Radiostereometric analysis of sacroiliac joint movement and outcomes of pelvic joint fusion

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Publications included

1. **Precision and accuracy measurement of radiostereometric analysis applied to movement of the sacroiliac joint.**  
   Kibsgård TJ, Røise O, Stuge B, Röhrl SM.  
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2. **Radiosteriometric analysis of movement in the sacroiliac joint during a single-leg stance in patients with long-lasting pelvic girdle pain.**  
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4. **Pelvic joint fusions in patients with chronic pelvic girdle pain: a 23-year follow-up.**  
   Kibsgård TJ, Røise O, Sudmann E, Stuge B.  
   Epub 2012 Sep 23.
<table>
<thead>
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<th>Abbreviation</th>
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<tbody>
<tr>
<td>CI</td>
<td>Confidence Interval</td>
</tr>
<tr>
<td>CT</td>
<td>Computer Tomography</td>
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<tr>
<td>MRI</td>
<td>Magnetic Resonance Imaging</td>
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<tr>
<td>RSA</td>
<td>Roentgen Stereophotogrammetric Analysis</td>
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<tr>
<td>VAS</td>
<td>Visual Analogue Scale</td>
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<td>ODI</td>
<td>Oswestry Disability Index</td>
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<tr>
<td>SF-36</td>
<td>Short Form-36</td>
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<tr>
<td>SIJ</td>
<td>Sacroiliac Joint</td>
</tr>
<tr>
<td>ME</td>
<td>Mean Error of Rigid Body Fitting</td>
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<tr>
<td>CN</td>
<td>Condition Number</td>
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<tr>
<td>MIS</td>
<td>Minimal Invasive Surgery</td>
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<tr>
<td>LBP</td>
<td>Low Back Pain</td>
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<tr>
<td>PGP</td>
<td>Pelvic Girdle Pain</td>
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<tr>
<td>BMI</td>
<td>Body Mass Index</td>
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<tr>
<td>A-P</td>
<td>Anterior - Posterior</td>
</tr>
<tr>
<td>ASLR</td>
<td>Active Straight Leg Raise</td>
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<td>LOS</td>
<td>Limit of significans</td>
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**Summary**

The sacroiliac joint (SIJ) might be the source of pain for 13-30% of patients with low back pain and possibly an even greater proportion of patients suffering from “failed back surgery”. This pain can be caused by specific pathology of the joint, but the specific role of the SIJ in unspecific pelvic girdle pain (PGP) disorder remains unknown. PGP is a common complaint in pregnancy that can cause disability, and in some women, the complaint continues after delivery. The origin and diagnosis of PGP are also unclear because radiological findings are often absent, and the diagnostic criteria lack sufficient evidence. However, it has become increasingly apparent that patients with PGP have different clinical presentations than patients suffering from low back pain. Based on the theory of pathological joint mobility, SIJ fusion combined with symphysis pubis fusion is a therapeutic option when conservative treatment has been unsuccessful.

This thesis includes four papers. In the first two papers, we used a specialized x-ray method, called radiostereometric analysis (RSA), to evaluate the movement of the SIJ, and in the last two papers, we evaluated the outcomes after SIJ fusion.

In paper I, we evaluated the RSA method when this method was applied to the SIJ. We used a phantom model to measure the true values of movements, and these values were compared to the measurements obtained by RSA imaging. By this process, we could determine whether there was any bias that had to be corrected for when this method was used. Furthermore, we measured the precision of the method in the phantom model and in patients. The main results were that the accuracy and precision of the RSA method were high, and the method could be used to measure SIJ movement.

In paper II, we used the RSA method to measure the movement in the SIJ in the single-leg stance. Chamberlain described a method for indirectly measuring SIJ movement by measuring the movement in the pubic symphysis on anterior-posterior (A-P) x-rays. This procedure was performed in the single-leg stance, and Chamberlain attempted to correlate the pubic movement with SIJ pain. However, there have been different reports regarding this relationship, which have made it difficult for clinicians to use the results of the Chamberlain
test in the diagnosis of PGP, particularly when normal variations in the movement of the pubic symphysis have also proven to be large. We used RSA to measure the movement in the SIJ in the single-leg stance in 11 patients, and the movements were small and almost undetectable using the method. We measured a mean rotation of 0.5° on both the standing- and hanging-leg SIJs, and no translation was detected. There were no differences in total movement between the standing- and hanging-leg SIJs. From the results of this study, we consider the Chamberlain examination to likely be inadequate for evaluation of SIJ movement in patients with PGP.

In paper III, we used a single-subject design study to evaluate the outcomes of pain, disability and health-related quality of life 1 year after SIJ fusion in 8 patients with severe PGP. These patients were included by applying strict inclusion and exclusion criteria, and they were submitted to surgery with anterior unilateral SIJ fusion combined with a fusion to the pubic symphysis. One year after open unilateral anterior SIJ fusion combined with symphysis pubis fusion, positive and significant changes in both physical function and pain were observed. Despite these positive results, this procedure was associated with adverse events and complications.

In the last paper, paper IV, we performed a long-term follow-up of 50 patients who underwent SIJ fusion performed by Sudmann in Hagavik, Bergen, Norway. All of the patients completed a questionnaire that measured the outcomes of pain, disability and health-related quality of life, and these outcomes were compared with the 1-year outcomes collected by the Sudmann. A comparison group of 28 patients who did not receive SIJ fusion completed the same questionnaire. Patients with chronic PGP who underwent SIJ fusion reported being moderately disabled, with moderate or severe pain intensity 23 years after surgery. Approximately half of these patients had successful 1-year outcomes, and in these patients, good results were sustained 23 years after surgery. Two-thirds of the patients experienced a positive long-term effect from fusion surgery, and 20% reported no effects from the surgery. It appeared that this surgery was an appropriate treatment option for a select group of patients with severe PGP, but which patients would benefit from surgery remains unclear.

Denne avhandlingen inkluderer fire artikler. I de to første artiklene brukte vi en spesiell røntgenmetode, kalt radiostereometrisk analyse (RSA), for å vurdere bevegelsen i iliosacralleddet og i de to siste har vi evaluert utfallet etter avstivningsoperasjon av iliosacralleddet.

I artikkel I evaluerte vi RSA metoden anvendt på iliosacralleddet. Vi brukte en fantommodell for å måle den sanne verdi av bevegelsen, og disse verdier ble sammenlignet med resultater av RSA målingene. Av dette kunne vi se hvor nøyaktig metoden var og om det forelå systematisk feil som innvirket på resultatene. Vi målte også presisjonen av RSA metoden både på en fantommodell og på pasienter. Hovedresultatene var at nøyaktigheten og presisjonen av RSA metoden var høy, og at RSA kan brukes til å nøyaktig måle bevegelse i iliosacralleddet.

I artikkel II brukte vi RSA metoden for å måle bevegelse i iliosacralleddet i forbindelse med single leg stance test (stående vekselvis på ett og ett ben med det andre hengende ned). I 1930 beskrev Chamberlain en metode for å indirekte måle iliosacroalleddets bevegelse ved å måle bevegelsen i symfysen på et vanlig front røntgenbilde av symfysen. Bildene ble tatt i
forbindelse med single-leg stance test og disse resultatene ble forsøkt korrelert med smerter i iliosacralleddet. Sprikende resultater med tanke på denne korrelasjonen har gjort det vanskelig for klinikere å bruke resultatene av Chamberlain test i diagnostisering av bekkenleddsmerter. Ved hjelp av RSA målte vi bevegelsen i iliosacralleddet i forbindelse med single-leg stance test hos 11 pasienter med bekkenleddsmerter. Vi fant at bevegelsen i iliosacralleddet var veldig liten, så liten at den nesten var umulig å oppdage ved bruk av RSA. Vi målte en gjennomsnitts rotasjon på 0,5 grader på både det stående og hengende bens iliosacralledd, og ingen translasjon ble funnet. Det var ingen forskjell i total bevegelse mellom de leddene med smerter og de uten smerter. Resultatene i denne studien tyder på at den tradisjonelle Chamberlain-undersøkelsen trolig ikke har noen plass i utredningen av pasienter med mistenkt iliosacralleddssmerte.

I artikkel III brukte vi et single subject design studie for å vurdere utfallet av smerte, fysisk funksjon og helselatert livskvalitet 1 år etter avstivning av iliosacralledd og symfyse hos åtte pasienter med uttalte bekkenleddsmerter. Pasientene ble inkludert med veldefinerte inklusjons- og eksklusjonskriterier. Ett år etter åpen ensidig fremre avstivning av iliosacralleddet kombinert med avstivning av symfysen fant vi positive og betydningsfulle endringer i både fysisk funksjon og smerte. Til tross for disse positive resultatene så vi dessverre at noen pasienter fikk tildels alvorlige komplikasjoner.

Thesis at a glance

Paper I

**Background** Different techniques have been used to quantify the movement of SIJ's. These include RSA, but the accuracy and precision of this method have not been properly evaluated and it is unclear how many markers are required and where they should be placed to achieve proper accuracy and precision. The purpose of this study was to test accuracy and precision of RSA, applied to the SIJ, in a phantom model and in patients.

**Methods** We used a plastic phantom attached to a micrometer to obtain a true value of the movement of the SIJ and compared this value with the measured value obtained by RSA; the difference represented the accuracy. The precision of the system was measured by double examination in the phantom and in six patients, and was expressed by a limit of significance (LOS). We analyzed different marker distributions to find optimal marker placement and number of markers needed.

**Results** The accuracy was high and we identified no systematic errors. The precision of the phantom was high with a LOS less than 0.25° and 0.16 mm for all directions, and in patients, the precision was less than 0.71° for rotations and 0.47 mm translations. No markers were needed in the pubic symphysis to obtain good precision.

**Conclusions** The accuracy and precision are high when RSA is used to measure movement in the SI joint and support the use of RSA in research of SIJ motion.

Paper II

**Background** Chamberlain’s projections (anterior-posterior x-ray of the pubic symphysis) have been used to diagnose SIJ mobility during the single-leg stance test. This study examined the movement in the SIJ during the single-leg stance test with precise RSA.

**Methods** Under general anesthesia, tantalum markers were inserted into the dorsal sacrum and the ilium of 11 patients with long-lasting and severe pelvic girdle pain. After two to three weeks, a RSA was conducted while the subjects performed a single-leg-stance.

**Results** Small movements were detected in the SIJ during the single-leg stance. In both the standing- and hanging-leg sacroiliac join, a total of 0.5° rotation was observed; however, no translations were detected. There were no differences in total movement between the standing- and hanging-leg SIJ.

**Interpretation** The movement in the SIJ during the single-leg stance is small and almost undetectable by the precise RSA. A complex movement pattern was seen during the test, with a combination of movements in the two joints. The interpretation of the results of this study is that, the Chamberlain examination likely is inadequate in the examination of SIJ movement in patients with PGP.
Paper III

**Background** The fusion of the pelvic joints in patients with severe PGP is a controversial and insufficiently studied subject. The aims of this study were to evaluate physical function and pain after SIJ fusion.

**Methods** A single-subject research design study with repeated measurements was conducted; pre-operatively and 3, 6 and 12 months post-operatively. The outcome measures considered were the Oswestry disability index (ODI), visual analogue scale (VAS), and SF-36. Eight patients with severe PGP received open-accessed unilateral anterior SIJ fusion and concomitant fusion of the pubic symphysis.

**Results** Seven patients reported positive results from the surgery. At 1 year post-operation, significant (p<0.001) reductions in ODI (54 to 37) and VAS (82 to 57) were reported. The physical functioning, bodily pain, and social functioning scores in the SF-36 were also improved.

**Conclusion** Positive and significant changes in disability and pain at 1 year after SIJ fusion were observed. Despite these positive results, open accessed anterior fusion of the SIJ was associated with adverse events and complications such as infection and nerve damages.

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Paper IV

**Purpose** Fusion of the SIJ has been a treatment option for patients with severe PGP. The primary aims were to evaluate the long-term outcomes in patients who underwent SIJ fusion and to compare 1-year outcomes with long-term outcomes. The secondary aim was to compare patients who underwent SIJ fusion with a comparable group who did not.

**Methods** This study includes fifty patients that underwent SIJ fusion between 1977 and 1998. Function (ODI), pain intensity (VAS) and health-related quality of life (SF-36) were determined according to a patient-reported questionnaire. The questionnaire scores were compared with previously recorded 1-year outcomes and with questionnaire scores from a group of 28 patients who did not undergo SIJ fusion.

**Results** The patients who underwent SIJ fusion reported a mean ODI of 33 (95 % CI 24–42) and a mean VAS score of 54 (95 % CI 46–63) 23 years (range 19–34) after surgery. Regarding quality of life, the patients reported reduced physical function, but mental health was not affected in the same manner. The patients with successful 1-year outcomes (48 %) retained significantly improved function and reduced pain levels compared with the subgroup of patients with unsuccessful 1-year outcomes (28 %). The patients who underwent surgery did not differ from the non-surgery group in any outcome at the long term follow-up.

**Conclusions** Patients treated with SIJ fusion had moderate disability and pain 23 years after surgery, and the 1-year outcomes were sustained 23 years after surgery. Although many fused patients reported good outcome, this group did not differ from the comparable non-surgical group.
1.0 Introduction

1.1 Pelvic girdle pain

The sacroiliac joint (SIJ) can be a source of pain for 13-30% of patients with low back pain (LBP) (Vleeming et al. 2008) and for possibly an even greater proportion of patients suffering from “failed back surgery” (DePalma et al. 2011, Katz et al. 2003). This pain may be caused by a specific pathology of the joint (Bellamy et al. 1983), but the specific role of the SIJ in unspecific pelvic girdle pain (PGP) disorder remains unknown. PGP is a common complaint in pregnancy that can cause disability, and in some women, the complaint continues after delivery (Albert et al. 2002, Vleeming et al. 2008). The origin and diagnosis of PGP are also unclear because radiological findings are often absent, and the diagnostic criteria lack sufficient evidence. It has, however, become increasingly clear that the clinical presentation and disability level in patients with PGP differ from those in patients suffering from LBP (O’Sullivan and Beales 2007, Robinson et al. 2010).

1.1.1 Terminology

Many different terms have been used to describe pelvic pain (Wu et al. 2004), and some of these terms describe possible etiologies, such as "relaxation", "instability" and "arthropathy". Because the origin of pain in PGP is uncertain, these terms might be incorrect or misleading. To obtain a single term, the authors of the European guidelines for the diagnosis and treatment of pelvic girdle pain proposed the term PGP, together with the following definition (Vleeming et al. 2008).

"Pelvic girdle pain generally arises in relation to pregnancy, trauma, arthritis and osteoarthritis. Pain is experienced between the posterior iliac crest and the gluteal fold, particularly in the vicinity of the SIJ. The pain may radiate in the posterior thigh and can also occur in conjunction with/or separately in the symphysis. The endurance capacity for standing, walking, and sitting is diminished. The diagnosis of PGP can be reached after exclusion of lumbar causes. The pain or functional disturbances in relation to PGP must be reproducible by specific clinical tests."
This definition has been proposed for pelvic musculoskeletal pain to exclude gynecological and/or urological disorders and to promote consistent use of terminology.

### 1.1.2 Epidemiology

PGP is most commonly reported in pregnancy, but some patients also develop PGP after minor trauma or without any specific reason. The prevalence of PGP during pregnancy has been estimated to be approximately 20% (Vleeming et al. 2008), and approximately 50% of patients report LBP during pregnancy (Berg et al. 1988, Wu et al. 2004, Robinson et al. 2006, Ostgaard et al. 1991). Most women recover, but approximately 10-25% of women continue to have complaints after delivery (Albert et al. 2001, Wu et al. 2004, Larsen et al. 1999, Vleeming et al. 2008, Bjelland et al. 2013b), and approximately 5% suffer from pain that is sufficiently severe to require medical assistance (Wu et al. 2004). Furthermore, it seems that patients with symptoms in all three pelvic joints during pregnancy have an increased risk of suffering from disabling PGP 2 years after delivery compared to patients with pain in one or two joints (Albert et al. 2001). PGP occurs frequently during pregnancy and has a good prognosis of rapid regression of symptoms, but in some cases, the pain becomes long-lasting and debilitating.

The SIJ has also been suggested to be a possible source of pain in non-pregnant patients, such as patients with non-specific LBP. The prevalence reported has varied greatly and has been estimated to be somewhere between 10% and 62% (Simopoulos et al. 2012). Without a gold standard to diagnose the SIJ pain, the prevalence has been difficult to establish. Intra-articular SIJ injections have been used as a gold standard to estimate the prevalence of SIJ pain in patients with LBP. The effects of these injections have been reported to be positive or negative, however, with different cut-off values defining a positive test result. Some investigators used a single injection as the cut-off, whereas others did not define the test as positive if the test was not replicated with a control block (Simopoulos et al. 2012). The selection of patients is another factor that has had a large influence on prevalence. The lowest prevalence has been observed in unselected groups of patients with unspecific LBP, and in studies including patients with a probability of SIJ pain, the prevalence has increased. Although the prevalence varies, it is highly possible that the SIJ is a source of pain in patients with non-specific LBP, but without a gold standard, these numbers are uncertain.
1.1.3 Etiology

The etiology of PGP is poorly understood, but there is agreement that the cause is multi-factorial, and it must be viewed in a bio-psycho-social framework (O'Sullivan and Beales 2007). Many attempts have been undertaken to understand the origin of PGP, and the factors that are believed to be of importance include hormonal, genetic, psychological, neurophysiological, biomechanical, pathoanatomical and social factors (Kanakaris et al. 2011, O'Sullivan and Beales 2007).

All of these different factors can contribute to PGP, but the extent to which each factor contributes is likely different in each patient. The hormonal influence of relaxin and progesterone on the ligaments, with smoothening and relaxing effects, has been well established (Albert et al. 1997), but the association between hormone levels and PGP has been debated (Albert et al. 1997, Vollestad et al. 2012, Bjelland et al. 2013a). The sacropelvic ligaments have been of interest because many patients with PGP report tenderness in these ligaments (Torstensson et al. 2009, Palsson and Graven-Nielsen 2012). The relaxing effects of hormones on the ligaments during pregnancy have contributed to the biomechanical understanding or misunderstanding of pelvic relaxation and instability. Separation of the pubic symphysis has been used as an objective measurement of pelvic joint movement, and pubic movement has been reported to be greater in patients with PGP than in controls (Mens et al. 2009). However, the variations in the movements and the overlap in range between patients with and without PGP are too large to use these measurements as diagnostic tools. Although increased movement can be observed during and shortly after pregnancy, there has been no documentation of a correlation between sacroiliac mobility and symptoms in patients with long-term PGP (Sturesson et al. 1989, Vleeming et al. 2012). However, a correlation has been found between asymmetrical laxity of the SIJ and the intensity of symptoms (Damen et al. 2001). Because pelvic relaxation during pregnancy is a normal physiological response, and the majority of pregnant women do not experience pain, the importance of this minimal increase in pelvic joint movement is uncertain. Although many factors have been proposed to be important when PGP develops and persists, it has been suggested that pain caused by dysfunction in the SIJ is a plausible explanation (Ostgaard et al. 1991).

Optimal muscular control is important for stabilizing the SIJ as well as the entire pelvic girdle (Snijders et al. 1993), and a treatment program, including training in motor control, has been shown to reduce PGP (Stuge et al. 2004a, O'Sullivan and Beales 2007, Stuge et al. 2004b).

When researching PGP, the bio-psycho-social model should be applied because psychological and social factors also seem to contribute when a patient develops chronic pain syndrome. Psychological factors, such as emotional distress and catastrophizing during pregnancy, have been shown to increase the risk of PGP (Beales et al. 2009, Bjelland et al. 2013b, Olsson et al. 2012), and social factors, such as physically demanding work and inconvenient work hours, have also been found to increase the risk of PGP (Juhl et al. 2005). These findings are in contrast to the strict reductionist biological model of medicine, in which the disease can be explained by an underlying pathological process or a developmental abnormality.

### 1.2 Diagnostics

#### 1.2.1 Medical history

The clinical presentation of patients with PGP varies, but there are certain common characteristics. Often, the pain is located over the SIJ region and extends below the posterior spine, along the long dorsal ligament (Figure 4), deep into the gluteal region and into the pubic symphysis. The pain worsens with standing, sitting and walking (Vleeming et al. 2008, Wu et al. 2004), and many patients report "catching" of the leg (Sturesson et al. 1997). Although many patients have reported these clinical symptoms, the medical history alone has been reported to have limited value compared to an SIJ injection as the gold standard (Dreyfuss et al. 1996).

#### 1.2.2 Clinical tests

There have been many reports with very different results regarding the reliability and validity of clinical tests in the diagnosis of PGP. The main reason for this variation has been the lack of a gold standard with which to compare the tests (Vleeming et al. 2008). Dreyfuss et al. (1996) did not find any reasonable value of medical history or clinical testing compared to diagnostic SIJ block, but other researchers have found adequate sensitivity and specificity with clinical testing, particularly if multiple tests were used (Laslett et al. 2005a). In two
systematic reviews (van der Wurff et al. 2000b, van der Wurff et al. 2000a), the authors reported both the reliability and validity of clinical tests to be poor; however, later studies of higher quality reported more promising results (Laslett et al. 2006). These tests can be divided into provocation tests and functional tests (Vleeming et al. 2008).

Provocation tests aim to stress the SIJ and the surrounding ligaments and to trigger actual pain. Laslett (2005) emphasized that the test should be regarded as positive if it reproduces familiar pain. The use of multiple tests strengthens the probability of the diagnosis (Laslett et al. 2003, Laslett et al. 2005b, Slipman et al. 1998, Stanford and Burnham 2010, van der Wurff 2006, van der Wurff et al. 2006, Vleeming et al. 2008). The above-mentioned studies used 3 of 5-6 positive provocation tests as a cut-off and SIJ injection as the gold standard. The studies reported sensitivity of 82-94% and specificity ranging from 57% to 78%. As shown in Table 1, if only 1 positive test was chosen, the sensitivity was high because a patient with PGP most likely tested positive on one test. Because these tests loaded the SIJ in different ways, the patients most likely did not respond to all tests; consequently, the sensitivity decreased, together with an increased cut-off for positive clinical tests (Table 1). The specificity was low if only 1 test of 5 was positive, but if 5 of 5 tests were positive, the specificity was 88-100%.

Table 1
Sensitivity and specificity of combinations of tests

<table>
<thead>
<tr>
<th>Author</th>
<th>Tests</th>
<th>Number of positive tests</th>
<th>Se</th>
<th>Sp</th>
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<th>Sp</th>
<th>Se</th>
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<th>Se</th>
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<tr>
<td>Van der Wurff</td>
<td>1, 2, 3, 4, 5</td>
<td>&gt;50% reduction in VAS</td>
<td>100</td>
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Se=sensitivity, and Sp=specificity of clinical tests. Tests used: 1-Distraction, 2-Compression, 3-Thigh trust, 4-Patrick sign, 5-Gaenslen, 6-Sacral thrust, 7-Bilateral Gaenslen
To provoke the pubic symphysis pain, two tests have been used: the modified Trendelenburg test and palpation of the pubic symphysis (Albert et al. 2000, Vleeming et al. 2008). The modified Trendelenburg test is considered positive if the patient experiences pain in the symphysis when standing on one leg with the other hip in 90° of flexion (Albert et al. 2000). The sensitivity has been reported to be 40-62% and the specificity to be 99% (Vleeming et al. 2008). Gentle palpation of the symphysis, with pain 5 seconds after removal of the hand, has been shown to have sensitivity of 60-81% and specificity of 85-99% and to have good inter-examiner agreement (kappa=0.89) (Albert et al. 2000, Kristiansson and Svardsudd 1996).

The most widely used functional test is the active straight leg raise (ASLR) (Figure 1) (Mens et al. 1999). When Mens et al. (1999) applied this test in 200 patients with suspected PGP and
compared these patients to 50 healthy controls, they found, when a cut-off score between 0 and 1 was chosen, sensitivity of 0.87 and specificity of 0.94. The same test proved to be well-correlated with the severity of symptoms (Mens et al. 2002).

1.2.3 Radiological examinations

Computer tomography is the best examination for visualizing the bony anatomy of the SIJ, but one challenge is that radiological findings of degeneration can be observed in normal subjects as well as in patients with SIJ pain. In a study of 45 asymptomatic subjects, Vogler et al. (1984) reported symmetrical and normal CT scans in subjects younger than 30 years old, but with increasing age, the radiological findings changed. The joint space became less uniform, and subchondral sclerosis was observed. In the oldest participants (older than 59 years old), the investigators discovered high percentages of osteophytes, cysts, erosions and ankylosis. These findings were verified by Shibata et al. (2002), who reported joint space narrowing (87%), sclerosis (52%) and osteophytes (68%) in 190 patients who were asymptomatic for SIJ, with a higher prevalence with increasing age. Subsequently, Elgafy et al. (2001) compared degenerative CT findings with SIJ injections to examine the relationship between radiological findings and symptoms. Of 62 patients with positive SIJ injections, 42.5% had normal CT findings, and when these cases were compared to a control group of asymptomatic patients, the authors found sensitivity of 58% and specificity of 69% for CT. In summary, these CT findings could be found in patients with PGP and suspected SIJ pain, as well as in asymptomatic subjects, which limited the diagnostic value of CT scans for the diagnosis of PGP.

Magnetic resonance imaging has been reported to play an important role in diagnosing inflammatory SIJ pathology and particularly in diagnosing early changes in SIJ spondyloarthropathies (Puhakka et al. 2004b, Puhakka et al. 2004a, Vleeming et al. 2008). The use of MRI can be used to identify these changes, but it has primarily been used to exclude serious pathology.

Radionuclide bone scanning has also been used to diagnose SIJ pathology, but when scintigraphy is compared to SIJ injections, the sensitivity of the former has proven to be low. When scans were considered either positive or negative, Slipman et al. (1996) reported sensitivity of 13%, indicating that 87% of the patients with positive SIJ injections had
negative bone scans. Subsequently, Maigne et al. (1998) used quantitative radionuclide bone scans and found higher uptake in joints with positive SIJ injections, and they reported sensitivity of 46% and specificity of 89%. The radionuclide bone scan is therefore an inadequate tool for screening the SIJ as the origin of pain.

1.3 The sacroiliac joint

The SIJ has been considered one of many etiologies of PGP and LBP. Before herniated discs were discovered to be a cause of LBP, the SIJ was believed to play a central role as a pain generator. Subsequently, however, focus moved away from the SIJ toward the herniated disc. More recent injection studies have discovered that a significant proportion of LBP patients have SIJ pain (Schwarzer et al. 1995, Simopoulos et al. 2012), and the SIJ has also been suggested to be a significant contributor to failed back surgery (Katz et al. 2003, Maigne and Planchon 2005). Hence, the SIJ might play a role in the development of PGP, as well as having a potentially important role in patients with LBP.

1.3.1 The sacroiliac joint in historical perspective

There has been great interest in the SIJ in the medical literature, and according to several authors (Buchowski et al. 2005, Weisl 1955, Lynch 1920, Vleeming et al. 2012), the SIJ was first described by Hippocrates as a source of pain. Hippocrates described the "disjunctio pelvica" as a reason for pain during pregnancy, but it was originally believed that the SIJ only became mobile during pregnancy. Since the time of Hippocrates, debate has persisted regarding the degree of mobility of the SIJ. According to Weisl (1955), Dimerbroeck stated in 1689 that the SIJ was likely also mobile also in men and in women apart from pregnancy, and this fact was later confirmed by multiple cadaver studies (Weisl 1955). From these cadaver studies, evidence emerged that the SIJ is a synovial joint and therefore must move. Subsequent measurements of the true conjugate distance showed differences between different postures, indicating movement of the sacrum relative to the innominate bone (Von Schubert 1929, Weisl 1955). Later authors concluded that the movement of the SIJ was mostly rotational around an axis perpendicular to the joint surface.
In 1930, Chamberlain described an x-ray method for indirectly measuring SIJ movement. Because SIJ movements were assumed to be mostly rotational, the movement measured in the pubic symphysis was interpreted as an indirect measurement of SIJ movement (Chamberlain 1930). Chamberlain also found that the movement observed in the pubic symphysis, and indirectly in the SIJ, could be correlated with pain. This relationship between Chamberlain’s x-ray studies and pain was also reported by other researchers, but the findings were not consistent (Anderson and Peterson 1944, Mens et al. 1999). During this time period, the SIJ was a well-established cause of ischialgia and LBP, but it lost attention when Mixter and Barr (Mixter and Barr 1934) first described a ruptured intervertebral disc as a source of ischialgia. Despite Anderson’s (1944) statement, "There seems to be no question at the present time that the SIJ is a movable joint," and the convincing results of Weisl (1955) showing SIJ movement, interest in the SIJ declined. Subsequently, Solonen (1957) stated that because of the strong ligaments and irregular shape of the SIJ, the joint is immobile, and this observation was more or less considered to be true for a long period of time. In the 1980s, interest in the SIJ returned, and there were several studies during this period that attempted to establish and understand the biomechanical properties of the SIJ (Smidt et al. 1995, Smidt et al. 1997, Sturesson 1999, Sturesson et al. 1989, Sturesson et al. 1999, Sturesson et al. 2000b, Sturesson et al. 2000a, Weisl 1955).

1.3.2 Anatomy

The SIJ is a diarthrodial joint, but it is unique because the sacral surface has hyaline cartilage, and the ilial surface has fibrocartilage (Foley and Buschbacher 2006, Forst et al. 2006, Vleeming et al. 2012). The joint’s anatomy is variable between subjects with regard to its size and shape, and the joint changes over one’s lifetime (Vleeming et al. 1990, Vogler, III et al. 1984). The joint is L-shaped and is formed from the sacral bodies from S1 to S3. The joint line is smooth in childhood but becomes more irregular in adulthood, which minimizes movement (Vleeming et al. 2012). In addition to the irregular joint surface, primary stabilization of the joint is accomplished by different strong ligaments (Figure 3, Figure 4). The anterior ligament is more or less a thickening of the anterior capsule, and it is not as strong as the dorsal ligaments. The dorsal ligaments consist of different defined ligaments, of which the interosseous ligament is the strongest. This ligament is multidirectional and is an important stabilizer, allowing great forces to be transferred from the spine to the lower extremities. The other ligaments that stabilize this joint include the long and short dorsal
sacroiliac ligament, the sacrotuberous ligament and the iliolumbar ligament (Figure 3, Figure 4). The bony anatomy and these ligaments ensure that the pelvic girdle is not a rigid ring but instead works as a suspension mechanism that allows forces to be transferred without causing a fracture to the pelvis (Vleeming et al. 2012).

No muscles directly cross the SIJ, but the interactions of adjacent muscles and fascial structures are dynamic stabilizers of the SIJ. The biceps femoris, the gluteus maximus and the piriformis are connected to the ligaments around the SIJ and contribute to the functional stability of the joint (Snijders et al. 1993). Additionally, the pelvic floor muscles have been reported to add stiffness to the pelvic ring (Pool-Goudzwaard et al. 2004, Snijders et al. 1993). Furthermore, the deep abdominal muscles connecting to the thoracolumbar fascia are believed to contribute to the stability of the SIJ (Vleeming et al. 1995).
1.2.3 Stability of the SIJ

A biomechanical model has been created to describe the forces that contribute to the stability of the SIJ. Snijders et al. (1993) described a model based on the theory of form and force closure. Form closure refers to a situation in which the joint is stable, without any need for additional stabilizing forces, and form closure is a situation in which the joint is stabilized by friction and compression forces. The SIJ is believed to be stabilized by a combination of form closure (ridges and grooves in the SIJ) and force closure (ligaments and muscles) (Snijders et al. 1993, Vleeming et al. 1990) (Figure 5).

The range of motion in the SIJ is small, both in patients with PGP and in asymptomatic individuals (Goode et al. 2008, Vleeming et al. 2008, Vleeming et al. 2012). Hence, the stability of the SIJ is more closely related to how a load can be smoothly and effortlessly transferred across the SIJ than it is related to the degree of mobility. Non-optimal joint stability is defined in the European guidelines (Vleeming et al. 2008) as an "altered laxity or stiffness leading to new joint positioning and/or exaggerated/reduced joint compression, with a disturbed performance/effort ratio."
1.3.4 Innervation

The innervation of the SIJ has been reported in many studies, but there is no agreement regarding the exact innervation of the SIJ (Cohen 2005, Vleeming et al. 2013). In two systematic reviews (Cohen 2005, Vleeming et al. 2013), the innervation of the dorsal part of the joint was suggested to arise primarily from the lateral branches of L4-S3, although different authors have suggested that different levels are involved. The anterior part is assumed to be innervated by the ventral rami, varying from L2 to S4. Further immunohistochemical analyses have been performed on the ventral capsule, interosseous ligaments, cartilage and bone, and there has been evidence of sensory nerves in all of these structures (Szadek et al. 2008, Szadek et al. 2010). The presence of calcitonin gene-related peptide and substance P immunoreactive fibers has been believed to provide morphological and physiological bases for pain signals originating from these structures (Szadek et al. 2008, Szadek et al. 2010), which could be why SIJ injections have effects and might also be why fusion to the joint can be effective for alleviating PGP.

1.3.5 Referred pain

Referred pain has been reported to coexist with SIJ pain, and in a study of 25 patients with PGP, verified by positive SIJ injections, as many as 60% had either thigh or leg pain (Laplante et al. 2012). In another study of 50 patients, the authors found buttock pain in 94% of the patients and referred pain to the leg in more than 50% of the cases but with 18 different
pain distributions (Slipman et al. 2000). Before the herniated disc was discovered, the SIJ was regarded as an important etiology of sciatica, and it has been questioned whether nerves can be affected by disturbances in the SIJ (Fortin et al. 1994). Using arthrography, extravasation of contrast agent has been observed in many subjects, and different pathways between the SIJ and neural structures have been identified. Fortin et al. (1994) reported 61% SIJ extravasation in 76 injections, and these cases followed 5 patterns; ventral (16%), dorsal to the first sacral foramen (8%), dorsal sub-ligamentous (24%), superior (3%) and inferior (12%) to the sacral ala. Ventral extravasation of inflammatory agent could theoretically affect the lumbosacral plexus and S1 foramen all the way up to the L5 foramen. The neurotransmitter substance P has been identified as a possible cause of “neurogenic inflammation”. The SIJ is innervated and can therefore be a pain generator. Different referred pain patterns have been observed and can be explained by individual variations in innervation, direct nerve involvement or different sclerotomes (Slipman et al. 2000).

1.3.6 Biomechanical considerations of SIJ movement

"Interestingly, studies that demonstrated the highest levels of quality and that offered the lowest levels of error in measurement also reported the lowest values [of movement] available at the SIJ."

Adam Goode, 2008

As mentioned in the historical overview, several attempts have been undertaken to establish movement in the SIJ, both in healthy subjects and in patients with PGP. Many different techniques have been used, such as cadaver studies, studies using different markers (skin markers, palpation of the bony landmarks and k-wires) and radiological studies (x-ray, CT, RSA) (Hungerford et al. 2004, Hungerford et al. 2007, Jacob and Kissling 1995, Lavignolle et al. 1983, Smidt et al. 1995, Smidt et al. 1997, Sturesson 1999, Brunner et al. 1991, Vleeming et al. 1992a, Sturesson et al. 1989, Sturesson et al. 1999, Sturesson et al. 2000b, Sturesson et al. 2000a). All of these techniques have obvious advantages and disadvantages. The cadaver studies lacked muscular influence on stabilization, and the sample tended to come from an older population. The different experimental settings have different levels of precision and accuracy, and it seems that the methods with the best precision have the lowest measured SIJ motion (Goode et al. 2008). Although the literature regarding analysis of movement has reported various results, there are some points on which these reports have generally agreed.

2. There are different theories regarding the motion of the sacrum relative to the innominate bone and the center of rotation. Because the sacrum is L-shaped and has an irregular surface, and because there is a large variation among subjects, a fixed center of rotation has been difficult to find (Walker 1992). In Figure 6, different theories that have been proposed are presented: (a) sacral tilt with the center of rotation inside the SIJ; (b) sacral rotation with the center of rotation located immediately dorsal to the SIJ; (c) rotation with the center of rotation in front of the SIJ; and finally, (d) translation with no rotation (Alderink 1990). The evidence is not in agreement regarding this subject, but the strongest evidence has supported that the center of rotation is most likely located dorsal to the SIJ as a transverse axis and in close proximity to the iliac tuberosity (Figure 6b) (Brunner et al. 1991, Vleeming et al. 1992b, Egund et al. 1978, Jacob and Kissling 1995), although there are likely large individual differences.

3. The movements in the SIJ are small, and the total rotation has varied in different studies but has seldom exceeded a mean value of 2° (Egund et al. 1978, Goode et al. 2008, Jacob and Kissling 1995, Vleeming et al. 1992a, Vleeming et al. 2012). This movement has seemed to be greater in an unloaded pelvis than in a loaded pelvis (Goode et al. 2008, Sturesson et al. 1989, Sturesson et al. 2000b, Sturesson et al. 2000a).

4. There do not seem to be differences in movement between symptomatic and asymptomatic SIJs (Sturesson et al. 1989).

5. There is evidence that women tend to have greater mobility than men. In healthy volunteers, Jacobs (1990) did not find any differences in SIJ movement with regard to age, sex or parturition. Other studies have reported less movement in men than in women (Brunner et al. 1991, Bussey et al. 2009, Sturesson et al. 1989). It also seems that multiparous women have greater movement of the pelvic joints than nulliparous and men (Garras et al. 2008, Mens et al. 2009).
To measure SIJ movement accurately, the RSA technique has been applied to the SIJ. One-millimeter markers were implanted in patients, and with a specialized x-ray set-up and a computer program, the \textit{in vivo} movement could be measured with high precision (Sturesson et al. 1989). These markers were attached to a segment in each ilium and to one in the sacrum, and the movement between these segments was then measured (for a more detailed description, see section 4.5). The RSA studies have, in general, reported less movement than other studies using methods with questionable precision (Goode et al. 2008), and because the RSA showed less motion than other methods, the RSA method has been questioned. The RSA studies have measured movement between the sacrum and the ilium with dorsally placed RSA markers, and the markers were placed near the joint line. Because of the flat anatomy of the
bones, the markers became collinear (in the same plane, which is not necessarily the optimal 3D distribution of the markers (Cibulka 2001).

Guidelines for the standardization of RSA of implants have recommended at least three non-collinear RSA markers in each segment (rigid body), which should be compared to one another (Valstar et al. 2005). A good 3D configuration of the segments relies on the distance between the markers and the distribution of the markers on all three axes; a condition number (CN) expresses the quality of a marker segment (Makin 2004). The CN is a mathematical expression of how the markers relate to a straight line that passes through the segment (Ryd et al. 2000). A low CN represents a good scatter of markers in the segment. A CN below 110 is considered a reliable distribution (Valstar et al. 2005), and an upper limit of 150 is suggested. This CN will consequently influence the precision and accuracy.

Additionally, another factor of importance is how well the RSA computer identifies and calculates the placement of each individual marker. The precision of each marker can be influenced by soft tissue disturbances and by the stability of the markers. If the markers are not thoroughly inserted into the bone and end up in the soft tissue, the markers can become unstable. This instability can occur in the sacrum because of the thick and strong dorsal ligaments covering the bone, particularly in the cranial portion. To ensure including only stable markers in the analysis, unstable markers should be excluded if they move more than 0.35 mm between two examinations (ME; mean error of rigid body fitting) (Valstar et al. 2005). Uncertainties with the RSA method, when applied to the pelvic joints, were addressed in a letter to the editor by Cibulka (2001), in which he asked:

"I question whether using this sort of marker arrangement can accurately define the fixed segments (especially the innominate bones) and therefore truly describe sacroiliac joint motion."

"Would a different configuration (e.g., wider distribution) of pelvic markers show different results?" (Cibulka 2001)

These questions formed the basis for our first research question: What are the accuracy and precision of RSA when applied to the SIJ, and was the marker distribution used in the available RSA studies useful?
1.3.7 The Chamberlain technique

"The place to look for evidence of sacroiliac joint motion is at the symphysis pubis, where it is magnified and measurable."

Chamberlain (cited in Andersson 1944)

All of the experimental techniques used to quantify SIJ movement have been impractical in clinical practice. In 1930, Chamberlain described an easy and practical method for measuring pubic movement on anterior-posterior (AP) pelvic x-rays while the patient stood on one leg with the other leg hanging down (single-leg stance) (Chamberlain 1930) (Figure 7). In patients with SIJ pain, Chamberlain found that weight bearing caused cranial displacement of the pubic bone to the side of the painful joint. This displacement was explained by rotation around the axis that was perpendicular to the SIJ surface. The Chamberlain technique has since been used to examine pubic bone movement and, indirectly, SIJ hyper-mobility (Mens et al. 1999).

Since the Chamberlain technique was first described, researchers have attempted to correlate pubic movement with SIJ pain (Anderson and Peterson 1944, Siegel et al. 2008, Mens et al. 1999). Chamberlain found a clear pattern in his patients, but Mens et al. (2009) subsequently found the exact opposite pattern, in which the hanging leg caused downward displacement of
the pubic bone on the side of the painful joint. These differences have made it difficult for clinicians to use the results of this test in the diagnosis of PGP, particularly when normal variations in the movement of the pubic symphysis have proved to be large (Garras et al. 2008). Measurements of the movement of the SIJ with the subject in the single-leg stance have been obtained using k-wires; however, those authors only measured healthy subjects without SIJ pain (Jacob and Kissling 1995). Because the Chamberlain technique is an indirect measurement of SIJ movement, what really occurs in the SIJ during the single-leg stance test in patients with PGP remains unknown.

This uncertainty formed the basis for our second research question: What is the movement in the SIJ during the single-leg stance in patients with severe PGP?

### 1.4 SIJ fusion as a treatment for SIJ pain

"Cases of relaxation of the sacroiliac joint which have had the above type of arthrodesis performed have been uniformly successful."

Smith-Peterson, 1921

The role of the SIJ as a pain generator has interested orthopedic surgeons for almost a century. Smith-Petersen described a method for SIJ fusion in 1921 (Smith-Peterson MN 1921), and since then, several different attempts have been made to select and operate on patients with suspected SIJ pain. In the beginning, a large proportion of the patients had joint infections (especially tuberculosis), and many of the first surgical techniques were developed to treat these infections, but patients with "pelvic relaxation" during pregnancy have also comprised a large proportion of the patients receiving SIJ fusion (Hagen 1974, Smith-Petersen and Rogers 1926, Smith-Peterson MN 1921) (Appendix 1). SIJ fusion followed the same popularity curve as the knowledge of SIJ movement, most likely because movement, pain and fusion are closely related in an orthopedic surgeon's mind. If there is mobility that causes pain, fusion can cure the pain. Before the herniated disc was discovered, SIJ fusion was a novel treatment for low back pain and disruption after pregnancy, and the treatment was described in several case series in the period from 1921 to the 1940s (Appendix 1). After the 1940s, there were no papers in the literature until the 1970s, when new reports of SIJ fusion started to appear. In the last few years, the role of the SIJ in orthopedic surgery has again been gaining popularity.
Almost a century has passed since Smith-Petersen published his experiences with SIJ fusion in 1921. Despite this long history, only a few papers can be found in the literature, and to locate these papers, searches were conducted in Medline/Ovid, Embase and Google Scholar, combining the terms "sacroiliac joint", "arthrodesis" and "fusion". From the articles retrieved from this search, all of the articles and references were cross-checked to find further possible evidence. The results included only 30 papers and book chapters (Appendix 1).

When planning the study in 2005-2006, only 18 papers were available, describing 277 patients and 13 different surgical techniques. Although many different techniques were used, they all involved either open anterior or open dorsal fusion. In the beginning (1921-1940), there were mostly descriptions of these new surgical techniques and short descriptions of the results obtained for the first patients. In these materials, a large proportion of the patients were surgically treated for infections or for SIJ arthritis. The authors reported, in general, excellent to good outcomes in 50-60% of the patients, fair results in 20% and poor results in 20%. No further documentation was found from between 1941 and 1974, likely because the SIJ lost attention to the herniated disc. Until 2006, there were only case series, and only 4 of these 18 series were prospective registrations of outcomes. Only one larger study was available when we started our investigation, and this large study did not actually evaluate SIJ fusion but instead fixation with SIJ screws and without fusion (van Zwienen et al. 2004). All of these studies had short follow-up periods, except for one that had 5.8 years of follow-up (Buchowski et al. 2005). Since 2006, 12 more case series have been published, all of which were designed as retrospective reviews of prospective registered outcome measurements, and these studies are referred to as prospective in the appendix because the outcome measures were collected pre- and post-operatively. These studies reported the surgical outcomes of 354 patients, and all of these reports were the results of minimally invasive surgery (MIS).

The diagnostic criteria and the criteria for surgery are not standardized, and surgery for PGP is controversial. There have been few studies and only limited knowledge about this treatment option (Appendix 1). Hence, it is difficult for healthcare providers to give proper advice to patients with PGP regarding surgery.

1.4.1 History of pelvic joint fusions in Norway

In 1974 orthopaedic surgeon Rolf Hagen, Martina Hansens Hospital, published his experience with conservative and surgical treatment of 23 patients with SIJ pain. Over a 20 year period
from 1951-71, eight patients were operated on with the surgical technique described by Smith-Peterson (Hagen 1974). Six out of these 8 had a good result, 1 had a fair result and 1 did not have any effect at all. From the middle of 1970's to late 1990's a few orthopedic surgeons in Norway performed SIJ fusions and some also did fusion to the pubic symphysis. Especially, orthopaedic surgeon Einar Sudmann at Hagavik Hospital, developed a systematic approach to the SIJ problem, as he registered the surgical outcome of 81 patients together with complications in a database. Einar Sudmann and the rest of the orthopaedic surgeons performing SIJ fusions did however not achieve the results they wanted. Because the outcomes were unpredictable, the complication rate appeared unacceptably high and the need for additional spinal surgery, the enthusiasm diminished. During the 1990's most orthopedic surgeons had stopped performing SIJ fusions except professor Olav Røise, orthopedic pelvic trauma surgeon at Oslo University hospital. In the early 2000s the medical literature on surgical treatment of PGP was sparse and without high quality studies, and because of this professor Røise decided to stop doing the surgery until a study protocol was established. In 2004 a pilot study with 4 patients was conducted and in 2005 Olav Røise together with Britt Stuge, PT, PhD, Finnur Snorrason, MD, PhD and May Arna Risberg, PT, professor at OUS, started to plan this project.

The lack of documentation regarding the results after SIJ fusion and the fact that Sudmann performed SIJ fusion on more than 80 patients between 1977 and 1998 led us to the last two research questions:
What are the outcomes of SIJ fusion, and what are the long-term results after SIJ fusion?
2.0 Aims of the thesis

The main questions and aims of the thesis are:

**Paper I**
Is the RSA method valid to measure pelvic movement?
The aims were to (1) measure the accuracy, precision, and condition numbers of pelvic RSA with different marker distributions in a phantom model, (2) explore whether frontal markers around the symphysis improve the condition number and precision and whether it is possible to avoid markers in the cranial part of the sacrum, and (3) to compare the precision obtained by a phantom with the precision in patients.

**Paper II**
What is the movement in the SIJ during the single-leg-stance test?
The aims were to (1) measure movement in the SIJs during the single-leg stance test by using RSA, in patients with severe PGP and to (2) identify whether there were any differences between movements in the SIJs of the standing leg and the hanging leg.

**Paper III**
What is the outcome of unilateral anterior SIJ fusion combined with fusion of the pubic symphysis?
The primary aim of this prospective study was to examine changes in pain and physical function at 3, 6, and 12 months after SIJ fusion. The secondary aims were to evaluate post-operative health-related quality of life and patient satisfaction with treatment.

**Paper IV**
What are the long-term results of SIJ fusion?
The main purpose was to evaluate long-term functioning, pain and health-related quality of life (HRQoL) in patients who had previously undergone pelvic joint fusion surgery. Further aims were to compare the 1-year outcomes with the long-term results and to compare patients who underwent surgery with PGP patients who did not undergo surgery.
3.0 Materials

3.1 Patients

Papers I, II and III

The patients in papers I, II and III consisted of patients from the same cohort. Originally, we planned to include and operate on 10 patients at Oslo University Hospital, Norway, and 10 patients at Ängelholm Hospital, Sweden, during the inclusion period from 2007 to 2010. In Norway, 20 patients were examined, but only 9 met the criteria for participation (Figure 8). The patients were included according to the criteria provided in Table 2.

Table 2
Inclusion and exclusion criteria

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<td>2. Minimum two positive out of five clinical tests:</td>
<td>2. Other spine pathology</td>
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<td>* Posterior pelvic pain provocation test (P4) test</td>
<td>3. CT-verified ankylosis at baseline</td>
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<td>* Active straight leg raise (ASLR) test</td>
<td>4. Body mass index over 30</td>
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<td>* Palpation of the long dorsal sacroiliac-ligament</td>
<td></td>
</tr>
<tr>
<td>* Modified Trendelenburg test</td>
<td></td>
</tr>
<tr>
<td>* Palpation of the symphysis</td>
<td></td>
</tr>
<tr>
<td>3. High pain and disability score</td>
<td></td>
</tr>
<tr>
<td>* Visual analogue scale &gt;50 and/or</td>
<td></td>
</tr>
<tr>
<td>* Oswestry disability index &gt;40</td>
<td></td>
</tr>
<tr>
<td>4. The patients should have performed adequate physiotherapy over time without positive effects</td>
<td></td>
</tr>
</tbody>
</table>

In Sweden, 10 patients were also selected for surgery, but unfortunately, there were problems with the collection of questionnaires, primarily due to large administrative changes and the loss of key personnel in the Orthopedic Department at Ängelholm Hospital. Some preoperative and 1-year data were available, but the collection of questionnaires was incomplete. Hence, these patients were excluded from paper III.

All of the Swedish patients, however, had RSA markers implanted and were thus available for the RSA study (paper II). After evaluating the RSA data from all of the patients, six patients were excluded because of poor x-ray quality, so 11 patients were eligible for inclusion in paper II (Figure 8). The patients were excluded because of misplaced markers in the soft
tissue or insufficient visualization of the markers on radiographs during the software analysis. In paper I, we were able to use RSA pictures from 6 of the 9 Norwegian patients (Figure 8). The Swedish patients did not undergo double examinations; therefore, they were not included in paper I.

Figure 8
Flow chart of patients in study I, III and IV.
Paper IV

The patients in paper IV were operated on at Hagavik Orthopaedic Hospital between 1977 and 1998 by Sudmann and co-workers. The data came from 50 subjects at 1 year after SIJ fusion and from long-term follow-ups. Eighty-one patients underwent SIJ fusion during this period. These patients were registered in a database and were asked in 2009 to participate in long-term follow-up. The study population is described in Figure 9. During the 1990s, the surgeons became increasingly reluctant to perform SIJ fusion, and a number of patients were refused surgery. Twenty-eight of these patients constituted the non-surgery group.

![Flow chart of patients in paper IV](image-url)
The patients were selected and operated on primarily by one of the authors (E.S.). The criteria for surgery were based on the patient history and on radiological and clinical examinations. The inclusion criteria were pain in the SIJ >1 year after pregnancy or trauma, pain with an idiopathic origin, severe disability and resistance to conservative treatment. The clinical tests performed included tenderness at the superior and inferior posterior iliac spines, active and passive straight leg raise tests, Patrick Faber’s test, passive hip rotation, forcible inward rotation and extension of the hip joint. Further tests included normal neurological and gynecological exams, normal spinal x-rays, symphysis movement of less than 3 mm on plain radiographs during a one-leg stance, normal radiculography, negative rheumatology tests and negative blood tests.
4.0 Methods

4.1 Design

Paper I
In paper I, the accuracy and precision of pelvic RSA were evaluated in an experimental setting using a phantom model, and the precision was also measured *in vivo* by double examinations. We used a plastic pelvic phantom (Sawbones® 1301; Pacific Research Laboratories, Inc., Vashon, WA, USA) attached to a micrometer to measure the true value of movement, and these values were compared to the values obtained from the RSA measurements. Because the number of markers needed has been questioned, the accuracy and precision were also evaluated with different numbers and distributions of markers.

Paper II
In paper II, we used RSA to measure the *in vivo* movement of the SIJ in patients with PGP in the single-leg stance.

Paper III
In paper III, a single-subject research design was used to evaluate the individual response and outcomes of pain, disability and health-related quality of life after SIJ fusion. The use of multiple measurements, at baseline and after the intervention, allowed us to consider the patients as their own controls. Five data collection sessions were conducted in each of the following 4 phases: prior to surgery (baseline) and at 3, 6, and 12 months after surgery.

*Single-subject research design (SSRD)*
A randomized controlled trial is the gold standard for examining the effects of an intervention. Because SIJ fusion is performed on few patients, a single-center randomized controlled design was difficult to apply due to the small number of available participants. Single-subject research designs (SSRDS), however, have been recommended as useful for examining clinical accountability (Engel RJ and Schutt RK 2009). If properly applied, an SSRD can provide a systematic approach for documenting clinical changes and can also provide evidence regarding the efficacy of a treatment modality (Engel RJ and Schutt RK 2009). SSRD refers to a study of a single patient or a small number of patients observed over time, during which
the treatment and outcome variables are controlled. The design consists of multiple measurements before (baseline) and at different phases after the intervention (Engel RJ and Schutt RK 2009). The data are then presented graphically with a mean value in each phase, and the changes between the phases are shifts in level (Figure 10).

![Graphical presentation of data](image)

**Figure 10**
Example of graphical presentation of data from one patient. Mean value (dotted line); all measurements in each phase (black line); the four phases (phase 1 before the intervention and phases 2, 3 and 4 after the surgery).

An SSRD focuses on individual responses and repeated measures, which improve the validity of the study. When an SSRD is replicated across patients, the internal and external validity is strengthened, allowing inferences to be made about effectiveness (Gonnella 1989, Logan et al. 2008, Zhan and Ottenbacher 2001).

**Paper IV**

Paper IV was a cross-sectional study of the patients who underwent SIJ fusion at Hagavik Hospital in Norway between 1977 and 1998, and the data consist of surgical results 1 year after surgery and over a long-term follow-up. In 2009, all of the eligible patients (Figure 9) received by mail invitations to participate and a questionnaire. The long-term outcomes of the
patients who underwent SIJ fusion were compared to those of a semi-matched group of patients who did not undergo this surgery. At 1 year, the surgeon graded each joint as good, fair or poor. The clinical outcomes were graded according to the following criteria. A joint with negative SIJ tests and no or minor pain that did not interfere with the patient’s work was graded “good”. A joint with obvious improvement compared to the pre-operative status and little pain but with pain that interfered with work (professional or at home) was graded “fair”. A joint was graded “poor” if there was no relief from pain or if the joint deteriorated after surgery. In cases of bilateral surgery, each of the patient’s joints could receive a different grade. According to the grading of the joints one year after surgery, the patients were allocated to three different subgroups (Figure 11).

![Diagram showing short-term grading of specific joints and patient subgroups.]

Figure 11
Each joint was graded after 1 year as good, fair or poor. For comparison purposes, three different subgroups were created based on this 1-year grading.
Twenty-four patients (48%) had all of their joints classified as “good” and were assigned to the “successful” subgroup. Fourteen patients (28%) had at least one joint classified as “poor” and were assigned to the “unsuccessful” subgroup. Twelve patients (24%) had their worse joint scored as “fair” and represented the “partly successful” subgroup (Figure 11). These three subgroups formed the baseline for comparing the long-term effects. The patients’ 1-year outcomes were prospectively registered in a DOS Advanced Revelation relational database by the surgeons responsible for the operations.

4.2 Data collection

Papers I, II and III

The patients in papers I, II and III were included after a baseline evaluation. At baseline, a clinical evaluation was performed, and the patients completed questionnaires. It has previously been shown that female patients with PGP have variations in pain intensity during the menstrual cycle, with a relapse around menstruation (Mens et al. 1996). For this reason, the patients completed a questionnaire every Thursday for 5 weeks during each phase, to ensure that the evaluations were performed throughout the entire menstrual cycle (Figure 12). The questionnaires were returned weekly by mail. All of the patients underwent 3 clinical examinations, and in all but two cases, CT-guided SIJ injections were administered before the decision to perform SIJ fusion was made. The CT-guided injections were administered by two experienced radiologists, and the patients filled out a VAS scale before and at 2 hours after the injections. The SIJ injections were not used as inclusion criteria but rather as one of several factors to strengthen the diagnosis before surgery. The patients underwent surgery to fuse the more painful SIJ, and the pubic symphysis was operated on in all of the cases.

RSA images were obtained pre-operatively and after 3, 6 and 12 months, but the pre-operative images were used in papers I and II because a pre-operative clinical test (single-leg stance) was evaluated.
Paper IV

In paper IV, the patients received a similar questionnaire to that of the patients in paper III. They completed it and returned it in a pre-paid envelope.

4.3 Outcome measures

In papers III and IV, the patients completed a questionnaire consisting of the Norwegian versions of the Oswestry disability index (ODI), a visual analog scale (VAS) and the short form-36 (SF-36). Our main outcome was the ODI, and the secondary outcomes were the VAS, the SF-36 and self-reported satisfaction with treatment. In paper III, the patients also registered their pain distribution on a pain diagram.

4.3.1 Oswestry disability index (ODI)

The ODI was initiated by John O'Brian in 1976, and it has become one of the most commonly used condition-specific outcome measures for patients with LBP (Fairbank and Pynsent 2000). The questionnaire measures limitations in various activities of daily living, and the disability is graded using 10 items, for a total score ranging from 0 to 100. Each item consists of 6 statements, and these statements are scored from 0 to 5, with 0 indicating normal function and 5 a grade of high disability. A maximum score of 50 can be achieved, and this score sum is doubled and expressed as a percentage. A high score indicates a high grade of disability,
and a 10-point difference represents a significant clinical change (Fairbank and Pynsent 2000, Hagg et al. 2003). The Norwegian version of the questionnaire was used (Grotle et al. 2003). The ODI was tested for test-retest reliability, and at 24 hours, the reliability was 0.99. When the interval was increased to 4 days and one week, the test-retest reliability values were 0.91 and 0.83, respectively (Fairbank and Pynsent 2000).

4.3.2 Visual analogue scale (VAS)

Each patient’s most severe morning and evening pain intensity was assessed using a 100 mm VAS (0=no pain, 100=worst possible pain) (Revill et al. 1976). The patients answered two questions: (1) How severe is your pain in the morning, immediately after you leave your bed? and (2) How severe is your pain in the evening, immediately before you go to bed? The VAS was found to be sensitive to changes in pain intensity, and it has been validated for this use (Hagg et al. 2003, Von et al. 2000).

4.3.3 Pain diagrams

In addition to the VAS, a pain diagram was used to localize the pain and pain referrals (Figure 13), and when used as a pain locator, it has shown reliable results (Ohlund et al. 1996). The patients were asked to draw a cross where they experienced pain.

![Pain diagram](image)

**Figure 13**
Pain diagram. The patients were asked to draw crosses to describe and localize their pain.
4.3.4 Short form-36 (SF-36)

Health-related quality of life was assessed using the Norwegian version of the SF-36 (Loge et al. 1998). This questionnaire is divided into 8 sub-scales: physical function, physical role, bodily pain, generic health, vitality, social function, emotional role and mental health. The score is converted to a 0-100 scale for each of these 8 items, and a high score indicates good health status.

4.3.5 Self-reported satisfaction with treatment

Additionally, the patients answered the following two questions: "Have you experienced any effects of the surgery? If so, would you grade these effects as excellent, good, some, minor or no effects?" and “How do you tolerate physical activity now, compared to before surgery?”

4.4 Surgical intervention

All of the patients in paper III received unilateral SIJ fusion combined with symphysiodesis. An anterior approach with a skin incision over the iliac crest was used to reach the SIJ. The joint was partially resected, and the bone was grafted with cancellous bone from the ipsilateral iliac crest. Two AO (Arbeitsgemeinschaft für Osteosynthesefragen) reconstruction plates or AO-DC plates (Synthes®, Synthes GmbH, Switzerland) were used (Figure 14) to achieve stabilization. The pubic symphysis was accessed through a bikini line incision. A 2 × 2 cm bone block was removed and replaced with a bone graft from the iliac crest, and a Matta plate was applied (Figure 14). Post-operatively, the patients received epidural anesthesia pain relief and 1-2 days of wound drainage. The patients were advised to avoid full weight-bearing activities for 8 weeks after the surgery.

The patients in paper IV were operated on using a dorsal approach, with either trans-iliac fusion or intra/extra-articular fusion between the ilium and the sacrum. When the trans-iliac fusion was performed, an iliac window was constructed to access the joint (Smith-Peterson MN 1921). The joint surface was cleared of cartilage and was decorticated. The cortical iliac window was used as a graft and was typically hammered into the sacrum to promote intra-articular bone formation and conduction. Additional cancellous bone was compacted around the cortical graft. In dorsal intra/extra-articular fusion, iliac crest autografts were added after
joint removal and bone decortication (Waisbrod et al. 1987). The pubic symphysis was fused in four patients using an open technique, with an iliac crest block bone autograft and plating.

4.5 Radiostereometric analysis (RSA)

RSA was invented by Selvik in 1974 and has primarily been used to measure the 3-dimensional motion of implants and to obtain measurements of implant wear. Small tantalum (1 mm) markers were implanted into the bone segments under general anesthesia through a small skin incision in the dorsal part of the sacrum and in both ilia.

Both in the phantom and in the patients, approximately 8 markers were inserted into each bony segment (Figure 15), and the movement between these marker segments was later measured. The calibration cage contained markers, so the markers in the patients could be assigned to a 3D coordinate system (Figure 16). Two to 3 weeks after RSA x-rays were

Figure 14
X-ray of a unilateral anterior fusion combined with fusion to the pubic symphysis.
obtained with two angulated x-ray tubes (approximately 40°). The x-ray films were placed behind a calibration cage (Figure 17).

Figure 15
Markers placed in the sacrum and the innominate bone. The markers represent a marker segment, and the movement between these segments is calculated by the computer.

Figure 16
The 3D coordinate system. The rotational movements are Euler angles around the axis of this coordinate system.
4.5.1 Accuracy and precision of the pelvic RSA

Paper I

A good 3-D configuration relies on the distance between the markers and the distribution of the markers on all three axes, and a condition number (CN) expresses the quality of a marker segment (Mäkinen et al. 2004). Guidelines for standardization of the RSA of implants have recommended at least three noncollinear markers in each segment (rigid body) (Valstar et al. 2005). The CN is a mathematical expression of how the markers relate to a straight line passing through the segment (Ryd et al. 2000). A low CN represents a good distribution of markers in the segment. A CN of less than 110 is considered a reliable distribution (Valstar et al. 2005), and an upper limit of 150 has been recommended. The CN will influence the precision and accuracy, and a factor of importance is how well the RSA system calculates the placement of each marker. The precision of each marker can be influenced by soft tissue disturbances and by the stability of the markers. If the markers are not thoroughly inserted into the bone and are partially or completely in the soft tissue, the markers can become unstable. This instability can occur in the sacrum because of the thick and strong dorsal...
interosseous ligaments covering the bone, particularly in the cranial part. Unstable markers should be excluded if they move more than 0.35 mm between two examinations (mean error [ME] of rigid body fitting) (Valstar et al. 2005).

The accuracy and precision of RSA have been tested in different settings and have been reported to be high. Despite the use of pelvic RSA in clinical research, the accuracy and precision have not been fully evaluated. The accuracy of the measurement is the closeness of the measurement to its true value. In phantom models, accuracy reflects the level of agreement between the true value of movement and the results obtained using RSA. Systematic error (bias) of the system occurs when the differences between systems and experiments are uniform (Ranstam et al. 2000). In paper I, the true value of movement was measured with a phantom attached to a micrometer (Figure 18), and these values were compared to the measurements from pelvic RSA. This value should ideally be zero.

Figure 18
The setup of the pelvic phantom with the sacrum attached to a translation stage and a rotation rod are shown for (A) Y rotation and all translations, (B) X rotation, and (C) Z rotations.
The precision (spread) of the measurement is the degree of closeness of repeated measurements under unchanged conditions. Under optimal conditions, the difference between two examinations should be zero (Ranstam et al. 2000). The precision was measured in the phantom model and was evaluated in the patients using 17 double examinations in six patients.

To evaluate the influence of different marker distributions on accuracy and precision, the eight markers in each segment were divided into four different marker segments (Figure 19). We wanted to determine whether the markers in the pubic symphysis could increase the precision and how many markers were needed in each segment to achieve precision and accuracy. To examine the need for frontal markers, we performed tests with and without frontal markers. To examine the need for cranial markers in the sacrum, we performed tests with and without these markers. Finally, three dorsal markers in the ilium and three markers in the sacrum were randomly selected (Figure 19) and were tested against eight markers.

**Figure 19**
The RSA markers were divided into different marker segments (MS): MS A = five dorsal markers in the ilium; MS B = three frontal markers in the inferior pubic ramus; MS C = six sacral markers; and MS D = two cranial markers in the sacrum. Circles = three randomly selected markers in the ilium and three randomly selected markers in the sacrum.
In paper II, three pairs of images were obtained: one with the subject standing on both feet, one with the subject standing on the right foot and one with the subject standing on the left foot (Figure 20). The sacrum was defined as the fixed segment, and the movement of the innominate bone was relative to the sacrum.

Figure 20
RSA setup. A- Patient standing on both feet, B - Patient standing on left leg with the right leg hanging down, C - Patient standing on right leg with the left leg hanging down.
5.0 Main results

Paper I

Overall, the RSA method applied to the SIJ was found to be accurate and to have high precision.

Accuracy

When we applied translations and rotations to the micrometer, the RSA had good ability to detect these movements. With 8 markers in both the sacrum and ilium, the mean accuracy was between -0.03° and 0.05° for the rotations (Table 3). For translation, there was a small underestimation in the Z-direction of 0.07 mm. The resolution of the micrometer was 0.04° and 0.01 mm; therefore, these deviations from zero were not sufficiently large to be considered a systematic error that required correction.

<table>
<thead>
<tr>
<th>Table 3</th>
<th>Accuracy of the phantom</th>
</tr>
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<tbody>
<tr>
<td></td>
<td>Bias (mm)</td>
</tr>
<tr>
<td>Rotations X</td>
<td>-0.025</td>
</tr>
<tr>
<td>Y</td>
<td>0.051</td>
</tr>
<tr>
<td>Z</td>
<td>0.003</td>
</tr>
<tr>
<td>Translations X</td>
<td>-0.003</td>
</tr>
<tr>
<td>Y</td>
<td>-0.012</td>
</tr>
<tr>
<td>Z</td>
<td>0.065</td>
</tr>
</tbody>
</table>

| Accuracy expressed by bias; Bias=mean of the difference between the true value (TV) and the measured value (MV); SD_{bias}=standard deviation of the difference between TV and MV; 95% CI=95% confidence interval for the bias. |

Precision

For the RSA measurements in the phantom, there was high precision in all of the analyses, from 8 markers in each segment down to 3 markers in each segment. The precision (LOS) in the phantom was between 0.06° and 0.25° for rotation and between 0.03 mm and 0.16 mm for translation with 8 markers in each segment; with 3 markers in each segment, the precision was between 0.14° and 0.26° and between 0.09 mm and 0.21 mm. The precision in the patients was lower than in the phantom, with precision for rotation between 0.2° and 0.7° and for translation between 0.3 mm and 0.5 mm.
Analysis of different marker distribution and numbers of markers

The CN in the phantom varied from 17 to 59 in the sacrum and from 29 to 117 in the ilium, with varying numbers of markers and different marker distributions. When the markers placed in the symphysis were removed from the phantom, the CN increased from 29 to 92. The ME was reduced (p=0.001) when the frontal markers were removed. There was no difference in the accuracy, but the precision decreased from 0.11° to 0.22° in the Z rotation (p = 0.010) and from 0.08 mm to 0.18 mm in the X translation (p = 0.003). In vivo, the removal of the frontal markers did not affect the precision. The CN in the ilium increased from 38 to 96 (p=0.001), and the ME had a tendency (p = 0.048) to be lower in the ilium without the frontal markers. When the two cranial markers were removed, there was a reduction (p = 0.012) in the accuracy of the Y translation from -0.03 mm to -0.01 mm, and the precision was reduced from 0.11° to 0.22° in the Z rotation (p = 0.023), as well as in the X translation (p = 0.005), where it decreased from 0.08 mm to 0.24 mm. When three randomly selected markers were analyzed compared with eight markers, there were reductions in the precision of the Y and Z translations (p = 0.016 and p = 0.013, respectively), but there was no reduction in accuracy.

Paper II

Eleven patients (4 from Norway and 7 from Sweden) with long-term PGP were analyzed. Only small movements in the SIJ were detected, and only 15% of the measurements exceeded the precision of the RSA. Although some of the mean values were significantly different from zero, all but one of these mean values was less than the precision of the RSA.

The main findings in this study were as follows.

- When the patients performed a single-leg stance, there was almost no detectible movement.

- There was a mean of 0.5° of rotation on both sides around a helical axis (the true axis of rotation).

- When the movements were assessed based on the coordinate system (Figure 16), a small, 0.3° (SD 0.2) rotation around the Z-axis in the SIJ of the standing leg (p<0.001) was observed. This rotation was significantly different from that of the hanging-leg SIJ (p=0.036).

- No translations were detected.
• With the exception of the 0.2° difference observed in the Z-axis rotation (p=0.036), no differences were observed in the movement between the SIJs of the standing leg and the hanging leg (p-values between 0.055-0.978).

• There were no differences between the 18 symptomatic joints and the four asymptomatic joints with regard to the total amount of rotation (diff: -0.2, p=0.335 on the standing side; diff: 0.0, p=0.896 on the hanging side) or translation (diff: -0.1, p=0.398 on the standing side; diff: 0.1, p=0.687 on the hanging side).

**Paper III**

Nine consecutive patients received unilateral anterior SIJ fusion with concomitant fusion of the pubic symphysis. One patient developed chronic fatigue syndrome during the follow-up and dropped out of the study after 6 months. The remaining eight patients followed the study protocol; the baseline characteristics of these patients are presented in Table 4.

The ODI scores for each patient are presented in Figure 21. All but one patient exhibited a decrease of more than 10 points on the ODI from the pre-operative period to the 1-year follow-up. One patient experienced no effects. There was a strong association between ODI and time, with a 17-point decrease (p<0.001) at 1 year after surgery (Figure 21). The graphs showed significant variations in the measurements at each time point. At baseline, a difference of more than 40

<table>
<thead>
<tr>
<th>Table 4</th>
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<tbody>
<tr>
<td>Pre-operative patient characteristics</td>
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<tr>
<td>Mean</td>
</tr>
<tr>
<td>---</td>
</tr>
<tr>
<td>Age (years)</td>
</tr>
<tr>
<td>Sex: female/male</td>
</tr>
<tr>
<td>BMI (kg)</td>
</tr>
<tr>
<td>Children</td>
</tr>
<tr>
<td>Duration of symptoms (years)</td>
</tr>
<tr>
<td>Disability pension</td>
</tr>
<tr>
<td>• 100%</td>
</tr>
<tr>
<td>• Graded</td>
</tr>
<tr>
<td>Sick leave</td>
</tr>
<tr>
<td>• 100%</td>
</tr>
<tr>
<td>• Graded</td>
</tr>
<tr>
<td>Unilateral/bilateral SIJ symptoms</td>
</tr>
<tr>
<td>Pain in the pubic symphysis</td>
</tr>
<tr>
<td>Etiology</td>
</tr>
<tr>
<td>• Post-pregnancy</td>
</tr>
<tr>
<td>• Trauma</td>
</tr>
<tr>
<td>Positive clinical test</td>
</tr>
<tr>
<td>1. Posterior pelvic provocation test</td>
</tr>
<tr>
<td>2. Active straight leg raise</td>
</tr>
<tr>
<td>3. Modified Trendelenburg</td>
</tr>
<tr>
<td>4. Pain with palpation over the long dorsal ligament</td>
</tr>
<tr>
<td>5. Palpation of the symphysis</td>
</tr>
</tbody>
</table>

* One patient could not perform the test.
points between the maximum and minimum values was observed in two patients, and only two patients had less than a 10-point difference.

The VAS scores of each patient are presented in Figure 22. The patients experienced a reduction in pain, with a decrease from 82 points at baseline to 57 points after 1 year (p<0.001, regression coefficient of -8.4) (Figure 22). All of the patients reported a decrease in pain. Pre-operatively, a difference of 43 points between the maximum and minimum scores was observed in one patient, and none of the patients had variations of less than 10 points.

At baseline, seven of eight patients had bilateral SIJ symptoms. At the 1-year follow-up, only two patients experienced pain in the fused joint; however, six of the seven patients reported discomfort on the contralateral side. Seven patients had pain in the pubic symphysis before surgery, and five continued to have pain in this area at the 1-year follow-up.

On the SF-36, the patients experienced a mean 20-point improvement in physical function and bodily pain (p<0.001), a 15-point improvement in social functioning (p=0.008) and a 6-point improvement in general health (p=0.009).
All of the patients reported that the surgery had positive effects; one patient reported minor effects, two reported some effects, and five reported good effects of the surgery. None of the patients reported excellent results. With regard to tolerance of physical activity, seven patients reported some improvement, and one patient reported major improvement.

There were 3 major complications: one infection, one case of complex regional pain syndrome with drop-foot and one loss of bladder sensation. There were also 3 cases of transient sensitivity loss of the lateral femoral cutaneous nerve as a possible complication of bone harvesting from the iliac crest. All of the patients reported high post-operative pain levels, and they required epidural treatment for 5-7 days. They were hospitalized for 7-10 days and were discharged with prescribed opioids.

Figure 22
The mean VAS of each patient is presented, together with the regression line from the mixed model (VAS = 81.7 - 8.4 × time).
Paper IV

The demographics of the patients who did and did not undergo surgery are presented in Table 5. The non-surgical group was younger and had a shorter follow-up period than the surgical group.

<table>
<thead>
<tr>
<th>Table 5</th>
<th>Characteristics of the participants in the long-term follow-up study.</th>
</tr>
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<tbody>
<tr>
<td></td>
<td>Surgery group (n=50)</td>
</tr>
<tr>
<td>Age (years)</td>
<td>58 (56-61)</td>
</tr>
<tr>
<td>Follow-up (years)</td>
<td>23 (22-24)</td>
</tr>
<tr>
<td>Male/female</td>
<td>3/47</td>
</tr>
<tr>
<td>Etiology</td>
<td></td>
</tr>
<tr>
<td>Pregnancy</td>
<td>60%</td>
</tr>
<tr>
<td>Trauma</td>
<td>16%</td>
</tr>
<tr>
<td>Idiopathic</td>
<td>24%</td>
</tr>
<tr>
<td>Disability pension</td>
<td></td>
</tr>
<tr>
<td>0%</td>
<td>30%</td>
</tr>
<tr>
<td>50%-99%</td>
<td>16%</td>
</tr>
<tr>
<td>100%</td>
<td>54%</td>
</tr>
<tr>
<td>Pain medication</td>
<td></td>
</tr>
<tr>
<td>None</td>
<td>28%</td>
</tr>
<tr>
<td>Seldom</td>
<td>10%</td>
</tr>
<tr>
<td>1-6 days/week</td>
<td>16%</td>
</tr>
<tr>
<td>Daily</td>
<td>46%</td>
</tr>
</tbody>
</table>

The means are presented with a 95% confidence interval. Comparisons were performed with a two-sample t-test. p-values marked * were tested with a chi-squared test.

The patients who underwent surgery had a mean ODI of 33 (95% CI 24-42) and an evening VAS of 54 (95% CI 46-63) 23 years after SIJ fusion (Table 6). The subgroup of patients with a successful 1-year outcome had significantly lower scores on the ODI and the VAS than the patients with unsuccessful outcomes at 1 year. There was a 16 point difference in ODI score between these two groups, and this difference was regarded as both clinically and statistically significant (p=0.034). The difference in VAS between these two groups was 28 (p=0.011).

There were positive correlations between the 1-year outcome and three different long-term outcomes; VAS in the morning (ρ = 0.34, p = 0.0016), VAS in the evening (ρ = 0.42, p = 0.013) and ODI (ρ = 0.43, p = 0.002). There were no significant differences in ODI (p = 0.54), morning VAS score (p = 0.54), evening VAS score (p = 0.50) or SF-36 score between the group that underwent surgery and the non-surgery group at the long term-follow-up.
Table 6
Results of Oswestry disability index, visual analogue scale and SF-36

<table>
<thead>
<tr>
<th>Scores in the three subgroups with different short-term outcomes</th>
<th>Patients with SIJ fusion (n=50)</th>
<th>Successful (n=24)</th>
<th>Partly successful (n=12)</th>
<th>Unsuccessful (n=14)</th>
<th>Non-surgery group (n=28)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td>Adjusted means (95% CI)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Oswestry disability index (ODI)</td>
<td>33 (24-42)</td>
<td>27 (20-34)</td>
<td>37 (26-47)</td>
<td>43 (34-53)</td>
<td>37 (31-43)</td>
</tr>
<tr>
<td>Morning VAS score</td>
<td>44 (31-57)</td>
<td>38 (27-49)</td>
<td>41 (24-57)</td>
<td>62 (48-76)</td>
<td>50 (41-59)</td>
</tr>
<tr>
<td>Evening VAS score</td>
<td>54 (46-63)</td>
<td>42 (31-53)</td>
<td>60 (44-77)</td>
<td>71 (56-85)</td>
<td>60 (46-74)</td>
</tr>
<tr>
<td>SF-36</td>
<td></td>
<td></td>
<td></td>
<td></td>
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</tr>
<tr>
<td>Physical functioning</td>
<td>45 (36-54)</td>
<td>56 (45-68)</td>
<td>47 (30-63)</td>
<td>32 (17-48)</td>
<td>48 (34-62)</td>
</tr>
<tr>
<td>Physical role</td>
<td>25 (12-37)</td>
<td>27 (9-44)</td>
<td>31 (5-56)</td>
<td>18 (-6-41)</td>
<td>19 (1-39)</td>
</tr>
<tr>
<td>Bodily pain</td>
<td>39 (32-47)</td>
<td>46 (36-55)</td>
<td>42 (28-56)</td>
<td>28 (15-41)</td>
<td>39 (28-51)</td>
</tr>
<tr>
<td>Vitality</td>
<td>46 (40-53)</td>
<td>49 (41-57)</td>
<td>51 (40-62)</td>
<td>42 (31-52)</td>
<td>36 (26-45)</td>
</tr>
<tr>
<td>Social functioning</td>
<td>62 (54-71)</td>
<td>65 (56-75)</td>
<td>69 (56-83)</td>
<td>53 (41-66)</td>
<td>59 (47-72)</td>
</tr>
<tr>
<td>Emotional role</td>
<td>63 (49-76)</td>
<td>61 (44-79)</td>
<td>66 (41-92)</td>
<td>60 (37-83)</td>
<td>61 (49-76)</td>
</tr>
<tr>
<td>Mental health</td>
<td>73 (67-79)</td>
<td>72 (64-80)</td>
<td>76 (64-87)</td>
<td>76 (66-87)</td>
<td>71 (62-80)</td>
</tr>
</tbody>
</table>

Scores for ODI (0 indicates no disability, 100 indicates a high degree of disability), visual analogue scale (VAS; 0 indicates no pain, 100 indicates the worst possible pain) and SF-36 (0-100; 100 indicates the best health-related quality of life). The means are presented as least-square means, as they were adjusted for body mass index, age and time at follow-up, and are presented with a 95% confidence interval.
6.0 Discussion

6.1 Methodological considerations

6.1.1 Study designs and patient selection

The study design and the patient selection are important for drawing scientific conclusions from studies and, furthermore, to make the results generalizable. When these subjects are discussed, the terms internal and external validity are often used, and these terms address different quality aspects of the study. Internal validity is e.g. the extent to which the observed effect of an intervention can be explained by the treatment itself, rather than by chance or other systematic errors. Internal validity must be good to be able to draw conclusions that can be generalized (van der Worp et al. 2010). The study must be reproducible and reliable, and any form of bias should be minimized. External validity, in contrast, is the extent to which the results can be generalized.

Paper I

Paper I reported a methodological study in which we used a phantom model to attempt to explain what occurs in vivo. The external validity of a phantom model study can be questioned because the results from a phantom study cannot be transferred directly to the analysis of patients. Therefore, we analyzed precision in both the phantom and in patients. When the 3D position of each individual marker was calculated, the soft tissue was an important factor that could disturb x-rays, and the markers were likely not as stable in patients as in the phantom. These factors rendered the measurements in patients less accurate and less precise. Hence, patient precision should always be determined by double examination. We used a micrometer as the “gold standard” or true value. This micrometer had a resolution of 0.04° and 0.01 mm. The accuracy was less than 0.07 mm for translation and 0.05° for rotation; better resolution would be required to allow the conclusion that this deviation constituted actual bias (systematic error). The bias measured in our study was small and could be explained by the resolution of the micrometer. An interpretation of this bias might be that the RSA method had no bias that had to be corrected for when the measurements were obtained.
As expected, the precision was better in the phantom than in the patients. In the patients, the precision of the rotations had an LOS of less than 0.7° in all directions, and the precision of the translations was less than 0.5 mm. These values were comparable to the precision measurements reported by others (Tullberg et al. 1998, Sturesson et al. 2000b). The RSA method has been widely used in the study of hip prostheses. Using double examinations, precision has been reported to be between a low of 50 μm and 150 μm (Bragdon et al. 2002). When RSA is used in the SIJ, the segments are large, although the spread of the markers occurs mostly along a craniocaudal straight line in the ilia. However, the distance between the segments (sacrum and ilium) is larger than in the analysis of the implants. Abdominal and pelvic soft tissue can also decrease the quality of marker visualization on radiographs. Furthermore, absolutely zero motion between two examinations can hardly be expected. Because the patients were allowed to move between examinations, it is possible that the joints might have aligned differently between the two examinations. Hence, when precision is evaluated in patients, the joint alignment must be considered. All of these factors could explain why the precision was lower than the numbers reported in the analysis of the implants.

We evaluated the RSA in our lab with our set-up and equipment. One could argue that this evaluation weakened the external validity. However, the software that we used had the ability to compensate for the deviation automatically because it retrograde-calculated the positions of the tubes depending on the control markers in the calibration cage. As long as the same software is used, our results should be transferable to other studies as well.

*Paper II*

We believe the internal validity of measuring movement in the SIJ during the single-leg stance to be good. Both the method and the patients were well described, and the study could easily be replicated. We knew about the high accuracy and precision from paper I, and we did not find any bias for which we had to correct. We also included patients suffering from clinically verified severe PGP; hence, we believe these results to be sustainable.

External validity might have been influenced because only 11 patients were included. They were, however, recruited from two different clinics, and the results were uniform between the two clinics, strengthening the validity of our results.
The study design is an important factor in strengthening internal validity. In a prospective registration with repeated measures, such as an SSRD, some of the threats to internal validity can be controlled. Recommendations have been made to rate the quality of an SSRD, and a 14 question checklist was developed to obtain a quality rating (Logan et al. 2008). According to this quality rating, a study can be categorized as strong if more than 11 out of 14 questions can be answered in the affirmative. The 14 questions are divided into five different items: descriptions of the participants, independent variables, dependent variables, design and analysis.

First, the participants should be well described to enable replication of the study. There are no defined criteria to select patients for SIJ fusion. The diagnosis of PGP is mostly based on medical history and a combination of different clinical tests (Vleeming et al. 2008). Radiological examinations are primarily used to exclude serious pathologies that could explain the symptoms, and SIJ injections are administered to attempt to establish evidence for the SIJ as the origin of PGP. In our inclusion criteria (Table 2) we used the medical history of pelvic joint pain, a combination of 5 different clinical tests, a high level of disability measured by the ODI and a high level of pain measured by the VAS. In addition, the patients had to have undergone conservative treatment without positive effects. We did not include the SIJ injection among the inclusion criteria because there is uncertainty about the diagnostic value of SIJ injections, particularly when this procedure is performed infrequently (Simopoulos et al. 2012). We performed SIJ injections in most of the patients, and the results might have influenced the final decision regarding whether to perform surgery as the effect of the injection was not blinded for the research team. Similar inclusion criteria have been used in many of the studies published in recent years. Hence, we believe that our study can be reproduced, and furthermore, the results can be compared with those of other studies in which SIJ fusion is performed (Cummings and Capobianco 2013, Endres and Ludwig 2013, Rudolf 2012). Concerning the first item regarding description of participants, one point could be added to the total score.

The next two items on the checklist consisted of six questions about independent and dependent variables. The intervention was well described in our study and could be easily replicated. We also believe that the independent variables were well defined because a
thorough presentation of the patients was undertaken. The dependent variables are well known and described. The ODI, VAS and SF-36 have been tested for test-retest properties, and the questionnaires have been evaluated for use in the Norwegian population (Grotle et al. 2003, Loge et al. 1998). To score six of six points on these questions, the assessors had to be unaware of the phases of the study, and in our study, we had no opportunity to comply with this requirement. Both the patients and the examiners were fully aware of the phase in which they were. The last quality criterion in item covering the dependent variables was the stability of the measurements. Our patients did not show stability of measurements during any phase. Patients with PGP have reported cyclic variations in symptoms, and as many as 72% report relapses during menstruation (Mens et al. 1996). To account for these potential variations, we repeatedly collected the patients’ data for a 5-week period and discovered a large variation in the values during each phase. For some patients at baseline, a 40-point difference in the ODI and a 43-point difference in the VAS were observed over the 5-week period. Although the stability of the measurements is important for internal validity, one strength of an SSRD is its ability to detect these individual variations, which is important for studying patients with PGP. Some of these variations could be corrected for in large group studies, but conclusions from small case series, with single measurements, should be interpreted with caution because these threats to the internal validity cannot be controlled. Another finding that strengthened the validity of this study was that we measured both morning VAS and evening VAS. Patients with PGP have reported experiencing more pain in the evening than in the morning (Stuge et al. 2004b), and we found a difference of more than 20 points in the VAS between the morning and the evening. Furthermore, the evening pain showed a greater decrease between baseline pre-operatively and at 1 year of follow-up. Hence, when the VAS score is used, this variation must be taken into consideration. According to the SSRD quality checklist, our study scored 4 of 6 points in the questions regarding independent and dependent variables.

The next item covers the description of the design. The study was conducted according to guidelines for SSRDs (Engel RJ and Schutt RK 2009, Grotle et al. 2003, Zhan and Ottenbacher 2001), and an adequate number of participants (more than 3) was investigated. To score the maximum on this item, the effect should be observed in three or more subjects, and in our study, 7 of 8 experienced positive effects of the intervention. Because all of the questions on this item were answered affirmatively, 3 more points could be added for a total score of maximum 14.
The last item covers the analysis of the data. We presented the data according to the guidelines, and because we had 8 patients, we were also able to conduct a statistical analysis of the whole group. All 4 of the criteria concerning analysis of the data were met, and the total of the SSRD rating was 12 of 14, which is regarded as strong quality by the authors of the rating system (Logan et al. 2008).

One other threat to the internal validity, which is not addressed in the rating, is that the observed results were normal variations in symptoms, and there was another event between baseline and follow-up that could explain this effect. The patients had a chronic condition with mean 11-year duration, so we believe the baseline phase was representative of the pre-operative status. To correct for events in between that could explain the measured effect, we obtained measurements in 3 post-operative phases.

With an SSRD, the findings are generalizable if the study is replicated among 3 or more subjects (Logan et al. 2008). The patients included in papers I, II and III had severe PGP, and they had symptoms from the pelvic girdle for an average of 11 years. They had a mean baseline ODI value of 54, a mean morning VAS of 60 and a mean evening VAS of 82, and only patients with an ODI greater than 40 and/or a VAS greater than 50 were included. Hence, this was a selected group of patients with PGP. In comparison, patients with PGP in the weeks after pregnancy were reported to have an ODI of 42 and an evening VAS of 58 (Stuge et al. 2004a). The patients reported by Stuge et al. (2004a) did not reach a chronic phase and responded well to conservative treatment. Our patient selection was undertaken by applying strict inclusion criteria, and lumbar spine pathology was excluded by lumbar MRI. Hence, we believe that these patients were representative of patients with severe and long-lasting chronic PGP. The results, however, should be interpreted with the knowledge that these patients were a selected group of patients.

A randomized controlled trial is the gold standard for examining the effects of an intervention, and an understanding has developed that only group studies can produce scientific data (Gonnella 1989). Because SIJ fusion has been performed on few patients, a single-center randomized controlled design was difficult to establish due to the small number of participants. However, a single-subject research design (SSRD) has been recommended as a useful method for examining clinical accountability (Zhan and Ottenbacher 2001).
SSRD is replicated across patients, the internal and external validity is strengthened and allows inferences to be made about effectiveness. Pelvic fusion in patients with PGP is a rare procedure and is only performed in severe cases in which conservative treatment modalities have been unsuccessful. A randomized controlled trial of this procedure was difficult to perform because the alternative treatment modality (conservative treatment) had already been attempted by most of the patients. Hence, compliance in such a conservative group could have been a great challenge. Because an SSRD with multiple measurements is designed to study small samples of patients, this design was chosen. A limitation of our study was the short follow-up period of 1 year, which is regarded to be too short for a clinical trial, but in paper IV, we reported that the 1-year outcomes after SIJ fusion were sustained 23 years later. Hence, a 1-year follow-up might be sufficient. Despite the limitations of the SSRD, we believe that our study contributes valuable information regarding the effects of pelvic joint fusion.

**Paper IV**

The design of the study in paper IV was cross-sectional and included 50 patients with SIJ pain who underwent SIJ fusion, combined with a long-term follow up. The 1-year outcomes were compared to outcomes recorded in 2009 and to a comparison group. To compare the 1-year results, a sub-group classification was created (Figure 11), and we were aware of the limitations of this subgroup classification. The sub-grouping might not have been optimal, although the post-operative joint pain was classified according to specific criteria. The primary aim of the study was to examine the long-term outcomes of SIJ fusion, not to evaluate the effects of the surgery compared to a control group. There was great selection bias in the two groups. First, they differed in age and length of follow-up, and second, they most likely did not have the same baseline status in pain and function. The comparison must be interpreted with these limitations in mind.

We found a correlation between the 1-year results and the long-term results. With the same inclusion criteria and the same surgical technique, these results could be generalized to this population. With a long-term follow-up, there are several opportunities for bias. We only examined 50 patients, and because the follow-up was as long as 23 years, some of these patients could have experienced other events that would explain the findings.
6.1.2 Outcome measures

Because PGP is a multi-factorial condition, a questionnaire should address different aspects of the patient's experience. To detect changes in pain, disability, and health-related quality of life, we used three different questionnaires: the ODI, a VAS and the SF-36. There are several generic instruments that measure pain perception, functional disability and health-related quality of life, but when we started this project, no condition-specific questionnaire was available (Stuge et al. 2011). Because PGP is a subtype of low back pain with a unique clinical presentation (O'Sullivan and Beales 2007), a condition-specific questionnaire, the Pelvic Girdle Questionnaire, was later developed (Stuge et al. 2011). The closest to a condition-specific questionnaire that was available were the ODI, the Quebec Back Pain Disability Scale and the Roland Morris scales, and these instruments have frequently been used in research on PGP (Vleeming et al. 2008). We used the ODI because most studies have used this questionnaire to investigate interventions in PGP, and it has been shown that the ODI is an appropriate instrument to measure changes in functional status in patients with chronic LBP (Grotle et al. 2004).

We decided to use a VAS instead of a numeric rating scale because most of the studies used this outcome measure after SIJ fusion (Keating et al. 1997, Schutz and Grob 2006, Ziran et al. 2007). A numeric rating scale would most likely have been better than a VAS to detect changes in chronic patients (Grotle et al. 2004), because the responsiveness has proven to better for NRS compared to VAS when these scales are used on chronic patients. In addition to the VAS, a pain diagram was used to localize local pain and its referrals; for this use, the pain diagram has shown reliable results (Ohlund et al. 1996).

As a generic instrument, we used the SF-36, which has also been used in the evaluation of patients undergoing SIJ fusion (Buchowski et al. 2005). The Norwegian versions of the questionnaires have been evaluated, and they have sufficient properties with regard to validity, reliability and responsiveness (Loge et al. 1998). With the selection of these outcome measures, we first hoped to detect possible changes in pain, physical function and health-related quality of life after SIJ fusion, and second, we were able to compare our results to the available literature.
6.2 The SIJ

6.2.1 Movement in the SIJ

The literature has disagreed regarding the amount of movement that occurs in the SIJ, with reports ranging from movement that can be detected by an examiner (Hungerford et al. 2007) to RSA studies demonstrating almost no movement (Sturesson et al. 1989, Sturesson et al. 1997, Sturesson et al. 1999, Sturesson et al. 2000b, Sturesson et al. 2000a, Tullberg et al. 1998). The first aim of thesis was to determine whether the RSA was reliable (paper I). In the phantom model, we measured the accuracy and did not find any bias that had to be corrected for when this examination was used. In the patients, the RSA had a precision for translation of less than 0.5 mm in all directions, and for rotation, the precision was less than 0.3° around the Y and Z axes and 0.7° around the X axis (Figure 16). We also discovered that the markers only needed to be placed in the dorsal part of the pelvis and that 3-4 markers were sufficient if the marker distribution was good (appropriate three-dimensional distances between the markers). We found the RSA technique to be highly accurate and precise. We therefore concluded that the RSA results were reliable and valid because these reports used the same set-up. Nevertheless, some questions regarding the SIJ movements should be addressed.

*What is the movement in the SIJ?*

There has been evidence that, during and after pregnancy, movements are increased in the pelvic joints and that patients with pain have greater movement than asymptomatic controls (Mens et al. 2009). However, the importance of SIJ movement when patients develop chronic PGP is more uncertain. Some RSA studies have been performed, and the overall findings were that the movement in the SIJ is small, and it seems to be normally distributed (Sturesson et al. 1989). When changing from a supine to a standing position, a total of 1.2° of forward rotation of the sacrum has been reported relative to the innominates, and the movement is 1.6° when the patients change from a supine to a sitting position (Sturesson et al. 1989). In the standing position, the SIJ seems more or less locked. Using RSA, Sturesson et al. (2000b) found a total of 1.2° of rotation in each SIJ when alternating among the straddle position, standing with the left hip maximally extended and right hip maximally flexed and standing with the right hip maximally extended and the left hip maximally flexed. When these authors analyzed the movement during the standing hip flexion test (movement between standing on both feet and standing on one foot with the other hip maximally flexed), a small total
movement of 0.6° was reported (Sturesson et al. 2000a). This finding contradicted those of Hungerford et al. (2007), who stated that the SIJ movement could be felt by hand during the Stork test. In addition, Sturesson et al. (1989) did not find any differences in movement between symptomatic and asymptomatic SIJs. Hence, there is movement in the SIJ, but there does not seem to be hyper-mobility of the SIJ in patients with chronic PGP.

If the SIJ has a limited range of motion, why should we even attempt to measure the SIJ movement in the diagnosis of PGP?
The Chamberlain x-ray projection has been used to attempt to diagnose SIJ movement, and in paper II, we wanted to determine whether there was movement in the SIJ during the single-leg stance test. We only found minor movement, with total rotation of 0.5° in both the standing leg SIJ and the hanging leg SIJ. Researchers have used this method to establish a relationship between movement detected in the pubic symphysis and pain in the SIJ (Anderson and Peterson 1944, Chamberlain 1930, Mens et al. 1999, Siegel et al. 2008), but the findings have not been uniform. Hence, based on the results of paper II, the Chamberlain examination was likely inadequate for examining SIJ movement in patients with PGP. Furthermore, there did not seem to be any difference between symptomatic and asymptomatic SIJs. Based on the range of motion being limited and the measuring methods lacking proper precision, it does not seem valid to measure SIJ movement in clinical practice.

If the range of movement in the SIJ is limited, why would SIJ fusion help these patients?
As described in the European guidelines, the stability of the SIJ is more a matter of altered laxity or stiffness than an increased range of motion (Vleeming et al. 2008). Using Doppler imaging and small vibrations made by a vibrating device, authors have been able to measure laxity (Damen et al. 2002a, Damen et al. 2002c). During pregnancy, increased laxity of the SIJ has been reported, but the values returned to pre-pregnancy status within 8 weeks (Damen et al. 2002a). In this study, the authors found that asymmetrical laxity during pregnancy was associated with increased pain post-partum, but the magnitude of the laxity was not important. In another paper, the same authors demonstrated that a pelvic belt reduced laxity (Damen et al. 2002b), and a pelvic belt was also shown to increase the functional abilities of patients with PGP because the ASLR improved with a pelvic belt (Mens et al. 1999). Sturesson et al. (1999) demonstrated that the movement was reduced by 50% when a pelvic frame was applied to the pelvis. The use of an external frame has been reported to reduce pain in patients
with PGP (Slatis and Eskola 1989). Hence, pelvic mobility could be more a matter of laxity within a small range of motion, and because a stabilizer such as a pelvic belt or frame can reduce symptoms, SIJ fusion might be an appropriate treatment option in some severe cases.

6.2.2 The SIJ as a pain generator

The SIJ has neural structures in the joint and in the surrounding ligaments, and these structures can be stressed, with pain as a consequence. However, there have been many theories and models created to explain the underlying causes of PGP and SIJ pain. It has been emphasized that the disorder of PGP must be viewed in a bio-psycho-social framework because many factors are important when treating a patient with a long-term pain syndrome (O'Sullivan and Beales 2007).

In our studies, we primarily examined the bio-mechanical properties of the SIJ. Sacroiliac fusion immobilized the SIJ, and the effects could be observed as a direct consequence of increased stabilization of the pelvic joints. With this approach to the problem, the premise was that the SIJ was the major pain generator. According to the results of SIJ injections in patients with PGP, as well as in patients with non-specific LBP, this premise could be true (Simopoulos et al. 2012). With a single injection and with a cut-off of between 50% and 80% pain relief, an average prevalence of 30-35% was observed, and a higher cut-off (>80%) yielded a slightly lower prevalence (Simopoulos et al. 2012). With a dual block (confirmatory blocks), the prevalence was estimated to be 25%, but with a wide range of 10-40%. When SIJ injections have been used as a "gold standard", the prevalence has varied because the "gold standard" is not standardized. Furthermore, the patients have often been selected differently, resulting in a prevalence with a broad range. Schwarzer et al. (Schwarzer et al. 1995) studied 100 patients with low back pain, and after screening (pain below L5-S1 and over the SIJ), 43 patients were assigned to SIJ injections due to suspected SIJ pain. Thirteen of 43 experienced positive effects from the injections (more than 75% relief), representing a 30% prevalence in patients with suspected SIJ pain, but in the entire sample of LBP patients, the prevalence was 13%. Hence, in a selected population, the prevalence became greater. Using injections only, Laplante et al. (Laplante et al. 2012) found a 17.8% prevalence of SIJ pain in 153 cases of patients with non-specific LBP, and interestingly, the prevalence in a population of 54 patients with suspected SIJ pain was 18.9% (Maigne et al. 1996). The highest prevalence
reported was 62% in a highly selected group of patients (Slipman et al. 1998). These patients had 3 positive SIJ provocation tests, and they were assigned to a rehabilitation program. Those who failed this program received diagnostic SIJ injections. Thirty-one of 50 had positive responses to the injections, yielding a prevalence in this population of 62%. Based on these investigations, it seems that the SIJ could be a source of pain in a large proportion of patients with suspected PGP, as well as in patients with non-specific LBP.

Some researchers have proposed that the pain is caused by subluxation or displacement of the SIJ, and the treatment has thus primarily been based on manipulation to correct this (O'Sullivan and Beales 2007). Positive short-term effects of this treatment have been reported (Wright 1995), but the effects did not seem to last. Although these manipulations have effects on the symptoms, the position of the SIJ does not seem to be altered during these treatments (Tullberg et al. 1998). Hence, subluxation of the joint as a reason for PGP seemed not to be correct.

Disturbances in the motor control of the lumbo-pelvic musculature has been emphasized as a possible cause of PGP because this impairment alters the ability to transfer load through the pelvis and triggers nociceptive structures (O'Sullivan and Beales 2007). A treatment program focusing on training in motor control has been shown to have statistically and clinically significant and long-lasting effects on PGP post-partum (Stuge et al. 2004b, Stuge et al. 2004a), but some patients did not respond to this treatment. Different patterns of motor control impairments could explain these findings, and PGP has been associated with both excessive and insufficient motor activation of the pelvic musculature (O'Sullivan and Beales 2007). It has been suggested that over-activity of the pelvic floor muscles might reduce force closure by counter-nutation in the SIJ, and it could be a possible mechanism for maintaining pain and disability in patients with PGP (Pool-Goudzwaard et al. 2004). Stuge et al. (2012, 2013) found that women with PGP had a statistically significantly smaller levator hiatus, even at rest, and a tendency for higher vaginal resting pressure might indicate increased activity of the pelvic floor muscles. The reason for this is unknown, but could be a response to SIJ pain. Under pelvic floor muscle contraction, the coccyx moves in a ventral and cranial direction (Bo et al. 2001), and it most likely generates counter-nutation in the SIJ (Pool-Goudzwaard et al. 2004). Because counter-nutation might lead to increased tension in the ligamentous structures (Snijders et al. 1993), sustained contraction of the pelvic floor muscles might be a
non-optimal strategy, leading to stress on the ligaments and resulting in pain. Palsson and Graven-Nielsen (2012) recently showed that superficial ligament structures were potential pain sources in PGP.

Another group receiving increasing interest is patients with failed back surgery. In patients with persistent pain after spinal fusion, Depalma et al. (DePalma et al. 2011) used SIJ injections and found the SIJ was the source of pain in 43%, and 83% of these patients had fusion to the sacrum. These findings were similar to those of another study that found 35% positive blocks in patients with pain after spinal fusion (Maigne and Planchon 2005). Increased SIJ degeneration was found, compared to un-operated subjects, 5 years after spinal fusion, particularly in cases with lumbosacral fusion (Ha et al. 2008). Patients with a suspected SIJ origin of pain after spinal fusion also reported a different type of pain than that for which they primarily underwent surgery (Maigne and Planchon 2005). The importance of SIJ degeneration in patients with suspected SIJ pain is not fully understood because degenerative changes have been observed in both asymptomatic and symptomatic patients (Elgafy et al. 2001). However, with the use of finite element analysis, increased stress in the SIJ has been reported, particularly if there was a sacro-lumbar fusion (Ivanov et al. 2009), and increased tension on the SIJ structures can possibly cause pain. Based on these studies, the SIJ could be a pain generator in patients with failed back surgery; furthermore, these patients have reported responding positively to SIJ fusion (Rudolf 2013).

6.3 Surgery as a treatment for PGP

In paper III, we found a mean 17-point decrease in the ODI (from 54 to 37) and a mean 25-point decrease in the VAS (from 82 to 57) one year after SIJ fusion was performed. Pre-operatively, these patients had a high degree of disability and high levels of pain. One patient did not experience any effects from the intervention, but the remainder had significant decreases in ODI and VAS scores. The minimal clinically significant difference has been suggested to be 10 points in the ODI (Hagg et al. 2003), and in our study, 7 of 8 patients achieved this difference. For the VAS, the minimal clinically significant difference has been suggested to be 18 (Hagg et al. 2003). Six of 8 patients had a change of 18 or greater on the VAS: one had a 16-point change, and one had a small change of only 3. As a group, the
patients experienced positive effects from surgery, and the changes seemed to be of statistical and clinical significance.

In the long-term follow-up (reported in paper IV), the patients had a mean ODI score of 33 (95% CI: 24-42) and a mean VAS score of 54 (95% CI: 46-63), comparable to the 1-year results reported in paper III. Compared to a comparable non-surgery group, the operated patients in the long-term follow-up had lower ODI (33 compared to 37) and VAS scores (54 compared to 60), but these differences were not statistically significant. There are possible explanations for this lack of difference. The most tempting interpretation to make of this finding is that SIJ fusion had no effects, but the study was not designed to draw such a conclusion. The non-surgery group differed from the surgery group with regard to age and length of follow-up. The non-surgery group was diagnosed with severe PGP by the same surgeon, but the patients in that group were likely less disabled at baseline than the group of patients who underwent surgery; hence, the comparison might have had limited validity. What was discovered, however, was that the patients with successful short-term results had significantly better outcomes than the patients with poor short-term results, and the short-term results were correlated with both the ODI and VAS over the long-term follow-up. The studies in both paper III and paper IV had specific selections of patients, and with the use of the inclusion criteria described in paper III, it seems that positive effects from surgery could be expected.

In our study (paper III), there was clinical improvement, but the patients continued to have moderate disability and relatively high pain scores after 1 year, and one patient did not experience any effects at all. One explanation for this finding is the limitation of the use of outcomes. Most of the patients had bilateral SIJ pain at inclusion (7 of 8), but only the most painful joint and the symphysis were fused. Because the outcome measures tested the overall pain and functioning of the patients, the non-fused joints influenced the measured values of the ODI, VAS and SF-36. In paper III, 6 of 8 patients were pain-free in the fused joint, but six of seven reported pain on the contralateral side, which could indicate that the function and pain could have been further improved if the patients had undergone fusion of the other joint as well.
How do our results compare to those of other studies?

When the literature was explored, only 28 original papers and 2 book chapters were found (Appendix 1), and most of the older papers did not include any validated outcome measures. There were only case series without control groups. The patient selection and surgical techniques also differed in almost every paper, and no studies reported the outcomes of our surgical method. Hence, a comparison of our results to others is difficult, but there have been several studies that have reported the outcomes of SIJ fusion using similar inclusion criteria. A positive effect of surgery has been observed in 50% to 90% of patients (Appendix 1), and this finding is in accord with the positive effects observed in our 1-year outcomes. In a prospective study of 58 patients, van Zwienen et al. (van Zwienen et al. 2004) reported a mean increase in physical outcomes from 37 to 61 (p < 0.001) as measured using the Majeed score (0 - poor, 100 - good), and the same positive results have been reported in several papers (Appendix 1, Table 7).

Although positive results have been reported, a number of patients have not experienced effects from surgery. In our long-term study, 28% of patients did not experience any effects from surgery, and these findings were comparable to those of other studies (Buchowski et al. 2005, Duhon et al. 2013, Rudolf 2012, Smith et al. 2013, van Zwienen et al. 2004, Wise and Dall 2008). Most of these patients had complications or non-union events, but some experienced no effects without any clear explanation. Open surgery causes extensive surgical trauma, and some of these patients may have experienced pain due to soft tissue trauma caused by surgical exposure. It has been shown that 10-15% of patients develop chronic pain after simple iliac crest bone harvesting (Delawi et al. 2007, Laurie et al. 1984), which is a similar, but much smaller, procedure to open SIJ fusion. In our study, one patient did not experience any effects from the surgery. This patient had a more generalized pain pattern than the others and it is possible that the SIJ was not the major source of pain in this case, although this patient had a positive response to the SIJ injection. In contrast, the patients who reported sharp and localized pain in the SIJ area did benefit from surgery. It has been reported that the outcomes improved if patients with psychosomatic disorders were excluded, with the success rate increasing from 50% to 70% (Waisbrod et al. 1987). Because SIJ fusion has effects in many patients with PGP, the SIJ or the surrounding ligaments could be the origin of pain in many of these patients, but in patients with more generalized pain patterns, other pain generators might influence the outcome of surgery. The reason for the effect of SIJ fusion is,
however, not known. In theory, SIJ fusion can correct many biological disturbances, but it also has a psychological influence on the patient, including placebo effects.

The placebo effect after surgery has been shown to be an important factor in short-term efficacy (Turner et al. 1994) and particularly in the treatment of chronic pain, in which the psychological component is believed to be an important factor. For example, in a study using sham surgery as much as 43% of the patients in the placebo group had pain relief after sham lumbar discectomies (Spangfort 1972), and other similar sham procedures have also reported to have a significant effect on clinical outcome (Vandana and Tushar 2001). There are several factors in both the patients and examiners that contribute to the placebo effect. Patients with long-lasting diseases seems to be poorer placebo responders, but on the other hand placebo tend to work better in patients expecting to have changes in sensation of pain (Vandana and Tushar 2001). Our patients was included according to specific criteria and might feel a special need to become pain free, and this can enhance the placebo effect. They also got a relationship with the examiners because we used multiple testing. Hence, some of the positive effect of SIJ fusion seen in paper III can of course be placebo. In our long-term study (paper IV), almost three-quarters of the patients reported to be relieved, or almost relieved, from pain after 1 year, and 28% were not. The 1-year results for the operated patients seemed to be sustained throughout life because patients with successful 1-year results had both significantly less pain and less disability than the patients with unsuccessful 1-year results. With these results, it is difficult to believe that the placebo effect is the only contributor to successful surgical outcomes.

6.3.1 Future of SIJ fusions
It has been questioned whether MIS implants can improve the outcomes of SIJ fusion. Over the last decade, minimally invasive procedures have been introduced, and the use of implants has become popular. Between 2009 and 2013, more than 5000 patients were treated with a specific implant (Miller et al. 2013). In the last few years, several manufacturers have created MIS implants for use in SIJ fusion, but studies have been published for only three of these implants. In 2008 and 2009, two papers were published regarding the use of a traditional, hollow, cylindrical screw across the SIJ (Al-Khayer et al. 2008, Khurana et al. 2009). To achieve fusion, the study authors filled the screw with BMP-2 or demineralized bone matrix. They reported the outcomes of 24 patients with suspected SIJ pain; the fusion rate was 100%,
and the clinical outcomes were promising (Table 7). Later, Mason et al. (Mason et al. 2013) reported the outcomes of 55 patients using the same technique and reported no implant failures, two cases of nerve damage and no other complications. Although the complication rate was lower, the clinical outcomes were not better than those of our study (paper III) in which open anterior surgery was performed (Table 7).

Table 7
Outcome of studies using ODI or/and VAS/NRS as outcome measures.

<table>
<thead>
<tr>
<th>Study</th>
<th>VAS pre op</th>
<th>VAS post op</th>
<th>Δ</th>
<th>Change in %</th>
<th>ODI pre op</th>
<th>ODI post op</th>
<th>Δ</th>
<th>Change in %</th>
</tr>
</thead>
<tbody>
<tr>
<td>Al-Khayer 2008*</td>
<td>8.1</td>
<td>4.6</td>
<td>3.5</td>
<td>43%</td>
<td>59</td>
<td>45</td>
<td>14</td>
<td>24%</td>
</tr>
<tr>
<td>Mason 2013*</td>
<td>8</td>
<td>4.5</td>
<td>3.5</td>
<td>44%</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sachs 2012§</td>
<td>7.9</td>
<td>2.3</td>
<td>5.6</td>
<td>71%</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Rudolf 2012§</td>
<td>7.6</td>
<td>3.3</td>
<td>4.3</td>
<td>57%</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sachs 2013§</td>
<td>8.7</td>
<td>0.9</td>
<td>7.8</td>
<td>90%</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Cunnings 2013§</td>
<td>8.9</td>
<td>2.3</td>
<td>6.6</td>
<td>74%</td>
<td>53</td>
<td>13</td>
<td>40</td>
<td>75%</td>
</tr>
<tr>
<td>Duhun 2013§</td>
<td>7.6</td>
<td>2.9</td>
<td>4.7</td>
<td>62%</td>
<td>55</td>
<td>39</td>
<td>16</td>
<td>29%</td>
</tr>
<tr>
<td>Endres 2013†</td>
<td>8.5</td>
<td>6</td>
<td>2.5</td>
<td>29%</td>
<td>64</td>
<td>57</td>
<td>7</td>
<td>11%</td>
</tr>
<tr>
<td>Kibsgård 2014</td>
<td>8.2</td>
<td>5.7</td>
<td>2.5</td>
<td>30%</td>
<td>54</td>
<td>37</td>
<td>17</td>
<td>31%</td>
</tr>
<tr>
<td>Mean</td>
<td>8.2</td>
<td>3.6</td>
<td>4.6</td>
<td>56%</td>
<td>57</td>
<td>38</td>
<td>19</td>
<td>34%</td>
</tr>
</tbody>
</table>

All studies used MIS to achieve SIJ fusion. Marks according to the implants used in the study: *=hollow cylindrical screw, §=iFuse, †=DIANA

Recently, two other MIS techniques have been developed: the iFuse implant (SI-bone Inc., San Jose, CA, USA) and the Distraction Interference Arthrodesis Neurovascular Anticipating (DIANA) implant (Signus Medizintechnik, Alzenau, Germany). Outcomes with the iFuse implant have been reported in 5 studies (Table 7), and the complication rate has been reported to be between 0% and 20%. The implant is a triangular, coated implant that is applied across the SIJ, and the SIJ is indirectly fused as the bone grows into the implant. The implant seems promising with regard to clinical outcomes and complication rates, and it has been reported to be cost-effective in the treatment of SIJ pain (Ackerman et al. 2013). In a retrospective, multi-center study of 263 patients, the implant showed better clinical outcomes than open surgery (3.5 points lower on a VAS at 12 months in the MIS group), fewer complications and a shorter hospitalization time (Smith et al. 2013). Unfortunately, the authors of these studies have close connections to the corporation that produces these products (SI-bone) as consultants, stockholders or employees of the company, and the company has sponsored some
of the studies. Hence, these results should be interpreted with caution and replicated by independent researchers. The DIANA is another type of titanium implant that is inserted dorsally into the recess. Only one original paper with 19 patients has been published, with a fair fusion rate and no infections or no nerve damage. The clinical outcomes, in contrast, were worse than our results (Table 7), with a decrease in the ODI from 64 to 57 and a reduction in the VAS from 85 to 60.

In conclusion, the new MIS implants seem safer than open access surgery, with low reported complication rates. The surgical trauma is far less when using MIS, and consequently, the length of the hospital stay is shorter. The clinical outcomes have been reported to be as good, or even better, than the old open technique. However, more studies have to be performed; the studies must be of higher quality; and importantly, the studies must be performed independently of industry. Additionally, future studies should focus on optimal inclusion criteria for SIJ fusion.
7.0 Conclusions

This thesis showed the following.

- Radiostereometric analysis applied to the SIJ has high accuracy and precision and is suitable as a tool for investigating SIJ movement. In patients, more than 4 markers in each segment are recommended to ensure obtaining a proper segment to use in the analysis. The use of frontal markers does not improve the precision and is therefore unnecessary.

- In patients with PGP, the movements in the SIJ in the single-leg stance are small and almost undetectable by the precise RSA method. We measured a mean rotation of 0.5° in both the standing- and hanging-leg SIJs, and no translation was detected. There were no differences in total movement between the standing- and hanging-leg SIJs. The interpretation of the results of this study is that the Chamberlain examination is likely inadequate for examining SIJ movement in patients with PGP.

- One year after open unilateral anterior SIJ fusion combined with symphysis pubis fusion, positive and significant changes were observed in both physical function and pain. Despite these positive results, this procedure was associated with adverse events and complications.

- Patients with chronic PGP who underwent SIJ fusion reported being moderately disabled, with moderate or severe pain intensity, 23 years after surgery. Approximately half of these patients had successful 1-year outcomes, and in these patients, their good results were sustained 23 years after surgery. Two-thirds of the patients experienced positive long-term effects from fusion surgery, and 20% reported no effects from the surgery.


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Appendix I

Tables of former studies reporting surgical outcomes after sacroiliac joint fusion
<table>
<thead>
<tr>
<th>Author/year</th>
<th>N</th>
<th>Surgical method</th>
<th>Fusion rate</th>
<th>Functional outcome</th>
<th>Result</th>
</tr>
</thead>
<tbody>
<tr>
<td>Smith-Petersen 1921</td>
<td>13</td>
<td>Retrospective</td>
<td>Trans-iliac-window (Smith-Petersen)</td>
<td>NA</td>
<td>Authors opinion</td>
</tr>
<tr>
<td>Smith-Petersen 1926</td>
<td>26</td>
<td>Retrospective</td>
<td>Smith-Petersen</td>
<td>95% on x-ray</td>
<td>Recovery - absolute relief</td>
</tr>
<tr>
<td>Gaenslen 1927</td>
<td>9</td>
<td>Retrospective</td>
<td>Smith-Petersen</td>
<td>NA</td>
<td>Very good</td>
</tr>
<tr>
<td>Mittner 1931</td>
<td>12</td>
<td>Retrospective</td>
<td>Smith-Petersen</td>
<td>NA</td>
<td>Good Fair Poor</td>
</tr>
<tr>
<td>Mitchell 1938</td>
<td>15</td>
<td>Retrospective</td>
<td>Smith-Petersen Campbell</td>
<td>NA</td>
<td>Classified by surgeon</td>
</tr>
<tr>
<td>Avilia 1941</td>
<td>4</td>
<td>Retrospective</td>
<td>3 Antero-lateral 1 Smith-Petersen</td>
<td>NA</td>
<td></td>
</tr>
<tr>
<td>Hagen 1974</td>
<td>8</td>
<td>Retrospective</td>
<td>Smith-Petersen 2 Pubic symphysis only</td>
<td>6/8</td>
<td>Good Fair Poor</td>
</tr>
<tr>
<td>Olerud 1984</td>
<td>8</td>
<td>Retrospective</td>
<td>Fusion PS 5 SIJ fusion NA</td>
<td>100% SIJ NA</td>
<td>Residual pain</td>
</tr>
<tr>
<td>Rand 1985</td>
<td>1</td>
<td>Retrospective</td>
<td>Anterior fusion</td>
<td>NA</td>
<td></td>
</tr>
<tr>
<td>Waisbrod 1987</td>
<td>22</td>
<td>Retrospective</td>
<td>Dorsal resection, bone graft</td>
<td>20 (90%)</td>
<td>Unsatisfactory (US) 50% pain relief</td>
</tr>
<tr>
<td>Lippit 1995</td>
<td>15</td>
<td>Retrospective</td>
<td>SI screws</td>
<td>No fusion 1 loosening</td>
<td>Classified by surgeon</td>
</tr>
<tr>
<td>Keating 1997</td>
<td>39</td>
<td>Prospective</td>
<td>Debridement and screw</td>
<td></td>
<td>Pain level VAS</td>
</tr>
<tr>
<td>Güner 1998</td>
<td>1</td>
<td>Retrospective</td>
<td>Endoscopic anterior fusion</td>
<td>100%</td>
<td></td>
</tr>
<tr>
<td>Belanger 2001</td>
<td>4</td>
<td>Retrospective</td>
<td>Dorsal resection + pedicle screw</td>
<td>100%</td>
<td>Self-reported satisfaction</td>
</tr>
<tr>
<td>Giannikas 2004</td>
<td>5</td>
<td>Prospective</td>
<td>Sacroiliac bone plugs</td>
<td>100%</td>
<td>Complete relief</td>
</tr>
<tr>
<td>Van Zwienen 2004</td>
<td>58</td>
<td>Prospective</td>
<td>Plating PS Bilateral SI screws PS 49/58 (84%) SIJ no fusion was performed</td>
<td></td>
<td>Majeed</td>
</tr>
<tr>
<td>Buchowski 2005</td>
<td>20</td>
<td>Prospective</td>
<td>Modified Smith-Petersen Plate-screws</td>
<td>17/20 (85%) SF-36</td>
<td>Physical sum Mental sum</td>
</tr>
<tr>
<td>Author</td>
<td>Year</td>
<td>Study Type</td>
<td>Procedure</td>
<td>Results</td>
<td></td>
</tr>
<tr>
<td>-------------------------</td>
<td>-----------</td>
<td>------------</td>
<td>---------------------------------------------------------------------------</td>
<td>---------------------------------------------------------------------------------------------------</td>
<td></td>
</tr>
<tr>
<td>Scütz 2006</td>
<td>Retrospective</td>
<td>Bilateral SIJ fusion with SI screws and ilium to ilium connecting bolt</td>
<td>Questionable fusion 4; Non-union 7; Fusion 6; VAS; 1 excellent, 2 good, 15 no improvement, 7 temporary</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Ziran 2007</td>
<td>Prospective</td>
<td>SI screws CT guided</td>
<td>No fusion was performed; VAS; Pre-post 8.3-3.3, 2 no effect</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Wise 2008</td>
<td>Prospective</td>
<td>MIS-threaded fusion cage BMP-2</td>
<td>89% fusion rate (17/19); LBP VAS score; Improvement: 4.9, 77% do it again</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Al-khayer 2008</td>
<td>Prospective</td>
<td>Hollow cylindrical screw</td>
<td>100% fusion; ODI; VAS; Pre-post: 59-45, 8.1-4.6, 1 no effect</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Khurana 2009</td>
<td>Prospective</td>
<td>Hollow cylindrical screw, Demineralized bone matrix</td>
<td>100% fusion; SF-36, Physical sum, Mental sum, Majeed; Pre-post: 29 - 52, 47 - 57, 37 - 79, 13 excellent</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Stark 2012</td>
<td>Prospective</td>
<td>Diana</td>
<td>92% fusion; Million VAS; Return to work; Improvement in total score (P&lt;0.0001), 28/41 work candidates</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sachs 2012</td>
<td>Prospective</td>
<td>iFuse</td>
<td>No revision; VAS; Pre-post: 7.9-2.3, 80% over 2 point on VAS</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Rudolf 2012</td>
<td>Prospective</td>
<td>iFuse</td>
<td>20% complication, 95% ingrowth after 6 m; NRS, Pain; Pre-Post 7.6-3.3, 20% partly satisfied</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mason 2013</td>
<td>Prospective</td>
<td>Hollow modular screw, DBM</td>
<td>NA; No late failures; VAS, Majeed; Pre-1 year 8-4.5, 36-65</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sachs 2013</td>
<td>Prospective</td>
<td>iFuse</td>
<td>No revisions; VAS; Pre-1 year 8.7-0.9, 1 failure</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Cunnings 2013</td>
<td>Prospective</td>
<td>iFuse</td>
<td>VAS, ODI; Pre-1 year 8.9-2.3, 53-13</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Duhun 2013</td>
<td>Prospective</td>
<td>iFuse</td>
<td>VAS, ODI; Pre-6 m 7.6-2.9, 55-39</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Endres 2013</td>
<td>Prospective</td>
<td>DIANA</td>
<td>79% fusion; VAS, ODI; Pre-13.2 m 8.5-6, 64-57</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Total</td>
<td>Total</td>
<td>631 patients</td>
<td>18 different techniques</td>
<td>Fusion rate 24%-100%</td>
<td></td>
</tr>
</tbody>
</table>
Precision and Accuracy Measurement of Radiostereometric Analysis Applied to Movement of the Sacroiliac Joint

Thomas J. Kibsgaard MD, Olav Roise MD, PhD, Britt Stuge PT, PhD, Stephan M. Rohrl MD, PhD

Abstract

Background Different techniques have been used to quantify the movement of sacroiliac (SI) joints. These include radiostereometric analysis (RSA), but the accuracy and precision of this method have not been properly evaluated and it is unclear how many markers are required and where they should be placed to achieve proper accuracy and precision.

Purpose The purpose of this study was to test accuracy and precision of RSA, applied to the SI joint, in a phantom model and in patients.

Methods We used a plastic phantom attached to a micrometer to obtain a true value of the movement of the SI joint and compared this value with the measured value obtained by RSA; the difference represented the accuracy. The precision of the system was measured by double examination in the phantom and in six patients, and was expressed by a limit of significance (LOS). We analyzed different marker distributions to find optimal marker placement and number of markers needed.

Results The accuracy was high and we identified no systematic errors. The precision of the phantom was high with a LOS less than 0.25 mm and 0.16 mm for all directions, and in patients, the precision was less than 0.71 mm for rotations and 0.47 mm translations. No markers were needed in the pubic symphysis to obtain good precision.

Conclusions The accuracy and precision are high when RSA is used to measure movement in the SI joint and support the use of RSA in research of SI joint motion.

Introduction

Because of high accuracy and precision, radiostereometric analysis (RSA), has become a well-established method for three-dimensional (3-D) measurements of micromotion in joints [9, 21]. Different radiographic modalities have been used to measure movement in the sacroiliac (SI) joint objectively [7, 12, 15], whereas five studies have used RSA to evaluate movement between the sacrum and the ilium [16–20]. With RSA, a maximum rotation of 3.6° has been measured and the translation never exceeded 2 mm in the SI joint, which is less movement than other methods have revealed [5], and for this reason the method has been questioned [3]. Despite the use of pelvic RSA in clinical research, the accuracy and precision have not been fully evaluated.

Accuracy of the measurement is the closeness of the measurement to its true value. In phantom models accuracy
reflects the level of agreement between a true value of movement and the results obtained using RSA. Systematic error (bias) of the system occurs when the differences between systems and experiments are uniform and if they can be corrected [13]. The precision (spread) of the measurement is the degree of closeness in repeated measurements under unchanged conditions. Under optimal conditions, the difference between two examinations should be zero [13].

Earlier studies have measured movement between the sacrum and the ilium with dorsally placed markers [16–20]. In these studies, the markers were placed close to the joint line, and because of the flat anatomy of the bones, the markers become collinear (in the same plane). As this is not necessarily the optimal 3-D distribution, the use of this technique has been questioned [3]. Guidelines for standardization of RSA of implants recommend at least three non-collinear markers in each segment (rigid body) [21]. A good 3-D configuration relies on the distance between the markers and the distribution of markers in all three axes, and a condition number (CN) expresses the quality of a marker segment [11]. The CN is a mathematical expression of how the markers relate to a straight line going through the segment [14]. A low CN represents a good scatter of markers in the segment. A CN less than 110 is considered a reliable distribution [21], and an upper limit of 150 is suggested. The CN will influence the precision and accuracy, and a factor of importance is how good the RSA system calculates the placement of each marker. The precision of each marker can be influenced by soft tissue disturbances and stability of the markers. If the markers are not thoroughly inserted in the bone, but end up in the soft tissue, the markers become unstable. This may occur in the sacrum because of the thick and strong dorsal ligaments covering the bone, especially in the cranial part. Unstable markers should be excluded if they move more than 0.35 mm between two examinations (mean error [ME] of rigid body fitting) [21]. Although RSA has been validated with the use of a phantom in a fracture model of the distal radius and hip and knee prostheses [8, 10, 11], it has not been validated for use in the pelvis.

We therefore (1) measured the accuracy, precision, and CNs of pelvic RSA with different marker distributions in a phantom model, (2) explored whether frontal markers around the symphysis improve the CN and precision and whether it is possible to avoid markers in the cranial part of the sacrum, and (3) compared the precision obtained by a phantom with the precision obtained by double examinations in patients.

**Patients and Methods**

We used a phantom model to measure accuracy and precision while precision was measured in six patients. The phantom was a full male plastic pelvis (Sawbone® 1301; Pacific Research Laboratories, Inc, Vashon, WA, USA) with detached ilium and sacrum. The left ilium was rigidly fixed to a platform and the sacrum was attached to a combined X, Y, Z translation (25 mm-PT3/M X, Y, Z Travel Translation Stage; Thorlabs, Inc, Newton, NJ, USA) and rotation stage (PR01A/M Precision Rotation Platform, Thorlabs) (Fig. 1). According to the manufacturer, the translation stage has a resolution of 0.01 mm and the rotation stage has a resolution of 1/25° (2.4 arcmin). The movement was performed around an X, Y, Z coordinate system with the X and Y axes in the plane of the table imitating a supine position (Fig. 1). The rotations were performed in three different setups (Fig. 1). One-mm markers were used. Five markers were inserted into the ilium posteriorly and three markers into the inferior pubic ramus. Another eight markers were inserted into the dorsal aspect of the sacrum (Fig. 2). The markers defined two rigid body segments, where the ilium was defined as the fixed segment and movement of the sacrum relative to the ilium was measured. We conducted 10 double examinations to analyze the precision. Between these examinations, the equipment was fully dismantled and reset.

To evaluate different marker distributions, the eight markers in each segment were divided into four different marker segments (MS): MS A with five dorsal markers in the ilium; MS B with three frontal markers in the inferior pubic ramus; MS C with six sacral markers; and MS D with two cranial markers in the sacrum (Fig. 2). To examine the need for frontal markers, we tested MS AB against MS A. In this setting, all eight sacral markers were included. To examine the need for cranial markers in the sacrum, we tested MS CD against MS C. All eight markers in the sacrum were included. Finally, three dorsal markers in the ilium and three markers in the sacrum were randomly selected (Fig. 2, circles) and tested against eight markers.

To determine the in vivo precision, six patients received 17 double examinations. The six patients were women, selected from a group of nine patients with long-lasting pelvic girdle pain after pregnancy included in an ongoing study where SI fusion of the affected SI joint was performed. These six patients had a mean age of 38 years (range: 33–47 years), BMI of 24 (range: 21–30), and pelvic girdle pain for 10 years (range: 5–25 years). The patients were analyzed with RSA at baseline, 3 months, and at 12 months after surgery. The maximum level of CN was set to 150 and the mean error (ME) was set to 0.35. With these limitations we had six patients with one, two, three, and six double examinations respectively. When the markers in the symphysis were removed from the analysis, 11 examinations had sufficient CN and ME to perform a comparison of precision. The patients were repositioned between the examinations, without moving
As there is movement in two joints in each examination (one in each SI joint), movement in the right SI joint was converted to represent the left SI joint. The total movement in one double examination was the mean of these two measurements.

To conduct the displacements, three different setups were required to cover all the translations and rotations. We performed all translations and Y rotations in one setup (Fig. 1A). We performed X rotations (Fig. 1B) and Z rotations (Fig. 1C) in separate setups. To measure accuracy, the sacrum was translated and rotated using the micrometers to measure the true value of movement. The sacrum was moved from Point 0 and the translations were performed in all three directions in one move. Film pairs were taken at 0.01, 0.02, 0.04, 0.08, 0.5, 1, 2, 3, and 5 mm translation. The phantom was set to zero before rotation. Every position was examined by double examinations. To simulate a change in position, a 1-cm support was placed under the phantom between the double examinations. The rotation was measured at 0.2, 0.5, 1, 3, and 5°. A total of 66 film pairs was taken.
Each pair of radiographs was taken with two x-ray tubes (GE Proteus XR/A™ system [GE Healthcare, Piscataway, NJ, USA] and Philips OPTIMUS [Philips Healthcare, Best, The Netherlands]) at an approximately 40°-angle to each other. An UmRSA Calibration Cage Number 43 (UmRSA Biomedical, Umeå, Sweden) was used and there was a film-focus distance of 155 cm. An exposure of 133 kV and 6.5 to 8 mAs was used. The digital images were analyzed using UmRSA Version 6.0 software (UmRSA Biomedical), and the markers were identified with user-assisted edge detection (UmRSA digital measure) [22]. The maximum limit for the CN was set to 150 and the maximum level of ME was set to 0.35.

We calculated the accuracy of the RSA system using the mean difference between the measured value and the true value ($d_{true}$), and the margin of error was expressed by a 95% CI with $n - 1$ degrees of freedom [13]. The precision for translation and rotation was expressed by the mean of the absolute value of the difference between two double examinations + $t_{0.025, n-1} \times SD_{absolute value}$ where $t$ follows the Student’s t-distribution with $n - 1$ degrees of freedom and 99% level of confidence. Some authors have used the expression limit of significance (LOS) [1, 4, 6] as an alternate way to express this precision. When a movement beyond this limit is found, an actual movement between the segments has occurred and the detected movement is larger than what can be explained by the measurement error. We used a paired t-test to detect any differences in accuracy and precision between different marker setups. We used SPSS® Version 18 (SPSS Inc, Chicago, IL, USA) for statistical analysis.

### Results

The CN in the phantom varied from 17 to 59 in the sacrum and 29 to 117 in the ilium (Table 1). With eight markers in the ilium and eight in the sacrum the accuracy of the phantom for rotation was between $-0.025^\circ$ to $0.051^\circ$. The largest error was observed in the Y rotation where the system underestimated the result by $0.051^\circ$. The translation accuracy was better in the in-plane directions (X, Y) than in the out-of-plane direction (Z). In the Z direction, the system underestimated the value by 0.065 mm (Table 2). The precision (LOS) in the phantom was between 0.06° and 0.25° for rotation and between 0.03 and 0.16 mm for translation (Table 3).

When the markers placed in the symphysis were removed, the CN went from 29 to 92. The ME was reduced ($p < 0.001$) when the frontal markers were removed (Table 3). There was no difference in the accuracy, but the precision was reduced in the Z rotation ($p = 0.010$) and the X translation ($p = 0.003$). However, when the two cranial-placed markers were removed (MS ABCD versus MS ABC), there was a reduction ($p = 0.012$) in the accuracy of the Y translation, and the precision was poorer in the Z rotation ($p = 0.023$) and the X translation ($p = 0.005$). When three randomly selected markers were analyzed compared with eight markers, there were reductions in precision of the Y and Z translations ($p = 0.016$ and $p = 0.013$, respectively), but there was no reduction in accuracy (Fig. 3).

The precision of the measurement in the phantom was better than in the patients (Table 3). Similar CN was achieved, but the mean ME was greater in the patients than in the phantom. When the frontal markers were removed, six double examinations could not be analyzed owing to insufficient CN, and therefore they were excluded. The 11 examinations remaining for analysis showed no change in the precision (Table 3). The CN in the ilium had a mean 38 to 96 increase ($p < 0.001$) and the ME had a tendency ($p = 0.048$) to be lower in the ilium without the frontal markers (Table 3).

### Discussion

RSA as a tool to measure 3-D movement in the SI joint has been used in a limited number of studies [16–20], but none...
Table 3. Limit of significance in the phantom with different marker distributions and in patients before and after the frontal markers were removed

<table>
<thead>
<tr>
<th>Model</th>
<th>Number of examinations</th>
<th>X Rot</th>
<th>Y Rot</th>
<th>Z Rot</th>
<th>X Trans</th>
<th>Y Trans</th>
<th>Z Trans</th>
<th>ME sacrum</th>
<th>ME ilium</th>
<th>CN sacrum</th>
<th>CN ilium</th>
<th>Markers in sacrum</th>
<th>Markers in ilium</th>
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<tbody>
<tr>
<td>Phantom model</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>With frontal markers (MS ABCD)</td>
<td>10</td>
<td>0.06</td>
<td>0.25</td>
<td>0.11</td>
<td>0.08</td>
<td>0.03</td>
<td>0.16</td>
<td>0.04 (0.01)</td>
<td>0.08 (0.02)</td>
<td>17</td>
<td>29</td>
<td>8</td>
<td>8</td>
</tr>
<tr>
<td>Without frontal markers (MS ACD)</td>
<td>10</td>
<td>0.19</td>
<td>0.18</td>
<td>0.22</td>
<td>0.18</td>
<td>0.09</td>
<td>0.23</td>
<td>0.04 (0.01)</td>
<td>0.02 (0.01)</td>
<td>17</td>
<td>92</td>
<td>8</td>
<td>5</td>
</tr>
<tr>
<td>p values (MS ABCD versus MS ACD)</td>
<td></td>
<td>0.254</td>
<td>0.801</td>
<td>0.010</td>
<td>0.003</td>
<td>0.284</td>
<td>0.313</td>
<td>&lt; 0.001</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Without cranial sacral markers (MS ABC)</td>
<td>10</td>
<td>0.09</td>
<td>0.22</td>
<td>0.22</td>
<td>0.24</td>
<td>0.07</td>
<td>0.18</td>
<td>0.02 (0.01)</td>
<td>0.08 (0.02)</td>
<td>43</td>
<td>29</td>
<td>5</td>
<td>8</td>
</tr>
<tr>
<td>p values (MS ABCD versus MS ABC)</td>
<td></td>
<td>0.077</td>
<td>0.568</td>
<td>0.023</td>
<td>0.005</td>
<td>0.067</td>
<td>0.876</td>
<td>&lt; 0.001</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>With 3 random markers in each segment</td>
<td>10</td>
<td>0.24</td>
<td>0.26</td>
<td>0.14</td>
<td>0.11</td>
<td>0.09</td>
<td>0.21</td>
<td>0.02 (0.01)</td>
<td>0.01 (0.01)</td>
<td>59</td>
<td>117</td>
<td>3</td>
<td>3</td>
</tr>
<tr>
<td>p values (MS ABCD versus MS circles)</td>
<td></td>
<td>0.053</td>
<td>0.457</td>
<td>0.191</td>
<td>0.140</td>
<td>0.016</td>
<td>0.013</td>
<td>&lt; 0.001</td>
<td>&lt; 0.001</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Patients</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
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<td></td>
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<tr>
<td>With frontal markers (n = 17)</td>
<td>17</td>
<td>0.71</td>
<td>0.24</td>
<td>0.28</td>
<td>0.47</td>
<td>0.28</td>
<td>0.38</td>
<td>0.09 (0.04)</td>
<td>0.13 (0.07)</td>
<td>27 (24)</td>
<td>43 (15)</td>
<td>6.4 (1.3)</td>
<td>5.6 (1.3)</td>
</tr>
<tr>
<td>Before removing markers in the symphysis</td>
<td>11</td>
<td>0.52</td>
<td>0.21</td>
<td>0.18</td>
<td>0.15</td>
<td>0.30</td>
<td>0.39</td>
<td>0.08 (0.02)</td>
<td>0.12 (0.05)</td>
<td>20 (8)</td>
<td>38 (9)</td>
<td>6.8 (1.4)</td>
<td>6.8 (1.3)</td>
</tr>
<tr>
<td>After removing markers in the symphysis</td>
<td>11</td>
<td>0.55</td>
<td>0.40</td>
<td>0.14</td>
<td>0.14</td>
<td>0.23</td>
<td>0.45</td>
<td>(0.08) (0.02)</td>
<td>0.08 (0.05)</td>
<td>20 (8)</td>
<td>96 (28)</td>
<td>6.8 (1.4)</td>
<td>4.3 (1.3)</td>
</tr>
<tr>
<td>p values (patients with versus without frontal markers)</td>
<td></td>
<td>0.213</td>
<td>0.248</td>
<td>0.249</td>
<td>0.592</td>
<td>0.811</td>
<td>0.711</td>
<td>0.341</td>
<td>0.048</td>
<td>&lt; 0.001</td>
<td>&lt; 0.001</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Precision expressed as limit of significance = \(\text{Mean of the absolute values} + 1.645 \times \text{SD of the absolute values}\); ME = mean error of rigid body fittings; CN = condition number; Rot = rotation; Trans = translation.
of these has assessed the accuracy and precision of the method in an experimental setting, as in the current study. We therefore (1) measured the accuracy, precision, and CNs of pelvic RSA with different marker distributions in a phantom model, (2) explored whether frontal markers around the symphysis improve the CN and precision and whether it is possible to avoid markers in the cranial part of the sacrum, and (3) compared the precision obtained by a phantom with the precision obtained by double examinations in patients.

We acknowledge limitations to our study. First, precision from phantom studies cannot be extrapolated to clinical data. Obviously, a phantom is the best tool to evaluate the accuracy of RSA as it ensured standardization and reproducible conditions for the test. However, these results cannot be transferred directly to the analyses of patients. When the 3-D position of each marker is calculated, the soft tissue is an important factor that can disturb x-rays and the markers probably are not as stable in patients as in a phantom. These factors make measurements less accurate and less precise. In patient series, therefore, precision always should be determined by double examination. Second, a potential source of error in our model is that the actual movement of interest exceeds ROM in our phantom. All clinical RSA studies describe small movements in the SI joint [5, 16–20]. In these studies, a maximum rotation of 3.6° is seen and the translation never exceeds 2 mm. Our displacement protocol included a range of 0 to 5 mm for translations and 0° to 5° for rotation and therefore should cover the normal and potentially pathologic interval of movement. Third, we used a micrometer as a gold standard or true value. This micrometer has a resolution of 0.04 mm and 0.01 mm. The accuracy was less than 0.07 mm for translation and 0.05° for rotation, and to conclude that this is an actual bias (systematic error), the resolution should have been better. The bias measured in our study is small, and can be explained by the resolution of the micrometer. There is no other logical explanation for the bias and in our opinion, based on our results, no correction of the measurements should be done.

The accuracy of the RSA method depends on the marker distribution (CN) and stability of the markers (precision of calculating the 3-D positions of individual markers) [21]. In the phantom experiment, the markers are inserted and glued to the pelvis and become 100% stable. When the precision of each marker is high, a phantom experiment is useful to calculate CN and highest possible precision for different marker distributions. In the phantom a CN never exceeded 117, and no more than three markers were needed in each segment to achieve this. With eight markers in each segment, the accuracy was less than 0.07 mm in all directions for translation and 0.05° for rotations. There were some minor differences between the different marker distributions, but we found no systematic error that influenced our measurements. The precision was less than 0.16 mm for translations and less than 0.25° for rotations, and these results are comparable to precision measured in pelvic phantoms by others (Table 4) [19, 20]. The phantom experiments qualify pelvic RSA as a proper tool for analysis of 3-D movement in the SI joint, but they do not provide a prediction of precision in a clinical situation where stability of the markers is different. With frontal markers around the symphysis, the CN was 29 in the ileum, but the CN was never greater than 117 using dorsal markers only. The ME was reduced when the frontal markers were removed. Different marker distributions showed some differences, but these differences were extremely small. In all setups, the precision never exceeded a LOS of 0.24 mm for translations and 0.26° for rotations. The use of frontal markers was an easy way to decrease the CN, but this study does not take the biomechanical properties of the ilium and the pubic bone into consideration. Possible plasticity of the iliac and pubic bones can disqualify the use of frontal markers, but that is beyond the scope of this study. When three markers were used, the CN was 117 in the ilium. This is not optimal [21]. However, the accuracy and precision were almost as good as when eight markers were used. We therefore conclude, as long as the markers in the dorsal aspects of the ilium are placed...
with the best spread possible, there is no need for frontal markers.

In the initial patients, we found that sacral markers, especially the cranial ones, were hard to get into the bone and often ended up in the soft tissue. This probably is attributable to the strong dorsal ligaments covering the sacrum. CT scans performed to evaluate the quality of the SI joint arthrodesis documented this. When these cranial markers were excluded in the phantom, the CN ranged from 17 to 43, which is acceptable. Without these markers, there were reductions in accuracy in the Y translation and in precision of the Z rotation and X translation. As in the evaluation of the frontal markers, these differences were small, and the precision was good without these markers. Precision was better in the phantom than in the patients. In the patients, the precision of the rotations had a LOS less than 0.7° in all directions, and for translation the precision was less than 0.5 mm (Table 3). These values were comparable to precision measurements by others (Table 4) [19, 20]. The RSA method has been widely used in the study of hip prostheses. Using double examinations, precision was reportedly between 50 and 150 \( \mu \text{m} \) [2]. When RSA is used in the SI joint, the segments are large and the distance between the segments (sacrum and ilium) is larger than in analysis of implants. The ME also tended to be larger in the patients with frontal markers, and all these factors might have a negative influence on the precision [9]. Abdominal and pelvic soft tissue also might decrease the quality of marker observation on the radiograph and these factors probably explain why our results are somewhat poorer. Furthermore, zero motion between two examinations cannot be expected. As the patients were allowed to move between the examinations, it is possible that the joints might align differently between the two examinations. Therefore, when precision in patients is evaluated, the joint alignment must be taken into consideration. It was difficult to obtain a proper CN in patients when the frontal markers were removed. All examinations that were excluded had less than four stable markers in the ilium. It seems that four markers with a good spread is enough to obtain good precision, however, we recommend putting more than four markers in the dorsal aspects of the ilium to ensure that no examination ends up being excluded. This is important, especially in patients with implants, were markers can be hidden by the implant.

Our data suggest RSA is a reasonable method to measure SI joint movement in patients. The accuracy was high and we identified no systematic error in the phantom study that would require correction. Precision was high in the phantom and as long as the CN was less than 120 and the ME less than 0.35, we found no need for more than three to four properly placed markers in the back of the pelvis to measure SI joint movement. In patients however, we recommend more than four markers in each segment to ensure an appropriate segment for the analysis. The use of frontal markers did not improve precision, and therefore are not needed.

**Acknowledgments** We acknowledge the assistance of Alexis Hinojosa (MRI radiographer; Department of Radiology and Nuclear Medicine, Oslo University Hospital, Oslo, Norway) with the RSA radiographs and analyses and Ingar Holme PhD (statistician, Department of Biostatistics and Epidemiology, Oslo University Hospital, Oslo, Norway) for help with statistics.

**References**


Radiosteriometric analysis of movement in the sacroiliac joint during a single-leg stance in patients with long-lasting pelvic girdle pain

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A B S T R A C T

Background: Chamberlain’s projections (anterior–posterior X-ray of the pubic symphysis) have been used to diagnose sacroiliac joint mobility during the single-leg stance test. This study examined the movement in the sacroiliac joint during the single-leg stance test with precise radiostereometric analysis.

Methods: Under general anesthesia, tantalum markers were inserted into the dorsal sacrum and the ilium of 11 patients with long-lasting and severe pelvic girdle pain. After two to three weeks, a radiostereometric analysis was conducted while the subjects performed a single-leg stance.

Findings: Small movements were detected in the sacroiliac joint during the single-leg stance. In both the standing- and hanging-leg sacroiliac joint, a total of 0.5 degree rotation was observed; however, no translations were detected. There were no differences in total movement between the standing- and hanging-leg sacroiliac joint.

Interpretation: The movement in the sacroiliac joint during the single-leg stance is small and almost undetectable by the precise radiostereometric analysis. A complex movement pattern was seen during the test, with a combination of movements in the two joints. The interpretation of the results of this study is that the Chamberlain examination likely is inadequate in the examination of sacroiliac joint movement in patients with pelvic girdle pain.

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1 Introduction

The sacroiliac joint (SIJ) is a possible cause of pain in patients with low back and pelvic girdle pain (PGP), with a reported prevalence ranging from 10% to 60% depending on the patient population and choice of diagnostic criteria (Cohen et al., 2013; Simopoulos et al., 2012; Vleeming et al., 2008). A large amount of force is transferred from the spine to the legs through the pelvis and SIJ joints. In an upright position, when gravitational forces are transferred through the sacrum, the anatomy of the SIJ ligaments and muscles locks the joint and stabilizes the pelvic girdle (Snijders et al., 1993). Locking occurs when the sacrum rotates forward (Sturesson et al., 1989) and is explained by the theory of form and force closure (Sturesson et al., 2000a). An increased movement in the SIJ might reduce stability and result in stress of the SIJ ligaments, impaired motor control and consequently pain (Mens et al., 2009; Siegel et al., 2008; Snijders et al., 1993). However, the exact movements that occur in the SIJ are still debated (Goode et al., 2008; Jacob and Kissling, 1995; Mens et al., 1999; Walker, 1992).

Despite the difficulty in measuring true movement of the SIJ, several attempts have been made using different experimental techniques, such as k-wires, CT, cadaver studies, skin markers, X-rays and radiostereometric analysis (RSA) (Hungerford et al., 2004; Jacob and Kissling, 1995; Lavignolle et al., 1983; Smidt et al., 1995; Sturesson et al., 1989; Sturesson et al., 1999; Sturesson et al., 2000a; Sturesson et al., 2000b). However, these techniques are impractical in a clinical practice. In 1930, Chamberlain described an easy and practical method to measure pubic movement on an anterior–posterior (AP) pelvic X-ray while the patient stands on one leg with the other leg hanging down (single-leg stance) (Chamberlain, 1930). In patients with SIJ pain, Chamberlain found that weight bearing caused a cranial displacement of the pubic bone on the side of the painful joint. This displacement was explained by a rotation around the axis that was perpendicular to the SIJ surface. The Chamberlain technique has since been used to examine pubic bone movement and indirect SIJ hypermobility (Mens et al., 1999).

Since the Chamberlain technique was first described, researchers have attempted to correlate pubic movement to SIJ pain (Anderson...
and Peterson, 1944; Mens et al., 1999; Siegel et al., 2008). Chamberlain found a clear pattern in his patients, but later Mens et al. (2009) found the exact opposite where the hanging leg caused a downward displacement of the pubic bone on the side of the painful joint. These differences have made it hard for clinicians to use the results of the test in the diagnosis of PGP, especially when normal variations of the movement in the pubic symphysis have proven to be large (Garras et al., 2008). Measurements of the movement in the SIJ during the single-leg stance have been done using k-wires, however only measured on healthy subjects without SIJ pain (Jacob and Kissling, 1995). As the Chamberlain technique is an indirect measure of SIJ movement, it is still unknown what really occurs in the SIJ during the single-leg stance test in patients with PGP.

In a systematic review Goode et al. (2008) concluded that the measurement techniques with the lowest level of error also reported the lowest values of movement in the SIJ. The RSA technique is highly accurate and precise (Kibsgård et al., 2012), but has not been utilized to examine SIJ movements during the single-leg stance test. Therefore, the aims of the present study were to measure movement in the SIJs of the standing leg and the hanging leg.

2. Methods

We used RSA to measure the in vivo movement of the SIJ in patients with PGP. All patients signed an informed consent, and the study was approved by the Regional Committee for Medical and Health Research Ethics (Number: 1.2006.1574).

2.1. Patients

From 2007 to 2010, 17 patients with severe PGP were assigned for SIJ fusion at two orthopedic centers, Oslo University Hospital, Norway and Ängelholm Hospital, Sweden. The inclusion criteria were long-lasting pain localized to one or both SIJs, minimum of two out of five positive SIJ tests (posterior pelvic pain provocation test, active straight leg raise, palpation of the long dorsal sacroiliac ligament, modified Trendelenburg test, palpation of the symphysis (Vleeming et al., 2008)) and a high degree of pain and disability as measured by the visual analog scale (VAS) and the Oswestry disability index (ODI). Patient characteristics at inclusion are presented in Table 1. All patients had normal spinal MRIs, and the patients had either CT scan or/and MRI of the SIJ. The pelvic MRI or CT was primary done to exclude patients with sacroiliitis. Seven out of 11 did not have any radiographic abnormalities. In three patients there were light unilateral degenerative changes in the side that were later operated on, and one had bilateral degenerative changes. Two out of these patients also had anterior osteophytes on the side that were operated. After evaluating the RSA data, six patients were excluded because of poor X-ray quality, leaving 11 patients for the final analysis (10 females and 1 male). The patients were excluded because of misplaced markers in the soft tissue or insufficient visualization of the markers on radiographs during the software analysis.

2.2. RSA protocol

Under general anesthesia, 1 mm RSA tantalum markers were inserted into the dorsal sacrum and the ilium with a marker gun through small skin incisions. An imaging intensifier was used to assure proper placement. RSA X-rays were taken after 2–3 weeks. Three pairs of X-rays were taken under the following conditions: 1) standing on both legs, 2) standing on the right leg and 3) standing on the left leg, with full weight bearing according to Chamberlain examination procedure (Fig. 1). Each pair of radiographs was taken with two X-ray tubes. As we used the standard set-ups in Norway and Sweden respectively, the RSA set-ups were slightly different in the two centers. The software program has, however, the ability to compensate for the deviation automatically because its retrograde calculates the position of the tubes depending on the control markers in calibration cage. In Norway, X-ray tubes from the GE system (GE Healthcare, Piscataway, NJ, USA) and Philips OPTIMUS (Philips Healthcare, Best, The Netherlands) were used. The tubes were at an approximately 40° angle to each other, with a film-focus distance of 155 cm, an exposure of 133 kV and 6.5 to 8 mAs using UmRSA Calibration Cage number 43. In Sweden, two GE systems (GE Healthcare, Piscataway, NJ, USA) tubes were used. The tubes were at a 30° angle to each other, with a film-focus distance of 130 cm, an exposure of 125 kV and 13 mAs using UmRSA Calibration Cage number 41. The digital images were analyzed using UmRSA Version 6.0 software (UmRSA Biomedical), and the markers were identified with user-assisted edge detection (UmRSA digital measure). The RSA software calculates the translation and rotations in a x,y,z

Table 1

<table>
<thead>
<tr>
<th>Patient characteristics, n = 11.</th>
<th>Mean (Range)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (years)</td>
<td>39 (29–47)</td>
</tr>
<tr>
<td>Body mass index</td>
<td>24 (19–30)</td>
</tr>
<tr>
<td>Female/male</td>
<td>10/1</td>
</tr>
<tr>
<td>Duration of symptoms (years)</td>
<td>8 (1.5–20)</td>
</tr>
<tr>
<td>Oswestry disability index</td>
<td>56 (26–76)</td>
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<tr>
<td>Evening VAS</td>
<td>75 (53–91)</td>
</tr>
<tr>
<td>Etiology (numbers)</td>
<td>6</td>
</tr>
<tr>
<td>Post-pregnancy</td>
<td>4</td>
</tr>
<tr>
<td>Trauma</td>
<td>1</td>
</tr>
<tr>
<td>Idiopathic</td>
<td>7/4</td>
</tr>
</tbody>
</table>

VAS = visual analog scale.

Fig. 1. RSA setup. A—Patient standing on both feet, B—Patient standing on left leg with the right leg hanging down, C—Patient standing on right leg with the left leg hanging down.
The patients were examined with weight-bearing on the right side followed by the left side. Since there are differences in interpretation of positive and negative movement between left and right side, we converted the sign. So all calculations were performed in a setting where the patients were standing on the right leg with the left leg hanging down. The mean of the two sides was then used in the analysis. The movement in each direction is presented as the mean, standard deviation and range. The $H_0$ hypothesis was that there was no movement in the SIJ during the single-leg stance. A one-sample t-test was used to determine whether the mean was significantly different from zero. To determine if there were any differences between the hanging leg and standing legs SIJ a paired sample t-test were used. Because the measurement values were close to the RSA precision, the fraction of measurements that exceeded this threshold is presented along with the range. We used SPSS® Version 18 (SPSS Inc., Chicago, IL, USA) for the statistical analysis.

### 3. Results

Eleven patients (4 from Norway and 7 from Sweden) with long-lasting PGP were analyzed. Only small movements in the SIJ were detected, and only 15% of the measurements exceeded the RSA precision. Although some mean values were significantly different from zero, all but one of these mean values was below the RSA precision.

When the patients performed a single-leg stance there were almost no detectible movement (Table 2). There were mean 0.5° of rotation on both sides around a helical axis (the true axis of rotation). When the movements were assessed based on the coordinate system (Fig. 2), a small 0.3-degree (SD 0.2) rotation around the z-axis in the SIJ of the standing leg ($P < 0.001$) was observed (Fig. 3), and this rotation was significantly different from the hanging-leg SIJ ($P = 0.036$). Although the 0.3-degree rotation was significantly different from zero, only 50% of the patients had a movement that exceeded the threshold of precision, with a maximum value of 1.0°. No translations were detected (Table 2). With the exception of the 0.2 degree difference observed in the z-axis rotation ($P = 0.036$), no differences were observed in the movement between the SIJs of the standing leg and the hanging leg ($P$-values between 0.055 and 0.978).

Four patients presented with clear unilateral symptoms. All other patients had one or more positive SIJ provocation test(s) or pain localized bilaterally on pain diagrams. There were no difference between the 18 symptomatic joints and the four asymptomatic joints with regard to the total amount of rotation (diff: $-0.2, P = 0.335$ on the standing side; diff: $0.0, P = 0.896$ on the hanging side) and translation (diff: $-0.1, P = 0.398$ on the standing side; diff: $0.1, P = 0.687$ on the hanging side).

### Table 2

Movement in the sacroiliac joint during the one-leg stance ($n = 11$).

<table>
<thead>
<tr>
<th>Movement in the standing-leg SIJ (right)</th>
<th>Movement in the hanging-leg SIJ (left)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mean (SD)</td>
<td>$P$-value</td>
</tr>
<tr>
<td>-----------</td>
<td>---------</td>
</tr>
<tr>
<td><strong>Rotation</strong></td>
<td></td>
</tr>
<tr>
<td>X</td>
<td>0.1 (0.2)</td>
</tr>
<tr>
<td>Y</td>
<td>0.1 (0.2)</td>
</tr>
<tr>
<td>Z</td>
<td>0.3 (0.2)</td>
</tr>
<tr>
<td><strong>Translation</strong></td>
<td></td>
</tr>
<tr>
<td>X</td>
<td>0.0 (0.2)</td>
</tr>
<tr>
<td>Y</td>
<td>0.0 (0.1)</td>
</tr>
<tr>
<td>Z</td>
<td>0.1 (0.1)</td>
</tr>
<tr>
<td><strong>Helical axis</strong></td>
<td></td>
</tr>
<tr>
<td>Rotation</td>
<td>0.5 (0.3)</td>
</tr>
<tr>
<td>Translation</td>
<td>0.0 (0.1)</td>
</tr>
</tbody>
</table>

Mean = mean movement in the sacroiliac joint; rotations in degrees and translations in mm; $P$-value = t-test whether the mean values were different from zero; % above precision = the fraction of the measurements that are above the RSA precision; helical axis = the true axis of rotation.

Fig. 2. The x, y, z coordinate system.
Only two patients had a visible step in the symphysis during the single-leg stance (Fig. 4), and these patients had bilateral symptoms, with the right side being more painful. On the X-ray of the pubic bone, there was a visible caudal shift of the pubic bone on the side of the hanging leg, and this shift was observed on both sides. The RSA measures showed a combination of rotation in the SIJ of the standing leg and in the SIJ of the hanging-leg side, with a relative forward rotation of the hanging leg's innominate (Fig. 4). When the patients went from standing on the right leg to standing on the left leg a 1 and 1.4° of rotation around the x-axis were seen between the two innominates.

Because the patients were observed in two different centers with different laboratory setups, the results from the two labs were compared. There was a 0.3 degree difference in the X-axis rotation of the standing legs SIJ, but this statistical significant difference was only a small variation below the precision of 0.7°. Except for this, no differences were found between the two labs.

4. Discussion

This is the first study to directly measure the movement in the SIJ during the single-leg stance in patients with PGP. Only small movements were detected. A total of 0.5° of movement were observed on both sides, and there were no differences in total movement between the SIJ of the hanging and standing legs.

One limitation of our study is that only 11 patients were included. They were recruited from two different clinics. However, the results were uniform between the two clinics, which strengthen the validity of our results. A strength of this study is that the patients were selected by strict criteria and had a long-standing history of severe PGP. The patients reported a high degree of disability and pain and were referred for SIJ fusion due to the severity of their symptoms and the failure of non-surgical treatment. We believe that these patients are representative of patients with severe PGP.

A major strength of this study is that we used the RSA technique, which has a high degree of precision and accuracy (Kibsgård et al., 2012). To our knowledge, more precise techniques evaluating movements than RSA do not exist. Because the tantalum markers are stable in the bone segments and are situated close to the joint line, RSA is more precise than other techniques that are used to examine SIJ movement. A possible limitation is that we only used RSA radiographs, and

Fig. 3. Mean movement in the SIJ with weight-bearing on the right leg. A mean z-rotation of 0.3° is observed and corresponds to the force of the hanging leg on the sacrum.

Fig. 4. Sacroiliac joint movement in the two patients with a detectible movement in the pubic symphysis on an anterior–posterior RSA X-ray. A–Standing on both legs, B–Standing on the right leg, C–Standing on the left leg. Numbers presented are degree of rotation around the x-axis (positive value = forward rotation, negative value = backward rotation).
not AP X-rays, to measure and identify patients with a large and clear pubic movement. Hence, we might have missed those with small movements in the pubic symphysis. The RSA X-rays are, however, sufficient to identify large and abnormal movements. It would have been an advantage to have included the Chamberlain X-ray in our protocol, even though the Chamberlain X-ray is associated with measurement errors and normal variations (Garras et al., 2008; Ruch and Ruch, 2005). According to Garras et al. (2008) the normal variation could be up to 5 mm in asymptomatic volunteers (1.5 mm in men and nulliparous women and 3.1 mm in multiparous woman).

Chamberlain reported a clear pattern of cranial displacement of the pubic bone on the symptomatic side upon weight bearing (Anderson and Peterson, 1944; Chamberlain, 1930). These findings contradict those of Mens et al. (2009), who observed a caudal displacement on the hanging-leg side when symptoms were localized to the hanging-leg SIJ. However, both Chamberlain and Mens observed a caudal slip of the hanging-leg pubic symphysis. Their reported differences in the correlation between pubic movement and clinical manifestations may be a result of the different patient populations (Mens et al., 1999).

Chamberlain (1930) included patients with acute low back pain and Mens et al. (1999) included women with SIJ pain after delivery. Although there are some discrepancies among the studies that have used the Chamberlain technique (Anderson and Peterson, 1944; Chamberlain, 1930; Mens et al., 1999), these studies conclude that hypermobility in the painful SIJ is responsible for movement in the pubic symphysis. In our study, we observed only a small amount of movement in the SIJ. Moreover, there were similar movements in both the standing- and hanging-leg SJs. We discovered two cases with a visible step in the pubic symphysis on X-ray. However, these two patients reported bilateral SIJ pain and possibly the results might have been different if their symptoms were unilateral. Nonetheless, four cases with unilateral symptoms did not have any asymmetrical movement in the SIJ. Based on these results, the movement pattern of the SIJ appears to be more complex than a single unidirectional rotation. Instead, SIJ movement patterns are likely best explained by a combination of movements between the two innomates. With this level of complexity, the Chamberlain technique is likely insufficient to quantify SIJ movement. Hence, our interpretation is that the Chamberlain examination should not be recommended in the examination of patients with PGP.

The literature disagrees on the degree of SIJ movement that occurs during a single-leg stance, with reports ranging from movement that can be felt by an examiner (Hungerford et al., 2007) to our RSA studies that have demonstrated almost no movement. The results of our current study were similar to the RSA study by Sturesson et al. (2000a) in which the patients performed a standing hip flexion test (with the hip maximally flexed). Sturesson et al. (2000a) found 0.6° of rotation in the SJs of both the standing and contra-lateral legs. In the standing flexion test the joint is theoretically compressed and stabilized, and one could have expected more movement during the Chamberlain examination because the absence of muscular stabilization of the joint. In addition, Sturesson et al. (1989) reported no differences between the symptomatic and asymptomatic joints. Jacob and Kissling (1995) used k-wires to measure movement during the single-leg stance and found a total movement of 1.5° between the two innomates; however, these analyses were performed with healthy participants aged between 20 and 50 years. Our study supports previous studies that demonstrate small movement in the SIJ in standing positions. Based on this observation, the SIJ pain is unlikely to be caused by hypermobility of the SIJ and the SIJ movement is probably too small to assess with palpation (Freburger and Riddle, 2001). It is important to note that these studies were performed in the standing position, during which movement is expected to be small due to the theory of form and force closure (the self-bracing mechanism of the pelvis).

Future studies will need to address the movements of the SIJ in an unloaded pelvis.

With the minimal movement detected in this study it could be questioned whether it is theoretically possible that these small movements could result in a detectible movement in the pubic symphysis. In this scenario, most of the rotation would be around the x-axis. The normal distance from the sacral promontory to the pubic symphysis is approximately 10–12 cm, and the distance likely increases to 15 cm because the markers are placed in the dorsal parts of the pelvis. With these assumptions and simple trigonometry, we calculate that a rotation of 2–3° would be necessary to trigger a 5 mm shift of the pubic symphysis. In the two patients with a visible step, a 1.0 and 1.4° of rotation between the two innomates were observed (Fig. 4), and this rotation should theoretically result in a 3–4 mm step. The total amount of rotation around the helical axis (true axis of rotation) was in the same patients 1.1 and 1.5, which support earlier assumptions of the axis of rotation close to the x-axis. Mens et al. (1999) observed a caudal shift of the pubic symphysis during the single-leg stance at the symmetrical hanging leg and concluded that a SIJ rotation had occurred on the side of the hanging leg. We found the same caudal shift, but the rotation in the SIJ was a combination of rotation (rotation around the x-axis) of the hanging-leg SIJ and a rotation of the standing-leg SIJ. Although our theoretical assumption is simplified, it is unlikely that this minimal SIJ movement can cause a clear and large movement in the pubic symphysis. This can easily be explained by differences in X-ray projection between the three different positions, a factor which has been reported to be of importance in the analysis of Chamberlain test (Ruch and Ruch, 2005). An alternative explanation is the plasticity of the bone and deformation of the innomates. There are some indications that deformation of the innominate bone exists and could contribute to this phenomenon (Pool-Goudzwaard et al., 2012).

5. Conclusion

In patients with PGP the movements in the SJs during the single-leg stance are small and almost undetectable by the precise RSA method. We measured a mean rotation of 0.5° on both the standing- and hanging-leg SJs, and no translation was detected. There were no differences in total movement between the standing- and hanging-leg SJs. The interpretation of the results of this study is that the Chamberlain examination is likely inadequate in the examination of SIJ movement in patients with PGP.

Acknowledgments

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References


Pelvic joint fusion in patients with severe pelvic girdle pain – a prospective single-subject research design study

Thomas J Kibsgård1*, Olav Røise1,2 and Britt Stuge1

Abstract

Background: The fusion of the pelvic joints in patients with severe pelvic girdle pain (PGP) is a controversial and insufficiently studied procedure. The aims of this study were to evaluate physical function and pain after sacroiliac joint (SIJ) fusion.

Methods: A single-subject research design study with repeated measurements was conducted; pre-operatively and at 3, 6 and 12 months post-operatively. The outcome measures considered were the Oswestry disability index (ODI), visual analogue scale (VAS), and SF-36. Eight patients with severe PGP received open-accessed unilateral anterior SIJ fusion and fusion of the pubic symphysis.

Results: Seven patients reported positive results from the surgery. At 1 year post-operation, significant (p < 0.001) reductions in ODI (54 to 37) and VAS (82 to 57) were reported. The physical functioning, bodily pain, and social functioning scores in the SF-36 were also improved.

Conclusion: Positive and significant changes in disability and pain at 1 year after SIJ fusion were observed. Despite these positive results, open accessed anterior fusion of the SIJ was associated with adverse events and complications such as infection and nerve damage.

Keywords: Sacroiliac joint, Fusion, Pain, Arthrodesis, Surgery, Pelvic girdle pain

Background

The sacroiliac joint (SIJ) may be the source of pain for 13-30% of patients with low back pain [1], and possibly an even higher proportion of patients suffering from “failed back surgery” [2,3]. This pain may be caused by specific pathology of the joint [4], but the specific role of the SIJ in unspecific pelvic girdle pain (PGP) disorder remains unknown. PGP is a common complaint in pregnancy that might cause disability, and in some women the complaint continues after delivery [1,5]. The origin and diagnosis of PGP are also unclear, as radiological findings are absent and the diagnostic criteria lack sufficient evidence. However it has become increasingly clear that patients with PGP have a different clinical presentation than patients suffering from low back pain [6]. Based on the theory of pathological joint mobility, SIJ fusion associated with symphysis pubis fusion is a therapeutic option when conservative treatment is unsuccessful [7].

SIJ fusion was first described by Smith-Petersen in 1921 [8]. Pelvic joint fusion has since been reported in a number of studies, but there is limited evidence in support of its efficacy [9-13]. The results of pelvic joint fusions have mostly been reported in small case series, with the exception of one study that included 58 patients, however without a control group [12]. The reported short-term results have been mainly positive [9,10,12,13], but poor results have also been reported [11]. One recent study showed that among patients with successful short-term outcomes, the effect was sustained 23 years post-operatively [14].

A randomized controlled trial is the gold standard to examine the effect of an intervention. As SIJ fusion is performed on few patients, a single-center randomized controlled design is difficult to establish due to the low
number of participants. However single subject research design (SSRD) have been recommended as a useful method to examine clinical accountability [15]. If properly applied, a SSRD can provide a systematic approach to documenting clinical change, and also provide objective evidence regarding the efficacy of a treatment modality [15]. SSRD refers to a study of a single patient or a small number of patients, observed over time, in which the treatment and outcome variables are controlled. The design comprises of multiple measurements before (baseline), and at different phases after, the intervention [15,16]. SSRD focuses on individual responses and repeated measurements that improve the validity of the study. When the SSRD is replicated across patients, the internal and external validity is strengthened and allows inferences to be made about effectiveness.

The primary aim of this prospective study was to examine changes in pain and physical function at 3, 6, and 12 months after SIJ fusion. The secondary aims were to evaluate post-operative health-related quality of life and patient satisfaction with treatment.

**Methods**

During the study period, from 2007 and 2010, a total of 20 patients with PGP were referred to our pelvic centre, but only 9 patients met the study’s inclusion criteria (Figure 1). A SSRD was used to evaluate the outcomes for pain, disability and health-related quality of life [15,16]. Five data collection sessions were conducted in each of the following 4 phases: prior to surgery (baseline) and at 3, 6, and 12 months after surgery. Inclusion and exclusion criteria are presented in Table 1. Based on previous findings that female patients with PGP have variations in pain intensity during their menstrual cycle [17], the patients filled out a questionnaire every Thursday for 5 weeks in each phase to ensure that evaluations were made throughout the menstrual cycle. The questionnaires were returned weekly by mail. All patients underwent 3 clinical examinations and CT guided SIJ injections before the decision for SIJ fusion was taken. The CT guided injections were performed by two experienced radiologists, and the patients filled out a VAS scale before and 2 hours after the injection. The patients received surgery to fuse the most painful SIJ, and the pubic symphysis was fused in all cases. The fusions were evaluated with a CT scan at 1-year follow-up.

All patients signed a written informed consent for participation. The project was approved by the Regional committees for medical and health research ethics, Region South East, Norway (number: 1.2006.1574) and registered in the Clinical Trials Database (reference number: NCT00900601).

**Outcome measurements**

Function was measured according to the Oswestry disability index (ODI) [18], each patient’s most severe morning and evening pain intensity was assessed using a 100 mm visual analogue scale (VAS) (0 = no pain, 100 = worst possible pain), and health-related quality of life was assessed using the SF-36 [19]. The ODI is a 10-item questionnaire that assesses function (0–100, with lower scores indicating less disability), for which a 10-point difference represents a significant clinical change [18,20]. In addition to the VAS, diagrams were used to record pain localization (Figure 2). The SF-36 questionnaire contains 36 items representing 8 subscales, including physical functioning, role limitations due to pain, bodily pain, general health, vitality, social functioning, role limitations due to emotional challenges, and mental health. The SF-36 scores are transformed to a 0–100 scale for each subscale. The higher the score, the
better the health status. Additionally, the patients answered the following two questions: “Have you experienced any effect of the surgery? If so, would you grade this as excellent, good, some, minor or no effect?” and “How do you tolerate physical activity now, as compared to before surgery?”

Surgical procedure

The patients received unilateral SIJ fusion, of the most painful SIJ, and symphysiodesis. An anterior approach with a skin incision over the iliac crest was used to reach the SIJ. The joint was partially resected, and grafted with cancellous bone from the ipsilateral iliac crest. Two AO reconstruction plates or AO-DC plates (Synthes®, Synthes GmbH, Switzerland) were used (Figure 3) to achieve stabilisation. The pubic symphysis was accessed through a bikini line incision. A 2 × 2 cm bone block was removed and replaced with a bone graft from the iliac crest, and a Matta plate (Stryker®, Michigan, United States) was applied (Figure 3). Post-operatively, the patients received epidural anaesthesia pain relief and 1–2 days of wound drainage. The patients were advised to avoid full weight bearing activities, on the operated side, for 8 weeks after the surgery.

Data analysis

The graphed data were analysed according to the guidelines for SSRD [16]. The levels (mean measurements over the 5-week period) and variability of the measurements are presented graphically. To analyse the changes in ODI, VAS, and SF-36, a mixed model for repeated measurements was used. The statistical analyses were performed using STATA 12.0 (Statacorp, Texas, USA).

ODI, VAS, and individual items of the SF-36 were used as dependent variables, and time was used as an independent variable. This provided a regression line = constant value (baseline) + regression coefficient × time, where time was defined as either 0 = baseline, 1 = 3 months, 2 = 6 months or 3 = 1 year. The correlation structure was specified as independent, and the regression slopes were allowed to vary at random. The correlation matrix was also tested as unstructured, but this did not alter the regression slope. We considered differences significant if the p value was less than 0.05.

Results

Nine consecutive patients received unilateral anterior SIJ fusion and fusion of the pubic symphysis. One patient developed chronic fatigue syndrome during the follow-up and dropped out of the study after 6 months. The remaining eight patients followed the study protocol; the baseline characteristics of these patients are presented in Table 2. Seven of the patients reported bilateral SIJ symptoms, mostly marked on one side, and six of them also had pain in the pubic symphysis. One patient reported unilateral SIJ pain and pain in the pubic symphysis. All patients had CT guided injection before surgery. Five of these patients experienced more than 70% pain relief from the injection (patient 1, 4, 6, 7 and 8) and 3 patients (2, 3 and 5) experienced no effect from the injections.

The ODI scores of each patient are presented in Figure 4. All but one patient exhibited a decrease of more than 10 points on the ODI from the pre-operative period to the 1-year follow-up. One patient experienced no effect. There was a strong association between ODI and time, with a 17-point decrease (p < 0.001) at 1 year after surgery (Table 3, Figure 4). The graphs show significant variations in the measurements at each time point. At baseline, a difference of more than 40 points between the maximum and minimum values was observed in two patients, and only two patients had less than a 10-point difference.

The VAS scores of each patient are presented in Figure 5. The patients showed a reduction in pain, with a decrease from 82 points at baseline to 57 points after 1 year (p < 0.001, regression coefficient of –8.4) (Figure 5, Table 2). All patients reported a decrease in pain. The difference between the pre-operative status and the 1-year follow-up scores was greater than 40 points in three patients, between 22 and 29 in three patients, and 15 points in one patient. One patient had only a minor change (3 points); although she had a positive SIJ injection she showed a more generalised pain pattern than the other patients (Figure 2). Pre-operatively, a difference of 43 points between the maximum and minimum scores was observed in one patient, and no patient had variations of less than 10 points.

Table 1 Inclusion and exclusion criteria

<table>
<thead>
<tr>
<th>Inclusion criteria</th>
<th>Exclusion criteria</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Pain located to one or more pelvic joints</td>
<td>1. Known psychiatric diagnosis,</td>
</tr>
<tr>
<td>2. Minimum two positive out of five clinical tests:</td>
<td>2. Other spine pathology</td>
</tr>
<tr>
<td>*Posterior Pelvic Pain Provocation test (PPPT)</td>
<td>3. CT verified ankylosis at baseline</td>
</tr>
<tr>
<td>*Active Straight Leg Raise (ASLR)</td>
<td>4. Body mass index over 30</td>
</tr>
<tr>
<td>*Palpation of the long dorsal SI-ligament</td>
<td></td>
</tr>
<tr>
<td>*Modified Trendelenburg test</td>
<td></td>
</tr>
<tr>
<td>*Palpation of the symphysis</td>
<td></td>
</tr>
<tr>
<td>3. High pain and disability score</td>
<td></td>
</tr>
<tr>
<td>*Oswestry Disability Index &gt;40 and/or</td>
<td></td>
</tr>
<tr>
<td>*Visual Analogue Scale &gt;50</td>
<td></td>
</tr>
<tr>
<td>4. The patients should have performed adequate physiotherapy over time without positive effect</td>
<td></td>
</tr>
</tbody>
</table>

*One could not perform the test.
At baseline, seven out of eight patients had bilateral SIJ symptoms. At the 1-year follow-up, only two patients experienced pain in the fused joint; however, six of the seven patients reported discomfort in the contralateral side. Seven patients had pain in the pubic symphysis before surgery, and five still had pain in this area at the 1-year follow-up (Figure 2).

The patients showed low health-related quality of life scores at baseline as compared to the general Norwegian population [19]. These patients also scored lower on the physical items of the SF-36 than on the items covering mental health. One year after surgery, there was a 20-point improvement in physical function and bodily pain \((p < 0.001)\), a 15-point improvement in social functioning...
and a 6-point improvement in general health ($p = 0.009$) (Table 3).

All patients reported that surgery had a positive effect; one patient reported a minor effect, two reported some effect, and five reported a good effect from the surgery. None of the patients reported an excellent result. Concerning tolerance of physical activity, seven patients reported some improvement, and one patient reported major improvement.

The fusion was evaluated with CT at the 1-year follow-up, and all patients had either solid fusion or significant bone bridging in the SIJ. However, it was difficult to evaluate the fusion in the pubic symphysis because of the plate artefacts, but no patient had plate or screw loosening or other signs of non-union.

There were 3 major complications: one infection, one complex regional pain syndrome with drop-foot and one loss of bladder sensation. In addition, there were 3 patients with transient sensitivity loss to the lateral femoral cutaneous nerve, a possible complication of bone harvesting from the iliac crest. All patients reported high post-operative pain and required epidural treatment for 5–7 days. They were hospitalised for 7–10 days and were discharged with opioids.

**Discussion**

The primary aims of this study were to evaluate changes in disability and pain intensity after SIJ fusion in patients with severe PGP. Pre-operatively, these patients showed severe disability and high pain levels. One-year post-operatively, clinically significant reductions in both disability and pain were observed. The SF-36 scores for physical function, bodily pain and social functioning also

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### Table 2 Patient characteristics pre-operatively

<table>
<thead>
<tr>
<th>Characteristic</th>
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<tbody>
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<td>Age (years)</td>
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<td>BMI (kg)</td>
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<td>(20–30)</td>
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<td>Children</td>
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<tr>
<td>Duration of symptoms (years)</td>
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<td>(2–25)</td>
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<tr>
<td>Disability pension</td>
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<tr>
<td>• 100%</td>
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<tr>
<td>Sick leave</td>
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<tr>
<td>• 100%</td>
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<td>• Graded</td>
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<td>Unilateral/Bilateral SIJ symptoms</td>
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<tr>
<td>Etiology</td>
<td></td>
<td></td>
</tr>
<tr>
<td>• Post pregnancy</td>
<td>6</td>
<td></td>
</tr>
<tr>
<td>• Trauma</td>
<td>2</td>
<td></td>
</tr>
<tr>
<td>Positive clinical test</td>
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<tr>
<td>1. Posterior pelvic provocation test</td>
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<tr>
<td>2. Active straight leg raise</td>
<td>8</td>
<td></td>
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<tr>
<td>3. Modified Trendelenburg</td>
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</tr>
<tr>
<td>4. Pain while palpation over the long dorsal ligament</td>
<td>8</td>
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<tr>
<td>5. Palpation of the symphysis</td>
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</tr>
</tbody>
</table>

*One could not perform the test.
Figure 4 (See legend on next page.)
Improved significantly. Seven out of eight patients reported a positive effect from the procedure.

Pelvic fusion in PGP is a rare procedure and is only performed in severe cases where conservative treatment modalities have been unsuccessful. A randomized controlled trial of this procedure is not possible, as the alternative treatment modality (conservative treatment) has already been tried. For this reason, an alternative study design was sought. The SSRD with multiple measurements is designed to study small samples of patients. A sample size of three patients is considered sufficient for external validity [15,16] and a study with 8 patients is scientifically generalizable. Patients with PGP have reported cyclic variations in symptoms, and as many as 72% report relapses during menstruation [17]. To capture these potential variations we repeatedly collected patients’ data for a 5-week period and discovered a large variation in the values during each phase. For some patients at baseline, a 40-point difference in ODI and a 43-point difference in VAS were observed during the 5-week period. One strength of the SSRD is its ability to uncover these individual variations, which is important for studying patients with PGP. Some of these variations may be corrected for in large group studies, but conclusions from small case series, with single measurements, should be interpreted with caution. A limitation of our study is the short follow-up period of 1 year. Although a 1-year follow-up period for clinical trials is commonly regarded as being too short, a recent study showed that the 1-year outcome after SIJ fusion was sustained 23 years later [14]. Despite the limitations of the SSRD, we believe that our study contributes valuable information on the effects of pelvic joint fusion.

Outcomes for SIJ fusion have been reported in several case series [9-14]. A positive effect of the surgery was observed in 50% to 90% of patients and this is in accordance with the positive effects seen in our 1-year outcomes. In a case series of nine patients, Al-Khayer et al. [9] observed decreases from 59 to 45 for mean ODI (p < 0.005) and from 8.1 to 4.6 for mean VAS (0–10) (p < 0.002). The same positive outcomes were reported by van Zwienen et al. [12], who found that 58 patients exhibited an increase in physical outcome from 37 to 61 (p < 0.001) as measured using the Majeed score (0-poor, 100-good) [21]. Although a mean improvement in physical function was observed in this study, 27% of patients reported a poor result with no effect from the surgery. Most of these patients had complications or non-union events, but some experienced no effect without any proper explanation. In our study, one patient did not experience any effect from the surgery. This patient had a more generalised pain pattern than the others (Figure 1) and it is possible that the SIJ was not the major source of pain in this case, although she had a positive response to the SIJ injection. In contrast the patients who reported a sharp and localised pain in the SIJ area did benefit from

Table 3 Mixed model for repeated measurements

<table>
<thead>
<tr>
<th></th>
<th>Pre-operative score constant value (95% CI)</th>
<th>Regression coefficient (95% CI)</th>
<th>Score at 1-year follow-up</th>
</tr>
</thead>
<tbody>
<tr>
<td>ODI</td>
<td>54.2 (48.4-59.9)</td>
<td>−5.7 (−7.6 to −3.8)</td>
<td>p &lt; 0.001</td>
</tr>
<tr>
<td>VAS in the morning</td>
<td>59.5 (45.6-73.5)</td>
<td>−4.8 (−6.0 to −3.7)</td>
<td>p = 0.019</td>
</tr>
<tr>
<td>VAS in the evening</td>
<td>81.7 (76.3-87.2)</td>
<td>−8.4 (−12.3 to −4.5)</td>
<td>p &lt; 0.001</td>
</tr>
<tr>
<td>SF-36</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Physical functioning</td>
<td>21.5 (8.8-34.3)</td>
<td>7.0 (3.2-10.8)</td>
<td>p &lt; 0.001</td>
</tr>
<tr>
<td>Role physical</td>
<td>2.8 (−6.7-12.3)</td>
<td>2.6 (−1.1-6.3)</td>
<td>p = 0.169</td>
</tr>
<tr>
<td>Bodily pain</td>
<td>13.1 (4.7-21.6)</td>
<td>6.8 (4.2-9.4)</td>
<td>p &lt; 0.001</td>
</tr>
<tr>
<td>General health</td>
<td>48.4 (34.9-61.9)</td>
<td>2.0 (0.5-3.5)</td>
<td>p = 0.009</td>
</tr>
<tr>
<td>Vitality</td>
<td>42.8 (33.3-52.4)</td>
<td>1.5 (−0.7-3.6)</td>
<td>p = 0.174</td>
</tr>
<tr>
<td>Sosial functioning</td>
<td>41.8 (22.4-61.2)</td>
<td>5.2 (1.3-9.0)</td>
<td>p = 0.008</td>
</tr>
<tr>
<td>Role emotional</td>
<td>55.1 (24.8-86.2)</td>
<td>2.6 (−1.7-7.0)</td>
<td>p = 0.240</td>
</tr>
<tr>
<td>Mental health</td>
<td>75.1 (64.1-86.1)</td>
<td>−1.0 (−2.7-0.7)</td>
<td>p = 0.224</td>
</tr>
</tbody>
</table>

ODI, VAS and individual items of SF-36 as dependent variables and time as independent variable. Regression line: constant + regression coefficient × time (time is defined as; 0 = baseline, 1 = 3 months, 2 = 6 months, 3 = 12 months). Score at 1-year follow-up = constant value at baseline + 3 times regression coefficient.
Figure 5 (See legend on next page.)
surgery. Hence a major challenge for clinicians is to identify patients who could possibly benefit from surgery. In our study, seven out of eight patients had a positive effect, indicating that our patient selection criteria were reasonably successful. However further studies must be conducted to identify the optimal criteria for the identification of patients to be offered surgical treatment.

Surgery is generally associated with a risk of complications. Because of the location of the SIJ an open approach to this structure is quite an aggressive surgical procedure. One of our patients developed a complex regional pain syndrome despite displays of normal neurological function in the first two post-operative days. This phenomenon has been found to occur after anterior SIJ fusion and is most likely due to nerve root compression [22]. When performing SIJ fusion the most serious and common complications are non-union, infection and nerve damage [10,12]. Van Zwienen et al. operated on 58 patients with bilateral SI screws and plating of the pubic symphysis and reported a 46.6% complication rate [12]. Patients experiencing complications report poorer outcomes than those without complications [10-13]. In our study, three patients experienced a major complication or adverse event but still reported satisfaction with the surgery because their SIJ pain had been relieved.

We fused the pubic symphysis for every patient based on experience that this procedure increases pelvic ring stability in patients operated for unstable pelvic ring fractures [23]. After 1 year five patients still had some pain in the symphysis. Due to artefacts, CT scans could not verify fusion in all cases. However, there were no indirect signs of non-union so it remains unclear why these patients reported persistent pain in the pubic area. Few studies have reported clinical results after symphysis plating in PGP patients [12,24], and it could be questioned whether fusion of the pubic symphysis is necessary in patients with PGP.

Non-specific PGP is thought to be a multi-factorial disorder with genetic, social, psychological, neuro-physiological and patho-anatomical factors involved in the pain syndrome [6]. SIJ fusion is used to treat these patients based on a biomechanical understanding of the disorder. Although it is difficult to evaluate whether the pain originates from the synovial joint or the surrounding ligaments, fusion will most likely, aside from stabilising the joint, also reduce the stress on the surrounding ligaments. Hence the positive results might be a consequence of greater SIJ stability. On the other hand, the placebo effect of surgery might also have had an impact on patient outcomes [25].

Because conservative treatment has proven effective for patients with PGP [26], this should be the first choice for therapy [6]. However some patients remain severely disabled with persistent pain despite appropriate conservative treatment [27]. Because of the possibility of complications, and a lack of randomized controlled trials, SIJ fusion needs to be further studied. Recently, new percutaneous devices for SIJ fusion have been introduced, and first reports [28-30] show a low complication rate together with a high fusion rate. These new techniques may reduce complications, however evidence of their efficacy has yet to be demonstrated.

Conclusion

One year after open unilateral anterior SIJ fusion combined with symphysis pubis fusion, positive and significant changes in both physical function and pain were observed. Despite these positive results, this procedure was associated with adverse events and complications.

Abbreviations

SIJ: Sacroiliac joint; PGP: Pelvic girdle pain; ODI: Oswestry disability index; VAS: Visual analogue scale; SSRI: Single subject research design.

Competing interests

This study was supported by grants from the Norwegian Foundation for Health and Rehabilitation and Sophies Minde Ortopedi AS. The authors declare that they have no competing interest.

Authors’ contributions

All authors have been involved in the planning of the study, data collection, data analysis and the writing. All authors read and approved the final manuscript.

Acknowledgements

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Pelvic joint fusions in patients with chronic pelvic girdle pain: a 23-year follow-up

Thomas J. Kibsgård · Olav Røise · Einar Sudmann · Britt Stuge

Abstract
Purpose Fusion of the sacroiliac joints (SIJ) has been a treatment option for patients with severe pelvic girdle pain (PGP). The primary aims were to evaluate the long-term outcomes in patients who underwent SIJ fusion and to compare 1-year outcomes with long-term outcomes. The secondary aim was to compare patients who underwent SIJ fusion with a comparable group who did not.

Methods This study includes fifty patients that underwent SIJ fusion between 1977 and 1998. Function (the Oswestry disability index; ODI), pain intensity (visual analogue scale; VAS) and health-related quality of life (SF-36) were determined according to a patient-reported questionnaire. The questionnaire scores were compared with previously recorded 1-year outcomes and with questionnaire scores from a group of 28 patients who did not undergo SIJ fusion.

Results The patients who underwent SIJ fusion reported a mean ODI of 33 (95 % CI 24–42) and a mean VAS score of 54 (95 % CI 46–63) 23 years (range 19–34) after surgery. Regarding quality of life, the patients reported reduced physical function, but mental health was not affected in the same manner. The patients with successful 1-year outcomes (48 %) retained significantly improved function and reduced pain levels compared with the subgroup of patients with unsuccessful 1-year outcomes (28 %). The patients who underwent surgery did not differ from the non-surgery group in any outcome at the long-term follow-up.

Conclusions Patients treated with SIJ fusion had moderate disability and pain 23 years after surgery, and the 1-year outcomes were sustained 23 years after surgery. Although many fused patients reported good outcome, this group did not differ from the comparable non-surgical group.

Keywords Sacroiliac joint · Pelvic girdle pain · Long-term follow-up · Fusion · Arthrodesis

Introduction

The sacroiliac joint (SIJ) is a source of pain for 13–30 % of patients with lower back pain [17] and can become painful following inflammatory diseases, metabolic disorders, post-traumatic arthritis, malalignment and infections [2]. The origin and diagnosis of pelvic girdle pain (PGP) are controversial when radiological findings are absent and objective diagnostic tests lack proper evidence. The cause of PGP is unclear, but it may be a biomechanical disorder in which SIJ hypermobility causes pain [5]. The theory of abnormal joint mobility is one reason some orthopaedic surgeons have fused pelvic joints to reduce PGP [7]. There is, however, no evidence of a correlation between sacroiliac instability and symptoms [14, 15, 19], whereas a correlation between asymmetrical movement in the SIJ and symptom intensity has been described [4].
Conservative treatment is the first choice for patients with PGP and has proven efficient [13]. Despite appropriate conservative treatments, some patients remain severely disabled by persistent pain; therefore, pelvic joint fusion has been performed on those patients. Pelvic joint fusions have been reported in a number of studies, but there has been no consistency in the inclusion criteria used [1, 3, 10, 16, 18]. The results of pelvic joint fusions are mostly reported in small case series, with the exception of one report that included 58 patients [16]. The reported short-term results have been mainly positive [1, 3, 16, 18], but poor results have also been reported [10]. To our knowledge, the longest follow-up reported in the literature is 5.8 years [3].

The main aims of this study were to evaluate long-term functioning, pain and health-related quality of life in patients who had previously undergone pelvic joint fusion surgery and to compare the 1-year outcomes with the long-term results. The secondary aim was to compare patients with PGP, who underwent surgery with a comparable group of patients who did not undergo surgery.

Materials and methods

Design and subjects

This cross-sectional study included patients with SIJ pain that had been operated at the Hagavik Orthopaedic Hospital between 1977 and 1998 (Fig. 1). During 2009 the patients received a questionnaire including self-report of pain and function. The material consists of data from 50 subjects 1 year after SIJ fusion and long-term follow-up. The regional committee of ethics approved the study (project number: 1.2006.1574).

Indications for surgery

The patients were selected and operated on primarily by one of the authors (E. Sudmann). The criteria for surgery were based on patient history and radiological and clinical examinations. Inclusion criteria were pain in the SIJ > 1 year after pregnancy or after trauma, pain with an idiopathic origin, severe disability and resistance to conservative treatment. The clinical tests performed included tenderness at the superior and inferior posterior iliac spines, active and passive straight leg raise, Patrick Fabere’s test, passive hip rotation, forcible inward rotation and extension of the hip joint. Further tests included normal neurological and gynaecological examinations, normal spinal X-rays, symphysis movement of less than 3 mm on plain radiographs during a one-leg stance, normal radiculography, negative rheumatology tests, and negative blood tests.

Fig. 1 Flow chart of the study population

Before the surgery only 5 (10 %) patients had the ability to work and 23 (46 %) had to use some kind of walking aids.

Surgical technique and post-operative management

The patients were operated on using a dorsal approach, with either a trans-iliac fusion or an intra/articular fusion between the ilium and the sacrum. When the trans-iliac fusion was performed, an iliac window was constructed to access the joint [12]. The joint surface was cleared of cartilage and decorticated. The cortical iliac window was used as a graft and was usually hammered into the sacrum to promote intra-articular bone formation and conduction. Additional cancellous bone was impacted around the cortical graft. In a dorsal intra/articular fusion, iliac crest autografts were added after joint removal and bone decortications [18]. The pubic symphysis was fused in four patients using an open technique with an iliac crest block bone autograft and plating. In some patients a Hoffman frame was tried to achieve post-operative fixation, but in most cases the patients were confined to bedrest, usually for 6 weeks.

One-year outcomes and the classification of subgroups

The patients were examined with CT after 1 year, and the clinical outcome was graded according to the following criteria: a joint with negative SIJ tests and no or minor pain that did not interfere with the patients’ work was graded “good”. A joint with obvious improvement in comparison to the pre-operative status and little pain, but pain that interfered with work (professional or at home) was graded...
“fair”. A joint was graded “poor” if there was no relief from pain or if the joint deteriorated after surgery. In cases of bilateral surgery, each of the patient’s joints could receive a different grade. According to the grading of the joints 1 year after surgery, the patients were allocated to three different subgroups (Fig. 2). Twenty-four patients (48 %) had all joints classified as “good” and were assigned to the subgroup “successful”. Fourteen patients (28 %) had at least one joint classified as “poor” and were assigned to the subgroup “unsuccessful”. Twelve patients (24 %) had their worse joint scored as “fair” and represented the subgroup “partly successful”. These three subgroups became the baseline for comparing the long-term effects. The patients’ 1-year outcomes were consecutively and systematically registered in a DOS Advanced relational database.

Non-surgery group

On the basis of their experiences with surgery failures, surgeons became increasingly reluctant to perform SIJ fusion. As a result, surgery was declined for a number of patients in the 1990s. These 28 patients constitute the non-surgery group. According to the patient charts 5 (17 %) patients had the ability to work at the time they were diagnosed and 17 (61 %) had to use some sort of walking aid. There were no differences in working ability \((p = 0.32)\) and the use of walking aids \((p = 0.21)\) between the surgical group and the non-surgical group at the time they were diagnosed. Beside these parameters, no other measurements of physical function or pain were available at that time.

Statistical analyses

The means were adjusted for BMI, age and time of follow-up (least square mean) and are presented with a 95 % confidence interval. The differences in the mean ODI, VAS and SF-36 scores between the three subgroups and between the patients who underwent surgery and those who did not were tested using analysis of covariance, adjusting for BMI, age and time of follow-up. Pair wise comparisons were performed using \(t\) tests with Bonferroni’s adjustment. The correlations between the 1-year outcome and ODI/VAS were expressed by Spearman’s \(\rho\). A \(p\) value of 0.05 was regarded as significant. Statistical analyses were performed using SPSS Version 18.

Results

Demographics and patient characteristics

Eighty-one consecutive patients were selected for this study. Nine of the 81 patients were dead at inclusion, and two patients were excluded because of insufficient baseline data. Of the remaining 70 patients, 50 (71 %) responded to the questionnaire. Among the 20 non-responders, 8 did not answer because of serious illness, 10 refused to participate and 2 had an unknown address (Fig. 1). The patient demographics at the long-term follow-up are presented in Table 1. The non-responders and the excluded patients did not differ from the included patients with regards to age, follow-up or short-term outcomes.

Of the 50 patients, 21 patients had a unilateral SIJ fusion, 25 patients had a bilateral fusion and 4 patients had a bilateral fusion + pubic symphysiodesis. The patients were allocated to three different subgroups (Fig. 2). Twenty-four patients (48 %) had all joints classified as “good” and were assigned to the subgroup “successful”. Fourteen patients (28 %) had at least one joint classified as “poor” and were assigned to the subgroup “unsuccessful”. Twelve patients (24 %) had their worse joint scored as “fair” and represented the subgroup “partly successful”. These three subgroups became the baseline for comparing the long-term effects. The patients’ 1-year outcomes were consecutively and systematically registered in a DOS Advanced relational database.

Outcome measures

All patients from the database were contacted and asked to complete and return a questionnaire. The questionnaire included validated measures for function (the Oswestry disability index) [6], pain intensity at worst, morning and evening was measured by 100 mm visual analogue scale (VAS; 0=no pain, 100=worst possible pain) and health-related quality of life (SF-36) [20]. The Oswestry disability index (ODI) is a 10-item questionnaire that assesses patient function (0 indicates the best, 100 indicates the worst). A 10-point difference represents a significant clinical change [6, 8]. The SF-36 questionnaire contains 36 items representing 8 subscales: physical functioning, role physical, bodily pain, general health, vitality, social functioning, role emotional and mental health. Additionally, if the patients experienced any effect from the operation, they graded the effect as excellent, good, some or minor.
received fusion of both the SIJ and the pubic symphysis. In total, 83 joints were surgically fused; only 7 joints had an inferior result that required a second operation, including 4 with a CT-verified pseudarthrosis. In total, there were eight joints (10 %) with a CT-verified pseudarthrosis. The patients with pseudarthrosis had a 29 % greater risk of a poor result compared with those with a solid fusion (8 %). Pseudarthrosis caused a 3.6-fold increased risk of chronic painful joints compared with a fused joint ($p = 0.009$).

There were few complications: one patient developed icterus of unknown aetiology, one developed a pulmonary embolism and one contracted a pin tract infection after the use of a Hoffman frame. In addition, unrelated to the fusion surgery, one patient required surgery because of immediate post-operative acute appendicitis, and one required surgery for a small bowel obstruction.

**Long-term follow-up**

The patients who underwent surgery had a mean ODI score of 33 (95 % CI 24–42; Table 2). Twenty-eight percent of the patients scored less than 20 on the ODI; 69 % had a score below 40. The patients had a mean evening VAS score of 54 (95 % CI 46–63; Table 2). Twenty percent of the patients had a VAS score under 20, and 53 % had a VAS score higher than 60. The SF-36 score indicated that the patients had impaired physical health, as represented by the four subscales for physical functioning, role physical, bodily pain and general health (Fig. 3). Patient mental health was not affected in a similar manner, but the patients presented lower scores than a representative sample from the general Norwegian population [9] (Fig. 3).

The three subgroups of patients did not differ significantly in terms of age, follow-up, BMI or level of education (Table 3). The subgroup with a “successful” 1-year result (48 %) reported significantly lower scores on the ODI and VAS compared with the subgroup with an “unsuccessful” 1-year result (28 %; Table 2). The observed 16-point (95 % CI 1–32) difference in mean ODI was regarded as both clinically and statistically significant ($p = 0.034$; Table 2). The difference in VAS score was 24 (95 % CI 2–47) in the morning ($p = 0.029$) and 28 (95 % CI 6–51) in the evening ($p = 0.011$). There was a positive correlation between the 1-year outcome and three different long-term outcomes; VAS in the morning ($q = 0.34$, $p = 0.001$), VAS in the evening ($q = 0.42$, $p = 0.013$) and ODI ($q = 0.43$, $p = 0.002$). For the SF-36 subscales, there were no significant differences between the subgroups, except for the physical functioning subscale, on which the subgroup with a “successful” short-term result had a 24-point higher score than the “unsuccessful” group ($p = 0.040$).

At follow-up, 65 % of the patients reported that the surgery had a positive effect, and 74 % of these reported a good or excellent result. Eighteen percent of patients reported no effect, and the remaining patients were uncertain. Among the patients with a “successful” 1-year outcome, 80 % reported experiencing a good or excellent effect. Despite these results, 54 % of the patients who underwent surgery

<table>
<thead>
<tr>
<th>Table 1 Characteristics of the participants in the long-term follow-up study</th>
</tr>
</thead>
<tbody>
<tr>
<td>Surgery group (n = 50)</td>
</tr>
<tr>
<td>Age (years) 58 (56–61)</td>
</tr>
<tr>
<td>Follow-up (years) 23 (22–24)</td>
</tr>
<tr>
<td>Education (years) 12 (11–13)</td>
</tr>
<tr>
<td>Male/female 3/47</td>
</tr>
<tr>
<td>Body mass index (BMI) 27 (26–28)</td>
</tr>
<tr>
<td>Duration of SIJ pain (years) 5 (range 1–21)</td>
</tr>
<tr>
<td>Aetiology</td>
</tr>
<tr>
<td>Pregnancy</td>
</tr>
<tr>
<td>Trauma</td>
</tr>
<tr>
<td>Idiopathic</td>
</tr>
<tr>
<td>Disability pension received at long-term follow-up</td>
</tr>
<tr>
<td>0 %</td>
</tr>
<tr>
<td>50–99 %</td>
</tr>
<tr>
<td>100 %</td>
</tr>
<tr>
<td>Pain medication use at long-term follow-up</td>
</tr>
<tr>
<td>None</td>
</tr>
<tr>
<td>Seldom</td>
</tr>
<tr>
<td>1–6 days/week</td>
</tr>
<tr>
<td>Daily</td>
</tr>
</tbody>
</table>

* Chi-squared test
received a 100% disability pension, and 46% used some sort of pain medication daily (Table 1).

There were no significant differences in ODI ($p = 0.54$), morning VAS ($p = 0.54$), evening VAS scores ($p = 0.50$) or SF-36 scores between the group that underwent surgery and the non-surgery group at the long term-follow-up (Table 2). The non-surgery comparison group was in average 6 years younger ($p = 0.003$) and had a 6-year shorter follow-up time ($p < 0.001$) than the patients who underwent surgery. Aside from these differences, there were no significant differences between these two groups in terms of BMI, aetiology or education level (Table 1).

### Discussion

The patients who underwent pelvic joint fusion reported having a moderate disability, with moderate to severe pain 23 years after surgery. They had impaired physical function, but their mental health was not reduced in a similar manner. The 1-year outcomes appeared to predict long-term outcomes with regard to pain and disability. The patients who underwent surgery did not differ from a non-matched comparison group of PGP patients who did not undergo surgery.

There are several strengths and limitations to this study. Its strengths include the long follow-up time and high response rate. To our knowledge, no other studies in the literature have a 23-year follow-up. Fifty out of 70 eligible patients were included, and the drop-outs did not differ significantly from the included patients. Moreover, most patients were selected and operated on by one surgeon, and the 1-year outcomes were consecutively and systematically registered. Another strength is the use of multiple clinical tests for inclusion [17] and validated questionnaires for the follow-up. However, a limitation is the subgroup

<table>
<thead>
<tr>
<th>Table 2: Scores for ODI (0 indicates no disability, 100 indicates a high degree of disability), visual analogue scale (VAS) (0 indicates no pain, 100 indicates the worst pain) and SF-36 (0–100; 100 indicates the best health-related quality of life)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Scores in the three subgroups with different short-term outcomes</td>
</tr>
<tr>
<td>Patients with SIJ fusion (n=50)</td>
</tr>
<tr>
<td>Oswestry disability index (ODI)</td>
</tr>
<tr>
<td>Morning VAS score</td>
</tr>
<tr>
<td>Evening VAS score</td>
</tr>
<tr>
<td>SF-36</td>
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<tr>
<td>Role physical</td>
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<td>Vitality</td>
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<td>Social functioning</td>
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<td>Role emotional</td>
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<tr>
<td>Mental health</td>
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</tbody>
</table>
The primary aim of this study was to evaluate long-term patient outcomes after SIJ fusion. The patients reported being moderately disabled, with 70% of their scores below the limit of severe disability [6]. One-third of the patients reported no or minimal disability, with ODI scores below 20. Half of the patients had chronic pain and used pain medication daily. Their reported pain intensity was quite high and was worse in the evening. As many as 70% of the patients received some sort of disability pension. Despite these moderate results, 65% of the patients reported that surgery had a positive effect, mostly good or excellent. It is known that patients tend to report a high grade of satisfaction, even when the clinical outcome is moderate [11]. Because few studies have examined the long-term outcomes after surgery, comparison with other studies is difficult. The longest follow-up study that we found was 5.8 years [3]. That study reported a positive effect of surgery in terms of satisfaction and health-related quality of life, with SF-36 scores that were similar to those reported in our study.

Few studies used more than one post-operative measurement [10, 16]. Consequently, the long-term outcomes after SIJ fusion are difficult to predict. Interestingly, we found that the 1-year outcomes predicted the long-term outcomes, even 23 years after surgery. The subgroup of patients with successful 1-year outcomes reported significantly reduced disability and pain levels when compared with the subgroup with an unsuccessful outcome. van Zwienen et al. [16] reported that surgery had a positive effect on functional outcomes after 1 year, with further improvement after 2 years. Most of the patients in our study reported to be relieved (48%) or almost relieved (24%) from pain at the 1-year point, but 28% were not; this outcome distribution was relatively constant. The 1-year outcomes of our study were similar to those of other studies [1, 16, 18] reporting a positive effect from surgery; however, a fraction of our patients reported that the surgery had no effect. The effect of SIJ fusion for patients with PGP, has however to be examined in future studies with proper designs.

Pelvic joint fusion is a controversial surgical procedure. The scientific evidence supporting the use of this surgery is weak, as only case series are available [17]. In our study, the patients who underwent surgery showed no differences in the outcome measurements when compared with the non-surgery patients. As surgery appears to have provided positive and long-lasting results in some patients, future research should address the question of who will benefit from this surgery. However, a poor understanding of the pathogenesis and aetiology of PGP persists, and there is no consensus on which clinical and radiological tests should be used to select patients for surgery. It is hoped that improved insights into the pathogenesis of PGP and improved diagnostic tools will allow the correct patients to be selected for surgery, resulting in improved outcomes.

**Conclusions**

Patients with chronic PGP who underwent SIJ fusion reported being moderately disabled, with moderate or
severe pain intensity 23 years after surgery, and the 1-year outcomes were sustained 23 years after surgery. Two-thirds of the patients experienced a positive long-term effect from the fusion surgery, and 20% reported no effect from the surgery. Although some patients report to have a successful long-term outcome, the group of patients who underwent SIJ fusion did not differ from the group of patients that were not operated, in regard of physical function, pain and HRQoL. These two groups of patients were not matched at baseline, hence further studies are needed to address the efficacy of surgery for patients with PGP.

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Conflict of interest None.

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