Hip pain in general practice

Exploration and classification

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Hip pain in general practice
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Heupklachten in de huisartspraktijk
Exploratie en classificatie

Proefschrift

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Gean no fierder, en komst ea yn 'e knipe,
tink dan oan dy lêste sneontemoarn,
Maitiid, sinne, blommen, 't wie dochs skoan.
Henk Zeinstra, 1980

voor Mem
voor Menno

Contents

INTRODUCTION

Musculoskeletal diseases are the second most costly group of disorders in primary care in the Netherlands.[1] Of the musculoskeletal disorders, hip problems occur most frequently in the aged.[2] Therefore, in the coming decades, prevalence of hip disorders, of which an important part is attributed to osteoarthritis of the hip,[3] is expected to increase due to ageing of the population. The report 'Public Health Status and Forecasts 1997' predicts that by the year 2015 the incidence of osteoarthritis will have increased 36% compared to the current incidence.[4]

Besides generating high costs, hip disorders have a major impact on the life of individuals, not only due to the presence of pain, but also because of the adverse effects on the mobility and daily activities of the elderly[2]. A recent Dutch study in the open population aged 55 years and over and living independently revealed that 16.6% of the women and 8.3% of the men reported hip pain.[5] In the Netherlands, the general practitioner is the first physician who is consulted and therefore plays an important role in the management of these disorders. Optimal management, however, requires accurate diagnosis. The Dutch College of General Practitioners has published national guidelines for the diagnosis, treatment and referral of many disorders in general practice.[6] For hip disorders, however, no such guidelines are yet available. International (ICPC) codes for several hip disorders in general practice have been introduced[7], but these lack clear guidelines as to what clinical criteria constitute these conditions. The possibility in the ICPC to code a hip problem as 'unspecified' is eagerly accepted by general practitioners; at least one third of the patients with hip complaints receives no specific diagnosis.[2]

As in other musculoskeletal disorders[8-10] it is plausible that a valid classification system for hip disorders in adult patients in primary care is lacking. In primary care, most musculoskeletal conditions are identified as clinical syndromes. A syndrome is a combination of symptoms that frequently appear together, attributed to a specific underlying condition. The diagnosis rests on a total evaluation of the clinical picture and a decision has to be made whether the patient sufficiently resembles this to allow such a diagnosis to be made. Clinical syndromes in general, syndromes without a pathognomonic sign, cause the greatest problems with respect to classification of diseases.[11]

However, as long as no generally accepted definition of clinical syndromes (diagnostic criteria) exists, no collective experience of clinical course and treatment results in these syndromes will be gained, and no scientific studies (e.g. randomised trials) will be reproducible and generalisable. Organised collective experience and scientific studies on hip syndromes in primary care are essential to give patients with hip problems an optimal treatment.

The main research question in this thesis is to identify valid diagnostic and/or classification criteria that may be applied to hip disorders in general practice.

Chapter 1 of this work gives an overview of current diagnostic criteria for hip disorders in the middle-aged and elderly, which were assessed based on a review of the literature. Subsequently, *chapter 2* presents our analysis of the actual management of middle-aged and elderly hip patients by 20 general practitioners in the Rotterdam region. One of the specific problems identified in this latter study, as well as in the literature study, is the diagnosis of osteoarthritis of the hip. Therefore, *chapter 3* presents the results of a study that investigated in general practice the validity of available diagnostic criteria for osteoarthritis of the hip. One of the major criteria for classifying the type and severity of a condition seems to be the restriction of motion. This led to a study designed to compare different methods of measuring joint motion in order to obtain the most reliable outcomes; the results of this study are described in *chapter 4*.

Based on the first three studies we concluded that no adequate classification system for hip disorders is available in general practice. Therefore, we performed a study with the aim to develop a new classification system based on cluster analysis and then validated in several ways. The results of this study are presented in *chapter 5*. The feasibility of the use of such a classification system in general practice is addressed in *chapter 6*.

Radiographic examination of the hip joint is a well known additional investigation in primary care, but the additional value of a sonographic examination in adults with hip disorders in primary care is still unknown. *Chapter 7* addresses the diagnostic value of sonographic examination of the hip joint in adults with hip problems in general practice.

Patients with the disorder 'meralgia paresthetica', identified in the new classification, showed a previously unknown relationship with radiographic abnormalities of the pubic symphysis. *Chapter 8* presents the results of a study designed to confirm this relationship in another study population.

The methodological problems in classifying diseases are addressed in *chapter 9* and suggestions for future research are made.

The studies described in chapters 3, and 5-7, made use of the same study population. This will cause some overlap of the method sections of these chapters, but also allows them to be read as separate chapters.

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1

DIAGNOSTIC CRITERIA FOR HIP DISORDERS IN ADULTS: A REVIEW OF LITERATURE

Introduction

Hip complaints in the middle-aged and elderly have a high prevalence and are generally attributed to osteoarthritis[1]; however, under-diagnosing of disorders of soft tissue in this group of patients is frequently reported.[2-6] Although osteoarthritis of the hip is often the primary diagnosis of the general practitioner concerning middle-aged and elderly patients with hip complaints, other treatable causes of hip pain need to be considered. Even when the X-ray shows osteoarthritic changes, these may not necessarily explain the patient's complaints. From population screening it is known that only a minority of the patients with moderate osteoarthritic changes on X-ray of the hip joint, show joint-specific pain or decreased joint mobility.[7,8] Additionally, only a minority of middle-aged and elderly persons with pain in the hip display such degenerative changes on X-ray.[8]

This review of literature presents a concise survey of hip disorders causing chronic pain and disability in middle-aged and elderly patients, as well as a description of the symptoms and available diagnostic criteria of these disorders with the aim to support general practitioners in their diagnosis.

Methods

A Medline search was conducted on the abstract word 'hip' in combination with 'diagnosis' or 'diagnostic criteria'. The same procedure was repeated for the index-category 'hip osteoarthritis'. Finally, we searched on 'differential diagnosis' in combination with the abstract word 'hip'. Abstracts were screened and selected when the article was written in English, when they addressed chronic hip disorders in adults, and when symptoms or diagnostic criteria were described. From references in selected articles and from a Medline search on the names of the disorders mentioned in these articles, additional publications could be traced.

All articles were screened to ascertain the aim of the study, the study design, number of patients, population from which the patients were derived, inclusion criteria, and the symptoms described.

The hip disorders found are grouped according to soft tissue disorders, disorders of bone and cartilage, and differential diagnosis.

Table 1
Studies on symptoms in trochanteric bursitis

	Aim	Cases/ controls	Population	pros-pective	Selection criteria	Symptoms
Collee[12] 1991	- prevalence trochanteric bursitis in low back pain - symptoms	50/154	Patients with low back pain in general practice, occupational health service, rheumatologic outpatient clinic	Yes	Criteria of Little	- pain in trochanter region and lateral leg - paresthesia in the leg - tenderness of iliotibial tract - pain ↑ with walking, standing, lying on affected side, crossing legs.
Schapira[13] 1986	- comorbidity - treatment - symptoms	72/0	Patients in rheumatologic outpatient clinic	Yes	Criteria of Little	- pain in lateral upper thigh radiating to the knee - pain ↑ with lying on affected side, walking, skatting, climbing stairs
Karpinsky[14] 1985	- symptoms - treatment	15/0	Patients in orthopaedic clinic	No	Referred to the clinic for trochanteric pain	- pain and tenderness on top of trochanter - painfull resisted abduction - pain ↑ with activities (especially walking)
Rasmussen[1] 1985	- treatment - symptoms	36/0	Patients in physical and rheumatologic clinic	No	Criteria of Andersson	- pain with external rotation combined with abduction
Swezey[17] 1976	- prevalence trochanteric bursitis in low back pain - symptoms - treatment	31/39	Patients from a geriatric home, referred for low back pain	No	Tenderness of greater trochanter	- pain upper thigh and buttock - limited internal rotation - pain ↑ with lying on affected side

Gordon[9] 1961	- symptoms - comorbidity - treatment	61/0	Patients in private practice for orthopaedic surgery	No	Not defined	- tenderness of trochanter - pain in lateral thigh radiating to knee - pain with external rotation in combination with abduction - painfull resisted abduction
Andersson[18] 1958	- symptoms - morbidity	45/0	Patients in physiatric and rehabilitation clinic	No	Not defined	- tenderness of trochanter (often post.aspect) - pain on extremes of internal rotation and/or abduction - painfull resisted abduction
Leonard[19] 1958	- symptoms	18/0	Patients of orthopaedic surgeons	No	Not defined	* - pain on lateral aspect of the hip - gluteal limp - local tenderness - passive internal rotation painful - active abduction painful
Spear[10] 1952	- symptoms in bursitis - symptoms in peritendinitis calcarea	40/0 24/0	Patients in unspecified clinic	No	Not defined for bursitis, roentgenographic calcification for peritendinitis calcarea	- dull aching pain in trochanter region - painfull active abduction and passive adduction - pain ↑ with climbing stairs, walking, standing
Goldenberg[2] 1936	- prevalence supratrochanteric calcification - frequency of location	30/520	Patients in hospital for joint diseases for whom roentgenograms of the hip joints were available	No	Calcified deposits in the region of the greater trochantor seen on roentgenograms	- limitation of abduction and internal rotation

* impossible to detect if the symptoms described derive from the case series or from literature

Results

Disorders of the soft tissues

Bursitis

Inflammation of bursae in the hip region is reported regularly. Inflammations similar to that in the synovial joint (such as rheumatoid arthritis, gout, and infections) may occur in bursae, but usually the inflammation is of the non-specific type. Injuries, including repetitive micro-trauma, and excessive pressure are considered as causal factors.[9]

Trochanteric bursitis

Trochanteric bursitis is a frequently reported disorder of the hip; Larsson[4] refers to it as a frequently overlooked disorder in the hip. The trochanteric bursa consists of three separate parts. The most extensive part is situated between the tendon of the gluteus maximus muscle and the insertion of the gluteus medius muscle. The second part lies between the greater trochanter and the gluteus medius muscle. The third part (not always present) lies between the greater trochanter and the gluteus minimus muscle.[10,11]

Studies concerning the symptoms in trochanteric bursitis are presented in Table 1. In some studies, specific criteria were used to define subjects as cases. Two studies used the criteria of Little: by palpating the lateral aspect of the thigh with the patient lying on the side with the painful side uppermost, the greater trochanter is markedly tender and other pelvic bony prominences are not. Little[20] described this palpation technique in 1979; however, because they are not based on his own series it is unclear how he derived these criteria. Rasmussen[15] used the criteria of Andersson[17] who did not present these as diagnostic criteria in his original article but only gave the percentage of his cases in which the different symptoms occurred. In none of these studies the diagnoses of bursitis was confirmed by visualisation techniques or histopathological examination. There are some recent case reports about MRI and/or scan findings and/or sonographic findings in trochanteric bursitis, but they present only one case each.[11,21,22]

Several authors[12,14,18] prefer the name trochanteric pain syndrome because tendons as well as bursae may be involved. Calcifications in or about the trochanteric bursa are often reported and are said to occur in about 40-50% of the cases.[4,9,11,23] Spear[10] described the calcifications in the surrounding tendons as separate diagnoses, although he found similar symptoms in 24 patients with radiological signs of peritendinous calcarea of the trochanteric region as well as in 40 patients with trochanteric bursitis. In the 13 patients with peritendinous calcification of the tendon of the gluteus medius the pain was more severe.

Ischiogluteal bursitis

The ischial bursa is situated between the ischial tuber and the gluteus maximus muscle. Although this disorder, also known as 'weaver's bottom', is mentioned in many reviews about bursitis[4,23,24], only one original articles on this disorder could be traced. Swartout and Compere[25] described 19 patients with this syndrome over a period of 20 months, after one of the authors had developed the condition himself. They do not give any inclusion criteria for the cases. In the description of clinical characteristics of these patients they do not give percentages but only general statements. They mention a dominating pain in the buttock which is greatly aggravated by sitting, an extreme tenderness on the ischiogluteal bursa, painful abduction in combination with flexion and external rotation, and a positive Lasègue sign.

Iliopectinal bursitis

The iliopectinal bursa is situated at the front of the hip, concealed by the iliopsoas muscle. A communication exists between the bursa and the cavity of the hip joint in about 15% of all cases.[26] Although a large swelling is not necessarily present in bursitis, most reports on iliopectinal bursitis considered the condition to result from an increase in bursal volume. The direction and the degree of bursal enlargement determine the mode of clinical presentation. Recent application of diagnostic imaging studies has led to an increased awareness of iliopectinal bursitis, and the diagnosis of this syndrome may become more common.

Studies concerning the signs and symptoms in this syndrome, based on more than one case are presented in Table 2. All studies were case reports that generally described a small number of cases. In several studies the majority of the cases had an underlying hip disease, such as rheumatoid arthritis or osteoarthritis, which also cause clinical symptoms of their own.

Disorders of the fibrous capsule and ligaments

We traced three articles dealing with a primary capsular constriction without a reasonable explanation. Lequesne[37] called this syndrome idiopathic capsular constriction of the hip. Another name for this syndrome is the 'frozen hip'[38,39] which suggests similarity with a syndrome in the upper extremities; i.e. the 'frozen shoulder'. The authors describe the capsular constriction as self-limiting after several months. All three studies were descriptive studies of cases from rheumatology clinics without control patients, with 13, 3, and 1 case(s), respectively. In the study by Lequesne[37]

Table 2
Studies on symptoms in iliopectinal bursitis

	Aim	Cases/ controls	Population	pros-pective	Selection criteria	Symptoms
Fortin[27] 1995	- diagnostic criteria	4 case reports	Physiatric clinic	No	Inguinal pain Hip disease absent	- inguinal pain - pain on passive hip movement (especially flexion/internal rotation) - no pain upon resisted hip movements - significant clinical response to bursography with steroid injection
Meany[28] 1992	- role of imaging comparing the various imaging modalities	14/0	3 radiodiagnostic institutions	No	Iliopectinal bursal enlargement	- pain and/or groin mass and/or lower limb oedema - underlying hip disease - hip effusion on ultrasound
Toohey[29] 1990	- description of 4 cases - review - differential diagnosis - comorbidity	4 case reports	Unspecified	No	Iliopectinal bursitis Demonstrated by CT or MRI	- anterior hip pain or inguinal mass - history of trauma or underlying hip disease - tenderness beneath the midpoint of the inguinal ligament - hip effusions
Underwood[30] 1988	- description of varied presentations of iliopsoas bursitis - role of CT	8/0	Clinic for rheumatologic and internal medicine	No	Iliopectinal bursitis Demonstrated by CT	- underlying hip disease - hip pain and/or groin mass and/or unilateral leg swelling - retroperitoneal extension (pelvic mass) is also possible
Binek[31] 1987	- description of 3 cases	3 case reports	Radiodiagnostic clinic		Iliopectinal bursal enlargement demonstrated by CT and arthrography	- underlying hip disease - often connection of bursa with joint space - femoral vessel displacement (CT)

Sartoris[32] 1985	- description of 9 cases - develop imaging strategy	9 case reports	Unspecified	No	Radiologically demonstrated iliopectinal cyst	- palpable groin mass (+ underlying hip disease) - pain in hip region with anterior radiation to the knee - shortening of the stride - point tenderness inferior to the inguinal ligament
Armstrong[33] 1972	- description of 2 cases of interest for radiologists	2 case reports	Radiodiagnostic clinic	No	accidently entered bursa during femoral artery or vein puncture	- femoral vein obstruction
Melamed[34] 1967	- description of 3 cases	3 case reports	Radiodiagnostic clinic	No	retroperitoneal extension due to synovitis of the hip via the iliopsoas bursa	- palpable mass - extrinsic pressure on adjacent structures - radiological advanced degenerative or destructive arthritis
Huchersson[3] 1946	- description of 2 cases - review	2 case reports	Unspecified	No	not defined	- pain in the hip region - tenderness just below the inguinal ligament/ lateral to femoral pulse - pain elicted by extension, abduction, internal rotation
O'Connor[36] 1933	- diagnostic criteria for early recognition of iliopectinal bursitis	33/0	Clinic for orthopedic surgery	Unknown	not defined	- pain anterior aspect of the hip joint, frequently radiating to anterior knee - ↑ pain by activity, - atrophy of the psoas muscle, - limitation of hyperextension - tenderness just below inguinal ligament and lateral to femoral pulse - extreme abduction/ internal rotation causes pain at site of the bursa

the intracapsular capacity was measured and was found to be decreased compared with normal values. Luukainen[38] and Chard[39] both reported an increased uptake on an isotope bone scan around the hip joint in this syndrome. The symptoms described in this syndrome were persisting pain during several months with slightly[37] to moderate[38,39] limitation of hip movement (rotations and flexion).

Meralgia Paresthetica Syndrome

The syndrome called meralgia paresthetica describes an entrapment of the cutaneous lateral nerve of the femur in, or under the inguinal ligament. Probable causes, although not confirmed scientifically, include tight trousers or corsets[40], and adipositas or a changed gait pattern.[6,41] This syndrome is also frequently noticed after bone graft harvesting. We traced four articles describing symptoms in case series ranging from 3 to 150 cases. From the two largest studies (74[42] and 150[43] cases) it could be determined that the studies were retrospective and that the patients were derived from a neurology clinic. All case series described a dull ache and numbness of the anterio-lateral aspect of the thigh as the most common symptom in the cases. Accentuation of symptoms when standing and walking (hyperextension) was described in three case series[43-45], as well as a tender point just medial and below the anterior iliac spine[42, 44-45]; increased irritation by adduction was described in one case series.[45] An injection with lidocaine/procaine in the lateral femoral cutaneous nerve, which should result in immediate relief of pain, is mentioned as a sort of 'gold standard' for diagnosis.[42-45] However, none of the case series used this 'gold standard' as inclusion criterion for the cases.

Muscular disorders

Disorders of the muscles in the hip region, except for sport injuries, is seldom reported in medical literature. Some reviews described gluteal tendinitis in elderly people, often occurring in combination with other hip disorders such as osteoarthritis, and difficult to differentiate from trochanteric bursitis.[46,47] No articles describing diagnostic criteria in this tendinitis were found. The only description of symptoms, based on case series, was found in an article on peritendinous calcifications of the tendon of the gluteus medius[10] (Table 1). General statements in reviews revealed that the patient will complain about pain located over the insertion of the tendon possibly with a caudal radiating pattern. Resisted abduction causes discomfort and there is local tenderness upon palpation.[5,46]

Piriformis syndrome

A frequently described muscular disorder in the hip is the piriformis syndrome, probably because of the entrapment of the ischial nerve in this syndrome and therefore mimicking lumbar sciatica. For many years this syndrome was a purely clinical syndrome. However, recent reports on imaging techniques such as scans and MRI suggest an affected piriformis muscles in selected cases.[48-51] In a study with both cases and control patients (although not blinded), Fishman[52] showed a delayed H-reflex in the affected limb of the cases with the piriformis syndrome compared to the controls; inclusion criteria for the cases were: 1) positive Lasègue sign at 45°; 2) tenderness at the sciatic notch; 3) increased pain within the distribution of the sciatic nerve when the hip was positioned in a combination of adduction, internal rotation and flexion. All authors describe local tenderness at the piriformis muscle as the most specific sign.[48-56] The diagnostic manoeuvre most frequently used and referred to is that of Page and Nagle.[57] Their report covered 45 cases with piriformis syndrome from a group of 750 patients admitted to the 'back service' clinic. They found pain and weakness on resisted abduction-external rotation of the hip a more consistent finding in the cases than the until then used Freiberg sign (pain on internal rotation of the hip, described by Freiberg in a review[58]). However, no exact data on the symptoms were given and no inclusion criteria for the cases were defined; symptoms in control patients were not given at all. Another diagnostic manoeuvre, pain caused by lifting the affected leg when lying on the side, was described by Beatty.[55] He found this manoeuvre to be positive in 3 cases and negative in 100 patients with surgically documented unilateral lumbar disc degeneration, and in 27 patients with hip abnormalities.

Disorders of bone and cartilage

Tumours

Reviews reveal that, in the elderly, primary tumours in the hip region are very seldom seen and, if they are present, usually involve osteosarcoma or multiple myeloma.[6] Metastatic tumours are more common in this region.[6,41] About 50% of the hip metastases are secondary to breast cancer,[41] followed by lung, prostate, kidney, and thyroid cancer as being mainly responsible.[6] Local pain, due to a pathological fracture or impending, can be the first symptom of a bone tumour. Plain X-ray and especially bone scans will show pathology; the definite diagnosis can be made from bone biopsy. Meals[59] described a series of seven cases with malignant disease (diagnosed by bone scan and biopsy) masquerading as hip osteoarthritis. The patients sought medical care for hip complaints, but on physical examination showed very few abnormal findings.

Hidden fractures

Schon[6] and McBeath[60] state in their reviews that femoral neck fractures in elderly people can be present without a preliminary major trauma. They may present with an antalgic gait[6], and a pain in the groin or a referred pain in the knee.[61] Morgan[62] published a series of five cases, all elderly women, with an undisplaced femoral neck fracture. The fracture was not recognised on the initial plain X-ray, but could be revealed with bone scans or on repeat X-ray after several weeks. Three of the five patients were able to walk but all had significant pain on weight bearing. The classical finding of external rotation and shortening was absent, and the range of hip movement was often well maintained. Underlying bone disease was common, with radiological evidence of osteopenia.

Transient osteoporosis

Several papers have been reported on the syndrome transient osteoporosis of the hip. This is an entity of unknown aetiology characterised radiographically by diffuse osteoporosis of the femoral head and acetabulum. The syndrome is called 'transient' because the clinical and radiological signs disappear after several months. Although this syndrome previously was only described as occurring in pregnant women, it was later also recognised in middle-aged persons, mainly men. [64-66] The term 'transient marrow oedema' has been suggested rather than 'transient osteoporosis' because the osteoporotic changes were quite variable in the 10 cases Wilson[67] investigated; however, an oedema of the bone marrow shown by MRI, was always present. Hoffman examined nine patients with this syndrome and on biopsy he found no histological evidence for osteoporosis, but did find that this syndrome is accompanied by increased intraosseous pressure. Clinical symptoms in this syndrome are often only defined as 'disabling hip pain', probably because of the emphasis in most articles on the radiological signs[67-73]. However, Bruinsma[74] described clinical symptoms based on seven cases; progressive groin pain upon weight bearing, increased hip discomfort at the extremes of movement, and minor limitations in range of movement usually in abduction and internal rotation. Additionally, Hunder[75] described more moderate limitations of hip movements in seven of the nine cases. Schils[76] reported that the focus of pain in his seven patients was consistently the groin area and anterior aspect of the thigh, and found normal range of movement in six patients.

Avascular necrosis

As in transient osteoporosis, the diagnosis avascular necrosis is made on radiological grounds. With positive findings at bone biopsy used as gold standard, MRI shows the highest sensitivity (89%) and specificity (100%) of the radiological techniques.[77] An insufficient vascularisation of the bone of the femoral head can induce avascular necrosis, with a fracture of the neck or a luxation of the head of the femur being the most common cause. [78] Insufficient vascularisation of the femoral head without previous trauma is also possible and can cause non-traumatic avascular necrosis. Its precise aetiology is still uncertain but an interosseous, intravascular venous blockage seems to be important.[79,80] There are predisposing factors for the non-traumatic avascular necrosis such as steroid therapy, alcoholism, sickle-cell disease, lupus, renal transplant, Cushing's syndrome, or Gaucher's disease in 75% of the cases[6] From an extensive epidemiological study in Japan we know that in about 35% of the cases steroids had been used over a long period.[81]

A well-known staging system of this disease is described by Ficat[82], who also described the clinical symptoms during the four clinical stages (Table 3). More recently, an accurate staging system was developed by Steinberg[83] based on the type of radiological changes and the extent of involvement; however, no clinical symptoms are incorporated in this staging system.

Osteoarthritis

Osteoarthritis of the hip is usually divided into two arbitrary groups: idiopathic or primary osteoarthritis, and secondary osteoarthritis. Secondary osteoarthritis of the hip is characterised by a recognised underlying hip abnormality such as trauma, osteonecrosis, congenital hip deformity, Perthes' disease, slipped epiphysis, etc. The idiopathic form develops without a known prior event or predisposing disease. Primary osteoarthritis of the hip can be local but can even be a part of a generalised osteoarthritis, and often has a familial predisposition.[84] Other published classifications of osteoarthritis are radiologically derived: classifications of hip osteoarthritis according to its radiographic pattern[86], classifications according to atrophic or hypertrophic bone response, and according to severity.[87] The radiological classification of osteoarthritis of the hip by Kellgren and Lawrence has been used as classification criteria in many epidemiological studies.[88] For the same purpose, the American College of Rheumatology presented in 1991 classification criteria for osteoarthritis of the hip based on clinical signs alone, and in combination with radiological signs and/or laboratory signs.[89] In a study with 114 patients with osteoarthritis of the hip and 87 control patients with other hip disorders, they determined the sensitivity and specificity of the features. These features were selected by an expert team as impor-

Table 3
Staging of osteonecrosis of the femoral head[82]

	Stage 1	Stage 2	Stage 3	Stage 4
Pathology	Cell death of osteocytes, the bone is still strong	Resorption of necrotic bone and production of new woven bone which is mechanically weak, the bone is now vulnerable for subchondral fracture	A subchondral fracture has taken place	Sequesters and collapse in the femoral head which leads to progressive deformation, resulting in osteoarthritis
Clinical symptoms	Sudden pain in groin, anterior thigh and medial knee especially under load, limitation of movement, especially internal rotation and abduction	Pain in groin, anterior thigh and medial knee, limitations of movement	Pain in groin, anterior thigh and medial knee, limitations of movement	Pain in groin, anterior thigh and medial knee, severe limitations in all planes
Rradiological signs	X-ray: normal Scan: increased uptake (+ cold spot) MRI: irregular belt around a necrotic region	X-ray: sclerosis with osteopenia, sometimes cysts Scan: as in stage 1 MRI: a crescent sign is sometimes visible	X-ray: a crescent sign becomes visible, some sequesters might already be visible	X-ray: sequesters, flattened sclerotic head with in the long run irregular contours and a narrowed joint space

tant in diagnosing hip osteoarthritis. The diagnosis established by the physician in one of the clinics included in the study served as gold standard that was subsequently controlled by an expert team. With multivariate methods diagnostic criteria sets were derived (see chapter 3, Table 1; this thesis). Studies describing the clinical symptoms in hip osteoarthritis, based on case series, are presented in Table 4. In reviews and overviews the pain distribution in osteoarthritis of the hip is described differently; groin, outer or inner aspect of the thigh, knee, buttock, and posterior thigh are all mentioned.[3,6,41,90-92] Neither the restricted or painful movements in osteoarthritis are described equally.

Arthritis

An infectious inflammation of the hip joint is very rare, but if this does occur there is severe pain in the hip region that increases with every movement of the joint. Weight bearing is almost impossible and a 'muscular defence' may be present. The diagnosis can become be confirmed by a joint punction.

Generally, arthritis of the hip joint will be non-infectious. Rheumatoid arthritis, gout, psoriatic arthritis, Reiters' disease, and ankylosing spondilitis are all conditions which can affect the hip joint with rheumatoid arthritis being the most common cause.[41] The hip joint becomes affected relatively late in this disease and generally the patient is already diagnosed as having this disease.[41] The hip joint will often be affected bilaterally, and patients will often show limitation of internal rotation and abduction of the hip.[95] Some important symptoms that distinguish rheumatoid arthritis from osteoarthritis are rheumatic radiological changes, which are junta-articular osteopenia and subchondral erosion, and possibly later, a protrusio acetabuli.[6,96,97] Furthermore, the morning stiffness will persist longer than at least 15 minutes in rheumatoid arthritis.[6,89] Laboratory findings will also be different in rheumatoid arthritis; the RA-factor may be present even as an increased sedimentation rate in direct relation to the activity of the disease.[6,89,96] Sonographic investigation may show joint effusion, but this feature is not specific for rheumatoid arthritis.[98]

Differential diagnosis of hip disorders

Obviously, it is not absolutely certain that a disorder is located in the hip if the patient complains about pain in this region. Extrinsic causes have also to be considered.

Table 4
Studies on symptoms in osteoarthritis of the hip

	Aim	Cases/ controls	Population	prospective	Selection criteria	Symptoms
Altman[89] 1991	Define diagnostic criteria for more uniform reporting of osteoarthritis	114/87	Patients from rheumatology clinics	Observational (prospectively collected)	diagnosis of osteoarthritis derived by physician, controlled by expert team	- pain location: lateral thigh 57%, groin 39%, radiating to knee 64% - pain on prolonged ambulation 97% - morning stiffness < 60 minutes in 91% - flexion ≤ 115° in 96%, internal rotatation <15° in 66% - pain at: flexion 80%, extension 64%, abduction 76%, adduction 68%, internal rotation 82%, external rotation 79% - antalgia 85%, Trendelenburg sign positive in 37% - shortened leg length in 42% - ESR < 20 mm/h in 58%, < 40 mm/h in 85%
Macys[93] 1980	Describe observations in osteoarthritis patients and suggest radiological staging	183/0	Patients in surgery clinic	Unspecified	patients with primary osteoarthritis (criteria described) undergoing hip replacement	- flexion contracture in 90% - ESR elevated in 60% - radiological bilateral signs in 78% - classification depending on location of joint space narrowing: lateral 16%, superolateral 46%, superior 16%, intermediate 3%, and medial 3%
Danielsson[94] 1964	Describe the natural history of osteoarthritis of the hip by observing the desease over a 10-year period	168/0	Patients who attended an orthopaedic clinic	Prospective observational with follow-up	patients with hip complaints and who had radiologically verified structural and/or joint-space changes	- pain location: greater trochanter 54%, groin 24%, 22% knee - starting pain in 62%, pain when walking in 72%, resting pain 25% - flexion contracture in 75%, external contracture in 27%, adduction contracture in 22%, abduction contracture in 0.8% - Trendelenburg test positive in 26% - bilaterality in 20%

Pearson[86] 1962	Observe clinical state and progress of disease of patients with primary osteoarthritis of the hip	400/0	Patients from orthopaedic clinics	Observational (prospectively collected)	Patients with primary osteoarthritis in whom sufficient information was available and in whom the osteoarthritis was not considered as secondary to previous hip disorder	- pain referred from hip to: greater trochanter 62%, back of thigh 13%, groin 9%, knee 4% - low back pain in 6% - initial loss of movement always internal rotation and extension. Further limitation leads to flexion deformity and limitation of flexion - if the maximal loss of joint space and greatest degree of sclerosis and cyst formation appear in the superior part of the joint (78%) the abduction and external rotation will decrease with subsequent progress - if the radiological signs appear medially in the deepest part of the joint the adduction and the external rotation will decrease with subsequent progress of the disease.

Referred pain from abdominal and pelvic organs

Malignancy of women's internal genitals can cause groin pain. In the initial stage of these malignancies, groin pain can be the only symptom. In men it is possible that during the initial stage of an epidymitis a vague groin pain is felt. Groin pain, often in combination with low back pain, can also be present in prostatitis.[99] Further examination, however, should make the diagnosis obvious. D'Ambrosia[100] emphasised the importance of a careful examination of the lower part of the abdomen in hip evaluation. As an example, he described appendicitis causing pain in the groin with an irritation of the iliopsoas muscle to such an extent that the leg goes into spasm with the hip held in a flexed, externally rotated position.

Pierron[41] and Schon[6] described (in reviews) that inguinal or femoral hernia may cause pain in the groin. Schon[6] mentioned that symptoms such as swelling in the groin that enlarges with sneezing, straining or coughing are also present in this disorder.

Referred pain or neuralgia from the back

Compression fractures of the first and the second lumbar vertebra can cause referred pain in the anterior upper leg. Usually, a pain in the back will also be present.[41]

A frequently seen disorder of the back in the elderly is spinal stenosis, which may cause radiating pain in the hip region and the anterior upper leg[3,6,41,101,102]; Thorne[103] described pain in buttock and posterior leg in a case report. The pain will decrease or disappear with forward bending of the back and will increase with extension of the back. If the patient feels pain during walking or standing, this will usually disappear when in the sitting or lying position. Spinal stenosis is caused by a narrowing of the spinal canal, and neural structures or other sensitive tissue can be compressed. The narrowing is often the result of a deflated intervertebral disc; the ligaments of the adjoining vertebra fold more or less together[102], and/or degeneration of facet joints.[103]

A herniation of the intervertebral disc in persons older than 50 years is rare and will therefore seldom be a differential diagnosis.[103] Posterior facet joint degeneration may cause a posterior facet syndrome with pain in low back, buttock, trochanteric region, and posterior thigh. Often, there is limitation of spinal movement. Especially lateral bending in combination with extension causes pain.[104]

Ankylosing spondylitis, psoriatic arthritis, Reiter's disease, Crohn's disease and ulceric colitis can (besides arthritis of the hip) cause sacroiliitis with referred pain in the hip region.[41]. Sacroiliitis results usually in pain in the upper inner gluteal square with a referring pattern to the groin and the medial and posterior upper leg. During physical examination, pain can be provoked by testing the sacroiliac joint.[104] Radiological findings will show a vague joint space with adjacent sclerosis and erosions.[6]

Vascular disorders

Atherosclerosis in the abdominal aorta, iliac arteries, and femoral arteries can cause stenotic lesions resulting in a claudication syndrome. Lesions at, or proximal to, the hypogastric arteries will cause gluteal pain or fatigue. Lesions proximal to the deep femoral artery will cause thigh pain. Typically the pain is characterised by a deep aching sensation that worsens with walking and is relieved after 2-5 minutes of inactivity.[6,101] Clinically, these vascular disorders are often difficult to distinguish from spinal stenosis. A major difference is that pain due to a vascular disturbance often disappears when standing still[101,103], whereas bending forward or sitting or lying down is necessary to relieve the pain due to spinal stenosis.[101] Another difference is that pain is present during bicycling in vascular disease; whereas in spinal stenosis bicycling is comfortable.[103]

Discussion

We found several articles describing hip disorders in adults, resulting in a report of disorders which can cause hip complaints in especially middle-aged and elderly patients. However, there were few studies that aimed to diagnose the separate disorders and distinguish them from one another. With some exceptions, the studies were mainly descriptive ones based on case reports. Only Collee[12] and Altman[89] described symptoms in control patients also. In studies concerning purely clinical syndromes, the physicians' diagnosis, without reporting on which grounds this diagnosis was made, often determined the cases. Such studies do not provide new knowledge. In syndromes where visualisation techniques can determine the diagnosis, this often served as gold standard or as inclusion criterion for the cases.

Although patients with hip complaints will visit a general practitioner at first, only one study was partly conducted in the primary care sector. Symptoms and/or diagnostic criteria in a patient population seen in primary care may, of course, differ from those in a population of hospital patients. Furthermore, some of the described syndromes are extremely rare in primary care, and in some syndromes the emphasis in the studies is put on diagnostic techniques which are not available in primary care. Without availability of bone scans and/or MRI, it is easy to mistake a 'transient osteoporosis' or an 'avascular necrosis' for an osteoarthritis due to the clinical symptoms in these syndromes. For a patient with avascular necrosis this may be harmful because diagnosis at an early stage might prevent the subchondral fracture or further destruction.[81]

Because of the high prevalence among middle-aged and elderly and the impact on daily living activities, a disorder such as osteoarthritis of the hip has often been the subject of epidemiological studies. Therefore, more effort may be made to standardise the diagnosis. For a long time, the radiological diagnostic criteria of Kellgren and Lawrence[87] were used for this purpose. Presence of radiological osteoarthritis according to Kellgren and Lawrence relies heavily on osteophytes and joint-space narrowing. However, there are good reasons to believe that osteophytes are independent age-related variables[94,105], and also that joint-space narrowing in older age is a normal feature.[106] Many studies which investigated the relation between clinical and radiological signs used 'pain in the hip' as clinical sign[8,88] Pain in the hip, however, may be present due to reasons other than osteoarthritis. The American College of Rheumatology conducted a more precise study to achieve criteria based on clinical signs, or a combination of clinical signs and radiological and/or laboratory signs[89]. Although this study, in deriving classification criteria for osteoarthritis, is the best to date, its shortcomings have been reviewed by McAlindon and Dieppe.[108] The controls were not matched for age and gender, were younger than the patients with osteoarthritis, and included many patients with rheumatoid arthritis. Important is that the patient sample in the ACR study does not represent patients in primary care. Furthermore, these criteria were intended for epidemiological research and not to diagnose an individual patient. These criteria sets contain the major characteristics of osteoarthritis of the hip and do not include the entire spectrum of disease manifestations.

In many articles the description of the symptoms in hip osteoarthritis differ widely. This difference may be caused by the different (or lack of) inclusion criteria for the cases. It may also depend on the stage or severity of the disease, or on the peri-articular syndromes such as trochanteric tendinitis/bursitis. Such peri-articular syndromes may be present besides or even because of the osteoarthritis. The pain location over the greater trochanter, described as one of the pain locations in osteoarthritis of the hip in several papers, may be caused by a tendinitis/bursitis but may also be caused by the osteoarthritis. Arnoldi[80] described an increased interosseous pressure in many of the osteoarthritis patients not only in the femoral head but even in the greater trochanter. Local tenderness at the trochanter should be a differentiating factor[109], but the papers describing the symptoms in patients with osteoarthritis did not investigate this.

The purpose of recognising diagnostic categories is, of course, not a goal in itself, but a tool for prognosis and/or therapy. Some of the described disorders in this review are self-limiting, whereas others may be more chronic. However, the present study of literature showed a lack of reliable diagnostic criteria for hip disorders in adult patients to be used in primary care. This conclusion forms the basis of our main study described in this thesis.

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2

HOW DO GENERAL PRACTITIONERS MANAGE HIP PROBLEMS IN ADULTS?

Bierma-Zeinstra SMA, Lipschart SS, Njoo KH, Bernsen RMD, Verhaar JAN, Prins A, Bohnen AM. How do general practitioners manage hip problems in adults? *(Submitted)*

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Introduction

Hip problems have an adverse effect on mobility and daily functioning, and with the increasing number of elderly, the prevalence of hip problems is expected to increase.[1,2] A recent Dutch study in the open population aged 55 years and over and living independently (n=2895) revealed that 16.6% of the women and 8.3% of the men reported hip pain.[3]

In case of these problems, the general practitioner is often the first physician to be consulted. This and the fact that not all hip problems are caused by osteoarthritis (OA) of the hip and only a minority of patients with hip OA finally undergo joint replacement surgery, results in a major role for the general practitioner (GP) in the management of hip problems in adult patients.

The GP has a number of diagnostic and therapeutic interventions at his disposal for patients with hip problems, but there is a lack of practice guidelines for these patients in primary care. However, some recommendations for the management of OA of the hip have been published.[4-6] These recommendations contain an initial treatment with paracetamol in order to avoid the side effects of non-steroidal anti-inflammatory drugs (NSAIDs). They also recommend, although on the ground of weak evidence, physical therapy to improve range of movement and muscle strength in patients with OA of the hip.

A large practice variation and/or deviation from available recommendations would necessitate the need for standardised and widely accepted guidelines. No detailed information about medical management, and consistency of such management, in different hip disorders is available. The aim of this study was to investigate the actual management in general practice of adult patients with hip disorders, and the consistency and determinants of this management.

Methods

General practitioners' management of hip disorders in middle-aged and elderly patients was studied in two ways. In method I, four 'paper patients' with hip disorders were presented to 20 GPs. In method II, the computerised patient records (CPRs) of 400 patients with new hip problems were analysed over a two-year period. Both studies took place in 1996.

General Practitioners (GPs)

Twenty GPs (15 male and 5 female) in fourteen practices in or near Rotterdam participated. They all worked for more than five years with CPRs using the same software (ELIAS), and none used paper records. The same GPs participated in both methods I and II.

Method I; 'Paper Patients'

The four paper patients were based on existing patients (Table 1). These cases were presented to the GP in an interactive way, which simulated a clinical setting. The researcher visited the general practitioner and confronted the GP with four different case scenarios. The general practitioner was presented with the written patient record consisting of the previous medical history of the patient together with the present reason for encounter (pain in the hip). After this the GP was asked to perform history taking and to state physical examination. For each paper patient a list with the results of all possible items of history taking and physical examination was prepared beforehand. The researcher played the role of the patient and answered all questions according to the list of predetermined answers and noted all items that the GP addressed. At the end of the consultation, pathological results of items that had not been addressed were reported to the GP in order to ensure that they all had the same clinically important information before they decided about medical management. Patient A was a 70-year-old woman with mild OA of the hip; patient B was a 52-year-old man with moderate OA of the hip; patient C was a 60-year-old man with a periarticular syndrome of the hip; and patient D was a 72-year-old woman with a hidden fracture of the hip (without major trauma, caused by secondary osteoporosis). Patients A, B and C had 3 consecutive consultations at 1, 6 and 18 months, whereas patient D had these three consecutive consultations within one month. Medication was defined as the prescription of paracetamol, non-steroidal anti-inflammatory drugs (NSAIDs) or corticosteroid injections.

Method II; Computerised patient records (CPR)

The investigator identified eligible CPRs of patients aged 50 years and over. Search criteria included terminology concerning hip problems in the free text, and diagnostic codes (ICPC: L13, L14, L89)[7] related to hip problems. From each participating GP, 20 CPRs of the identified patients who complied with the inclusion criteria were selected at random.

Table 1
Short description of abnormal findings in the paper patients

Paper patient A (female, age 70, mild hip OA)
– aching pain in left groin for three months, dull pain in left anterior thigh especially with prolonged walking or standing
– morning stiffness
– decreased walking distance
– Trendelenburg sign positive on the left side
– internal rotation, flexion, abduction and extension of the hip slightly painful in active movement and painful in passive movement, slightly decreased movement in all directions on the left side
– mild radiological osteoarthritis on the left side, doubtful radiological osteoarthritis on the other side

Paper patient B (male, age 52, moderate OA)
– dull pain in left groin for three months which is progressing
– pain continuously, worsening in walking and by lying on the side
– after overuse pain in the evening, pain when lying on the left side
– varus deviation in both knees
– hip joint capsule in left groin painful at palpation
– active and passive movements of the left hip, except adduction, painful; also decreased motion in these movements with the accent on internal rotation, abduction extension and flexion
– extension of both knees and flexion of one knee slightly decreased
– muscle resistance test for internal and external rotation of the left hip painful
– pain on sacro-iliacal provocation on the left side
– moderate radiological osteoarthritis on the left side

Paper patient C (male, age 60, soft tissue diagnosis)
– dull pain for one month on the right side in groin, buttock and inner thigh
– pain almost continuously, but worsens during and after walking and during sitting
– active and passive adduction, extension, and flexion are painful on the right side but not decreased
– groin and buttock muscles painful at palpation on the right side

Paper patient D (female, age 70, hidden fracture)
– heavy pain in right groin referred to knee, especially during walking and standing
– pain appeared two days ago after raising from a chair
– pain continuously
– 10 mg prednisolone daily (oral) for polymyalgia rheumatica
– decreased load on the painful side under walking
– Trendelenburg sign not possible on the right side because of pain
– active and passive internal and external rotation painful, no decreased motion on the right side
– thigh muscles painful at palpation (both sides)
– resistance test for the thigh muscles painful (both sides)
– radiological intertrochanteric fracture on the painful side

Inclusion criteria:

- Presenting with a new hip problem (pain in the hip region without a consultation for hip problems over the last three years).
- Aged 50 years or over at the time of the consultation for a new hip problem.
- A CPR available for the three years preceding and two years following the current consultation for a new hip problem.

Exclusion criteria:

- Presence of a hip prosthesis
- Presenting after an acute trauma of the hip.

The 400 patients with new hip problems were followed in the CPR for two years. The patient's symptoms, results of examination, diagnosis and management registered in the CPR, were noted by the investigator.

Statistical analyses

Differences in examination or management between the four paper patients were tested with a Chi-square test. Results were considered statistically significant at p-value < 0.05. On the basis of the 400 CPRs, determinants of management were analysed in multilevel logistic regression analysis. This 'multilevel' technique takes both the variation due to GPs and the variation due to patients into account.[8] Sampling units on level 1 were patients and sampling units on level 2 were GPs (patients nested within GPs). As independent factors were defined: age and gender of the patient, recorded diagnosis, and number of visits made per patient for hip problems during the two-year follow-up. In each analysis one type of management during this period was defined as the dependent variable. Receiving medication as well as receiving NSAIDs were analysed separately. Finally, the variance in management due to the general practitioner, estimated by multilevel logistic regression analysis, was translated into a 95% tolerance interval of the probability to receive one type of management during the first visit given a specific age, gender and hip disorder of the patient.

Results

Paper patients

Table 2 presents an overview of the medical history taking, physical examination and medical management of the four paper patients; with increasing unanimity be-

Table 2
Medical history taking, physical examination and medical management of the four paper patients by 20 GPs. The percentages represent the proportion of 20 GPs who addressed these items of examination or types of management.

	Paper Patient A female, age 70, mild hip OA	Paper Patient B male, age 52, moderate hip OA	Paper Patient C male, age 60, soft tissue	Paper Patient D female, age 70, hidden fracture
Medical history				
Pain location	100%	100%	100%	85%
Pain character	45%	65%	75%	55%
Pain appearance	95%	85%	95%	85%
Pain duration	85%	100%	75%	75%
Continuance of pain	15%	15%	20%	10%
Nocturnal pain	60%	60%	35%	25%
Morning stiffness	25%	5%	10%	5%
Joint stiffness	20%	10%	15%	5%
Daily functioning	10%	5%	5%	5%
Trauma	25%	25%	45%	15%
Overuse*	25%	25%	50%	10%
Self medication	10%	0%	5%	25%
Inspection and physical investigation				
Gait	60%	45%	45%	55%
Position of knee	35%	30%	20%	15%
Position of hip	45%	35%	30%	25%
Hip movements (active)	55%	50%	55%	35%
Hip movements (passive)	90%	90%	90%	75%
Back movements	25%	30%	35%	15%
Knee movements	15%	15%	10%	15%
Neurological investigation	10%	15%	30%	5%
Sacroiliacal provocation	10%	5%	15%	0%
Palpation*	35%	60%	75%	30%
Muscle resistance test	15%	15%	20%	15%

Management	A 1[a]	A 2	A 4	B 1	B 2	B 4	C 1	C 2	C 4	D 1	D 2	D 4
X-ray*(visit 1)	40%	70%	95%	60%	80%	80%	25%	45%	75%	70%	75%	90%
Blood sampling	10%	10%	15%	35%	40%	40%	25%	30%	35%	20%	25%	30%
Medication*(visit 1,4)	55%	70%	85%	20%	40%	60%	50%	55%	60%	15%	30%	30%
Physical therapy*(visit 2,4)	5%	55%	75%	20%	55%	80%	25%	75%	90%	10%	10%	10%
Orthopaedic surgeon*(visit 2,4)	0%	5%	60%	5%	35%	90%	0%	0%	30%	5%	70%	95%

* statistically significant difference between the four patients ($p < 0.05$)
a: 1 =management at the 1st visit, 2 =cumulative management after 2 visits, 4 =cumulative management after 4 visits

tween the GPs, the values in the table should approximate 0% or 100%.

History taking
All GPs asked about the exact location of the pain; most GPs asked when the pain had started and about factors influencing the pain. The GPs inquired about nocturnal pain more often with patient A and B ($p = 0.05$), and about overuse of the hip with patient C ($p = 0.04$).

Physical examination
Almost all GPs examined passive joint motion; half of them investigated gait and half examined active joint motion. The examination was similar for all patients except for patient B and C who underwent more often palpation ($p = 0.01$). Only a minority of the GPs made more extensive examination than described above.

Management
The GPs were most consistent with patient D. In the first consultation 70% of the GPs requested an X-ray of the hip and after having received the result the patient was referred to an orthopaedic surgeon. The management was less consistent in the other cases except for non-referral to an orthopaedic surgeon on the first consultation. Few GPs referred patients for physical therapy or blood sampling on the first visit. On subsequent consultations blood sampling did not increase, but referral to physical therapy did. The types of management showed significant differences between the four paper patients at one or more visits. NSAIDs were prescribed (80%) more often than paracetamol (20%). Corticosteroid injections were not prescribed.

Computerised patient records (CPR)

Demographics
CPRs of 400 patients with new hip problems were analysed: there were 259 CPRs (64%) concerning female patients mean age 67 years (standard deviation 11 years) and 141 (36%) concerning males mean age 64 years (standard deviation 10 years).

During the two years of follow-up most patients (67%) visited their GP only once for hip problems, 25% of the patients visited their GP twice, and 8% of the patients more than twice.

Management
The data from the CPRs showed a large variation in management between GPs (Figure 1). The largest variation was related to X-ray requests and prescription of medication. During the two years of follow-up 141 patients were prescribed NSAIDs, 31 patients paracetamol and 8 patients local corticosteroids injections.

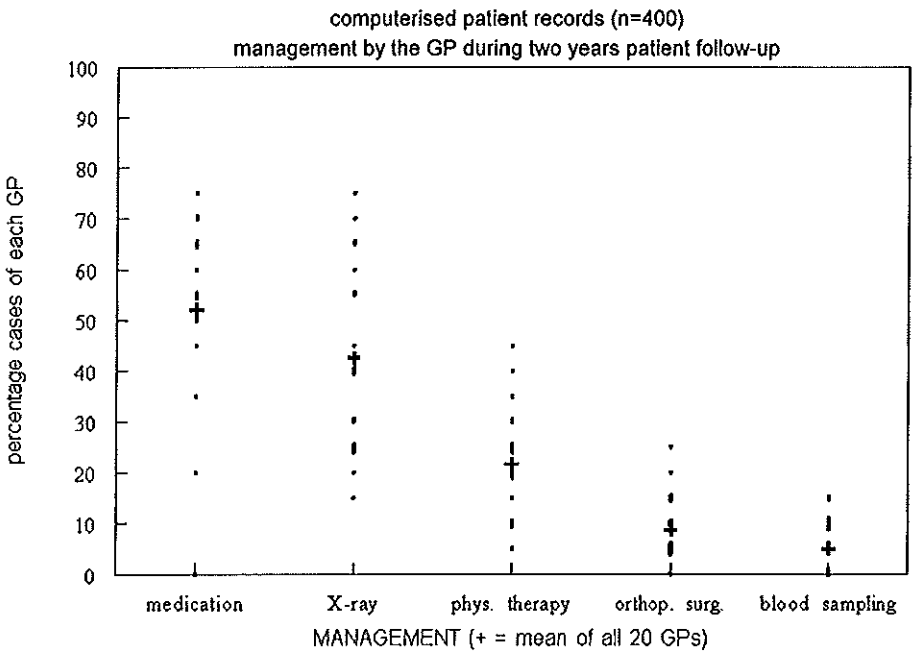

Figure 1
Mean management of each GP in 20 randomly selected patients aged 50 years and over with hip problems.

Diagnosis

For most patients (68.3%) no specific diagnosis for the hip problem was recorded in the CPR during the two-year period. The specific diagnoses that were recorded in the CPR can be categorised as: hip OA (18.5%), low-back disorders (4.8%), bursitis (3%), muscular disorders (3%) and other disorders (2.6%). The distribution of diagnoses varied between GPs; for example, the number of patients who received the diagnosis hip OA varied between GPs from 5% to 50%. In multilevel logistic regression model after adjustment for age, gender and number of visits per patient, still a large part of the variation, though on the border of significance ($p = 0.07$), was due to the different GPs. Being a female of age 60, the 95% tolerance interval of the probability to receive the diagnosis OA would range from 0.04 to 0.39 depending on the different GPs.

Determinants

The influence of different patient factors and diagnoses on the management was also investigated. Gender did not influence the management. Only the prescription of medication was related to age: older patients were more likely to get medication (odds ratio 1.03, $p < 0.05$). Patients diagnosed as hip OA and patients with a soft tissue diagnosis (bursitis and tendinitis) were less likely to be referred for X-ray investigation compared to patients with no specific diagnosis (odds ratio 0.38, $p = 0.003$ and odds ratio 0.24, $p = 0.014$, respectively). Patients with the diagnosis of hip OA also were less often referred to physical therapy compared to patients with no specific diagnosis (odds ratio 0.41, $p = 0.028$). The increasing number of visits made by the patients for hip problems resulted in an increase in prescription of medication, referral to physiotherapy, and referral for X-ray investigation (odds ratio ranging from 2.18 to 3.4, $p < 0.0001$). GPs contributed considerably to the variation in X-ray requests, medication prescription and NSAID prescription. Table 3 shows the variation in probability due to GPs to receive X-ray examination or medication at the first visit.

Table 3
Tolerance interval of the probability (i.e. probability ± 1.96.sd) to receive an X-ray investigation or medication at the first visit given a specific age, diagnosis and gender. Patients 2-4 resemble paper patients A-C.

	X-ray request	Medication	NSAID
1) female, 60 years, no specific diagnosis	0.25 - 0.68	0.19 - 0.75	0.10 - 0.64
2) female, 70 years, osteoarthritis	0.12 - 0.46	0.16 - 0.70	0.10 - 0.64
3) male, 52 years, osteoartritis	0.12 - 0.46	0.10 - 0.58	0.10 - 0.64
4) male, 60 years, soft tissue diagnosis	0.08 - 0.35	0.24 - 0.80	0.19 - 0.75

Discussion

Examination of both the paper patients and the CPR revealed inconsistencies in medical management. The four paper patients were based on existing patients and were presented to the GP in an interactive way; this method has proven valid for the study of clinical decision making.[9] However, using paper patients carries the risk that GPs act in a more idealistic manner than in daily practice. It is therefore likely that the differences revealed in the present study may be underrated. For this reason we used two types of study design (paper patients and CPRs) and based our conclusions on both studies.

Although patient D with a hidden fracture had the most consistent management, only 70% of the GPs requested an X-ray at the first consultation. Taking into account that prolonged load on this fracture increases the risk of displacement, thus worsening the prognosis, an X-ray request by all GPs would have been preferred. Palpation or muscle resistance tests were not routinely performed, though it is considered important for diagnosing soft tissue syndromes both in presence and absence of osteoarthritis[4]. Neither were the patients questioned about daily functioning when it is known that patients with hip problems often have such difficulties.[2] Hip patients are reported to have decreased function in sleeping, walking, personal care, household, mobility, and even in social interaction and cognitive functioning[1] and sexual activity.[10]

Demographic characteristics of the 400 patients corresponded with data of the National Survey of Morbidity and Interventions[2] on patients who consult a GP for hip problems, indicating that we obtained a representative study sample. Aspects of medical history and physical examination in the CPRs were not analysed in the present study because we suspected an underrating of these examinations in the CPR, where often only abnormal findings are recorded. X-ray requests and laboratory tests, prescription of medication, and referrals are always registered in the CPR and these data are therefore considered reliable.[11] Over 50% of Dutch GPs use CPRs to register the clinical data of the patient. We do not think the selection of only general practitioners with CPRs would be of influence on the results.

The CPRs showed a very high variation between GPs for X-ray requests and prescription of medication (particularly NSAIDs). Also disturbing is the large number of patients with hip problems for whom no diagnosis was entered in the CPR; many of these patients may have remained untreated due to a lack of diagnostic certainty. The variability in the number of patients registered by the GP as having OA is probably caused by the lack of standardisation of this diagnosis. In 1991 the American College of Rheumatology published criteria for the classification of OA of the hip.[12] These criteria were established in a rheumatology clinic and cannot simply be transferred to general practice; thus criteria specific for general practice should be established. During the two-year follow-up, 67% of the patients saw their GP only once. This may easily suggest that much of the hip pain is self-limiting. From other data we know that this is not the case. Miedema[2] showed that half of the patients who had consulted the GP for hip pain had persistent hip problems a year after the first consultation.

Medical management of patients in the present study differs from published recommendations for the management of osteoarthritis.[4-6] In our study most of the prescriptions were NSAIDs, particularly for the patients with OA; however, there are strong indications that these patients receive no additional benefit from NSAIDs compared with paracetamol.[13,14] In our study only a minority of the GPs referred patients with hip problems to a physiotherapist during the first consultation. During the two-year follow-up patients with the diagnosis OA in the CPR, for whom physical therapy was recommended[4-6], received even less physical therapy than patients with no diagnosis.

In our opinion the large variation between GPs in the diagnosis and treatment of hip disorders in adult patients is not acceptable. If evidence is available about a clinically relevant superiority of a certain treatment, this should be the preferred treatment. However, if evidence is lacking, further research is needed. The lack of proper diagnosis, the practice variation in diagnosis and therapy, and the use of non-recommended treatments may have a negative effect on patient outcome and may increase costs. Therefore, widely accepted and evidence based guidelines for the diagnosis and treatment of adult patients in primary care with hip problems are needed.

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3

VALIDITY OF ACR CRITERIA FOR DIAGNOSING HIP OSTEOARTHRITIS IN PRIMARY CARE RESEARCH

Bierma-Zeinstra SMA, Bohnen AM, Ginai AZ, Prins A, Verhaar JAN. Validity of ACR criteria for diagnosing hip osteoarthritis in primary care research. J Rheumatol. *(In press)*

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Introduction

The prevalence of osteoarthritis of the hip is expected to increase with the ageing of the western society.[1] This disorder is an important cause of pain and disability in the middle aged and elderly population.[2,3] To exchange research results in clinical and epidemiological studies, disease classification is an indispensable tool. For a long time, radiological signs have served as criteria for the presence of hip osteoarthritis. Radiological signs, however, do not correlate well with clinical symptoms, and cannot serve as a single criterion.[4] Therefore, the American College of Rheumatology[5] created three different sets of criteria for hip osteoarthritis: a set of clinical criteria (in classification tree format), and two combined sets of clinical criteria and radiographic criteria (one in traditional format and one in classification tree format). It is claimed that all three sets, separate from each other, can be used to diagnose hip osteoarthritis.[5] Since these ACR criteria were established in patients in rheumatology clinics, they cannot be transferred to general practice without validation. Although the validity of ACR criteria for osteoarthritis of the knee already has been studied in the general population, no validity studies are available for the ACR criteria for hip osteoarthritis.

As no gold standard for osteoarthritis of the hip exists, we studied the validity of the ACR criteria for this disease in primary care by investigating the agreement between the three ACR criteria sets (cross-validation) in hip patients in general practice.

Patients and methods

During 1996 (Jan-Dec) at the radiological departments of two hospitals in Rotterdam, consecutive patients who had consulted the general practitioner with pain in the hip region, and had been referred for X-ray investigation of the hip, were recruited for a standardised physical examination. The inclusion criteria were: age 50 years and older, and pain duration of at least one month and not longer than two years.

Patients with a hip arthroplasty on the painful side and patients in whom the general practitioner suspected a fracture or tumour were excluded. Patients were also excluded in whom history taking or physical examination was impossible due to co-morbidity. Written informed consent of the patient was obtained before they were examined in accordance with a standardised protocol, which included history taking, physical examination, and evaluation of the radiographs. History taking and physical examination in all patients was performed by the same observer. Data concerning pain location, pain endurance and character, pain aggravation, morning stiffness and daily activities were collected. Physical examination included pain provocation on palpation, on active/passive hip movement and on muscle resistance tests, goniometric measurements of passive and active hip joint motion, and muscle weakness testing. An examination of the back and knee was also performed. Anterior-posterior radiographs of the pelvis and axial radiographs of both hips separately ('frog-leg' position) were taken. The radiographs were evaluated according to a standardised protocol. The qualitative assessment of radiographic features of hip osteoarthritis, expressed in the Kellgren score[6] (see Appendix A), and other pathology was performed by two investigators; a radiologist specialised in skeletal radiology and a medical scientist with special training. Both investigators were blinded for each other and for the results of the physical examination. After assessment of the total sample, consensus was reached when both observers disagreed. This final outcome was used for the criteria sets. Measurements of joint space distance (superiolateral, superior and axial) and size of osteophytes were performed by one investigator (postgraduate medical student with special training) on the anterior-posterior radiograph. The repeatability of these measurements between two observers (postgraduate medical student and medical scientist) was tested in a representative subset of 64 radiographs.

Table 1
The three sets of ACR criteria for osteoarthritis of the hip.

ACR 1 Clinical criteria (classification tree format)	ACR 2 Combined clinical and radiographic criteria (traditional format)	ACR 3 Combined clinical and Radiographic criteria (classification tree format)
Hip pain + hip internal rotation < 15º and ESR < 45 mm/h (if ESR not available, hip flexion ≤ 115º) or + hip internal rotation ≥ 15º and pain on internal rotation and morning stiffness of the hip ≤ 60 min and age > 50 years	Hip pain + at least 2 of the following 3 features: - ESR < 20 mm/h - Radiographic femoral or acetabular osteophytes - Radiographic joint space narrowing (superior, axial and/or medial)	Hip pain + radiographic femoral or acetabular osteophytes or + ESR ≤ 20 mm/h and radiographic axial joint space narrowing

ESR = one-hour erythrocyte sedimentation rate

The patients were classified on the basis of the three separate ACR criteria sets for osteoarthritis of the hip (table 1). For each patient, only the (most) symptomatic hip, indicated by the patient, was used for all separate criteria sets. Further on in this paper referred to as 'the (most) symptomatic hip. The percentage agreement and the Kappa between the separate sets were calculated. The percentage agreement between two criteria sets is the percentage of patients who with both criteria sets are unanimous labelled as either osteoarthritis patient or non-osteoarthritis patient. The Kappa is the proportion of the percentage agreement, which is not attributable to chance. In the sets in which joint space narrowing was one of the criteria, we defined joint space narrowing at four different cut-off points (superior-lateral joint space: <3 mm, <3,5 mm, <4 mm, <4.5 mm, and axial joint space: <2.5 mm, <3 mm, <3.5 mm, <4 mm). Additionally, the agreement between the ACR criteria sets and radiographic hip osteoarthritis (Kellgren score of 2 or more), as well as the agreement between the ACR criteria sets and a criteria set based on hip pain and joint space narrowing alone, was studied. Approval for this study was given by the local medical ethical committees.

Results

During one year, 276 patients fulfilled the inclusion criteria of which 32 patients were omitted by the exclusion criteria. Of the remaining 244 patients, 227 gave informed consent and were subsequently studied using the standardised protocol. These 227 patients had a mean age of 66 years (SD 9.6) and 73% of them were women. Of these patients, 13% had bilateral hip problems. The (most) symptomatic hip showed a Kellgren score of 2 or more in 33.8% of the cases. Qualitative assessment of radiographic osteoarthritis according to the Kellgren score of 2 or more showed an agreement between two observers of 89% and a Kappa of 0.75. The presence or absence of the osteophytes, which were measured by two observers in 64 cases, showed an agreement of 97% and a Kappa of 0.94. Superolateral joint space and superior joint space measurements dichotomised at cut-off levels of <4 mm showed an agreement of 86% and 88%, and a Kappa of 0.66 and 0.74, respectively. Axial joint space measurements dichotomised at <3 mm showed an agreement of 82% and a Kappa of 0.62.

Table 2 shows the prevalence of the different signs in our study group and in the study population used to derive the ACR criteria.

The percentage agreement and Kappas between the three different ACR criteria sets, as well as with the radiographic osteoarthritis, are presented in Table 3.

There is low agreement between the set of clinical criteria and the two sets in which radiological criteria were included (Kappa 0.11 or lower). This agreement is not exceeded when the diagnosis osteoarthritis only is based on hip pain in combination with narrowed joint space. The agreement between the two combined sets is much higher (Kappa from 0.81 to 0.94). Radiological presence of osteoarthritis (Kellgren score ≥2) showed the highest agreement with the combined ACR criteria (Kappa 0.13 to 0.48).

Table 2
The prevalence of the different symptoms and signs in our study population (general practice) and in the study population used to derive the ACR criteria (rheumatology clinics).

	Hip patients in General practice (most symptomatic hip) N=227 % (n)	Hip patients in Rheumatology clinics* N=201 % (n)
Age > 50 years	96 (218)#	83 (165)
Internal rotation < 15°		49 (97)
Passive	3 (7)	
Active	5 (11)	
Pain on internal rotation		73 (140)
Passive	64 (145)	
Active	56 (127)	
Flexion ≤ 115°		89 (178)
Passive	73 (166)	
Active	84 (191)	
Morning stiffness ≤ 60 minutes	97 (221)	77 (150)
Osteophytes (acetabular and/or femoral)	45 (102)	55 (111)
Narrowed superior joint space		63 (127)
< 4.5 mm	57 (129)	
< 4 mm	30 (68)	
< 3 mm	12 (27)	
< 2.5 mm	11 (25)	
Narrowed axial joint space		58 (117)
< 4 mm	74 (168)	
< 3.5 mm	67 (152)	
< 3 mm	29 (66)	
< 2.5 mm	19 (43)	
ESR		
≤ 45 mm/hour	99 (216)	74 (93)
< 20 mm/hour	85 (185)	46 (58)

* Altman *et al.*[5]
\# Age 50 or older was an inclusion criterion in this study population

We also analysed the agreement in those patients in whom pain persisted longer than three months (144 patients). The agreement between the clinical set and the combined sets never exceeded a Kappa of 0.1, i.e. a low agreement.

Discussion

In the present study, we found that the clinical ACR criteria are not concordant with the two other ACR criteria sets and therefore show no cross-validity. We used different cut-off points for narrowed joint space and different combinations of narrowed axial joint space and narrowed superior joint space. The American College of Rheumatology[5] gave no exact data on cut-off points. Therefore, we used cut-off

Table 3

Percentage agreement and Kappa between the three different sets of ACR criteria, the Kellgren score of 2 or more, and the criteria based on narrowed joint space, in 227 patients with hip pain in general practice.

	Number of patients	Kellgren score ≥ 2 (n=75)		ACR 1		ACR 2	
		Agreement %	Kappa	Agreement %	Kappa	Agreement %	Kappa
ACR 1	135	48	0.03				
ACR 2							
Sjs <4.5 mm, ajs <3.5 mm	175	50	0.16	58	0.06		
Sjs <4 mm, ajs <4 mm	176	49	0.16	56	0.03		
Sjs <4 mm, ajs <3 mm	128	69	0.42	52	0.02		
Sjs <3.5 mm, ajs <2.5 mm	117	73	0.48	52	0.03		
ACR 3							
Ajs <4 mm	175	48	0.13	55	0.00	97	0.94
Ajs <3.5 mm	166	52	0.18	53	0.03	94	0.85
Ajs <3 mm	123	67	0.36	51	0.00	91	0.81
Ajs <2.5 mm	115	71	0.42	50	0.11	92	0.84
Hip pain and joint space narrowing							
Sjs <4.5 mm, ajs <3.5mm	183	48	0.13	58	0.04	87	0.60
Sjs <4 mm, ajs <4 mm	179	48	0.12	56	0.01	85	0.57
Sjs < 4 mm, ajs < 3mm	101	73	0.43	51	0.03	83	0.66
Sjs <3.5 mm, ajs < 2.5 mm	80	78	0.52	50	0.06	80	0.59

ACR 1 = clinical ACR criteria set
ACR 2 = combined clinical and radiographic ACR criteria set in traditional format
ACR 3 = combined clinical and radiographic ACR criteria set in classification tree format
sjs = supero-lateral joint space
ajs = axial joint space

points mentioned in their references[7], as well as lower cut-off points like those presented in the study by Croft.[2] However, there was little influence of these different cut-off points or the different combinations on the agreement between the clinical criteria and the combined criteria. Furthermore, analyses were carried out for both passive and active motion because it neither was mentioned if hip motion should be measured actively or passively, but also this resulted in only slightly different results. We had no access to measurements of the medial joint space; this sign is used in the combined set of clinical and radiographic criteria in traditional format. However, if accessible, even more patients might have been selected as osteoarthritis patients with the ACR criteria set, which gave already the highest percentage of osteoarthritis patients.

In earlier studies, the ACR criteria for knee osteoarthritis appeared not to be valid in the general population.[8,9] In our study, we investigated the criteria in a patient population seeking medical care, (albeit in general practice), and one might expect a better agreement here than in the general population. However, in our study the agreement between the clinical and the combined criteria was even lower than that reported in the study of Schouten and Valkenburg[9]. Several symptoms showed a highly dissimilar prevalence in our patient population compared with the patient population of rheumatology clinics used for development of the criteria. In general practice, a sedimentation rate above 45 mm/hour is rare, whereas this rate is often present in rheumatology clinics. Additionally, almost none of our patients showed morning stiffness more than 60 minutes. Therefore, these features are no good discriminators between osteoarthritis patients and non-osteoarthritis patients in general practice. Also striking was the low prevalence of internal rotation <15° in our sample compared with the ACR sample. Contributing factors to this low prevalence may be that our patients had less severe complaints and that only 27% of our patients were male, resembling the gender distribution of the middle-aged and elderly with pain in the hip in the general population[10] and in general practice.[1] In the ACR study population, however, the majority of the patients were male. It is known that men show less internal rotation than women[11,12] which underlines the risk of transferring criteria developed in one patient population to another, dissimilar, patient population.[13]

Consequences of relying on the ACR criteria for epidemiological studies in general practice are that, depending on which criteria set is used, different determinants may be shown.[9] In trials examining specific treatment in osteoarthritis patients, the power of such studies might be a major concern when too many patients with hip pain not arising from osteoarthritis are included in the study. By applying the combined clinical and radiographic sets, a sign of major importance for the diagnosis osteoarthritis is the presence of osteophytes. In the traditional format set (ACR 2), after fulfilling the criterion 'ESR < 20 mm/hour' (which applies to almost all patients in general practice) the presence of osteophytes is sufficient to classify a patient as an osteoarthritis patient. In the tree format (ACR 3), the presence of osteophytes alone is sufficient. Several studies have shown that osteophytes alone are more often associated with ageing than with other changes in osteoarthritis.[14] The major impact of this feature has consistently been reported as a concern in the Kellgren grading system, which also largely depends on the presence of osteophytes.[2,15] However, also the combination hip pain and narrowed joint space alone, which should be more applicable in the general population and possibly therefore also in general practice[2], did not show a better agreement with the clinical criteria. In the clinical criteria set (ACR 1), the pain at internal rotation mainly divided our patients in non-osteoarthritis and osteoarthritis patients because morning stiffness ≤ 60 minutes and ESR ≤ 45mm/h was present in almost all patients, and internal rotation ≤. 15° in almost none. Although it seems reasonable for clinicians to attribute a combination of decreased or painful internal rotation and morning stiffness ≤ 60 minutes to hip osteoarthritis, it is possible that the importance of these symptoms are derived by means of circular reasoning. In deriving the ACR criteria, the diagnosis made by clinicians, controlled by ACR committee members, served as 'gold standard'. It may be stated that the ACR criteria describe what the members of the committee consider the features of osteoarthritis of the hip to be.[16]

We conclude that for research in general practice the ACR criteria, at least one or more of them, are not valid. Further efforts are needed to establish valid criteria for different patient populations, especially for the general practice population in which hip pain and/or disability in middle-aged and elderly patients are highly prevalent.

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4

COMPARISON BETWEEN TWO DEVICES FOR MEASURING HIP JOINT MOTIONS

Bierma-Zeinstra SMA, Bohnen AM, Ramlal R, Ridderikhoff J, Verhaar JAN, Prins A. Comparison between two devices for measuring hip joint motions. Clin Rehab 1998; 12: 496-504.

Introduction

Limitations of hip motion are important signs of hip disease, such as osteoarthritis,[1,2] and accurate measurement of range of motion is essential for monitoring hip disease and for evaluation of treatment. Reliable and comparable measurements are especially important in research projects. Nevertheless, there is no universally accepted standard device for measuring hip joint motion.[3]

Two devices are available for measuring joint motion in a clinical setting: a two-arm goniometer and an inclinometer. Of these two instruments, the goniometer is most frequently used,[3,4] but the inclinometer is claimed to have greater reliability.[4] Many studies have shown superiority of the inclinometer in measuring complex motions such as movements of the spine,[5-7] only a few studies have compared the reliability of inclinometers and goniometers for measuring joint motion in the extremities.[8,9] None of these studies focused on the hip joint.

An inclinometer is driven by gravity, making measurements in the horizontal plane impossible with this device. Hip rotation and abduction and adduction with the patient lying supine, as recommended by the American Academy of Orthopaedic Surgeons (AAOS),[10] cannot be measured with an inclinometer. Most studies on normal range of motion (reference values) followed the AAOS methods of measurements and used a two-arm goniometer.[11-15]

When using a new instrument it should be verified whether it measures the same range of motion as the traditional instrument. For some movements, measurements with the inclinometer entail different positioning of the subject, and it is important to establish what influence these positions have on measurement outcomes. The present study was set up to address these two topics. In addition, it aimed to clarify which of the two instruments provides the most reliable measurements of hip motion in a clinical setting by determining the intra- and inter-observer variability of both instruments.

Methods

Subjects and Observers

The subjects were nine healthy persons (2 males, 7 females); age range 21-43 years. All volunteers were familiar with the purpose of the study. Ten medically educated observers, with previous experience in using a two-arm goniometer, received identi-

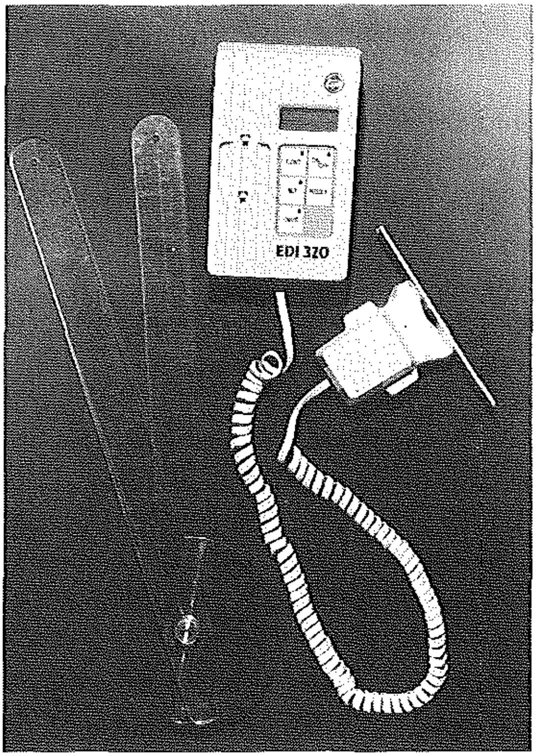

Figure 1
The two-arm goniometer (left) and the electronic inclinometer (right). The measuring unit of the inclinometer is extended with the 'long bone' attachment.

cal instruction and training in the procedures and measurement techniques of the two instruments before the study.

Material

The two-arm goniometer (Enraf Nonius, Delft, Netherlands) is a plastic long-armed (50 cm) instrument with a 180° scale marked in one-degree increments (Figure 1). The inclinometer, the EDI-300 (Cybex, New York, USA), is an electronic digital device with a measuring unit (which is placed on the moveable part of the body) and a display unit containing a microprocessor that processes and displays the range-of-motion in one-degree increments. This instrument also has an extra 'long bone' attachment for additional stabilisation of the measuring unit on the leg when testing hip motion.

Procedure

Systematic differences between instruments and positions

Simultaneous measurement with both instruments enabled systematic differences between measurements obtained with the both devices to be studied. The influence of the different positions of the subject was assessed by measuring the internal and external rotation in three positions (lying prone, sitting, and lying supine), and the abduction and adduction in two positions (lying on the side and lying supine).

Intra-observer variability

A 'test-retest' design was used with separate tests at short intervals; this type of test is accepted as the most accurate design for assessment of the reliability of instruments.[16] To study intra-observer variability, one observer measured the various hip movements 10 times consecutively in the nine subjects. The inclinometer and two-arm goniometer were removed and repositioned between each measurement. The observer was blinded for the results of the electronic inclinometer. The observer ensured that the alignment of the two-arm goniometer was correct, kept the goniometer fixed in this position, and read the results of the measurements.

Inter-observer variability

To examine the variability between observers, 10 different observers measured the internal and external rotation of nine subjects in sitting position. The measurements were made simultaneously with the inclinometer and the goniometer at short intervals (test-retest design). The same procedure was followed as used in assessing the intra-observer variability.

Movements in the horizontal plane can only be measured with the two-arm goniometer. Therefore, abduction and adduction and the internal and external rotation with the subject lying supine were measured with the goniometer but not with the inclinometer. Abduction and adduction were measured with the inclinometer with the subject lying on his or her side (lateral decubitus position); measurements in this position were not possible with the goniometer because the arm of this instrument is too long for exact alignment over the anatomical landmarks. Internal and external rotation with the subject sitting as well as lying prone, and flexion (lying supine) and extension (lying prone), were all measured simultaneously with the goniometer and the inclinometer. In all movements maximal passive and active measurements were obtained, except for extension and abduction, which were only measured passively (due to muscle fatigue during the active movements). Detailed information about the alignment of the instruments and the limb positioning during the measurements is given in Appendix B.

Data analyses

Differences in measurement between the different positions and between the two instruments were tested separately for each movement by means of ANOVA. Fixed effects in the model were: instruments, position, active versus passive movements, random effect: subjects. Ten measurements taken with one instrument in one position from one subject were considered repeated measurements.

In order to estimate the variability per instrument we used ANOVA (for each instrument separately and for active and passive separately) as recommended for experiments with more than two repeated measurements.[17] This was done for each movement separately and also for all movements in one model. The square root of the mean square error is an estimate of the intra-observer variability (within-observer standard deviation (SD)). The ratio of the two means square errors (obtained separately for the two instruments) has under H_0 an F distribution with degrees of freedom corresponding to the degrees of freedom of the two mean square errors. A p-value of 0.05 was considered statistically significant.

A similar procedure was followed to determine the inter-observer variability.

Table 1

Mean range of motion (ROM) in degrees in nine subjects measured with the electronic inclinometer and with the two-arm goniometer in different positions.

	Active mean ROM		Passive mean ROM	
	Two-arm goniometer	Electronic inclinometer	Two-arm goniometer	Electronic inclinometer
Extension	-	-	21.5	27.6
Flexion	116.1	120.5	126.6	132.0
Internal rotation				
- Lying supine	36.0	32.0	39.9	37.5
- Lying prone	46.3	44.2	53.2	50.4
- Sitting	33.6	-	38.2	-
External rotation				
- Lying supine	33.1	26.4	34.2	33.0
- Lying prone	47.0	38.0	51.9	43.0
- Sitting	33.9	-	37.6	
Adduction				
- Lying supine	19.2	-	17.1	-
- Lateral decubitus position	-	28.7	-	28.7
Abduction				
- Lying supine	38.0	-	43.2	-
- Lateral decubitus position	-	-	-	48.5

Results

Systematic differences between measurements and positions

Table 1 shows the mean outcomes of all movements measured. There were significant differences in measurement outcome in several movements that were measured with both instruments. Significantly (p=0.003) more flexion and extension with the inclinometer was observed. Significantly (p=0.016) more external rotation in the sitting position was measured with the two-arm goniometer. In the prone position more external rotation (p<0.001) was measured with the goniometer.

The position of the subject resulted in significant differences in the outcomes of measurements. More internal and external rotation was measured in prone position compared to sitting and supine position (p<0.001). Adduction was increased in lateral decubitus position compared to adduction in supine position (p=0.006). Whether this difference in adduction depends on the position or the instrument is not clear. No significant differences were found between measurement of the internal and external rotation in sitting and supine positions, and between passive abduction in lateral decubitus and supine positions.

Intra-observer variability

No significant differences in intra-observer variability were found when comparing measurements obtained with the two instruments when testing equality in overall variability for all movements at once. When testing equality in variability for each movement separately, however, significant differences were found. In measurements of passive rotations the variability was lower when using the inclinometer. In active rotation the results were contradictory; measurements of external rotation lying prone and internal rotation in sitting showed lower variability with the inclinometer but internal rotation in sitting showed lower variability with the two-arm goniometer. Table 2 gives the p-values for the tests of equality of intra-observer variance in measurements with the two instruments, the within-observer standard deviation (SD) of the different hip movements in the nine subjects, and the overall standard deviation within observers.

Inter-observer variability

Table 3 presents p-values for the tests of equality of inter-observer variance in measurements with the two instruments, and the between observers SD for external and internal rotation separately as well as the overall SD for these movements. In

Table 2
Variability within observers (intra-observer SD) in degrees in the nine subjects separately for each movement and in different positions, and an overall variability for all movements at once, separately for the two-arm goniometer and the inclinometer. In the analysis of overall variability only the movements which could be measured simultaneously with both instruments were included.

	Active			Passive		
	Two-arm Gonio-meter	Electronic Inclino-meter	*p*-value*	Two-arm Gonio-meter	Electronic inclino-meter	*p*-value*
Extension	-	-	-	2.8	3.3	>0.1
Flexion	3.0	2.8	>0.25	3.5	3.7	>0.25
Internal rotation						
- Lying supine	4.0	-	-	3.0	-	-
- Lying prone	4.4	5.5	<0.05	2.8	3.7	<0.05
- Sitting	4.4	3.0	<0.01	4.4	2.9	<0.01
External rotation						
- Lying supine	3.1	-	-	4.4	-	-
- Lying prone	4.1	3.2	<0.05	4.0	3.2	<0.05
- Sitting	3.0	3.1	>0.25	3.2	2.6	<0.05
Adduction						
- Lying supine	2.2	-	-	2.2	-	-
- Lateral decubitus position	-	2.9	-	-	2.9	-
Abduction						
- Lying supine	4.2	-	-	2.9	-	-
- Lateral decubitus position	-	-	-	-	3.4	-
All movements (overall SD)	3.8	3.7	>0.25	3.5	3.3	>0.25

* *p*-value for the test of equality of variance

Table 3
Variability between observers (inter-observer SD) in degrees in nine subjects in measurements of internal and external rotation in sitting position.

	Active			Passive		
	Two-arm Gonio-meter	Electronic Inclino-meter	*p*-value*	Two-arm Gonio-meter	Electronic inclino-meter	*p*-value*
Internal rotation						
- Sitting position	4.8	3.0	<0.01	4.9	4.2	>0.1
External rotation						
- Sitting position	4.1	3.6	>0.1	5.2	4.4	>0.1
Both movements (overall SD)	4.5	3.3	<0.01	5.1	4.3	<0.05

* *p*-value for the test of equality of variance

active internal rotation a significant difference was found; i.e. the SD was significantly smaller with the inclinometer. Overall variability for internal and external rotation together, was also lower with the inclinometer.

Discussion

Systematic differences between instruments and positions

An important observation in the present study was that we found that the two devices do not measure the same range of motion; this means that the instruments are not interchangeable for subsequent measurements. In addition, range of motion measured with the inclinometer cannot always be compared with reference values of normal range of motion measured with the two-arm goniometer.

The dissimilarities in outcomes found in the different positions have the same consequences. The degree of hip flexion is probably responsible for the differences in rotational movement when in sitting or supine position compared to prone position. The different outcomes of active adduction are probably caused, in part, by gravity, which supports the movement when the subject is lying on the side. Furthermore, adduction in this position (lying on the side) forces the hip in slight flexion. This means that the movement cannot be performed accurately in the frontal plane.

The question of which of the two instruments is the most valid was not investigated, but it was demonstrated that they do not measure the same range of motion. This was also found by Petherick *et al.*[9] in measurements of elbow movements. In clinical practice, a difference in range of motion between two sides, or between subsequent measurements to evaluate improvement or deterioration, is very important. For these measurements, a systematic error will not influence the results on condition that the same type of instrument is used.

Variability

Based on previous studies on measurement of movements of the spine[5,6,7] one would expect the intra-observer variability for the measurements with the inclinometer to be smaller than those with the goniometer. Although this was the case in our study in passive, and some active, hip rotations, we found an opposite result in active internal rotation lying prone. For hip movements in general, no significant difference in variability between the two instruments was found. For measurement with a goniometer, a precise alignment of the instrument is necessary. Precise alignment of the goniometer is probably easier for hip measurement than for measurement of complex movements of the spine; this may explain the superiority of the inclinometer in spinal measurements.

A drawback of the present study is that the observer was blinded for the results of the inclinometer, but not for the results of the goniometer. This bias was minimised by not allowing the observer to read the results on the goniometer before he had ensured the precise alignment of the goniometer and that the maximal range-of-movement was reached. In view of these precautions, it is not thought that this factor influenced the results obtained.

The inter-observer variability of the two instruments was significantly smaller with the inclinometer for active internal rotation and contributed to a difference in overall variability. However, the interobserver variability was determined only in rotational movement in the sitting position. In flexion/extension and abduction/adduction the results may be different.

In previous studies on measurement of motion, the variability between observers was larger than the variability within observers.[18-21] These findings were corroborated by us, but were more pronounced in passive rotations. An absence of uniformity in the amount of passive force applied by the 10 observers is the most likely explanation for the greater variability in passive movements between observers.

The coefficient of variation (CV), i.e. the standard deviation divided by the mean times 100%, is a well-established measure of variation in variability studies on measurement of movement. However, when the variation between repeated measurements is independent of the range of motion, as in this study, variation is better reflected in SD values. If the range of motion is small, the CV may become large, and vice versa. In this study an extreme low CV in flexion and extreme high CV in extension would have been found. Only when comparing variability of measurements with different dimensions, would the CV the more appropriate choice of index.

All subjects in the present study reported discomfort during measurement in the lateral decubitus position. In patients with hip problems, discomfort and pain may be intensified in this position, which will not contribute to reliable measurements. Some authors[3,4] reported poor stabilisation of the inclinometer on the body. In the present study, the extra attachment was used for additional stabilisation on the thigh. This was not possible in the rotational movements, and thus the measurement unit was used without the extra attachment. However, during measurement of the rotational and other movements the instrument was found to be firmly positioned on the body.

Reliability of a new instrument to measure range of movement should always be tested in healthy persons first. Results of intra- and interobserver variability studies in patients generally support the findings reported for healthy subjects.[16] However, it is possible that some specific patient problems make one instrument preferable above the other.

Unlike mechanical inclinometers, the electronic inclinometer used had to be recalibrated before each single measurement against a known vertical or horizontal axis. Also, with the different placements and the necessity of firm fixation of the instrument on the body, use of the electronic inclinometer may be time consuming. Another consideration is that measurements of adduction are not possible with the inclinometer. A disadvantage of the two-arm goniometer is that it is difficult to accurately position the goniometric arms and passively move a joint at the same time.

Conclusions

If consecutive measurements of hip movements are made by the same observer, the inclinometer is no more reliable than the two-arm goniometer, except for measurements of passive hip rotations. When different observers perform the measurements, the inclinometer is more reliable in active internal rotation; however, the differences are small.

The two instruments show systematic differences in range of motion of the hip. Therefore, these instruments are not interchangeable during consecutive measurements and corrected reference values for measurements with the inclinometer are needed. Rotational movement of the hip increases in prone position and is, therefore, not interchangeable with the other positions (lying supine and sitting).

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5

CLASSIFICATION OF HIP DISORDERS IN ADULTS

Bierma-Zeinstra SMA, Bohnen AM, Bernsen RMD, Ridderikhoff J, Verhaar JAN, Prins A. Hip problems in adults: classification by cluster analysis. *(Submitted)*

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Introduction

Of the population aged 55 years and over 8-14% suffers from pain in the hip region.[1] These hip disorders cause problems with respect to daily living activities[2], and are expected to increase with the ageing of the Western society.[3]

However, no universally accepted classification scheme or diagnostic criteria are available for patients with hip problems in primary care. In a previous study we showed a high variability between general practitioners in diagnosing 'hip osteoarthritis' patients (chapter 2, this thesis). These findings, and the fact that in the majority of hip patients the general practitioner did not reach a diagnosis, highlight the need for diagnostic criteria.

In clinical research, disease classification is an indispensable tool to compare research results. In clinical practice it is necessary in order to predict patients' prognosis and outcome of treatment. In many reports, the diagnostic criteria for hip osteoarthritis of the American College of Rheumatology are used.[4] However, these criteria appeared not to be applicable to patients with pain in the hip in general practice (chapter 3, this thesis). Radiological signs of osteoarthritis, which for long served as research criteria for the absence or presence of hip osteoarthritis, do not correlate well with the clinical symptoms and, therefore, may not be considered as gold standard.[5] Furthermore, not all hip complaints in middle-aged and elderly patients are caused by osteoarthritis. In many hip patients, we can not define the syndromes in chemical, physiological or immunological terms and are confronted with pure clinical syndromes.

When no gold standard is available, a syndrome has to be identified by a group of symptoms. When a certain pattern of symptoms occurs repeatedly in different patients, then the basic assumption is made that there is probably a common cause of the medical problem in these patients.[6] However, for hip disorders these patterns of clinical syndromes are poorly described and, if present, the methods of deriving them are often of inferior methodological quality or ill defined (chapter 1, this thesis). In a classification study such as by Altman *et al.*[4], methods are well defined and of high quality, but a serious problem in their study is 'circular reasoning'. In deriving such a classification 'experts' provide the diagnosis and will give symptoms that they already believe to be important more weight in their sorting decision. These symptoms will now correlate highly with the presence or absence of the disorder, and if diagnostic criteria are developed from such classifications especially these symptoms will appear to be important.

The aim of this study was to determine whether it is possible to obtain a valid classification scheme, of middle-aged and elderly patients with hip problems seen in primary care, free from preconceptions by using the methods of numerical classification (numerical taxonomy). In numerical classification the patients are grouped by mathematical algorithms on the basis of similarity or dissimilarity in symptoms.

Subjects and Methods

Patients and patient examination

During 1996, consecutive patients from two hospitals in Rotterdam with pain in the hip region who were referred by the general practitioner for radiological investigation of the hip, were extensively examined according to a standardised protocol. Written informed consent of the patient was obtained.

Inclusion criteria for the subjects were:
– aged 50 years or over on the day of examination,
– pain in the hip region during minimal 1 month and maximal two years,
– no hip arthroplasty at the painful side.

Patients were excluded when the general practitioner explicitly mentioned a suspected fracture or tumour on the X-ray request, or when history taking or physical examination was impossible due to co-morbidity.

The patients underwent a standardised history taking and physical examination of low back, hip and knee. All patients were examined by the same observer. Hip joint motion, measured with a two-arm goniometer, was later converted to an ordinal scale (not decreased, slightly decreased, and moderately decreased). Normal values for range of motion in adults were used to define these cutoffs.[7,8,9] In sensory examination we defined sensation to light touch as normal, decreased or increased. The radiographs taken were anterior-posterior exposures of both hips and the pelvic region, and a frog-leg position exposure from both hips separately. The X-rays were scored according to a standardised protocol by two investigators, independently of each other and blinded for the results of the clinical examination. The degree of hip osteoarthritis was scored according to the Kellgren scale (see Appendix A).[10] Signs of degeneration of the lumbosacral joints, symphysis joint and sacroiliacal joints, as well as radiographic signs of osteoporosis, were scored as present or absent. When both observers disagreed, consensus was reached. Laboratory tests included the one-hour erythrocyte sedimentation rate. Additionally, a sonographic examination was performed. Two consecutive measurements of the ultrasonic intra-capsular distance of each hip joint as described by Koski[11] were done with a 5 Hz convex array transducer. This was followed by sonographic examination of the trochanteric region where we looked for an ultrasonic visible increase of bursal fluid and oedema around the trochanteric tendons. Sonographic images were stored and checked and approved by an independent radiologist. All the above-mentioned patient examinations were carried out on the same day.

Classification method

In numerical classification two main types of approaches are available: the phenetic classification and the phylogenetic classification. The phylogenetic classification (cladistics) hypothesises an evolutionary structure, whereas the phenetic classification is an ordering based on resemblance in externally observable features[12]. Cluster analyses, the method for phenetic numerical classification, groups the cases into clusters on the basis of a set of variables which are collected in all cases. In our study an agglomerative hierarchical sorting algorithm was used which implies that the cluster analysis starts from single cases, which are step by step agglomerated into larger clusters.[13,14] The cases are placed in a multi-dimensional space, with as many axes as variables (one can imagine such a space with a maximum of three axes but conceptually there is no difference if more axes are used). The position of the case is obtained by establishing how this individual case scored for all these variables. Different methods can be used to decide which cases should be combined at each step. We used the Ward's method which at step one combines those cases to a cluster that show mutually the lowest squared Euclidean distance, i.e. the shortest distance in a multi-dimensional space. For each cluster of cases the means of all variables are calculated. Then for each case, the Euclidean distance to the cluster mean is calculated.[15] These distances are summed for all cases. At each step, the two clusters that merge are those that result in the smallest increase in the overall sum of the squared within-cluster distances.

The optimum number of groups has to be chosen by compromising between loss of clinical detail, clarity in structure and reproducibility.[12,13] We defined a 'stopping rule' for the clustering when the already largest cluster would agglomerate to an even larger cluster in the next clustering step. Because a solution with a small number of clusters is more stable than a solution with a large number, not more than 10 clusters were allowed. From large groups we identified the subgroups also. All analyses were done with SPSS+ software.

Variables

For the classification we used the variables from the patients' history and physical examination of the hip region on the painful side. If the patients had bilateral complaints, the most painful side was included in the analysis. Thus, variables derived from examination of low back and knee, as well as radiological, sonographic and blood sample variables were not used for the classification. Furthermore, case characteristics as age and gender were left out of the classification analysis because we did not want a clustering on the grounds of these variables. Strongly correlated variables ($r > 0.5$ and $p < 0.05$) were identified and only one of these variables was chosen for further use. Variables present in less than 5% of the cases or variables that had missing values were omitted from the analysis.

To achieve a standardised contribution of the variables, independent from the scale in which they were measured, the scores of each variable were converted into *z*-scores[13,15,16] (an average of 0 and a standard deviation of 1).

Reproducibility of the classification

Reproducibility of the obtained classification was tested in 10 sub-samples.[13] Each of these samples contained a random 75% sub-sample of the total sample. These samples were checked with repeated cluster analysis for recurrence of the separate clusters, which we had obtained in the total sample. A cluster was defined as present in the 75% sub-sample if the majority of the members of the cluster found in the total sample, constituted a cluster in the 75% sub-sample where they again represented the majority of this cluster.

The reproducibility was also checked by performing cluster analysis using the complete linkage algorithm on the total sample of patients, another method within the agglomerative hierarchical algorithm. If the classification is stable, the same clusters should be recognised in a classification with another cluster method.[14] The decision as to whether this new cluster method showed the same clusters was made in the same way as described above in the sub-samples.

Validity of the classification

The validity of the obtained syndromes was checked on the basis of external variables; variables which not were included in the cluster analysis.[13] These comprised variables from radiological, sonographic, and laboratory examination, and variables from knee and low back examination. Differences between the clusters concerning these variables were tested by means of a chi-square test at a significance level of $p < 0.05$.

Recognition of several syndromes in the classification by clinicians will also strengthen the opinion that we approached a real classification. The syndromes were presented to 20 clinicians (10 general practitioners with special interest in musculoskeletal diseases and 10 orthopaedic surgeons) containing the variables which were present (or absent) in each syndrome more than 0.5 standard deviation (SD) from the mean in the total sample. The clinicians had to state whether they had patients with similar symptoms, whether they recognised this as a syndrome and, if yes, which syndrome. The experts had no access to information about the external variables. Furthermore, the combination of symptoms in the clusters were compared with the combination of symptoms in hip syndromes described in a study on hip disorders in the middle-aged and elderly (chapter one, this thesis).

Results

Demographics of the study population

During one year, 244 consecutive patients complied with the inclusion criteria; 227 patients gave informed consent and were examined. In two cases we could not achieve a full physical examination; in one case deficient memory caused problems in history taking. Excluding these three patients resulted in a sample of 224 cases for the cluster analysis. In this sample the patients' age ranged from 50 to 86 years (mean 65.6; SD 9.6) with 27% men and 73% women. The following radiological diagnoses were made: 34% hip osteoarthritis at the (most) symptomatic side (Kellgren score ≥ 2), 46% degenerative lumbosacral joint, 18% degenerative signs of symphysis, 17% degenerative sacroiliacal joint, 18% osteoporosis. Reliable ultrasonic measurements were obtained in 213 patients. In our study population 80 patients (37%) showed an intra-capsular distance of 7 mm or more at the (most) symptomatic side or an increase of 1 mm or more compared to the contralateral side, indicating a joint effusion or synovitis.[11] Twenty patients (11%) showed a major effusion (ultrasonic intra-capsular distance of 9 mm or more) and 47 patients (22%) showed an increase of 1 mm or more on the (most) symptomatic side. An ultrasonic oedema around the trochanteric tendons was registered in 7 patients and an ultrasonic visible effusion of the trochanteric bursa in 3 patients, both at the (most) symptomatic side. Blood samples were obtained in 218 patients; the mean ESR was

Table 1
Variables used in the cluster analysis and their presence in the study population (n=224)

Symptom	Presence
Pain excitation/character	
pain onset after trauma	08%
pain onset after overuse	14%
pain increased by movement	33%
pain increased by sitting	31%
pain increased by lying	17%
pain increased by lying on the side	62%
pain increased by standing	57%
pain increased by walking	68%
pain increased after load	50%
pain after prolonged inactivity	76%
nocturnal pain	15%
morning stiffness	35%
continuance of pain (1,2,3)	12% (1), 50% (2), 38% (3)
pain endurance (1,2,3)	36% (1), 38% (2), 26% (3)
pain severity (0-10)	mean score: 6.24
Pain location (0,1,2)	
low back	23% (1), 5% (2)
groin	31% (1), 22% (2)
buttock	32% (1), 29% (2)
greater trochanter	34% (1), 31% (2)
anterior thigh	33% (1), 9% (2)
posterior thigh	08% (1), 3% (2)
lateral thigh	33% (1), 7% (2)
medial thigh	10% (1), 3% (2)
anterior knee	21% (1, 6% (2)
posterior knee	06% (1), 3% (2)
medial knee	06% (1), 1% (2)
lateral knee	19% (1), 4% (2)
lower leg	27% (1), 7% (2)
Tenderness on palpation	
iliopsoas muscle	17%
tensor muscle	23%
gluteus max. muscle	40%
glutteus med. muscle	41%
piriformis muscle	50%
hip capsule in groin	25%
linguinal igament	30%
greater trochanter	61%
ischial tuber	21%
posterior iliac spines	36%

Table 1 - continued

Symptom	Presence
Pain on muscle resistance tests	
hip flexion	36%
hip extension	14%
hip abduction in 90° flexion	24%
hip abduction in 0° flexion	42%
hip adduction	27%
hip internal rotation	27%
hip external rotation	41%
Weakness on muscle resistance tests	
hip abduction in 0° flexion	13%
hip internal rotation	09%
hip external rotation	09%
Decreased passive hip motion (0,1,2)	
flexion	20% (1)= <100° or ≥5° dif, 24% (2)= <80° or ≥10° dif
extension	33% (1)= <5° or ≥5° dif, 5% (2) = <-5° or ≥10° dif
abduction	30% (1)= <21° or ≥5° dif, 29% (2) = <11° or ≥10° dif
adduction	19% (1)= <10° or ≥5° dif, 8% (2)= <5° or ≥10° dif
internal rotation	18% (1)= <21° or ≥5° dif, 23% (2)= <10° or ≥10° dif
external rotation	24% (1)= <21° or ≥5° dif, 16% (2)= <11° or ≥10° dif
Pain at passive hip motion	
flexion	65%
extension	43%
abduction	71%
adduction	58%
internal rotation	64%
external rotation	43%
End-feel	
bony end - feel at passive hip movements	28%
Other tests	
pain on provocation sacro-ilacal joint	36%
pain on straight leg raising	11%
pain at joint compression	19%
decreased sensation anterio-lateral thigh	06%

Continuance of pain (1,2,3): 1= continuously, 2= several times a day, 3= several times a week
Pain endurance (1.2.3): 1= < 3 months, 2= 3-12 months, 3=12-24 months
Pain severity (0,10): analog scale from 0= no pain to 10=worst imaginable pain
Pain location (0,1,2): 0 = no pain here, 1= pain here, but not the worst pain, 2 = worst pain here.
Decreased passive hip motion (0,1,2) : 0 = not decreased, 1= slightly decreased, 2 = moderate or severe decreased; 1 and 2 based on range of motion and decrease of motion compared with the other side (dif).

12mm/h (range2-87 mm/h). 17 patients (7.8%) had an increased ESR (25 mm/h or more).

Variables

The variable 'Trendelenburg sign' had too many missing values in our study population because of pain or decreased balance and was therefore not used in the cluster analysis. Pain on active and passive hip movements, as well as decreased range of motion in active and passive movements, was strongly correlated. Decreased active motion and pain on active motion were therefore not used in the cluster analysis. The symptoms locally decreased or increased sensation of buttock, inner and posterior thigh, and muscle weakness at muscle resistance tests for extension, adduction and flexion of the hip were present in less than 5%. The remaining 65 variables used in the analysis are shown in table 1. All, but four variables (nocturnal pain, pain after prolonged inactivity, pain onset after overuse, and decreased external rotation) significantly contributed in a univariate analysis (p <0.05) to the classification into nine groups in the total sample. The exact z-scores of the 65 variables for the nine clusters are presented in Appendix C.

Obtained classification

Applying the 'stopping rule', the cluster analysis resulted in nine separate clusters (see Appendix D). Allowing one cluster step more, clusters 5 and 8 would have merged to a very large cluster of 62 cases. Further clustering, applying the same rule, resulted in five separate clusters (Figure 1). Because we aimed to achieve a specific as possible classification, the 'nine cluster' solution was validated. The largest cluster contained 44 cases and the smallest contained 8 cases. In 6 clusters, because they contained more than 15 cases, we identified their subgroups. In the ten 75% samples, 8 clusters were the best solution. The nine-cluster solution in the total sample, with the most discriminating variables in the separate clusters, is shown in Table 2.

Reproducibility of the classification

The reproducibility of the clusters is shown in Table 2. All nine clusters found in the total sample appeared in five or more of the ten 75% samples. The subgroups in clusters 2, 4 and 6 were also found in the ten 75% samples. In clusters 3, 5, and 8 only the larger subgroups (of clusters 3 and 5) were seen in the 75% samples. In the total sample, using the method of complete linkage clustering, 7 of the 9 clusters were found. Clusters 8 and 5 did not appear but some of their subgroups did.

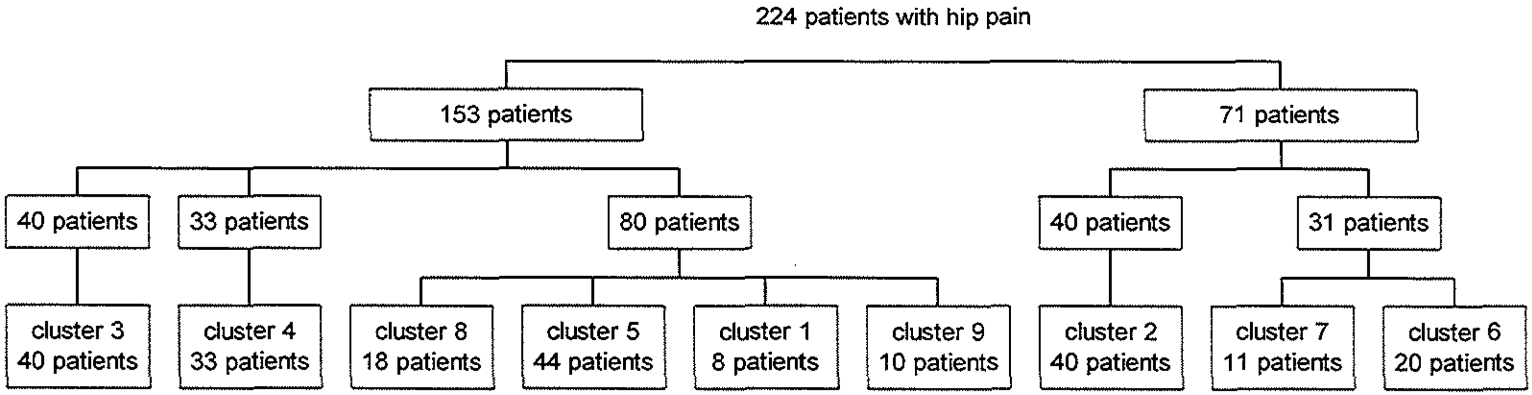

Figure 1
Schematic representation of the cluster tree, starting from the level of nine clusters, obtained by cluster analysis (Ward method) of patients (n=224) with hip pain based on 65 local symptoms from medical history and physical examination.

Table 2
Symptoms frequently present in the separate clusters obtained with cluster analysis in the total sample (*n*=224)

Clusternr, *n*	Variables which are 0.5 x SD or more than the mean present in the syndrome	Stability I*	Stability II**	Validation with external variables#
1, *n* = 8	***Pain excitation/character***: pain increased with standing, morning stiffness ***Pain location***: lateral thigh†, anterior knee ***Painful passive joint movements***: extension, adduction ***Other tests***: decreased sensation anterio-lateral thigh	7	yes	Radiological degenerative symphysis ($p = 0.015$)
2, *n* = 40	***Pain excitation/character***: pain increased by movements, walking, long pain endurance ***Pain location***: groin, anterior thigh, medial thigh ***Pain on palpation***: tensor muscle, gluteus med.muscle, inguinal ligament, hip capsule in groin ***Pain on muscle resistance***: internal rotation, abduction 0° ***Painful passive joint movements***: flexion, abduction, adduction, internal rotation ***Decreased passive motion***: flexion, extension, abduction, adduction ***End-feel***: bony end-feel ***Other tests***: hip pain at joint compression	8	yes	- radiological OA of the hip ($p < 0.001$) – ultrasonic major joint effusion ($p = 0.002$)
3, *n* = 40		8	yes	
4, *n* = 33	***Pain excitation/character***: discontinuous pain ***Pain location***: groin	8	yes	
5, *n* = 44	***Pain location***: lateral thigh, lateral knee, lower leg	7	only sub-groups	

6, n = 20	***Pain appearance/character***: pain increased with sitting, severe pain ***Pain location***: low back, buttock, posterior thigh, posterior knee, lower leg ***Pain on palpation***: greater trochanter, glut.med.muscle, glt.max.muscle, ischial tuber, posterior iliac spines ***Pain on muscle resistance***: flexion, extension, abduction 0°, adduction, external, internal rotation ***Painful joint movements***: external rotation ***Other tests***: pain on sacroiliacal provocation, pain on straight leg raising	8	yes	- sonographic effusion of trochanteric bursa (p <0.001) - decreased sensation and/or muscle strength in lower leg (p = 0.002) - painful extension (p = 0.011), flexion (p = 0.013) in low back and rotation/lateroflexion of the low back to the painful side (p = 0.008)
7, n = 11	***Pain excitation/character*** :pain onset after trauma, pain increased with lying, standing, and after prolonged inactivity, severe pain ***Pain location***: low back, anterior thigh, medial thigh ***Pain on palpation***: iliopsoas muscle, glut.max.muscle, piriformis muscle, ischial tuber, inguinal ligament, posterior iliacal spine ***Pain on muscle resistance***: flexion, extension, abduction 0°, adduction, external and internal rotation ***Weakness on muscle resistance***: external, internal rotation, abduction ***Painful passive joint movements***: abduction, adduction, eternal rotation, extension ***Decreased passive motion***: flexion, adduction ***Other tests***: pain on straight leg raising, sacroiliacal provocation, hip pain at joint compression	6	yes	- decreased sensation and/or muscle strength in lower leg (p = 0.001) - painful flexion in low back (p = 0.007) and rotation/lateroflexion in low back to the painful side (p = 0.006) - sonographic effusion of trochanteric bursa (p = 0.022)
8, n = 18	***Pain excitation/character***: increased by lying ***Pain location***: greater trochanter, lateral thigh ***Pain on palpation***: greater trochanter, posterior iliacal spine ***Weakness on muscle resistance***: abduction, internal rotation	5	only sub-groups	- sonographic oedema around trochanteric tendons (p = 0.001)
9, n = 10	***Pain excitation/character***: pain increased after load, morning stiffness ***Pain location.***: posterior knee, medial knee, lateral knee, anterior knee ***Pain on muscle resistance***: flexion, extension ***Painful passive joint movements***: abduction	6	yes	- knee effusion (p = 0.014)

† variables more than mean + 1x standard deviation (SD) present are underlined
* stability I, number of the ten 75% sub-samples in which the clusters were present
** stability II, presence of the cluster in the total sample using the complete linkage cluster method
# external variables which were more present (p <0.05) in patients of a certain group then in the other patients

Table 3
Clusters recognised as syndromes by the experts and in the literature.

	Orthopaedic surgeons (n = 10)	General practitioners (n = 10)	Study of literature
	Syndrome	Syndrome	Syndrome
Cluster 1	Meralgia paresthetica (n = 7) (pseudo) root syndrome (n = 3)	Meralgia paresthetica (n = 7) Osteoarthritis of the hip (n = 1)	Meralgia paresthetica
Cluster 2	Osteoarthritis of the hip (n = 10)	Osteoarthritis of the hip (n = 9)	Advanced degenerative hip joint disease (e.g. Osteoarthritis)
Cluster 3	Overuse (n = 1) Extra-articular referred pain (n = 1) Psychogenic (n = 1)	Referred pain from low back (n = 2) Coxalgia (n = 1) Sacro-ilacal joint (n = 1) Extra-articular referred pain (n = 1)	Stenosis low back Referred pain abdominal and pelvic organs ('no local signs')
Cluster 4	(Osteo)arthritis/synovitis of the hip (n = 8) Tumor (n = 1) Inguinal herniation (n =1)	(Osteo)arthritis (n = 4) Adductor tendinitis (n = 2) Tumor (n = 1) Iliopsoas bursitis (n = 1)	Early stage degenerative hip joint
Cluster 5	Tendinitis (Iliotibial tract) (n = 3) (Pseudo)root syndrome (n = 4) Osteoarthritis of lateral knee (n = 1) Pes planus (n = 1) Meralgia paresthetica (n = 1)	Low back disorder (n = 4) Tendinitis (Iliotiabial tract) (n = 3) Bursitis (n = 1) Knee (n = 1) Neuralgia (n = 1) (pseudo) root syndrome (n = 1)	Soft tissue pathology of lateral thigh
Cluster 6	Low back disorder (with radiation) (n = 8) Sacro-iliacal joint (n = 1) Hamstrings (n = 1)	Low back disorder (with radiation) (n = 5) Sacro-iliacal joint (n = 2) Osteoarthritis of the hip (n = 1) Baker's cyste (n = 1)	Low back disorder

Cluster 7	Pubic fracture (n = 5) Low back disorder (n = 1) Traumatised osteoarthritis (n = 1)	Fracture (n = 4) Symphysis pubica (n = 1) Post traumatic (n = 1) Coxalgia (n = 1)	Low back disorder Osteoarthritis of the hip Piriformis syndrome
Cluster 8	Trochanteric bursitis (n = 9)	Trochanteric bursitis (n = 9)	Trochanteric tendinitis/bursitis
Cluster 9	Knee problem (n = 5) Thigh problem (n = 1)	Knee problem (n = 6) Trochanteric tendinitis (n = 1) Compartment syndrome (n = 1)	Knee pathology

Validity of the classification

Signs from radiological, sonographic and blood examination, and symptoms from knee and low back investigation, which were seen significantly more in patients of one particular cluster than in the other patients, are shown in Table 2. Radiographic degeneration of the sacro-iliacal joint, lumbosacral joint, and osteoporosis were not seen significantly more in any of the clusters; neither did an increased ESR.

The combination of symptoms characterising the separate clusters was (except for cluster 3) recognised by the experts as syndromes (Table 3). Almost all experts recognised osteoarthritis of the hip in cluster 2 and trochanteric bursitis in cluster 8. The majority of experts recognised meralgia paresthetica in cluster 1, (osteo)arthritis in cluster 4, low-back problems in cluster 6, and knee problems in cluster 9. In clusters 5 and 7 the answers were more diverse. Tendinitis, (pseudo)root syndromes and low back disorders were equally frequently mentioned in cluster 5. In cluster 7 the suspicion of a fracture (often a pubic fracture) was the most frequently noted diagnosis.

The syndromes described in literature that are similar to the clusters show similarity are also listed in Table 3.

Discussion

In the present study we achieved a symptom-based classification as free as possible from preconceptions. Obviously, also in cluster analysis, the input variables determine the classification found. However, we used numerous and unselected local hip variables and not a selection of symptoms which we believed to be important. Therefore, we may assume that the achieved classification is based on an unbiased selection of hip symptoms.

The best way to test reproducibility of such a classification is to analyse whether a similar classification is reached in a new sample of hip patients.[12-14] Because we used a large number of variables (65) our population sample was not large enough to divide the group of patients in two different samples. However, we could reproduce the classification in the majority of the 75% sub-samples and with use of a different clustering technique on the same set of data; these are two other methods to test reproducibility of the obtained classification.[13,14]

Due to lack of a gold standard it is difficult to establish whether we reached a valid classification. However, if the classification makes sense, physicians must have met patients with symptoms similar to those described in the clusters and therefore must be able to recognise the combination of symptoms in at least some of the clusters as a syndrome. Most clusters were recognised as syndromes by our 20 experts. In most clusters the experts mentioned the same syndrome; however, in some clusters the answers were more diverse. In cluster 7 a fracture was frequently mentioned, probably because of the large number, and severity of symptoms. Radiological examination, however (which the experts had no access to), showed no fracture in any of the cases.

The set of variables in cluster 1 corresponds with the symptoms described in case series for the syndrome meralgia paresthetica.[17,18,19,20] The only symptoms often present in our cluster and not mentioned in these case series were morning stiffness and pain location at the anterior knee, all other symptoms corresponded. These two symptoms may also relate to the radiological degenerative symphysis, which was more often seen in this cluster.

The symptoms often present in cluster 2 resembled those described in osteoarthritis of the hip. Painful and/or decreased passive movements appeared as described in osteoarthritis of the hip.[21-23] Also, radiological hip osteoarthritis, major sonographic joint effusion, although not included in the cluster analysis, were present more often in this cluster than in the other patients of the total sample. Dividing the cluster in two subgroups, one group had worse symptoms than the other.

Cluster 4 showed a discontinuous pain in the groin as major characteristic with absolutely no pain in the trochanteric region or lateral thigh. Although several symptoms as described in osteoarthritis of the hip were more present than on average in this cluster, the level of 0.5 x standard deviation more than the average was only reached in the two subgroups of this cluster. These were pain location in anterior thigh, pain at palpation of the iliopsoas muscle and hip capsule in the groin, painful joint motions (internal rotation and flexion) and decreased hip abduction. This cluster fits the description of early stage osteoarthritis[23], frozen hip[24,25] or even an early stage of transient osteoporosis or avascular necrosis.[26,27]

Clusters 6 and 7 show symptoms which are reported in patients with low back disorders[28] but there are also some local hip symptoms such as pain at palpation of the greater trochanter and gluteal muscles, and painful muscle resistance tests. The symptoms described in the Piriformis syndrome[29-39] are, although among many other symptoms, present in this cluster. However, pain in the anterior and medial thigh; pain after prolonged inactivity, and decreased hip joint motions, which were also often present in cluster 7 are also described in osteoarthritis.[4,21-23] A combination of low back disorder or Piriformis syndrome and osteoarthritis of the hip may be present in this cluster. Surprisingly, sonographic effusion of the trochanteric bursa was only found in clusters 6 and 7. Collee *et al.*[31] described a trochanteric syndrome, originating from the low back. However, local signs of bursitis are not expected in this syndrome. These findings must be interpreted with caution because of the low prevalence of this sonographic sign.

Cluster 8 shows a combination of symptoms, described in trochanteric tendinitis or trochanteric bursitis.[32-34] The external variable 'sonographic effusion around the trochanteric tendons', was significantly more present in this cluster; however, a sonographic effusion of the bursa was not seen in this group. This may indicate that this cluster represents the patients with a trochanteric tendinitis or that a trochanteric tendinitis is more often visible with sonography than a trochanteric bursitis.

Cluster 9 was characterised by the major pain location around the knee, a painful muscle resistance test and a painful hip abduction. Although knee pain secondary to hip disease is described[23,36] the pain in the hip may also be secondary to a painful knee. Gait disturbance by knee pathology may cause painful hip muscles; the pain in the hip appears in this cluster after load. Because the external variable 'knee swelling' was significantly more present in this cluster we found the latter explanation more likely.

Clusters 3 and 5 caused problems in identifying them as described syndromes. Although the patients in cluster 3 were seen by the general practitioner for pain in the hip, they were characterised by the absence of local symptoms at physical examination. In this cluster, pathology outside the hip region has to be considered (e.g. referred pain). In cluster 5 the patients were characterised by pain on the lateral thigh, lateral knee and the lower leg. Because no external low-back variables were seen more often than in other cluster and because of the absence of pain in the groin, the description could fit tendinitis of the iliotibial tract. It is also possible that these clusters cover many separate syndromes (or yet unknown syndromes) which, above all, correspond to each other in the absence of any of the investigated specific symptoms.

Concluding, with numerical techniques we were able to make a classification in which the clusters could be reproduced, in which most clusters were recognised as syndromes by experts, in which most separate clusters showed significant relations with external variables, and which showed similarity with syndromes described in literature. Whether the classification is useful in clinical practice, which is the ultimate validation of it, has to be tested in further research. A classification not leading to a correct prognosis or specific treatment is of little use in clinical practice. The prognostic value of the classification revealed by cluster analysis in the present study should be compared with the prognostic values of radiological classifications, sonographic classifications, and combinations of these.

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6

ASSIGNMENT OF INDIVIDUAL PATIENTS TO SPECIFIC CLUSTERS OF HIP PROBLEMS: IDENTIFICATION IN CLINICAL PRACTICE

Introduction

The study described in chapter 5 identified several clusters of hip patients. When no gold standards are available a syndrome has to be identified by a group of symptoms. When a certain pattern of symptoms occurs repeatedly in different patients, then the basic assumption is that there is probably a common cause of the medical problem in these patients. A specific combination of symptoms constitutes the similarity between the patients within the cluster and the dissimilarity from the patients in the other clusters.

The obtained classification described in chapter 5 was based on 65 variables. In clinical practice it is not realistic to collect information on so many symptoms in a patient. Furthermore, even if all these 65 variables could be examined, how is this information then combined to assign a patient one particular cluster? We therefore have to identify the symptoms that are important to enable distinction between the clusters, and to develop a procedure for predicting cluster membership for new individual patients whose cluster membership is yet undetermined. In this way it may be possible to reduce the number of variables needed to classify individual patients. It also allows us to define diagnostic criteria for clinical syndromes, which do not have a pathognomonic sign or test as gold standard.

The aim in the present study is to develop a diagnostic function for hip disorders in middle-aged and elderly patients based on as few symptoms as possible that are also easy to examine by the general practitioner.

Material and methods

A study population of 224 patients with hip problems was used who had consulted the general practitioner were referred for radiographic investigation of the hip. All patients were assessed using a standardised protocol and formed the study population in the previous classification study (chapter 5). The assessed variables, related to medical history and physical examination and used for the classification are given in chapter 5 (Table 1).

With discriminant analysis the most discriminating variables (good predictors) in relation to the cluster membership were identified. We used a stepwise selection; a combination of forward selection and backward elimination with Wilks' minimalisation method.[1] This implies that variables that have small within-cluster variability compared to the total variability are entered first in the model. The entry of variables continues until the total variability is no longer attributable to the different cluster means of variables. When no more variables meet the entry (or removal) criteria the maximal allowed number of variables are selected. With these variables the classification function coefficients (Fisher's linear discriminant function coefficients) are computed. These classification function coefficients can be used directly for classification. A set of coefficients is obtained for each cluster and a patient is assigned to the cluster for which it has the largest discriminant score. We analysed the percentage of patients placed into the same cluster as obtained in the cluster analysis using only the five most discriminating variables increasing to the maximal allowed (by Wilks' method) number of variables. We assessed the prediction into nine different clusters, as well as the prediction into five different clusters, since some clusters may be very similar (e.g. soft tissue disorders).

Results

Aiming at the classification with nine different clusters we found 43% of the patients placed in the 'right' cluster (i.e. the same cluster as in the cluster analysis) using a discriminant function based on only five variables. With the maximal allowed number of variables 97% of the patients were placed in the right cluster. Prediction into five clusters resulted in a percentage patients placed in the right cluster ranging

Table 1
Percentage of patients (*n*=224) placed into the 'right' cluster, separately shown for classification into nine clusters and into five clusters.

	Percentage of patients placed into the right cluster of nine	Percentage of patients placed into the right cluster of five
Maximal set of variables allowed by Wilks	96.9 (49 variables)	98.6 (51 variables)
45 variables	95.1	98.6
40 variables	92.4	96.7
35 variables	88.8	95.3
30 variables	85.3	94.4
25 variables	83.5	93
20 variables	81	84.6
15 variables	71.9	81.2
10 variables	62.9	79.9
5 variables	43.3	63.6

from 63% with five variables to 99% with the maximal allowed number of variables (Table 1). To achieve a percentage of 80% of the patients placed in the right cluster, a physician would have to examine 20 variables if he wants to classify the patients into nine different clusters. To achieve the same percentage for classifying the patients in five different clusters, the physician has to examine only 10 variables. The 20 respectively 10 variables needed are listed in Table 2. Table 3 shows the examination scheme required for the physician to obtain information on all 20 variables. The exact classification function coefficients when 20 variables are used to classify the patients into nine different clusters are shown in Appendix E.

Table 2
The variables with the highest discriminating value for classifying patients into nine clusters or into five clusters; in both cases 80% of the patients are placed in the right cluster.

Variables	Outcomes	9 clusters	5 clusters
History			
Pain location:			
- groin	no=0, yes, but not worst=1, worst=2	X	X
- medial knee	no=0, yes, but not worst=1, worst=2	X	-
- posterior knee	no=0, yes, but not worst=1, worst=2	X	-
- lateral thigh	no=0, yes, but not worst=1, worst=2	-	X
- posterior thigh	no=0, yes, but not worst=1, worst=2	X	X
- medial thigh	no=0, yes, but not worst=1, worst=2	X	-
- greater trochanter	no=0, yes, but not worst=1, worst=2	X	-
- lower leg	no=0, yes, but not worst=1, worst=2	X	-
Pain excitation:			
- pain onset after trauma	no=0, yes=1	X	X
- pain increased with lying	no=0, yes=1	X	-
Physical Examination			
Passive hip motion:			
- decreased abduction	no=0, slightly=1, moderate/severe=2	X	-
- pain at maximal external rotation	no=0, yes=1	X	X
- bony end-feel	no=0, yes=1	X	X
Muscle resistance:			
- weakness at hip external rotation	no=0, yes=1	X	-
- weakness at hip internal rotation	no=0, yes=1	X	-
- weakness at hip abduction	no=0, yes=1	X	-
- pain at hip abduction	no=0. yes=1	X	X
- pain at hip internal rotation	no=0, yes=1	X	X
Palpation:			
- tenderness of posterior superior iliac spines	no=0, yes=1	X	X
- tenderness of groin	no=0, yes=1	X	X
Other tests:			
- decreased sensation of antero-lateral thigh	no=0, yes=1	X	-

Table 4 shows the distribution of patients who did not get the same cluster membership from the prediction with the discriminant function as in cluster analysis. The highest percentage of 'misclassified' occurred in patients with the cluster membership 'advanced hip joint'. However, most of the 'misclassified' moved to the cluster 'early stage hip joint'. Patients with cluster membership 'soft tissue' also had a high percentage of 'misclassified'. Most of these 'misclassified' patients moved to the cluster with no local signs.

Table 3
Examination scheme to identify cluster membership (nine clusters).

What is the exact pain location? Was the pain onset after a trauma? Does the pain increase with lying?
Passive abduction and external rotation (decreased, painful, bony end-feel)?
Muscle resistance tests abduction and rotations (painful or weakness)?
Pain on palpation at groin and at posterior iliac spines?
Decreased sensation to light touch over the anterio-lateral thigh?

Table 4
Number of patients with cluster membership received in cluster analysis (A), and from the prediction with the discriminant function using 20 variables (B).

		B Predicted cluster membership								
		1	2	3	4	5	6	7	8	9
A	1 'Meralgia Paresthetica' (*n*=8)	8								
	2 'Advanced hip joint' (*n*=40)	3	30		6	1				
	3 'No local signs' (*n*=40)			35	2	3				
	4 'Early stage hip joint' (*n*=33)	1	1	2	28	1				
	5 'Soft tissue'(*n*=44)	1	2	7		29	2		3	
	6 'Low back' (*n*=20)	1	1		1	1	16			
	7 'Low back + hip joint' (*n*=11)							11		
	8 'Trochanteric tendinitis/bursitis' (*n*=18)					1			16	1
	9 'Knee' (*n*=10)			1		1				8

n; number of patients

Discussion

By identifying the most discriminating variables for the classification we could considerably reduce the number of variables. With 10-20 variables we were able to place 80% of the patients into the right cluster, depending on the choice to classify into five different clusters or, for a more specific breakdown, into nine different clusters.

Of the 20 variables used to classify patients into nine different clusters almost half of the variables were variables derived from medical history taking. The physical examination does not have to be greatly extended to classify into nine different clusters rather than into five different clusters. Because the more specific classification into nine clusters may give more tools for prognosis and intervention, one should aim at the classification into nine clusters. The presence of seven pain location variables in the function emphasises the importance of the exact pain location for distinguishing pain syndromes in the hip region. However, a symptom such as painful or decreased internal rotation, which was found to be important in distinguishing patients with hip osteoarthritis from other hip patients[2], was not one of the 20 most discriminating variables. This symptom showed a very high prevalence in our study population and although this symptom occurred more in one of the clusters ('advanced hip joint'), it had a low discriminating value for the classification into nine different clusters.

The classification and prediction of cluster membership are carried out in the same study population. Because these very patients constituted the population on which the classification by cluster analysis was based, the prediction (however with less variables) may be better than in new patients who did not contribute to the classification. However, our first concern should be the clinical importance (prognosis and therapeutic consequences) and thereby the validity of this classification. We have shown that, provided this classification is clinically important, it is possible to introduce it into clinical practice without obliging clinicians to examine 65 items in each patient.

With the introduction of personal computers in general practice, the use of a diagnostic function is easy to incorporate in the clinical diagnostic process. After programming the diagnostic function into the personal computer the general practitioner has only to fill in the outcomes of the examination. The program will calculate the most probable cluster membership and can even show the probability size. Many other classification criteria lead to a division in absence or presence (with all uncertainty in it) of one specific disorder. However, getting the probability of the presence or absence of a certain disorder is more realistic than a mere division into presence or absence.[3] Presence or absence classifications for rheumatic diseases are often formatted as a certain number of criteria present, or formatted as classification trees.[4] These criteria sets are derived for one disorder at a time. However, we want to differentiate between nine different syndromes at once, in the most efficient way. In this study we have shown that with the use of a diagnostic function this is possible on the basis of a relatively brief medical examination and makes introduction of such a classification in general practice feasible.

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7

SONOGRAPHY FOR HIP JOINT EFFUSION IN ADULTS WITH HIP PAIN

Bierma-Zeinstra SMA, Bohnen AM, Verhaar JAN, Prins A, Ginai AZ, Laméris JS. Sonography for hip joint effusion in adults with hip pain. *(Submitted)*

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Introduction

The medical history and physical examination, sometimes supplemented with X-ray investigation, are the usual tools for the general practitioner to reach a diagnosis in adults with hip pain. Recently, ultrasonic examination for joint and soft tissue evaluation has been added to the available techniques. The diagnostic value of ultrasonic examination of hip joints in children has been extensively studied, but there are few reports on hip joint sonography in adults.

Until recently, joint effusion of the hip joint remained 'hidden' for the physician because it is difficult to detect by physical examination. It is now established that even small intra-articular effusion can be detected with sonography by measuring the distance between the collum of the femur and the joint capsule.[1-3] It is also known that inflammatory joint diseases, such as rheumatoid arthritis and septic arthritis, are often accompanied by joint effusion.[4-6] The prevalence of joint effusion in less severe, or early stage hip disorders is, however, not well documented.

That an enlarged ultrasonic distance reflects joint effusion has been confirmed by joint aspiration in previous research.[2,5] In false positive cases, instead of joint effusion often an active synovitis (without joint effusion) was shown.[5]

Koski *et al.*[5] showed a decrease of the enlarged ultrasonic distance after intra-articular corticosteroid injections in patients with chronic inflammatory joint disease. Several authors reported[2,7] that their management was influenced by detecting joint effusion. Even in less severe or early stage hip disorders a detection of hip joint effusion may have consequences for diagnosis and/or therapy. We do not know which symptoms in early or less severe hip disorders relate to joint effusion. Foldes *et al.*[7] demonstrated a positive correlation between nocturnal pain and joint effusion in patients with hip pain. However, as in all other studies on ultrasonic examination of the hip joint, these patients showed severe and/or late stage of disease and were not representative for hip patients in general practice.

Before considering a more routine application of ultrasonic investigation in general practice, the diagnostic value of this examination in relation to other symptoms and signs in patients with hip pain should be studied. The purpose of the present study was to investigate the prevalence of hip joint effusion depicted by sonography, and its relation with physical symptoms, radiological signs, and laboratory results in adult patients consulting the general practitioner with pain in the hip.

Table 1
Presence of variables from medical history and physical investigation in the study population (n = 224) in the (most) symptomatic hip and their relationship with hip joint effusion (odds ratio) adjusted for radiological osteoarthritis of the hip and age.

Variable	Presence in our study population N = 224	Odds ratios (p<0.05)
Nocturnal pain	15%	
Morning stiffness	35%	
- Pain onset		
after trauma	8%	
after overuse	14%	
- Pain aggravation		
by lying on the side	62%	1.9[1]
by walking	68%	
after load	56%	
after prolonged inactivity	76%	
- Location of worst pain		
groin	22%	2.5[3]
greater trochanter	31%	
medial thigh	3%	17.8[2]
anterior thigh	8%	
lateral thigh	7%	
- Inspection		
Trendelenburg sign positive	38%	2.7[2]
- Pain on palpation present		
iliopsoas muscle	17%	
gluteus maximus muscle	40%	
gluteus medius muscle	41%	
hip capsule in groin	25%	2.9[2],2.1[3]
inguinal ligament	30%	1.9[1]
greater trochanter	61%	
- Decreased passive hip joint motion present		
flexion	44%	2.2[3]
extension	38%	2.1[3]
abduction	59%	6.8[2]
adduction	27%	
internal rotation	41%	2.2[3]
external rotation	40%	
- Painful passive hip joint movements present		
flexion	65%	
extension	43%	
abduction	71%	
adduction	58%	
internal rotation	64%	
external rotation	43%	1.9[1],2.4[3]

[1] Effusion defined by Koski (n = 80); [2] Major effusion (n = 20); [3] Unilateral effusion based on left/right difference (n = 47); flexion: decreased = <100° or ≥ 5° decrease in relation to the other side (d); extension: decreased = < 5° or ≥ 5°d; abduction: decreased = < 21° or ≥ 5° d; adduction: decreased = < 10° or ≥ 5° d; internal rotation: decreased = < 21° or ≥ 5° d; external rotation: = < 21° or ≥ 5° d

Patients and methods

Patients

During 1996, patients who visited the general practitioner with pain in the hip and who were referred for X-ray investigation of the hip were eligible for inclusion. The inclusion criteria were: age 50 years and over, and pain in the hip region during minimal one month and maximal two years. Patients in whom the general practitioner suspected a fracture or tumour were excluded. Also excluded were patients in whom history taking or physical examination was impossible due to co-morbidity, and those who had a hip arthroplasty on the painful side. All patients were asked to give written informed consent.

Method

A standardised medical history taking and physical examination was performed. The items investigated are listed in Table 1. Additionally, a standardised sonographic examination was performed.[1] During this examination the patient was lying supine with the heels together and the hip externally rotated 10-15°. A 5 Hz convex array transducer was applied on the skin in the direction of the neck of the femur. When the joint capsule could be followed from the acetabulum to the point of its fixation to the collum, measurements on a magnified picture on the monitor were made. The longest distance, perpendicular to the joint capsule, from the joint capsule to the femur was measured (Figure 1). The measuring points were the lower edge of the capsule and the upper edge of the osseous echo. Two measurements were made on each side. For each hip, the mean outcome of the two consecutive measurements

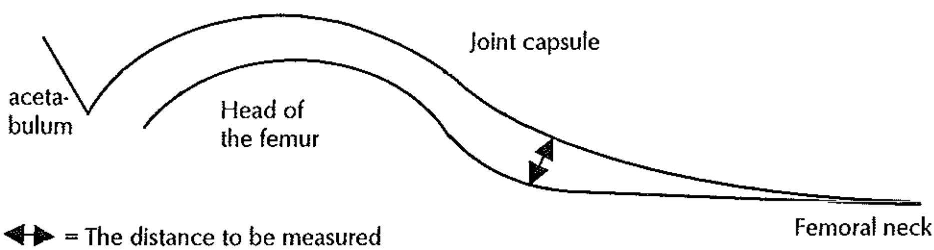

Figure 1
Diagram of the hip joint as seen during sonographic examination (anterior view).

was used as final result. All above-mentioned examinations were done by the same investigator. The ultrasonic images and measurements were stored and were checked and approved by another independent radiologist. Non-reproducible measurements due to unclear images were excluded from analysis. An ultrasonic distance of 7 mm or more, or a difference between the hips of 1 mm or more were considered as a intra-capsular 'effusion' in the hip joint (Koski's definition[1]). Besides this definition, we also used two other definitions. When an ultrasonic distance of 9 mm or more was measured we defined this as a 'major effusion'. In children, a left/right difference in ultrasonic distance between the two hips is considered to be of more diagnostic value than to a high ultrasonic distance alone.[8] Therefore, we also defined 'unilateral effusion' based on only a left/right difference in ultrasonic distance of 1 mm or more (the most symptomatic hip compared to the other hip). Intra-observer variability in these measurements of ultrasonic distance in healthy persons has been shown to be small ($r = 0.94$)[1].

After the ultrasonic examination, an anterior-posterior X-ray of the pelvis was made, as well as a frog-leg position X-ray of each hip. The radiological features used in this study were scored blinded for the results of other examinations. Axial and superior joint space were measured, and the presence or absence of femoral and acetabular osteophytes were noted. Radiological osteoarthritis was scored according to the Kellgren scale (see Appendix A).[9] Radiological osteoarthritis in the dichotomous scale was defined as a Kellgren score ≥ 2. Finally, a blood sample was taken and the one-hour erythrocytes sedimentation rate (ESR) was determined. In our study population increased ESR in the dichotomous scale was defined as ESR ≥ 25 mm/h.

Statistical analysis

The relationship between the presence of hip joint effusion and other symptoms and signs was tested with chi-square (dichotomous and ordinal variables) and with Pearson's correlation test (numerical variables). The percentage explained variance in joint effusion by medical history and physical investigation was assessed with logistic regression analysis with as independent variables those variables which in univariate testing showed a relation with joint effusion of $p < 0.05$.

Also in a logistic regression model the relationship between joint effusion and symptoms/signs from history, physical examination adjusted for possible confounders as radiological osteoarthritis of the hip (Kellgren score ≥2) and age was assessed.

In people with unilateral hip problems (patients consulting for bilateral pain or BSE >25mm/h were excluded) we assessed if left/right difference in various joint motions could predict an effusion based only on side differences in ultrasonic distance. Linear regression analysis was used (left/right differences of joint motion as independent variables, and left/right difference of ultrasonic distance as dependent variable) to identify the most related joint motion(s). We also assessed pre and post test probabilities of an effusion based on unilateral effusion by examining side difference in range of joint motion (dichotomy, side difference in extension >5°).

All analyses were performed with the SPSS+ package for Windows-95; results were considered statistically significant at a p-level <0.05.

Results

A total of 244 consecutive patients during one year complied with the inclusion criteria of which 227 patients gave informed consent. Of these, three cases were excluded; two because a full physical examination was not possible and in one patient impaired memory precluded history taking. Therefore, the study population comprised 224 patients, with a mean age of 66 (SD 9.6) years. Of these, 164 patients (73%) were female and 29 patients (12.9%) consulted for bilateral hip problems. X-ray's of the (most) symptomatic hip were missing in two patients. The number of patients with Kellgren score 0-4 in the (most) symptomatic hip were 88, 59, 44, 23, and 8 respectively. Blood samples were obtained in 218 patients; the mean ESR was 12 mm/h (range 2-87 mm/h). 17 patients (7.8%) had an ESR of 25 mm/h or more.

Reliable ultrasonic measurements of the (most) symptomatic hip were obtained in 212 patients and of the less or non-symptomatic hip in 213 patients. Due to a hip arthroplasty on the non-symptomatic side measurements could not be made on this side in 8 patients. Measurements could not be obtained due to poor ultrasonic visibility in 3 patients on both sides, and in 6 patients on the (most) symptomatic side. Three other cases were excluded by the independent radiologist due to non-reproducible measurements on the (most) symptomatic side.

The mean ultrasonic distance in the (most) symptomatic hip was 6.8 mm compared with 6.5 mm in the less or non-symptomatic hip. Joint effusion as defined by Koski was found in 80 patients (38%) in the (most) symptomatic hip and in 28% of the patients in the less or non-symptomatic hip. A major hip joint effusion in the (most) symptomatic hip was found in 20 patients (9.4%) compared with 16 (4.7%) patients in the less or non-symptomatic hip. In 47 patients (22%) there was a unilateral effusion in the (most) symptomatic hip and in 18 patients (8.5%) in the less or non-symptomatic hip. There was a significant correlation between the ultrasonic intra-capsular distance of both sides of the hip, even when 29 patients who had consulted for bilateral complaints were excluded ($r = 0.74, p < 0.001$).

Variables from medical history and physical examination, with their significance level in relation to joint effusion in the (most) symptomatic hip, adjusted for age and radiological osteoarthritis of the hip, are presented in Table 1. Pain aggravated by lying on the side and pain at passive abduction had shown a significant relationship with unilateral joint effusion in univariate analysis ($p < 0.05$). After adjustment for age and radiological osteoarthritis of the hip they showed no longer a significant relationship with joint effusion. More severe decreased extension (< -5° or ≥ 10° left/right difference) showed, adjusted for age and osteoarthritis of the hip, very high relationship with major effusion and with unilateral effusion (odds ratio = 10.7, $p = 0.0025$ and odds ratio = 7.5, $p = 0.006$ respectively). This was not the case for the other joint motions.

Significant relationships between radiological signs and laboratory signs with joint effusion were found. Major joint effusion showed a positive relationship with the presence of radiological hip osteoarthritis, both in the dichotomous scale (p = 0.013) and the ordinal Kellgren scale ($p = 0.032$) for radiological osteoarthritis. A major effusion also showed a relationship on the border of significance ($p = 0.057$) with femoral osteophytes. Joint effusion according to the other definitions showed no relationship with osteoarthritis of the hip, osteophytes or dichotomised joint space narrowing. A significant relationship existed between major effusion and increased ESR in a dichotomous scale ($p = 0.023$) which adjusted for age and radiological osteoarthritis still was significant. An even stronger relationship ($p = 0.002$) with increased ESR was found in bilateral major joint effusion, present in 9 patients.

Severity of pain, as determined by a linear analogue scale (zero representing no pain and 10 the worst imaginable pain) showed no correlation with the ultrasonic distance in the (most) symptomatic hip; neither did the diameter of the head of the femur. There was a slight negative correlation between the superior joint space respectively the axial joint space, measured on the X-ray, and the ultrasonic distance (r = 0.18 and $r = 0.19$ respectively). We also found a correlation between age and ultrasonic distance ($r = 0.21$); the correlation remained ($r = 0.30$) when we selected those cases with no radiological signs of osteoarthritis (Kellgren score 0) at all.

Logistic regression showed that maximal 24% (Nagelkerke R^2) of the variance in major joint effusion could be explained by medical history and physical examination. This increased by 3% when age was added and once more with 0.3% when radiological osteoarthritis (Kellgren scale 0-4) was added. Using the definition of

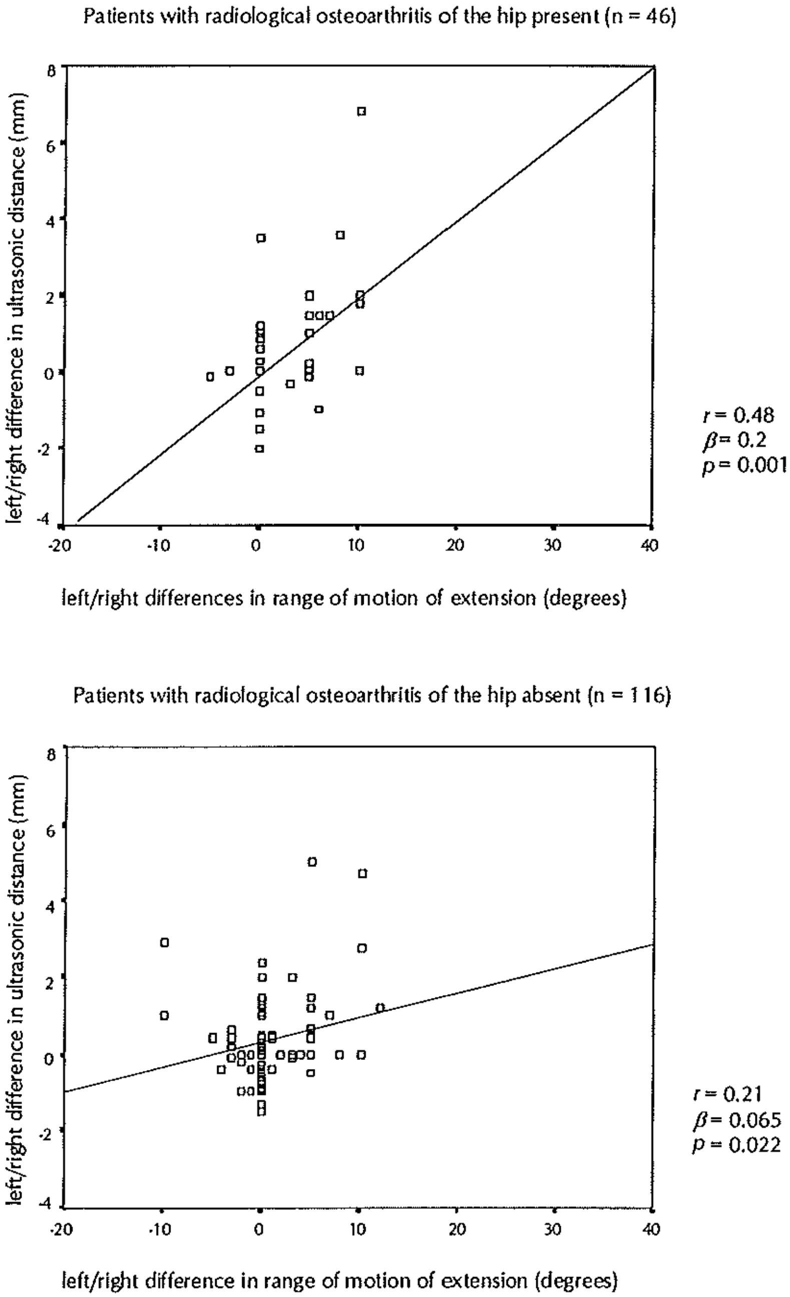

Figure 2
Linear relation (regression line) between left/right differences in range of motion of extension and left/right differences in ultrasonic distance, separately for patients with and without radiological osteoarthritis of the hip. Patients consulting for bilateral problems and with increased ESR are excluded.

Table 2
Distribution and predictive value of left/right difference in extension in unilateral hip patients (n = 163). Patients consulting for bilateral problems and with increased ESR are excluded.

		Left/right difference in ultrasonic distance		Total
		Absent	Present	
Left/right difference in extension	Absent	120	29	149
	Present	4	10	14
Total		124	39	163

Pre test probability of hip effusion = 24%
Predictive value of negative test: 80% ($CI_{95\%}$ = 0.79;0.86)
Predictive value of positive test: (post test probability) 71% ($CI_{95\%}$ = 0.44;0.98)
Likelyhood-ratio = 7.9

Koski for joint effusion the maximal percentage was 10%, and using the definition based on left/right differences it was 21%.

Linear regression analysis showed that of all side differences in hip movements as independent variables together in one model, only the side differences of extension was explanatory for left/right difference in ultrasonic distance (p <0.001). Other variables from medical history and physical examination added to the model showed no significant explanatory effect. Regression analysis separate for patients with and without radiological osteoarthritis showed that in the group with radiological osteoarthritis the relationship is stronger (Fig 2).

Table 2 shows the predictive values of a side difference in extension of more than 5° extension on unilateral effusion, as well as the pre- and post test probability on this effusion.

Discussion

Measuring the ultrasonic intra-capsular distance in a standardised manner has been shown to have a low intra-observer variability.[1] In the present study we measured the distance twice on each side in the same standardised manner; together with final approval of these data by an independent radiologist, we assume that we achieved reliable ultrasonic measurements.

Using the definition of Koski[1] for hip joint effusion, a relative high prevalence of joint effusion in hip patients from general practice was detected. When a definition based on intra-individual left/right differences in ultrasonic distance was used, fewer patients with effusion were found. However, in this latter group of patients, as in the group with major effusion, a stronger relation between joint effusion and clinical symptoms was shown. In healthy adults an ultrasonic distance exceeding 7 mm was found to be abnormal by Koski *et al.*[1] and by Kang *et al.*[10] In our study population, however, showed even a relative high percentage of patients (28%) effusion according to Koski's definition in the non-symptomatic hip. Additionally, a high correlation between the measurements of both sides was shown, even when patients consulting for bilateral pain were excluded. These inter-individual differences and intra-individual similarities do not seem to depend on differences in gender or bone-size. On the basis of the higher relation with clinical symptoms, we assume that hip joint effusion is better reflected by a definition based on intra-individual differences, especially in the case of unilateral problems. The strong relation between major bilateral effusion and an increased ESR might indicate a more systemic disorder. In these cases a definition based on intra-individual differences will not be suffice.

In our study no relationship was found between nocturnal pain or pain severity and joint effusion, in contrast to other reports in cases with increased intra-articular pressure.[7,11] This discrepancy may be explained by different study populations. In the studies by Foldes *et al.*[7] and Goddard *et al.*[11], the population consisted of pre-operative osteoarthritis patients. Our patients had a pain duration of maximal two years; moreover, not all patients in our study were osteoarthritis patients.

Especially decreased extension, but also internal rotation and flexion in our study showed a significant relationship with joint effusion independent from age and radiological osteoarthritis. This corresponds with the findings of Goddard et al.[11] and Eyring *et al.*[12] who, during the same movements reported an increase in intra-articular pressure, especially with extension and internal rotation. Earlier, Lloyd-Roberts[13] demonstrated that the hip capsule is tightest during extension, followed by internal rotation and abduction. These authors all assumed that increased intra-articular pressure causes a reflectory decreased motion in just these movements. Pearson *et al.*[14], studying patients with osteoarthritis of the hip, found that the initial loss of movement is always extension and internal rotation, followed by decreased flexion.

In the present study, several symptoms from medical history and physical examination had a relation with joint effusion as well as with radiological osteoarthritis. In several symptoms these relations existed independently from each other, indicating the diagnostic value of ultrasonic investigation in addition to radiological investigation. However, worst pain located at medial thigh and severe decreased extension showed a very high relationship with joint effusion but not with radiological osteoarthritis; therefore, these symptoms may be specific for hip joint effusion.

We also showed that in patients with unilateral hip problems, the examination of side difference in hip extension is useful to predict a 'unilateral' effusion. This test may be useful in general practice in cases where a referral for ultrasonic examination is inconvenient.

In contrast to Koski *et al.*[1] we found age and ultrasonic intra-capsular distance positively correlated, even in subjects with no radiographic signs of osteoarthritis. It is plausible that this relation is also due a degenerative process that is not yet visible on the X-ray. In these cases the effusion may be a preliminary sign of a degenerative process. Therefore, the prognosis of effusion in the hip joint in patients with early or less severe hip problems, as well as the role of effusion in selecting efficient therapy, should be further investigated. In patients with knee osteoarthritis, intra-articular injection with steroids was more effective if joint effusion was present.[15] Studies into the superior effect of non-steroidal anti-inflammatory drugs above pure analgetics in patients with knee osteoarthritis lacked power to explore the predictive therapeutic value of effusion by subgroup analysis.[16]

We conclude that our results indicate the need for more studies on ultrasonic examination of hip effusion in adults with hip pain in general practice, particularly because of the relation between effusion and clinical symptoms, and the relatively high prevalence. However, the prognostic and therapeutic relevance of hip effusion in these patients should be evaluated before ultrasonic examination of the hip joint, or a prediction of it by examining the hip extension, is generally recommended.

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8

DEGENERATIVE PUBIC SYMPHYSIS MAY CAUSE MERALGIA PARESTHETICA

Bierma-Zeinstra SMA, Ginai AZ, Prins A, Geleijnse JM, van den Berge JH, Bernsen RMD, Verhaar JAN, Bohnen AM. Degenerative pubic symphysis may cause meralgia paresthetica. *(Submitted)*

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Introduction

Meralgia paresthetica is a mononeuropathy involving the lateral femoral cutaneous nerve, a purely sensory nerve that innervates the anterolateral part of the thigh. It is a condition that can be confused with symptoms produced by entrapment of the upper lumbar nerve roots or with symptoms of hip diseases. The most common features of meralgia paresthetica are numbness, decreased sensation to pinprick, pain and burning of the anterolateral aspect of the thigh.[1,2] The syndrome starts most often in middle age.[2] The most specific feature is aggravation of the symptoms when the hip is extended, a well-defined area of sensory loss, and a lack of motor abnormalities.[3] A valuable test to aid in the diagnosis is an injection with local analgesics in the lateral femoral cutaneous nerve.[4]

The reasons for the onset of meralgia paresthetica are poorly understood and are often considered idiopathic.[5] Entrapment of the lateral femoral nerve at the inguinal ligament (just below the anterior iliac spine) is believed to be the major cause, especially because autopsy has revealed local demyelination of the nerve at that site.[6] At the same site, due to the human erect position, the nerve undergoes a sharp angulation; Stookey[7] believes this angulation to be the main contributing factor to the onset of meralgia paresthetica.

Although this syndrome is described as uncommon, a recent observational study of 224 elderly patients in primary care with pain in the hip region revealed that 4% suffered from this condition (chapter 5, this thesis). Also Jones[4] reported that the diagnosis was made in 7% of patients referred to a neurological clinic for the evaluation of leg discomfort. In the study of Bierma et al. (chapter 5, this thesis) a positive relationship was found between meralgia paresthetica and radiological degeneration of the pubic symphysis (RDSP); half of the patients with meralgia paresthetica showed a degenerative pubic symphysis, whereas only 20% of the patients with other hip disorders showed this feature. Earlier, a degenerative pubic symphysis was not considered to cause much discomfort and a relationship with meralgia paresthetica has never been reported. The present study explores the relationship between radiological degenerative pubic symphysis and meralgia paresthetica in another population using a different research design.

Methods

Population

The relationship between radiological degeneration of the pubic symphysis and meralgia paresthetica was investigated in middle-aged and elderly persons because of the higher prevalence of both features in these age groups. The cases were patients with meralgia paresthetica, aged 40 years and older, who underwent surgical release of the lateral femoral cutaneous nerve due to the meralgia paresthetica syndrome during the period 1993 to 1998 in St Clara Hospital, Rotterdam. The diagnosis in these patients was confirmed by an injection with local anaesthetics. Only patients for whom X-rays of the pelvis were available three years before or up to three years after the operation were included. The control group included persons who participated in a prospective follow-up study (Rotterdam study) in the open population of persons aged 55 years and over;[8] X-ray investigation of the pelvis under 1990-1993 was a routine examination in this population study. Controls and cases were matched for gender and age (4 controls for each case) as far as possible; cases of 60 years and younger were matched with controls of age 55 years, cases older than 60 were matched with controls with the same age.

Measurements

The X-rays of cases and controls were scored by one experienced radiologist and two physicians according to our standardised protocol for the presence of radiological degeneration of the pubic symphysis. These observers were blinded for the aim of the study and for each other's assessments. The standardised protocol comprised four items of pubic symphysis degeneration: osteophytes, discongruence, marginal irregularity, and sclerosis. When two of these four items were scored positive we defined this as radiological degeneration of pubic symphysis. Narrowing of the pubic symphysis was scored separately. The distance between the two pubic bones was measured (in mm) in the middle, and at the superior and the inferior edge, by two independent observers. The radiologist also noted the presence or absence of radiographic sacroiliacal and lumbosacral degeneration; the presence of radiological osteoarthritis of the hip was scored 0-4 on an ordinal scale according to the Kellgren assessment.[9] In women, the number of children delivered is known to be positively related to degeneration of the pubic symphysis.[10] Because the number of children delivered could be a mutual cause of both degeneration of the pubic symphysis and meralgia paresthetica and thereby bring about the relationship, a separate analysis was done for males only. Patient records in general practice of all controls were checked to ensure the absence of meralgia paresthetica during the previous ten years (1988-1998).

Statistical analysis

The relationship between radiological degeneration of the pubic symphysis and meralgia paresthetica was assessed with the chi-square test at 0.05 significance level, stratified for osteoarthritis of the hip and age (Mantel-Haenszel procedure). For this final analysis, radiological degeneration was defined as present when two of the three observers found radiological degeneration of the pubic symphysis. Radiological osteoarthritis of the hip was defined as Kellgren grade ≥ 2 at the left and/or right side.

To demonstrate a difference in distribution of degenerative pubic symphysis in 50% of the cases with meralgia paresthetica and in 20% of the controls without meralgia paresthetica, a minimum of 26 patients and 104 controls are needed ($\alpha = 0.05$ and power = 80%).

Results

During a five-year period (1993 to 1998) 103 patients underwent surgical release of the lateral femoral cutaneous nerve due to meralgia paresthetica. Of these 103 patients, 82 were older than 40 years; of this latter group an X-ray of the pelvis was available for only 25 patients of which 16 were female. For each case 4 controls, matched for age and gender, were included in the study. This resulted in a study

Table 1
Characteristics of the study population with the *p*-value for the differences in distribution between the cases and controls.

	Cases $N = 25$	Controls $N = 100$	*p*-value
age (mean value in years)	59	61	0.166[a]
Hip joint degeneration (*n*)	8	11	0.009[b]
Sacroiliacal degeneration (*n*)	7	32	0.861[b]
Lumbosacral degeneration (*n*)	4	42	0.031[b]

[a] by means of Chi-square
[b] by means of T-test

population of 125 persons; 25 cases and 100 controls. Controls matched with cases younger than 60 were all 55 years of age; this resulted in a slightly older control group. Radiological osteoarthritis of the hip was more common in the cases whereas radiological degeneration of the low back was more common in the control group. Data on characteristics of the two groups are given in Table 1. None of the controls had recorded symptoms of meralgia paresthetica in general practice during the past ten years.

The two physicians showed fair agreement (66% agreement, Kappa = 0.29) in recognising radiological degeneration of the pubic symphysis. The inter-observer variability between the radiologist and the two physicians was slightly better (66% agreement, Kappa = 0.30 and 73% agreement, Kappa = 0.40, respectively). The two physicians found 58 and 33 persons, respectively, with degeneration of the pubic symphysis, whereas the radiologist found 51 persons positive for radiological degeneration of the pubic symphysis. For the three observers separately a positive relationship was found between meralgia paresthetica and degeneration of the pubic symphysis ($p = 0.008$, $p = 0.001$, and $p = 0.004$). The final diagnosis 'radiological degeneration of the pubic symphysis' (scored positively by two or more observers) was present in 42 persons (16 cases and 26 controls).

The Mantel-Haenszel procedure stratified for age (age ≤ 60 years, age > 60 years) and for radiological osteoarthritis of the hip (Kellgren < 2, Kellgren ≥ 2) showed a significant positive relationship between meralgia paresthetica and radiological degenerative pubic symphysis (Table 2) with a compounded p-value of 0.004, and a weighted odds ratio of 4.38. Additional stratification for gender (the matching

Table 2
Presence of radiological degeneration of the pubic symphysis (RDPS) in cases with meralgia paresthetica and in controls without meralgia paresthetica stratified by radiological osteoarthritis (OA) of the hip and age.

	Strata	Cases RDSP		Controls RDSP	
		+	-	+	-
Radiological OA of the hip	age > 60 yrs	5	1	5	3
	age ≤ 60 yrs	1	1	2	1
No radiological OA of the hip	age > 60 yrs	3	1	10	22
	age ≤ 60 yrs	7	6	9	48

Compounded p-value by Mantel-Haenszel procedure = 0.004
+ = present
- = absent

for gender preserved[12]) resulted in empty cells in two strata (n=6). The Mantel-Haenszel procedure for the remaining six strata resulted in a weighted odds ratio of 4.28 with a *p*-value of 0.005.

Analysis limited to the study population without radiological osteoarthritis, stratified for age (age $\leq$ 60, age > 60), resulted in a weighted odds ratio of 6.32 with a *p*-value of 0.002.

Analysis for the relationship between radiological osteoarthritis of the hip and meralgia paresthetica, stratified for age (age $\leq$ 60 years, age > 60 years) and radiological degeneration of the pubic symphysis did not show a significant relationship (Mantel-Haenszel $p = 0.157$, weighted odds ratio 2.69).

An analysis that takes all the individual matched sets into account is conditional logistic regression. This analysis, with meralgia paresthetica as dependent variable, and as independent variables radiological osteoarthritis of the hip and radiological degenerative pubic symphysis, yielded similar results (odds ratio = 2.76, $p = 0.143$ and odds ratio = 4.58, $p = 0.006$, respectively).

Also separate analysis for males only ($n = 46$), conditional logistic regression with adjudgement for osteoarthritis, yielded similar result as for all patients (odds ratio = 9.2, $p = 0.05$), though the estimated odds ratio is less precise due to the lower number of observations.

Narrowing of the symphysis, in both the dichotomous scale and the numerical measurements, showed no relation with meralgia paresthetica; moreover there was no relationship with age or with radiological osteoarthritis of the hip.

Discussion

This study has shown a positive relationship, independent of age and radiological osteoarthritis of the hip, between degeneration of the pubic symphysis and the syndrome meralgia paresthetica. This relationship was also present in men, indicating that the relationship is not caused by the number of children delivered, i.e. a factor that could cause both entities.

Radiological assessment of the pubic symphysis showed only fair agreement between the observers. For this reason we defined degeneration of the symphysis as present only when two or more observers recognised this degeneration on the X-ray. However, separate analysis for all three observers revealed a significant relationship between radiological degeneration of the pubic symphysis and meralgia paresthetica, suggesting that the inter-observer variability has minor impact on the results of the study.

In this study, the cases were collected from a hospital population and the controls from the general population. We allowed our controls to derive from a population which would be referred to the hospital in case they developed the illness to the same extend as our cases. The hospital in which the cases were collected has a regional function in surgical treatment of this condition. For these reasons, we selected the references from the general population in the same region. Our cases may not be representative for all patients aged 40 years and older with meralgia paresthetica, because they represent a group in whom the symptoms indicate a need for intervention. Persons with mild symptoms of meralgia paresthetica are probably not included in the cases; the relationship found in our study may therefore only apply to persistent complaints. Further, because the selected cases already had an X-ray of the pelvic region available, co-morbidity may have been suspected. One of the pitfalls of such a selection could be an overrepresentation of the exposure in the cases if the X-ray request is related to the presence of degenerative pubic symphysis and we admit that it would be methodologically more correct to have X-rays of all patients. However, we assume that an X-ray request is mainly related to suspect osteoarthritis of the hip. Indeed, our cases presented with more osteoarthritis of the hip than the control group. Osteoarthritis of the hip has also been proposed as one of the possible causes of meralgia paresthetica.[12] However, separate analysis for the study population without radiological osteoarthritis showed a higher relationship between meralgia paresthetica and degenerative pubic symphysis. Furthermore, we showed that meralgia paresthetica had a non-significant and weaker relationship with osteoarthritis of the hip than with degeneration of the pubic symphysis, although patients with osteoarthritis of the hip probably are over-represented in our cases due to our selection method. We therefore assume that degenerative symphysis has a stronger independent relationship with meralgia paresthetica than osteoarthritis of the hip. On the basis of our results we can conclude that the relationship between meralgia paresthetica and degenerative pubic symphysis is not caused by hip osteoarthritis.

With respect to age we were not able to match all cases exactly to the controls resulting in a slightly older control group. However, a degeneration of the pubic symphysis is more common with increasing age.[10] Therefore, the age difference, still present in the younger stratum, probably only weakened the relationship.

Although the relationship between meralgia paresthetica and degenerative pubic symphysis has been shown both in the present study and in a former study using a different study population (chapter 5, this thesis), the physiological explanation is not obvious. Because the inguinal ligament is inserted on the pubic bone, this ligament might be 'tightened' by the degenerative process of the symphysis leading to the entrapment of the lateral femoral cutaneous nerve under this ligament. The position of the pubic bone might become less flexible, or muscular tension might be changed by the degenerative process. It is also reported that several muscles and fascial attachments to the inguinal ligament have a direct effect on its tension.[13] Occasionally, direct entrapment of the nerve by the iliac fascia, the tensor fascia latae muscle and the sartorius muscle is reported.[1,7,14]

Meralgia paresthetica has also been reported during and directly after pregnancy[14], often believed to relate to the changes in weight[2,15]; also in these cases, abnormalities in pubic symphysis may play a role.

Until now, degenerative changes of the pubic symphysis were not considered to cause much discomfort. However, the present study confirmed the positive relationship between degeneration of the pubic symphysis and meralgia paresthetica, indicating that such degenerative changes might influence surrounding and connected tissues. The presence of a degenerative pubic symphysis might be of importance for the prognosis and/or therapy choice in patients with meralgia paresthetica and should be addressed in future research.

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9

GENERAL DISCUSSION

The studies reported in this thesis focused on the classification and diagnosis of hip disorders in the middle-aged and elderly as seen in general practice. In this final chapter we describe our starting position, after critically reviewing the results, draw our main conclusions. We will reflect on the methodological problems encountered and give recommendations for future research.

The work presented in chapter 1 revealed that the medical literature provides no valid diagnostic criteria in general practice for middle aged and elderly persons with hip disorders. Diagnostic criteria, and thereby classification into separate diagnostic groups, often form the basis for studies on treatment in these separate diagnostic groups. Therefore, we were not surprised by the large variation between general practitioners (as reported in chapter 2) in the diagnosis, referral and treatment of middle-aged and elderly hip patients in primary care. In chapter 3 we showed that the American College of Rheumatology (ACR) classification criteria for osteoarthritis of the hip, which were introduced for use in research, are not valid in primary care patients.

The lack of diagnostic criteria led us to the root of clinical practice, namely the disease classification. Diagnosis is established when the physician makes a choice among a series of classes.[1] A disease classification has to be realised in order to establish diagnostic criteria. Back in the 1600s, Sydenham introduced a medical classification system, inspired by zoology and botany;[1] observation and description of symptoms provided the features for discrimination of different diseases. In the 1700s de Sauvage also provided a classification of diseases ordered in a botanical manner (classes, orders, genera, species). In the subsequent decades structural and physiological signs that were assumed to indicate causes replaced symptoms as privileged diagnostic categories[1]. In medicine nowadays there is a wide variety of diagnostic categories based on physiology, pathophysiology, nosology, functional complaints, symptom diagnoses and problem behaviour[2].

King[1] emphasised the purpose of the classification systems by asking: "for whom is this classification useful and under what condition?" A classification of hip disorders useful for a general practitioner should categorise reasonable and logical entities that are recognisable in the general practice setting. Furthermore, it should of course be the basis for prognosis, and subsequently, suitable and adequate therapy. In general practice a symptom-based (syndrome) classification may be advantageous to a classification based on e.g. radiological signs of osteoarthritis. This is reflected by the example of a clinical active tendinitis in a patient with radiological, but no clinical signs of osteoarthritis. Treatment of the clinically active syndrome should be the first concern of the general practitioner. Thus, we consider the detection of separate syndromes as the first step in a classification useful for a general practitioner. Also for purposes of medical research in the domain of a complaint (e.g. 'pain in the hip'), the first-order of concern should be to enable diagnosis of the underlying condition. It is essential to operationalise the potential underlying disorder as focal entity for the study of aetiology, clinical course and intervention effect.[3]

Chapter 1 in this thesis revealed that, in general practice, hip disorders with a specific pathognomonic sign are rare and that in clinical practice we are mostly confronted with clinical syndromes. A classification of clinical syndromes depends on the perception of similarity. Such classifications are often derived from literature reviews and/or the classification designer's own judgement.[4] To realise a symptom-based classification in a more experimental way, two conceptually different methods are available. The first method is based on a consensus of 'experts' and the second is a mathematical method. In the consensus method 'experts' decide whether patients show similar symptoms, or show symptoms similar to the syndromes that they know on an empirical basis. An advantage of the first method is that the use of empirical knowledge may increase the acceptance of such a classification. However, a serious problem with this method is the circularity of reasoning. In their 'sorting' process experts will give extra weight to those symptoms which they already believe to be important. If diagnostic criteria are developed from such classifications, especially these symptoms will appear to be important (discriminating). Felson and Andersson[5] stated that this could be avoided when the group of clinicians who decide on the basis of which 'important' symptoms the patients should be classified, are separate from the group of clinicians who actually sort the patients. In our opinion, however, the circularity of reasoning in this case still exists; both groups of clinicians will use similar empirical knowledge. As long as the diagnosis is made by clinicians and/or 'experts' the classification will be no more than a consensus of what we already think we know.

The second method for classifying patients is a mathematical method, also known as numerical taxonomy. Cluster analysis, the classification method used in phenetic numerical taxonomy, sorts cases into classes or clusters on the basis of similarity or dissimilarity in a set of symptoms that are collected in all the cases[6-8]. After being widely used in the natural science, numerical taxonomy had a limited introduction into medical science. The international DSM-III classification system of psychological syndromes is largely based on numerical classification[8]. More recently, cluster analysis was also used to classify facial pain syndromes,[10,11] shoulder syndromes[12,13] and non-specific low back pain syndromes.[14] Because the similarity between patients on the basis of symptoms is determined by the cluster analysis, the circularity of (human) reasoning is probably avoided. This was our rationale to use cluster analysis to explore the classification of hip disorders in the middle-aged and elderly (chapter 5).

Although cluster analysis is an objective method, during the classification process it was shown to be less straightforward than it first appeared. The method involves many choices, each with consequences for the final classification result. A major difficulty lies in choosing one specific clustering method from the many available, and in the choice of which similarity or distance measure to use. No technique can be judged to be 'best' in all circumstances.[7] For medical classification it seemed to us more reasonable to select an agglomerative method for clustering rather than a divisive method. When 'sorting' the patient, the agglomerative method allows the total clinical picture of the patient in relation to the others to be taken into account. From the agglomerative methods available we selected Ward's method with the Euclidean distance matrix because it is known to provide a clear cluster structure.[11] In previous classification research on musculoskeletal disorders[13,14] this method showed stable results. However, because this method is known to create groups more easily than others, stability has to be tested.[6] The use of several techniques should help to prevent misleading results being accepted. Therefore we also used an alternative cluster method, the complete linkage algorithm, to test the stability of the obtained classification.

The choice of variables is also crucial for the generation of a classification system; the variables should, of course, be relevant to the purpose of the classification. As we aimed at a classification of hip syndromes we included only the symptoms from medical history and local physical examination of the hip region. To obtain an objective as possible classification, free from preconceptions, we included all these symptoms instead of only the pre-assumed important ones.

Because we used variables that employ different scales, the outcomes had to be standardised, which was achieved by converting outcomes to z-scores. However, because this standardisation tends to over-weight the rare outcomes and down-weight the common outcomes, we excluded symptoms, which are very rare (less than 5% present in the total sample).

The next decision concerned the number of clusters; here there is a trade-off between clinical detail and reproducibility. When the clusters become smaller, the chance of coincidental effects becomes larger. Milligan and Cooper[15] described a procedure for determining the number of clusters. They recommended that the number of clusters should not exceed the square root of the number of variables used for the classification and the square root of the number of patients used for the classification. If we had applied this rule the maximum number of groups (clusters) would have been eight. This number of clusters would have been achieved when we

allowed one cluster step more; the two clusters with soft-tissue problems would have been combined to one cluster, with the result that the already largest cluster would have been enlarged. To avoid loss of clinical detail we did not allow this before we had tested the reproducibility. Specifically these 'soft-tissue' clusters, although better results were reached in their subgroups, showed the lowest stability of all clusters. This may indicate that a clear distinction within local (lateral) soft-tissue disorders may be difficult. One could also wonder whether prognosis or therapy within this group would differ to the extent that differentiation is indicated. This should, in our opinion, be the most important indicator for the number of groups.
Buchbinder et al.[4] made a critical appraisal of existing classifications of soft-tissue disorders of the neck and upper limbs using methodological criteria including purpose, validity, reliability, feasibility, and generalisability (Table 1). They found that these classifications, of which the ICD-9[16] showed the lowest score, did not meet the standards of face validity, content validity, and feasibility. Construct validity and reliability could not be assessed at all because these studies had not been performed or reported.

Applying these standards to our classification we consider that the purpose of our classification, as well as the population and setting, are satisfactorily described.

Concerning the content validity, we specified the inclusion and exclusion criteria for the domain, and intended to include the relevant categories (i.e. middle-aged and elderly patients with benign hip problems, which might become chronic). In our classification the categories are mutually exclusive. However, whether or not the breakdown of the categories satisfies our purpose has to be shown in future research. Because we are confronted with clinical syndromes without a gold standard, we consider the method of development by identifying groups of patients with similar symptoms acceptable. Cluster analysis (with all its drawbacks) is the (only) method allowing objective classification on the grounds of symptoms. Our classification solution was tested for validity by comparing the obtained clusters for variables not used to generate the classification. For almost all groups in our classification, significant differences in distribution of these variables (variables from radiological, sonographic, low back and knee examination) were found. In addition, our classification produced a previously unknown relationship between radiological degeneration of the symphysis pubis and meralgia paresthetica. Because we also found this relationship in a different independent study population (chapter 8) we preclude coincidental effect and take this as an additional validation.

To meet the criteria of face validity, the nomenclature to label the categories should be satisfactory. Most groups in our classification received a similar label from each of the 20 experts; these labels were also similar to ours. We did not create diagnostic criteria for each separate group, but rather a diagnostic function, which shows a probability for all groups at once. If required, criteria for each separate group are easy to create. The outcomes of variables are clearly defined, whereby the definitions of the criteria are specified.

The feasibility is considered satisfactory because the classification is easy to understand, relies on clinical examination alone, is easy for general practitioners to perform, and requires no special skills or training. Furthermore, data collection can be quickly and efficiently performed (chapter 6).

That a classification discriminates between entities that are considered to be different in a way appropriate to the purpose, is an important issue for construct

Table 1
Items used in the methodological scoring list for critical appraisal of classification systems (from Buchbinder et al[4]).

Purpose
- Is the purpose, population, and setting clearly specified?

Content validity
- Is the domain and all specific exclusions from this domain clearly specified?
- Are all relevant categories included?
- Was the method of development appropriate?
- If multi-axial, are criteria of content validity satisfied for each additional axis?

Face validity
- Is the nomenclature used to label the categories satisfactory?
- Are the items used based upon empirical (i.e. directly observable) evidence?
- Are the criteria for determining inclusion into each category clearly specified?
  - If yes, do these criteria appear reasonable?
- Have the criteria been demonstrated to have validity and/or reliability?
- Are the definitions of criteria clearly specified?
- If multi-axial, are criteria of face validity satisfied for each additional axis?

Feasibility
- Is the classification simple to understand?
- Is the classification easy to perform?
- Does it rely on clinical examination alone?
- Are special skills, tools, and/or training required?
- How long does it take to perform?

Construct validity
- Does it discriminate between entities that are thought to be different in a way appropriate to the purpose?
- Does it perform satisfactorily when compared to other classification systems which classify the same domain?

Reliability
- Does the classification system provide consistent results when classifying the same conditions (test-retest)?
- Is the intraobserver and interobserver reliability satisfactory?

Generalizability
- Has it been used in other studies and/or setting?

validity. Our ultimate aim is to create a classification with which the general practitioner can predict the prognosis or outcome of specific treatment. For this purpose we consider a classification by syndromes, attributed to specific underlying conditions, as the most reasonable. Most of the separate groups in our classification could be recognised as syndromes described in medical literature. It is therefore not surprising that it also shows, albeit more detailed, similarities with the ICPC classification[17] for hip complaints in primary care. The reliability of the classification was tested in ten 75% sub-samples and was found to be satisfactory. All variables were examined in a standardised manner, but only some facets of physical examination were tested for intra- and interobserver variability. However, the physical examination performed, is included in the standard physical examination of the musculoskeletal system conducted by every physician.

With respect to generalisability of our classification, some concerns have to be discussed. The patients in the classification study were referred for X-ray investigation by the general practitioner; this may have resulted in a specific selection of the hip patients in our study population. Theoretically, disorders for which the general practitioner never requests an X-ray are not represented in this classification. However, as shown in a previous study (chapter 3) the attitude of the general practitioner largely determines a referral to the radiological department, and in any group of hip patients some always receive an X-ray investigation. It was also shown that patients in whom the GP had diagnosed a soft-tissue disorder or osteoarthritis of the hip, were less often referred to the radiological department than patients with unspecified hip problems. Patients in whom the GP had difficulties with the diagnosis may be over-represented; this may explain the large cluster with unclear symptoms (cluster 3) and the relatively small cluster with very clear symptoms of trochanteric tendinitis/bursitis (cluster 8). Looking at the separate clusters and their symptoms, it appears that soft-tissue disorders, neurological disorders, joint disorders, and disorders causing secondary pain in the hip are all represented. The distribution of these syndromes in our population is similar to the distribution of disorders in a randomly selected population of middle-aged and elderly hip patients (chapter 2) and with the distribution in the National Survey of Morbidity and Interventions in General Practice.[18] We therefore assume that no major selection bias was caused in our study by including only those hip patients who received X-ray investigation. If patients in whom the GP had difficulty in reaching a diagnosis are slightly over-represented, this should not be a problem as long as diagnostically 'clear' cases are also included in the classification. Our goal was not to investigate incidence and prevalence of the different disorders, but to create a classification of the different disorders seen in general practice.

Felson and Andersson[4] stated that the ability of a classification to predict long-term outcomes is critical for the validation of a classification. To enable the general practitioner to predict outcome in middle-aged and elderly hip patients is, besides predicting outcome of treatment in the separate diagnostic groups, also our ultimate goal. The predictive value of the obtained symptom-based classification with respect to prognosis will be investigated in future studies. The predictive value of this classification has to be compared with the predictive value of radiological and sonographic signs, and of course with single symptoms. Especially the prognostic value of hip joint effusion, detected by sonography, is totally unexplored and because of the relation with clinical symptoms (chapter 7) it is a very interesting one. Furthermore, outcomes of specific treatment in the different diagnostic groups within the obtained classification need to be investigated. Although treatment by the general practitioners may not differ on the grounds of radiological signs, a sonographic sign of hip joint effusion may support anti-inflammatory treatment; this also needs additional study.

Finally, in our opinion, future intervention research on chronic disabling hip disorders should not only focus on pain management, but also on methods to improve the functional status of these patients as has been done in a recent study.[19] However, combined forces are needed to create and agree on diagnostic criteria for research purposes in primary care. This could lead to more collective experience and subsequently evidence-based guidelines for managing adults with hip pain in primary care.

Concluding this discussion, there is a lack of validated criteria for diagnosing hip problems in adults in general practice, both for clinical use and for research purposes. A new symptom-based classification of hip disorders, together with a diagnostic function, was developed and proved valid in several ways. An additional important symptom-related distinction in the actual clinical status of the patient is the presence of joint effusion detected by sonography. Implications for clinical practice are not yet fully elucidated, but are expected in the near future. The first important step, namely an attempt to classification, has been taken and will be the basis of research on the usefulness and applicability of this classification for the general practitioner in managing patients with hip disorders.

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SUMMARY

Hip problems are increasingly seen in general practice due to the ageing population, and represent a major cause of chronic pain and disability. This thesis describes the problems general practitioners experience in finding and using valid criteria to diagnose the various hip problems presented in general practice. It also presents the construction of new diagnostic criteria based on cluster analysis of patients with hip pain seen in general practice. Additionally, two relatively unexplored features in patients from general practice with hip pain, namely ultrasonic hip joint effusion and radiological degeneration of the pubic symphysis, are further investigated.

Chapter 1 presents an overview of studies that describe syndromes and available diagnostic criteria of hip disorders in adults seen in general practice. A Medline search was conducted and selected articles were screened and reviewed.

The review showed that few studies are performed to diagnose and distinguish the separate disorders. Most available studies are descriptive ones based on case reports. In studies describing pure clinical syndromes, the inclusion criteria are seldom clearly specified. In syndromes where imaging techniques may allow determining the diagnosis, these radiological/imaging signs often serve as inclusion criteria for the cases. These techniques are, however, not readily available in general practice. Only one study was found which was conducted (partly) in primary care.

Although many descriptions of symptoms in hip disorders were found, our analysis of literature showed an important lack of reliable studies of diagnostic criteria for hip disorders for use in adult patients in primary care.

Chapter 2 addresses the actual medical management of hip disorders in adults in general practice as well as the consistency and determinants of this management. Additionally, the medical management of patients with osteoarthritis was compared with available recommendations.

In this investigation, physical examination, diagnosis and treatment of hip patients by 20 general practitioners (GPs) were analysed in two different manners. First, four 'paper patients' based on existing patients were used. Second, computerised patient records of 400 patients (20 per GP) with new hip problems were assessed.

Use of 'paper patients' showed that medical history taking consisted mainly of questions concerning the localisation and onset of pain, and physical examination focused mainly on checking passive hip motion. It was also found that the paper patient with alarming symptoms received the most consistent management; for the other three patients, variation in management between GPs was high, particularly for the prescription of medication and for X-ray requests. The 400 computerised patient records showed the same high variation between GPs in prescription of medication and X-ray requests. During the two-year follow-up a specific diagnosis was registered for only one third of the 400 patients. Main factors influencing medical management were the age of the patient, number of consultations, and the attitude of each individual GP. The proportion of patients who were diagnosed with osteoarthritis varied highly between GPs. Patients who had received the diagnosis osteoarthritis were more often prescribed NSAIDs than paracetamol (the latter are recommended); they also received less referrals for physical therapy than patients without a specific diagnosis.

We conclude that there is no consistent medical management by GPs in adult patients with hip problems. Diagnosing of hip disorders varies widely between GPs, and the medical treatment of patients with osteoarthritis is not consistent with published recommendations. These conclusions underline the need for widely accepted and evidence-based guidelines for diagnosis and treatment of adult patients with hip problems in primary care.

Chapter 3 presents a study on the validity of the ACR criteria for hip osteoarthritis, developed by the American College of Rheumatology, in hip patients from primary care. Three different sets of criteria are available, one set with pure clinical symptoms, two sets with clinical symptoms and radiological and/or laboratory signs combined. It is claimed that all three sets, separately from each other, can be used to diagnose hip osteoarthritis for research purposes.

Consecutive patients (n = 227) consulting for pain in the hip in general practice and who had been referred for X-ray investigation of the hip were recruited for a standardised history taking and physical investigation. The radiographs taken were evaluated according to a standardised protocol. The patients were classified on the basis of the three separate sets and the cross-validity of the three different ACR criteria sets was assessed by calculating the percentage agreement and the Kappa between the separate sets. Various cut-off points for joint-space narrowing were used.

There was low agreement between the set based on clinical criteria only and the two sets where radiological signs were included (Kappa 0.1 or lower). The two sets with radiological signs included showed high mutual agreement (Kappa 0.81–0.94, depending on cut-off points for joint-space narrowing). These sets also showed the highest agreement with the radiological osteoarthritis defined as Kellgren score 2 or more (Kappa 0.13 – 0.48).

We conclude that the clinical ACR criteria show poor cross-validity with the two other ACR criteria sets. For research in primary care the ACR criteria, at least one or more of them, are not valid. Further efforts are needed to establish valid criteria for hip patients in primary care.

Limitation of hip joint motion seems to be an important sign of hip disease. Therefore *chapter 4* presents a comparison between the reliability of measurements of hip motions obtained with two instruments: an electronic inclinometer and a two-arm goniometer. It was also investigated whether the two instruments, and different patients' positions, would produce the same measurement results.

Maximal active and passive hip movements were measured simultaneously with both instruments, in nine subjects during 10 consecutive measurements at short intervals.

Intra-observer variability was lower with the inclinometer in measurements of passive hip rotations. The two instruments showed equal intra-observer variability for hip movements in general. The inclinometer also showed lower inter-observer variability in the measurements of active internal rotation. A higher range of rotational movement was measured with the two-arm goniometer and more extension and flexion with the inclinometer. Also, more rotational movement was found in the prone position compared with sitting and supine positions.

The inclinometer was more reliable in measurements of hip rotation. For hip movements in general the two-arm goniometer is just as accurate when used by only one observer. The two instruments, and some positions, are not interchangeable during repeated measurements.

Chapter 5 explores whether it is possible to obtain a valid classification scheme of hip problems in middle-aged and patients seen in primary care with the method of numerical classification. In numerical classification, the patients are grouped by mathematical algorithms on the basis of similarity or dissimilarity in symptoms.

Patients (*n* = 224) aged 50 years or older, who had consulted the general practitioner for pain in the hip region, and had been referred for X-ray investigation of the hip, underwent a standardised history taking and physical examination of low back, hip and knee. In addition, a sonographic examination of the hip region was performed and the one-hour erythrocyte sedimentation rate was determined. To obtain a classification scheme, the patients were grouped into clusters with cluster analysis (Ward method). In this cluster analysis the variables from the patient's history and physical examination of the hip region on the (most) painful side were used (65 variables).

The cluster analysis resulted in a classification with nine different clusters. These clusters could be reproduced in the 10 sub-samples and with the alternative clustering method (complete linkage method). Significant relationships ($p < 0.05$) of various radiological and sonographic signs with the separate clusters were found; this was also the case for variables of low-back and knee examination. A group of 20 experts could recognise the symptoms in seven of the nine clusters as known syndromes seen in clinical practice. Seven clusters showed symptoms similar to syndromes described in literature, namely: moderate or severe osteoarthritis of the hip, early stage osteoarthritis of the hip, trochanteric tendinitis, meralgia paresthetica, low-back problems, a combination of low-back problems and osteoarthritis of the hip, and knee problems in combination with lateral thigh symptoms. Two clusters were more difficult to identify; a cluster with no local hip symptoms apart from pain, and a cluster with suspected soft-tissue problems. Further study of these groups of patients may help to clarify the situation

We achieved a symptom-based classification of patients with hip problems in general practice, which was reproducible in sub-samples and showed a relationship with radiological and sonographic signs. Most of the clusters in the classification showed similarity with the various hip disorders described in literature and were as such recognised by experts. However, a classification not leading to discrimination in prognosis or discrimination in effectiveness of specific treatment has no clinical importance. This should be tested in future research.

Chapter 6 presents the development of a diagnostic function for hip disorders in middle-aged and elderly patients (based on the classification described in chapter 5), in order to classify an individual patient into the right cluster. We aimed at a function with as few as possible variables that are easy to examine by the general practitioner.

Applying discriminant analysis (Wilks method) on the same 224 patients used for the classification, the most discriminating variables (of the 65 available) related to the separate clusters were identified. With these variables a diagnostic function, which can predict cluster membership in individual patients, was developed. Diagnostic functions, ranging from 5 variables to 50 variables were developed and the percentage of patients placed into the 'right' cluster was assessed.

It was shown that with the use of only 20 variables, 80% of the patients was still placed into the 'right' cluster as in the original classification. Half of these 20 variables were derived by medical history taking, with the exact pain location being the most important variable.

We used the same patients for the classification study that were used for testing the diagnostic function, which may give better results than in new patients. If the clinical importance of the obtained classification can be shown in future research, it is feasible to introduce this new classification in clinical practice without forcing clinicians to examine all 65 variables in patients with hip problems.

In *chapter 7* the prevalence of ultrasonic hip joint effusion and its relation with physical symptoms, radiological signs and laboratory results was investigated in the same patient population used in the studies presented in chapters 5 en 6 (n = 224).

The standardised ultrasonic examination of the hip joint included measurement of the distance between the ventral capsule and the femoral collum, an increase of which represents a joint effusion. Hip joint effusion was defined in three ways: 1) 'effusion' according to Koski's definition, defined as an ultrasonic distance of ≥ 7 mm or left/right difference of ≥ 1 mm, 2) 'major effusion', i.e. an ultrasonic distance of ≥ 9 mm, and 3) 'unilateral effusion', based on only left/right differences of ≥ 1 mm.

Effusion (Koski's definition) of the (most) symptomatic hip was present in 80 (37%) patients, a major effusion was present in 20 (11%) patients. Unilateral effusion was found in 47 (22%) patients. Adjusted for age and radiological osteoarthritis, the following variables showed significant positive relationship with hip joint effusion: pain located in groin or medial thigh, pain aggravation by lying on the side, positive Trendelenburg sign, decreased hip extension, abduction, internal rotation or flexion, painful external rotation, and pain on palpation of the groin. Only major joint effusion showed significant positive relationship with an increased one-hour erythrocyte sedimentation rate (ESR). If patients with bilateral pain and increased

ESR were excluded, a side difference in the range of motion of hip extension showed to be a good predictive test for unilateral effusion (predictive value for a positive and a negative test 71% and 80%, respectively).

In our study population, the high prevalence of hip joint effusion detected by sonography, and the relation with clinical symptoms and signs, underline the need for more studies on the therapeutic and prognostic relevance of hip effusion in adults with hip pain in general practice.

Chapter 8 describes a case-control study in which the relationship between meralgia paresthetica and radiological degenerative signs of pubic symphysis is explored. This previously unknown relationship was shown in the classification obtained by cluster analysis described in chapter 5. The relationship was tested again in another study population using a different research method.

In this case-control study the cases were patients with meralgia paresthetica aged 40 years and older who underwent surgical release of the lateral femoral cutaneous nerve; cases were only included when an X-ray of the pelvis was available. The control group was derived from a population study in persons aged 55 years and older. The controls were matched to the cases (4 references per case) for gender and for age as far as possible. The patients records in general practice were checked to ensure that persons from the control group had not had symptoms of meralgia paresthetica during the previous ten years. Radiological degeneration of the pubic symphysis was defined as present when two of three independent observers verified this degeneration on the X-ray.

The Mantel-Haenszel procedure (strata for age and radiological osteoarthritis of the hip) showed a positive relationship ($p = 0.004$, odds ratio = 4.38) between radiological degeneration of the pubic symphysis and meralgia paresthetica. A separate analysis for men only also confirmed this positive relationship.

In this study population the previous unknown relationship between meralgia paresthetica and radiographic degenerative pubic symphysis was confirmed. Degenerative changes in the pubic symphysis may influence surrounding and connected tissues or anatomic relations, thereby causing meralgia paresthetica.

Chapter 9 comprehends a general discussion and the main conclusion of this thesis. It describes the methodological problems in classification studies when no gold standard is available. The method of numerical classification, presented as a objective method which avoids circularity of reasoning and used for the study in chapter 5, showed to be a method involving many choices, each with consequences for the final classification results. The rationale behind the choices that were made is discussed. Additionally, the classification obtained by numerical methods for medical classification (cluster analysis) is appraised based on criteria for purpose, validity, reliability, feasibility, and generalisability. This classification was found to meet most of these criteria. However, because the ability to predict prognosis or therapeutic effect is not yet established, the clinical relevance of this classification in general practice requires further investigation. This applies also for the presence of (ultrasonic detected) hip joint effusion in patients from general practice.

SAMENVATTING

Door het verouderen van de bevolking komen heupproblemen in de huisartspraktijk in toenemende mate voor en vormen bij ouderen een belangrijke oorzaak van chronische pijn en handicap. Dit proefschrift beschrijft de problemen bij het stellen van de diagnose bij patiënten met heupklachten in de huisartspraktijk. Tevens wordt in dit proefschrift de constructie van nieuwe diagnostische criteria, gebaseerd op clusteranalyse van heuppatiënten afkomstig uit de huisartspraktijk, beschreven. Als aanvulling worden twee nog weinig bestudeerde aspecten bij patiënten met heupklachten in de huisartspraktijk verder onderzocht, namelijk een echografisch zichtbare gewrichtszwelling van de heup en een radiologische degeneratie van de symphysis pubica.

Hoofdstuk 1 bevat een overzicht van studies die de syndromen en de beschikbare diagnostische criteria beschrijven van heupaandoeningen bij volwassenen in de huisartsenpraktijk.

Het bleek dat weinig studies zijn verschenen waarin is getracht diagnostische criteria te construeren voor heupaandoeningen. De meeste studies waren beschrijvende studies van patiëntenseries. Het was vaak onduidelijk hoe de betreffende patiënten werden geselecteerd. Bij aandoeningen waar met behulp van radiologische technieken de diagnose bepaald kan worden, dienden deze radiologisch vastgestelde kenmerken vaak als inclusiecriteria voor de beschreven patiënten. Deze radiologische technieken zijn echter voor de huisarts niet direct toegankelijk. Slechts één studie was (gedeeltelijk) uitgevoerd in de huisartsenpraktijk.

Dit hoofdstuk liet zien dat er belangrijke hiaten bestaan in de kennis over betrouwbare diagnostische criteria voor heupaandoeningen bij volwassenen in de huisartsenpraktijk.

Hoofdstuk 2 beschrijft het beleid van de huisarts bij volwassenen met heupklachten en de consistentie en determinanten van dit beleid. Tevens wordt het beleid bij patiënten met coxartrose vergeleken met aanbevelingen uit de literatuur. Voor dit onderzoek zijn het lichamelijk onderzoek, de diagnose en de behandeling van heuppatiënten door 20 verschillende huisartsen onderzocht op twee verschillende manieren: aan de hand van 4 'papieren patiënten' en aan de hand van gecomputeriseerde medische dossiers van 400 nieuwe heuppatiënten (20 per huisarts).

Uit het onderzoek met de papieren patiënten bleek dat de anamnese voornamelijk bestond uit vragen naar de lokalisatie en het ontstaan van de pijn. Het lichamelijk onderzoek was voornamelijk gericht op het passieve bewegingsonderzoek. De 'papieren patiënt' met alarmerende symptomen onderging het meest consistente beleid. Bij de drie andere 'papieren patiënten' varieerde het beleid sterk tussen de verschillende huisartsen, met name ten aanzien van het voorschrijven van pijnstillende medicatie en het aanvragen van röntgenonderzoek. De gecomputeriseerde medische dossiers lieten dezelfde hoge variatie zien tussen huisartsen met betrekking tot het voorschrijven van medicatie en het aanvragen van röntgenonderzoek. In slechts een derde van de gecomputeriseerde medische dossiers van patiënten met heupklachten was gedurende een vervolgperiode van twee jaar een specifieke diagnose genoteerd. Belangrijke factoren die het beleid beïnvloeden waren het aantal consulten, de leeftijd van de patiënt, en de huisarts zelf. Het aantal patiënten die van de huisarts de diagnose coxartrose kregen varieerde sterk tussen de verschillende huisartsen. Patiënten met de diagnose coxartrose kregen vaker NSAID's voorgeschreven dan paracetamol (het laatste middel is aanbevolen als eerste keuze); deze patiënten werden tevens minder vaak naar de fysiotherapeut verwezen dan patiënten met een niet gespecificeerde diagnose.

We concludeerden uit dit onderzoek dat er geen consistent beleid is bij volwassen patiënten met heupklachten. Het stellen van een diagnose varieert sterk tussen huisartsen en de behandeling van patiënten met coxartrose komt niet overeen met aanbevelingen in de literatuur. Deze conclusies onderstrepen de behoefte aan geaccepteerde en op onderzoek gebaseerde richtlijnen voor diagnose en behandeling van patiënten met heupklachten in de huisartsenpraktijk.

In *hoofdstuk 3* werd de validiteit van de ACR-criteria voor coxartrose, ontwikkeld door het American College of Rheumatology, onderzocht in heuppatiënten in de eerstelijnsgezondheidszorg. Drie verschillende combinaties van criteria zijn beschikbaar, één met alleen klinische symptomen en twee waarbij ook radiologische symptomen zijn betrokken. Er wordt gesteld dat deze drie combinaties van criteria, onafhankelijk van elkaar, gebruikt kunnen worden voor het stellen van de diagnose coxartrose voor onderzoeksdoeleinden.

Patiënten die de huisarts consulteerden voor heupklachten en werden doorgestuurd voor radiologisch onderzoek (*n* = 227) werd gevraagd om een gestandaardiseerde anamnese en lichamelijk onderzoek te ondergaan. Ook de röntgenfoto's werden op een gestandaardiseerde manier geëvalueerd. De patiënten werden volgens de drie verschillende combinaties van criteria geclassificeerd en de 'kruisvaliditeit' van de drie verschillende combinaties van criteria onderling werd onderzocht door het percentage overeenstemming en de Kappa te berekenen.

Een zeer geringe overeenkomst werd gevonden tussen de combinatie van klinische criteria en de twee combinaties waarbij ook radiologische symptomen waren betrokken (Kappa 0.1 of lager). Deze twee laatste combinaties van criteria gaven onderling wel een goede overeenkomst (Kappa varieerde van 0.81 tot 0.94 afhankelijk van welk afkappunt werd genomen voor gewrichtsspleetvernauwing). Deze combinaties gaven ook de grootste overeenkomst met een uitsluitend radiologische diagnose van coxartrose, gedefinieerd als een Kellgren score van 2 of meer (Kappa 0.13-0.48).

We concludeerden dat de klinische ACR criteria een slechte 'kruisvaliditeit' vertonen met de twee andere ACR criteria combinaties. Voor onderzoeksdoeleinden in de eerstelijnsgezondheidszorg zijn de ACR criteria (één of meer combinaties) niet betrouwbaar.

Bewegingsbeperkingen van de heup zijn belangrijke symptomen van heupaandoeningen. In *hoofdstuk 4* is daarom nagegaan welke van de twee beschikbare instrumenten om bewegingsbeperkingen vast te leggen, de goniometer en de inclinometer (elektronisch), de meest reproduceerbare resultaten oplevert. Tevens werd nagegaan of de twee instrumenten en verschillende uitgangshoudingen van de patiënt dezelfde meetresultaten geven. Maximale bewegingsuitslag tijdens actief en passief bewegingsonderzoek van de heup werd simultaan gemeten met beide instrumenten bij negen proefpersonen tijdens 10 herhaalde bewegingen met korte tussenpozen.

Gemiddeld vertoonden beide instrumenten dezelfde intra-beoordelaarsvariabiliteit. De intra-beoordelaarsvariabiliteit was echter lager met de inclinometer bij de metingen van passieve heuprotaties. De inclinometer vertoonde tevens een lagere inter-beoordelaarsvariabiliteit bij de metingen van actieve endorotatie.

Er werd een hogere bewegingsuitslag van de rotaties gemeten met de goniometer. De inclinometer liet echter een hogere bewegingsuitslag van extensie en flexie zien. Liggend op de buik werd een hogere bewegingsuitslag van de rotaties gemeten ten opzichte de uitgangshoudingen liggend op de rug en zittend.

Dit onderzoek leidde tot de conclusie dat de inclinometer beter reproduceerbare metingen van heuprotaties oplevert. Voor metingen van heupbewegingen in het algemeen is de goniometer even reproduceerbaar wanneer uitgevoerd door één beoordelaar. De twee instrumenten, en sommige uitgangshoudingen, zijn niet uitwisselbaar bij herhaalde metingen.

In *hoofdstuk 5* werd nagegaan of het mogelijk is een valide classificatieschema te verkrijgen van heupklachten bij patiënten in de huisartspraktijk met behulp van de numerieke classificatietechniek. Bij deze wiskundige techniek worden onderzoekseenheden (in dit geval patiënten) gegroepeerd door mathematische algoritmen op grond van gelijkheid in kenmerken, in ons geval symptomen.

Patiënten (n = 224) van 50 jaar en ouder die de huisarts hadden geconsulteerd voor heupklachten en werden doorverwezen voor röntgenonderzoek van de heup, ondergingen een gestandaardiseerde anamnese en lichamelijk onderzoek van lage rug, heup en knie. Vervolgens werd een echografisch onderzoek uitgevoerd en werd de bezinkingssnelheid van de erytrocyten (BSE) bepaald. Voor de numerieke classificatie werden de patiënten in groepen geplaatst met behulp van cluster analyse (Ward methode). Voor deze clusteranalyse werden de gegevens afkomstig uit anamnese en lichamelijk onderzoek van de heup gebruikt (65 variabelen) hetgeen resulteerde in een classificatie met negen verschillende clusters. Herhaling van de analyse in 10 subgroepen van de patiëntpopulatie en met een alternatieve cluster analyse (complete linkage methode) leidde tot vergelijkbare resultaten. Verschillende clusters vertoonden een significante relatie met röntgenologisch en echografisch vaststelbare symptomen en met rug- en kniesymptomen. Een groep van 20 experts herkende de combinatie van symptomen in zeven van de negen clusters als bekende syndromen uit de klinische praktijk. Deze zeven clusters konden aan de hand van de combinatie van symptomen benoemd worden als syndromen beschreven in de medische literatuur, namelijk respectievelijk matige of ernstige coxartrose, beginnende coxartrose, trochantaire tendinitis, meralgia paresthetica, lage-rugklachten, knieklachten, en met een combinatie van coxartrose en lage-rugklachten. Twee clusters waren moeilijk te identificeren; een cluster met, behalve pijn in de heup, totaal geen locale symptomen en een cluster met vermoedelijk aandoeningen van de weke delen. Verder onderzoek zal moeten uitwijzen welke syndromen aan deze clusters ten grondslag liggen.

In dit hoofdstuk werd een op symptomen gebaseerde classificatie geponeerd van patiënten met heupklachten in de huisartspraktijk die reproduceerbaar bleek te zijn en een relatie vertoonde met radiologische en echografische symptomen. De meeste clusters vertoonden een vergelijkbare combinatie van symptomen als beschreven in de medische literatuur en werden als zodanig herkend door de experts. Een classificatie die echter geen onderscheid in prognose of in effectiviteit van een bepaalde behandeling aanbrengt heeft geen klinische betekenis; dit dient te worden bestudeerd in toekomstig onderzoek.

Hoofdstuk 6 beschrijft de ontwikkeling van een diagnostische functie voor heuppatiënten van middelbare leeftijd en ouder, gebaseerd op de classificatie beschreven in hoofdstuk 5, met het doel een individuele patiënt in het juiste (d.w.z. oorspronkelijke) cluster te plaatsen. We trachtten een diagnostische functie te ontwikkelen met zo weinig mogelijk en makkelijk door de huisarts te onderzoeken variabelen.

Met behulp van de discriminantanalyse (Wilks methode), op dezelfde 224 patiënten die voor de classificatie waren gebruikt, werden de meest onderscheidende variabelen in relatie tot de verschillende clusters geïdentificeerd. Hierop werd de diagnostische functie gebaseerd. Het aantal variabelen voor de diagnostische functie werd gevarieerd van 5 tot 50 variabelen en het percentage patiënten dat werd geplaatst in het juiste cluster met behulp van de diagnostische functie werd geanalyseerd.

Bij gebruik van 20 variabelen werd 80% van de patiënten nog steeds in hetzelfde cluster geplaatst als bij de oorspronkelijke classificatie. De helft van deze 20 variabelen bestond uit variabelen uit de anamnese, de exacte locatie van de pijn bleek de meest belangrijke.

Omdat dezelfde patiënten voor de oorspronkelijke classificatie werden gebruikt als voor het testen van de diagnostische functie kunnen betere resultaten zijn geboekt dan wanneer voor het laatste andere patiënten waren gebruikt. Wanneer de classificatie na toekomstig onderzoek een klinische betekenis blijkt te hebben, is deze classificatie goed bruikbaar voor de huisarts.

Hoofdstuk 7 beschrijft het vóórkomen van echografische gewrichtszwelling van de heup en de relatie hiervan met lichamelijk, röntgenologisch en haematologisch onderzoek (BSE) in dezelfde patiëntenpopulatie als in de beide voorgaande hoofdstukken.

Tijdens het gestandaardiseerde echografisch onderzoek van de heup werd de afstand tussen het gewrichtskapsel en de collum femoris aan de voorzijde van het gewricht gemeten. Een toename van deze afstand duidt op een gewrichtszwelling van de heup. De gewrichtszwelling was gedefinieerd op drie verschillende manieren: een 'gewrichtszwelling' in overeenstemming met Koski's definitie (afstand ≥ 7 mm óf een links/rechts verschil ≥ 1 mm), een 'ernstige gewrichtszwelling' (afstand ≥ 9 mm), en een 'eenzijdige gewrichtszwelling' (links/rechts verschil ≥ 1 mm).

'Gewrichtszwelling' van de (meest) symptomatische heup was aanwezig bij 80 (37%) patiënten, 'ernstige gewrichtszwelling' bij 20 (11%) patiënten en 'unilaterale' gewrichtszwelling bij 47 (22%) patiënten. Gecorrigeerd voor leeftijdsverschillen en de aanwezigheid van radiologische coxartrose vertoonde gewrichtszwelling van de heup een significant positieve relatie met de volgende symptomen: pijnlocatie in de lies of het mediale bovenbeen, verergering van pijn bij liggen op de zij, teken van Trendelenburg, bewegingsbeperking van de extensie, abductie, endorotatie en/of flexie, pijnlijke exorotatie en palpatiepijn in de lies. Alleen een ernstige gewrichtszwelling vertoonde een positieve relatie met een verhoogde BSE. Na uitsluiten van patiënten met een verhoogde BSE en met bilaterale klachten bleek een eenzijdige beperking van de extensie een goede voorspeller voor het hebben van een eenzijdige gewrichtszwelling aan dezelfde zijde (positief voorspellende waarde 71% en negatief voorspellende waarde 80%).

In deze studie werd een hoge prevalentie van een echografische gewrichtszwelling van de heup gevonden met een duidelijke relatie met klinische symptomatologie. Deze bevinding vraagt om vervolgonderzoek naar de prognostische en therapeutische waarde van de aanwezigheid van gewrichtszwelling bij volwassenen met heupklachten in de huisartsenpraktijk.

Hoofdstuk 8 beschrijft een patiënt-controle onderzoek waarbij de relatie tussen het hebben van meralgia paresthetica en de aanwezigheid van een radiologisch vastgestelde degeneratieve symphysis pubica werd onderzocht. Deze tot nu toe onbekende relatie werd gevonden in de met clusteranalyse gevonden groep van patiënten met meralgia paresthetica, beschreven in hoofdstuk 5.

De patiënten in dit patiënt-controle onderzoek waren 25 personen ouder dan 40 jaar die een chirurgische neurolyse ondergingen vanwege een meralgia parethetica en waarvan een röntgenfoto van het bekken beschikbaar was. De controlepersonen werden getrokken uit een studiecohort uit de open populatie van 55 jaar en ouder (ERGO) met hetzelfde geslacht en vergelijkbare leeftijd als de patiënten (4 controles per patiënt). Van de controlegroep werden de patiëntendossiers bij de huisarts gecontroleerd op de afwezigheid van symptomen van meralgia paresthetica in de afgelopen 10 jaar. Degeneratie van de symphysis pubica werd als aanwezig gedefinieerd wanneer twee van de drie onafhankelijke waarnemers dit als aanwezig hadden opgemerkt op de röntgenfoto.

De Mantel-Haenszel procedure (strata voor leeftijd en radiologische coxartrose) toonde een positieve relatie tussen meralgia paresthetica en een radiologisch degeneratieve symphysis pubica (odds ratio = 4.38, $p = 0.004$). Een afzonderlijke analyse voor alleen mannen vertoonde dezelfde positieve relatie.

In deze studie werd de in de clusteranalyse gevonden, maar tot nu toe onbekende relatie tussen meralgia paresthetica en een degeneratieve symphysis pubica bevestigd. Het is mogelijk dat degeneratieve veranderingen in de symphysis pubica omliggende weefsels of anatomische verhoudingen beïnvloeden en daardoor meralgia paresthetica veroorzaken.

Hoofdstuk 9 bevat de algemene discussie en de belangrijkste conclusies uit dit proefschrift. Het beschrijft tevens de methodologische problemen in classificatiestudies wanneer een gouden standaard ontbreekt. De numerieke classificatiemethode, vaak gepresenteerd als een objectieve methode die cirkelredenatie ontwijkt en gebruikt voor de classificatie in hoofdstuk 5, bleek een methode die gepaard gaat met vele keuzes die elk consequenties hebben voor het uiteindelijke resultaat. De beweegredenen bij de gemaakte keuzes worden besproken. In aansluiting werd de met numerieke methoden verkregen classificatie van heupklachten beoordeeld op basis van criteria voor doel, validiteit, betrouwbaarheid, haalbaarheid en generaliseerbaarheid. De classificatie voldeed aan de meeste van de bovengenoemde criteria. Of deze classificatie echter een leidraad zou kunnen zijn voor het voorspellen van prognose of therapeutisch effect, en daarmee klinisch relevant zou zijn, is nog niet vastgesteld en vereist vervolgonderzoek. Dezelfde vraag geldt voor de aanwezigheid van een (echografisch vastgestelde) gewrichtszwelling van de heup.

APPENDICES

Appendix A

Kellgren grading system for radiological osteoarthritis of the hip

Grade	Description
0 No osteoarthritis	
1 Doubtful	Possible narrowing of joint space medially and possible osteophytes around femoral head; or osteophytes alone.
2 Mild	Definite narrowing of joint space, definite osteophytes and slight sclerosis.
3 Moderate	Marked narrowing of joint space, definite osteophytes, some sclerosis and cyst formation and deformation of femoral head and acetabulum.
4 Severe	Gross loss of joint space with sclerosis and cysts, marked deformity of femoral head and acetabulum and large osteophytes.

Appendix B

Detailed information about limb positioning and instrument alignment for comparing two devices for measuring hip joint motion.

All movements were measured from the zero starting position as recommended by the AAOS.[10] This was also the position at which the inclinometer had to be re-calibrated to zero. When maximal movement was reached, the observer had to press the ENTER button of the measuring unit of the inclinometer. An assistant read the results on the display and noted these. The results from the goniometer were read by the observer and noted by the assistant.

Flexion of the hip was measured with the subject lying supine on a flat surface. The flexion was measured as deviation from the neutral zero position, in which the leg and body are in the horizontal plane. The maximal hip flexion was defined as the point at which rotation of the pelvis was felt by the observer. During hip flexion the knee was also flexed. The stationary arm of the goniometer was aligned over the horizontal axis of the body. The goniometer's moveable arm was aligned over the lateral midline of the thigh. The greater trochanter was used to centre the fulcrum of the goniometer. The measuring unit with the 'long bone unit' of the inclinometer was firmly attached to the anterior surface of the thigh, parallel to the longitudinal midline of the thigh.

Hip extension was measured in prone position with a small pillow placed under the abdomen. The pelvis was stabilised on the surface by means of a belt. The knee was flexed to 90° and the extension was measured as amount of deviation from the neutral zero position. The anatomical landmarks used for alignment of the goniometer were the same as used for measuring flexion. The inclinometer with the 'long bone unit' was attached to the posterior surface of the thigh, parallel to the longitudinal axis of the thigh.

Abduction and adduction in supine position were only measured with the two-arm goniometer; these movements were measured from the neutral zero position in which the longitudinal axis of the leg is perpendicular to the transverse line across the anterior superior spines of the pelvis. These latter anatomical landmarks were also used for alignment of the stationary arm of the goniometer. The unilateral anterior superior spine was used to centre the fulcrum of the goniometer. The moveable arm of the goniometer was aligned over the midline of the femur pointing at the centre of the patella. In measuring adduction, the subject had to lift the contra-lateral leg over the unilateral knee to allow the leg to pass under it. To measure abduction and adduction in lateral decubitus position, the measuring unit of the inclinometer with the 'long bone unit' was firmly attached to the lateral surface of the thigh, parallel to the lateral midline of the thigh. The movement was measured, as in supine position, as deviation from the neutral zero position. In this position the longitudinal axis of the leg was situated perpendicular to the transverse line across the superior anterior spines of the pelvis.

Rotations of the hip in supine position could only be measured with the two-arm goniometer. The subject had the hip and the knee of the leg flexed to 90°. The stationary arm of the goniometer was aligned parallel to the transverse line across the superior anterior spines of the pelvis. Internal and external rotation were measured as deviation from the zero starting position in which the longitudinal axis of the lower leg was perpendicular to the transverse line across the anterior iliac spines. Rotations in the sitting and prone positions were measured with both the inclinometer and the goniometer. In sitting position the knees and hips were flexed to 90°. In prone position the hips were in neutral zero position and the knee flexed to 90°. In both positions, the zero starting position was reached when the longitudinal axis of the lower leg was perpendicular to the flat surface of the bench (horizontal plane). The rotations were measured as deviations from this position. As in supine position, the fulcrum of the goniometer was centred over the patellar apex and the moveable arm of the goniometer was aligned over the longitudinal axis of the lower leg. The measuring unit of the inclinometer (without the 'long bone attachment') was placed on the tibial tubercle. Compensatory movement of the pelvis or lifting of the thigh was not allowed.

Appendix C

Means of *z*-scores of variables used in the cluster analysis separate for every cluster. Average of the z-score of a variable for the total study population (*n* = 224) is 0 with a standard deviation of 1

Symptom	cluster 1 n = 8	cluster 2 n = 40	cluster 3 n = 40	cluster 4 n = 33	cluster 5 n = 44	cluster 6 n = 20	cluster 7 n = 11	cluster 8 n = 18	cluster 9 n = 10
- Pain excitation/character									
pain onset after trauma	0.14	-0.30	-0.30	0.02	-0.14	0.41	1.98	-0.10	0.05
pain onset after overuse	-0.04	-0.18	-0.18	-0.35	-0.32	-0.11	-0.14	-0.08	0.18
pain increased by movement	-0.18	0.51	-0.55	0.06	-0.37	0.24	0.06	0.47	0.35
pain increased by sitting	0.41	-0.29	0.19	0.38	-0.37	0.52	0.32	-0.43	-0.23
pain increased by lying	-0.44	-0.31	-0.24	-0.36	0.04	0.23	1.75	0.75	-0.18
pain increased by lying on the side	0.27	-0.14	-0.08	-0.39	0.18	0.38	0.04	0.44	-0.44
pain increased by standing	0.62	0.32	-0.24	-0.35	0.28	0.27	0.51	-0.69	-0.74
pain increased by walking	-0.11	0.47	-0.38	-0.22	0.25	0.15	0.10	-0.50	-0.17
pain increased after load	0.26	0.41	-0.19	-0.44	0.05	0.09	-0.26	-0.21	0.81
pain after prolonged inactivity	0.29	0.15	-0.25	-0.00	-0.03	-0.02	0.56	0.04	-0.37
nocturnal pain	0.29	0.01	0.01	0.07	-0.22	0.29	0.35	-0.10	0.15
morning stiffness	0.57	0.31	-0.53	0.02	0.07	0.20	0.23	0.31	0.53
continuance of pain (1,2,3)	0.20	-0.13	0.28	0.48	0.12	-0.44	-0.85	-0.28	-0.52
pain endurance (1,2,3)	-0.40	0.60	-0.33	0.04	0.18	0.01	-0.23	-0.66	-0.52
pain severity (0-10)	0.25	0.20	-0.49	0.23	-0.30	0.58	0.50	-0.30	0.37
- Pain location (0,1,2)									
low back	0.30	0.12	-0.01	-0.16	-0.30	0.74	0.70	-0.39	-0.41
groin	-0.47	0.82	-0.59	1.08	-0.51	-0.49	0.32	-0.45	-0.44
buttock	0.44	-0.14	0.20	-0.05	-0.09	0.68	0.25	-0.61	-0.60
greater trochanter	-0.09	-0.00	-0.22	-0.53	0.23	0.18	-0.50	1.08	-0.06
anterior thigh	-0.78	0.65	-0.40	0.36	-0.40	-0.48	0.86	0.14	-0.03
posterior thigh	0.23	-0.22	0.12	-0.33	-0.23	1.02	1.10	-0.33	-0.33
lateral thigh	1.24	-0.44	-0.24	-0.71	0.48	0.12	-0.17	0.84	0.36
medial thigh	-0.36	0.51	-0.36	0.06	-0.31	-0.25	1.11	0.02	0.10
anterior knee	0.92	0.29	-0.22	-0.30	0.06	-0.14	0.21	-0.37	0.46
posterior knee	0.03	-0.22	-0.09	-0.28	-0.28	1.12	0.18	-0.28	1.50
medial knee	0.11	-0.27	-0.04	-0.27	-0.27	-0.12	0.28	0.23	2.76

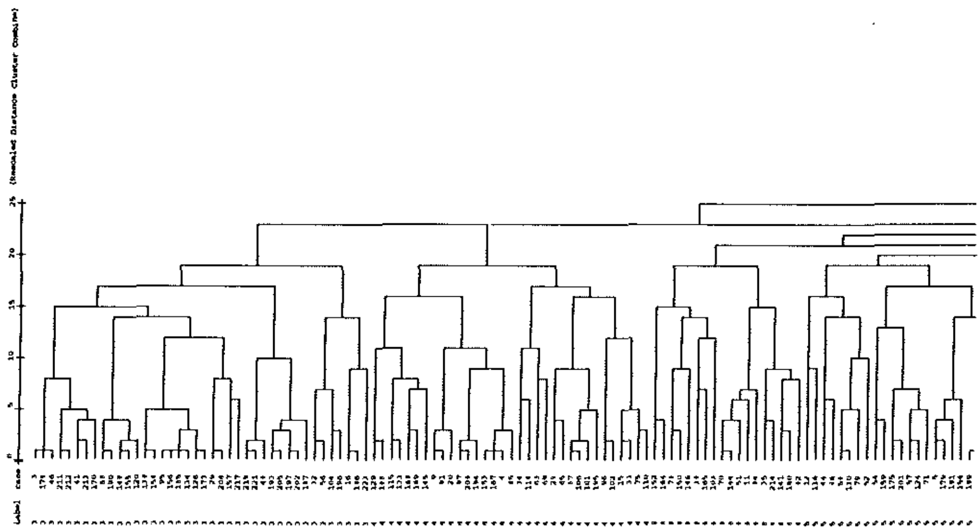
Symptom	cluster 1 n = 8	cluster 2 n = 40	cluster 3 n = 40	cluster 4 n = 33	cluster 5 n = 44	cluster 6 n = 20	cluster 7 n = 11	cluster 8 n = 18	cluster 9 n = 10
- Pain location (0,1,2) - cont'd									
lateral knee	0.21	-0.42	-0.13	-0.40	0.49	0.16	-0.17	0.23	0.64
lower leg	0.05	-0.50	0.10	-0.47	0.58	0.71	-0.37	-0.30	-0.34
- Tenderness on palpation									
iliopsoas muscle	-0.46	0.40	-0.39	0.18	-0.22	-0.06	1.22	-0.02	-0.46
tensor muscle	-0.25	0.63	-0.43	-0.48	0.15	0.04	0.09	0.11	0.08
gluteus max. muscle	-0.57	0.05	-0.46	-0.08	-0.01	0.61	0.85	0.20	0.00
gluteus med. muscle	-0.82	0.60	-0.47	-0.09	-0.32	0.49	0.28	0.42	-0.42
piriformis muscle	-0.25	0.15	-0.25	-0.09	-0.32	0.30	0.81	0.33	0.20
hip capsule in groin	-0.57	0.47	-0.51	0.34	0.12	-0.34	0.27	-0.18	-0.34
inguinal igament	0.44	0.76	-0.49	-0.45	0.19	-0.43	0.73	-0.41	-0.00
greater trochanter	-0.22	0.14	-0.52	-0.20	-0.03	0.49	-0.06	0.58	0.39
ischial tuber	-0.21	0.22	-0.27	-0.07	-0.35	0.59	0.60	0.17	-0.02
posterior iliac spines	-0.22	0.09	-0.69	-0.24	-0.08	0.92	0.96	0.53	-0.12
- Pain on muscle resistance tests									
hip flexion	-0.23	0.34	-0.28	-0.44	-0.28	0.81	0.95	-0.40	0.70
hip extension	-0.05	0.09	-0.26	-0.32	-0.21	0.86	0.63	-0.41	1.02
hip abduction in 90° flexion	-0.56	0.50	-0.14	0.08	-0.40	0.15	-0.13	0.10	0.15
hip abduction in 0° flexion	0.15	0.10	-0.70	-0.12	-0.12	0.86	0.98	0.04	0.36
hip adduction	-0.05	0.23	-0.55	-0.20	-0.30	0.96	0.98	0.13	0.06
hip internal rotation	-0.33	0.45	-0.44	-0.34	-0.15	0.73	0.92	0.82	-0.39
hip external rotation	-0.32	0.34	-0.37	-0.39	0.01	0.60	1.21	-0.60	0.19
- Weakness on muscle resistance tests									
hip abduction in 0° flexion	-0.02	0.05	-0.39	-0.21	0.21	-0.10	0.67	0.58	-0.39
hip internal rotation	0.11	-0.06	-0.32	-0.32	0.07	0.02	0.92	0.82	-0.32
hip external rotation	0.11	0.02	-0.32	-0.32	-0.24	0.02	2.48	0.44	-0.32
- Decreased passive hip motion (0,1,2)									
flexion	0.24	0.51	-0.42	0.19	0.08	-0.46	0.33	-0.09	0.20
extension	-0.09	0.63	-0.26	-0.16	-0.07	0.29	0.04	-0.35	-0.56
abduction	0.01	0.55	-0.63	0.19	0.10	0.03	0.16	-0.58	0.36
adduction	-0.56	0.74	-0.31	-0.16	-0.11	-0.31	0.48	0.16	-0.40
internal rotation	-0.01	0.32	-0.43	0.18	0.08	-0.46	0.33	-0.09	0.20
external rotation	0.25	0.25	-0.41	-0.06	0.13	0.05	0.22	-0.01	-0.22

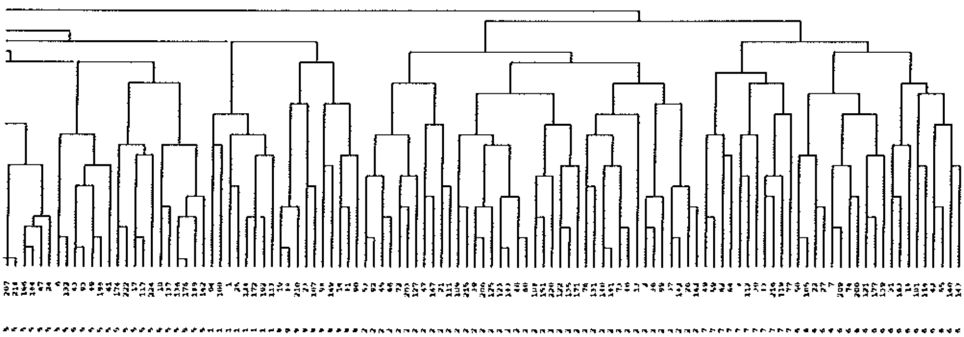
- Pain at passive hip motion									
flexion	-0.06	0.57	-0.84	0.41	-0.41	0.42	0.35	0.03	0.31
extension	0.65	0.35	-0.36	0.11	-0.59	0.35	1.15	-0.19	0.14
abduction	0.09	0.53	-0.74	-0.02	-0.11	0.20	0.64	-0.22	0.64
adduction	0.60	0.65	-0.66	0.06	-0.38	-0.05	0.67	0.30	-0.15
internal rotation	-0.03	0.48	-0.87	0.11	0.22	0.22	0.37	-0.41	0.12
external rotation	0.40	0.44	-0.81	0.11	-0.27	0.55	0.79	0.14	-0.26
- End-feel									
bony end-feel on passive hip movements	-0.07	0.76	-0.35	-0.36	0.13	-0.40	0.18	-0.38	0.26
- Other tests									
pain on provocation sacro-ilacal joint	-0.49	0.29	-0.28	-0.44	0.10	0.70	0.76	-0.29	-0.34
pain on straight leg raising	0.04	-0.12	-0.27	-0.26	0.01	1.23	0.51	-0.18	-0.35
pain at compression	-0.17	0.78	-0.49	-0.26	-0.01	0.15	1.13	-0.49	0.02
decreased sensation anterio-lateral thigh	3.86	0.05	-0.26	-0.13	-0.16	-0.05	-0.26	-0.26	-0.26

When not specified, dichotomous variables (0,1): 0=absent, 1=present
Continuance of pain (1,2,3): 1= continuously, 2= several times a day, 3= several times a week
Pain endurance (1.2.3): 1= < 3 months, 2= 3-12 months, 3=12-24 months
Pain severity (0,10): analog scale from 0= no pain to 10=worst imaginable pain
Pain location (0,1,2): 0 = no pain here, 1= pain here, but not the worst pain, 2 = worst pain here.
Decreased passive hip motion (0,1,2) : 0 = not decreased, 1= slightly decreased, 2 = moderate or severe decreased.

Appendix D

Dendrogram (cluster tree) of 224 patients with hip pain and their grouping by cluster analysis (Wards' method) based on the similarity which the patients show in 65 local symptoms. The separate bars at the bottom of the dendrogram represent the individual cases.





Appendix E

Classification function coefficients (Fisher's linear discriminant function) for classifying hip patients into nine different clusters with 20 variables.

	cluster membership								
	1	2	3	4	5	6	7	8	9
pain location groin (0,1,2)	1,519	4,526	1,019	5,342	1,312	2,450	4,321	1,353	1,641
pain location medial knee (0,1,2)	-1,578	-,301	,772	-,839	-,729	-1,237	,526	2,888	16,974
pain location posterior knee (0,1,2)	1,405	1,910	,307	1,389	-,321	5,551	1,742	1,163	11,470
pain location posterior thigh (0,1,2)	-,381	-,230	1,287	,414	,128	3,763	6,649	,561	-4,496
pain location medial thigh (0,1,2)	2,453	5,539	,605	2,424	1,382	3,574	9,900	2,879	5,494
pain location greater trochanter (0,1,2)	1,578	1,777	1,754	1,069	2,557	2,865	1,825	4,286	2,577
pain location lower leg (0,1,2)	2,603	,522	1,759	,490	3,419	2,216	1,119	1,623	-,993
pain onset after trauma (0,1,2)	,477	,796	1,202	3,474	1,730	7,086	15,698	2,021	,927
pain increased with lying (0,1,2)	2,273	2,421	1,318	1,456	3,543	5,303	13,293	7,323	2,658
decreased passive abduction (0,1,2)	1,475	2,950	,659	2,120	1,695	2,010	2,249	,567	2,330
painful passive external rotation (0,1)	3,005	3,225	-8,34E-02	2,708	1,105	3,651	4,292	2,797	1,418
bony end-feel (0,1)	2,737	4,360	,344	1,015	2,097	,578	2,969	,327	1,765
weakness on muscle resistance of abduction (0,1)	-,869	-1,658	,357	-1,598	2,502	-,667	-1,194	4,117	-2,043
weakness on muscle resistance of external rotation (0,1)	-2,905	2,988	,352	,549	-,583	2,468	23,171	1,660	-,126
weakness on muscle resistance of internal rotation (0,1)	,564	-2,990	-,120	-2,271	1,824	-1,514	-6,743	6,480	-,967
pain on muscle resistance of abduction (0,1)	4,036	4,823	,542	3,717	2,687	6,448	8,610	2,780	3,751
pain on muscle resistance of internal rotation (0,1)	4,108	4,956	,391	1,900	1,254	5,414	7,899	,245	2,795
tenderness on palpation of posterior iliac spines (0,1)	4,560	4,372	,759	2,813	2,974	7,151	7,630	4,784	2,549
tenderness on palpation of inguinal ligament (0,1)	4,300	5,038	,844	,174	3,415	-8,20E-02	4,143	,684	2,824
decreased sensibility anterio-lateral thigh (0,1)	42,461	5,839	-,520	1,946	1,702	3,889	-1,917	-,799	-,389
(Constant)	-29,987	-16,684	-3,865	-10,123	-9,026	-18,772	-44,262	-13,643	-20,501

Dankwoord

Aan het begin van een promotieonderzoek is een proefschrift nog een onwaarschijnlijke gedachte. Dat zoiets toch lukt binnen de gestelde termijn is de verdienste van velen. Personen die een bijzondere gaven aan dit proefschrift hebben gegeven wil ik hier noemen. Allereerst dank aan dr. Ridderikhoff, de geestelijke vader van het 'classificatieproject'. Zijn begeleiding en onvoorwaardelijke inzet tot aan het bereiken van zijn pensioen hebben de eerste twee jaar van het onderzoek kleur gegeven.

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About the author

Sita Bierma-Zeinstra was born in Leeuwarden on the 14th of February, 1961. Together with her two brothers she spent a happy childhood on their parents' farm situated on one of Friesland's *terps*. She attended the *Oostergo* College in Dokkum and after graduation in 1978 began physical therapy studies. After working for some time as a physiotherapist in northern Friesland she broadened her horizon by moving to Sweden. There she worked as a physiotherapist for nine years: two years in a rehabilitation clinic of the Swedish National Social Insurance Board and seven years in primary care. From 1988 until her return to the Netherlands in 1992 she was head of the department for physical therapy in the *Strängnäs* primary care region. Back in the Netherlands, she studied Health Sciences at the Catholic University of Nijmegen, majoring in Human Movement Sciences. After graduation in 1994 she became a research fellow at the department of General Practice of Erasmus University Rotterdam on the project 'Classification of hip disorders in the elderly'. Beside her research activities she teaches one day a week practical skills in physical examination of the musculoskeletal system to postgraduate medical students. At the same department, she will continue with both research and teaching related to musculoskeletal problems. The author lives with her husband Menno in the center of Haarlem.