the treatment had failed (8 patients). Another possibility is that for some of the 87 (of 95) patients, NHI was detected but for some reason this was not reported to the MBRN. A more likely explanation may be that severe hip dysplasia could develop after birth. Some studies support such an interpretation (Aronsson et al. 1994). If this is the situation, a change in the current screening program of newborns in Norway should be considered. It is also important to state that the patients in this study were young, and younger than the average for those in general receiving THR in the NAR (Furnes et al. 2001). When those with NHI become older, the risk of THR may increase further.

In the 1990s, a selective ultrasound screening program was introduced in Norway. Children with any risk factors for hip dysplasia were examined using ultrasonography. It is assumed that the ultrasound screening program would reduce the prevalence of late-detected unstable hips (Rosendahl 1994), and in the future this may affect the incidence of THR in young adults.

Contributions of authors

IØE: planning the study, analysis and interpretation, drafting of the article. LBE: conception and design, data acquisition,

and critical revision of the manuscript. OF, SEV, TGL: participating in the analysis and interpretation of the results, and critically revising the manuscript. All authors gave final approval of the manuscript.

- Albinana J, Dolan L A, Spratt K F, Morcuende J, Meyer M D, Weinstein S L. Acetabular dysplasia after treatment for developmental dysplasia of the hip. Implications for secondary procedures. *J Bone Joint Surg (Br)* 2004; 86: 876-86.
- Aronsson D D, Goldberg M J, Kling T F. Developmental dysplasia of the hip. *Pediatrics* 1994; 94 (2 Pt 1): 201-8.
- Cyvin K C. Congenital dislocation of the hip. *Acta Paediatr Scand (Suppl 263)* 1977: 1-67.
- Espehaug B, Furnes O, Havelin L I, Engesaeter L B, Vollset S E, Kindseth O. Registration completeness in the Norwegian Arthroplasty Register. *Acta Orthop* 2006; 77: 49-56.
- Furnes O, Lie S A, Espehaug B, Vollset S E, Engesaeter L B, Havelin L I. Hip disease and the prognosis of total hip replacements. A review of 53 698 primary total hip replacements reported to the Norwegian Arthroplasty Register 1987-99. *J Bone Joint Surgery (Br)* 2001; 83: 579-86.
- Furnes O, Havelin L I, Espehaug B, Fenstad A M, Steindal K. *The Norwegian Arthroplasty Register 2006*. ISBN-13 978-82-91847-09-2. ISBN-10 82-91847-09-6. Bergen, Norway: 2005.
- Harris W H. Etiology of osteoarthritis of the hip. *Clin Orthop* 1986; (213): 20-33.
- Havelin L I, Engesaeter L B, Espehaug B, Furnes O, Lie S A, Vollset S E. The Norwegian Arthroplasty Register: 11 years and 73,000 arthroplasties. *Acta Orthop Scand* 2000; 71 (4): 337-53.
- Helsedirektøren. Veiledning for Leger og jordmødre om medisinsk registrering av fødsel. Fastsatt av helsedirektøren november 1966.
- Holen K J, Tegnander T, Eik-Nes S H, Terjesen T. The use of ultrasound in determining the initiation of treatment in instability of the hip in neonates. *J Bone Joint Surg (Br)* 1999; 81 (5): 846-51.
- Irgens L M. The Medical Birth Registry of Norway. Epidemiological research and surveillance throughout 30 years. *Acta Obstet Gynecol Scand* 2000; 79 (6): 435-9.
- Jacobsen S, Sonne-Holm S, Søballe K, Gebuht P, Lund B. Hip dysplasia and osteoarthritis. *Acta Orthop* 2005; 76 (2): 149-58.
- Kubon C, Sivertsen Å, Vindenes H A, Åbyholm F, Wilcox A, Lie R T. Completeness of registration of oral clefts in a medical birth registry: a population-based study. *Acta Obstet Gynecol Scand* 2007; 11: 1-5.
- Lehmann H P, Hinton R, Morello P, Santoli J. Developmental Dysplasia of the Hip Practice Guideline: Technical Report. *Pediatrics* 2000; 105 (4): E57.
- Lohmander L S, Engesaeter L B, Herberts P, Ingvarsson T, Lucht U, Puolakka T. Standardized incidence rates of total hip replacement for primary hip osteoarthritis in the 5 Nordic countries: similarities and differences. *Acta Orthop* 2006; 77: 733-40.

- Pedersen A B, Johnsen S, Overgaard S, Soballe K, Sorensen H T, Lucht U. Registration in the Danish hip arthroplasty registry: completeness of total hip arthroplasties and positive predictive value of registered diagnosis and postoperative complications, *Acta Orthop Scand* 2004; 75 (4): 434-41.
- Reikerås O, Hinderaker T, Steen H. Reduced acetabular depth in hip instability in the newborn, *Orthopaedics* 1999; 22: 943-6.
- Rosendahl K, Markestad T, Lie R T. Ultrasound screening for developmental dysplasia of the hip in the neonate: The effect of treatment rate and prevalence of late cases. *Pediatrics* 1994; 94 (1): 47-52.
- Rosendahl K, Markestad T, Lie R T. Developmental dysplasia of the hip. A population-based comparison of ultrasound and clinical findings. *Acta Paediatr* 1996; 85 (1): 64-9.
- Sahin F, Aktürk A, Beyazova U, Cakir B, Boyunaga Ö, Tezcan S, Bölükab i S, Kanatli U. Screening for developmental dysplasia of the hip: Results of a 7-year follow-up study, *Pediatr Int* 2004; 46: 162-6.
- Tegnander A, Holen K J, Anda S, Terjesen T. Good results after treatment with the Frejka pillow for hip dysplasia in newborns: a 3-year to 6-year follow-up study, *J Pediatr Orthop B* 2001; 10 (3): 173-9.
- Weinstein S L, Murbarak S J, Wenger D R. Developmental hip dysplasia and dislocation part I, *J Bone Joint Surg (Am)* 2003; 85: 1824-32.