ORIGINAL RESEARCH ARTICLE



Generalized joint hypermobility and the risk of pregnancyrelated pelvic girdle pain: Is body mass index of importance?—A prospective cohort study

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Abstract

Introduction: Pelvic girdle pain (PGP) affects approximately 50% of pregnant women. The mechanisms are multifactorial but not fully understood. Women with generalized joint hypermobility (GJH) may be vulnerable to load in the pelvic joints during pregnancy. Our aim was to investigate if women with GJH had an increased risk of PGP and higher pain intensity during and after pregnancy, compared with women with normal joint mobility. We also studied if body mass index (BMI) in early pregnancy influenced that risk.

Material and methods: A prospective cohort study of 356 women, whose data were collected by self-reports and clinical examinations in early and in late pregnancy and 9 months after childbirth. GJH was present with $\geq 5/9$ points on the Beighton score. PGP was defined by a pain drawing and ≥ 1 positive test. Pain intensity was measured with a visual analogue scale (0–100 mm). We adjusted for age and origin in logistic regression and ordinal logistic regression analysis.

Results: In early pregnancy, 47.1% of the women with GJH had PGP vs 32.6% of women with normal joint mobility (adjusted odds ratio [aOR] 1.76; 95% confidence interval [CI] 0.86-3.62) and had higher odds of reporting higher pain intensity (aOR 2.04; 95% CI 1.02-4.07). The odds of PGP were highest for women with GJH and BMI $\geq 25 \, \text{kg/m}^2$ (aOR 6.88; 95% CI 1.34-35.27) compared with women with normal joint mobility and BMI $< 25 \, \text{kg/m}^2$. The estimated associations were weaker and not statistically significant in late pregnancy or after childbirth.

Conclusions: Women with GJH did not have an increased risk of PGP during or after pregnancy but reported higher pain intensity in early pregnancy compared with women with normal joint mobility. Since women with combined GJH and BMI ≥25 kg/m² had the highest odds of PGP in early pregnancy, our results may suggest that

Abbreviations: BeS, Beighton score; BMI, body mass index; GJH, generalized joint hypermobility; GW, gestational week; PGP, pelvic girdle pain; VAS, visual analogue scale.

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health care needs to pay attention to and develop methods to reduce the risk of PGP and delay the onset of pain during pregnancy in women with this combination.

KEYWORDS

body mass index, generalized joint hypermobility, pelvic girdle pain, pregnancy, Uppsala pelvic pain study

1 | INTRODUCTION

Pelvic girdle pain (PGP) affects ≥50% of women during pregnancy¹ and may cause disability with adverse impact on daily life and perceived health.² The symptoms disappear in most women within 6 months after delivery,³ but PGP may persist for several years in 10% of the affected women.⁴ Parity, overweight, strenuous work, and emotional distress are previously reported risk factors for PGP during pregnancy.⁵ The mechanisms of PGP are multifactorial but not fully understood.⁶ The high incidence of PGP in the first half of pregnancy and their regression shortly after delivery, indicate that pregnancy-related factors might be involved.⁶ Hormonal influences and non-optimal pelvic stability may be a driver for PGP⁶ but the evidence is inconsistent.^{8,9} In women with fragile connective tissue, the pelvic joints may be more vulnerable to load and to pain development.¹0 Studies have reported an association between increased pelvic mobility and PGP.¹1

Generalized joint hypermobility (GJH), defined as the ability of several joints to move beyond the normal range of motion, ¹² is a collagen phenotype that impacts the entire body. ¹³ The reported prevalence in women varies between 6% and 9% ¹⁴ and varies by sex, age, and race. ^{12,15} The association between GJH and PGP is sparsely investigated and inconclusive. ^{16,17} Women with self-reported GJH had higher odds of reporting PGP during pregnancy, especially in the first trimester. ¹⁷ However, using a clinical examination to assess the association between GJH and PGP is recommend. ¹⁸ The Beighton score (BeS) is the most commonly used instrument to assess GJH, ¹⁵ with good inter- and intra-rater reliability but with validity shortcomings. ¹⁸

Overweight is a risk factor for PGP during pregnancy¹⁹ and women with GJH and pre-pregnancy body mass index (BMI) ≥25 kg/m² reported higher evening pain than women without GJH and BMI <25 kg/m².¹⁶ The aim of the study was to investigate if women with clinically assessed GJH had an increased risk of PGP and reported higher pain intensity during and after pregnancy, than women without GJH. Our secondary aim was to study the importance of early pregnancy BMI for the risk of PGP in women with GJH.

2 | MATERIAL AND METHODS

2.1 | Study population

In this prospective cohort study, the Uppsala Pelvic Pain Study, we consecutively invited pregnant women attending three maternity care centers in two medium-sized cities in the middle of Sweden,

Key message

Pregnant women with generalized joint hypermobility reported higher pelvic pain intensity in early pregnancy. Women with a combination of general joint hypermobility and early pregnancy body mass index ≥25 kg/m² had an increased risk of pelvic girdle pain in early pregnancy.

from February 2014 to June 2019. During the study period approximately 8000 women attended the maternity care centers. Midwives invited the women to participate and offered written information by hand or post. Women interested in participation sent an email to the research group. Time for study inclusion was booked by telephone. Inclusion criteria were an ongoing pregnancy of ≤15 completed gestational weeks (GW) according to the last menstrual period and ability to read Swedish. Women with recent musculoskeletal injury or pain were excluded from the assessment of the GJH. Gestational age was revised after the ultrasound examination, if necessary.

Data were collected by self-reported web-based questionnaires and by clinical examinations at visit 1, at \leq 15 GW, at visit 2 at 36 GW, and at visit 3, 9 months after childbirth. One of the assessors (city A: a general practitioner and a physiotherapist, city B: two physiotherapists), conducted the clinical examinations and was blinded to all self-reported data.

The web-based questionnaires included questions about maternal age, non-European origin one or both parents with origin outside Europe (yes/no), completed GW, previous childbirth (yes/no), university education (yes/no), marital status (married/partnership, yes/no), use of tobacco 1 month before the current pregnancy (yes/no), history of lumbar pain and/or PGP (yes/no), and included a pain drawing with additional questions about pain onset in relation to the current pregnancy and pain intensity. The weight (kg) and height (cm) were clinically assessed and BMI was calculated as ≥25 kg/m² (yes/no).

2.2 | Exposure variables

We assessed GJH at visit 1 using the BeS, 15 following a standardized protocol and use of a goniometer. 20 The BeS comprises assessments of nine joints, passive: dorsiflexion of the fifth metacarpophalangeal joints (\leq />90°), apposition of the thumbs to the flexor aspect of the

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forearm, hyperextension of the elbows (\le />10°), hyperextension of the knees (\le />10°) and forward flexion of the trunk with knees extended and the palms easily resting on the floor. Each measurement is answered yes/no and a hypermobile joint yields one point, with a total score from 0 to 9. We used a BeS \ge 5 as the cut-off level for GJH. As in a previous study, 16 we additionally categorized the women into four subgroups based on their GJH and BMI status in early pregnancy: (1) BeS <5/9 and BMI <25 kg/m² (the reference group); (2) BeS \ge 5/9 and BMI <25 kg/m²; (3) BeS <5/9 and BMI \ge 25/ kg/m²; and (4) BeS \ge 5/9 and BMI \ge 25 kg/m².

2.3 | Outcome variables

The woman was diagnosed with PGP if she had indicated pelvic pain on the pain drawing (Figure 1) and the location was verified by at least one positive provocation test: an ipsilateral modified posterior pelvic pain provocation test (P4) for dorsal pain and a symphysis pubis pain provocation test for ventral pain. ^{6,21} P4 was modified to a semi-quantitative test with a predefined load of 1, 5, 10 or 15 kg applied to the flexed knee to put load along the longitudinal axis of femur. The test was considered positive when a familiar pain was felt in the posterior part of the pelvis on the provoked side and the lowest painful load was recorded. Palpation of the pubic symphysis was conducted with the woman in a supine position. The assessor palpated gently along the pubic symphysis. If the palpation caused pain, persisting >5 s after removal of the assessor's hand the presence of pain (yes/no) and intensity (visual analogue scale [VAS] 0–100) were recorded. ²¹

The women estimated their previous week PGP intensity by VAS, from 0 (no pain) to 100 (worst imaginable pain). The VAS is a reliable and valid tool with which to assess pain intensity.²² In the analyses,

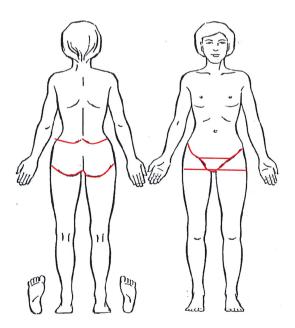


FIGURE 1 A pain drawing to indicate the distribution of pain. Pelvic girdle pain was defined as pain indicated within the red borders (not shown to the women).

pain intensity was categorized into four categories, $(0=no pain, 1-38=mild pain, 39-57=moderate pain, and <math>\ge 58=severe pain)$.

2.4 | Statistical analyses

Descriptive data are given as frequencies, proportions, or medians with range or interquartile range. Group differences were tested using the proportion test and the Wilcoxon rank-sum test (Mann-Whitney *U* test). We used logistic regression analyses to test the association of GJH with the presence of PGP and in subgroups of women who differed in GJH and BMI, estimated as crude odds ratios (OR) and adjusted odd ratios (aOR), with 95% confidence interval (95% CI). Due to a large number of zero values on the VAS, we tested differences in previous week worst pain according to GJH with ordinal logistic regression analyses, estimated as crude OR and aOR and tested for proportional odds assumption. All regression analyses were adjusted for age and origin based on directed acyclic graphs²⁴ (Figure S1) and literature search¹² to identify possible confounders of a causal association between GJH and PGP.

In sensitivity analyses, we tested if the association between GJH and PGP differed according to parity, according to time for PGP onset, including onset before current pregnancy and women with pregnancy-induced onset and to BMI ≥25 kg/m² in early pregnancy. The 5% significance level was chosen for all analyses. We used STATA V.14.0 (Stata Corp) for all analyses.

2.5 | Ethics statement

Informed written consent was obtained from all participants. The study was approved by the Regional Ethical Review Board in Uppsala, Sweden on August 28, 2013 (reference number 2013/186). The study is reported according to the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) guidelines.

3 | RESULTS

A total of 356 women conducted visit 1, (median 12 GW, range 5–18 GW), which included nine women in ultrasound-adjusted GW 16–18. In late pregnancy, 299 women (84%) conducted visit 2 (median 36 GW, range 31–39 GW) and 270 women (76%) conducted visit 3, 9 months after childbirth (median 9 months, range 7–14 months) (Figure 2).

Table 1 shows the characteristics of all the women and of women divided into without and with GJH. The median age at visit 1 was 30 years with median BMI 23.7 kg/m², 47.3% were multipara and 9.6% had GJH. The proportion of women with a university education was lower in women with GJH compared with women without GJH.

At visit 1, 47.1% of the women with GJH had PGP compared with 32.6% of women without GJH (age and origin aOR 1.76; 95% CI 0.86-3.62). At visit 2, the proportions of women with PGP had increased in both groups to 72.4% vs 70.7% (aOR 1.07; 95% CI

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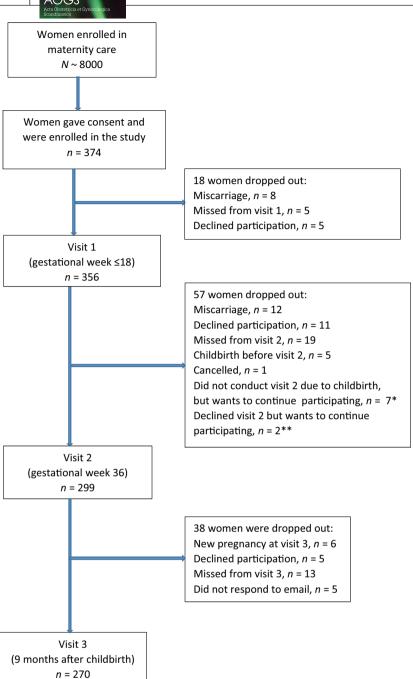


FIGURE 2 Flow chart of the participating pregnant women in the Uppsala Pelvic Pain Study. *Childbirth before visit 2, but participated at visit 3, n=7; **declined visit 2, but participated at visit 3, n=2.

0.45–2.54). At visit 3, 25.0% of the women with GJH had PGP compared with 23.2% of the women without GJH (aOR 1.01; 95% CI 0.37–2.74) (Table 2). The risk of having PGP at visit 1 was highest for women with GJH and BMI $\geq 25 \, \text{kg/m}^2$ (aOR 6.88; 95% CI 1.34–35.27) compared with women with normal joint mobility and BMI $< 25 \, \text{kg/m}^2$. At visits 2 and 3, the corresponding estimated associations were weaker and did not reach statistical significance (Table 2).

At visit 1, 121 of 356 women (34%) were diagnosed with PGP, of which 57% had a unilateral or bilateral positive P4-test, 21.5% had a positive symphysis pain provocation test, and 21.5% had a combination of both. At visit 2, 212 of 299 women (70.9%) were diagnosed with PGP, of which 30.7% had a unilateral or bilateral positive P4-test, 22.6% had a positive symphysis pain provocation test,

and 46.7% had a combination of both. At visit 3, 63 of 270 women (23.3%) were diagnosed with PGP, of which 60.3% had a unilateral or bilateral positive P4-test, 23.8% had a positive symphysis pain provocation test, and 15.9% had a combination of both (not shown).

Women with GJH had a higher risk of reporting higher pain intensity during the last week compared with women with normal joint mobility (aOR of 2.04; 95% CI 1.02–4.07) at visit 1, but not at visit 2 and 3 (Table 3).

A higher proportion of women with GJH reported PGP onset before the current pregnancy compared with women without GJH, 29.4% vs 14.3% (p=0.02). The proportion of women with PGP onset during pregnancy (n=192) was higher among women without GJH, 55.6% vs 38.3 (p=0.05), where a higher proportion of women with

TABLE 1 Characteristics of the study population (n=356) in visit 1 and stratified by generalized joint hypermobility.

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	All women		gene	Women with generalized joint hypermobility ^a		Women without generalized joint hypermobility ^a		
	N=3	56	N=	34 (9.6%)	N=32	22 (90.4%)		
Variable	n		n		n			
Age (years), median (range)	356	30 (19-45)	34	30 (20-38)	322	31 (19-45)		
BMI ^b (kg/m²), median (IQR)	353	23.7 (21.7-26.5)	34	22.4 (19.8-24.9)	319	23.7 (21.8-26.5)		
Origin outside Europe ^c , <i>n</i> (%)	355	29 (8.2)	34	5 (14.7)	321	24 (7.5)		
Gestational week ^d , median (range)	356	12 (5-18)	34	11 (5-15)	322	12 (6-18)		
Parous, n (%)	355	168 (47.3)	34	17 (50.0)	321	151 (47.0)		
University education, n (%)	354	246 (69.5)	34	18 (52.9)	320	228 (71.2)		
Marriage/ partnership, n (%)	354	337 (95.2)	34	32 (94.1)	320	305 (95.3)		
Pre-pregnancy smoking, n (%)	352	17 (4.8)	34	2 (5.9)	318	15 (4.7)		
Pre-pregnancy snuffing, n (%)	354	27 (7.6)	34	2 (5.9)	320	25 (7.8)		
History of back pain, n (%)	354	119 (33.6)	34	13 (38.2)	320	106 (33.1)		
History of pelvic girdle pain, n (%)	355	35 (9.9)	34	4 (11.8)	321	31 (9.7)		

Abbreviations: BMI, body mass index; IQR, interquartile range.

GJH had onset in early pregnancy compared with the women without GJH, 46.2% vs 32.4% (p=0.31) (Table 4).

We observed a tendency in sensitivity analyses that childbirth increased the odds for women with GJH to have PGP in early pregnancy and 9 months after childbirth. Increased odds for PGP were also seen for women with early BMI \geq 25 kg/m² during and after pregnancy and for women with pregnancy-induced PGP, in early pregnancy. None of the analyses were statistically significant (Table 5).

4 | DISCUSSION

We saw no statistical evidence that women with GJH had an increased risk of PGP during pregnancy or 9 months after childbirth. Women with clinically assessed GJH had a higher risk of reporting

higher pain intensity in early pregnancy compared with women without GJH. Furthermore, women with GJH and early pregnancy BMI $\geq 25\,\mathrm{kg/m^2}$ had the highest odds of having PGP in early pregnancy compared with women without GJH and early pregnancy BMI $<25\,\mathrm{kg/m^2}$.

The prospective study design including a 9-month follow up is a strength in our study. We collected data from web-based questionnaires, pain drawings, and clinical assessments. With predetermined criteria, we identified PGP by pain drawings and via clinical tests. The experienced assessors used a goniometer and followed a standardized protocol when assessing GJH. The follow-up rate was high, 83.9% from early to late pregnancy and 75.8% from early pregnancy to 9 months after childbirth. With the recommended cut-off value \geq 5/9 in the BeS, the prevalence of GJH was 9.6%. This is in accordance with a recent study from Norway on pregnant women.

^aGeneralized joint hypermobility was assessed with clinical assessment and defined as sum score ≥5 in the Beighton score (0-9).

^bHeight and weight were measured at visit 1 for calculation of body mass index.

^cOrigin outside Europe, defined as ≥1 parent with origin outside Europe.

^dGestational week, primarily based on ultrasound and secondarily on the last menstrual period.

TABLE 2 The associations between generalized joint hypermobility and pelvic girdle pain in early pregnancy (GW^g 5-18), late pregnancy (GW^g 31-39) and 9 months after childbirth, expressed as crude and adjusted odds ratios with 95% confidence intervals. The association is also presented for subgroups of women (N = 353)8 who differ on generalized joint hypermobility and early pregnancy body mass index (kg/m²).

Allw	All women, N Cases PGP^a , n (%)		Crude OR 95	95% CI	p value	Adjusted OR ^c	95% CI	p value
Early pregnancy								
GJH ^b no 322	105 (32.6)		Reference Re	Reference		Reference	Reference	
GJH ^b yes 34	16 (47.1)		1.84 0.9	0.90-3.75	0.09	1.76	0.86-3.62	0.12
Late pregnancy								
GJH ^b no 270	191 (70.7)		Reference Re	Reference	0.85	Reference	Reference	
GJH ^b yes 29	21 (72.4)		1.09 0.4	0.46-2.55		1.07	0.45-2.54	0.88
9 months after childbirth								
GJH ^b no 246	57 (23.2)		Reference Re	Reference		Reference	Reference	
GJH ^b yes 24	6 (25.0)		1.10 0.4	0.42-2.92	0.84	1.01	0.37-2.74	0.72
Combined GJH ^b and BMI ^d	All women ^f , N	Cases PGPª, n (%)	%) Crude OR	95% CI	p value	Adjusted OR ^c	95% CI	p value
Early pregnancy (GW ^e ≤18)								
GJH no and BMI $<$ 25 kg/m 2	202	59 (29.2)	Reference	Reference		Reference	Reference	
GJH yes and BMI $<$ 25 kg/m 2	2 25	9 (36.0)	1.36	0.57-3.26	0.49	1.31	0.54-3.14	0.55
GJH no and BMI $\geq 25 \text{kg/m}^2$	118	46 (39.0)	1.55	0.96-2.50	0.07 < 0.02	1.52	0.94-2.46	0.09 < 0.03
GJH yes and BMI ≥25kg/m²	8	6 (75.0)	7.27	1.43-37.07		6.88	1.34-35.27	
Late pregnancy (GW ^e >32–39)								
GJH no and BMI $<$ 25 kg/m 2	178	121 (68.0)	Reference	Reference		Reference	Reference	
GJH yes and BMI $<$ 25 kg/m 2	2 20	13 (65.0)	0.87	0.33-2.31	0.79	0.85	0.32-2.28	0.75
GJH no and BMI ≥25 kg/m²	91	70 (77.0)	1.57	0.88-2.81	0.13	1.60	0.89-2.86	0.12
GJH yes and BMI ≥25kg/m²	89	7 (87.5)	3.37	0.40-27.44	0.27	3.31	0.40-28.22	0.26
After childbirth (≤9 months)								
GJH no and BMI $<$ 25 kg/m 2	166	34 (20.5)	Reference	Reference		Reference	Reference	
GJH yes and BMI $<$ 25 kg/m 2	2 17	3 (17.6)	0.83	0.23-3.06	0.78	0.73	0.19-2.79	0.65
GJH no and BMI $\geq 25 \text{kg/m}^2$	78	23 (29.5)	1.62	0.88-3.00	0.12	1.71	0.92-3.19	0.09
GJH yes and BMI ≥25kg/m²	7	3 (42.9)	2.91	0.62-13.63	0.18	2.83	0.59-13.52	0.19

Note: The association between generalized joint hypermobility and pelvic girdle pain was estimated by using crude and adjusted logistic regression analysis.

Abbreviations: BMI, body mass index; CI, confidence interval; GJH, generalized joint hypermobility; GW, gestational week; OR, odds ratios; PGP, pelvic girdle pain.

^eGestational week primarily based on ultrasound and secondarily on the last menstrual period.

^aPelvic girdle pain was indicated on a pain drawing and verified with clinical tests.

^bGeneralized joint hypermobility was assessed with clinical assessment and defined as sum score ≥5 in the Beighton score (0-9).

Odds ratios adjusted for age (based on year of birth) and origin outside Europe, defined as ≥1 parent with origin outside Europe.

^dBody mass index, height and weight were measured at visit 1.

 $^{^{}f}$ N=353 due to exclusion of three women with missing data values on body mass index.

TABLE 3 The odds ratio of women with generalized joint hypermobility to report higher pain scores (divided into no pain, mild pain, moderate pain, and severe pain) compared with women without generalized joint hypermobility, in early pregnancy, in late pregnancy, and 9 months after childbirth.

GJHª	Women, N	Median (IQR)	q d	VAS ^c , 0, <i>n</i> (%)	VAS ^c , 1-38, n (%)	VAS ^c , 39-57, VAS ^c , ≥58, n (%) n (%)	VAS ^c , ≥58, n (%)	Crude OR ^d	95% CI	a	Adjusted OR ^e	95% CI	a
Early pregnancy GW ^f 5-18	y GW ^f 5-18												
No GJH	321	0 (0-19)		220 (68.5)	50 (15.5)	20 (6.2)	32 (10.0)						
GJH	34	0 (0-40)	0.04	18 (52.9)	3 (8.8)	7 (20.6)	6 (17.7)	2.16	1.09-4.28	0.03	2.04	1.02-4.07	0.04
Late pregnancy GW ^f 31–39	, GW ^f 31-39												
No GJH	270	37 (0-64)		84 (27.0)	50 (16.1)	38 (12.2)	139 (44.7)					0.58-2.29	
GJH	29	49 (0-64)	0.52	8 (25.0)	4 (12.5)	4 (12.5)	16 (50.0)	1.21	0.61-2.40	0.59	1.15		69.0
9 months after childbirth	childbirth												
No GJH	246	(0-0) 0		190 (60.9)	26 (8.3)	11 (3.5)	85 (27.3)						
СЈН	24	0 (0-1)	0.78	18 (54.5)	2 (6.1)	1 (3.0)	12 (36.4)	1.38	0.69-2.79	0.37	1.24	0.60-2.54	0.56

Note: The association between generalized joint hypermobility and pelvic girdle pain intensity was estimated by using crude and adjusted ordinal logistic regression analysis. Abbreviations: CI, conficence interval; GJH, generalized joint hypermobility; GW, gestational week; IQR, interquartile range; OR, odds ratios; VAS, visual analogue scale.

^a Generalized joint hypermobility was assessed with clinical assessment and defined as sum score ≥5 in the Beighton score (0-9).

bThe probability of no difference in median difference in pelvic girdle pain intensity between the groups was tested with Wilcoxon rank-sum test (Mann–Whitney U test).

Visual analogue scale 0-100, divided into 0=no pain, 1-38=mild pain, 39-57=moderate pain and ≥58=severe pain.

^dOR, odds ratio for a women with generalized joint hypermobility to be included in a higher category of pain intensity compared with women with normal mobility.

eOdds ratios adjusted for age (based on year of birth) and origin outside Europe, defined as ≥1 parent with origin outside Europe.

^fGestational week based on ultrasound and on the last menstrual period.

TABLE 4 The timing of pelvic girdle pain onset in all women (N=356), and stratified by joint mobility.

	All women	Women with generalized joint hypermobility ^a	Women without generalized joint hypermobility ^a	
	N=356	N = 34 (9.6%)	N=322 (90.4%)	p value
Onset of pelvic girdle pain ^b				
Onset before pregnancy, n (%)	56 (15.7)	10 (29.4)	46 (14.3)	0.02
Onset during pregnancy, n (%)	192 (53.9)	13 (38.3)	179 (55.6)	0.05
Onset in $GW^c \le 18$, n (%)	64 (33.3)	6 (46.2.)	58 (32.4)	0.31
Onset after $GW^c > 18$, n (%)	128 (66.7)	7 (53.8)	121 (67.6)	0.31
Onset after childbirth d , n (%)	5 (1.4)	1 (2.9)	4 (1.2)	0.40 ^f
Unknown debut of PGP ^e , n (%)	27 (7.6)	3 (8.8)	24 (7.5)	0.73 ^f

Note: The probability of no difference in proportion of pelvic girdle pain onset between the groups was tested with proportional test.

Abbreviations: GW, gestational week; PGP, pelvic girdle pain.

TABLE 5 Sensitivity analyses for the relation between generalized joint hypermobility and pelvic girdle pain in women having previous childbirths, pelvic girdle pain onset before the current pregnancy, onset during pregnancy, and women who differed in early pregnancy body mass index.

The association of generalized joint					
hypermobility ^d with pelvic girdle pain ^e	Women n	Crude OR	95% CI	Adjusted OR ^f	95% CI
Parous women					
Visit 1 ^a	168	2.49	0.46-3.73	2.67	0.90-7.89
Visit 2 ^b	143	0.67	0.21-2.10	0.83	0.24-2.83
Visit 3 ^c	127	1.71	0.48-6.14	1.87	0.46-7.67
Women with pelvic girdle pain onset before the	e current pregnancy				
Visit 1 ^a	56	4.24	0.88-0.52	3.92	0.79-13.94
Visit 2 ^b	45	2.25	0.26-9.26	2.29	0.26-20.35
Visit 3 ^c	34	3.30	0.56-19.5	3.79	0.59-24.31
Women with pregnancy-induced pelvic girdle p	ain				
Visit 1 ^a	192	1.79	0.57-5.56	1.58	0.50-4.99
Visit 2 ^b	176	1.00	omitted	1.00	omitted
Visit 3 ^c	156	0.58	0.12-2.82	0.49	0.09-2.54
Women with BMI ^g ≥25 kg/m ²					
Visit 1 ^a	126	4.70	0.91-4.27	4.47	0.86-23.30
Visit 2 ^b	99	2.10	0.24-8.05	2.00	0.23-17.52
Visit 3 ^c	85	1.79	0.37-8.66	2.38	0.44-13.01

Note: The association between generalized joint hypermobility and pelvic girdle pain was estimated by using crude and adjusted logistic regression analysis.

Abbreviations: BMI, body mass index; CI, confidence interval; OR, odds ratios.

 $^{^{}a}$ Generalized joint hypermobility defined as sum score ≥5 in the Beighton score (0–9).

^bPelvic girdle pain was indicated on a pain drawing and verified with clinical tests.

^cGestational week based on ultrasound or last menstrual period.

^dOnset after childbirth within 9 months after childbirth.

 $^{^{\}mathrm{e}}$ Unknown debut of PGP refers to women without PGP who dropped out after visit 1.

^fFisher's exact test.

^aVisit 1, in gestational week 5–18.

^bVisit 2, in gestational week 31–39.

^cVisit 3, 9 months after childbirth.

^dGeneralized joint hypermobility was assessed with clinical assessment and defined as sum score ≥5 in the Beighton score (0-9).

^ePelvic girdle pain was indicated on a pain drawing and verified with clinical tests.

fOdds ratios adjusted for age (based on year of birth) and origin outside Europe, defined as ≥1 parent with origin outside Europe.

 $^{^{\}rm g}$ Body mass index (kg/m²), height and weight were measured at visit 1.

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Despite the experienced assessors and the use of the standardized protocol, some women may have been misclassified by GJH. However, we assume the risk of misclassification of GJH in relation to PGP was low, because the assessors were blinded to the initial self-reported data and started the clinical examination with the assessment of GJH.

The prevalence of PGP during pregnancy varies between 7% and 76%.^{6,7} The variation may rely on differences in design, diagnostic procedure, GW for PGP assessment, and the definition of PGP.⁶ In our study, the prevalence of PGP increased from 34.0% in early pregnancy to 70.9% in late pregnancy, where 15.7% of the women reported pain onset before current pregnancy. A limitation of our study is that we only used an ipsilateral P4 test to verify dorsal pelvic pain on the pain drawing and a symphysis pain provocation test to verify ventral pelvic pain. The fact that we did not use several pain provocation tests may have increased the risk of misclassification of PGP, which could have influenced the association between GJH and PGP.⁶

In our study, about one-fourth of the women had PGP 9 months after childbirth regardless of GJH. However, 29.4% of the women with GJH and 14.3% of the women without GJH reported PGP before the current pregnancy, which may be an explanation for the high prevalence of women where PGP remains. The prognosis of PGP is good and the symptoms regress shortly after delivery for most women.²⁵ But the prevalence of women with long-standing PGP after childbirth varies.^{6,25} Previous low back pain and early PGP onset during pregnancy have been reported as risk factors for more severe PGP.²⁶ One study showed that 37% of the women reported "some back pain" 12 months after childbirth and 7% reported "serious back pain" 18 months after childbirth.²⁷

We found no statistical evidence that women with GJH had an increased risk of having PGP during or after pregnancy, which is partially in accordance with a previous study showing no increased odds of having PGP in women with GJH in GW 30.¹⁶ However, we observed some trends that women with GJH may suffer from PGP earlier in pregnancy than women without GJH. A larger proportion of women with GJH had pelvic pain onset before the current pregnancy and earlier pelvic pain onset in pregnancy-induced PGP compared with women with normal joint mobility. In contrast to the present study, where our estimates were imprecise with wide confidence intervals, we found an association in a previous study between self-reported GJH and PGP in women with GJH in early pregnancy (aOR 1.54: 95% CI 1.20–1.96) and for the entire pregnancy (aOR 1.27: 95% CI 1.11–1.47).¹⁷

A higher proportion of women with GJH reported moderate and severe pain in the beginning of pregnancy compared with women without GJH. But with few women in the GJH-group, our results must be interpreted with caution because of the risk of imprecise estimates and low statistical power. The reflection on "worst pain last week", could also have introduced some recall bias. A previous study reported no increased evening pain in women with GJH compared with women without GJH in GW 30. But comparison with our study is difficult because of differences in reported outcomes.

As PGP often starts in the early stages of pregnancy and commonly disappear shortly after childbirth, pregnancy-related factors have been suggested as one of the causes of PGP. 7,25 Relaxin is a polypetide hormone that relaxes the connective tissues in the pelvis during pregnancy.²⁸ The hormone level increases and peaks during the first trimester.8 As relaxin has been reported as a driver for collagen remodeling with relaxation of the pelvic ligaments, women with GJH are perhaps more vulnerable to its rapid increase in early pregnancy. Even though the association between female sex hormones and PGP is disputed, 8,9 our result with higher odds for PGP in early pregnancy in women with GJH and BMI ≥25 kg/m² may correspond to increasing pregnancy hormone levels in early pregnancy.⁸ The increased hormone levels may alter joint mobility, 6,11 which perhaps has a stronger impact in women with a combination of GJH and high BMI, due to elastic connective tissues²⁹ and high levels of estrogen or inflammation in adipose tissues. 30

Our results should be interpreted with caution because low statistical power may have affected our findings. These results need to be verified in future studies with a larger samples of women with GJH before we can tell whether women with GJH or a combination of GJH and high BMI have an increased risk of pelvic pain during and after pregnancy.

5 | CONCLUSION

GJH did not increase the risk of PGP during or after pregnancy. However, women with combined GJH and overweight in early pregnancy showed an increased risk of PGP in early pregnancy. In addition, women with GJH had higher pain intensity in early pregnancy compared with women without generalized joint mobility. These results need to be verified in future studies with a larger sample of women with GJH before we can tell if pregnant women will benefit from GJH assessment in order to reduce the risk of PGP, reduce the pain intensity, and delay the pain onset in pregnancy, particularly for women with a combination of GJH and high BMI.

AUTHOR CONTRIBUTIONS

Concept and design: KA, EKB, LNW, MP, and PK. Drafting the manuscript: KA. Statistical analysis: KA, and RP. All authors: acquisition, analysis and interpretation of the data and critical revision of the manuscript for important intellectual content. All authors approved the final submitted manuscript.

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CONFLICT OF INTEREST STATEMENT

The authors have stated explicitly that there are no conflicts of interest in connection with this article.

DATA AVAILABILITY STATEMENT

The data used for this study will be publicly available through the link http://urn.kb.se/resolve?urn=urn:nbn:se:uu:diva.

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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