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Hypermobility among patients with greater trochanteric pain syndrome

Lisa C. U. Reimer[1], Julie S. Jacobsen[2] & Inger Mechlenburg[1, 3, 4]

ABSTRACT

INTRODUCTION: Greater trochanteric pain syndrome (GTPS) is a common and disabling hip condition. Hypermobility has been suggested as a possible cause of GTPS. The purpose of this study was to report the prevalence of hypermobility and to investigate its impact on hip-related function and awareness in patients with GTPS.

METHODS: This cross-sectional study was based on a cohort of patients diagnosed with GTPS in the 2013-2015 period. Hypermobility was investigated with the Beighton Score and defined by a cut-off score ≥ 5. Data on patients' current hip function and awareness were collected with the questionnaires the Copenhagen Hip and Groin Outcome Score and the Forgotten Joint Score.

RESULTS: A total of 612 patients with GTPS were identified based on the diagnosis system; out of those, 390 patients were assessed for eligibility, and 145 (37%) were included. The prevalence of hypermobility within this cohort was estimated to be 11% (95% confidence interval (CI): 3-26%) for males and 25% (95% CI: 17-34%) for females. No significant association was found between hypermobility and self-reported hip function and awareness.

CONCLUSIONS: The prevalence of hypermobility in patients with GTPS was high, but the prevalence of hypermobility did not influence hip function and awareness. The results were based on a very low response rate and should be interpreted with this in mind.

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Greater trochanteric pain syndrome (GTPS) is pain located to the lateral side of the hip and encompasses trochanteric bursitis, gluteal tendinopathy, partial and total tears in the gluteal tendons, and external snapping hip [1]. These diagnoses are often seen together, and only the surgical management of the diagnoses varies [1]. GTPS has been estimated to affect 10-25% of the general population, and the incidence of new cases has been reported to be 2-6 per 1,000 per year [2]. The incidence has been estimated to be higher among patients with low-back pain, and the condition is most commonly seen in females aged 40-60 years [1]. The clinical presentation of GTPS is movement-related pain, particularly during external rotation and abduction [3]. Normal activities like getting in and out of a car; sleeping and lying on the affected side, and running often aggravate or extend the syndrome [3]. Also, poor quality of life, high levels of pain and physical impairments and a lower rate of fulltime employment are found among patients with GTPS [2]. A retrospective study by Lievense et al found that 36% of patients visiting primary care due to trochanteric pain still had problems one year after their initial visit and that 29% still experienced pain after five years [4].

The cause of GTPS is multifactorial. Generalised joint hypermobility (GJH) has been suggested as a possible cause of GTPS [5, 6]. When the hip is hypermobile, the need for dynamic stabilisers is increased to ensure that the femoral head is contained in the acetabulum during walking [5]. The increase in muscle work and possible dysfunctional gait and movement patterns could result in tightness of the iliotibial band, tendinopathy and/or bursitis [5, 6]. According to Fearon et al, research involving risk factors for GTPS is rare and more risk factors should be identified [7]. High levels of physical disability among patients with GTPS could lead to a high use of healthcare resources and have an impact on quality of life and metal health [2]. With this in mind, we found it important to investigate the possible predisposing factor, hypermobility, and the impact of this factor. The primary aim of this study was to report prevalence of GJH and to investigate the impact of GJH on self-reported outcome in patients 2-4 years after their initial GTPS diagnosis. GJH has been found to be related to reduced muscle strength and a lower level of physical activity [8]. Therefore, we hypothesised that patients with GJH would report lower Copenhagen Hip and Groin Outcome Scores (HAGOS) [9] and lower Forgotten Joint Scores (FJS) [10] than the patients without GJH.

METHODS

The Danish Data Protection Agency approved the study (record number: 2012-58-006). Data collection was initiated on 11 October 2017 and concluded on 28 February 2018.

Subjects

Patients were identified from the Danish Diagnosis-

ORIGINAL ARTICLE

1) Department of Orthopaedics, Aarhus University Hospital
2) Department of Physiotherapy, Faculty of Health Sciences, VIA University College, Aarhus
3) Department of Clinical Medicine, Aarhus University
4) Department of Public Health, Aarhus University, Denmark

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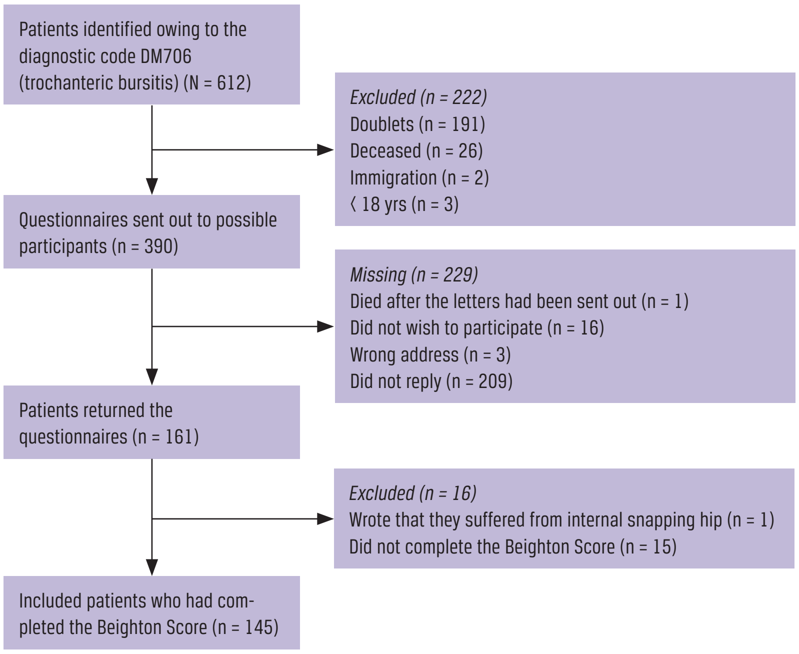
FIGURE 1

Flow chart of the patients included and excluded. Doublets were patients with multiple visits with an identical diagnostic code and thus appearing more than once in the diagnostic system.



Related Group (DRG) database based on the diagnostic code DM706, which covers GTPS. GTPS encompasses trochanteric bursitis, gluteus medius and minimus tendinopathy, partial and total tears in the gluteal tendons, and external snapping hip [1]. We identified 612 eligible patients. Patients were excluded if they were under the age of 18 years, no longer lived in Denmark or had died. A total of 390 questionnaires were mailed in October 2017; after one month, another 240 letters were sent out as a reminder to those who did not answer initially. Since the questionnaires were written in Danish, inclusion indirectly required that patients were able to read and understand Danish.

Study design

This study was cross-sectional and based on a cohort of patients diagnosed with GTPS in the 2013-2015 period from the Orthopaedic Department at Aarhus University Hospital, Denmark. Information about exposure was investigated with the self-reported Beighton Score, and patients with a score of five or more were defined as having GJH [11, 12].

Self-reported hip function

Information on the patients' current hip function was collected with the HAGOS questionnaire [9]. HAGOS was developed to evaluate symptoms related to the hip in patients aged 18-63 years [9]. Hip function is measured by six subscales including; Symptoms, Pain, Physical function in daily living, Physical function in Sport and Recreation, Participation in Physical Activities, and Quality of Life, and consist of 37 questions [9]. Every question has five answer options ranging from never to always, corresponding to the categories 0-4. Each subscale was converted into a score from 0-100, where 100 indicates no problems and 0 indicates severe problems. Kemp et al found that the minimal important change (MIC) for HAGOS ranged 1-10 by the different subscales in a population of Australian patients who had undergone hip arthroscopic surgery [13]. Thomeé et al found that for the Swedish version of HAGOS, MIC ranged from 8.8 to 13.1 in a population of Swedish patients scheduled for hip arthroscopy due to symptomatic femoroacetabular impingement [14].

Self-reported hip awareness

The patients' awareness of their affected hip was examined with the FJS questionnaire, where each patient completed the 12 questions about awareness of their affected hip [10]. Each question was answered with one of the following options; never, almost never, seldom, sometimes and mostly, corresponding to a score of 1-5. The sum of the scores was converted into a score ranging 0-100. A high score indicates lack of awareness and a low score indicates great awareness [10]. The minimal detectable change of the Danish version of FJS has been reported to be 32 points among patients with femoroacetabular impingement who had undergone hip arthroscopic treatment [15]. Bramming et al found that the Danish version of the FJS had a high reliability, responsiveness and no floor or ceiling effect in this patient group [15].

Covariates

The patients also answered a number of baseline questions, including questions about height, weight, educational level, consumptions of painkillers and pain measured with the visual analogue scale at rest and in activity [6]. Furthermore, the participants were asked to complete the University of California at Los Angeles Activity Score, which is a questionnaire in which the patient chooses one out of ten options that best describes their current physical activity level [16].

Statistical analysis

Continuous variables are presented as mean values with one standard deviation (SD) when normally distributed and elsewise as medians and interquartile ranges. Categorical data are presented as prevalences and prevalence proportions. The unpaired t-test was used to investigate differences between hypermobile and non-hypermobile patients for normally distributed data. Wilcoxon's rank-sum test was used when data were not normally distributed. Fisher's exact chi-squared test was used for categorical data, and variables with more than two categories were tested for trend. Differences in outcomes between hypermobile and non-hypermobile patients were examined using linear regression. The analysis was adjusted for the possible confounders of age, gender and Body Mass Index (BMI), which were decided prior to the data collection. All statistical analyses were performed in Stata 15 (StataCorp LLC, College Station, TX, USA).

Trial registration: not relevant.

RESULTS

A sample of 612 patients with the DRG code DM706 were identified; out of those, 390 patients were assessed for eligibility and 145 (37.2%) patients were included in this study (**Figure 1**).

There were no differences between patients with and without GJH (**Table 1**), and we found no difference in age and gender among the included patients (37.2%) and excluded/missing patients (62.8%). The mean age was 55.3 years (SD: ± 18.6 years) for included patients and 55.7 years (SD: ± 19.4 years) for excluded and missing patients (p = 0.84). The proportion of males was 24.8% for included patients and 30.5% for excluded and missing patients (p = 0.25).

The prevalence of GJH within this cohort was estimated to be 11.1% (95% confidence interval (CI): 3.1-26.1%) for males and 24.8% (95% CI: 17.0-34.0%) for females, using a five-point Beighton Score cut-off. The mean age of the included patients was 63.0 years (SD: ± 16.81 years) for males and 53.0 years (SD: ± 18.33 years) for females. Having GJH did not affect the self-reported hip outcome 2-4 years after the initial diagnosis (**Table 2**).

DISCUSSION

In an unsystematic review, GJH has been suggested to be associated with GTPS and the underlying diagnoses [5]. Nevertheless, no studies have systematically investigated the association between GJH and GTPS. We found it relevant to estimate the prevalence of GJH in patients diagnosed with GTPS and to investigate its impact on self-reported hip function. The prevalence of GJH within this cohort was 11% for males and 25% for females when applying a five-point Beighton Score cut-off level. Compared with a study on the prevalence of GJH in healthy Caucasians living in New Zealand, GJH was much more frequently seen in patients with GTPS in our study [17]. Klemp et al estimated the prevalence of GJH to be 2% for healthy Caucasian males and 6% for healthy Caucasian females based on a cut-off level of four on the Beighton Score [17]. The mean age in the study performed by Klemp et al was 46 years for both male and females, whereas it was 63 and 53 years for male and females in our study. Both the mean age and cut-off level on the Beighton score were higher in our study, and therefore it is possible that the difference in prevalence of GJH between our patients and the general population is even bigger.

This study did not establish any significant associ-

TABLE 1

Baseline characteristic for the patients with greater trochanteric pain syndrome, divided into two groups: hypermobile and non-hypermobile patients.

	Hypermobile[a] (n = 31)	Non-hypermobile[b] (n = 114)	p-value
Beighton Score, mean (± SD)	5.9 (± 1.1)	1.6 (± 1.6)	–
Men, n (%)	4 (12.9)	32 (28.1)	0.10
Age, yrs, mean (± SD)	56.5 (± 19.0)	55.2 (± 18.3)	0.73
BMI, kg/m^2, mean (± SD)	26.4 (± 4.9)	25.6 (± 4.8)	0.42
Educational level, n (%)			0.23
Primary school	8 (25.8)	20 (17.7)	
High school	4 (12.9)	9 (8.0)	
Short higher education	6 (19.4)	24 (21.2)	
Medium long higher education	9 (29.0)	46 (40.7)	
Long higher education	4 (12.9)	14 (12.4)	
Received physical therapy, n (%)	24 (77.4)	81 (72.3)	0.65
Z-plastic operation, n (%)			
Previous	11 (39.3)	35 (32.4)	0.51
Scheduled	0	5 (4.5)	0.58
Steroid injection, n (%)	16 (53.3)	71 (65.7)	0.29
Pain related to snapping or clicking within the last 14 days, n (%)	8 (25.8)	49 (45.0)	0.06
Use of painkillers, n (%)			0.50
Never	9 (32.1)	27 (25.0)	
Monthly	4 (14.3)	18 (16.7)	
Weekly	5 (17.9)	18 (16.7)	
Daily	10 (35.7)	45 (41.7)	
VAS score, median (IQR)			
At rest	15.0 (2.0-32.0)	25.0 (4.0-54.0)	0.11
At activity	42.0 (10.0-70.0)	59.5 (18.0-80.1)	0.13
Playing sport, n (%)	21 (67.7)	54 (48.2)	0.07
Time used weekly on sport, h, median (IQR)	3.0 (1.5-4)	2.5 (2-4)	0.99
UCLA activity score, mean (± SD)	5.9 (± 2.2)	6.1 (± 2.3)	0.72
FJS, median (IQR)	33.90 (23.6-44.2)	29.74 (24.7-34.7)	0.46
HAGOS, median (IQR)			
Pain	59.74 (49.8-69.7)	56.01 (50.9-61.1)	0.50
Symptoms	60.41 (51.9-68.9)	54.57 (50.2-59.0)	0.22
Activities of daily living	58.63 (47.9-69.4)	54.54 (49.0-60.0)	0.49
Sport and recreation	50.80 (39.6-62.0)	44.43 (38.5-50.4)	0.32
Participation in activity	53.45 (41.7-65.2)	39.70 (33.2-46.2)	0.05
Quality of life	51.45 (40.7-62.2)	43.69 (38.8-48.5)	0.15

FJS = Forgotten Joint Score; HAGOS = Copenhagen Hip and Groin Outcome Score; IQR = interquartile range; SD = standard deviation; UCLA = University of California at Los Angeles; VAS = visual analogue scale.
a) Patients with a Beighton Score ≥ 5.
b) Patients with a Beighton Score ≤ 4.

TABLE 2

Linear regression analysis for the association between the presence of generalised joint hypermobility and the score on the Copenhagen Hip and Groin Outcome Score (HAGOS) and Forgotten Joint Score (FJS).

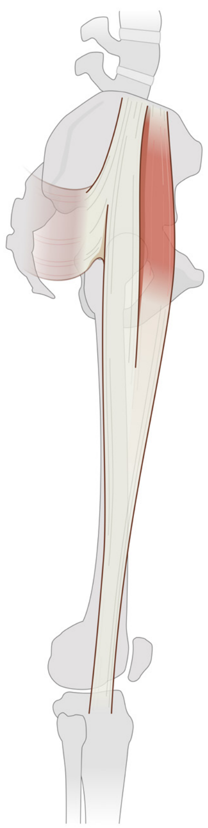
	Unadjusted			Adjusted[c]		
Questionnaire	n[a]	difference in score[b], average (95% CI)	p-value	n[a]	difference in score[b], average (95% CI)	p-value
HAGOS						
Pain	141	3.72 (-7.16-14.61)	0.50	139	4.41 (-6.55-15.38)	0.43
Symptoms	144	5.83 (-3.62-15.29)	0.22	142	6.61 (-3.05-16.28)	0.18
Activities of daily living	142	4.09 (-7.65-15.83)	0.49	140	5.32 (-6.42-17.07)	0.37
Sport and recreation	138	6.36 (-6.12-18.84)	0.32	136	7.25 (-5.28-19.77)	0.26
Participation in activity	137	13.75 (0.01-27.49)	0.05	135	12.91 (-1.23-27.05)	0.07
Quality of life	143	7.76 (-2.94-18.46)	0.15	141	7.48 (-3.37-18.33)	0.18
FJS	143	4.16 (-6.88-15.21)	0.46	141	4.58 (-6.65-15.82)	0.42

CI = confidence interval.
a) Patients who had completed the Beighton Score, the relevant subscale and the question about BMI.
b) Between hypermobile and non-hypermobile patients defined by a cut-off at Beighton Score = 5, non-hypermobile patients are the reference group.
c) Adjusted for sex, age and self-reported BMI.

ations between GJH and self-reported hip function measured with HAGOS or hip awareness measured with FJS. Mayes et al found no difference on the HAGOS subscale of pain between a group of ballet dancers and a group of matched non-dancing athletes [18]. The ballet dancers had a significantly higher prevalence of GJH than the non-dancing athletes, measured with the Beighton Score and using a cut-off level of five [18]. The result of our study is therefore consistent with the results by Mayes et al [18].

The mean age of our patients was 55 years for included patients and 56 for non-participating patients. The proportion of males within this cohort was 25% for included patients and 31% for missing patients. Patients diagnosed with GTPS have been described to be around 40-60 years of age and mostly females [1], which is in line with our findings. The relatively high age of the patients in our study increases the likelihood that our patients suffer from other diagnoses as well as GTPS, since the risk of having different diagnoses is higher among older patients [19].

Greater trochanteric pain syndrome is pain located to the lateral side of the hip and encompasses trochanteric bursitis, gluteal tendinopathy, partial and total tears in the gluteal tendons, and external snapping hip.



Study limitations

A major limitation of the present study is the low response rate (37.2%), which might be associated with the use of diagnostic codes. Since the diagnostic code covering GTPS is still named bursitis trochanteric and encompasses trochanteric bursitis, gluteal tendinopathy, partial and total tears in the gluteal tendons, and external snapping hip, it is possible that some of the patients had never heard of GTPS. These patients are expected to have influenced the response rate due to the possibility that they have not found the study relevant for them and thus did not participate. The inclusion of patients based on a diagnostic code can also result in a risk of including patients who do not suffer from the diagnosis, due to the risk that the examining doctor may have made a mistake when entering the patient's diagnose in the patient chart. One patient reported that he had never had GTPS but internal snapping hip, indicating that this could have occurred. To increase the quality and verify that patients actually did suffer from GTPS 2-4 years ago, there should have been a question in the questionnaire clarifying this.

Another limitation is the data collection. The patients' self-reported hip function could be a result of other diagnoses than GTPS or a combination of diagnoses, if the patient had suffered from other hip-related diagnoses since being diagnosed with GTPS or at the time the diagnosis was given. We did not have the possibility to check this in the patient charts, and we did not ask the patients if they had been diagnosed with other hip-related conditions, which would have increased the validity of the results of this study. The use of the specific diagnostic code (DM706) could also result in the exclusion of patients suffering from gluteal tendinopathy since this condition is covered by another diagnostic code (DM76.0). During the inclusion period, the diagnostic code DM76.0 was used 31 times at the Orthopaedic Department at Aarhus University Hospital. Due to the approval of this study, which did not include the diagnostic code DM76.0, the number has not been checked for doublets or exclusion criteria. Since GTPS is an unspecific syndrome that encompasses most diagnoses related to pain at the lateral side of the hip, it is possible that the diagnosis is given when a more precise diagnose cannot be made. The 2013-2015 period was chosen because we were interested in the hip function 2-4 years after the diagnosis was given since we assumed that the patients had regained or reached a stable level of hip function and physical activity at this time. Furthermore, the choice of this period made it possible to include patients who had undergone surgery, since these patients were also expected to have reached a stable level of function and physical activity 2-4 years after surgery.

It is possible that the prevalence of GJH has been overestimated due to selection bias. If the patients without GJH are the ones who did not participate due to a lack of interest, then the prevalence may be overestimated. The estimate of the prevalence should therefore be interpreted with the low response rate in mind. As for all self-reported questionnaires, misreports of results due to patients not understanding the questions could be a possible bias. Since the HAGOS and the FJS have been found to be valid and reliable among patients with hip-related problems, misreporting is not expected to have occurred for these questionnaires [9, 15]. The agreement of the self-reported Beighton Score and the examiner-assessed Beighton Score has been demonstrated to be good, which indicates that the validity and the reliability are good [20]. Having this in mind, we do not expect misreports and thereby information bias to be a concern in our study.

CONCLUSIONS

This study indicates that hypermobility is common among patients with GTPS, but the presence of hypermobility does not influence hip function and awareness, 2-4 years after the initial diagnosis. The results are based on a very low response rate and should be interpreted with this in mind.

CORRESPONDENCE: *Lisa C. U. Reimer*. E-mail: lisaurup@gmail.com
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