Hip dysplasia in young adults

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SCIENTIFIC ENVIRONMENT

This thesis is based on data from the Norwegian Arthroplasty Register, the Medical Birth Registry of Norway, and data from the 1989 Hip Project.

The study was initiated in 2004 and in the beginning the work was carried out while the author was finishing medical school and internship. From 2010 full-time financial support was provided by the Western Norway Regional Health Authority. The different projects have been funded by the Department of Surgical Sciences at the University of Bergen, the Western Norway Regional Health Authority, the Departments of Orthopaedic Surgery and Radiology at Haukeland University Hospital, the Frank Mohn Foundation, the Research Council of Norway and Arthritis Research UK.

Supervision has been provided by staff at Department of Radiology and Department of Orthopaedic Surgery at Haukeland University Hospital and staff of the Norwegian Arthroplasty Register.

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2. ABSTRACT

According to data held by the Medical Birth Registry of Norway, approximately 1% of all newborns in Norway are diagnosed annually with hip instability at birth. Abduction treatment (Frejka pillow) for 6-12 weeks is the standard treatment in Norway, with which the majority will develop normal hips. Additionally to those detected by the neonatal screening program, some are late-diagnosed cases (diagnosis >1 month of age). For this group, the treatment is usually more demanding and prolonged. The older the child is at start of treatment, the poorer is the prognosis. A dysplastic hip causes altered mechanical conditions, predisposing for increased wear of the cartilage and development of osteoarthritis of the hip in young adult age. The final treatment option for this condition could be a total hip replacement.

In Paper I, we linked two national registries, the Norwegian Arthroplasty Register and the Medical Birth Registry of Norway, using the unique national identification number. The Birth Registry contains information on all newborns in Norway from 1967 and the Arthroplasty Register includes all total hip replacements inserted in Norway from 1987. The study found a 2.6 times increased risk for a total hip replacement in young adulthood for patients reported with hip instability in the newborn period. The absolute risk was however low at only 57 in $10^5$ for patients with hip instability compared to 20 in $10^5$ for those with stable hips. Only 8% of those who underwent a total hip replacement due to hip dysplasia were reported to have had instable hips at birth.

In Paper II, we validated the dysplasia diagnosis reported to the Norwegian Arthroplasty Register for subjects born after 1967. Medical records were reviewed and we also investigated age at dysplasia diagnosis, previous treatment and quality of life. We found the dysplasia diagnosis reported to the Arthroplasty Register to be correct in 88% of the hips. Median age at time of diagnosis was as high as 7.8 years: 4.4 years for females and 22 years for males. 75% of the patients had undergone different hip-preserving treatments before their prosthesis, and the dysplasia patients
scored poorer in quality of life (EQ-5D) compared to the age-matched general population in Sweden and the UK.

In Paper III, we aimed to validate a digital measurement programme for hip dysplasia at skeletal maturity. Ninety-five radiographs were measured by three independent observers in both a newly developed digital measurement programme and manually in AgfaWeb1000. Eleven radiological measurements, all relevant for hip dysplasia at skeletal maturity, were evaluated. We found acceptable inter- and intra-observer reproducibility for most measurements, but with poorer accuracy for measurements with small absolute values. The reproducibility was relatively similar for the two methods used, but the digital measurements were performed much faster.

In Paper IV, we used data from the 1989 Hip Project and reported on the prevalence of hip dysplasia in 2081 19-year old Norwegians. The prevalence of hip dysplasia in the cohort varied from 1.7% to 20% depending on the radiological measurement used. A Wiberg’s angle <20° was seen in 3.3% of the cohort: 4.3% in women and 2.4% in men. We found no association between subjects with radiological signs indicating hip dysplasia and body mass index (BMI), Beighton hypermobility score, EQ-5D score or WOMAC score.

The overall conclusions of this thesis are as follows: About 25% of all total hip replacements in young adults (< 40 years) are performed due to an underlying hip dysplasia. The dysplasia diagnosis is in general detected late, indicating that clinical testing for hip instability in newborns is an insufficient screening method to detect hips that require a total hip replacement in young adulthood. Several radiographic measurements for hip dysplasia are proposed in the literature. The reproducibility for these measurements varies, but with acceptable results for the more common measurements such as Wiberg’s centre-edge and Sharp’s acetabular angle. The prevalence of hip dysplasia is highly dependent on the radiographic measurements used, but a high prevalence for some of the measurements is found in skeletally mature Norwegians as compared to other studies on Caucasians in the literature.
3. NORSK SAMMENDRAG

Hofteledddsysplasi er en tilstand hvor hofteleddene er ufullstendig utviklet og hofteskålen er grunnere enn normalt. Dette disponerer for instabilitet i hofteleddet og lårhodet kan helt eller delvis gå ut av ledd. Basert på tall fra Medisinsk Fødselsregister, rapporteres årlig rundt 1 % av alle nyfødte i Norge med hofteleddssinstabilitet. Den vanligste behandlingen er 6-12 ukers abduksjonsbehandling med Frejkas pute, og med denne utvikler de fleste normale hofter. I tillegg til dem som diagnostiseres ved fødsel, er det noen som er sen-dagnostiserte (alder >1 måned ved diagnose). For denne gruppen kan behandlingen være mer komplisert og langvarig. Jo eldre barnet er ved behandlingsstart, dess årligere prognose. En dysplastisk hofte medfører endrete mekaniske forhold i hofteleddet. Dette disponerer for økt bruskslitasje, og fare for utvikling av artrose i hoften i ung voksen alder som kan ende opp med en totalprotese i hoften.

I artikkel I koblet vi to nasjonale helseregister; Nasjonalt Register for Leddproteser og Medisinsk Fødselsregister. Fødselsregisteret inneholder informasjon om alle nyfødte i Norge fra 1967 og Leddregisteret omfatter alle totalproteser i hofteleddet satt inn i Norge fra 1987. Studien fant en 2,6 ganger økt risiko for en hofteprotese i ung voksen alder for pasienter diagnostisert med hofteleddssinstabilitet i nyfødtperioden. Imidlertid var den absolute risikoen lav, bare 57 av 10^5 for pasienter med hofteleddssinstabilitet sammenlignet med 20 av 10^5 for dem med stabile hofter. Kun 8 % av dem med en totalprotese på grunn av hofteledddsysplasi var rapportert med instabile hofter ved fødsel.

I artikkel II validerte vi diagnosene/årskene til en totalprotese i hofteleddet som var rapportert til Nasjonalt Register for Leddproteser for pasienter født etter 1967. Medisinske journaler ble gjennomgått og vi noterte alder ved diagnose, tidligere behandling og i tillegg ble pasientene spurt om livskvalitet (EQ-5D). Vi fant at 88% av hofteledddsysplasi-diagnostics rapportert til registeret var korrekte. Median alder ved diagnosetidspunkt var hele 7,8 år; 4,4 år for kvinner og 22 år for menn. 75 % av pasientene hadde gjennomgått ulike hoftebeværende operasjoner før proteseinsetting,
og dysplasipasientene scoret dårligere på livskvalitet sammenlignet med den alders-
matchete generelle befolkningen i Sverige og Storbritannia.

I artikkel III validerte vi et digitalt røntgenmåleprogram for hofteleddsdysplasi hos
skjelettmodne. 95 røntgenbilder ble målt av tre uavhengige personer i både et digitalt
måleprogram (DDH_Adult) og manuelt i AgfaWeb1000. 11 røntgenmål, alle
relevante for hofteleddsdysplasi, ble vurdert. Vi fant akseptabel inter- og intra-
observatør reproduserbarhet for de fleste målingene, men med dårligere nøyaktighet
for målinger med lave absoluttverdier. Reproduserbarheten var relativt lit for de to
metodene, men de digitale målingene var mye raskere å utføre.

I artikkel IV brakte vi data fra Hofte89-studien og undersøkte prevalensen av
hofteleddsdysplasi blant 2081 norske 19-åringar. Utbredelsen av hofteleddsdysplasi i
kohorten varierte fra 1,7 % til 20 % avhengig av hvilket røntgenmål som ble benyttet.
Wibergs vinkel <20° ble funnet hos 3,3 % av personene: 4,3 % hos kvinner og 2,4 %
hos menn. Vi fant ingen sammenheng mellom personer med radiologiske tegn på
hofteleddsdysplasi og body-mass-index (BMI), Beighton hypermobilitets score og
livskvalitets-score (EQ-5D og WOMAC).

Hovedfunnene i denne avhandlingen er at rundt 1/4 av alle totalproteser hos unge
voksne (<40 år) skyldes en underliggende hofteleddsdysplasi. Dysplasidiagnosen er
generelt sent diagnostisert (median 7,8 år), noe som indikerer at klinisk undersøkelse
for hofteleddsdysplasi hos nyfødte ikke er en tilstrekkelig screeningmetode for å
oppdage de hoftene som krever totalprotese i ung voksen alder. Flere ulike
røntgenmål for hofteleddsdysplasi er beskrevet i litteraturen. Reproduserbarheten for
disse er varierende, men vi fant akseptable resultater for de vanligste målene som
Wibergs centre-edge (CE)-vinkel og Sharps vinkel. Prevalensen av
hofteleddsdysplasi er avhengig av hvilket røntgenmål som benyttes, men vi fant
høyere forekomst for enkelte røntgenmål blant skjelettmodne nordmenn
sammenlignet med andre studier på kaukasiske.
4. LIST OF PUBLICATIONS

This thesis is based on the following papers, referred to in the text by their Roman numerals:


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<table>
<thead>
<tr>
<th>Abbreviation</th>
<th>Description</th>
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<tbody>
<tr>
<td>AA</td>
<td>Acetabular roof Angle of Tönnis</td>
</tr>
<tr>
<td>AAOS</td>
<td>American Academy of Orthopedic Surgeons</td>
</tr>
<tr>
<td>ADR</td>
<td>Acetabular Depth-width Ratio</td>
</tr>
<tr>
<td>AP</td>
<td>Anteroposterior</td>
</tr>
<tr>
<td>ARO</td>
<td>Acetabular Roof Obliquity</td>
</tr>
<tr>
<td>ATD</td>
<td>Articulo-Trochanteric Distance</td>
</tr>
<tr>
<td>AVN</td>
<td>Avascular Necrosis</td>
</tr>
<tr>
<td>BMI</td>
<td>Body Mass Index</td>
</tr>
<tr>
<td>CE angle</td>
<td>Centre-Edge angle of Wiberg</td>
</tr>
<tr>
<td>CDH</td>
<td>Congenital Dislocation of the Hip</td>
</tr>
<tr>
<td>CI</td>
<td>Confidence Interval</td>
</tr>
<tr>
<td>CLP</td>
<td>Calvé-Legg-Perthes’ disease</td>
</tr>
<tr>
<td>CT</td>
<td>Computed Tomography</td>
</tr>
<tr>
<td>DDH</td>
<td>Developmental Dysplasia of the Hip</td>
</tr>
<tr>
<td>DICOM</td>
<td>Digital Imaging and Communication in Medicine</td>
</tr>
<tr>
<td>EQ-5D</td>
<td>EuroQol Five-Dimensional Questionnaire</td>
</tr>
<tr>
<td>EQ-VAS</td>
<td>EuroQol Visual Analogue Scale</td>
</tr>
<tr>
<td>FHEI</td>
<td>Femoral Head Extrusion Index</td>
</tr>
<tr>
<td>HTE</td>
<td>Horizontal Toit Externe</td>
</tr>
<tr>
<td>HUS</td>
<td>Haukeland University Hospital</td>
</tr>
<tr>
<td>ICC</td>
<td>Intraclass Correlation Coefficient</td>
</tr>
<tr>
<td>ICD</td>
<td>International Classification of Diseases</td>
</tr>
<tr>
<td>IOE</td>
<td>Ingvild Øvstebø Engesæter</td>
</tr>
<tr>
<td>JSW</td>
<td>Joint Space Width</td>
</tr>
<tr>
<td>Abbreviation</td>
<td>Full Form</td>
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<tr>
<td>KR</td>
<td>Karen Rosendahl</td>
</tr>
<tr>
<td>LBE</td>
<td>Lars Birger Engesæter</td>
</tr>
<tr>
<td>LBL</td>
<td>Lene Bjerke Laborie</td>
</tr>
<tr>
<td>MBRN</td>
<td>Medical Birth Registry of Norway</td>
</tr>
<tr>
<td>MDC</td>
<td>Minimal Detectable Change</td>
</tr>
<tr>
<td>MRI</td>
<td>Magnetic Resonance Imaging</td>
</tr>
<tr>
<td>n</td>
<td>Number</td>
</tr>
<tr>
<td>NAR</td>
<td>The Norwegian Arthroplasty Register</td>
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<tr>
<td>NHI</td>
<td>Neonatal Hip Instability</td>
</tr>
<tr>
<td>NSAIDs</td>
<td>Non-Steroidal Anti-Inflammatory Drugs</td>
</tr>
<tr>
<td>OA</td>
<td>Osteoarthritis</td>
</tr>
<tr>
<td>OR</td>
<td>Odds Ratio</td>
</tr>
<tr>
<td>PAO</td>
<td>Periacetabular Osteotomy</td>
</tr>
<tr>
<td>POSNA</td>
<td>The Pediatric Orthopedic Society of North America</td>
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<tr>
<td>PPV</td>
<td>Positive Predictive Value</td>
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<tr>
<td>QoL</td>
<td>Quality of Life</td>
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<tr>
<td>RCT</td>
<td>Randomised Controlled Trial</td>
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<tr>
<td>ROM</td>
<td>Range Of Motion</td>
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<tr>
<td>SCFE</td>
<td>Slipped Capital Femoral Epiphysis</td>
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<tr>
<td>SD</td>
<td>Standard Deviation</td>
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<tr>
<td>TGL</td>
<td>Trude Gundersen Lehmann</td>
</tr>
<tr>
<td>THR(s)</td>
<td>Total Hip Replacement(s)</td>
</tr>
<tr>
<td>UK</td>
<td>United Kingdom</td>
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<tr>
<td>US</td>
<td>Ultrasonography</td>
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<tr>
<td>VAS</td>
<td>Visual Analogue Scale</td>
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<tr>
<td>Acronym</td>
<td>Description</td>
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<tr>
<td>WHO</td>
<td>World Health Organization</td>
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<tr>
<td>WOMAC</td>
<td>Western Ontario and McMaster Universities Osteoarthritis Index</td>
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6. BACKGROUND

6.1 INTRODUCTION TO THE FIELD

Hip pain is a frequent problem and is caused by numerous conditions. Degeneration of the hip joint (osteoarthritis) is one of the more common causes and often presents with groin pain that worsens during activity and improves with rest. The cause of osteoarthritis (OA) may be unknown (idiopathic osteoarthritis), or secondary to other conditions such as fractures, rheumatologic diseases, childhood hip diseases, or side effects of medical treatments and drugs. The end-stage treatment for hip osteoarthritis could be a total hip replacement (THR).

Hip dysplasia is the most common of the childhood hip diseases and an important cause to secondary hip osteoarthritis. It is normally diagnosed in the newborn period by clinical examination supplemented with ultrasound. The hips present with a shallow acetabulum (hip socket) (Figure 1B), most often associated with an unstable

Figure 1 Illustration of A) a normal hip joint with the femoral head nicely located inside the acetabulum, and B) a dysplastic acetabulum with a subluxated femoral head.
or dislocatable femoral head. In severe cases the hip is dislocated. The aim of the
treatment is to maintain the femoral head within the acetabulum, leading to a normal
development of the hip joint. Different treatment techniques are in use, ranging from
various harnesses (the Frejka pillow being used in Norway), orthosis, hip spica cast
and several surgical procedures. If the mechanical configuration is not satisfactorily
re-established, the patient is at risk of developing hip osteoarthritis in early adulthood.
Early treatment improves the outcome.

Hip dysplasia is an important underlying cause of total hip replacement in young
adults. In Norway, all total hip arthroplasties performed since 1987 are reported
to the Norwegian Arthroplasty Register. The register holds information on the
patient, the operation, implants used and any revisions. Almost 140 000 primary total
hip replacements have been registered, in which hip dysplasia was the underlying
cause of 8%. This is a higher percentage than in the other Nordic countries, which
report figures of 2-3%. This may indicate a relatively high prevalence of hip
dysplasia among Norwegians.

In order to make a correct diagnosis in medicine, one has to know what diagnostic
tools to use and how to use them. For hip dysplasia, the diagnosis is based on clinical
and radiological examination. Multiple radiological measurements are proposed in
order to define hip dysplasia at skeletal maturity, but the reported validity of the
different markers varies. This is of relevance when the prevalence is studied and
related to findings in the literature. Comparing findings based on different diagnostic
criteria may lead to incorrect conclusions.

This thesis focuses on hip dysplasia in young adults. By using national registers (the
Norwegian Arthroplasty Register and the Medical Birth Registry of Norway),
medical records, and data from a follow-up hip study (The 1989 Hip Project), I have
tried to enhance our understanding of diagnosis and occurrence of hip dysplasia at
skeletal maturity, and total hip replacement due to hip dysplasia in young adults.
6.2 HIP DYSPLASIA

Hip dysplasia is a complex disorder, both in diagnosis and treatment. A poorly developed (=dysplastic) acetabulum is found in these patients, and without proper treatment, the hip is at risk of developing osteoarthritis.

6.2.1 A historical overview

Hip dysplasia is a condition known since ancient times. The disease was already described by Hippocrates (460-370 B.C.) who stated that it might be congenital and caused by an injury to the mother’s abdomen. More than 1500 years later, Ambroise Paré (1510-1590) suggested that the condition might be hereditary and that a shallow acetabulum developed when the femoral head was not exactly reduced. In 1783, the first description of a dislocated hip in a post-mortem specimen was given by Dr Paletta from Vienna, and in 1826 the French surgeon Guillaume Dupuytren (1777-1835) presented a dissertation on hip joint dislocations. He noted that in one type of dislocation there was “a defect in the depth or completeness of the acetabulum” and named it “original or congenital dislocations”. “Congenital dysplasia of the hip” (CDH) was accepted as a distinct clinical term for most of the next century.

The first report of hip dysplasia as a separate entity was written by Dr Parise in 1842, who reported on autopsies in two infants who had subluxated hips and flattened acetabula. In 1906, Gourdon reported three patients (5, 15 and 19 years of age), all with mal-developed upper acetabular borders. The patients reported pain and easy fatigability, and this was the first association of hip dysplasia with clinical symptoms. The first report of an association of hip dysplasia and osteoarthritis was compiled in 1924 by Le Scolan who studied 28 patients with both acetabular dysplasia and hip subluxation. Six of these patients aged 16-46 years had osteoarthritis.

Pravaz (1847) and Paci (1887) were the first to report on closed reduction of the femoral head using prolonged traction and bed rest as treatment. Roser (1879) suggested that hip dislocation could be diagnosed in the newborn period and that
proposed that an abnormal foetal position might cause the hip dislocation\textsuperscript{136}. Further, he described that unstable hips in newborns could be reduced by abduction and re-dislocated by adduction. The first successful open reduction of the femoral head was performed by Hoffa in 1890. Several reports on surgical intervention were presented during the 19\textsuperscript{th} century, but all had many complications (Poggi 1888, Hoffa 1890, König 1891).

The Austrian orthopaedic surgeon Lorenz (1854-1946) became an important advocate of conservative treatment of dislocation of the hip. Due to the development of a severe skin allergy, he could not perform traditional surgical operations and became known as “the Bloodless Surgeon of Vienna”\textsuperscript{115}. Dr Lorenz was renowned for his conservative technique of reducing the femoral head into the acetabulum under light anaesthesia with a subsequent hip spica cast in abduction followed by external rotation as the child matured. He was also the first to add a walking frame to the cast so that the child was somewhat mobile. Lorenz reported good results with this method of treatment and it became the most popular method for a long time. Dr Lorenz stated that treatment with reduction should start at 2-3 years of age, as a diagnosis made before the child started to walk was uncertain. This idea of late onset of the treatment was popular well into the 20\textsuperscript{th} century, but was also countered by many opponents. One of them was Hilgenreiner (1870-1954) (famous for describing Hilgenreiner’s line on radiographs) who claimed that reduction was very easy in children 3-6 months of age. Dr Hilgenreiner also suggested the name dysplasia with or without dislocation in 1925\textsuperscript{101}.

Dr Putti (1880-1940) was an orthopaedic surgeon from Bologna, an area in Italia with a high prevalence of hip dysplasia. He was the first physician to realise the importance of early recognition and treatment of CDH, and recommended all newborn babies to be screened by x-rays in order to detect CDH. He also described that a child with a CDH of one hip could have a located or minimally subluxated hip with a shallow acetabulum on the contralateral side\textsuperscript{184}. 
Dr Marino Ortolani (1904-1983) studied medicine in Bologna in the 1920s and was a student of Dr Putti. He opened a paediatric clinic outside Bologna, where one of his patients was a 5-month-old boy whose mother had recognized a “click” every time the baby was washed in the perineal region. Dr Ortolani concluded that the “click” occurred with abduction-adduction motion of the thigh, and described his famous method of assessing hip instability (“Ortolani’s manoeuvre”) in 1937. The clinical test had already been described by Le Damany and Saiget in 1910, but Dr Ortolani brought it to prominence. Dr Ortolani reported good results on early diagnosis and treatment using a pillow keeping the hips in a flexed and abducted position and this soon became the dominant way to deal with neonatal hip instability. A modification of this pillow was introduced in 1941 by the Czech orthopaedic surgeon Bedrich Frejka (1890-1972). The Frejka pillow is today the preferred treatment option of dysplastic hips in newborns in Norway.

Sherman Coleman (1922-2004), an orthopaedic surgeon from Salt Lake City, and Kurt Palmén, a paediatrician from Sweden, noted that some babies with CDH did not have a positive Ortolani sign, but only a “jerk of exit”. The hip could be provoked to subluxation. In 1962, the English orthopaedic surgeon T. G. Barlow used the term dislocatable. He studied 10 000 newborns in the first week of life for hip instability and found a higher incidence (1:60) than previously anticipated. The majority of the unstable hips did not have a positive Ortolani sign. They were located at rest, but were dislocatable when tested. Barlow also noticed that the incidence was greater in the first days of life (0-3½ days, 1:25) as compared to slightly older babies (3½ -7 days, 1:100). This indicated that the instability resolves quickly.

In the late 1970s, the Austrian orthopaedic surgeon Dr Reinhard Graf noted that the cartilaginous femoral head and acetabulum could be imaged by ultrasound. Subsequently, Dr Harcke, a radiologist from Wilmington, Delaware, demonstrated a dynamic ultrasound of the dynamic hip. Harcke made it possible to see what Ortolani and Barlow could only feel.
Several important works on hip dysplasia have been written in Scandinavia. In 1939, the Swedish orthopaedic surgeon Gunnar Wiberg (1902-1988) published his thesis on the dysplastic acetabulum and congenital subluxation of the hip joint. He introduced the so-called Wiberg’s centre-edge (CE) angle which became a classic radiographic measurement describing the acetabular coverage of the femoral head. Two years later, the work of Severin (1941) showed that the Lorenz method of late onset of treatment had very poor long-term results and found an improved outcome in infants with early treatment start. The paediatrician Kurt Palmén introduced clinical screening of the hip joints for all newborns in Falköping in Sweden in the 1950s, and based on these experiences, Fredensborg (1976) concluded that late detected hip dysplasia could almost be eliminated by a dedicated clinical examination. In Norway, Walther and Moe (1954) were the first to report results of neonatal hip screening of all newborns240. Later, the studies of Bjerkreim in 197429 and Cyvin (1928-1990) in 197755 were the most important in Norway. In recent decades, diagnostic approaches and screening strategies of hip dysplasia have received much attention. Ultrasonography has been introduced and the theses of Dr Rosendahl (1995)189 and Dr Holen (1999)106 have shed light on the role of ultrasonography in the diagnosis and treatment of hip dysplasia in newborns.

### 6.2.2 Normal development of the hip joint

The hip joint starts to develop from mesenchymal cells as early as the 4th-6th week of gestation. In the 7th week, a cleft appears in the pre-cartilage cells defining the femoral head and the acetabulum, and at 11 weeks the femoral head is fully formed with a spherical configuration209. In late gestation the further growth of the femoral head is more rapid than the growth of the acetabular cartilage, so that the femoral head is less than 50% covered at birth. At birth the acetabulum is at its most shallow and most lax in order to maximise the hip range of motion which facilitates the delivery process. However, it is extremely difficult to dislocate a normal infant’s hip as the retaining forces are similar to that of a suction cup. Accordingly, dislocatable
hips in newborns are not merely normal hips with increased capsular laxity, they are pathological cases.

Post-natally the acetabular cartilage develops faster than the femoral head, which progressively allows more coverage. At this time the cartilage is tri-radiated medially and cup-shaped laterally. The acetabulum is formed by the ilium above, the ischium below and the pubis anteriorly (Figure 2).

At birth the proximal end of the femur is composed entirely of cartilage. Between the 4th and 7th months of life, the proximal femoral ossification centres begin to appear. These bony centres continue to enlarge and replace the cartilage until adult age when only a thin layer of cartilage remains over them. The development of the hip joint until skeletal maturity occurs through balanced growth of the proximal femur, the acetabular tri-radiate cartilage and the adjacent bones. This balance is probably genetically determined, and the familial predisposition of hip dysplasia may partly be due to this. The replacement of the cartilage by bone in the proximal femur and the increase of the normal depth of the acetabulum require a femoral head placed in the acetabulum. In conditions where this does not happen, due to e.g. a (sub)luxated femoral head, the development is delayed and the bony hip joint will not be mechanically satisfactory. In cases of hip dysplasia it is therefore important to reduce the femoral head as soon as possible, and the reduction must be maintained to provide the normal stimulus for acetabular development.
6.2.3 Definition and terminology

Hip dysplasia includes a wide spectrum of pathology, ranging from only mild radiological changes without subjective symptoms to a dislocated hip joint showing severe radiographic deformities and disabling ailments for the patient. The terminology of hip dysplasia has changed over time and various terms have been proposed and used. Variation in both severity of the disorder and age at diagnosis may partly explain this conceptual dissatisfaction and confusion.

In the beginning of the 19th Century the term congenital dislocation of the hip (CDH) was introduced and soon became an accepted term, reflecting all kinds of hip joint instability, at birth or later. But the fact that a dysplastic hip often does not develop into a dislocated hip and even when dislocation occurs, this often happens post-natally, resulted in a change of the term to developmental dysplasia of the hip (DDH). The American Academy of Orthopedic Surgeons (AAOS) and The Pediatric Orthopedic Society of North America (POSNA) have advocated the use of DDH, as the term includes all cases that are clearly congenital and those that are developmental, incorporating subluxation, dislocation and dysplasia of the hip. Some authors differentiate between teratologic and typical dislocation of the hip. A teratologic dislocation is associated with other malformations (chromosomal abnormalities and neuromuscular disorders). A typical dislocation occurs in an otherwise healthy newborn.

Hip dysplasia is another commonly used term, also in this thesis where it is defined as including the same abnormalities as described for DDH, but the hip dysplasia term does not have the challenges regarding time of diagnosis. Neonatal hip instability (NHI) is used for hip joint instability detected at birth (Paper I).

In addition to the already mentioned terms, there are some other commonly used definitions (Figure 3). Some of the terms refer to the morphology of the hip and others to the stability. Acetabular dysplasia is a morphological term and refers to a condition with shallow acetabulum with an increased slope. Immature hip is used to describe a morphological borderline group when neonatal hips are examined with
ultrasound. Stability terms include dislocatable and subluxatable. A totally *dislocated hip* refers to the situation in which the whole femoral head is outside the acetabulum, in contrast to a *subluxated hip* where the femoral head is partially outside the acetabulum and thus the femoral head is poorly covered by the acetabulum. In a *subluxatable hip* the hip can be pushed into this position, i.e. partly out of the acetabulum, usually due to ligamentous laxity. In a *dislocatable hip* the femoral head is reduced at rest (i.e. within the acetabulum), but can be dislocated in specific positions or examination manoeuvres. This is thus a hip with instability. In a *reducible hip*, the hip is dislocated at rest, but can be reduced into the acetabulum with manipulation.

### 6.2.4 Epidemiology

The exact prevalence of hip dysplasia is controversial and comparing the literature is challenging as variations in definitions of diagnosis, methods of diagnosis, age groups, clinical experience of the examiner, ethnicity and geography are intermingled. Furthermore, the results are affected by the period when the studies were conducted. Before the 1950s the prevalence was estimated arbitrarily, from the 1950s to the 1980s the prevalence was based on unstable hips from the neonatal clinical examination plus the addition of late-diagnosed patients, and from the 1980s ultrasound was introduced in additional to clinical examination.\(^{149}\)
A review of studies from 44 unscreened populations of predominately northwest European ancestry found the median birth prevalence of persistent and clinically diagnosed hip dysplasia to be 1.3 per 1000 (range 0.8-1.5)\textsuperscript{143}. By contrast, the prevalence of neonatal hip instability, ascertained through clinical examination, was higher, ranging from 1.6 to 28.5 per 1000. In 2000, a meta-analysis on the prevalence of DDH found the following estimates: 9 per 1000 based on clinical screening by paediatrician, 12 per 1000 based on screening by an orthopaedic surgeon, and 25 per 1000 on ultrasound screening (different techniques used)\textsuperscript{146}. In Norway, about 1-2% of all newborns are found to have unstable hips at birth assessed by Ortolani’s and/or Barlow’s test\textsuperscript{56,76,103}. The corresponding number for hips examined with ultrasound is between 2 and 4%\textsuperscript{194}. Moreover, 0.5-3 in 1000 are late-detected hip dysplasia (age at diagnosis >1 month) in Norway.

The reported prevalence of adult hip dysplasia varies throughout the literature according to sex, ethnicity, different radiological measurements with corresponding cut-off values and different radiological material. However, many studies on Caucasians report a hip dysplasia prevalence of approximately 2-4% (Wiberg’s centre-edge angle <20\textdegree)\textsuperscript{53,120}. Females have a higher prevalence than males and the prevalence is also higher in the Sami population\textsuperscript{125}.

### 6.2.5 Aetiology

The complete aetiology for hip dysplasia is unknown, but several risk factors have been identified.

*Family history.* Although no genes have been identified, we know that there is a genetic influence as some families are more affected than others. Two different components regarding the genetic mechanisms in hip dysplasia have been proposed, one related to the connective tissue laxity, and the other to the shape of the acetabulum. The degree of interaction between these two components remains unknown. A polygenic or monogenic autosomal dominant inheritance is assumed for
connective tissue laxity, and the acetabular component is presumed to have a polygenic inheritance. Increased concordance of hip dysplasia is found in monozygotic compared to dizygotic twins. Although monozygotic twins are not invariably concordant, they are concordant much more frequently than dizygotic twins, indicating a part genetic and part environmental aetiology. In a study based on the Norwegian Twin Registry, the odds ratio for prevalence of hip dysplasia was reported to be much higher for mothers (OR 35.8) than for siblings (OR 12.7), fathers (OR 8.1), and offspring (OR 3.3), which suggests a maternal effect.

**Female gender.** Females are affected more than four times as often as males and a hormonal influence has been proposed. Relaxin is a maternal hormone which contributes to ligamentous laxity in the mother’s pelvis and to neonatal ligamentous laxity (especially in the female foetus). The hormone passes freely through the placenta into the newborn and may increase the risk of hip dysplasia. However, a study found no difference in mean relaxin concentration between cord bloods of a group of 24 newborns with hip dysplasia, and a control group of normal babies matched by sex and gestation. But, as suggested by the authors, the relaxin receptor expression of the developing foetal hip joint needs to be explored. A 1997 study of 90 newborns reported an insignificant decrease of relaxin concentrations in babies with increased sonographic hip instability. Other hormones being investigated are urinary oestrogen, serum β-estradiol and serum cord blood relaxin, but without any clear-cut conclusions.

**Mechanical factors/limited foetal mobility.** Lack of space in the uterine cavity predisposes for hip dysplasia. This is seen in breech presentation, first-time mothers (probably due to the unstretched abdominal muscles) and decreased amount of amniotic fluid. Increased birth weight (>4000g) is reported as a risk factor in some studies, whereas others are not able to confirm these findings. A higher risk is seen among children with other malformations in the musculoskeletal system that can be related to abnormal positioning and/or limited foetal mobility; i.e. metatarsus adductus and congenital muscular torticollis. An association with clubfoot is inconsistent. Further, breech presentation is more common in infants with hip
dysplasia than in the general population (16 versus 3%)\textsuperscript{161}. Chan and colleagues found in 1997 that vaginally delivered breech births had a significantly higher risk for hip dysplasia (OR 17) than breech births delivered by Caesarean section (OR 10)\textsuperscript{18}. These findings are in concordance with others\textsuperscript{81,151}, even though Hinderaker et al (1994) found no significant difference in the occurrence of hip dysplasia based on method of delivery using data from the Medical Birth Registry of Norway\textsuperscript{103}.

Postnatal treatment and care can also influence the development of the hip joint. Tight swaddling has been popular in some cultures to help decrease crying and promote uninterrupted sleep in neonates. However, swaddling has been recognised as a risk factor for hip dysplasia\textsuperscript{182,235,241}. In certain cultures in which swaddling has been especially prevalent, e.g. Canadian Indian tribes\textsuperscript{197} and American Indians\textsuperscript{132}, a higher rate of hip dysplasia is seen. The same theory has been proposed for the Norwegian Sami population, as they traditionally used a special form of swaddling, with a higher prevalence of hip dysplasia in those areas\textsuperscript{125}. In Japan, a public health campaign to alter infant swaddling practices was initiated and resulted in a subsequent fall in the prevalence of hip dysplasia\textsuperscript{197,253}. The American Academy of Pediatrics Clinical Practice Guidelines\textsuperscript{10} recommend avoiding swaddling if hip dysplasia is found, but state that it is probably safe in hips with a normal hip ultrasound.

The left hip is three times more commonly affected than the right hip\textsuperscript{12,65}. This is due to the majority of infants being in the left occiput anterior position at the time of birth. In this position, the left hip lies posteriorly against the mother’s spine with left femur adducted, which may predispose the left hip to a higher incidence of hip dysplasia\textsuperscript{17}.

Other malformations. Hip dysplasia occurs most commonly in otherwise healthy infants, but is associated with certain conditions. Teratologic hip dysplasia is associated with various syndromes (e.g. Ehler Danlos syndrome, Down syndrome and arthrogryposis), and neuromuscular hip dysplasia occurs when spasticity in the hip muscles group (e.g. in spina bifida and cerebral palsy) pulls the femoral head out
of the acetabulum, resulting in unsatisfactory development and a dysplastic hip joint. The diagnosis and management of teratologic and neuromuscular hip dysplasia may differ from when hip dysplasia is seen in otherwise healthy children.

*Seasonal variation.* Some studies have reported a seasonal variation of hip dysplasia, with a peak incidence in autumn and winter months\(^{15}\). This may be due to increased swaddling or tight clothing to protect the baby from the colder weather. In a study from Norway, Cyvin found that girls showed a significant seasonal variation with peak incidence in September and October; whereas there was no seasonal peak for boys\(^{55}\). Bjerkreim observed a seasonal variation in late diagnosis of hip dysplasia, but found no seasonal variation in the neonatal group. Other studies have, however, not been able to confirm this seasonal variation\(^{48}\).

*Joint laxity.* Some studies have found increased joint laxity in patients with hip dysplasia. Wynne-Davies and colleagues found that a higher proportion of children with hip dislocation were lax-jointed than the control population. Further, they found that a higher proportion of neonates with congenital dislocation and their first degree relatives had joint laxity than the late diagnosis group\(^{251}\). In a Japanese study, it was found that female athletes with an anterior cruciate ligament injury had an increased prevalence of acetabular dysplasia and generalised joint laxity\(^{254}\).

### 6.2.6 Natural history of hip dysplasia

The natural history of hip dysplasia depends on age at diagnosis and the severity of the condition. A high proportion of babies diagnosed with hip dysplasia in the newborn period will develop normal and stable hip joints without any treatment. Observational studies have reported high rates of resolution without intervention\(^{47,219}\), including a randomised study that showed that mildly dysplastic but stable hips normalised spontaneously in around half of the cases\(^{42,190}\). Barlow, who studied more than 11 000 newborns in the period 1957-1962, reported that in 60% of the cases with
detected laxity, the hips recovered spontaneously after 3-4 days of life and 88% stabilised by two months of age\textsuperscript{22}.

Although most mildly dysplastic and/or unstable hips will recover spontaneously, some will remain dysplastic and even unstable. A few will deteriorate into severe dysplasia with or without a subluxated or dislocated femoral head. Hips that are severely dysplastic and dislocated at birth do not tend to reduce spontaneously. However, whether or not completely normal hips in the newborn period can turn into dysplastic, dislocated hips in later infancy in otherwise normal children is controversial.

A dysplastic hip without (sub)luxation will often remain asymptomatic for many years. However, the altered mechanical conditions predispose for development of osteoarthritis of the hip in young adult age and will present with typical symptoms of osteoarthritis. Women with moderate involvement typically begin to develop symptoms in association with their first or second pregnancy, men a couple of decades later. In a hip with subluxation, the degeneration process will normally begin at an earlier stage depending on the degree of subluxation. In situations with a complete dislocation, the symptoms depend on whether a false acetabulum has developed or not. In young children, pain is uncommon, but becomes more severe as the child matures. A limp is often seen and weakness in the hip abductor muscles may result in the classic Trendelenburg gait pattern. Patients with a bilateral dislocation may complain of back pain from a secondary hyperlordosis of the lumbar spine. Knee pain may be seen due to repeated excessive valgus stress and from increased load on the lateral compartment of the knee. Cooperman and colleagues studied 20 adults (32 hips) with hip dysplasia in order to determine the natural history of the disorder. At 22-year follow up, the majority (94%) had moderate osteoarthritis in the affected hip\textsuperscript{51}. 
6.2.7 Diagnosis in newborns and infants

Family history and clinical features are of great importance in the detection of unstable hips and hip dysplasia. A baby presenting with a family history of hip dysplasia, other malformations in the musculoskeletal system and/or a breech position should focus the physician on hip dysplasia.

Clinical examination

All newborns should be examined clinically in order to detect an unstable hip and it is recommended that the examinations continue until walking age. A dysplastic but stable hip reveals no clinical findings in the newborn period and has to be detected by ultrasound. Dislocated and unstable hips can be detected by careful clinical examination, and the diagnosis is confirmed by ultrasound.

Figure 4 The stability of the hip examined by Ortolani’s manoeuvre. A positive sign indicates that a dislocated hip is reduced.

Figure 5 Barlow’s manoeuvre is another test used to examine the stability of the hip. If the hip can be dislocated by manipulation, the test is positive.
Hip instability. The stability of the hip joint is examined in newborns by Ortolani’s and/or Barlow’s manoeuvre (Figure 4 and Figure 5). In a newborn with hip instability the tight fit between the femoral head and the acetabulum is lost. It is therefore possible to glide the femoral head in and out of the acetabular cavity. This gives a palpable sensation referred to as a positive Ortolani test when the hip is reduced. In a positive Barlow test the femoral head can be (sub)luxated by manipulation. The specificity for detecting an unstable hip by combining these tests is high (estimated to be approximately 98-99%)\textsuperscript{23,60}. The sensitivity varies, depending on the skill of the examiner, but experienced hands can have a sensitivity ranging from 65 to 97%\textsuperscript{181}. Range of motion. Reduced abduction of the hip in a newborn baby is highly suspicious of hip dislocation and this is the most important examination in children older than three months (Figure 6A). The baby is examined in supine position, with the pelvis in a neutral position. Normally >60° abduction is seen when the hip is flexed 90 degrees. Around 20-30% have bilateral dysplasia, which make the diagnosis more troublesome, as the asymmetry might be missing and the abduction is reduced in both sides. However, a total abduction of less than 120° indicates a possible bilateral hip dislocation. Unilateral reduced abduction can also be detected with the “heel-buttock test” (Figure 6B). In a normal hip the line between the sole of the feet and the gluteal cleft is continuous. In the case of unilateral reduced abduction, this line is interrupted and the feet deviate to the healthy side.

**Figure 6** Six-month-old girl with hip dislocation in the right hip presented with (A) reduced abduction and (B) a positive heel-buttock test.
Figure 7 Asymmetric skin folds in a six-month-old girl with hip dislocation in the right hip (A) and leg length discrepancy illustrated with a positive Galeazzi sign (B).

Asymmetry. Another sign indicating a dislocated hip can be asymmetric skin folds, which would reflect a unilateral dislocation of the hip (Figure 7A). However, asymmetric skin folds are also seen in 1/4 of children with normal hips.\textsuperscript{14} Leg length discrepancy may also indicate a dislocated hip. This finding may be evident by Galeazzi test (also called the Allis or Perkins test) performed with the infant supine, hips flexed to 90 degrees and knees flexed (Figure 7B). A lower knee level in one leg indicates a positive Galeazzi sign. The test is not specific for hip dysplasia, and other causes of congenital femoral length discrepancy must also be considered (e.g. hemihypertrophy, femoral hypoplasia, coxa vara).

Gait. After walking age (approximately 1 year), a child with a totally dislocated hip will often limp. This may be seen in both a unilateral and bilateral dislocation. In a unilateral dislocation the gait asymmetry is usually caused by leg-length discrepancy, resulting in gait changes and often toe walking on the affected side. Children affected bilaterally might be difficult to identify as asymmetry is lacking, but hyperlordosis and a waddling Trendelenburg gait are classic findings. The Trendelenburg sign is positive because the superiorly dislocated hip mechanically shortens the abductor
muscles and consequently decreases the muscle strength. These children may also present with widening of the perineum, symmetric limited abduction and short thigh segments relative to the child’s size.

**Ultrasonography**

Ultrasonography is an important supplement to clinical evaluation and an increasingly popular method since the late eighties (Figure 8). The newborn hip joint consists of a large proportion of cartilage and is therefore better visualised by ultrasound than by radiographs. Ultrasound waves are reflected by bony structures, but not by cartilage, and it is therefore possible to get an overview of both the femoral head and the acetabulum without exposing the child to ionising radiation. Ultrasonography is a demanding examination and accurate interpretation requires training and experience. However, performed by trained observers, ultrasonography is helpful in confirming findings from the clinical examination, evaluating patients with risk factors and making treatment decisions. An ultrasound examination includes a static and/or a dynamic evaluation of the hip. Static ultrasound images are used to assess the morphology of the largely cartilaginous hip joint, describing the acetabular inclination, the depth of the acetabulum and the location of the femoral head at rest. The dynamic assessment obtained during a modified Barlow’s test is used to determine hip stability. There is a strong association between hip morphology and stability, but morphologically normal hips may be unstable and vice versa.
Different ultrasound techniques have been described, and a combined technique assessing both acetabular morphology and hip stability is advised\textsuperscript{25,195}. Dr Graf started his studies on hip ultrasound in 1978, and described the standard coronal view through the centre of the acetabulum. Based on two angles, the acetabular inclination angle (\(\alpha\)) and the cartilage roof angle (\(\beta\)), he classified the hips into four main groups (\textbf{Figure 9})\textsuperscript{90,91}. However, Graf’s classification is based on both morphology and stability, and is thus rather imprecise. Furthermore, the indications and thresholds for treatment remain controversial. Alternative techniques have been established, such as Rosendahl’s method\textsuperscript{193}, by which hip morphology and stability are assessed separately (\textbf{Table 1}). Harcke\textsuperscript{96} and Novick\textsuperscript{168} have both reported on dynamic multiplanar approaches that focus on hip stability and the position of the femoral head relative to the acetabulum. The hip is accordingly classified as normal, subluxated, or dislocated. An alternative technique was proposed by Morin for assessment of the femoral head coverage\textsuperscript{158}. This method has been slightly modified by Dr Terjesen\textsuperscript{108,218}.
Table 1 Morphological classification of newborn hips assessed by ultrasound. Rosendahl's modification of Graf's method.

<table>
<thead>
<tr>
<th>Type</th>
<th>Description</th>
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<tbody>
<tr>
<td>Normal</td>
<td>Normal hip. A well-modelled bony acetabular roof with a sharp lateral edge and a narrow cartilaginous rim.</td>
</tr>
<tr>
<td></td>
<td>The alpha angle ≥ 60°</td>
</tr>
<tr>
<td>Immature</td>
<td>Adequately formed bony acetabular roof with a rounded lateral margin and a wide cartilaginous rim, i.e. a physiological retardation of the acetabular rim.</td>
</tr>
<tr>
<td></td>
<td>The alpha angle 50-59° (50° ≤ α &lt; 60°)</td>
</tr>
<tr>
<td>Dysplastic</td>
<td>Mild: Deficiently formed bony acetabular roof with a rounded to flattened lateral margin and a wide cartilaginous rim, i.e. a maturational deficit.</td>
</tr>
<tr>
<td></td>
<td>The alpha angle between 43° and 50° (43° ≤ α &lt; 50°). Corresponds to Graf type IIc</td>
</tr>
<tr>
<td></td>
<td>Severe: Poorly formed bony acetabular roof with a flattened lateral margin and a wide cartilaginous rim.</td>
</tr>
<tr>
<td></td>
<td>The alpha angle &lt; 43°</td>
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</table>

Radiographs
From 3-6 months of age the nucleus of the femoral head becomes visible in radiographs and this becomes a more valuable examination method. A dysplastic hip presents with a shallow acetabulum and in severe cases the femoral head is (sub)luxated. Different radiographic measurements are used to evaluate the dysplastic hip. One of the more common is the acetabular index. An increased acetabular index (mean ± 2SD) indicates a dysplastic acetabulum, but the angle decreases with age so the cut-off values are age-specific. A delayed appearance of the femoral ossific nucleus on the affected side, or dissimilar sizes of the nuclei, also indicates a dysplastic hip. Based on the radiograph, the hips can be classified as either 1) normal,
2) acetabular dysplasia (without subluxation), 3) subluxated with associated acetabular dysplasia, or 4) dislocated.

**CT/MRI**

CT and MRI are used on special occasions, but do not form part of the normal screening for hip dysplasia.

### 6.2.8 Diagnosis in adolescents and adults

Although most patients are diagnosed during the newborn period or early childhood, some have their diagnosis after skeletal maturity\textsuperscript{212}, particularly those with acetabular dysplasia only. Subjective symptoms such as pain/discomfort, limp and/or reduced range of motion are often present at diagnosis, but sometimes the dysplasia is diagnosed incidentally on the basis of radiographs taken for other reasons (e.g. abdominal radiographs where the hips are also visible).

**Clinical examination**

A good clinical examination is important both to diagnose and to monitor the condition. The first signs of hip dysplasia in young adults are typically hip/groin pain, reduced range of motion and/or a limp. In rare cases, clicking and popping may be presenting signs. It is important to note that these symptoms can also be caused by many other hip disorders. Initially, the symptoms may only appear after sport or other physical activity, but as the symptoms progress they may affect a person’s work, childcare, travel, etc. The onset of pain may be insidious, or start acutely after a period with increased activity. Pregnancy and weight gain may also cause a dysplastic hip to deteriorate.

**Range of motion.** The range of motion of both hips should be examined. A dysplastic hip will typically entail reduced abduction of the dysplastic joint. But the physical examination may also be normal, or with some discomfort at the extremes of ranges of motion, particularly abduction and internal rotation\textsuperscript{176}. 

Table 2 Different classification systems of hip dysplasia at skeletal maturity

<table>
<thead>
<tr>
<th>Grade</th>
<th>Crowe’s classification</th>
<th>Severin’s classification</th>
<th>Hartofilakidis’ classification</th>
</tr>
</thead>
<tbody>
<tr>
<td>I</td>
<td>Less than 50% subluxation</td>
<td>Normal appearance</td>
<td>Dysplasia</td>
</tr>
<tr>
<td>II</td>
<td>50-75% subluxation</td>
<td>Mild deformity</td>
<td>Low dislocation</td>
</tr>
<tr>
<td>III</td>
<td>75-100% subluxation</td>
<td>Dysplasia or moderate deformity of femoral head/neck or acetabulum.</td>
<td>High dislocation</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Wiberg’s CE angle&gt;20°</td>
</tr>
<tr>
<td>IV</td>
<td>More than 100% subluxation</td>
<td>Subluxation</td>
<td></td>
</tr>
<tr>
<td>V</td>
<td>False acetabulum</td>
<td></td>
<td></td>
</tr>
<tr>
<td>VI</td>
<td></td>
<td>Redislocation</td>
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on the basis of acetabular depth and femoral head location at skeletal maturity. Hartofilakidis has also proposed a scoring system assessing the acetabular abnormality and classifying the hips into three groups.\textsuperscript{98}

### 6.2.9 Treatment options

All treatment options aim to keep the femoral head located in the acetabulum, in order to secure a normal development of the hip joint. Starting treatment early leads to a better and more secure outcome. Most hip joints will develop normally if treatment is initiated before 6 months of age\textsuperscript{243}. A diagnosis after walking age requires more advanced treatment and may have a poorer outcome. In the first months of life, weeks of treatment will correspond to months of treatment for toddlers. The treatment options differ depending on the severity of the condition and the age of the patient at diagnosis.

The Frejka pillow is the standard treatment in Norway for hip instability in newborns

*Figure 10* The Frejka pillow is the standard treatment for newborns with hip dysplasia in Norway. The pillow is easy to remove and of little strain for the baby.
(Figure 10). This pillow comes in different sizes and facilitates flexion and abduction of both hip joints. The pillow should be used all day (22 hours per day) and is only removed during bathing or nappy changing. The standard treatment time is 3-12 weeks depending on the age of the child, but is prolonged if no satisfactory result is achieved. Norway is one of the few countries where the Frejka pillow is used. Worldwide, the Pavlik harness is more popular, while the von Rosen splint is used in Sweden. The efficiency of the Frejka pillow has been questioned, but favourable results regarding treatment efficiency and complication rate have been proven in Norwegian studies.\textsuperscript{36,104,216}

If the condition is diagnosed after 3 months of age and the abduction is only moderately reduced (>60°), an orthosis is the preferred treatment. In hips with abduction <60°, preoperative traction and reduction under general anaesthesia may be needed followed by a hip spica cast (Figure 11 & Figure 12). Often open reduction might be necessary for these hips. For children more than 18 months at diagnosis, surgical procedures are usually needed. Patients with hip dysplasia should be followed up in the outpatient clinic until the hip joint(s) have developed normally or at skeletally mature age. The treatment is more troublesome and prolonged with increasing age, and the long-term results are also more uncertain than with treatment at an earlier age. The goal of treatment of hip dysplasia is to have a hip as anatomically normal as possible by skeletal maturity.
Figure 12 Hip spica cast for a girl with hip dislocation in the right hip.

Unfortunately, this is not always achievable with only non-operative treatment. Studies have shown that surgery is needed by up to 5% of infants treated with abduction treatment. Osteotomies are needed in cases where the acetabulum is sloping and the coverage of the femoral head is poor. The osteotomies can be performed at the pelvic and/or the femoral side. A pelvic osteotomy aims to improve the socket. There are several different types of pelvic osteotomies (i.e. Salter, Dega, Pemberton) and their use depends on the shape of the acetabulum. In a Salter innominate osteotomy the pelvic bone is cut and the entire acetabulum is rotated into a better position on top of the femoral head using the symphysis as a hinge. This procedure is often used when the acetabulum is round, but not well placed on the top of the femoral head. This situation is more commonly seen when the hip has never been located in the acetabulum and the lateral edge of the socket is intact. The Dega osteotomy hinges the roof of the acetabulum down over the femoral head and is used when the acetabulum is too wide and shallow. The Pemberton osteotomy is similar, only differing in a slightly different final orientation of the acetabulum. In a proximal femoral varus osteotomy, the femoral head is tipped into the socket, redirecting the stress forces toward the middle of the acetabulum instead toward the lateral edge.
In young adults with residual hip dysplasia, a periacetabular osteotomy (PAO) is an option in which the acetabulum is completely re-orientated relative to the femoral head. This osteotomy is also called Ganz or Bernese osteotomy because it was developed by Professor Ganz in Berne, Switzerland. The procedure involves cutting the pelvis around the acetabulum and re-orientating the acetabulum into a better position. Joint replacement surgery is another option for severe hip dysplasia in adults. Total hip replacement and hip resurfacing have been used until recently (Figure 13). The results of total hip replacements after hip dysplasia are generally good72. In hip resurfacing, a metal cup is placed over the femoral head while a matching metal cup is placed in the acetabulum, but due to poor clinical results this method has now been abandoned183,205.

![Figure 13 Radiograph of a 32-year-old female with a total hip replacement on the left side due to hip dysplasia.](image-url)
6.3 OSTEOARTHRITIS OF THE HIP

Osteoarthritis (OA) of the hip is a condition caused by degeneration of the cartilage of the joint and is believed to be the most prevalent chronic joint disease. The prevalence increases with age, but the previous conception that OA was a normal consequence of aging has been disproven. The cause is multifactorial, including genetic predisposition, joint integrity, mechanical forces, local inflammation, and cellular and biomechanical processes. A combination of these factors results in cartilage loss and bony changes.

OA can be classified as either idiopathic or secondary. In a secondary OA, specific conditions cause or enhance the risk for developing OA. Such risk factors are fractures affecting the joint, childhood hip diseases (e.g. hip dysplasia, Perthes’ disease or slipped capital femoral epiphysis), and inflammatory joint conditions (rheumatoid arthritis and ankylosing spondylitis (Morbus Bechterew)). Age is another risk factor for OA. According to a Danish study, hip OA was found in 0.5-2% of the population below 60 years of age, and in 4-5% of the population of 60 years or more\(^\text{119}\). Females are more commonly operated with a total hip replacement, which indicates that the female sex is another risk factor. The reason for this is unclear, but it is probably partly related to hormones and genetics. Further, obesity is believed to increase the risk for OA\(^\text{77}\), but less consistent data has been reported on the relationship between hip OA and obesity than between knee/hand OA and obesity.

The severity of the OA can be classified according to different classification systems. Three well-known systems are the Tönnis Classification of Osteoarthritis by radiographic changes\(^\text{230}\), the Kellgren-Lawrence scale\(^\text{126,127}\) and the Croft classification\(^\text{52}\) (Table 3).

The treatment options for hip osteoarthritis depend on the patient’s symptoms, age and general health condition, in addition to the distribution of the arthritis. In general, it is advised to follow a treatment pyramid where all patients should be offered information and guidance on physical activity and in some cases on weight loss. Some would benefit from physical therapy, pain killers and/or non-steroidal anti-
### Table 3 Different classification systems for hip osteoarthritis

<table>
<thead>
<tr>
<th>Grade</th>
<th>Tönnis classification</th>
<th>Kellgren-Lawrence classification</th>
<th>Croft classification</th>
</tr>
</thead>
<tbody>
<tr>
<td>0</td>
<td>No signs of osteoarthritis</td>
<td>No changes of osteoarthritis</td>
<td>No changes of osteoarthritis</td>
</tr>
<tr>
<td>1</td>
<td>Slight narrowing of joint space, slight lipping at joint margin, slight sclerosis of femoral head or acetabulum</td>
<td>Possible narrowing of joint space medially and possible osteophytes around femoral head</td>
<td>Osteophytosis only</td>
</tr>
<tr>
<td>2</td>
<td>Small cysts in femoral head or acetabulum, increased narrowing of joint space, moderate loss of sphericity of femoral head</td>
<td>Definite narrowing of joint space inferiorly, definite osteophytes and slight sclerosis</td>
<td>Joint space narrowing only</td>
</tr>
<tr>
<td>3</td>
<td>Large cysts, severe narrowing or obliteration of joint space, severe deformity of femoral head, avascular necrosis</td>
<td>Marked narrowing of joint space, slight osteophytes, some sclerosis and cyst formation and deformity of femoral head and acetabulum</td>
<td>Two of osteophytosis, joint space narrowing, subchondral sclerosis and cyst formation.</td>
</tr>
<tr>
<td>4</td>
<td></td>
<td>Gross loss of joint space with sclerosis and cysts, marked deformity of femoral head and acetabulum and large osteophytes.</td>
<td>Three of osteophytosis, joint space narrowing, subchondral sclerosis, and cyst formation.</td>
</tr>
<tr>
<td>5</td>
<td></td>
<td></td>
<td>As in grade 4, but with deformity of femoral head</td>
</tr>
</tbody>
</table>

Inflammatory drugs (NSAIDs). The minority, who do not achieve acceptable results from such advice and treatment, should be offered a total hip replacement procedure. Surgery is an effective treatment regarding pain and functionality for many patients, but being an operative treatment, with its complications and cost, it should always be a deliberated decision. Reticence should be shown regarding young patients, as a revision is likely to be necessary due to the limited life expectancy of the prosthesis and a revision is a more complicated procedure than the primary operation. In Norway, all total hip arthroplasties performed since 1987 are reported to the Norwegian Arthroplasty Register.
6.4 THE NORWEGIAN ARTHROPLASTY REGISTER

The Norwegian Arthroplasty Register (NAR) has since September 15, 1987 registered all total hip replacements (THR) performed in Norway. The main reason for establishing the register was the introduction of many new hip implants without documentation from clinical studies in the 1970s. Several of the implants were in use for more than 10 years before they were identified with high failure rates. The main purpose of the register is to function as a surveillance tool to identify inferior implants as early as possible.

From 1994 the register was extended to include registration of all joint replacements and in 2004 the Norwegian Cruciate Ligament Register, in 2005 the Norwegian Hip Fracture Register, and in 2010 the Paediatric Hip Register were included. The register is owned by The Norwegian Orthopaedic Association, and receives funding from the Western Norway Regional Health Authority and Haukeland University Hospital. In 2002 the Norwegian Ministry of Health approved the register as a national centre of excellence of joint replacements.

A hip prosthesis operation is reported to the register by the surgeon, who completes a one-page standard form (Appendix 3). The form includes the identity of the patient, the date of the operation, and the reason for surgery as given by the surgeon. Both primary operations and reoperations are recorded, and by using the patients’ national personal identification number, the revisions can be linked to their primary operation.

Reporting to the NAR is not compulsory, but it has been estimated that at least 97% of joint replacements performed in Norway are reported. All patients give written consent to be included in the register. The register staff includes orthopaedic surgeons, statisticians, informatics specialists and secretaries.
6.5 THE MEDICAL BIRTH REGISTRY OF NORWAY

The Medical Birth Registry of Norway (MBRN) was established in 1967 and collects information about all births in Norway, including stillbirths after 12 weeks of pregnancy. Reporting to the MBRN is compulsory. The MBRN is a department of the Norwegian Institute of Public Health, which owns and administers the registry.

Information for the register is reported on a standard form. The identity of the child’s parents, complications during pregnancy or birth, child’s health (including hip instability), maternal health, and maternal occupation and smoking habits are recorded. In December 1998, the original registration form from 1967 (Appendix 1) was replaced by a new one (Appendix 2). At the same time the register changed its coding system from the International Classification of Diseases Version 8 (ICD-8) to Version 10 (ICD-10).

Information regarding hip instability has been recorded since commencement. ICD-8 codes reflecting hip instability were “dislocation of the hip” (755.6), “dysplasia of the hip” (755.7), and “positive Ortolani test” (778.5). The new form introduced in 1998 included a box for “dysplasia of the hip treated with Frejka pillow”. Other diagnoses in the category of congenital abnormalities of the hip (Q65.0–Q65.9) were also recorded. An important limitation of the register is that for patients diagnosed with unstable hips, the form does not record whether the left, the right, or both hips are affected.

6.6 THE 1989 BERGEN BIRTH COHORT STUDY

The 1989 Bergen Birth Cohort study consists of two parts. The first part is a randomised clinical trial on different screening strategies for hip dysplasia in children born at Haukeland University Hospital in the period January 1988 to June 1990 (The 1988-1990 Hip Cohort). The second part is a maturity review of the same population and was performed in the period February 2007 to March 2009 (The 1989 Hip Project).
6.6.1 The 1988-1990 Hip Cohort

The 1989 Bergen Birth Cohort initially comprised all newborns delivered at Haukeland University Hospital (HUS) in Bergen between January 1988 and June 1990. Those with birth weight <1500 grams, with severe malformations/disease or who died within the first month of life were excluded (n=103). A total of 11,925 newborn infants (>99.5% Caucasians) were enrolled in the randomised controlled trial (RCT) designed to evaluate the effectiveness of three different screening strategies for hip dysplasia: universal ultrasound screening (n=3,613), selective ultrasound screening (n=4,388) and clinical screening alone (n=3,924) (Figure 14). The randomisation procedure was area-based as three equally sized newborn nursery units separated from the delivery ward were in use at that time. Newborns admitted to unit one and certain parts of unit three were assigned to the selective group, and newborns admitted to unit two and the rest of unit three were assigned to universal screening. Infants born when ultrasound was not available constituted the clinical screening only group (no ultrasound). All infants received a detailed newborn clinical examination, including assessments of joint laxity and hip stability, and known risk factors for hip dysplasia (breech presentation at delivery, and/or family history (first or second degree)) were elicited and recorded. High-risk infants from the selectively screened group and all infants from the universally screened group were offered a single examiner hip ultrasound (Rosendahl’s method). Indications for treatment were dislocatable/dislocated hips or severe sonographic dysplasia irrespective of clinical or sonographic stability. Hips with a mildly dysplastic morphology (43° ≤ α <50°) were treated if they were also clinically or sonographically dislocatable/dislocated. If the hips were sonographically immature (50° ≤ α <60°) or mildly dysplastic (43° ≤ α <50°), but clinically stable, they had clinical and sonographic surveillance every fourth week until normalisation or until treatment was initiated due to lack of improvement. Babies with normal hips but with risk factors for hip dysplasia were referred for a radiograph of the hips at 4-5 months of age. Additionally, all children in Norway are regularly clinically examined during their first two years of life as part of the national health programme. Follow-up clinical and
Figure 14 Flow of participants in the 1989 Bergen Hip Cohort Study.
HUS: Haukeland University Hospital; US: ultrasound; RCT: Randomised Clinical Trial; N: Number
radiological data on those diagnosed as having hip dysplasia or other hip disorders were collected on subjects at all ages.

Outcome measures for the RCT were 1) rates of late-detected dysplasia (age >1 month) as defined by the acetabular index of Tönnis (AI)\textsuperscript{229} more than 2 standard deviations above mean for the age, and the femoral head position\textsuperscript{108,218} 2) ultrasound follow-up and 3) treatment rates. The RCT found that universal ultrasound screening tends to reduce the prevalence of late-detected hip dysplasia (0.3 per 1009), as compared to selective ultrasound examination (0.7 per 1000) and clinical examination alone (1.3 per 1000), though a statistically significant reduction was not found (p=0.11, test for trend). Treatment rates were, however, higher in the universal group (3.4%) than in the selective group (2.0%) and clinical screening group (1.8%) (p>0.001), and also resulted in a higher follow-up rate (13% vs. 1.8% and 0%).

### 6.6.2 The 1989 Hip Project

In the period February 2007 to March 2009, all those born at Haukeland University Hospital in 1989 (n=5068), as well as those born in 1988 and 1990 in whom neonatal hip abnormalities were detected (n=569), giving a sample of 5637, were invited to a follow-up study of their hips. Persons who were dead, had emigrated or were living outside a defined area of Bergen and surroundings were excluded (n=1128). The area was defined as the catchment area of Haukeland University Hospital, which included Bergen, Samnanger, Os, Austevoll, Sund, Fjell, Askøy, Osterøy, Meland, Øygarden, Radøy, Lindås, Austrheim, Fedje and Masfjorden. The remaining cohort was invited to participate in a long-term clinical and radiological follow-up of their hips (Figure 14). A total of 2390 subjects (53%) accepted and were included.

All subjects were approached by letter and invited to participate in the follow-up study (Appendix 6). In the invitation letter, the subjects were given time and date for an appointment at the Department of Paediatric Radiology, Haukeland University Hospital, where all the examinations were performed. They were contacted by phone
and/or SMS as a reminder 1-3 days prior to their appointments. All participants gave
written consent (Appendix 7).

**Questionnaires**

A questionnaire was attached to the invitation letter which participants were asked to
complete prior to the appointment. This contained questions regarding hip problems
in childhood, hip problems in the family and their parents’ height (Appendix 8). A
second questionnaire was filled in on arrival which consisted of three parts, including
standardised questionnaires on quality of life (EQ-5D ([www.euroqol.org](http://www.euroqol.org)) and hip
problems (WOMAC ([www.womac.org](http://www.womac.org))) (Appendix 9).

**Radiological examinations**

The radiological examinations were performed by a single specially trained
radiographer using a low-dose digital radiography technique (DigitalDiagnost
System, version 1.5, Philips Medical Systems, Hamburg, Germany). A strictly
standardised protocol was followed. Boys were offered a gonadal shield. Females,

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*Figure 15* Two radiographs were obtained: (A) one erect anteroposterior view and (B) one
supine frog-leg view.
who were pregnant or with an uncertain pregnancy status were excluded from the radiological examinations. Two standardised views were obtained, one weight-bearing anteroposterior (AP) view and one supine frog-leg view (Figure 15).

The AP view was taken with hips kept in a neutral abduction-adduction position, toes pointing forwards. The film-focus distance was 1.2 m and centred 2 cm proximal to the symphysis. For the frog-leg view it was centred at the symphysis. The radiographer ensured correct posture during the exposures. The total mean radiation dose for both the AP and the frog-leg view together was 0.5 Gycm².

**Clinical examination**
The clinical examination (Figure 16) was performed by one of five physicians, blinded to the results of the questionnaire and radiographs. One experienced senior orthopaedic surgeon (LBE) standardised the clinical examination and trained the four less experienced physicians. A standardised protocol including height, weight,

![Figure 16 Clinical examination of the hip: (A) external rotation, (B) internal rotation, (C) abduction, and (D) extension.](image)
assessment of hip range of motion (ROM) and Beighton score for joint laxity was performed (Appendix 10). Flexion, abduction and adduction were measured with the subject supine, while extension, internal and external rotation were measured prone with 90 degrees flexion of the knee joint.

**Genetic sample**

All participants were asked to provide salivary DNA. This was optional and a separate consent form was signed. Participants attending after September 1, 2007 were invited to provide a sample at the clinical visit. Those who already had met for the follow-up when the collection of DNA started were requested by post to return salivary DNA. Two ml of saliva was collected using Oragene DNA self-collection kits (DNA Genotek Inc., Ontario, Canada). The specimens were frozen and are ready for later genetic analysis.

**Data from Child Health Clinics**

Height and weight are routinely measured in all children in Bergen at one of the 26 child health clinics in the city. Weight is measured at 6 weeks and in most children at 4, 5, 6, 10, 11 and 18 months and at 2, 4, 6, 8 and 12 years. Height (length) is measured at 6 and 18 months and at 2, 5-6, 8 and 12 years. These data were recorded on paper, but have been digitised in a project related to the 1989 Hip Project.
7. AIMS OF THE STUDY

The overall objective of this thesis was to investigate hip dysplasia among young adults in Norway.

The specific aims of the four papers included in the thesis were:

I To investigate whether hip instability at birth predisposes to a total hip replacement in young adulthood.

II To validate the dysplasia diagnosis reported to the Norwegian Arthroplasty Register for patients born after 1967 and also to report on age at diagnosis of dysplasia for this patient group, previous treatment and quality of life.

III To validate radiographic measurements relevant for hip dysplasia at skeletal maturity, using both a manual and a digital measurement technique.

IV To report on the prevalence of radiological measurements indicating hip dysplasia among 19-year old Norwegians.
8. METHODS

8.1 DATA

This thesis is based on registry data from the Norwegian Arthroplasty Register (NAR) (Papers I & II), the Medical Birth Registry of Norway (MBRN) (Paper I), and clinical data from the 1989 Hip Project (Papers III & IV).

8.1.1 Paper I

In the first paper, we linked the Norwegian Arthroplasty Register with another national register, the Medical Birth Registry of Norway, by using the unique, 11-digit national personal identification number (Figure 17). Information on neonatal hip stability in the newborn is recorded in the Birth Registry. This information was combined with information from the Arthroplasty Register and it was studied whether an unstable hip predisposed for a total hip replacement or not. In total, 442 patients with 633 total hip arthroplasties were recorded in both registries.

Figure 17 The two national registries, the Norwegian Arthroplasty Register (NAR) and the Medical Birth Registry of Norway (MBRN), were linked using the national personal identification number. In total 442 persons were recorded in both registries.
8.1.2 Paper II

In the second paper, the same population as described in paper I was studied, i.e. those born after 1967 and registered with a total hip replacement in the Norwegian Arthroplasty Register. This cohort was contacted by post and invited to participate in a follow-up study (Appendix 4). The patients were asked whether they agreed on the diagnosis reported to the register. They were also asked to fill in and return a questionnaire, and we asked for consent to review their hospital medical records regarding their hip problems. For patients who consented, the medical records were retrieved from the hospital and reviewed in order to validate the diagnoses reported to the Arthroplasty Register. For patients diagnosed with hip dysplasia, Calvé-Legg-Perthes’ disease (CLP), or a slipped capital femoral epiphysis (SCFE), we also recorded data and symptoms at the time of diagnosis of the hip disease and any treatment given for the hip disorder prior to their total hip replacement (Appendix 5).

8.1.3 Paper III

The third study comprised 95 conventional AP pelvic radiographs from participants in the 1989 Hip Project. All images were measured both in a digital measurement programme (Adult-DDH, Iowa Hospital and Clinics, Iowa, USA) and in the standard manner in AgfaWeb1000. This study was performed to validate the new digital measurement programme. Two observers measured all images twice digitally and twice manually. One observer measured the images twice digitally and once manually. In total, all images were measured 11 times.

8.1.4 Paper IV

All participants in the fourth study were born during 1989 and included in the 1989 Hip Project with questionnaires, clinical examination and two radiographs (n=2072). The radiographs were measured by one of three observers using the digital
measurement programme (Adult-DDH). Radiographic measurements relevant for hip dysplasia at skeletal maturity were used to classify the radiographs and to group the hips into either “normal”, “borderline” or “dysplastic”. The radiographic findings were related to body mass index (BMI), range of motion of the hips, Beighton score, EQ-5D score and WOMAC score.

8.2 OUTCOME MEASURES

8.2.1 Radiographic parameters associated with hip dysplasia at skeletal maturity

Several different radiographic measurements have been proposed to define hip dysplasia, but there is no consensus on what markers with corresponding cut-off values to use. The markers can be grouped into three different categories according to what they are designed to measure.

*Acetabular anatomy (Figure 18)*

**Sharp’s acetabular angle.** This is defined as the angle between the horizontal teardrop line and a line through the inferior teardrop point and the lateral rim of the acetabulum\textsuperscript{201}. Synonyms used in the literature are “the acetabular angle” and “angle of inclination”.

\[ ADR = \left(\frac{A}{B}\right) \times 1000 \]

*Figure 18 The acetabular anatomy is assessed by Sharp's angle, the acetabular roof angle of Tönnis (AA) and the acetabular depth-width ratio (ADR).*
The acetabular roof angle of Tönnis (AA). This is the angle between a line parallel to the horizontal teardrop line and a line drawn through the medial point of the sourcil and the lateral acetabular rim\textsuperscript{227,229,231}. In the literature, synonymous names are “horizontal toit externe” (HTE) (Toit: French for roof), acetabular roof obliquity (ARO) and acetabular index (AI).

The acetabular depth-width ratio (ADR). The ratio of the distance between the inferior teardrop point and the lateral acetabular rim, and the depth of the acetabulum, multiplied by 100\textsuperscript{51,212}.

Position of the femoral head relative to the acetabular cavity (Figure 19)

Wiberg’s Centre-Edge (CE) angle. This is defined as a line through the centre of the femoral head, perpendicular to the horizontal teardrop line, and a line running from the centre of the femoral head through the lateral acetabular edge\textsuperscript{245}.

Ogata’s angle. This is a modification of Wiberg’s CE angle, differing only slightly from this as Ogata’s angle uses the lateral edge of the sourcil instead of the lateral acetabular edge\textsuperscript{169}.

Femoral head extrusion index (FHEI). This is defined as the percentage of the femoral head lying medial to the lateral acetabular edge\textsuperscript{100}. Some authors also describe the opposite, i.e. the percentage of the femoral head lying laterally to the

\[
\text{FHEI} = \frac{A}{B} \times 100
\]

Figure 19 The position of the femoral head relative to the acetabular cavity is defined using Wiberg’s Centre-Edge (CE) angle, a modification of this (Ogata’s angle) and the femoral head extrusion index FHEI.
acetabular edge\textsuperscript{155}, described as the “migration index”\textsuperscript{120}. The FHEI is also referred to as the “acetabular head index”\textsuperscript{141}.

Other measurements (Figure 20)
The maximum width of the teardrop. This is a landmark seen in the AP view\textsuperscript{6}. Its medial border consists of the cortical surface of the pelvis, and its lateral border consists of the cortical surface of the middle third of the acetabular fossa. The inter-teardrop line, connecting the inferior tip of both teardrops, was used as the transverse axis of the pelvis.

The joint space width (JSW) medially, laterally and in the middle. The JSW is a well-accepted method for quantitative assessment of OA\textsuperscript{8,84,120,139}. We measured the JSW at three locations, namely medially, in the middle and laterally.

The articulo-trochanteric distance (ATD). This can be assessed in two different ways. One method defines it as the distance between the tangents normal to the long axis of the femur to the superior margin of the trochanter major and to the superior contour of the femoral head\textsuperscript{67,137,172}. Another method assesses the ATD as the distance between a horizontal line between the superior margin of the trochanter on both sides and a perpendicular to this line to the superior margin of the femoral head.

\[\text{Figure 20} \text{ The joint space width (JSW) was measured in three places; medial, middle and lateral. Other measurements are the maximum width of the teardrop and the articulo-trochanteric distance (ATD).}\]
In a population with predominantly normal hips and where the radiographs are taken with approximately parallel femurs, there is only a minor difference in these two measuring techniques.

**Sourcil shape.** The “sourcil cotyloïdien” (Sourcil: French for eyebrow) represents the weight-bearing bony area of the hip joint, seen as a hyper-dense arched line along the acetabular roof. In a normal hip joint, this line is horizontal or somewhat curving downward laterally, whereas it has an upward orientation in the dysplastic hip. The lateral edge of the roof is often located more laterally than the lateral point of the sourcil.169

### 8.2.2 EQ-5D

Quality of life was assessed in papers II and IV. We used the EuroQol instrument ([www.euroqol.org](http://www.euroqol.org)), which is designed to describe and evaluate health-related quality of life. The instrument consists of a health status part (EQ-5DTM) and a VAS score (EQ-VAS).

The EQ-5D is a descriptive tool assessing five dimensions: level of mobility, self-care, usual activities, pain/discomfort and anxiety/depression. Each question has three levels: no problems, some problems, and severe problems. A single weighted utility score – the EQ-5Dindex – is calculated from the 5 dimensions. The “best imaginable health state” represents individuals who report that they are experiencing the highest level of functioning for the five conditions. The EQ-5Dindex for these patients is 100. Death scores zero, but conditions worse than death (<0) are possible. Each of the five categories has an individual weighting with pain and mobility having the highest weighting.

The EQ-VAS is a 20 cm vertical visual analogue scale ranging from 0 (signifying worst possible health) to 100 (signifying best possible health). The patients are asked to mark their current health situation on this scale.
8.2.3 WOMAC Osteoarthritis Index

The Western Ontario and McMaster (WOMAC) Osteoarthritis Index is used in paper IV of this thesis. The instrument is self-administered and uses a battery of 24 questions to assess the three dimensions of pain (5 questions), disability (17 questions), and joint stiffness (2 questions) in patients with hip and knee osteoarthritis (www.womac.org). All questions have the same five descriptors: none, mild, moderate, severe, and extreme. These correspond to an ordinal scale of 0-4. The scores are summed for the items in each subscale, with possible ranges as follows: pain=0-20, disability=0-68 and joint stiffness=0-8. Most commonly, a total WOMAC score is created by summing the items for all three subscales, but other methods of aggregating subscales have been used. With this method the maximum score is 96 and an increasing score indicates worse pain, stiffness and functional limitations.

8.2.4 The Beighton score

The Beighton score was first described in 1973 by Beighton and colleagues\(^\text{25}\) and was a modification of a six-level (0-5) scoring system described by Carter and Wilkinson in 1964 where they examined five joints\(^\text{46}\). Beighton modified two of these joints tests to include hyperextension (>10°) of knees and elbows, passively touching the forearm with the thumb, dorsiflexion of the fifth finger more than 90 degrees (Gorling’s sign), and flexion of the trunk (touching the floor with the palms of the hands without bending the knees). The latter manoeuvre was included in a later edition, replacing a test of dorsiflexion of the ankle with eversion of the foot (Figure 21). The range of the Beighton score changed into a 10-grade system (0-9) and no cut-off values were determined. A cut-off value of 4 or more is often used in the literature, but as claimed by some authors, this value is probably too low as too many are then defined with joint hypermobility\(^\text{121}\). It has also been discussed whether to use different cut-off values for different age groups.
The Beighton test assesses joint hypermobility: 1) touching the floor with the palms of the hands with the knees straight, 2) hyperextension of the knees, 3) hyperextension of the elbows, 4) touching the forearm with the thumb, and 5) dorsiflexion of the fifth finger more than 90 degrees.

8.3 DIGITAL MEASUREMENT PROGRAMME

Several radiographic markers are of interest when judging whether a hip is dysplastic or not. The time-consuming measurement process of many of these markers partly explains why a digital measurement programme for hip dysplasia has been developed at the University of Iowa Hospital and Clinics, Iowa City, United States (DDH_Adult).
The programme uses anteroposterior pelvic radiographs stored as DICOM files. The radiographs are opened in the programme and 46 specific landmarks are marked in a subsequent order (Figure 22). When finishing an image, the programme asks if you want to correct any of your landmarks before the image is stored and the radiographic measurements are automatically calculated and added to an Excel file.

Eleven radiographic markers all relevant for hip dysplasia at skeletal maturity are included in the programme. These are Wiberg’s, Sharp’s and Ogata’s angles, acetabular roof angle of Tönnis, femoral head extrusion index (FHEI), maximal width of the teardrop, acetabular depth-width ratio (ADR), articulo-trochanteric distance (ATD), and maximal joint space distance in three different locations (medial, middle and lateral). Calculations from the digital measurement programme can also be linked to a stress programme developed in Ljubljana, Slovenia.

Figure 22 The digital measurement programme

8.4 STATISTICAL ANALYSES

In all studies, numerical variables were expressed as median and range, mean and standard deviation or mean and range as appropriate. Categorical variables were described as relative frequencies.

In paper I, 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 end of study was considered to be a censored observation. Cox regression analysis was used to calculate risk estimates for undergoing total hip replacement at young age. We adjusted for sex, neonatal hip instability (as registered in the Medical Birth Registry of Norway) and time period.

In papers II and IV, comparison of mean values was made using t-tests, where a p-value of <0.05 indicated a statistically significant difference.

In paper III, the Bland-Altman method was used to examine the mean difference between the measurements\(^{32,34,35}\) with the corresponding standard deviation of the difference distribution. When we had multiple readings for one method/observer, we first calculated the mean for each method/observer on each subject and used these pairs of means to compare the two methods/observers. In these cases the standard deviation had to be recalculated as the standard deviation of the differences was too small, because some effect of the repeated measurement error had been removed. The correct standard deviation of differences was given by \(\sqrt{S_0^2 + (1 - \frac{1}{m_1})S_1^2 + (1 - \frac{1}{m_2})S_2^2}\), where \(S_0\) represented the standard deviation of the mean difference between the methods/observers, \(S_1\) and \(S_2\) the within-subject standard deviation for each method separately, and \(m_1\) and \(m_2\) the number of observations on each subject by the respective methods/observers. The 95% limits of agreement were estimated as the mean difference between the two measurements ± 1.96 standard deviations (SD). The assumption that the agreement was similar over the range of measurements was checked by plotting the differences against the mean of the two methods.

In addition to the Bland-Altman method, intra-class correlation coefficient (ICC) and minimal detectable change (MDC) were calculated in paper III. The ICC was calculated using a one-way random effect ANOVA table (formula ICC (1))\(^{156}\). For each observer, the inter-method reliability was also expressed in terms of ICC calculated using two-way random effect ANOVA table (formula ICC (A,1))\(^{156}\). The ANOVA tables used for ICC indices were used to calculate MDC as indicated by de Vet and colleagues\(^{58}\).
8.4.1 Statistical software

Most analyses were performed using the statistical software programmes SPSS (SPSS Inc. Chicago, IL, USA) Version 14.0 (Paper I), SPSS Version 17.0 (Paper II), and PASW Statistics Version 18.0 (Paper III and IV). Additionally, S-Plus 6.2 (Insightful Corp., Seattle, WA, USA) and Stata Statistical Software: Release 11 (StataCorp. 2009, College Station, TX: StataCorp LP) were used in study I, and studies III and IV, respectively.

8.5 APPROVALS

The linking of the Norwegian Arthroplasty Register (NAR) with the Medical Birth Registry of Norway (MBRN) was approved by the Regional Committee for Medical and Health Research Ethics, Western Norway and the Norwegian Data Inspectorate (No. 10485). The licensing to the Norwegian Arthroplasty Register allows validation of the recorded data, including the diagnoses. Additionally, the participants in paper II gave a written informed consent that their hospital medical records regarding their hip disease could be reviewed.

The 1989 Bergen Birth Cohort Study (including the 1989 Hip Project) was approved by the Regional Committee for Medical and Health Research Ethics, Western Norway (No. 018.06 and No. 003.07) and the Norwegian Data Inspectorate (No. 14124 and 15925). All participants in the 1989 Hip Project provided written informed consent according to the Helsinki declaration. Nine subjects presenting with uncertain or severe clinical and/or radiographic findings related to hip, back or pelvic pathology were immediately scheduled for a radiological follow-up consultation or for a consultation with a senior paediatric orthopaedic professor as appropriate. The Regional Ethics Committee also approved further analyses regarding non-responders.
9. SUMMARY OF PAPERS I-IV

The main results from the four papers (I, II, III and IV) are presented below.

9.1 PAPER I

A total of 2,218,596 newborns were registered in the Medical Birth Registry of Norway (MBRN) between 1967 and 2004. Of these, 19,432 (0.88%) were reported to have unstable hips at birth. The number of THR's registered in the Arthroplasty Register between 1987 and 2004 was 106,716, but only 633 of these (both primary operations and revisions) had been performed in the 442 patients (96 patients operated bilaterally) born after January 1, 1967, and were thus included in both the MBRN and the NAR. Of these 442 individuals, 2.5% (95% CI: 1.4–4.7) were born with NHI 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). The absolute risks were, however, low: 57 (95% CI: 30–105) in $10^5$ (11 of 19,421) for children with NHI and 20 (95% CI: 18–22) in $10^5$ (431 of 2,198,733) for children without unstable hips at birth. Of the 442 patients reported to the NAR, only 11 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.

9.2 PAPER II

Hip dysplasia accounted for 163 (26%) of the 634 hip replacements recorded in 540 patients born after January 1, 1967 and registered in the Norwegian Arthroplasty Register. Extended medical information was retrieved for 150 hips (13 missing). Of these 150 THR’s, the dysplasia diagnosis was confirmed by medical notes in 88% (132 hips, 114 patients).
Median age at the time of diagnosis of hip dysplasia was 7.8 (0–39) years: 4.4 years for females and 22 for males, with the more common symptoms/findings at presentation being limp (25%), hip pain (20%), and reduced hip abduction (18%).

Data on previous hip-preserving treatment (both surgery and non-operative treatment) were retrieved for 88 of the 132 confirmed dysplastic hips (67%). Previous surgery was reported for 81 of the 88 hips, femur osteotomy being the most common and seen in 63 (36%) of 176 surgical procedures (more than one procedure during each operation was possible). In 10 hips (9 patients, 12% of the total) early abduction treatment with a Frejka pillow was given, but all of them were in need of hip preserving surgery later, i.e. none of those reported with a THR had only been treated with the Frejka pillow.

The mean EQ-5D index for men (score = 74) was slightly better than for women (score = 67), but the difference was not statistically significant (p = 0.2). The self-reported health situation for patients who were operated due to DDH (score = 68) was worse than that reported for the general population aged from 18 to 39 years in Sweden (score = 86, p < 0.001) and in the United Kingdom (score = 87, p < 0.001).

9.3 PAPER III

A total of 95 radiographs were measured manually in AgfaWeb1000 (on 5 occasions) and in a digital measurement programme (on 6 occasions) by three observers independently. Eleven radiological measurements all relevant for hip dysplasia were evaluated. Large variations among the different radiological measurements were found. However, the variation was not related to the use of manual or a digital measuring technique. For measurements with greater absolute values (Sharp’s angle, femoral head extrusion index and acetabular depth-width ratio) the inter- and intra-observer and inter-method agreements were better than for measurements with lower absolute values (acetabular roof angle, teardrop and joint space width).

Measuring time was shorter for the digital technique (approximately 2 min) than for the manual technique (approximately 5 min 30 sec).
9.4 PAPER IV

The study aimed to report on the prevalence of radiological features associated with hip dysplasia in a population of 2081 19-year-old Norwegians. Hip dysplasia was defined by Sharp’s angle (>45°), acetabular depth-width ratio (ADR) (<250), Wiberg’s CE angle (<20°), femoral head extrusion index (FHEI)) (<75%) or a dysplastic shape of the sourcil judged subjectively. The proportion of dysplastic hips in our cohort varied depending on the radiological measurements applied, ranging from 1.7% to 20%. All parameters except ADR were more commonly seen among females than males. Wiberg’s CE angle <20° was found in 3.3% of the cohort; 4.3% in females and 2.4% in males. By using a cut-off value for Wiberg’s CE angle of 25°, 20% of the cohort was defined with a pathological angle: 23% of the females and 16% in males. A pathological Sharp’s angle was common among both females (19%) and males (6.4%).
10. GENERAL DISCUSSION

10.1 METHODOLOGICAL CONSIDERATIONS

10.1.1 Study design

Medical research is conducted in order to reveal new information on health and diseases. The different analytical approaches can basically be categorised into two groups: experimental studies and observational studies. In experimental studies the researcher affects (controls) what happens to all or some of the individuals. In an observational study the researcher collects information about one or more groups of subjects, but does not influence events. Further, the study design can be classified as either prospective or retrospective, or as longitudinal or cross-sectional (Figure 23). The most appropriate design for the study will depend upon what is being studied, e.g. occurrences, causations, diagnostics or prognoses.

Experimental studies include randomised controlled trials (RCT) and interventional studies. RCTs are often claimed to be the gold standard in methods to evaluate healthcare interventions. If correctly designed and with adequate power, they are the best method for comparative trials and they eliminate the problem of bias. Known and unknown prognostic factors are equal in the two groups compared, and blinding is possible in some cases. But RCTs are expensive and require considerable time for

Figure 23 Types of research design. From Altman DG. Practical Statistics for Medical research. Chapman & Hall 1991.
both planning and implementation. The study populations are often highly selective and the trials may use only selected, specialised study centres. In some cases industry sponsors the trials, with the results that studies with economic interests are performed more frequently. There may be surrogate endpoints and sometimes the RCTs are prematurely discontinued due to slow recruitment or economic reasons. Furthermore, RCTs are not possible for all research questions.

Observational studies include cross-sectional studies, case-control studies and cohort studies. Observational studies have lower costs than RCTs, greater timeliness, and a broader range of patients. In cross-sectional studies, hypotheses are studied in a cross-section of the population. This gives a picture of the current information, but cannot explain causes. Case-control studies are retrospective, as “cases” (disease) are compared with “controls” (healthy) in order to detect differences. This study design is suitable for rare conditions, but is influenced by bias and care must be experienced in choosing appropriate “controls”. A cohort study is prospective, comparing a group with a specific characteristic/exposure with a group without this characteristic/exposure. Such studies are often demanding and expensive.

Paper I is a prospective cohort study of all persons registered in the Medical Birth Registry of Norway (MBRN) in the period 1967-2004. As newborns they were categorised according to whether they had instable hips (NHI) or not. Reporting to the registry is compulsory. The MBRN data file was linked to the Norwegian Arthroplasty Register (NAR) using the unique national identity code, and a primary total hip replacement (THR) was used as an endpoint. Reporting to the NAR is not compulsory, but it has been estimated that at least 97% of joint replacements and 98% of THRs performed in Norway are reported. The high reporting reduces the risk of selection bias in this study. Regarding information bias, neither the dysplasia diagnosis in the MBRN nor in the NAR was validated at this time. The definition of hip instability registered in the MBRN may vary by both hospital and physician. This should therefore be considered when the results are interpreted. Classification bias was minimised since hip instability was evaluated regardless of the outcome (whether the hip needed a THR in the future or not). A THR was used as the endpoint and is an
objective outcome; however, the indications for a THR procedure may be evaluated differently. The risk estimates for undergoing THR at a young age were adjusted for sex, NHI and time period in order to reduce the risk of confounders.

In paper II, a retrospective design was used with a cross-sectional follow-up. All patients under the age of 40 registered with a total hip replacement in the Norwegian Arthroplasty Register were contacted by post and asked to fill in a questionnaire. The non-responders differed from the responders regarding gender, as females were more prone to answer. The same tendency has been demonstrated in other studies. The questionnaires regarding their initial hip disease introduce the problem of recall bias. However, the only retrospective information used in the paper was whether they confirmed the diagnosis resulting in a total hip replacement as registered in the NAR. For all patients with childhood hip diseases, this was also checked with information in medical records of the respective hospitals. Additionally, the medical records for e.g. patients with hip dysplasia were reviewed to find age at diagnosis. Also in this paper, the high rate of reporting to the NAR reduces the risk of selection bias. But 32 of 572 patients (5.6%) did not permit review of their medical records and these patients were excluded from the study. Five of the 32 patients (15%) had their THR due to hip dysplasia, but the corresponding percentage of THR due to hip dysplasia in the cohort was 26%. This indicates that fewer of the dysplasia patients were excluded as compared to patients with other diagnoses, minimalising the risk of selection bias.

In paper III we aimed to test the inter- and intra-observer and inter-method reliability for different radiological measurements all relevant for hip dysplasia. The radiographs were drawn from participants taking part in the 1989 Hip Project, a follow-up of a large RCT on screening strategies for hip dysplasia. The dataset was balanced by oversampling dysplastic hips as judged by an experienced paediatric musculoskeletal radiologist (KR), who did not take part in the readings. This was done to ensure that a substantial number of dysplastic hips were included in the validation. All radiographs (right and left hips) were measured by three observers individually. Two observers (IØE and LBL) measured the radiographs four times, twice using a digital measurement programme and twice manually using the
AgfaWeb 1000 system. A third observer (TGL) measured all images three times, twice using the digital technique and once using the manual AgfaWeb 1000 system. Before performing the measurements, all three observers discussed and agreed on the precise definitions of landmarks to be used for all the measurements. All radiographs were performed by one specially trained radiographer using a well-defined protocol, reducing the risk of systematic errors.

Paper IV is also based on data from the 1989 Hip Project, comprising all participants born in 1989. Being a follow-up study, it can be considered as a prospective cohort design, but as only information at follow-up was used in this paper, a cross-sectional design is a more appropriate description. However, the prevalences were adjusted for non-responders using data from the Medical Birth Registry of Norway (MBRN) on gender, birth weight, maternal age, marital status, parity, foetal position and multiple births. The results were not adjusted with data from the newborn period as these data were not available at that time. This was, however, done in another study on the same cohort and did not significantly influence the results. The response rate at follow-up was 52%: 58% for females and 42% for males. As hip dysplasia is more common among females, it was important that the prevalences were adjusted according to gender. The predominance of female attenders is a frequent situation and described by Hill et al., but we doubt that our results have been influenced by a selection bias due to this predominance.

### 10.1.2 Health registry studies – pros and cons

A basic premise of epidemiology is to study the distribution and patterns of health states or health-related conditions, and their causes or influences in a population. By using health registries, medical diseases and treatment modalities can be studied. The registries are suitable as they include an unselected population and the results are consequently generalisable to all patients. The large number of patients registered allows for the collection of infrequent events. Further, the studies are relatively inexpensive, and the use of clinically important hard endpoints is possible. However,
some weaknesses of registry studies may be mentioned. Firstly, the quality of data is inferior to that e.g. clinical trials designed to study a specific question. Secondly, the use of data in the registries for research purposes can be challenging as some authorities perceive the registries as a threat to privacy. Thirdly, confounding factors are impossible to adjust for despite complex statistical methods. Often advanced statistics with multivariate analyses are used, which can be difficult to understand and interpret.

The health registries are important for epidemiological and medical research in Norway. Combined with the unique national identity number, biobank data and health data from large population studies, the registers provide Norway with outstanding research possibilities of high quality. There are thirteen mandatory national health registries in Norway (e.g. the Medical Birth Registry of Norway (MBRN)), and more than 40 national clinical registries (e.g. the Norwegian Arthroplasty Register (NAR))\(^7\).\(^8\)\(^,\)\(^15\). The fact that Norway is a small country with a relatively stable and homogenous population is also an advantage.

Most studies originating from the NAR are prospective observational studies where the patients enter with a total joint replacement and are followed until death or revision. By combing data from the NAR with the Birth Registry (Paper I), the same study design was used, but the patients enter the study at birth with their hip stability status and are followed until death or insertion of a total hip replacement. Unique to the birth registry in Norway and the other Nordic countries are the data on risk factors, health (e.g. hip instability) and health outcomes that are collected for each birth and can be linked to other registries and past or future events. The enormous volume of data from each registry can be combined in new interesting ways, utilising the extensive possibilities of the data and opening for inter-disciplinary collaboration.
10.1.3 Quality of register data

The various health registries in Norway provide data for many important studies. However, in order to draw correct conclusions, it is essential that the data is of high quality as there are several potential sources of error when interpreting the results of epidemiological studies.

Information bias occurs when an exposure or disease is incorrectly registered. This can be caused by mistakes made by the reporting physician (or other health care workers) or by the technical staff at the registry. A badly designed registration form may also increase the risk of incorrect reporting. A paper form is used to report to the Norwegian Arthroplasty Register (NAR), and was also previously used in the Medical Birth Registry of Norway (MBRN). The forms are plotted by the secretary in the NAR, and this was also done in the MBRN until December 1998 when a new reporting form was introduced. From then the forms were electronically read, but from 2006 the transition to electronic notification began. Today all maternity units report electronically. The different registration methods have their benefits and disadvantages. By plotting the data manually, an extra person handling the data is introduced and the risk of incorrect registration increases. However, this person can also sense weaknesses in the registration form and ensure that it is corrected as new trends, e.g. surgical procedures, may require regular updating of the form. In the future, however, practically all reporting to the registries will probably be done electronically. In this way bias related to the registration process is eliminated and it provides savings in terms of reduced secretarial expenses. Electronic reporting will also reduce the delay from the event until it is registered. But converting to reporting electronically should not be implemented until the new system is both as effective as the traditional one and also as convenient for the health worker filling in the form. There have been many challenges related to the electronic notification to the MBRN, and a system of high quality is important to correspond to the high quality of the recorded data.
The diagnoses reported to the NAR have only been validated for subjects aged <40 years at the time of the primary operation\textsuperscript{71,147}. Two separate papers have been published on this issue, one dealing with hip dysplasia as an underlying cause of prosthesis (Paper II) and the other focusing on Perthes’ disease and slipped capital femoral epiphysis\textsuperscript{147}. A total of 540 of 713 (76\%) patients registered with a primary total hip replacement in the NAR and born after 1967 were included in the study with questionnaires and medical records. The diagnoses reported to the NAR were confirmed as correct in 91\% of all cases. The dysplasia diagnosis was confirmed in 88\%, whereas only 61\% of the hips reported as primary osteoarthritis were confirmed. Correctness of 91\% for all underlying diagnoses is acceptable and corresponds well with the findings of a Danish study\textsuperscript{178}. In that study medical records and preoperative radiographs from 459 patients in the Danish Hip Arthroplasty Register were reviewed, giving a positive predictive value (PPV) for all registered diagnoses of 84\%. For hip dysplasia, the Danish study reported a PPV of 94\% (95\% CI 79-99\%) for congenital hip dislocation and 81\% (95\% CI 61-93\%) for acetabular dysplasia, corresponding well with our results. In the NAR, the dysplasia diagnosis is also grouped into two categories; sequelae dysplasia and sequelae dysplasia with dislocation. After reviewing medical records, we noticed that the term “dysplasia with dislocation” was confusing as to whether it described a dislocation at the time of operation or a dislocation earlier in life. To handle the situation, the two dysplasia alternatives were merged into one output called “hip dysplasia”.

Validation of other parameters than the diagnosis reported to the NAR has been performed by Arthursson and colleagues\textsuperscript{18}. They validated 5115 operations performed at one hospital in Norway (Stavanger University Hospital) in the period 1987-2003 and registered in the NAR. This constituted 5\% of the total number of operations reported to the register for the same period. In the local hospital database, 5134 operations were registered, thus 19 operations (0.4\%) were missing in the NAR database. Date of operation (1.1\% incorrect), index hip (left/right) (0.2\% incorrect), and reoperation rate (1.2\% missing) were also validated. In conclusion, they found the information recorded in the NAR to be valid and reliable throughout the period.
Unfortunately there are no validation studies of the neonatal hip instability diagnosis in the MBRN. However, the register has different alarms designed to detect new trends in the data. Among numerous false alarms through the years, hip dysplasia was source to a rather curious epidemic. The reason was revealed to be a specialist course for neonatologists in which new diagnostic criteria had been introduced. But in addition to this peak, the annually reported number of patients with hip instability varies widely. This variation probably does not reflect a real variation in prevalence, but changing diagnostic criteria and reporting procedures. With the introduction of a selective hip ultrasound screening programme in the beginning of the 1990s, the diagnostics were altered and new routines were introduced throughout the country. Whether the hip instability diagnosis reported to the MBRN includes hips with an abnormal ultrasound or not may vary according to the reporting hospital. In some cases the neonatal clinical examination takes place prior to discharge from the maternity unit and an ultrasound examination is performed at the outpatient clinic if needed. In these cases, the instability diagnosis for the registry might be missed. But even though the incidence of unstable hips in the MBRN has not been validated, the incidence of 0.88% found in Paper I is in good agreement with the literature. This is also in concordance with a Norwegian study from 1994. Hinderaker studied neonatal hip instability in the MBRN for the period 1970-1988 and found an occurrence of 0.9%. This figure is based on the same numbers as used in Paper I, but with some other exclusion criteria, e.g. oligohydramnions. Another MBRN-based study from 1986, addressing newborns born in the maternity unit at Haukeland University Hospital, Bergen, Norway, found 0.88% with hip instability. Furthermore, a study on completeness of another congenital malformation (oral clefts) showed that 94% of cases of cleft lip and palate were reported, but the percentage varied with the severity of the malformation. Several other validation studies based on data from the MBRN have been performed. Rasmussen and colleagues reviewed clinical and autopsy data from 108 457 newborns in order to validate the diagnosis of an unexpected antepartum foetal death. They found a PPV of 88% when compared to clinical data, and 77% when compared to both clinical and autopsy data. Obstetric sphincter tears were validated by
Baghestan and colleagues, who found a PPV of 91-95% and a sensitivity of 85-92%. Another study evaluating the diagnosis of rheumatic disease in 169 mothers notified to the MBRN found a sensitivity of 88%.

Selection bias occurs when the sample is not representative of the study population. This may occur as a result of low participation or dropout during follow-up. Today, it is mandatory to report to some of the Norwegian registries (e.g. the MBRN), but there has also been good reporting to several national registries not covered by this obligation. Studies on the NAR have shown that more than 97% of all joint replacements are reported, which is also in concordance with findings from the Danish Arthroplasty Register with a reported completeness of 94%. Due to the high reporting, the data can be treated as population-based. The MBRN is routinely linked with the Central Population Register to ensure high data quality.

Confounding represents a variable that correlates with both the investigated cause and the effect variables without being part of the cause being studied.

Validity is another term regarding data quality. A study of high “internal validity” refers to a study where no substantial errors from the three previously mentioned factors occur. Internal validity is an assumption for “external validity”, implying that the results can be generalised to subjects outside the study population.

10.1.4 Patient reported outcome measures

In medical practice, the evaluation of treatment often focuses on hard end-points such as death, treatment/operation rate, radiological measurement values or survival of e.g. the prosthesis. However, the aim of all research should be to benefit the patients, and it is therefore essential to include them with their experiences. Several questionnaires have been designed to report on patient outcome. Some are general (not disease specific) while others evaluate specific medical conditions (disease-specific).
Table 4 Strengths and weaknesses of EQ-5D and WOMAC.

<table>
<thead>
<tr>
<th></th>
<th>EuroQol EQ-5D</th>
<th>WOMAC</th>
</tr>
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<tbody>
<tr>
<td><strong>Categories</strong></td>
<td>5 levels: mobility, self-care, usual activities, pain and anxiety/depression</td>
<td>24 questions: pain (5), stiffness (2), physical function (17)</td>
</tr>
<tr>
<td><strong>Score(^1)</strong></td>
<td>-0.594 (worst) to 1 (best).</td>
<td>0 (best) to 96 (worst)</td>
</tr>
<tr>
<td><strong>Strengths</strong></td>
<td>Valid, Reliable, Easy to use, Cost-effective</td>
<td>Valid, Reliable</td>
</tr>
<tr>
<td><strong>Limitations</strong></td>
<td>Complex calculation needed, High ceiling effect, Less sensitive</td>
<td>Influenced by factors other than of lower extremities. Statistician needed</td>
</tr>
</tbody>
</table>

\(^1\)Based on UK value set

Designing a good questionnaire is a challenging procedure. The aim of the survey should be outlined in detail prior to the design process. This ensures that information relevant to the purposes of the study is obtained and avoids redundant questions. In addition, the information should be collected with maximal reliability and validity\(^{170,213}\). The validity of a questionnaire is the degree the assessment measures what it is supposed to measure. The reliability reflects the extent to which a measure is stable or consistent when it is administered repeatedly.

A validated questionnaire has undergone a validation process to ensure that it measures what it is designed to measure, regardless of who responds and when they respond\(^{213}\). The measurement is compared to the available “Gold Standard” and with other sources of data. The instrument should also be evaluated with respect to reliability, as a high degree of reliability offers the opportunity to compare the results from one study with the work of others. Unfortunately, many questionnaires described in the literature are not validated, and, as pointed out by Sushil and Verma, questionnaire design and validation are often neglected or overlooked at the expense of study validation\(^{213}\).
The EQ-5D and the WOMAC are two validated questionnaires included in this thesis (Table 4). The EQ-5D is a standardised measure of health status designed by the EuroQol Group, aimed to provide a simple, generic measure of health for clinical and economic assessment. The questionnaire comprised of five dimensions (mobility, self-care, usual activities, pain/discomfort and anxiety/depression) with three levels each: no problems, some problems and extreme problems. The answers can be translated into an index reflecting the person’s general health state. Strengths of the questionnaire are the brief and simple form and the fact that it can be completed within minutes. The questionnaire has been validated for numerous medical conditions and compared to other outcome measures (e.g. WOMAC). It has also been validated for patients undergoing hip replacement and been proven valid and reliable. The Norwegian version has never been validated, but there are several validation studies from Sweden. A limitation of the instrument is the complex calculation necessary to determine the final score. However, when used for research purposes, this calculation is automatically performed by statistical software such as SPSS. Further, the weighting for calculating the EQ-5D score differs from one country to another. The US and the UK weight the questions differently; no specific weightings have been calculated for Norway, and the UK or European version is normally used. Naturally, this complicates the possibility to compare the score between countries. Another limitation is that the form suffers from a high ceiling effect. The sensitivity is also low compared to other generic health forms, due to its brevity. Further, several studies have proved a bimodal distribution of the EQ-5D. This can be explained by the crude scaling of the EQ-5D. For example regarding mobility, the alternatives are “no problems”, “some problems”, and “unable/confined to bed”. Almost no patients will be in the “unable” group, and therefore the only movement which can be detected will be between “no problems” and “some problems”. This implies that the mean EQ-5D score often reported does not reflect this bimodal distribution.

In 2005, a modified form of the EQ-5D questionnaire was introduced. In order to improve the instrument’s sensitivity and reduce the ceiling effect, each dimension
was extended from three to five levels. The original questionnaire was renamed EQ-5D-3L and the new edition was named EQ-5D-5L. EQ-5D-3L is the version used in the papers included in this thesis.

The WOMAC is a self-administered disease-specific health status measure for osteoarthritis of the hip/knee. The questionnaire was validated by Bellamy et al. for patients with osteoarthritis in the lower extremities. They found the instrument to be valid, reliable, with high responsiveness and relatively efficient. The instrument has been proven sensitive to changes, and a change of score of 9-12 points for patients with osteoarthritis has been shown to be a clinically important difference. However, the specificity of the WOMAC instrument has been questioned. Wolfe and colleagues found in their study that the score was also influenced by other factors than OA (e.g. fatigue, depression and lower back pain), and the results must therefore be interpreted with care. The score can be difficult to determine for a clinician and statistical calculations are needed to identify differences between groups. Interpretation of significant differences in mean WOMAC scores is influenced by sample size and the size of the differences in scores between patient groups, thus large numbers are needed.

10.1.5 Reproducibility in measurement studies

Several statistical strategies have been described for the evaluation of reproducibility in measurement studies, but there is disagreement as to which method is the most appropriate. The expression reproducibility includes both agreement and reliability, two terms often used interchangeably in the literature. However, it is important to emphasise that these terms focus on different aspects of reproducibility. The agreement parameters (e.g. the Bland-Altman method and minimum detectable change) are more related to the measurement instrument itself and assess closeness of scores in repeated measurements. They indicate “how well a measure produces the same value on repeated measurements”. The reliability parameter (e.g. intra-class
correlation coefficient) is related to how well different measurements/patients can be
distinguished from each other.

In general, the Bland-Altman approach, analysing the difference between
measurements by two methods/observers on each subject is the preferred method for
analysing agreement on continuous data. According to Bland and Altman, the
degree of agreement between measurements is a problem of estimation rather than a
hypothesis testing or correlation. A correlation coefficient is a measure of association
and a good correlation will for instance be seen for any two methods designed to
measure the same parameter, and does not automatically imply that there is a good
agreement. A correlation coefficient will also be influenced by the range of
measurements used, e.g. widely spaced observations will increase the correlation
coefficient. The result may be that data showing an apparently high correlation can,
for individual subjects, show very poor agreement between the methods of
measurement.

Despite some critical voices, the intra-class correlation coefficient (ICC) has become
a more commonly used method to measure reliability and more advanced versions of
this method avoid some of the problems related to correlation. ICC was included in
paper III, in addition to the Bland-Altman method and minimal detectable change. By
including ICC, we were able to compare our results with others as ICC is often the
only reproducibility measurement found in the literature.

10.1.6 Measurement error and measurement uncertainty

All studies using measurements may be affected by measurement error. Measurement
error is the difference between the measured and the true (correct) value. A
measurement result consists of a measured value and its interval, in which the true
value is probably included. Measurement uncertainty is defined as “a parameter
associated with the measurement that characterises the dispersion of the values that
could reasonably be attributed to the measurand”.


Measurement error will always be present in material to be measured. Measurements will never be perfect, and therefore it is advised to do multiple measurements to calculate the most accurate value. The errors can be both systematic and random. A systematic error is caused by incorrect measuring methods, circumstances of the measuring and factors connected to the observer.

Measurement error and uncertainty are relevant for radiographic measurements, clinical data (hip range of motion, mobility, height, and weight), and different patient reported questionnaires. Radiographic measurements are included in papers III and IV, clinical data in paper IV and patient reported questionnaires in papers II and IV.

For radiographic measurements a high degree of measurement uncertainty can be problematic as these are often used to determine whether a patient is healthy or ill, the stage of disease, etc. So what is an acceptable measurement error? Some state that an uncertainty of ± 10% of the true (correct) value is acceptable. However, this is not always possible. Everyday life in clinical practice is often far removed from the ideal world. The radiographs should always be standardised, but different radiographers, locations and equipment are challenging. In addition, if the patient is monitored over time, there might be physical changes (increase/decrease of weight, fractures, ongoing growth), and it can be problematic to state whether a change in the measured value reflects a true change or one caused by various external factors. A good radiographic measurement should be easy to perform, measurable in most patients based on well-defined radiological landmarks and not affected by external environmental factors.

A high quality image is essential in performing accurate measurements. In a pelvis image, the pelvic tilt and inclination should be evaluated. According to Anda et al.\textsuperscript{11}, pelvic inclination in standing and supine pelvis radiographs shows insignificant variation. However, standing radiographs were preferred by Jacobsen et al. when analysing hip dysplasia and coxarthrosis to obtain the most accurate representation of femoral head translation and joint space widths\textsuperscript{118}. The same line of argument was used when we decided on projections for the 1989 Hip Project; a standing AP and a
supine frog-leg view were obtained. Further, the view used should be noted when performing measurement studies as many are performed with different protocols, e.g. urograms vs. pelvic views. Some radiological measurements are affected by this factor, which may result in changes in prevalences. It is therefore essential to debate this when comparing prevalence studies.

In papers III and IV, common radiographic measurements indicating hip dysplasia at skeletal maturity were used. In paper III, the measurements were validated according to intra- and inter-repeatability. A thorough standardisation prior to commencement was performed and all three observers agreed on the different radiological landmarks. In spite of this careful process, the results in paper III revealed varying agreement for the different measurements. In general, measurements of a small absolute value had poorer results. Further, the entire range of each parameter was not represented in our validation study (Paper III). This is a limitation of the study as the variation of the measurement may be influenced by the value we are measuring, i.e. more uncertainty for larger measurement values resulting in poorer agreement, referred to as a dome or funnel effect.

In paper IV, the different radiological measurements were used to determine the prevalence of hip dysplasia among 19-year-old Norwegians. When comparing our results with the literature it became evident that several different cut-off values are used and the proposed cut-off values are often based on material of varying quality. All the radiographs in the 1989 Hip Project were performed by the same radiographer in a standardised manner. By using the same equipment for all examinations, the risk of potential biases was reduced. In addition, there are concerns as to whether our results can be generalised to general orthopaedic surgeons and radiologists. As stated by Mast and colleagues, the radiographic diagnosis of hip dysplasia relies heavily on the clinician’s experience of reading hip radiographs. But the technical proficiency in actually making the measurements will often be independent of experience. Clinical decision making based on these parameters should take this possible bias into account.
In the 1989 Hip Project, all participants were clinically examined, including height, weight, hip range of motion, and an impingement test. For examination of the weight, an analogue scale was used, and the physician read the numbers and noted them on a special form (Appendix 10). In order to reduce a potential systematic bias, the same scale was used for all participants. In addition, the scale was checked with reference weights of different heaviness. Further, the range of motion of the hips was examined for all participants. Unfortunately, the clinical examination was never validated with respect to repeatability and validity. However, the same senior orthopaedic surgeon was responsible for training the four less experienced physicians. A goniometer was used in cases of uncertainty. The participants were also asked to fill in a questionnaire upon arrival. As mentioned in the previous section, questionnaires must also be validated with respect to measurement error and measurement uncertainty.

10.1.7 Defining hip dysplasia at skeletal maturity

The diagnosis of hip dysplasia at skeletal maturity can be quite difficult as there are no well-defined and well-documented guidelines for the diagnosis. Several different radiographic measurements are proposed with different cut-off values, and the background material is often inadequate. In paper III different measurements relevant for hip dysplasia were validated with respect to inter- and intra-repeatability. In conclusion, measurements with a high absolute value were found to have more satisfactory repeatability than measurements with smaller absolute values. However, the different cut-off values used in the literature were not evaluated in this paper. In paper IV, the prevalence of hip dysplasia among 19-year old Norwegians was studied. Different radiographic markers were used, but the prevalences varied to a great extent depending on the variable used. This indicates that the variable to be used in order to define dysplasia is of great importance. As stated by Jacobsen et al. in 2004, there is no agreement on radiographic cut-off values of dysplasia leading to hip osteoarthritis and the used cut-off values of the radiographic parameters are chosen somewhat arbitrarily in cross-sectional studies. Another paper from the 1989
Hip Project aimed to establish new reference intervals for different radiological measurements indicating hip dysplasia. However, these cut-off values do not necessarily reflect hips that will develop hip osteoarthritis. Further, the different radiological measurements are designed to measure slightly different anatomical relations in the hip joint. As the hip joint is a three-dimensional structure, it is not surprising that one single measurement cannot describe the complex structure. A score which includes several different measurements may be a valuable technique in order to give a more accurate description. This was proposed by Dr Tön尼斯 in 1976, when he suggested a “hip value” including several radiological measurements. However, it is of little use today.

Measurements designed to measure a dysplastic hip can be divided into two main groups: (1) measurements describing the acetabular anatomy and (2) measurements describing the position of the femoral head relative to the acetabulum. The first group consists of Sharp’s angle, the acetabular depth-width ratio (ADR) and acetabular angle of Tön尼斯 (AA). The second group includes Wiberg’s centre-edge (CE) angle, Ogata’s refined centre-edge angle and the femoral head extrusion index (FHEI). In addition to these measurements, several others have been proposed, but as they were not included in paper III or IV, a further description of these is not relevant here.

**Sharp’s acetabular angle.** Sharp’s acetabular angle was described by the British orthopaedic surgeon Ian K. Sharp in 1961 as a method of measuring the degree of acetabular development in the radiograph of the adult pelvis (Figure 24). The angle was already described in 1939 by the German Dr. Ullmann, but nowadays it is the British name that is most commonly related to the measurement. Using two points of the acetabulum (lateral edge of the acetabular roof and the inferior tip of the teardrop) and a horizontal line between the teardrops, the angle of inclination of the acetabulum can be measured. Sharp’s angle includes only landmarks of the acetabulum and Sharp claimed that this increases the accuracy of the measurement. A further advantage of the measurement is that it can be used for both children and adults.
Modifications of Sharp’s method have been described by others. The measurement is affected by cases of severe hip dysplasia as the teardrop in these cases is often deformed and hard to identify, resulting in difficulty in defining the horizontal line. As a solution, Tönnis proposed a method by drawing the horizontal line at right angles to the longitudinal pelvic axis\textsuperscript{231}. Another modification of the method, inspired by Dr Ogata who refined Wiberg’s centre-edge angle (see below), is a refined Sharp’s acetabular angle. The horizontal reference line is used as originally described, but the lateral landmark is defined as the lateral margin of the sourcil (subchondral bony condensation in the acetabular roof)\textsuperscript{2,128}. Using this method, a significant difference from the original method is observed. It is therefore important to confirm what method is used when results are compared. In paper III and IV, the original method of Sharp was used.

In his original paper, Sharp described how the measurement was influenced by rotation and tilt of the pelvis. Using five different views, the variation of the angle was no more than 4 degrees. However, one should be aware that the measurement is influenced by the position of the centre beam, and measurements should not be performed on an abdominal radiograph. This is supported by Jacobsen and colleagues.
as they found a highly significant effect of rotation and inclination on Sharp’s angle. Furthermore, Sharp’s acetabular angle does not reflect the depth of the acetabulum and this is a major theoretical objection to the validity of the measurement. However, Sharp argued that the angle of slope of the acetabulum appeared to bear a fairly constant relationship to its depth and therefore the angle is a good index of the acetabular development. Tönnis stated that Sharp’s angle defined the degree of lateroinferior inclination of the acetabulum, which is largely an expression of the load transfer across the joint.

Sharp stated that the acetabular angle was not influenced by gender and age, and this was supported by Stulberg and Harris. However, several studies have reported opposite results. Both Han and Aktas found a significant difference for gender and age, and an Egyptian study found a decreasing angle with patient age. Age and gender variation were also described in the original paper by Ullmann.

The normal range for Sharp’s angle was originally evaluated by measuring 200 hips (fifty male and fifty female pelvises) arbitrarily chosen from patients with no radiological evidence of osteoarthritis and a lower age limit of 60 years. The normal values for this angle were between 33 and 38 degrees, with an upper limit of normality from 39-42 degrees and essentially the same findings in men and women. An angle of 47 degrees was found in a hip with congenital subluxation and angles between 42 and 47 were claimed to need further investigation. Stulberg and Harris examined 60 persons (30 males, 30 females, age range 30 to 85 years) with no evidence of arthritis of the hip, and 130 patients (53 males, 77 females) with degenerative joint disease considered as primary osteoarthritis. They found the normal range of Sharp’s angle to be between 25° and 41° and characterised the hip as dysplastic if the angle was greater than 43 degrees (mean±3SD). Tönnis found an average angle of 38.3° in adults, with an upper normal limit of 42.3°. A fourth study referred to angles of 42 degrees or more as pathological, 39-42° as borderline pathological and 33-38° as normal. In a Japanese study, Nakamuro and colleagues studied 254 normal hips and defined values between ±2SD from the mean as representing a normal range. A dysplastic hip joint was therefore defined as a
Sharp’s angle of greater than 45°. These findings were supported by Fowkes et al. who examined one hundred coronal CT localisers (50 males, 50 females, age range 20-30 years). They found a mean of 38.8° and subsequently an upper limit (mean±2SD) of 45.5°. Jacobsen and colleagues also decided to use a 45° upper limit in their work on hip dysplasia in a Danish population.

In paper IV an upper cut-off value of 45 degrees for Sharp’s angle is used to define dysplastic hips. As already mentioned, cut-off values of 42°, 42.3°, and 43° are proposed in the literature. However, the fact that we found a high prevalence of hip dysplasia judged by Sharp’s angle by using a cut-off of 45 degrees indicates that the prevalence would be even higher if a lower cut-off value was used. Further, it can be argued that the high prevalence of hips with a pathological Sharp’s angle in our cohort is due to the relatively young age as compared to the upper age limit for studies defining cut-off values for Sharp’s angle (Sharp: 60 years, Stulberg & Harris: 85 years). If the presumption of a decreasing angle with age is correct, this can partly explain our findings.

**Acetabular depth-width ratio.** The acetabular depth-width ratio (ADR) has been described by various authors using slightly different measurement techniques. The aim of the measurement is to measure the shallowness of the acetabulum. Heyman and Herndon proposed the original method where the width is measured from the inferolateral point of the acetabulum to the lateral rim of the acetabulum, and the depth is measured from the medial sourcil point and perpendicular to the width line. The ratio is multiplied by 100 and named the acetabular quotient (or acetabular index). A modification of this quotient was proposed in the same paper, as Heyman and Herndon realised that the true acetabular quotient may be difficult to measure in some radiographs due to a poorly defined lower margin of the acetabulum. Therefore an approximate acetabular quotient was described. This measurement uses the line from the upper margin of the acetabulum to the lower end of the tear figure as the base line. They stated that this will not measure the true acetabular quotient, but the quotient will remain approximately the same as for
measurements of the true acetabular quotient as long as there is no marked obliquity of the acetabular roof.

Stulberg and Cooperman have also described the modified technique, using the inferior tip of the teardrop rather than the inferolateral point of the acetabulum\textsuperscript{51, \textit{212}}. They multiplied the ratio by 1000 instead of 100. In papers III and IV, Stulberg and Cooperman’s method is used, but the depth is measured slightly differently as the digital measurement programme\textsuperscript{179} was used (\textit{Figure 25}). The difference is that the depth line was perpendicular to the midpoint of the width line and the depth was given as the point where the depth line crossed the acetabular sourcil. In some cases the depth line was located medial to the medial point of the sourcil and the continuation of the sourcil was then extrapolated, and the depth was given as the intersection of the depth line and the extrapolated line. However, in most cases the two methods agreed. In the literature, the acetabular depth alone is sometimes reported as a parameter indicating acetabular dysplasia\textsuperscript{53, \textit{163, \textit{206}}}. It should, however, be emphasised that this depth is often calculated in yet another way, and must therefore not be confused with the depth specified by ADR. Another possible source of confusion is that the ADR sometimes is referred to in degrees, even though it is not an angle. This annotation can be confusing and might be related to the ACM angle of

\textit{Figure 25} Acetabular depth-width ratio (ADR)
Idelberger and Frank. The ACM angle is measured from the acetabular edge to the lower posterior end of the lunate figure, and the depth is measured from the middle of this line. The coverage percentage is given with the formula 200 cos (ACM) in which an acetabulum with an angle of 45° corresponds to 100% coverage of the hemisphere, but an angle of 50° corresponds to only 83% coverage. According to Jacobsen, the ADR measurement is affected by pelvic rotation and inclination/reclination, and this should be kept in mind when results are analysed.

Cooperman described an ADR of less than 250 as pathological. However, he did not elaborate on the basis for this cut-off value, and other studies on the normal range of the ADR are hard to find in the literature. There are however a few studies on Heyman and Herndon’s ADR. Delaunay and colleagues mention in their paper that the values of the Heyman and Herndon ratio have been found in vivo to be around 60 in adults, and in a comparison of normal and dysplastic hips with osteoarthritis, all normal hips were shown to have a ratio over 38. As mentioned earlier, Heyman and Herndon multiplied their ratio by 100 instead of 1000. However, the values proposed by Le Damany and Murphy do not correspond well with the value of 250 proposed by Cooperman. A paper from our group on participants in the 1989 Hip Project evaluated the Stulberg and Cooperman ADR. This paper proposed a new cut-off value of <235 for males and <233 for females.

**Acetabular roof angle of Tönnis.** The acetabular roof angle of Tönnis (AA) was originally proposed as a measurement in children with open triradiate cartilage and Hilgenreiner’s line (inter-triradiate line) was used as a reference line. A corresponding measurement for adults has been proposed, using the teardrop line instead of Hilgenreiner’s line as a reference (Figure 26).

The clinical range for the measurement is reported to be from 4 to 10 degrees, whereas values greater than 10 or below 0 are referred to as abnormal. Unfortunately, the basis for these cut-off values is not clearly described. Massie and Howorth reported in 1950 on the normal value for different age groups. For adult males (87 hips) and adult females (128 hips) the mean value and its corresponding
standard deviation was 9(3.2) and 13(3.7) degrees, respectively. Lingg and von Torklus studied 100 asymptomatic adult males and found a normal value range from -9.61 degrees to +9.31 degrees. In a reproducibility study by Bouttier and colleagues\textsuperscript{38}, 12 degrees was used as a cut-off, Delaunay et al. used 10 degrees\textsuperscript{59}, whereas Jacobsen et al. used 15 degrees as a cut-off\textsuperscript{17}. There is clearly no consensus on what is the correct cut-off value to define hip dysplasia, even though 10 degrees is probably most commonly used. The acetabular roof angle is only included in paper III in this thesis, where no cut-off value was used.

**Wiberg’s centre-edge angle.** Wiberg’s centre-edge (CE) angle (Figure 27) is one of the most commonly used measurements for hip dysplasia and was proposed by the Swedish orthopaedic surgeon Gunnar Wiberg in his thesis on the dysplastic acetabulum and congenital subluxation of the hip joint (1939)\textsuperscript{245}. The angle is formed by two lines: one line through the centre of the femoral head and perpendicular to the transverse axis, and a second line from the centre of the femoral head to the lateral aspect of the acetabulum. Originally, Wiberg stated that the transverse axis should be formed by an inter-centre line between the two femoral heads, but the literature suggests modifications of this method. Maurice E. Müller suggested using a line running through the triradiate cartilage in children (Hilgenreiner’s line\textsuperscript{101}) as a
reference line. In adults, the cartilage is closed and not visible in radiographs. A line intersecting the inferior teardrop point on the two sides was proposed as an alternative reference line by Müller. The modified method of Müller, using the inter-teardrop line, is used in papers III and IV.

Some limitations to the CE-angle were stressed by Sharp when he introduced the acetabular angle in 1961: (1) The centre point of a deformed femoral head cannot be located accurately, (2) subluxation, or simple loss of joint space, alters the CE angle, giving false readings, and (3) subluxation of the contralateral hip also affects the CE angle. Further, other conditions of non-dysplastic origin that will influence the measurement are a tilted pelvis or a non-neutral position of the femur. Jacobsen found that Wiberg’s CE angle was significantly affected by varying rotation and inclination/reclination of the cadaver pelvises. Further, it should be noted that several papers have reported that the CE angle shows an upward tendency with age. This can be due to marginal, undetectable osteophytes or a decreasing joint space. In a Swedish study of persons aged 8-75 years, Fredensborg (1976) found the CE angle to increase up to the age of 15, but with only a minimal increase after this age. Aktas and colleagues studied the hip joint morphology and the acetabular dysplasia rate in Turkish adults (aged 20-79 years). They found a statistically
significant difference in the CE angle for persons in the oldest age group (70-79 years) compared to the younger patients. Similar findings have also been described in an Egyptian \(^9\) and Chinese population\(^{202}\). The increasing CE angle may explain the high occurrence of hips with a pathological CE angle in our cohort, described in paper IV.

In Wiberg’s thesis of 1939\(^{245}\), he proposed cut-off values for the CE angle. One hundred persons (50 males and 50 females, 20-35 years) with a normal clinical hip examination were studied. A pelvic radiograph was taken with a focal distance of 100 cm and with the rays centred two finger-breadths above the symphysis. The results were not mathematically analysed, as Wiberg found this unnecessary as it is impossible to set exact figures for the borderlines between normal and pathological. However, he considered a CE angle below 20 degrees to be pathological, values of >25° were considered normal, whereas values of 20-25° were considered as a borderline group. In paper IV, both a cut-off value of 20° and 25° were included. However, we claim that 20° is the appropriate cut-off value with respect to the mean-2SD found in our population\(^{134}\)(Paper IV). These findings are also supported by others\(^{16,120,123,124,202,251}\), although McWilliams and colleagues reported that the 25° cut-off value corresponded well with their 2.5 percentile\(^{157}\). In a paper published by Lane and co-workers, 30° was used as a cut-off\(^{135}\).

**Ogata’s refined CE-angle.** A modification of Wiberg’s CE angle was proposed by the Japanese Dr Ogata in 1990 (Figure 28)\(^{169}\). The refined CE angle of Ogata uses the lateral end of the sourcil, i.e. the weight-bearing area of the acetabulum, rather than the lateral edge of the acetabulum, when these two points do not overlap. The background for this modified technique was young patients with a normal CE angle who later developed a dysplastic acetabulum. In some of these cases, Ogata found that the lateral point of the bony condensation of the acetabular roof did not reach the lateral rim of the acetabular roof, but was situated more medially. Ogata stated that the head cover could be more accurately determined by using the refined CE angle than the original method of Wiberg. The repeatability of the method was evaluated in
paper III, but the technique was not implemented in paper IV as the two techniques overlapped for the majority of hips in our cohort.

Dr Ogata claimed that hips in which the sourcil did not extend to the lateral bony acetabular roof margin represented cases with a defect posterior part of the acetabular roof in varying degrees. Kim and colleagues observed that the most lateral point of the acetabular roof represented the anterolateral portion of the acetabulum and the most lateral point of the sourcil indicated the lateral margin of the midsuperior part of the acetabulum on a plain radiograph. These two observations demonstrate that an acetabular roof in which the sourcil does not extend to the lateral margin on a plain radiograph has a more developed anterior portion compared to the middle and/or posterior portions. This indicates that using the lateral point as a landmark, as proposed by both Sharp and Wiberg in their angles respectively, overestimates the acetabular slope in hips with a defective acetabular roof. This view was also supported by Ömeroğlu et al. who stated that the Ogata classification system was a reliable and reproducible radiological indicator for reflecting the acetabular cover.

To my knowledge, the only study defining the normal range of Ogata’s angle is based on data from the 1989 Hip Project. The proposed cut-off values in this paper are 3
degrees lower than those for Wiberg’s CE angle (17 or 18 degrees vs. 20 or 21 degrees for females and males, respectively). However, it appears in the literature that the cut-off values applied for Wiberg’s CE angle are also used for Ogata’s angle. Jacobsen refers to one of his measurements as Wiberg’s CE angle with the corresponding cut-off values. But he also states that they have chosen to designate the readily identifiable lateral margin of the subsclerotic sourcil as their lateral point of reference\(^2\). This corresponds to the lateral point defined in Ogata’s refined CE angle, meaning that Jacobsen measured Ogata’s angle and not Wiberg’s angle. By using the lateral end of the sourcil rather than the lateral edge of the acetabulum, the measured angle decreases in those cases these two points do not correspond. By using the same cut-off values as proposed for Wiberg’s CE angle, a higher proportion of the studied population will be defined with a pathological angle. In paper IV, our results for Wiberg’s CE angle are compared to Jacobsen’s results for Ogata’s refined CE angle. This is a source of error when the results are interpreted as the difference between the results would be greater if exactly the same measurement techniques were used.

**Femoral head extrusion index.** The femoral head extrusion index (FHEI) was proposed by Heyman and Herndon in 1950 as a measure of the coverage of the

![Figure 29 Femoral head extrusion index (FHEI)]
femoral head (Figure 29). The measurement is also referred to as “acetabular head index”\(^{141}\), the “femoral head coverage”\(^{59,211}\) or the “acetabulum-head quotient”. A smaller index indicates a more dysplastic hip and a coverage of less than 75% is often considered pathological\(^{10,117}\). Wiig et al. validated the measurement in patients referred due to Perthes’ disease in the contralateral hip\(^{246}\). The mean-2SD femoral head extrusion index in normal hips was reported to be 82% (ranging from 80%-82% depending on the observer measuring the radiographs). Thus, they argue that 80% can be regarded as a reasonable lower limit of normal variation. Some authors also use the FHEI to describe the opposite, i.e. how much of the femoral head lies laterally to the acetabular edge; also termed “the migration index”\(^{155}\). A study of an Egyptian population aged 18-60 found that the index increased with patient age\(^9\). The femoral head extrusion index was measured in paper IV.

**Subjective evaluation of the sourcil.** A subjective evaluation of the shape of the sourcil (Figure 30) or the lateral part of the acetabulum has been proposed as a technique to evaluate dysplastic hips. Normally the lateral part should have a concave margin. In borderline hips the lateral margin is horizontal and in dysplastic hips the lateral part is convex with a sloping margin\(^{93}\). As this technique is based on subjective evaluation alone, it is impossible to determine cut-off values and to compare findings between studies. This weakens the technique. The evaluation of the sourcil was included in paper IV.

**Figure 30** The shape of the sourcil in a normal, borderline and dysplastic hip.
10.1.8 Ethics

All sub-studies in this thesis were approved by the Regional Committee for Medical and Health Research Ethics. The major ethical consideration related to this thesis is the radiographic examination of healthy adolescents. The research protocol was approved according to standard regulations. All teenagers participated voluntarily and several precautions were taken. The examination consisted of two radiographs, but we strongly focused on minimising the radiation exposure. All males were offered gonadal protection, but not all were interested in using it. Females were not offered this protection as a lead protection covering the ovarian region would also hide several important radiological landmarks. However, females with an uncertain pregnancy status were excluded from radiographic evaluation. A specially trained radiographer performed all the examinations. She was instructed to adapt the radiation doses to each participant to make them as low as possible without compromising the quality of the radiographs. All radiation doses were recorded and the mean doses in the AP view was 0.09 mSv and 0.06 mSV in the frog-leg view, giving a total radiation dose of 0.15 mSv for each participant. According to the Norwegian Radiation Protection Authority, the natural radiation in Norway varies from 0.05 μSv/h to 0.20 μSv/h\(^{167}\). This is equivalent to an annual dose of 0.4 to 1.8 mSv. The general population receives an annual radiation dose of approximately 3.5 to 4.5 mSv and half of this is from radon radiation in people’s homes. The exposure to cosmic radiation increases the closer we are to outer space, i.e. during flights, and is also influenced by the elevation of the area of where we live. The estimated radiation dose for a flight is 0.02-0.05mSv, approximately half of the dose of a chest radiograph\(^{233}\). The annual threshold of exposure for health workers is 20mSv. This demonstrates that the attendees in our study were exposed to a low level of radiation, similar to 15-20 days of natural radiation, which is not expected to involve any health risk.
10.2 RESULTS

10.2.1 Is the screening programme for hip dysplasia sufficient?

Clinical screening for hip dysplasia in Norway started in Bergen in 1953, initiated by Walther and Moe. In this period there were no official health clinics in Norway, only those established and run by the Norwegian Women’s Public Health Association. In the 1960s it was decided to make the offer available to the whole population by letting it become a public concern. The “Act on Health Clinics and Health Measures for Children” came into force in 1974 and a national guide book for physicians on clinical examination, including screening for hip dysplasia, was produced. The instructions were much the same as today and the importance of the abduction test was stressed. In the mid-eighties, radiographic screening for those at increased risk for hip dysplasia was introduced. The screening programme for hip dysplasia in Norway nowadays is based on routine clinical screening during the first or second day after delivery. An additional ultrasound examination is offered to those at an increased risk for hip dysplasia. This programme was informed by two RCT’s and first introduced in Bergen in 1990. Later, similar programmes were introduced in several other centres.

Uncorrected hip dysplasia is associated with significant long-term morbidity including chronic pain, gait abnormalities and premature degenerative changes warranting a total hip replacement in young adulthood. Early screening for hip dysplasia has the potential to prevent these long-term effects. Different endpoints have been used in order to evaluate different screening programmes for hip dysplasia. One commonly used outcome is the incidence of late diagnosed dysplasia (> 1 month of age). Others may be treatment rates, open surgery, delayed abduction splinting, avascular necrosis (AVN), delayed walking, limb length discrepancy, gait abnormality, chronic hip pain, osteoarthritis (OA) of the hip and total hip replacement.

A total hip replacement (THR) is the end-stage treatment for hip osteoarthritis due to hip dysplasia. All THRs performed in Norway are registered in the Norwegian
Arthroplasty Register. The youngest patient in the register is 11 years and the youngest one who had a THR operation due to hip dysplasia is 13 years. If the current screening programme of hip dysplasia is to be evaluated on the basis of the percentage of subjects suffering from a severe end-stage OA in need of a THR during life, the follow-up period should be longer. However, the incidence of THRs due to hip dysplasia may reflect the effect of clinical screening alone (the screening programme until the 1990s). In paper I, we found that only 8% of those who underwent THR due to dysplasia (born after 1967, maximum age at follow-up 38 years) were reported to the Medical Birth Registry of Norway (MBRN) as having instable hips at birth. The young patients (<40 years) with a primary total hip arthroplasty registered in the Norwegian Arthroplasty Register were studied in paper II and the results from paper I were confirmed, as only 12% were treated with early abduction (Frejka pillow). All were in need of hip preserving surgery later, i.e. none of those with only the Frejka pillow as treatment received a THR later. It is therefore of interest to question why the majority were not diagnosed at birth. This can indicate weaknesses in the clinical screening programme as all the patients were born at a time when the screening programme only consisted of clinical examination. With the introduction of ultrasound in the beginning of the 1990s, our knowledge of the newborn hip has increased. Today, we know that most unstable hips will resolve spontaneously and this can be monitored by ultrasound. The hips are evaluated both dynamically and statically. A high correlation between the two examinations is seen, but we know that hips can be mildly dysplastic but stable, and are accordingly not detected in a screening programme based on clinical examination alone. A possibility is that hips in need of a THR in young adulthood represent such cases. However, the population included in the screening programme used today is still too young for a THR, so it is too early to conclude whether or not the addition of ultrasound to the screening programme will influence the need for a THR due to hip dysplasia. Another explanation, which has been confirmed by others, may be that the hips are actually normal in the neonatal period, but tend to develop into dysplastic hips during growth. Jones and colleagues reported on five patients with initially normal dynamic and static ultrasound, but who returned with a dislocation. They
recommend a pelvic radiograph at 4-6 months in patients with a normal ultrasound to prevent such late cases. Also Rafique et al. published a report of a breech-born girl with a normal clinical and static ultrasound examination as newborn, but who returned with a dislocated right hip at age 12 months\textsuperscript{185}. Even though there is good consensus that ultrasound is a safe and effective screening tool for hip dysplasia in infants, the recommendations regarding radiographic follow-up of hips deemed normal by ultrasound screening are less clear. Some authors claim that hip dysplasia is an evolving condition that usually improves, but can also worsen, and that the persons need to be monitored\textsuperscript{112}, but several authors have suggested that patients can be discharged following a normal screening ultrasound\textsuperscript{19,122,174}.

A review performed by the Cochrane Collaboration in 2011 of five independent studies found insufficient evidence to give clear recommendations for screening practice\textsuperscript{203}. They stated that neither universal nor selective ultrasound strategies have been demonstrated to improve clinical outcomes including late diagnosed hip dysplasia and surgery. Another review paper also concluded with a lack of evidence either for or against general ultrasound screening of newborn infants for hip dysplasia\textsuperscript{250}. But as late detected hip dysplasia and surgery are uncommon events, the studies were underpowered to detect significant differences. In one of the randomised clinical trials, this was caused by a lower incidence of late subluxation or dislocation in the study period than in the period prior to study start, causing insignificant results\textsuperscript{189}. However, when compared with the pre-study period, the rates of late cases for the universal and selective ultrasound groups were significantly lower than with clinical screening alone (0.3 and 0.7 per 1000 vs. 2.6 per 1000 live newborns). A study of the long-term outcome of the three different ultrasound screening strategies (universal ultrasound, selective ultrasound and only clinical screening) for hip dysplasia in the 1989 Bergen Hip Cohort comprised follow-up data on 2038 19-year-olds\textsuperscript{133}. No statistically significant difference in radiographic or functional status between the three screening groups at skeletal maturity was found. Accordingly, an added value of universal or selective ultrasound screening for hip dysplasia in newborns was not found at skeletal maturity.
Despite the lack of studies demonstrating a significant reduction in the late cases and surgery in populations screened with ultrasound, there are strong recommendations on clinical screening with an additional hip ultrasound. There are, however, varying preferences for a particular strategy. Increased treatment rates for universal ultrasound screening versus selective ultrasound screening are found in the literature. Further, we know that most of the hips receiving abduction treatment would have resolved spontaneously without treatment. It is, however, difficult to identify those hips in need of early treatment. The clinical practice is therefore an overtreatment as it is believed that it is of minimal strain and the consequences of not treating can be dramatic. But complications with abduction treatment (e.g. the Frejka pillow), the most common treatment for early hip dysplasia, are seen, including avascular necrosis of the femoral head, femoral nerve palsies, pressure sores and parental anxiety.

The introduction of ultrasound screening in the 1980s and 1990s represented progress in newborn screening for hip dysplasia as the hip joint now could be visualised. But the fact that treatment rates increase with universal ultrasound screening should be kept in mind. The Norwegian screening programme for hip dysplasia focuses on clinical screening with additional ultrasound screening for risk groups and if clinical symptoms are presented. Hopefully, this is a good approach. However, it should also be emphasised that some studies indicate that a normal hip can develop into a dysplastic hip. Careful examination during childhood is important, and there should be a low threshold for referral to radiographic examination and/or consultation by a paediatrician or orthopaedic surgeon if dysplasia is suspected. The importance of this is revealed in paper II, as we found that most young adults with a THR due to dysplasia are late detected. The median age of dysplasia diagnosis for persons with a THR due to dysplasia was 7.8 years. However, these data are based on persons born when clinical screening alone was the routine screening programme. Further follow-up studies are needed to study the long-term effects of the different screening programmes.
10.2.2 Treatment options for hip dysplasia

Treatment for hip dysplasia depends on age at diagnosis and the severity of the condition. In cases of severe dysplasia, or a persistent dislocatable hip in the newborn, abduction treatment (Frejka pillow used in Norway) is started immediately. In cases of mild dysplasia, sonographic surveillance is performed until normalisation or until further action is considered necessary. This approach has reduced overtreatment among neonates and infants.

Further possible treatment options are orthosis or a hip spica cast. In cases where the femoral head cannot be relocated to the acetabular cavity, traction or open reduction may be necessary prior to a hip spica cast. Hips with poor acetabular coverage may be in need of surgery. Different pelvic osteotomies and femoral rotation osteotomies are the most common procedures. In young adults with residual hip dysplasia, a periacetabular osteotomy (PAO) is an option to restore the shape of the hip joint. The procedure involves making several angular cuts on the pelvis, separating the hip socket from the pelvis. This allows free rotation of the socket in three dimensions and the socket can then be placed in essentially any position. A PAO reveals the need of a THR. However, PAO is major surgery and there is a risk of complications such as nerve injury, non-union of pelvic bones and infections.

Young adults with a total hip replacement due to hip dysplasia were studied in paper II. The medical records were reviewed and the treatment for their hip dysplasia was noted. Common to these patients was the high age at dysplasia diagnosis (median 7.8 years). This is noteworthy and in agreement with the findings in paper I which reported that only 8% of all patients who underwent THR due to an underlying hip dysplasia had no reported hip instability in the newborn period, as assessed by clinical examination. Further, we found that the majority of the patients had undergone hip preserving treatment prior to their THR. Hip preserving treatment was reported for 67% of the confirmed dysplastic hips. Twenty-six percent had no treatment prior to their THR and for 8% information on prior treatment was missing. Fifty-eight percent of those who had undergone treatment were treated with a hip
spica cast, 47% with traction and 5% with orthosis. Only 11% were reported as having had treatment with the Frejka pillow. However, this number might be higher as this is a simple treatment and might have been missed in the notes in the medical records among all the more complex and specialised treatment regimes. Ninety-two percent of the hips that had undergone treatment were treated with hip-preserving surgery. This is a very high number and indicates that the surgeon has tried to preserve the hip joint to postpone a THR. Femur osteotomy was the most common of the surgical procedures and seen in almost 2/3 of them. On the basis of our studies, we cannot determine the effect of hip preserving surgery. But a dysplastic hip diagnosed at an early stage, with instigation of early treatment, will be likely to develop normally, and is at low risk of developing premature osteoarthritis in need of a THR before the age of 40 years. Dudkiewicz and colleagues from Israel reported on outcome for 11 total hip replacement patients operated before the age of 30 years following developmental dysplasia of the hip$. The findings in paper II were confirmed, as most patients had previously undergone multiple operations including open reduction, osteotomies, covering procedures, tenotomies and more. Mean age of the patients at the time of surgery was 23.3 (16-30) years, which is in accordance with our findings of 32 (14-40) years.

A THR is the end-stage treatment for hip dysplasia. Fortunately, only a small proportion of hips are in need of a hip replacement in young adult age. However, hip dysplasia is a major cause of hip replacements among young adults. Total hip replacements due to paediatric hip diseases have been reported to have inferior results as compared to THR due to OA$. This has been explained by morphological deformities in the proximal femur or in the acetabulum and previous surgery prior to the THR, leading to technically more difficult procedures. However, the survival of THRs due to hip dysplasia in the Norwegian Arthroplasty Register was not found to be inferior to that of THRs after OA, if adjustments are made for age and for type of implant$. These findings have also been supported by studies from the Danish Arthroplasty Register of patients with childhood hip diseases and the Nordic Arthroplasty Register Association (NARA), although these studies found an
increased risk for reoperation due to dislocation the first 6 postoperative months for patients with acetabular dysplasia. \cite{22,24}

### 10.2.3 Outcome measures in hip dysplasia patients with a THR

Quality of life (QoL) has been defined by the World Health Organization (WHO) as a ‘‘multidimensional’’ model which includes physical, material, social and emotional wellbeing, as well as individual development and daily activities.\cite{223} The consequences of a dysplastic hip on everyday living and activities range from no problems to severe problems. This is to be expected since there is a wide range of severity of the condition. Mild acetabular dysplasia may remain untreated throughout life without giving the patient any problems. In cases of moderate and severe dysplastic hips, a tight treatment regime is often followed. Numerous hospital visits, weeks or months with a hip spica cast and/or orthosis, and sometimes several correctional interventions are the history told by some, but fortunately not most, dysplasia patients.

Patients with a total hip replacement (THR) due to hip dysplasia are in general younger than the average total hip replacement patient, and the expectations and demands regarding the prosthesis may differ. In paper II, the general health state of young adults with a THR due to hip dysplasia was assessed. The cohort reported relatively low scores for quality of life (EQ-5D score 67-72 out of 100), compared to those for the general, age-matched Swedish and UK populations (85-90). Two-thirds reported some degree of pain, anxiety, or depression, while about 60% experienced problems with mobility, and 50% had difficulties with daily activities.

Tellini and colleagues reported on quality of life in 31 patients (mean age 51 years) affected by arthritis secondary to hip dysplasia (Crowe’s classification type I or II) who underwent hip replacement surgery.\cite{217} Quality of life was assessed by the Western Ontario and McMaster Universities Osteoarthritis Index (WOMAC, disease-specific outcome measure) and the 36-Item Short Form Health Survey (SF-
36. generic quality of life measure). An improvement was found post-operatively compared to pre-operatively for both scores. The SF-36 score was also compared with a healthy Italian population aged 45-54 years, finding that the patient group post-operatively scored better than the healthy population. As the authors commented, this may be due to a recall bias as recently rehabilitated patients may have tended to overvalue their current physical state when comparing with their pre-operative condition. The findings of the Italian group also contrast with our findings, as we found a lower health score (EQ-5D) compared to the general population. But different quality of life instruments were used and the population in our study was younger than the Italians. At what time the post-operative questions are asked may also affect the result. In our study, all the THR patients were asked at the same time, regardless of when they had their THR. In the Italian study, the post-operative questionnaires were administered at a minimum of four months after rehabilitation was completed. It is possible that the prolonged follow-up period in our study might be affected by more persistent complications, e.g. prosthesis wear, infections, dislocations, etc., resulting in lower quality of life scores. In another study, 28 patients with an underlying diagnosis of hip dysplasia who had undergone periacetabular osteotomy (PAO) and were aged over 40 years were compared to a control group of 61 patients with a THR due to a primary diagnosis of hip dysplasia. All outcomes were based on self-assessment using the WOMAC, the SF-12, the UCLA (University of California, Los Angeles) Activity Index, and the Hip and Knee Arthroplasty Satisfaction Questionnaire. The authors found that patients with a THR had a superior QoL outcome as compared to PAO patients older than 40 years. Ostendorf and colleagues used SF-36 and EQ-5D to study patient-reported outcome in THR patients predominately operated due to primary osteoarthritis (age range 36-89 years). They found that the SF-36 and the EQ-5D scores at one year after operation approached those of the general population.
10.2.4 Repeatability of hip dysplasia measurements

As stated earlier in this thesis, several different radiographic measurements are proposed for hip dysplasia. Some are used for both children and adults, while others are used in only one of the groups. To define the “ideal” or “perfect” hip dysplasia measurement is probably impossible. There are both limitations and strengths associated with most measurements. In paper III, the reliability and agreement for several radiological findings for hip dysplasia at skeletal maturity was evaluated. Two different measurement techniques were used, one digital (DDH_Adult) and one manual (PacsWeb1000). The main finding of this study was the notable inter- and intra-observer variation across different radiological measurements, which also has been shown by others\textsuperscript{166}. We found Sharp’s angle and the femoral head extrusion index to be the most accurate, but acceptable results were also seen for Wiberg’s centre edge (CE) angle, articulo-trochanteric distance (ATD), and acetabular depth-width ratio (ADR) (Table 5 & Table 6).

Nelitz and colleagues measured 100 radiographs of patients aged between 16 and 32 years with unilateral hip dysplasia\textsuperscript{166}. They found a high correlation judged by the intraclass correlation coefficient (ICC) for Wiberg’s CE angle, acetabular roof angle (AA), femoral head extrusion index (FHEI) and Sharp’s angle, but a poorer correlation for the acetabular depth-width ratio (ADR). Accordingly they recommend the use of the former measurements with a high inter- and intraclass correlation in the radiographic assessment for treatment planning and outcome studies of dysplastic hips. They also evaluated other measurements for hip dysplasia which were not included in our paper, and stated that the decision on what radiographic measurement to be used depends on the clinical question. Clohisy and colleagues have also validated radiographic features based on plain radiographs\textsuperscript{50}. Six observers (five with more than five years of practice with hip dysplasia and femoroacetabular impingement) performed a blinded radiographic review of 77 hips (a mixture of normal, dysplastic and femoroacetabular impingement). The acetabular roof angle of Tönnis was the only measurement included that also has been validated in our study. Further, they categorised the values (normal, increased or decreased angle) and
### Table 5 Intra-observer variation reported in the literature

<table>
<thead>
<tr>
<th>Authors</th>
<th>Year</th>
<th>N radiographs</th>
<th>N observers</th>
<th>Diagnosis</th>
<th>Age group (years)</th>
<th>Sharp’s angle</th>
<th>AA</th>
<th>ADR</th>
<th>Wiberg’s CE angle</th>
<th>FHEI</th>
<th>ATD</th>
<th>JSW</th>
</tr>
</thead>
<tbody>
<tr>
<td>Nelitz et al.</td>
<td>1999</td>
<td>100 radiographs</td>
<td>3 observers</td>
<td>Unilateral hip dysplasia</td>
<td>16-32</td>
<td>0.70-0.82</td>
<td>0.86-0.89</td>
<td>0.65-0.69</td>
<td>0.88-0.92</td>
<td>0.73-0.87</td>
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<tr>
<td>Tan et al.</td>
<td>2001</td>
<td>30 radiographs</td>
<td>2 observers</td>
<td>Hip dysplasia</td>
<td>3-36</td>
<td>0.42-0.85</td>
<td>0.91-0.94</td>
<td>0.86-0.96</td>
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<td>Laquesne et al.</td>
<td>2004</td>
<td>30 radiographs</td>
<td>2 observers</td>
<td>Routine control, no hip/lumbar pain</td>
<td>18-89</td>
<td>0.90-0.97</td>
<td>0.83-0.98</td>
<td>0.77-0.90</td>
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<tr>
<td>Tamast et al.</td>
<td>2006</td>
<td>51 radiographs</td>
<td>2 observers</td>
<td>Patients from orthopaedic outpatient clinic</td>
<td>16-54</td>
<td>0.74-0.89</td>
<td>0.97-0.98</td>
<td>0.94-0.97</td>
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<tr>
<td>Im et al.</td>
<td>2009</td>
<td>50 radiographs</td>
<td>1 observer</td>
<td>Hip-erosion or routine health check</td>
<td>17-90</td>
<td>0.90</td>
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<tr>
<td>Mast et al.</td>
<td>2010</td>
<td>39 radiographs</td>
<td>2 observers</td>
<td>Femoroacetabular impingement, mild hip dysplasia</td>
<td>18-54</td>
<td>0.55-0.84</td>
<td>0.86-0.97</td>
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<td>Williams et al.</td>
<td>2010</td>
<td>25 radiographs</td>
<td>2 observers</td>
<td>Normal or osteoarthritis</td>
<td>&gt;45</td>
<td>0.76</td>
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<td></td>
<td>0.98</td>
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<tr>
<td>Lee et al.</td>
<td>2011</td>
<td>36 radiographs</td>
<td>3 observers</td>
<td>Urograms</td>
<td>25-48</td>
<td>0.78-0.90</td>
<td>0.50-0.92</td>
<td>0.69-0.84</td>
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<tr>
<td>Engesæter et al.</td>
<td>2011</td>
<td>95 radiographs</td>
<td>3 observers</td>
<td>General population</td>
<td>18-19</td>
<td>0.83-0.89</td>
<td>0.80-0.95</td>
<td>0.72-0.80</td>
<td>0.84-0.97</td>
<td>0.79-0.95</td>
<td>0.95-0.99</td>
<td>0.57-0.81</td>
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<tr>
<td>Bouttier et al.</td>
<td>2012</td>
<td>51 hips</td>
<td>2 observers</td>
<td>Hip pain and/or hip motion limitation</td>
<td>40-75</td>
<td>0.79-0.92</td>
<td></td>
<td></td>
<td>0.77-0.94</td>
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</table>

### Table 6 Inter-observer variation reported in the literature

<table>
<thead>
<tr>
<th>Authors</th>
<th>Year</th>
<th>N radiographs</th>
<th>N observers</th>
<th>Diagnosis</th>
<th>Age group (years)</th>
<th>Sharp’s angle</th>
<th>AA</th>
<th>ADR</th>
<th>Wiberg’s CE angle</th>
<th>FHEI</th>
<th>ATD</th>
<th>JSW</th>
</tr>
</thead>
<tbody>
<tr>
<td>Nelitz et al.</td>
<td>1999</td>
<td>100 radiographs</td>
<td>3 observers</td>
<td>Unilateral hip dysplasia</td>
<td>16-32</td>
<td>0.70-0.82</td>
<td>0.86-0.89</td>
<td>0.65-0.69</td>
<td>0.88-0.92</td>
<td>0.73-0.87</td>
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<tr>
<td>Wiig et al.</td>
<td>2002</td>
<td>38-136 hips</td>
<td>3 observers</td>
<td>Normal hips of patients with CUP in the contralateral hip</td>
<td>N/A</td>
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<td>0.25-0.72</td>
<td>0.65-0.90</td>
<td>0.78-0.92</td>
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<tr>
<td>Laquesne et al.</td>
<td>2004</td>
<td>30 radiographs</td>
<td>2 observers</td>
<td>Routine control, no hip/lumbar pain</td>
<td>18-89</td>
<td>0.90</td>
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<td></td>
<td>0.78</td>
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<tr>
<td>Tamast et al.</td>
<td>2008</td>
<td>51 radiographs</td>
<td>2 observers</td>
<td>Patients from orthopaedic outpatient clinic</td>
<td>16-54</td>
<td>0.61</td>
<td>0.92</td>
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<tr>
<td>Mast et al.</td>
<td>2011</td>
<td>39 radiographs</td>
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<td>18-57</td>
<td>0.63</td>
<td>0.73</td>
<td>0.90</td>
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<td>Stubbs et al.</td>
<td>2011</td>
<td>30 radiographs</td>
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<td>&quot;Normal&quot; population</td>
<td>18-40</td>
<td>0.63</td>
<td>0.75</td>
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<tr>
<td>Engesæter et al.</td>
<td>2012</td>
<td>95 radiographs</td>
<td>3 observers</td>
<td>General population</td>
<td>18-19</td>
<td>0.83</td>
<td>0.83</td>
<td>0.77</td>
<td>0.84</td>
<td>0.82</td>
<td>0.98</td>
<td>0.55-0.65</td>
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<tr>
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<td></td>
<td>0.82</td>
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</table>
Kappa values were calculated (intra-observer reliability $K = 0.73$, inter-observer variability $K = 0.64$). The results are therefore not directly comparable to ours, as we analysed continuous variables. However, they concluded that many of the standard radiographic parameters used to diagnose hip dysplasia and/or femoroacetabular impingement are not reproducible, and they requested more information regarding precise definitions of landmarks and measurements.

The acetabular angle of Sharp was validated by Agus et al., analysing 66 hips from 33 patients previously treated surgically for unilateral or bilateral hip dysplasia\textsuperscript{7}. Mean age at follow-up was 9.5 years (range 3.5-20 years). They reported a mean intra-observer variation of $1.8^\circ \pm 1.9^\circ$, and a mean inter-observer variation of $2.1^\circ \pm 1.9^\circ$. Mast et al. reported the minimal detectable change (MDC) for Sharp’s angle to be $3.4^\circ$-$4.3^\circ$, and $5.0^\circ$ for intra-observer and inter-observer variation, respectively\textsuperscript{155}. This corresponds well with our findings (Paper III), where we reported the MDC to be $2.9$-$3.1^\circ$ (intra-observer) and $3.5^\circ$ (inter-observer) using the digital measurement technique.

The acetabular angle of Tönnis (AA) was validated by Broughton et al., but in children between four months and 15 years, where the tri-radiate cartilage was still open\textsuperscript{41}. The reference line used is then Hilgenreiner’s line as originally proposed, and not the teardrop line as used in skeletally mature subjects as in our study. Broughton and colleagues found the 95\% prediction interval to be $\pm 6.1^\circ$ and $\pm 5.5^\circ$ for intra-observer and inter-observer variation, respectively. This is slightly better than in the case of Troelsen and colleagues, who reported the standard deviation (SD) of the mean difference to be $3.5$-$5.3^\circ$ and $4.8$-$5.4^\circ$ for intra-observer and inter-observer variation, respectively\textsuperscript{226}. Mast et al. reported the MDC for AA to be $2.0^\circ$-$5.5^\circ$ (intra-observer) and $9.3^\circ$ (inter-observer)\textsuperscript{155}. This corresponds with our results, as we found the MDC to be $5.6$-$6.1^\circ$ (intra-observer) and $6.2$ (inter-observer) using the digital measurement technique. This is a high variation, considering the mean value for the measurement is approximately $7^\circ$. 
The reliability of Wiberg’s CE angle was evaluated by Broughton and colleagues. They assessed the centre-edge angle in 170 hips of children between five and 15 years of age. They found the 95% prediction intervals for intra-observer and inter-observer to be ±9.3° and ±9.1°, respectively. No systematic bias was reported. Ömeroglu et al. validated the CE angle in 33 patients (66 hips) who had previously been operated due to unilateral or bilateral hip dysplasia. Both Wiberg’s and Ogata’s CE angle were measured. For Wiberg’s CE angle, they reported a mean intra-observer variation of 3.1° ±3.0°, and a mean inter-observer variation of 4.0° ±3.6°. For Ogata’s CE angle, they reported a mean intra-observer variation of 3.8° ±2.8°, and a mean inter-observer variation of 5.1° ±4.8°. Wiig et al. have also reported on the inter-observer reliability of Wiberg’s CE angle. They analysed subjects referred due to Perthes’ disease in the contralateral hip. Measurements in both the sick and the normal hips were evaluated, but the analyses were separated and only the results for the normal hips are referred to here. The images were measured by the local orthopaedic surgeons with a special interest in children’s orthopaedics at the referral hospital, and two experienced paediatric orthopaedic surgeons. The study demonstrated poorer reliability when the local orthopaedic surgeons measured the images as compared to the two experienced paediatric orthopaedic surgeons. The authors reported standard deviation (SD) of the mean difference between 3.8 and 7.8 degrees. Troelsen et al. reported slightly better results with the SD of the mean differences between 2.6 and 3.8 degrees for intra-observer variation and 3.2-3.4° for inter-observer variation, in agreement with our findings (Paper III). Mast et al. reported the minimal detectable change (MDC) for Wiberg’s CE angle to be 3.0°-6.0°, and 8.3° for intra-observer and inter-observer variation, respectively. In our study (Paper III), the MDC for the digital technique varied between 3.8° and 4.8° (intra-observer), and 3.5° (inter-observer).

Wiig et al. reported on the inter-observer agreement for the femoral head extrusion index. They reported the standard deviation of the mean difference between 3.2 and 5.3% coverage. Mast et al. reported the MDC for FHEI to be 3.5%-4.4%, and 5.0% for intra-observer and inter-observer, respectively.
10.2.5 Occurrence of hip dysplasia

The occurrence of hip dysplasia varies according to gender, race, age, type of ascertainment, definitions used and other factors, which make it challenging to compare the results from different studies. There are several studies on the prevalence of hip dysplasia, most of them on newborn and infants, but also quite a few on adults (Table 6). The incidence also depends on the study period. The 1920s to 1950s was before the introduction of routine screening programmes for neonatal detection of hip dysplasia. There was variation in the diagnostic tests used, the age at diagnosis, and the inclusion and exclusion criteria. In the 1950s to 1980s clinical neonatal screening was exclusively used, and from the mid-eighties radiographic hip screening at age 4-5 months was introduced. Some authors added late-diagnosed hip dysplasia and dislocation to the neonatal incidence, making comparison difficult. From the 1980s onward, a period with the introduction of sonographic techniques for neonatal screening followed.

In paper I, we found a prevalence of neonatal hip instability (NHI) of 0.88%. As mentioned in an earlier section, there are some limitations to this study as the NHI diagnosis in the Medical Birth Registry of Norway has never been validated. It was not the main aim of the study to estimate the prevalence. However, our findings are in agreement with those reported by others. Hinderaker reported a similar figure (0.9%) for the period 1970 to 1988 using approximately the same definition of NHI on the same data material. Stamnes also confirmed this in his study on newborns born at Haukeland University Hospital from 1953 to 1985. However, the incidence varied for different time periods: 1953-1962 0.27%, 1963-1979 0.92%, and 1980-1985 1.85%. As commented by the authors, these variations probably do not reflect real variations, but are a consequence of several physicians examining the newborns in the final period as compared to only one physician in the period from 1963-1979. This demonstrates the importance of an experienced person performing the examinations. Further, it is important to emphasise that the prevalence of individuals testing positively does not reflect the proportion in whom the condition itself is present. Almost every condition for which there is an antenatal or neonatal screening
Table 6 Occurrence in percent of adult hip dysplasia in the literature based on Wiberg’s centre-edge (CE) angle, acetabular depth-width ratio (ADR), Sharp’s angle and femoral head extrusion index (FHEI) for females (F) and males (M).

<table>
<thead>
<tr>
<th>Authors</th>
<th>Year</th>
<th>Population group</th>
<th>Type of radiograph</th>
<th>N hips</th>
<th>Age (years)</th>
<th>CE&lt;20° F</th>
<th>CE&lt;20° M</th>
<th>CE&lt;25° F</th>
<th>CE&lt;25° M</th>
<th>ADR&lt;250 F</th>
<th>ADR&lt;250 M</th>
<th>Sharp&gt;45° F</th>
<th>Sharp&gt;45° M</th>
<th>FHEI&lt;75% F</th>
<th>FHEI&lt;75% M</th>
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<td>74</td>
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<tr>
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<td>Johnsen et al.</td>
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* The lateral margin of the subcondral sclerotic sourcil was used as the lateral point of reference (Ogata’s point)
programme, including hip dysplasia, has a prevalence lower than the proportion of infants who test positive in the primary screening test, due to false positive tests\textsuperscript{144,238}.

The prevalence of adult hip dysplasia was studied in paper IV. The main finding in this paper was the great variation of occurrence of the different parameters. A pathological CE angle of Wiberg was found in 3.3\% of the population using a cut-off value of 20\(^\circ\), but in as many as 20\% if 25\(^\circ\) was used as the cut-off value. Twenty percent had one or more parameters indicating hip dysplasia, but only 0.45\% met all the five features studied. Our findings are partly in concordance with those reported by others, but there are great racial variations (\textbf{Table 6}). A high prevalence of hip dysplasia is reported in Japan\textsuperscript{113,255}, Malawi\textsuperscript{160}, Turkey\textsuperscript{88} and in the Sami population\textsuperscript{125}, while the disease is very rare in India, China and among African-Americans in the U.S. Several studies have reported a high incidence in the Sami population. A study by Johnsen and colleagues found that 17\% of the Sami had definite dysplasia (CE angle <20 degrees) and 62\% had normal hip joints\textsuperscript{125}. A high prevalence is also found in Canadian Indians, with a reported incidence of 188.5 per 1000 as compared to zero among Bantus in Africa\textsuperscript{188}. Paterson et al. stated in 1976 that hip dysplasia was also very rare among Aborigines\textsuperscript{177}. In 2007, Moussa et al. studied 104 persons (208 hips) aged 40 to 88 years\textsuperscript{159}. They found that 3 hips (1.44\%) had mild to moderate acetabular dysplasia (CE angle<25), but found no cases of severe dysplasia (CE angle<20). Only one hip was found to have a Sharp’s angle of 45 degrees.

Some protective factors regarding hip OA due to dysplasia are described in the literature. In many African countries the mothers carry their babies on their backs with the hips fully abducted and partially flexed for at least the first year of life. This is proposed as a beneficial method of carrying and may partly explain the reduced prevalence of hip OA in African countries\textsuperscript{1,6}. Further, there is a tradition of sitting on very low chairs with hips fully flexed\textsuperscript{105}. The frequent use of the squatting position in the Egyptian population has been proposed as a cause of the very low incidence of primary OA\textsuperscript{141}.  

11. CONCLUSIONS

In the present study, we found that 0.88% of all Norwegian newborns are reported to have neonatal hip instability according to data held by the Medical Birth Registry of Norway. This finding is in accordance with the findings of others. As known, hip dysplasia may cause altered mechanical conditions in the hip, predisposing for osteoarthritis in young adulthood. A hip with severe OA may be in need of a total hip replacement.

Of the young adults with a total hip replacement before age 40 due to hip dysplasia, only 8% were diagnosed with an instable hip at birth. This finding was partly confirmed when the medical records for these young THR patients were reviewed (Paper II). A high age at time of dysplasia diagnosis (7.8 years) was found and 75% of the patients had undergone different hip-preserving treatments before their prosthesis. Furthermore, the diagnoses reported to the Norwegian Arthroplasty Register were validated. We found the dysplasia diagnosis reported to the Arthroplasty Register to be correct in 88% of the hips. Twenty-five percent of all total hip replacements in young adults were due to an underlying hip dysplasia.

Defining hip dysplasia at skeletal maturity can be troublesome. There is no consensus on which radiographic measurement to use and which cut-off values are most appropriate. In paper III, different radiographic measurements indicating hip dysplasia were validated. The agreement varied from fair to good, but unfortunately no excellent radiographic features were found. When the different measurements were studies in paper IV, we found that the prevalences varied to a great extent, underscoring the importance of a nuanced approach when diagnosing hip dysplasia. The CE angle of Wiberg is commonly used in the literature. Both 20° and 25° are described as cut-off values, but based on the findings in a Norwegian cohort, a 20° cut-off is the more appropriate, as using a 25° cut-off would indicate that up to 20% of the Norwegians had hip dysplasia in one or both hips, which is an unrealistic high number.
12. IMPLICATIONS AND FUTURE RESEARCH

Hip dysplasia is a condition about which there are still many unanswered questions and where there is a need for further research. A genetic predisposing is proposed, but no genes defining hip dysplasia have been detected. In the 1989 Hip Project saliva samples were collected from most of the participants. These samples have yet to be analysed, but provide a unique collection in combination with phenotype data. A further line of study should be the considerable ethnic variations in hip dysplasia. It is also a matter of interest whether hip dysplasia describes several different conditions or sub-types with different risk factors and prognoses.

National health registries are important contributors to research, but it is essential that the recorded data is of high quality. Unfortunately, the hip instability diagnosis recorded in the Medical Birth Registry of Norway has not been validated. A validation study of this diagnosis and others would strengthen the register. A further validation of the Norwegian Arthroplasty Register would also be beneficial. In paper II, a validation of the underlying diagnosis reported to the arthroplasty register was validated for patients of 40 years of age or younger. A new study which also included older patients, who are in fact the majority, would provide useful information and form a suitable basis for other studies.
13. SOURCES OF DATA

18. Arthursson AJ, Furnes O, Espehaug B, Havelin LI, Søreide JA. Validation of data in the Norwegian Arthroplasty Register and the Norwegian Patient Register:


115. Jackson RW, Pollo FE. The legacy of Professor Adolf Lorenz, the “bloodless surgeon of Vienna”. *BUMC Proceedings* 2004;17:3-7.


14. ERRATA

**Paper II**
Page 151, Figure 1: 15.09.89 → Correct: 15.09.87

**Paper III**
Page 776, Figure 1a: ADR = (A/B)*100 → Correct: ADR = (A/B)*1000

Page 75. Det complete citation for the reference of Rasmussen et al. is included. The reference is number 256 in the source of data.

Page 77. Table 4. The score for the WOMAC has been corrected to "0 (best) to 96 (worst)". It has also been specified that the given values for the EQ-5D score is based on the UK value set.

Page 78. It has been specified that both the UK and the European value set are used in Norway.
## 15. APPENDICES

<table>
<thead>
<tr>
<th>Appendix</th>
<th>Description</th>
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<tbody>
<tr>
<td>1</td>
<td>Registration Form – Medical Birth Registry of Norway (1967-1998) (Norwegian)</td>
</tr>
<tr>
<td>2</td>
<td>Registration Form – Medical Birth Registry of Norway (1999-) (Norwegian)</td>
</tr>
<tr>
<td>3</td>
<td>Reporting Form – The Norwegian Arthroplasty Register (Norwegian)</td>
</tr>
<tr>
<td>4</td>
<td>Invitation letter, questionnaire and consent form – Paper II</td>
</tr>
<tr>
<td>5</td>
<td>Registration form – Paper II</td>
</tr>
<tr>
<td>6</td>
<td>Invitation letter – The 1989 Hip Project</td>
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<td>7</td>
<td>Consent form – The 1989 Hip Project</td>
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<td>Questionnaire II – The 1989 Hip Project</td>
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<td>10</td>
<td>Clinical examination form – The 1989 Hip Project</td>
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**Medisinsk registrering av fødsel**

Sendes 9. dag etter fødselen til fylkesseksjonen (stadstyskils) i det fylket der moren er bosatt.


**Barnet**

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**Fødested. Navn og adresse på sykehuset/fødestemmet**

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**Faren**

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<tr>
<td></td>
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</table>

<table>
<thead>
<tr>
<th>Sektjon?</th>
<th>Nei</th>
<th>Ja</th>
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</thead>
</table>

<table>
<thead>
<tr>
<th>Sted (sykehusets stempel)</th>
<th>Dato</th>
<th>Jordmor</th>
<th>Lege</th>
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<tbody>
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<td></td>
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**IK - 1002.**
<table>
<thead>
<tr>
<th>Innholdsområde</th>
<th>Forgjenger/hemmeligheter</th>
<th>Medfødte misdannelser</th>
<th>Fraværet under fødselen for bedre forståelse av forholdene</th>
<th>Komplikasjoner, som beskrevet nedenfor</th>
<th>Komplikasjoner hos mor etter fødsel</th>
<th>Enkelte innholdsområder</th>
</tr>
</thead>
<tbody>
<tr>
<td>Spesifikasjon av skader, neonatale diagnoser og medfødte misdannelser – utfylles av lege</td>
<td>Nei</td>
<td>Ja</td>
<td>Nei, ang. type</td>
<td>Nei</td>
<td>Nei</td>
<td>Ja</td>
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<tr>
<td>Fødselstilstand</td>
<td>Ja</td>
<td>Nei</td>
<td>Nei, ang. type</td>
<td>Nei</td>
<td>Nei</td>
<td>Ja</td>
</tr>
<tr>
<td>Spesifikasjon av forhold før eller under svangerskapet:</td>
<td>Ja</td>
<td>Nei</td>
<td>Nei, ang. type</td>
<td>Nei</td>
<td>Nei</td>
<td>Ja</td>
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<tr>
<td>Legemål i svangerskapet:</td>
<td>Ja</td>
<td>Nei</td>
<td>Nei, ang. type</td>
<td>Nei</td>
<td>Nei</td>
<td>Ja</td>
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<tr>
<td>Indikasjon for inngrep og/eller induksjon</td>
<td>Ja</td>
<td>Nei</td>
<td>Nei, ang. type</td>
<td>Ja</td>
<td>Nei</td>
<td>Ja</td>
</tr>
<tr>
<td>Komplikasjoner hos mor etter fødsel</td>
<td>Ja</td>
<td>Nei</td>
<td>Nei, ang. type</td>
<td>Ja</td>
<td>Nei</td>
<td>Ja</td>
</tr>
<tr>
<td>Inngrep og/eller induksjon</td>
<td>Ja</td>
<td>Nei</td>
<td>Nei, ang. type</td>
<td>Ja</td>
<td>Nei</td>
<td>Ja</td>
</tr>
</tbody>
</table>
### HOFTEPROTINTER

**ALLE TOTALPROTINTER I HOFTEMAIL REGISTRERES.** Innsætt, skifting og fjerning av totalprotinter i hofteledd, samt kantplastikk, bløtdelsrevisjon for infisert protese og hemiprotinter på annen indikasjon enn fraktur/fraktursekvele. Hemiprotinter for fraktur/fraktursekvele registreres på Hoftemailskjema.

### TIDIGERE OPERASJON I AKTUELLE HOFT (ev. flere kryss)

<table>
<thead>
<tr>
<th></th>
<th>0</th>
<th>1</th>
<th>2</th>
</tr>
</thead>
<tbody>
<tr>
<td>Osteosyntese for fraktur i prox. femurende</td>
<td>Nei</td>
<td>Ja</td>
<td></td>
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<tr>
<td>Hemiprotinter pga. fraktur</td>
<td>Nei</td>
<td>Ja</td>
<td></td>
</tr>
<tr>
<td>Osteotomi</td>
<td>Nei</td>
<td>Ja</td>
<td></td>
</tr>
<tr>
<td>Antrode</td>
<td>Nei</td>
<td>Ja</td>
<td></td>
</tr>
</tbody>
</table>

### OPERASJONSDATO (dd.mm.yr)...

### AKTUELLE OPERASJON (ett kryss)

- Primærоперasjon (også hvis hemiprotein tidligere)
- Reoperasjon (totalprotei tidligere)
- Primær hemiprotein for annen indikasjon enn fraktur/fraktursekvele

### AKTUELLE SIDE (ett kryss) (Bilateral op. = 2 skjema)

- Øvre
- Nedre

### ÅRSAK TIL AKTUELLE OPERASJON (KRYSS AV EN I A ELLER B)

**A Primæroperasjon pga. (ev. flere kryss)**

- Idiopatisk coxartrose
- Rheumatoid artritt
- Sekvele etter frakt. coll. femur.
- Sek. dysplasi
- Sek. dysplasi med total luksasjon
- Sek. Perthes
- Sekv. Epiphysealise
- Mb. Bechterew
- Akutt fraktura coll. femur

### NAVN/TYPE (f.eks. caputnekrose, tidl. antrode os.l.)

[f.eks navn]

### SYSTEMISK ANTIBIOTIKA

### OPERASJONSSTUE

- Vanlig operasjonsstue
- Operasjonsstue med laminær luftstrøm
- "Green house"

### FIBROINLYSEHEMMER

- "Green house"
- Operaasjonsstue med laminær luftstrøm
- Vanlig operasjonsstue

### TILLAGT (ett kryss)

<table>
<thead>
<tr>
<th></th>
<th>0</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
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<tbody>
<tr>
<td>Direkte lateral (Transgluteal)</td>
<td>Nei</td>
<td>Ja</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Direkte lateral (Mediam glut. medius og tensor)</td>
<td>Nei</td>
<td>Ja</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

### BENTAP VED REVISJON (Bruk klistrelapp på baksiden, eller spesifiser navnet)

### PROTESEKOMPONENTER

- Acetabulum
- Femur

<table>
<thead>
<tr>
<th></th>
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<tbody>
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<td>Navn/Type</td>
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### SYSTEMISK ANTIBIOTIKA

### OPERASJONSSTUE

- "Green house"
- Operaasjonsstue med laminær luftstrøm
- Vanlig operasjonsstue

### TILLAGT (hud til hud)...

### FIBROINLYSEMENGER

### PEROPERATIV KOMPLIKASJON

### ASA KLASSE (se baksiden for definisjon)

### Legen som har fylt ut skjemaet (navnet registreres ikke i databasen).

### BENTAKREPLANTASJON (ev. flere kryss)

<table>
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<tr>
<th></th>
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<th>3</th>
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<th>5</th>
</tr>
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<td></td>
<td></td>
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<tr>
<td>Femur</td>
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### LEIE

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<tr>
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<td>Nei</td>
<td>Ja</td>
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<tr>
<td>Rygg</td>
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### TROKANTEROESTOTOMI

<table>
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<tr>
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</thead>
<tbody>
<tr>
<td>Nei</td>
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### PEROPERATIV KOMPLIKASJON

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<tr>
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<tbody>
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<td>Moribund</td>
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### A˚RSAK TIL REOPERASJON (ev. flere kryss)

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<tbody>
<tr>
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### AKTUELLE SIDE (ett kryss)

### OPERASJONSSTUE

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### OPERASJONSSTUE

<table>
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<th></th>
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### TILLAGT (ett kryss)

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<td></td>
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<td></td>
</tr>
</tbody>
</table>

### TILLAGT (ett kryss) (se baksiden for definisjon)

### Legen som har fylt ut skjemaet (navnet registreres ikke i databasen).
AKTUELLE OPERASJON

Primæroperasjoner: Dette er de første totaleproteseoperasjoner.
Primær hemiprotese for annen indikasjon enn frakturfraktursevle: Hemiprotese for frakturfraktursevle registeres på Huftebruddskjema.

ÅRSAK TIL AKTUELLE OPERASJON

Kryss av under A ved primæroperasjoner og under B ved reoperasjoner. I B må du kryse av for alle årsakene til reoperasjon, eller forklare med fritekt.

REOPERASJONSTYP


TILGANG


BENTRANSPRAKTASJON

Benpropp som sementstopper regnes ikke som bentransplantat.

PROTESEKOMPONENTER: Acetabulum - Femur - Caput - Trokanterdel og hals


KOMPLIKASJONER

Også operasjoner hvor pasienter dør på operasjonsbordet eller rett etter operasjon skal meldes. Ved stor stor blødnings, angi mengde.

ASA-KLASSE (ASA=American Society of Anesthesiologists)

ASA-klasse 1: Friske pasienter som røyker mindre enn 5 sigaretter daglig.
ASA-klasse 2: Pasienter med en asymptomatisk tilstand som behandles medikamentelt (f.eks hypertensjon) eller med kost (f.eks diabetes mellitus type 2) og ellers friske pasienter som røyker 5 sigaretter eller mer daglig.
ASA-klasse 3: Pasienter med en tilstand som kan gi symptomer, men som holdes under kontroll medikamentent (f.eks moderat anging pectoris og mild astma).
ASA-klasse 4: Pasienter med en tilstand som ikke er under kontroll (f.eks hjertesvikt og astma).
ASA-klasse 5: Mortalit/ødende pasient

MINIMAAL SVkirurgi (MIS = Minimally Invasive Surgery)

Med MIS menes her at kirurgien har brukt kort snitt og at det er brukt spesialinstrument laget for MIS

SYSTEMISK ANTIBIOTIKAT

Her finnes det på hvilket antibiotikum som er blitt benyttet i forbindelse med operasjonen, f.eks.: Medikament 1: Keftin 2g x 4, med varighet 12 timer.

TROMBOSEPROFYLAKSE


FIBRINOLYSEHEMMER

Her finnes det på om en benyttet blødningsreducerende legemidler i forbindelse med operasjonen (f.eks. Cyklokapron).

BEINTAP VED REVISJON

Femur (Paprosky’s klassifikasjon)
Type I: Minimat tap av metafysæt ben og intact dalfaye.
Type II: Stort tap av metafysæt ben, men intact dalfaye.
Type IIA: Betydelig tap av metafysæt ben uten mulighet for proximal mekanisk støtte. Over 4 cm intakt corticalis i ishismusområdet.
Type IIB: Betydelig tap av metafysæt ben uten mulighet for proximal mekanisk støtte. Under 4 cm intakt corticalis i ishismusområdet.
Type IV: Betydelig tap av metafysæt ben uten mulighet for proximal mekanisk støtte. Bred ishismus med liten mulighet for cortical støtte.

Acetabulum (Paprosky’s klassifikasjon)
Type I: Hemisfærisk acetabulum uten kantdefekter. Intakt bakre og fremre kolonne. Defekter i forankringssull som ikke ødelegger subchondral benplate.
Type IIA: Hemisfærisk acetabulum uten store kantdefekter, intakt bakre og fremre kolonne, men med lite metafysæt ben igjen.
Type IIB: Hemisfærisk acetabulum uten store kantdefekter, intakt bakre og fremre kolonne, men med lite metafysæt ben igjen og noe manglende støtte superiort.
Type IIC: Hemisfærisk acetabulum uten store kantdefekter, intakt bakre og fremre kolonne, men med defekt i medial vegg.
Type IIIA: Betydelig komponentvandring, osteolyse og bentap. Bentap fra kl. 10 til 2.
Type IIIB: Betydelig komponentvandring, osteolyse og bentap. Bentap fra kl. 9 til 5.

Kopi beholdes til pasientjournalen, originalen sendes Haukeland universitetssjukehus.

Kontaktpersoner vedrørende registreringsskjema er

Overlege Leif Ivar Havelin, tlf.: 55 97 56 87 og klinikkoverlege Ove Furnes, tlf.: 55 97 56 80
Ortopedisk klinik, Haukeland universitetssjukehus, Besøksadresse: Møllestadsbakken 11.
Sekretær i Nasjonalt Register for Ledproteser, Ortopedisk klinik, Helse Bergen: Ingunn Vindenes, tlf.: 55 97 37 43 og Ruth Wasmuth, tlf.: 55 97 37 42
Epost nr@helse-bergen.no
Internet: http://www.haukeland.no/ntf/
Kjære .............


I følge Nasjonal Register for Ledproteser er du en av de yngre voksne som har fått innsatt et kunstig hofteledd. Vi tillater oss derfor å be deg svare på noen spørsmål angående din hofteleidelse. Dine svar vil selvsagt bli behandlet konfidensielt og ingen personidentitifserbare opplysninger vil bli offentliggjort. Studien er godkjent av Datatilsynet og Etisk komité.


På forhånd takk for hjelpen!

Med vennlig hilsen

Lars B. Engeseter
Professor/ Seksjonsoverlege i Barneortopedi, Haukeland Universitetssykehus
Nasjonalt register for ledproteser
E-post: Lars.Engeseter@helse-bergen.no
Tel: 55 97 56 84

Stein Atle Lie
Statistiker, dr. philos
Nasjonalt register for ledproteser
Haukeland Universitetssykehus

Trude Gundersen Lehmann
Lege
Haukeland Universitetssykehus

Ingvild Øvstebø Engeseter
Ingvild Øvstebø Engeseter

Med vennlig hilsen

Navn
PASIENTSPØRRESKJEMA

Navn:

1. Fødselstunenummer:

2. Dato for utfylling av skjema: [____ | _____ | ____]

3. I Nasjonalt Register for Ledproteser er du registrert med følgende lidelse som bakgrunn for innsattelse av kunstig hofteledd:

   Idiopatisk coxartrose (=slitasjegikt i hoften med ukjent årsak)
   Rheumatoid artritt (=leddgikt)
   Seqv. frakturna colli femoris (=senskader etter lårhalsbrudd)
   Seqv. dysplasi (=senskader på grunn av grunne hofte skåler)
   Seqv. dysplasi m/ luksasjon (=senskader på grunn av grunne hofte skåler med lårhodet ute av ledd)
   Seqv. Perthes/epifysiolysse (=følgetilstand av Calvé-Legg-Perthes eller glidning av lårhodet på lårhalsen)
   Bechterew (=arvelig tilstivning av rygg og hofteledd)
   Annen årsak
   Årsaken mangler

Stemmer denne opplysningen?

   Ja
   Nei. Venligst angi riktig diagnose: ...........................................
   Vet ikke
4. Hvor gammel var du første gang du fikk behandling for din hoftelidelse?

   Alder: ............. Vet ikke:

5. Hvor gammel var du da du merket de første symptomene på din hoftelidelse?

   Alder: ............. Vet ikke:

6. Ved hvilke(t) sykehus ble du behandlet for din hoftelidelse?

   ................................................................. Vet ikke:

7. Har du smerter fra den andre hoften?
   
   Ja
   
   Nei

8. Er det andre årsaker til at du har problemer med å gå?
   (For eksempel smerter fra andre ledd, ryggsmerten, hjerte-karsykdom eller andre sykdommer som påvirker gangevnen din)
   
   Ja
   
   Nei
9. Smerte

Sett ett kryss på den streken som du synes tilsvarer din gjennomsnittlige smerteopplevelse fra den aktuelle hoften den siste måneden:

<table>
<thead>
<tr>
<th>Ingen smerte</th>
<th>Maksimal smerte</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
</tr>
<tr>
<td>Lett</td>
<td>Moderate</td>
</tr>
<tr>
<td>Middels</td>
<td>Sterk</td>
</tr>
<tr>
<td>Uetholdelig</td>
<td></td>
</tr>
</tbody>
</table>

10. Tilfredshet

Sett ett kryss på den streken som du synes tilsvarer hvor fornøyd du er med operasjonsresultatet:

<table>
<thead>
<tr>
<th>Fornøyd</th>
<th>Ikke fornøyd</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
</tr>
<tr>
<td>Svært fornøyd</td>
<td>Fornøyd</td>
</tr>
</tbody>
</table>
I de neste 5 spørsmålene ønsker vi å vite hvordan livssituasjonen din er i dag (EQ-5D-spørsmålene (http://www.euroqol.org):

11. ** Hvordan opplever du gangevnen din?**
   - Jeg har ingen problemer med å gå omkring
   - Jeg har litt problemer med å gå omkring
   - Jeg er sengeliggende

12. ** Hvordan klarer du personlig stell?**
   - Jeg har ingen problemer med personlig stell
   - Jeg har litt problemer med å vaske meg eller kle meg
   - Jeg klarer ikke å vaske meg eller kle meg

13. ** Hvordan klarer du dine vanlige gjøremål (f.eks. arbeid, studier, husarbeid, familie- og fritidsaktiviteter)?**
   - Jeg har ingen problemer med å utføre mine vanlige gjøremål
   - Jeg har litt problemer med å utføre mine vanlige gjøremål
   - Jeg er ute av stand til å utføre mine vanlige gjøremål

14. ** Smerter eller ubehat?**
   - Jeg har verken smerte eller ubehat
   - Jeg har moderat smerte eller ubehat
   - Jeg har sterk smerte eller ubehat

15. ** Angst eller depresjon?**
   - Jeg er verken engstelig eller deprimert
   - Jeg er noe engstelig eller deprimert
   - Jeg er svært engstelig eller deprimert

16. ** Hvordan er din helsetilstand i dag sammenlignet med helsetilstanden like før din (förste) hofteproteseoperasjon?**
   - Bedre
   - Uforandret
   - Dårligere
17. **Din helsetilstand i dag.**

For å hjelpe folk til å si hvor god eller dårlig en helsetilstand er, har vi laget en skala (omtrent som et termometer) hvor den beste tilstanden du kan tenke deg er merket 100 og den verste tilstanden du kan tenke deg er merket 0.

Vi vil gjerne at du viser på denne skalaen hvor god eller dårlig helsetilstanden din er i dag, etter din oppfatning. Vær vennlig å gjøre dette ved å trekke en linje fra boksen nedenfor til det punktet på skalaen som viser hvor god eller dårlig din helsetilstand er i dag.
Navn

I forbindelse med den videre studien ønsker vi å innhente medisinske opplysninger om din hoftelidelse fra sykehuset/sykehusene som har behandlet deg. På grunn av sykehusene sin taushetsplikt, er vi nødt til å innhente tillatelse fra pasienten for å få utlevert denne informasjonen.

Vi håper du kan gi oss tillatelse til å kontakte sykehuset hvor du ble behandlet for å hente ut medisinske data om din hoftediagnose.

☐ Ja, jeg tillater at dere kan kontakte sykehuset for å hente ut medisinske data om min hoftediagnose.

Signatur:...........................................................................

☐ Nei, jeg ønsker ikke at informasjon knyttet til min hoftelidelse skal utleveres og brukes i denne studien.

Ved eventuelle spørsmål, vennligst kontakt professor/seksjonsoverlege i Barneortopedi Lars B. Engesæter på tlf. 55 97 56 84 eller e-post: Lars.Engeseter@helse-bergen.no.
# TOTALPROTESE HOS UNGE VOKSNE

## DIAGNOSE I NRL
- Idiotapisk coxartrose
- Rheumatoid artritt
- Sekvele etter FCF
- Sekvele dysplasi
- Sekvele dysplasi med total laksjon
- Sekv. Perthes/Epifysiolyse
- Mb. Bechterew
- Akut fraktura coll femoris
- Annet

## NRL-DIAGNOSE BEKREFTET MED JOURNALOPPLYSNINGER
- Nei, annen diagnose bekreftet
- Ja

## DIAGNOSEGRUPPE ETTER JOURNALOPPLYSNINGER
- Idiotapisk coxartrose
- Hofteledad dysplasi
- Hofteledad dysplasi med total laksjon
- Calvè-Legg-Perthes
- Epifysiolyse
- Usikkert

## RØNTGENBILDER REGISTRERT
- Nei
- Ja
- Dato for røntgenbilder

## FØRSTE LÆGEKONTAKT
(Enten ved det aktuelle sykehus eller dokumentert i journalen)

## DATO

## AKTUELLE SIDE (bilateral opr. = 2 skjema)
- Høyre
- Venstre
- Bilateralt
- Ikke angitt

## SYMPTOMER OG FUNN
- Nedsatt abduksjon
- Nedsatt innadrotasjon
- Nedsatt ekstensjon
- Halling
- Smertevæp ved gange
- Lyskesmerter
- Knesmerter
- Asymmetriske hudfolder
- Ortolani pos
- Barlow pos
- Forsinket gangutvikling
- Anisomeli
- Hål-seteprevar pos (froskprevar)
- Bevegelsesinnkrekkning
- Annet (spesifiser)

## ANNEN
- Ingen behandling før proteseinsetting
- Fritekst, se bakside

## BEHANDLING HOFELEDDSDYSPLASI
<table>
<thead>
<tr>
<th>Startdato/</th>
<th>Sluttato/</th>
</tr>
</thead>
<tbody>
<tr>
<td>alder</td>
<td>varighet</td>
</tr>
<tr>
<td>Freikja pute</td>
<td>...</td>
</tr>
<tr>
<td>Gips</td>
<td>...</td>
</tr>
<tr>
<td>Ortose</td>
<td>...</td>
</tr>
<tr>
<td>Strekkebe</td>
<td>...</td>
</tr>
<tr>
<td>Operasjon (spesifiser type operasjon og dato)</td>
<td>...</td>
</tr>
<tr>
<td>Annen behandling (spesifiser)</td>
<td>...</td>
</tr>
</tbody>
</table>

## BEHANDLING CALVÈ-LEGG-PERTHES
<table>
<thead>
<tr>
<th>Startdato/</th>
<th>Sluttato/</th>
</tr>
</thead>
<tbody>
<tr>
<td>alder</td>
<td>varighet</td>
</tr>
<tr>
<td>Abduksjons-ortose</td>
<td>...</td>
</tr>
<tr>
<td>Strekk</td>
<td>...</td>
</tr>
<tr>
<td>Fysioterapi</td>
<td>...</td>
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<tr>
<td>Operasjon (spesifiser type operasjon og dato)</td>
<td>...</td>
</tr>
<tr>
<td>Annen behandling (spesifiser)</td>
<td>...</td>
</tr>
</tbody>
</table>

## BEHANDLING EPFYSIOLYSE
- Operasjon (spesifiser type operasjon og dato)
- Annen behandling (spesifiser)

## KOMPLIKASJONER
- Ingen kjente
- Infeksjon
- Fraktur
- Avaskulerer nekrose
- Pinneperforasjon
- Chondrolyse
- Annen

## Dato, sign.
Navn
Adresse

Invitasjon til å delta i

Hofteundersøkelsen, Haukeland Universitetssykehus


Vi tror i dag at grunne høfteskåler i nyfødtperioden er en viktig årsak til slitasjegikst i høfteleddet senere i livet. Studien som du nå inviteres med i, vil for første gang i historien vise utviklingen av høftene fra fødsel og frem til avsluttet vekst. En tilsvarende studie har aldri blitt gjort tidligere i verden. Resultatene fra denne studien kan få stor betydning for fremtidig behandling og oppfølgelse av nyfødte med medfødt høftefeil, og forhåpentligvis redusere forekomsten av slitasjegikt hos voksne.

Å være med innebærer utfylling av et spørreskjema, to røntgenbilder av høftene og undersøkelse av bevegeligheten i høftene. Det medfører ikke noe ubeheg og resultatet får du med en gang. Det hele vil ta ca. 20 minutter.

Undersøkelsen vil foregå på Barnerøntgen (i Barneklinikken 2. etasje), Haukeland Universitetssykehus.

Det er selvagt frivillig å delta, men for kvaliteten på studien er det avgjørende at flest mulig deltakar. Som belønning vil du få en MP3-spiller eller 150 kroner!
Dokumenterte bussutgifter innen Stor-Bergen vil også bli refundert.

Du har fått time .... : ....-2008 kl:....................

Mange av dine jevnaldrende vil også bli bedt om å delta i denne studien. Hvis det er et sterkt ønske fra deg, kan vi prøve å ordne det slik at flere venner kan komme sammen. For å endre den timen du har fått, tar du kontakt med oss på telefon 55 97 64 66 eller 911 02 568 eller trude.gundersen.lehmann@helse-bergen.no
UTFYLLENDE INFORMASJON OM HOFTESTUDIEN

Medfødt hoftefeil (=hofteleddsdysplasi) forekommer hos ca 2-3 % av alle nyfødte. Tilstanden oppdages vanligvis ved nyfødtnundersøkelsen, og de fleste blir bra etter 3-4 måneders behandling med "høftepute". Hos enkelte blir imidlertid tilstanden oppdaget senere, og behandlingen er da mer omfattende med gips og/eller operasjon. Hos andre ser vi at hofteksålen utvikles ufullstendig; at den blir "grunnere" enn normalt. Det er grunn til å tro at dette kan disponere for høfteslitasje senere i livet. I tillegg tror vi at enkelte har hoftefeil som er normale ved fødselen, men utvikler hoftefeil i de første levårene. Hoftestudien som du inviteres til å være med på, vil kunne klarlegge dette.


Målet med denne studien er å finne ut om der er en sammenheng mellom "grunne" hofteksåler i nyfødtperioden og utvikling av røntgenologisk høfteslitasje ved 17-19 år. Med andre ord, om de som hadde normale hofteledd ved fødselen fortsatt har normale hofteledd og om de som hadde grunne hofteksåler fortsatt har grunnere hofteksåler enn de andre. Fundet vil få stor betydning for nyfødtundersøkelse av alle barn, samt oppfølgelse av dem som får påvist grunne eller dysplastiske hofteksåler.


Vennlig hilsen

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trude.gundersen.lehmann@helse-bergen.no

Karen Rosendahl
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Ingvild Øvstebø Engesæter
Stud. med.

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Tlf. 55 97 56 84
lars.engesæter@helse-bergen.no
Navn/personnummer…………………………

Samtykke til deltagelse i Hoftestudien, Haukeland Universitetssykehus


Bergen,………………2008 Signatur: ……………………………………………………………

Dersom du er under 18 år må en av dine foreldre/foresatte godkjenne at du deltar i studien.

Bergen,………………2008 Forelder/foresattes signatur:……………………………………

TA MED DENNE SAMTYKKE ERKLÆRINGEN NÅR DU MØTER TIL UNDERSØKELSE.
NAVN
Tidligere hofteplager eller andre leddplager


Ingen problemer
Medfødt hofteledds dysplasi
Serøs coxitt (ikke-bakteriell betennelse i hofteleddet)
Septisk artritt i hofteledd (bakteriell betennelse i hofteleddet)
Calvé Legg Perthes’ sykdom
Epifysiolyse
Brudd
Leddgikt (Reumatoid artritt)
Annet (Spesifiser:………………………………………………………………)
Vet ikke
Har oppsøkt lege / Legevakt pga. problemer med hofte. Spesifiser:…………………..

Har du noen sykdom som har vart over 3 måneder? Hvilken:.........................

Hofteplager i nærmeste familie

Har du søsken som har medfødt hofteleidelse og har vært behandlet med pute?
Ja Nei Vet ikke Hvis ”ja”, antall: ( eks. 1 bror 2 søstre )

......bror/brødre .......søster/søstre ......halvbror/halvbrødre ....halvsøster/halvsøstre

Har du foreldre som har hatt medfødt hofteleidelse? Ja Nei Vet ikke
Hvis ”ja”, hvem: mor far

Har foreldrene dine plager med hofteene i dag? Ja Nei Vet ikke
Hvis ”ja”, hvem: mor far

Mors høyde……..cm Fars høyde……..cm
HOfte 89
Haukeland Universitetssykehus


1. Deltakernummer: ...........................

2. Navn: ................................................................

3. Fødselsnummer: ____________ __________

4. Yrke skoleelev annet (Spesifiser:.................................)

5. Har du noen gang hatt plager fra høyre hofte (varighet over 1 måner)?  
   Ja  Nei
   Hvis "JA", spesifiser............................................

6. Har du hatt plager fra høyre hofte siste 3 måneder?  
   Ja  Nei
   Hvis "JA", spesifiser............................................

7. Har du noen gang hatt plager fra venstre hofte (varighet over 1 måner)?  
   Ja  Nei
   Hvis "JA", spesifiser............................................

8. Har du hatt plager fra venstre hofte siste 3 måneder?  
   Ja  Nei
   Hvis "JA", spesifiser............................................

9. Hvor ofte har du vondt i nakken?
   omtrent hver dag
   mer enn 1 gang pr uke
   omtrent hver uke
   omtrent hver måned
   sjelden eller aldri
10. Hvor ofte har du vondt i ryggen?
   omtrent hver dag
   mer enn 1 gang pr uke
   omtrent hver uke
   omtrent hver måned
   sjelden eller aldri

11. Har du problemer som du relaterer til hoften, som gjør at du har vansker med å gå?
   Ja  Nei  Hvis "JA", spesifiser......................................................

12. Er det andre årsaker enn hofteplager som gjør at du har vansker med å gå?
    (For eksempel smertene fra andre ledd, ryggsmerte, hjerte-karsykdom eller andre
    sykdommer som påvirker gangevnen din)
    Ja  Nei  Hvis "JA", spesifiser......................................................

13. Utenom skoletid: Hvor mange GANGER i uken driver du med idrett/mosjon slik at du
    blir andpusten og/eller svett?
    hver dag
    4-6 ganger i uken
    2-3 ganger i uken
    1 gang i uken
    1 gang i måneden
    mindre enn 1 gang i måneden
    aldri

14. Utenom skoletid: Hvor mange TIMER i uken driver du med idrett/mosjon slik at du blir
    andpusten og/eller svett?
    ingen
    ½ time
    1 time
    2-3 timer
    4-6 timer
    7 timer eller mer
SMERTE- Tenk på smerten du opplevde i HOFTEN i løpet av de siste 48 timer.

15. Hvor mye smerter har du når du går på flott underlag?
   ingen   litt   moderat   stor   svært stor

16. Hvor mye smerte har du når du går opp og ned trapper?
   ingen   litt   moderat   stor   svært stor

17. Hvor mye smerte har du om natten når du ligger i sengen?
   ingen   litt   moderat   stor   svært stor

18. Hvor mye smerter har du når du sitter eller ligger?
   ingen   litt   moderat   stor   svært stor

19. Hvor mye smerter har du når du står oppreist?
   ingen   litt   moderat   stor   svært stor

STIVHET- Tenk på stivheten du har opplevd i HOFTEN i løpet av de siste 48 timer

20. Hvor alvorlig er stivheten i hoften din med en gang du våkner om morgenen?
   ingen   litt   moderat   kraftig   svært kraftig

21. Hvor alvorlig er stivheten i hoften din etter at du sitter ligger eller hviler senere på dagen?
   ingen   litt   moderat   kraftig   svært kraftig

FUNKSJON- Tenk på hvor vanskelig det har vært å utføre følgende daglige fysiske aktiviteter i løpet av de siste 48 timene, som følge av SMERTE I HOFTEN. Med dette mener vi din bevegelsesevne og evne til å klare deg selv.

SPØRSMÅL: Hvor vanskelig har det vært å........

22. gå ned trapper?
   ingen   litt   moderat   svært   ekstremt

23. gå opp trapper?
   ingen   litt   moderat   svært   ekstremt

24. reise deg fra sittende?
   ingen   litt   moderat   svært   ekstremt

25. stå oppreist?
   ingen   litt   moderat   svært   ekstremt

26. bøye deg ned mot gulvet?
   ingen   litt   moderat   svært   ekstremt
27. gå på flatt underlag?
   ingen,litt,moderat,svært,ekstremt
28. komme deg inn/ut av en bil?
   ingen,litt,moderat,svært,ekstremt
29. gå på handletur?
   ingen,litt,moderat,svært,ekstremt
30. ta på strømper?
   ingen,litt,moderat,svært,ekstremt
31. stå opp fra sengen?
   ingen,litt,moderat,svært,ekstremt
32. ta av strømper?
   ingen,litt,moderat,svært,ekstremt
33. ligge i sengen?
   ingen,litt,moderat,svært,ekstremt
34. komme deg inn/ut av dusj/badekar?
   ingen,litt,moderat,svært,ekstremt
35. sitte?
   ingen,litt,moderat,svært,ekstremt
36. komme deg til toalettet?
   ingen,litt,moderat,svært,ekstremt
37. tungt husarbeid?
   ingen,litt,moderat,svært,ekstremt
38. lett husarbeid?
   ingen,litt,moderat,svært,ekstremt
I de neste 5 spørsmålene (EQ-5D-spørsmålene (http://www.euroqol.org) ønsker vi å vite hvordan livssituationen din er:

39. Hvordan opplever du gangevnen din?
   1 Jeg har ingen problemer med å gå omkring
   2 Jeg har litt problemer med å gå omkring
   3 Jeg er sengeliggende

40. Hvordan klarer du personlig stell?
   1 Jeg har ingen problemer med personlig stell
   2 Jeg har litt problemer med å vaske meg eller kle meg
   3 Jeg klarer ikke å vaske meg eller kle meg

41. Hvordan klarer du dine vanlige gjøremål (f.eks. arbeid, studier, husarbeid, familie- og fritidsaktiviteter)?
   1 Jeg har ingen problemer med å utføre mine vanlige gjøremål
   2 Jeg har litt problemer med å utføre mine vanlige gjøremål
   3 Jeg er ute av stand til å utføre mine vanlige gjøremål

42. Smerter eller ubehag?
   1 Jeg har verken smerte eller ubehag
   2 Jeg har moderat smerte eller ubehag
   3 Jeg har sterk smerte eller ubehag

43. Angst eller depresjon?
   1 Jeg er verken engstelig eller deprimert
   2 Jeg er noe engstelig eller deprimert
   3 Jeg er svært engstelig eller deprimert

Mange takk for at du tok deg tid til å svare på spørreskjema!

Trude G. Lehmann           Lene B. Laborie                          Ingvild Øvstebø Engesæter
Cand.med                Cand.med                                Stud.med
Karen Rosendahl  Lars Birger Engesæter
Seksjonsoverlege, Professor dr. med     Seksjonsoverlege, Professor dr. med
Radiologisk avdeling     Barneortopedisk avdeling
Klinisk undersøkelse

Us dato

Høyde: cm

Vekt: kg

<table>
<thead>
<tr>
<th>Status</th>
<th>Høyre</th>
<th>Venstre</th>
</tr>
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<tbody>
<tr>
<td>Fleksjon:</td>
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<td></td>
</tr>
<tr>
<td>Ekstensjon:</td>
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<td>Abdusjon:</td>
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<tr>
<td>Addusjon:</td>
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<tr>
<td>Innadrotasjon:</td>
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</tr>
<tr>
<td>Utradrotasjon:</td>
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<td></td>
</tr>
<tr>
<td>Forkortning</td>
<td>mm</td>
<td>mm</td>
</tr>
<tr>
<td>Impingement</td>
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</tbody>
</table>

<table>
<thead>
<tr>
<th>Mobilitet</th>
<th>Høyre</th>
<th>Venstre</th>
</tr>
</thead>
<tbody>
<tr>
<td>Hyberekstensjon i albu &gt; 10°?</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Hyberekstensjon i kne &gt; 10°?</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Legger tommel ned på underarm?</td>
<td></td>
<td></td>
</tr>
<tr>
<td>&gt;90° dorsalfleksjon i 5. fingers grunnledd?</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Ta i gulvet med håndflate med strake knær</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
16. PAPERS I-IV