

Risk factors for persistent pelvic girdle pain pregnancy-related (PPGP): a Systematic Review and meta-analysis approach

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July 1, 2022

Abstract

Our review want to identify most incident risk factors that determine the persistence of PGP at 3-6 months after childbirth in women with PPGP or PPGP and PLBP, because of about 1/3 does not recover after childbirth and continues to experience symptoms after three months and in some cases up to two years. The research was performed on the databases of Medline, Cochrane, Pedro, Scopus, Web of Science and Chinal from December 2018 to January 2022 following the indications of the PRISMA statement 2009 - and updated according to the PRISMA 2020- including observational cohort studies and prospective questionnaires in English. Two authors independently selected studies excluding specific, traumatic, gynecological / urological cause PGP or isolated PLBP and studies that did not include the primary outcome (presence / absence of PGP); studies with an initial assessment in pregnancy / within one month of delivery and with at least a follow-up at least 3 months after delivery were included. Two independent authors then performed an evaluation of the ROB using the QUIPS tool. Finally, in-depth qualitative analysis was conducted, since due to high degree of heterogeneity in the data collection of the included studies and lack of raw data suitable for quantitative analysis, it was not possible to carry out the originally assumed meta-analyses for subgroups. High levels of pain in pregnancy, high number of positive provocative tests, history of LBP / LPP, high levels of disability in pregnancy, neurosis and high levels of Fear Avoidance Belief are main predictors of PPGP.

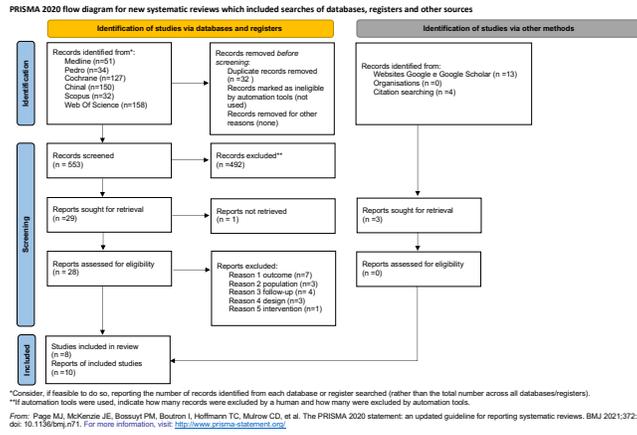


Figure 1

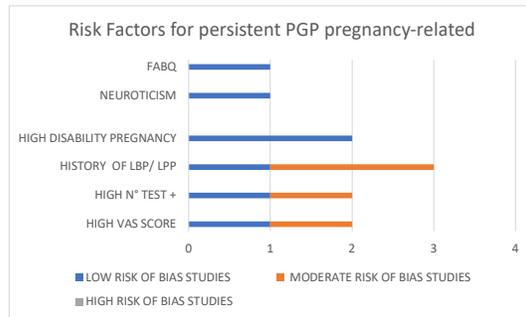


Figure 2

Risk factors for persistent pelvic girdle pain pregnancy-related (PPGP): a Systematic Review and meta-analysis approach

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Abstract

Background

PPGP is a condition that affects 20% of pregnant women; about 1/3 does not recover after childbirth and continues to experience symptoms after three months and in some cases up to two years. It is not clear why only some women with PPGP recover, and for this reason the identification of risk factors would enable the early identification of women at risk of chronicity and would make it possible to define prevention strategies for those modifiable risk factors.

Objectives

To identify the most incident risk factors that determine the persistence of PGP at 3-6 months after childbirth in women with PPGP or PPGP and PLBP.

Search Strategy

The research was performed on the databases of Medline, Cochrane, Pedro, Scopus, Web of Science and Chinal from December 2018 to January 2022 following the indications of the PRISMA statement 2009 - and updated according to the PRISMA 2020- including observational cohort studies and prospective questionnaires in English.

Selection criteria

Two authors independently selected studies excluding specific, traumatic, gynecological / urological cause PGP or isolated PLBP and studies that did not include the primary outcome (presence / absence of PGP); studies with an initial assessment in pregnancy / within one month of delivery and with at least a follow-up at least 3 months after delivery were included. Two independent authors then performed an evaluation of the ROB using the QUIPS tool.

Data collection and analysis

Finally, in-depth qualitative analysis was conducted, since due to high degree of heterogeneity in the data collection of the included studies and lack of raw data suitable for quantitative analysis, it was not possible to carry out the originally assumed meta-analyzes for subgroups.

Main results

The research process led to the inclusion of 10 articles which were evaluated using the QUIPS tool: 7 studies were evaluated as low ROB and three as moderate ROB.

The most predictive risk factors of persistent PGP 3-6 months after childbirth seem to be in line with the literary landscape relating to the topic.

Conclusions

High levels of pain in pregnancy, high number of positive provocative tests, history of LBP / LPP, high levels of disability in pregnancy, neurosis and high levels of Fear Avoidance Belief are predictors of persistent PPGP, while there is weak or contradictory evidence regarding emotional distress, catastrophization, sleep disturbances. The impossibility of carrying out the meta-analysis by subgroups, suggests the need for further research with greater methodological rigor in the acquisition of outcome measures.

Keywords:

PGP pregnancy-related, PGP post-partum, persistent PGP, risk factors, systematic review

Introduction

Pelvic Girdle Pain (PGP) identifies a pain that arises in relation to pregnancy, trauma, arthritis or osteoarthritis; it is felt between the posterior iliac crest and the gluteal line, in particular near the sacroiliac joint. Pain may be referred down the thigh posteriorly and may or may not occur in association with symphysis pain^[1].

It represents a multifactorial condition with partly unknown etiology: we are talking about biomechanical, traumatic, metabolic, genetic and degenerative factor^[2].

PGP is a frequent condition during pregnancy (Pregnancy-related Pelvic Girdle Pain-PPGP), it can occur together with PLBP (Pregnancy-related Low Back Pain) or less frequently separately. It has a prevalence ranging between 23% and 65% of women, with some variability based on how the disorder is defined and measured^[3]. 50% of pregnant women suffer from PLPP, and amongst them 20% experience PPGP.

Given the important heterogeneity of terminology in the literature, the incidence varies between 4% and 76%. Considering only the PPGP (excluding urological/gynecological causes) according to the definition of the "European Guidelines" of Cost Action 13, the incidence drops to 20%, with a high rate of misdiagnosis^{[4],[5]}.

PPGP can occur in the first trimester of pregnancy (usually at the end), during delivery or in the first month postpartum^[4]. The peak of symptoms generally occurs between the 24th and 36th week of gestation^{[6],[7]} and their localization may change during pregnancy ^{[8],[9],[10]}.

Although 78% of women recovers spontaneously 6 weeks after delivery ^[44], about 1/3 keeps on showing symptoms after three months and about 8.5% has important symptoms after two years.

According to Clinton's 2017 guidelines, the factors that determine the persistence of PGP in "late pregnancy" and post-partum are represented by: early onset of pain, localization of pain in several points, high number of positive provocation tests, dissatisfaction at work, low expectation of recovery^[11].

In particular, the high intensity of pain and the number of painful sites have been identified as important factors in the transition from acute / subacute pain to chronic pain and persistent disability^[12],^[13].

The altered body perception has been associated with altered motor control in subjects with LBP and the same relationship can be identified in individuals with persistent PPGP^[14].

Regarding the psychological domains, high levels of emotional distress^[15], catastrophization^[16] and the patient's poor expectation of recovery are identified as potential risk factors for persistence^[17]. In particular, emotional distress during pregnancy has been associated with persistent PGP at six months^[18].

Additional factors such as sleep disorder, back pain, headache and fibromyalgia are considered important for the persistence of PGP^[19].

Pain characteristics and psychological risk factors are therefore considered important in women who develop persistent PGP^[20] and for most of them no anatomical abnormalities and/or specific processes that can be identified through diagnostic tests have been found.^[21]

As shown by the 2018 review by Wuytack, F., despite the increase in interest and the number of international publications on PGP over the past twenty years, there are few studies dealing with PPGP, and it can be noted as the postpartum period is poorly studied in the literature. Studies dealing with it such as that of Sakamoto et al. of 2019 [53] do not comply with adequate methodological standards, giving us uncertain answers.

It is not clear why only some women with PPGP recover, and for that reason the identification of persistency risk factors would allow to early target women at risk of chronicity and then to work out management strategies for those modifiable risk factors.

The goal of this systematic review is to identify which are mostly affecting modifiable risk factors in determining the persistence of PGP at 3 and 6 months post-partum in women with PGP or PGP and pregnancy-related LBP.

METHODS

Research strategy

The search was performed by reviewing the literature on the Medline, Pedro, Chocrane, Scopus, Web of science and Cinhal databases from December 2018 to January 2022; only articles in English and published after 2000 were included.

The search string and keywords used were:

MEDLINE: (((“pelvic girdle pain”) OR “pelvic girdle pain”[MeSH Terms]) OR “pelvic girdle pain postpartum”) OR “pelvic girdle pain pregnancy-related”) AND ((“risk factors”) OR "risk factors"[MeSH Terms])

51 articoli

PEDRO simple search : pelvic girdle pain pregnancy **34 papers** (26 RCT, 8 SR)

COCHRANE: pelvic girdle pain **1 cochrane protocol e 127 trials**

CHINAL ((pelvic girdle pain OR pelvic girdle pain pregnancy-related OR pelvic girdle pain postpartum)) AND (risk factors or contributing factors or predisposing factors) **150 papers**

SCOPUS (TITLE-ABS-KEY (pelvic AND girdle AND pain OR pelvic AND girdle AND pain AND pregnancy-related OR pelvic AND girdle AND pain AND postpartum) AND TITLE-ABS-KEY (risk AND factors)) **32 papers**

WBOFSCIENCE ((pelvic girdle pain OR pelvic girdle pain pregnancy-related OR pelvic girdle pain postpartum)) AND

(risk factors or contributing factors or predisposing factors) **158 papers**

Study selection

Eligibility criteria for the selection of articles have been defined.

- ***Population:*** Articles concerning women with PGP or LPP or PGP + LBP with no age limit, without stratification by number of births, or by type of birth, were included in the study.
- ***Outcome assessment:*** women were followed in the studies in prospective longitudinal observational way, without therapeutic interventions, evaluated through self-reported questionnaires and/or clinical examination. Considering that the onset of PGP is located in time between the end of the first trimester of pregnancy and the first month postpartum (including the stage of labor), we have included studies with initial assessment in pregnancy or within 1 month of delivery and with at least one follow-up at least 3 months after delivery or beyond.
- ***Primary outcome:*** presence / absence of PGP according to the definition of Vleeming, 2008 and Kanakaris, 2011.
- ***Secondary Outcome:*** disability, depression, catastrophization, quality of life, sleeping disorder.
- ***Types of studies:*** observational studies of prospective cohorts and prospective questionnaires.

The **exclusion criteria** have also been defined

- Studies concerning specific PGP (inflammatory diseases, fractures, osteoporosis, neoplasia, other severe pathologies), traumatic PGP, PGP from gynecological/urological causes, PLBP taken in isolation and studies on biological risk factors only were excluded, while those studies in which both psychosocial and biological risk factors are analyzed were included.

- Failure to include the primary outcome presence/absence of PGP (VAS) is a criterion for exclusion of the studies; the same does not apply for secondary outcome measures which may appear heterogeneously in the studies, and which will be analyzed in the meta-analysis if the minimum homogeneity criteria are met.

The methodological process that led to the selection and inclusion of 10 articles by two independent authors (E.B. and S.M.) is summarized in ***Figure 1*** and ***Table 1***; any disagreement was resolved through discussion with a third author (G.G.). The table with the characteristics of the included studies is in the appendix (***Annex I***).

Risk of bias assessment

We analyzed the risk of bias (ROB) via evaluation by two independent authors using the *Quality In Prognosis Studies* (QUIPS) tool and resolved discrepancies via discussion.

For the application of QUIPS we referred to *Hayden A.* and colleagues^[22], while with reference to *Wuytack* we assigned each item a "-" when it was evaluated as *low risk of bias*, "+/-" as *moderate risk of bias* and "+" as *high risk of bias*.

The items considered are present in ***table 2***. For the global assessment of the degree of risk of the individual studies ("overall") we also referred to the *Scottish Intercollegiate Guidelines Network* (*SIGN*) for the evaluation of cohort studies which considers a high quality study ("+++") (*low ROB*) if the *majority* of the criteria is met, acceptable ("++") (*moderate ROB*) if *most* of the criteria are met, while low quality ("+") (*high ROB*), if many of the criteria are not met.

Referring to this we have given a *low* ROB to those studies in which more than half of the criteria were *low* ROB, a *moderate* ROB to those studies where there was not a majority of criteria met or they were partially met, and a *high* ROB to those studies where more than half of the criteria were *high* ROB.

To avoid interpretation mistakes, we have also kept *Wuytack, F* and colleagues symbology for the "overall" section. and we have not used that one present in the SIGN.

Meta analysis

Among the articles included it was possible to create 3 subgroups (VAS, previous LBP and number of positive tests), but it was not possible to conduct meta-analysis due to the heterogeneity of the outcome measures and the not sufficient amount of numerical data to enter in the quantitative calculation (Annex 3).

RESULTS

The 10 included studies were qualitatively assessed using the QUIPS tool.

During the evaluation with the QUIPS tool 7 studies were overall *low* ROB, 3 studies were evaluated as *moderate* ROB.

The final results of the ROB assessment are shown in **Table 2**, while in the appendix (**Annex 2**) there is the evaluation of the articles with the specifications for each domain considered by the QUIPS tool.

The 10 articles included in the systematic review are all prospective cohort studies. In 6 of the included studies the cohort was made up of pregnant women with PGP^{[28],[29],[24],[30],[25],[27]}, in 1 of healthy pregnant women^[23], while in 3 both women with LPP and healthy^{[26],[54],[52]}.

The origin of the cohort is from a previous cross-sectional in 2 studies^{[23],[29]} and from a previous retrospective study in 1^[25], while in Bergstrom, 2017^[24] the cohort was recruited in 2002.

In 6 out of 10 studies women were assessed both by questionnaire and by physical examination^{[28],[23],[30],[25],[27],[52]}, in 4 studies only by questionnaire^{[29],[24],[26],[54]}.

The primary outcome of all included studies is persistent PGP which is assessed as presence/absence in 3 out of 8 studies^{[24],[26],[28]}, by *VAS/NPRS* in 6 studies^{[29],[30],[25],[27],[52],[54]} and through *NPRS* and *Mc Gill* in 1^[23].

The baseline outcome assessment is done during pregnancy or within one month from delivery in all the included studies, while a certain variability is shown for the execution of the follow-up.

In 6 studies it covers beyond 6 months post-partum^{[28],[23],[29],[27],[52],[54]} while in 1 up to 12 years^[24].

The secondary outcomes analyzed are heterogeneous, but in 5 out of 10 studies one of the secondary outcomes is disability, which is evaluated with heterogeneous outcome measures: *PGQ*^{[23],[25]}, *DRI*^{[30],[54]}, *ODI*^[27]. Self-reported health status and quality of life are often considered the only factor and represent secondary outcomes in 5 out of 10 studies; also in this scenario we find a certain heterogeneity in the outcome measures: *HR-QoL*^[23], *EQ-5D*^[25], *SF-36*^[27], *SRH*^[29] and *NHP*^[54].

The risk factors analyzed are heterogeneous and therefore we decide to report the results for each study, trying finally to form subgroups for each risk factor.

Albert H. et al. 2001, identified in the subgroup of women with PGS (n=100) the group with the worst prognosis within which they identified six risk factors correlated with the persistence of pain at two years: advanced age (≥ 29 years; RR=1.9; $p \leq 0.05$), poor education (RR=2.3; $p \leq 0.05$), non-qualifying work or unemployment ($p \leq 0.05$), high pain intensity ($VAS \geq 6$; RR=1.6; $p \leq 0.05$), low test indices mobility (≤ 320) (RR=3.9; $p \leq 0.005$), high number of positive provocative tests (≥ 16) (RR=10.7; $p \leq 0.001$).

Beales D. et al., 2018 followed up on a group of women (n=29) on average at 15 months postpartum (SD=2.0) who had low to moderate levels of disability and pain at baseline (*PGQ*=28, SD=26; *NRS*=2, SD=3) and that in 41% of cases (n=12) reported continuous pain at follow-up. From the analysis made by using the Spearman correlation coefficient of three characteristics present at baseline (*ASLR* performance, sleep quality through *PSQI* and *PPT* in five parts of the body) and pain intensity (*NRS*), pain quality (*Mc Gill*), disability (*PGQ*) and quality of life (*SF-36*) at 15 months postpartum, it is highlighted that poor performance at *ASLR* during pregnancy correlates statistically significantly with

a low quality of life at 15 months postpartum ($p = -0.558$) and that a reduced PPT at the level of the sacrum during pregnancy correlates with high Mc Gill scores ($p = -0.384$). The presence of sleeping disorders in pregnancy is not correlated with any of the outcomes analyzed.

According to Bergstrom C. et al., 2014 women with a history of LBP before delivery were 2.47 times more likely to report "recurrent pain" (OR=2.47; p -value=0.030) and of 3.35 to report "continuous pain" (OR=3.35; p -value=0.024) at follow-up at 14 months postpartum compared to those who at 14 months had had a remission of symptoms ("no pain"); the presence of LBP before pregnancy is a strong predictor of pain 12-14 months postpartum.

High levels of pain during pregnancy and in the first six months postpartum identify a worse outcome at 14 months after delivery: this finding is in contrast to Olsson, 2012 which does not identify this as a risk factor for long-term LPP.

For Bergstrom C. et al., 2017 the most important predictor of poor outcome for women with PPGP at 12 years seems to be "wide spread pain": the outcome in fact finds a statistically significant correlation with the presence of sciatica (OR=3.4 (1.87-6.20); $p < 0.0001$) and neck/thoracic pain (NP/TP) (OR=2.50 (1.40-4.48); $p = 0.002$). The presence of NP/TP for more than 30 days in the last 12 months is associated with increase in the OR of 2.03 ((1.06-3.87); p -value=0.03).

According with Robinson H. et al., 2010 (b), women that would show 3-4 painful areas during physical examination in pregnancy (CI=18.7(7.9, 29.6); p -value=0.007) or more than 6-8 positive provocative test (CI=11.2(2.4, 19.8); p -value=0.04) had statistically significantly higher pain levels at 12 weeks postpartum. A pre-pregnancy BMI of ≥ 25 kg/m² (CI=5.7 (-0.3, 11.8); p -value = 0.05), is associated with the intensity of pain but not statistically significantly correlated.

The presence of pre-pregnancy LBP (CI=5.0 (0.5-9.5); p -value=0.03) and the sum of positive provocative tests on physical examination (CI=7.7 (1.1-14.3); p -value=0.03) correlate statistically

significantly with DRI at 12 weeks postpartum. Using "non-recovery at 12 weeks" as a dependent variable in a logistic regression model we see how the number of painful sites (CI=4.4(1.3-14.6); p-value=0.02) and the sum of positive provocative tests (CI=3.5(1.2-10.3); p-value=0.02) is statistically significantly associated with non-recovery at 12 weeks, while pre-pregnancy BMI is weakly associated (p-value=0.05). According to Gausel A. et al., 2015 the combined presence of three independent risk factors (age \geq 30 years (CI=2.9(1.3-6.8); p-value=0.012), moderate/high ODI in pregnancy (CI=5.1(1.7-15.0); p-value = 0.003) and PP with LBP in pregnancy (CI = 2.8 (1.2-6.4); p-value=0.017)) increases the risk of developing persistent PGP by 27 times compared to women with none of these factors, with an absolute risk of 35%. The investigated risk factors also correlate individually in a statistically significant manner with the investigated outcome.

Catastrophization (PCS), avoidance behaviors (FABQ), intensity of current pain and of worst perceived pain (VAS), disability (DRI), and quality of life (NHP) are six predictors of persistent LPP 6 months postpartum that Olsson C. et al. 2012, take into consideration. The presence of catastrophization (CI=1.06-3.98; p-value=0.034) and disability (CI=1.10-4.47; p-value=0.026) at 19-21 weeks of gestation statistically increases the risk of postpartum LPP for a significant value; they found no correlation for the other factors investigated.

Robinson H. et al., 2014, investigate pain and disability in women with PGP at the 30th week of gestation with one-year follow-up. Twelve weeks after delivery there are no significant differences in outcomes according to the considered variables (pain localization, ASLR, P4, PGP at the 30th week of gestation) except for the symphysis which correlates with higher levels of ache.

At one year after delivery, the average values of the disability variables are similar, while women who reported PGP at 30 weeks of pregnancy, and who had pelvic pain, or who had positive P4 and ASLR were found to have lower values for pain outcome. According to Xiangsheng et al. 2021 high levels of neurosis assessed with the Quick big five personality test (QBFPT) are associated with persistent PGP after pregnancy (OR = 2.12, P = 0.001), while extroverted and conscientious behaviors tend to have a protective action towards the disorder (OR = 0.65, P = 0.001; OR = 0.78, P = 0.010, respectively).

Finally Fernando et al. 2020 confirms that high levels of Fear Avoidance Belief (FABQ) at 34-37 weeks of pregnancy lead to a higher risk of having persistent low back pain at 6 months after delivery with an OR of 1.060 ($p \leq .05$).

Therefore, the risk factors that are most correlated with long-term PGP analyzed in the included studies are: high levels of pain in pregnancy (Albert, 2001-*moderate* ROB and Bergstrom, 2014-*low* ROB), high number of positive provocative tests (Albert, 2001-*moderate* ROB; Robinson, 2010(b)-*low* ROB), LBP/LPP history (Bergstrom, 2014-*moderate* ROB; Robinson, 2010(b)-*moderate* ROB; Gausel, 2015-*low* ROB), high levels disability in pregnancy (Gausel, 2015- *lw* ROB; Olsson, 2012(b)-*low* ROB), neurotic behavior (Xiangsheng, 2021-*low* ROB) and high levels of FABQ (Fernando 2020-*low* ROB) (***Figure 2.***).

DISCUSSION

In the analysis of 10 studies, we identified about 13 different risk factors and this important heterogeneity exposes to uncertainty in the conclusions.

The risk factors identified as the most predictive of persistent PGP 3-6 months after delivery seem quite in line with the scientific literature relating to the topic.

According to Van den Berg 2012^[23] and Eisenach 2008 ^[12], Albert 2001^[28], Bergstrom 2014^[29] and Gausel 2020^[44] eighlight how high levels of pain in pregnancy or in the immediate post-partum are related with disability at 6 weeks and with persistence of pain at 8/12 after delivery; this data is not confirmed by Olsson's study, 2012(b)^[26].

A large number of positive provocative tests in late pregnancy correlates with both pain and disability at 12 weeks postpartum^[30] and Elden,2016^[2] identifies that the most predictive tests of PGP up to 11

years are: the number of provocation tests positive (OR = 1.79), positive symphysis palpation test (OR = 2.01), positive Faber test (OR = 2.22) and positive modified Trendelenburg test (OR = 2.20). These data contrast instead with Robinson, 2014 who considers the influence of positive provocative tests in late-pregnancy to be minimal, in function and post-partum pain to be minimal^[27].

Comparing the literature with the data collected, the presence of LBP history appears to be associated both with the development of PGP in late pregnancy^{24]}, and with persistence of pain up to 6 months after delivery^{[18],[25]}. In our review, LBP as a predictor of persistent PGP is the risk factor that is correlated in the largest number of studies (Bergstrom, 2014^[29], Robinson, 2010(b)^[30], Gausel, 2015^[25]) although 2 of these were rated as *moderate ROB* (Bergstrom, 2014^[29], Robinson, 2010(b)^[30]); this data is also the only one in agreement with the review by Wuytack et al. 2018^[3].

Two studies evaluated as *low* ROB and therefore of good quality (Gausel, 2015^[25] e Olsson, 2012 (b)^[26]) bring to the conclusions that the presence of disability in pregnancy seems to predispose to persistence of PGP at 6 months after delivery in accordance with Sjudahl,2013 e Bjelland, 2012^{[25],[18]}. In our review only 5 studies rated as *low* ROB^{[25],[26],[23],[52],[54]} and 1 rated as *moderate* ROB^[30] consider psychosocial domains as risk factors for persistence and our analysis shows that the presence of emotional distress and depression in pregnancy are not associated with either pain or post-partum disability; although depressive symptoms are three times more frequent in women with LPP^[26], the study by Gausel, 2015^[25] cannot identify a cause-and-effect relationship. However, these results seem to contrast with the literature: Robinson, 2010 (a)^[27] highlights that emotional distress in early pregnancy correlates with PGP in late pregnancy and Bakker in 2013^[32] reports that Perceived Stress Scale (PSS) associated with Pregnancy Mobility Index (PMI) analyzed in early pregnancy are strong predictors of disability in late pregnancy; Wuytack,2018^[3] indicates a correlation between PGS and emotional distress, while Xiangsheng 2021 demonstrates an association between neurotic behaviors and persistence of PGP^[52].

Amongst the included studies only Olsson, 2012 (b)^[26] shows the tendency to catastrophize in pregnancy as a factor of persistence of PGP at 6 months after delivery , confirmed by a study by the

same author (Olsson, 2012 (a))^[16] in which evaluates the fluctuation in pregnancy and its correlation with postpartum pain and disability. In particular, according to Farzaneh et al. 2018 FABQ scores tend to vary with pain severity ^[45] and according to Fernando 2020, high FABQ scores at 34-37 weeks of gestation are predictive of PPGP^[54]. Despite the growing number of studies correlating central sensitization patterns with sleep disturbances ^{[28],[29]}, it is still a poorly investigated factor for PGP. Only one (Beales,2018)^[23] of the eight included studies investigates a possible association with PPGP without finding a correlation, in agreement with a previous study of the same author (Beales, 2016)^[20] where he finds an association between disorders of the sleep and PGP in pregnancy but not with persistent PGP. Other evidence instead reports that more than eight hours of sleep/rest the 30th week of pregnancy were associated with the persistence of pain at 12 weeks post-partum^[31].

As regards work, it seems that doing unskilled work, more than the workload itself, correlates with persistence of pain at 24 months after childbirth, while Van den Berg,2012^[31] highlights a correlation between maintaining uncomfortable postures at work, mainly intended as positions with repeated twists and bends, and PGP at the 30th week of pregnancy and 6 weeks after delivery.

The pre-pregnancy BMI shows conflicting evidence and appears to be associated, in a non-statistically significant way, with both disability and pain 3 months after delivery and Bjelland 2012^[18] confirms that a BMI>30 in pregnancy is associated with persistent PGP at 6 months after delivery. Also according to Matsuda ^[47] excessive weight gain in pregnancy is one of the risk factors for LBPP after delivery.

The characteristics of childbirth, the number of children, the number of pregnancies and the type of pregnancy, the weight and sex of the child, marital status, the use of contraceptives or other hormonal treatments, urinary infections in the years prior to pregnancy, smoking in pregnancy, are not correlated with persistence of pain in any of the studies and therefore does not contrast with the type of population

considered in the included studies. Diastasis of the rectus abdominis does not seem to affect PPGP either.^[57]

CONCLUSIONS

High levels of pain in pregnancy, a large number of positive provocative tests, having had a history of LBP and LPP, high levels of disability in pregnancy, neurotic behavior and high levels of Fear Avoidance Belief are the most predictors of PGP long-term 3-6 months after delivery^{[25],[26],[28],[29],[30],[52],[54]}, but given the extreme heterogeneity of the risk factors, and of the outcome assessment and follow-up times investigated in the included studies, it is not possible to define homogeneous subgroups by prognostic factor and therefore to draw strong conclusions on what are the risk factors of persistence of PPGP.

In the included articles, little attention was paid to the study of psychosocial domains or they were investigated together with numerous confounding factors, so that, despite the latest guidelines on the subject highlight how these factors are strongly correlated with the development of PGP in late-pregnancy and persistence postpartum (recommendation A)^[11], it was not possible to define their correlation with the persistence of pain.

Through neuroscience studies it has been seen that during pregnancy there is an increase in pain sensitivity not related to the pregnancy period ^[30] and therefore not entirely correlated to biomechanical or hormonal factors, even if they play an important role in the processes of pain modulation^[31].

The processes underlying the development of postpartum pain and disability are different from those identified for PGP in pregnancy^[35] and this complexity of the condition makes it necessary to frame the woman in early pregnancy both according to biomechanical and psychosocial factors. This type of management would allow for the implementation of chronicity prevention processes aimed at reducing the individual suffering of the woman, the costs for society but above all it would allow to reduce the risk of transition from acute pain to chronic pain^[32]. To avoid this transition, in fact, more and more

importance has been given, from the current state of the limb to chronic pain in the pelvis, to the factors that affect lifestyle, namely: low level of physical activity, poor sleep, periods of distress [56].

According to the "Fear-avoidance model" pain can be the result of catastrophising processes that lead to states of hypervigilance and avoidance behaviors, up to disability and chronic pain^[33] e and long-term low back pain^[54].

LIMITATIONS

The limitations of this study are represented by: a bibliographic search performed only in English, a limited number of included studies given by rather restrictive inclusion/exclusion criteria and studies of variable grade quality, strong heterogeneity of outcome measures^[48], resulting in the impossibility of meta-analysis.

Given the small number of studies dealing with the topic specifically and the poor methodological quality, it would be important to implement research through higher quality longitudinal studies based on existing standardized classification and evaluation systems ^{[34], [55]}. It would also be advisable for research in this area to be oriented towards the study of factors such as emotional distress, catastrophization, depression, sleep disturbances, job satisfaction and recovery expectations, factors for which we have identified few and low quality studies, as well as on standardized evaluation and self-screening tests as already proposed by Olsen in 2014 without however being able to guarantee high reproducibility and reliability of the same^[58], to lay the foundations for an early intervention in early pregnancy.

Aknoledgements

Thanks to everybody believes in this project and helps us to achieve it through suggestion or email support.

Disclosure of interests

The project was realized without financial, personal or political interests.

Contribution to Authorship

E.B. and G.G. start project during university career (Master of first level “Musculoskeletal and Rheumatological Physiotherapy”, Rome, Tor Vergata), S.M. continues this work under senior professor supervision of D.C. and F.B.

Statistic details was supervised by A.C., Associate Professor in Medical Statistics and Epidemiology.

Details of Ethics Approval

We don't ask ethics approval from the relevant regional or institutional ethics committee because of design of study, but we envelope the hole project involving ethical consideration, expecially about prevention of chronic pain and disability in a specific patient subgroup.

Funding

No one type of finding from commercial company or government department should started here.

Abbreviations

PGP: Pelvic Girdle Pain

PPGP: Pregnancy-Related Pelvic Girdle Pain

PLB: Pregnancy-Related Low Back Pain

LPP: Lumbopelvic Pain

PGS: Pelvic Girdle Syndrome

SIJ: Sacroiliac Joints

LDL: Long Dorsal Sacroiliac Ligament

P4: Posterior Pelvic Pain Provocation Test

ASLR: Active Straight leg raise

BMI: Body Mass Index

VAS: Visual Analogic Scale

PPT: Pain Pressure Threshold

PMI: Pregnancy Mobility Index

PSS: Perceived Stress Scale

DRI: Disability Rating Index

ODI: Oswestry Disability Index

PGQ: Pelvic Girdle Questionnaire

HR- Qol: Health Related Quality of Life

EQ-5D: EuroQol

SF-36: Short Form-36 Health Survey

SRH: Self-Rated Health

NHP: Nottingham Health Profile

PCS: Pain Catastrophizing Scale

PSQI: Pittsburgh Sleep Quality Index

FABQ: Fear-Avoidance Beliefs Questionnaire

NHP: Nottingham Health Profile

McGill: McGill Pain Questionnaire

NP= Neck Pain/ TC= Thoracic Pain

ROB: Risk of Bias

QUIPS: Quality In Prognosis Studies tool

OR: Odd Ratio

CI: Confidence Interval

Bibliography

- 1[]] Vleeming A., Hanne B. Albert, Hans Christian Ostgaard, Sturesson B. , Britt Stuge. European guidelines for the diagnosis and treatment of pelvic girdle pain. *Eur Spine J.* 2008; 17:794–819.
- 2[]] Elden H., Gutke A. et al. Predictors and consequences of long-term pregnancy-related pelvic girdle pain: a longitudinal follow-up study. *BMC Musculoskeletal Disorders* (2016) 17:276.
- 3[]] Wuytack F. PhD, Daly D. PhD, Curtis E. PhD, Begley C. PhD. Prognostic factors for Pregnancy-related Pelvic Girdle Pain, a Systematic Review. *Midwifery*, 2018; 66:70-78.
- 4[]] Kanakaris N., Roberts C., Giannoudis P. Pregnancy-related pelvic girdle pain: an update. *BMC Medicine* 2011; 9:15.
- 5[]] Verstraete E.H., Vanderstraeten G., Parewijck W. Pelvic Girdle Pain during or after Pregnancy: a review of recent evidence and a clinical care path proposal. *FVV in ObGyn.* 2013; 5 (1): 33-43.
- 6[]] Aslan E., Fynes M. Symphyseal pelvic dysfunction. *Curr. Opin Obstet Gynecol.* 2007; 19:133-139.
- 7[]] Depledge J., McNair PJ, Keal-Smith C., Williams M. Management of symphysis pubis dysfunction during pregnancy using exercise and pelvic support belts. *Phys Ther.* 2005; 85:1290-1300.
- 8[]] Vermani E., FRCA; Mittal R., FRCS; Weeks A., MRCOG. Pelvic Girdle Pain and Low Back Pain in Pregnancy: A Review. *World Institute of Pain* 2009; *Pain Practice*, Volume 10, Issue 1, 2010 60–71.
- 9[]] Mogren et al. BMI, pain and hyper-mobility are determinants of long-term outcome for women with low back pain and pelvic pain during pregnancy. *Eur Spine J.* 2006; 15: 1093–1102.
- 10[]] Leadbetter RE., Mawer D., Lindow SW. The development of a scoring system for symphysis pubis dysfunction. *J Obstet Gynaecol.* 2006; 26:20-23.

-
- ¹¹ Clinton S., Newell A., Downey P., Coleman-Ferreira K. Pelvic Girdle Pain in the Antepartum Population: Physical Therapy Clinical Practice Guidelines Linked to the International Classification of Functioning, Disability, and Health From the Section on Women's Health and the Orthopaedic Section of the American Physical Therapy Association, in *Journal of women's health physical therapy* 41(2):102-125 · May 2017.
- ¹² Eisenach JC., Pan PH., Smiley R., Lavand'homme P., Landau R., Houle TT. Severity of acute pain after childbirth, but not type of delivery, predicts persistent pain and postpartum depression. *Pain* 2008; 140:87–94.
- ¹³ Kamaleri Y., Natvig B., Ihlebaek CM., Benth JS., Bruusgaard D. Change in the number of musculoskeletal pain sites: a 14-year prospective study. *Pain* 2009;141:25–30.
- ¹⁴ Beales DJ., O'Sullivan PB., Briffa NK. Motor control patterns during an active straight leg raise in chronic pelvic girdle pain subjects. *Spine* 2009;34:861e70.
- ¹⁵ Bjelland EK., Stuge B., Engdahl B., Eberhard-Gran M. The effect of emotional distress on persistent pelvic girdle pain after delivery: a longitudinal population study. *BJOG* 2013;120:32e40.
- ¹⁶ Olsson CB., Grooten WJ., Nilsson-Wikmar L., Harms-Ringdahl K., Lundberg M. Catastrophizing during and after pregnancy: associations with lumbopelvic pain and postpartum physical ability. *Phys Ther* 2012;92:49e57.
- ¹⁷ Vollestad NK., Stuge B. Prognostic factors for recovery from postpartum pelvic girdle pain. *Eur Spine J* 2009;18:718e26.
- ¹⁸ Bjelland EK., Stuge B., Engdahl B., Eberhard-Gran M. The effect of emotional distress on persistent pelvic girdle pain after delivery: a longitudinal population study. *BJOG*. 2012;120:32–40.
- ¹⁹ Finan PH., Goodin BR., Smith MT. The association of sleep and pain: an update and a path forward. *J Pain* 2013;14:1539e52.

-
- ²⁰□ Beales D., Lutz A., Thompson J., Wand B.M., O'Sullivan P. Disturbed body perception, reduced sleep, and kinesiophobia in subjects with pregnancy-related persistent lumbopelvic pain and moderate levels of disability: An exploratory study. *Manual Therapy* 21 (2016) 69e75.
- ²¹□ Albert H. B., Godskesen M., Korsholm L., Westergaard J. G. Risk factors in developing pregnancy-related pelvic girdle pain. *Acta Obstetrica et Gynecologica* 2006; 85: 539/544.
- ²²□ Hayden J.A., Van Der Windt D. A., Cartwright, J. L., Cote P. & Bombardier, C. Assessing bias in studies of prognostic factors. *Annals of Internal Medicine*, 2013. 158, 280-6.
- ²³□ Stomp-van den Berg SG., Hendriksen IJ., Bruinvels DJ., Twisk JW., Van Mechelen W., Van Poppel MN. Predictors for postpartum pelvic girdle pain in working women: the Mom@Work cohort study. *Pain* 2012;153:2370e9.
- ²⁴□ Bakker E., van Nimwegen-Matzinger CW., Ekkel-van der Voorden W., Nijkamp MD, Völlink T. Psychological determinants of pregnancy-related lumbopelvic pain: a prospective cohort study. *Acta Obstet Gynecol Scand*. 2013 Jul;92(7):797-803.
- ²⁵□ Sjö Dahl J., Gutke A., Öberg B. Predictors for long-term disability in women with persistent postpartum pelvic girdle pain. *Eur Spine J*. 2013 Jul;22(7):1665-73.
- ²⁶□ Gutke A., Josefsson A., Öberg B. Pelvic girdle pain and lumbar pain in relation to postpartum depressive symptoms. *Spine (Phila Pa 1976)*. 2007 Jun 1;32(13):1430-6.
- ²⁷□ Robinson et al. Pelvic girdle pain - associations between risk factors in early pregnancy and disability or pain intensity in late pregnancy: a prospective cohort study. *BMC Musculoskeletal Disorders* 2010, 11:91
- ²⁸□ Dorheim SK., Bjorvatn B., Eberhard-Gran M. Insomnia and depressive symptoms in late pregnancy: a population-based study. *Behav Sleep Med* 2012;10:152e66
- ²⁹□ Sivertsen B., Lallukka T., Petrie KJ., Steingrimsdóttir ÓA., Stubhaug A., Nielsen CS. Sleep and pain sensitivity in adults. *Pain*. 2015 Aug;156(8):1433-9.

³⁰ Palsson T., Beales D., Slater H., O’Sullivan P., and Graven-Nielsen T. Pregnancy Is Characterized by Widespread Deep-Tissue Hypersensitivity Independent of Lumbopelvic Pain Intensity, a Facilitated Response to Manual Orthopedic Tests, and Poorer Self-Reported Health. *The Journal of Pain* 2015; Vol 16, No 3 .

³¹ Aloisi A., Marco Bonifazi. Sex hormones, central nervous system and pain. *Hormones and Behavior* 2006; Volume 50, Issue 1, Pages 1–7.

³² Bergström C., Persson M., Mogren I. Sick leave and healthcare utilisation in women reporting pregnancy related low back pain and/or pelvic girdle pain at 14 months postpartum *Chiropr Man Therap.* 2016 Feb 15; 24:7. doi: 10.1186/s12998-016-0088-9. eCollection 2016.

³³ Vlaeyen JW., Linton SJ. Fear-avoidance and its consequences in chronic musculoskeletal pain: a state of the art. *Pain.* 2000 Apr;85(3):317-32.

³⁴ Pennick, Liddle SD. Interventions for preventing and treating pelvic and back pain in pregnancy (Review). *The Cochrane Library* 2013; Issue 8.

^[43] Tavares P, Barrett J., Hogg-Johnson S. Prevalence of Low Back Pain, Pelvic Girdle Pain, and Combination Pain in a Postpartum Ontario Population *Women Health.* 2019

^[44] Gausel A., Malmqvist S., Andersen K. Subjective recovery from pregnancy-related pelvic girdle pain the first 6 weeks after delivery: a prospective longitudinal cohort study *European Spine Journal* 2020

^[45] Fakari F.R., Simbar M. The Relationship between Fear-Avoidance Beliefs and Pain in Pregnant Women with Pelvic Girdle Pain: A Cross-Sectional Study. *International Journal of Community Based Nursing and Midwifery* 2018

^[46] Davenport M., Marchand A.A., Mottola M. Exercise for the prevention and treatment of low back, pelvic girdle and lumbopelvic pain during pregnancy: a systematic review and meta-analysis. *British Journal of Sports Medicine* 2018; 23

-
- [47] Matsuda N., Kitagaki K., Perrein E., Tsuboi Y., Ebina A., Kondo Y., Murata S., Isa T., Okumura M., Kawaharada R., Horibe K., Ono R. Association Between Excessive Weight Gain During Pregnancy and Persistent Low Back and Pelvic Pain After Delivery. *Spine* 2020
- [48] Wuytack F., O'Donovan M. Outcomes and outcomes measurements used in intervention studies of pelvic girdle pain and lumbopelvic pain: a systematic review. *Chiropractic and Manual Therapies* 2019
- [49] Stafne S., Vøllestad N., Mørkveda, Salvesen K.A., Robinson H.S. Impact of job adjustment, pain location and exercise on sick leave due to lumbopelvic pain in pregnancy: a longitudinal study *Scandinavian Journal of Primary Health Care* 2019
- [50] Wuytack F., Begley C., Daly D. Risk factors for pregnancy-related pelvic girdle pain: a scoping review. *BMC Pregnancy and Childbirth* 2020
- [51] Ceprnja D., Chipchase L., Fahey P., Liamputtong P., Gupta A. Prevalence and Factors Associated with Pelvic Girdle Pain During Pregnancy in Australian Women. *Spine* 2021
- [52] Xiangsheng T., Long G., Yingying, Xiao A., Ping Y., Mingsheng T.. Personality traits predict regression of pelvic girdle pain after pregnancy: a longitudinal follow-up study. *BMC Pregnancy and Childbirth*. 2021
- [53] Asuka S., Gamada K. Altered musculoskeletal mechanics as risk factors for postpartum pelvic girdle pain: a literature review. *The Journal of Physical Therapy Science* 2019
- [54] Fernando M., Nilsson-Wikmar L., Olsson C.B. Fear-avoidance beliefs: A predictor for postpartum lumbopelvic pain. *Physiotherapy Research International* 2020
- [55] Remus A., Smith V., Gutke A., Saldaña Mena J.J., Mørkved S., Wikmar L.N., Olsson B.O.C., Robinson H.S. , Stuge B., Wuytack F. A core outcome set for research and clinical practice in women with pelvic girdle pain: PGP-COS. *PLOS ONE* 2021
- [56] Gutke A., Sundfeldt K., De Baets L. Lifestyle and Chronic Pain in the Pelvis: State of the Art and Future Directions. *Journal of Clinical Medicine* 2021

-
- [57] Gluppe S., Ellström Engh M., Kari Bøa. Women with diastasis recti abdominis might have weaker abdominal muscles and more abdominal pain, but no higher prevalence of pelvic floor disorders, low back and pelvic girdle pain than women without diastasis recti abdominis. *Physiotherapy Elsevier* 202.
- [58] Olsén M.F., Elden H., Gutke A. Evaluation of self-administered tests for pelvic girdle pain in pregnancy. *BMC Musculoskeletal Disorders* 2014.
- [59] Lindgren A., Kristiansson P. Finger joint laxity, number of previous pregnancies and pregnancy induced back pain in a cohort study. *BMC Pregnancy and Childbirth* 2014
- [60] Bergstrom C., Persson M., Mogren I. Psychosocial and behavioural characteristics in women with pregnancyrelated lumbopelvic pain 12 years postpartum. *Chiropractic & Manual Therapies* 2019
- [61] Kovacs F., Emma Garcia, Ana Royuela, Lourdes González, Víctor Abaira and the Spanish Back Pain Research Network. Prevalence and Factors Associated With Low Back Pain and Pelvic Girdle Pain During Pregnancy. *Spine* 2012
- [62] Lardon E., Audrey St-Laurent, Véronique Babineau, Martin Descarreaux, Stephanie-May Ruchat Lumbopelvic pain, anxiety, physical activity and mode of conception: a prospective cohort study of pregnant women. *BMJ Open* 2018
- [63] Rost C., Jaqueline J., Kaiser A., Arianne P. Verhagen e Bart W. K. Prognosis of women with pelvic pain during pregnancy: a long-term follow-up study. *Acta Obstetricia et Gynecologica*. 2006
- [64] Munro A., Pharm, Ronald B. George, Jill Chorney, Erna Snelgrove-Clarke; Natalie O. Rosen. Prevalence and Predictors of Chronic Pain in Pregnancy and Postpartum. *J Obstet Gynaecol Can* 2017

Included or excluded articles

<i>Article</i>	<i>Inclusion</i>	<i>Exclusion motivation</i>
Sjodahl, 2013 [33]	NO	- Lack of primary outcome - Baseline Time to follow-up not satisfy our criteria
Van den Berg, 2012 [31]	NO	- Population from clinical trial
Robinson, 2010 A [35]	NO	- Lack of follow-up post-partum
Bakker, 2013 [32]	NO	- Lack of follow-up post-partum
Albert, 2001 [28]	YES	
Olsson, 2012 A [16]	NO	- Primary Outcome: catastrophization
Eisenach, 2008 [12]	NO	- First follow-up at 8 week post-partum
Vollestad, 2009 [17]	NO	- Population from clinical trial
Elden, 2016 [2]	NO	- Population from clinical trial, Baseline Time to follow-up not clear
Beales, 2018 [23]	YES	
Bergstrom, 2014 [29]	YES	
Bergstrom, 2016 [40]	NO	- Primary Outcome not satisfy our criteria
Bergstrom, 2017 [24]	YES	
Bjelland, 2012 [18]	NO	- Primary Outcome evaluated like numbers of painful points
Robinson, 2010 B [30]	YES	
Gausel, 2015 [25]	YES	
Olsson, 2012 B [26]	YES	
Robinson, 2014 C [27]	YES	
Gausel, 2020	NO	- Follow-up not satisfy our criteria
Cepria, 2021 [51]	NO	- Cross-sectional design
Lindgren, 2014 [59]	NO	- Back pain (unclear terminology)
Bergstrom, 2019 [60]	NO	- Cross-sectional design
Kovacs, 2012 [61]	NO	- Cross-sectional design
Lardon, 2018 [62]	NO	- Main and secondary outcome not satisfy the requirements

Rost, 2006 [63]	NO	-	Therapeutical intervention
Fernando, 2020 [54]	YES	-	
Xiangsheng, 2021 [52]	YES	-	
Munro, 2017 [64]	NO	-	Outcome body pain ingeneral, not specific for PGP. Full text in French.
Notes:	"yes"= article included	"NO"= article excluded	-

Table 1

Assessment <i>Risk of bias</i> included articles through QUIPS tool							
Articles	Study participation	Study attrition	Prognostic factor measurement	Outcome measurement	Study confounding	Statistical Analysis and Reporting	Overall
<i>Albert, 2001</i>	-	-	+/-	-	+	+/-	+/- Moderate risk of bias
<i>Beales, 2018</i>	-	-	-	-	+	-	- Low risk of bias
<i>Bergstrom, 2014</i>	-	+	+/-	-	+/-	-	+/- Moderate risk of bias
<i>Bergstrom, 2017</i>	-	-	-	-	+	-	- Low risk of bias
<i>Robinson, 2010 (B)</i>	+/-	+	-	-	+	-	+/- Moderate risk of bias
<i>Gausel, 2015</i>	-	+	-	-	+	-	- Low risk of bias
<i>Olsson, 2012 (B)</i>	-	+/-	-	-	-	-	- Low risk of bias
<i>Robinson, 2014 (C)</i>	-	-	+/-	-	+/-	-	- Low risk of bias

<i>Fernando, 2020</i>	+/-	+	-	+/-	-	-	- Low risk of bias
<i>Xiangsheng, 2021</i>	+/-	+	-	-	+/-	-	- Low risk of bias
+ High risk of bias, +/- moderate risk of bias, - low risk of bias							

Table 2