



Incidence of perineal pain and dyspareunia following spontaneous vaginal birth: a systematic review and meta-analysis

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Received: 19 November 2018 / Accepted: 28 January 2019 / Published online: 15 February 2019
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Abstract

Introduction and hypothesis Perineal pain and dyspareunia are experienced by women undergoing a vaginal birth that can have short and longer term physical and psychological morbidities. This review aimed to determine the incidence of perineal pain and dyspareunia following spontaneous vaginal birth (SVB) with intact perineum, first and second-degree perineal trauma or episiotomy.

Methods Searches of MEDLINE, EMBASE, CINAHL, AMED and MIDIRS (inception – December 2017) were undertaken with selection criteria of any study evaluating the effect of intact perineum, first- or second-degree perineal trauma on perineal pain or dyspareunia in women with SVB.

Results Eighteen studies (8 RCTs and 10 NRSs) were included. Fourteen and 12 studies were undertaken to assess perineal pain and dyspareunia after SVB, respectively. Meta-analysis of 16 studies (3133 women) demonstrated that women at 2 days postpartum experienced nearly the same incidence of perineal pain whether perineal trauma existed or not. At 4–10 days postpartum there was a significant reduction in the incidence of perineal pain for both presence and absence of any perineal trauma. Episiotomy was associated with the highest rate of perineal pain. The incidence of dyspareunia was high at resumption of sexual intercourse following SVB with an intact perineum. At 12 months, women still experienced dyspareunia whether perineal trauma existed or not.

Conclusions Women experience perineal pain and dyspareunia regardless of the presence or absence of perineal trauma after SVB; nonetheless, the reported incidence is higher if perineal trauma occurred.

Keywords Perineal trauma · Perineal pain · Dyspareunia · Spontaneous vaginal birth · Systematic review

Introduction

Worldwide just over 60% of women have a spontaneous vaginal birth (SVB) equating to an estimate of 78.84 million

births per annum [1–5]. Childbirth is always an expected event full of emotions although not free of fears. Psychological concerns such as changing lifestyles, body image issues and psychosocial problems exist alongside physical

Conference Presentations IUGA 43rd Annual Meeting, Vienna, Austria. 27–30 June 2018.

Electronic supplementary material The online version of this article (<https://doi.org/10.1007/s00192-019-03894-0>) contains supplementary material, which is available to authorized users.

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concerns and morbidities of extreme tiredness, urinary and anal incontinence, wound infection or dehiscence, perineal pain or dyspareunia [6, 7].

Perineal pain and dyspareunia are commonly experienced in both the short and longer term by women undergoing a vaginal birth. Perineal pain can limit the woman's mobility, quality of life and ability to comfortably care for her baby, which can have a negative impact on a mother's adaptation to her new role [8]. Likewise, dyspareunia is usually reported in the long-term postnatal period, which can affect sexual health and relationships [9, 10].

Evidence shows that there is a significant association between perineal pain and dyspareunia and operative vaginal birth [11, 12]. Moreover, the best material and method to repair perineal trauma to reduce the incidence of perineal pain and dyspareunia following childbirth have been heavily researched [13, 14]. However, the incidence and ongoing rates of perineal pain and dyspareunia after SVB particularly when the perineum is intact are less clearly known.

This review aimed to systematically assess the current available evidence to determine the incidence of perineal pain and dyspareunia, at both the short and long term, following SVB with an intact perineum, first- and second-degree perineal trauma or episiotomy.

Materials and methods

Search strategy

A protocol using widely recommended methods for systematic reviews of observational studies was developed and registered with PROSPERO International prospective register of systematic reviews (CRD42017054281). The PRISMA statement and checklist were followed throughout review preparation (Appendix 1).

MEDLINE, EMBASE, CINAHL, AMED and Maternity and Infant Care (MIDIRS) databases were searched electronically from inception to December 2017. A combination of medical subject headings (MeSHs), to encompass both perineal pain and dyspareunia, keywords and word variants using Boolean operators 'OR' and 'AND' to capture relevant text citations were used. Search strategies were adapted for each database (Appendix 2). Terms of 'sphincter anal' or 'Obstetric Anal Sphincter Injuries (OASIS)' or 'operative vaginal birth' were not included in the search to reduce the risk of limiting access to all possible relevant articles. In addition, reference lists of relevant articles were manually searched to identify papers not captured by electronic searches. The aim of our systematic review was to assess the current evidence to determine the incidence of perineal pain and dyspareunia at both the short and long term, following spontaneous vaginal birth with either an intact perineum, first- and second-degree

perineal trauma or episiotomy. However, limiting our search to RCTs would have only enabled data capture for women who sustained second-degree trauma or an episiotomy. Therefore, a decision was made to include both randomised controlled trials (RCTs) and non-randomised studies (NRSS) reporting incidence of perineal pain or dyspareunia after SVB. Case series and case reports were excluded. Conference papers and abstracts were included if they reported enough information regarding study design and outcome data. No language restrictions were applied, but the search was limited to human studies. The search strategies were developed by MM. A database of all abstracts of citations was compiled.

Study selection

Studies were selected in a three-stage process. First, each title and abstract were assessed by two reviewers (MM and AP) and full articles of all references that were likely to fulfil predefined criteria were obtained. Second, selected articles were assessed by two independent reviewers (MM and AP), against pre-designed inclusion/exclusion criteria, with any discrepancies referred to a third party (SSW, EB or KMI) for the final decision. Third, studies were included if they gave information with supporting statistical evidence on perineal pain or dyspareunia for women after SVB. When necessary, the study's author was contacted to clarify or obtain additional data (Fig. 1).

Data abstraction and synthesis

Data were extracted on study quality, participants' characteristics, perineal trauma repair techniques, perineal pain including pain at different time periods and settings, use of analgesia, breastfeeding and dyspareunia using a pre-designed data capture form. Data extraction was performed by two reviewers (MM and AP), with assistance from a third reviewer in case of discrepancy (SSW). Primary outcomes were perineal pain and dyspareunia after SVB. Perineal pain was defined as pain in the area reaching from the symphysis pubis to the coccyx and across both ischial tuberosities [7, 15]. We chose this definition, which is different from that suggested by the International Urogynecology Association (IUGA) and the International Continence Society (ICS) [16] who both define it as the area between the posterior fourchette (posterior lip of the introitus) and the anus, in order to encompass pain related to perineal trauma when it occurs in the anterior perineum and to reflect the correct anatomical definition of the perineum. We used the dyspareunia definition of persistent or recurrent pain or discomfort associated with attempted or complete vaginal penetration [16]. When extracting data it was noted how the studies defined perineal pain and dyspareunia. Whenever possible, data were extracted to compute 2×2 tables.

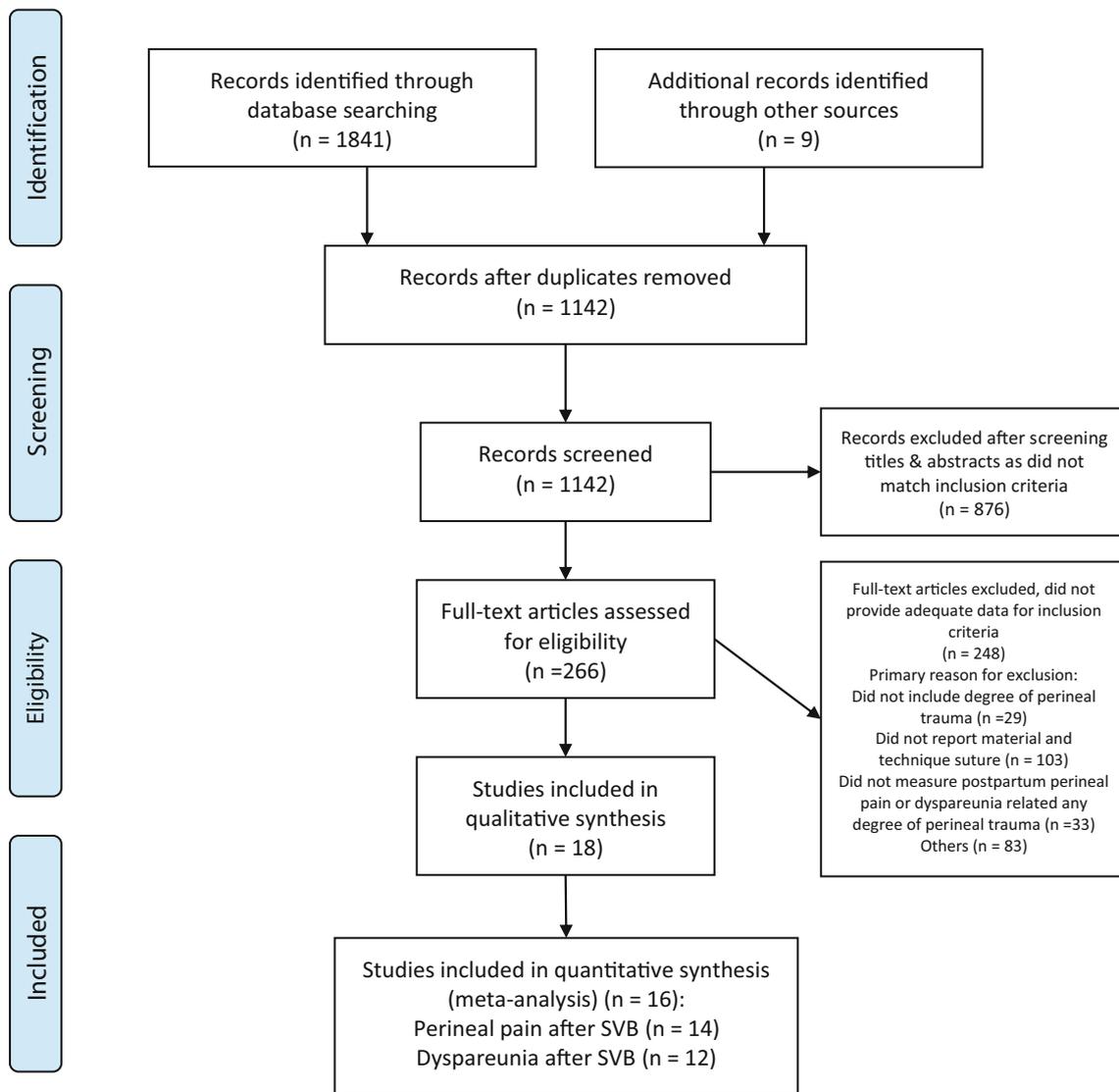


Fig. 1 PRISMA 2009 flow diagram

Although all studies used the Royal College Obstetricians and Gynaecologists (RCOG) classification for categorising perineal trauma, some studies undertook analysis by using alternative terminologies to describe the trauma or grouping the trauma for analysis. In five studies (27.7%) data were presented as spontaneous tear or perineal laceration [17–19] or as sutured/unsutured tears [11, 20], whilst in six studies [11, 21–25] data on second-degree perineal trauma and episiotomy were grouped. We contacted authors requesting the information be stratified by degree of trauma. However, in the majority of cases (72.7%), authors either could not provide this data or did not respond to our request.

Regarding second-degree perineal trauma and episiotomy, initially, our aim was to determine the incidence of the perineal pain and dyspareunia in relation to whether the trauma was repaired in line with Spanish National Healthcare System and RCOG evidence-based guidelines [26, 27]. Although all

the repairs were performed using a recommended absorbable suture material, the different interpretations of ‘continuous repair technique’ necessitated analysing incidence of perineal pain and dyspareunia in the following three repair technique categories: (1) continuous technique for all layers (vaginal mucosa, perineal muscles and skin), (2) continuous technique for vaginal mucosa only and (3) episiotomy repaired, at least, with the continuous technique for subcutaneous tissue. No other different repair techniques were found within studies that met inclusion criteria for this SR.

Validity assessment

The Cochrane Collaboration’s tool for assessing risk of bias in randomised trials [28] and the Joanna Briggs Institute Prevalence Critical Appraisal Tool (Table S1) [29] were used to assess risk bias and quality of included RCTs and NRSs

respectively. Quality assessment was then used to assess the methodological adequacies of the included studies and assist with interpretation of meta-analysis findings and possible bias resultant from study heterogeneity.

Statistical methods

R version 3.4.3 [30] with packages meta [31] and metaphor [32] were used for statistical analysis. Meta-analysis was performed if data from two or more eligible studies were available; all other eligible studies were analysed descriptively. The number of positive events and the total number of potential events (namely, women with the condition and total women examined) were analysed and summarised with the resulting incidence rate and its 95% confidence interval (CI). Meta-analytical estimates of the overall incidence rate (point estimate and 95% CI) were obtained fitting random-effects models because of the high likelihood of clinical and statistical heterogeneity; the inverse variance (IV) method with log transformation of the incidence rate was used. The preferred measure of statistical heterogeneity in the meta-analysis was the I^2 statistic (and, if obtainable, its 95% CI) [33]. Values below 25% were considered low, about 50% moderate and above 75% high [34]. Cochrane's Q statistic was reported as a χ^2 with its corresponding degrees of freedom and resulting P value; the between-study variance (τ^2) estimated using restricted maximum likelihood was also reported.

Results

Eight RCTs and ten NRSs (nine cohort studies and one cross-sectional survey study) from 11 countries were included (Table 1) in the review, of which eight RCTs and eight NRSs were included for quantitative synthesis by meta-analysis. No relevant systematic reviews were identified.

In relation to the primary objective, 14 of the total of 18 studies (7 RCTs and 7 NRSs) which satisfied the inclusion criteria were undertaken to assess perineal pain after SVB and 12 studies (6 RCTs and 6 NRSs) to assess dyspareunia after SVB. From all 18 of the included studies, a total of 3133 women were followed up after SVB to assess perineal pain and 1567 women to assess dyspareunia. Data on perineal pain were only available for meta-analysis on 2113 (67.4%) of the 3133 women from 12 studies. Data on dyspareunia were available for meta-analysis on 1468 (97.7%) of the eligible 1567 women from 9 studies.

Study quality and publication bias

Quality assessment of the included studies revealed deficiencies in many methodological areas (Fig. S1). For the RCTs, two studies (25%) met all six quality criteria. One study

(12.5%) fulfilled five criteria. The remainder (62.5%) met four or less of the quality criteria. One study failed to fulfil any of the quality criteria. For the NRSs, no studies met all ten quality criteria. Two studies (20%) met seven or more criteria; the remainder fulfilled six or less, where three studies (30%) met < 50% of quality criteria. No studies were excluded from the systematic review for failure to fulfil the quality criteria.

Validated measurement tools for perineal pain and dyspareunia were only used in nine studies (64.3%) and four studies (33.3%), respectively.

Regarding the definition of the perineal area or perineal pain used in the questionnaires, only three studies [18, 37, 38] detailed the specific area related to the perineum, albeit not clearly defined. Regarding dyspareunia, the majority of the studies reported on pain during sexual intercourse, similar to the ICS definition.

Only four studies provided information on sample size calculations.

Principal results

Perineal pain after SVB

Perineal pain after intact perineum

Meta-analysis of two cohort NRSs [38, 39] demonstrated that, at day 2 (Fig. 2), the incidence of perineal pain in women with an intact perineum following SVB was 42% (798 women; 95% CI 0.56 to 0.75; $I^2 = 0\%$), whereas at 4–10 days post birth (Fig. 3), the incidence of perineal pain with an intact perineum was 11% (789 women; 95% CI 0.09 to 0.13; $I^2 = 0\%$).

The intensity of pain experienced by women with an intact perineum following an SVB has only been reported by Harrison et al. (1984) [39]. In a small study of 19 primiparous women with an intact perineum following their first SVB, 12 (63%) women reported mild or moderate pain on the 1st day postpartum. However, this decreased to 32% at day 2 and by the 4th day the intensity of pain was reported by only 5% of these women, as mild only.

Meta-analysis of two studies [17, 38] (758 women; 95% CI 0.01 to 100; $I^2 = 98\%$) demonstrated an incidence of perineal pain of 11% for women with an intact perineum following SVB at 3 months postpartum. However, the very high heterogeneity of this meta-analysis ($I^2 = 98\%$) and the very wide CI must be acknowledged.

Perineal pain after first-degree perineal trauma

Albers et al. (1999) [38] reported the presence of perineal pain for women with perineal trauma in the outer vagina and labia as 64.3%, 25.5% and 5.5% at day 2, day 10 and 3 months,

Table 1 Study characteristics of all RCTs and NRSSs included in the systematic review

Study characteristics of all RCTs included in the Systematic Review of Perineal pain and Dyspareunia (N = 8) Author, country, language, year, reference	Study design	Population	Intervention/study intention regarding perineal pain (PP) or dyspareunia (dysp)	Number of women included at follow-up survey data with intact perineum and each degree of perineal trauma	Outcomes
Almeida, Brazil, Spanish, 2008 [21]	RCT Random sequence, allocation concealment, blinding for professionals-patients, blinding outcomes assessment, enrolment Cohort study: data collection, enrolment RCT Random sequence: computer controlled; allocation concealment: table given by the computer Blinding for professionals-patients: did not report Blinding outcomes assessment: the same midwives do the repair and postnatal assessment Enrolment: consecutive	Primiparous and multiparous with 2nd-degree perineal trauma or episiotomy after SVB	Compared two different suture techniques: all layers with continuous technique vs only vagina with continuous technique and interrupted stitches in muscles and skin	PP: 61 women Dysp: 23 women	Perineal pain and use of analgesia, wound healing and dyspareunia
Aslam, Pakistan, English, 2015 [22]	RCT Random sequence: computer controlled; allocation concealment: numbered, opaque and closed envelopes Blinding for professionals-patients: no comments were made regarding technique during labour or postnatal assessment Blinding outcomes assessment: third person blinded to treatment allocation Enrolment: consecutive	Primiparous with 2nd-degree perineal trauma or episiotomy after SVB	Compared two different suture techniques: continuous technique vs interrupted stitches (do not mention if all layers. This group: not included in the SR)	69 women in continuous technique group	Perineal pain
Beard, UK, English, 1974 [35]	RCT Random sequence: not reported Allocation concealment: not reported Blinding for professionals-patients: not reported Blinding outcomes assessment: not reported Enrolment: unclear	Nulliparous and multiparous with episiotomy after SVB	Compared two different suture materials: Vicryl st vs catgut (catgut group not included)	100 women (Vicryl group)	Perineal pain and use of analgesia
Graczyk, Poland, Polish, 1998 [36]	RCT Random sequence: even and odd; allocation concealment: even and odd Blinding for professionals-patients: no; blinding outcomes assessment: No Enrolment: unreported RCT	Primiparous with episiotomy after SVB	Compared two different suture techniques: subcuticular continuous repair vs unspecified technique repair	PP: 65 women (subcuticular group) Dysp: 24 women (subcuticular group)	Perineal pain and dyspareunia

Table 1 (continued)

McElhinney, UK, English, 2000 [23]	Random sequence: not reported; allocation concealment: two sets of sealed envelopes Blinding for professionals-patients: not reported; blinding outcomes assessment: not reported Enrollment: not reported RCT	Primiparous and multiparous with second-degree perineal trauma or episiotomy after SVB	Compared two different suture materials: Vicryl st vs Vicryl Rapid	Dyspareunia and perineal pain (PP: data unable to be included in MA)
Morano, Italy, English, 2006 [24]	Random sequence: computer controlled; allocation concealment: even and odd in cards placed in an opaque envelope and sealed Blinding for professionals-patients: blinded; blinding outcomes assessment: blinded Enrollment: unreported RCT	Primiparous with second-degree perineal trauma or episiotomy after SVB	Compared two different suture techniques: all layers with continuous technique vs vagina with continuous technique and interrupted stitches in muscles and skin	Perineal pain, use of analgesia, wound healing and dyspareunia
Perveen, Pakistan, English, 2009 [37]	Random sequence: alternating sequence in one of 4 groups; allocation concealment: no Blinding for professionals-patients: no; blinding outcomes assessment: unclear Enrollment: unreported RCT	Primiparous and multiparous with second-degree perineal trauma or episiotomy after SVB	Compared two different suture materials and two different suture techniques: Vicryl st vs catgut and all layers with continuous technique vs only vagina with continuous technique, muscles and skin interrupted stitches Catgut: not included	Perineal pain, wound healing and dyspareunia
Valenzuela, Spain, Spanish, 2008 [25]	Random sequence: computer controlled; allocation numbered opaque and closed envelopes Blinding for professionals-patients: blinded; blinding outcomes assessment: blinded Enrollment: unreported RCT	Primiparous and multiparous with second-degree perineal trauma or episiotomy after SVB	Compared two different suture techniques: all layers with continuous technique vs only vagina with continuous technique and interrupted stitches in muscles and skin	Perineal pain and dyspareunia
Study characteristics of all NRSs included in the Systematic Review of Perineal Pain (PP) and Dyspareunia (Dysp) (N = 10) Authors, country, language, year, reference	Enrollment: unreported Study design RCT: Random sequence, allocation concealment, blinding for professionals-patients, blinding outcomes assessment Cohort: data collection, enrolment Cohort study, prospective, consecutive	Population	Intervention/study intention regarding perineal pain (PP)	Outcomes
Albers, UK, English, 1999 [38]		Primiparous and multiparous women, included in an RCT previously, having SVB in two different hospitals	Incidence of PP-related intact perineum and degree of perineal trauma or episiotomy	Perineal pain

Table 1 (continued)

Harrison, Ireland, English, 1984 [39]	Cohort study, prospective, consecutive	Primiparous women having SVB	Incidence of PP-related intact perineum and degree of perineal trauma or episiotomy	Second degree and episiotomy not included; suture material and method of repair not reported 19 women with intact perineum Second degree and episiotomy not included; catgut material was used PP: 51 women Dysp: 51 women	Perineal pain
Kalis, Czech Republic, English, 2011 [40]	Cohort study; prospective, non-consecutive	Primiparous and multiparous women having SVB with an episiotomy at 60°	Incidence of PP and dysp-related episiotomy	PP: 110 women with intact perineum Dysp: 110 women with intact perineum Second degree and episiotomy not included; suture material and method not reported 3rd/4th-degree perineal injury excluded from this SR	Perineal pain, dyspareunia and anal incontinence
Klein, Canada, English, 1994 [17]	Cohort study; prospective, consecutive unclear	Primiparous and multiparous women, included in an RCT previously, having SVB	Incidence of PP and dysp-related intact perineum and degree of perineal trauma or episiotomy or 3rd/4th-degree perineal injury	24 women with intact perineum Second degree not included; method not reported	Perineal pain, dyspareunia and pelvic floor strength
Layton, UK, English, 2004 [41]	Cross-sectional survey study, retrospective	Primiparous after SVB	Incidence of dysp-related intact perineum and first- or second-degree perineal trauma	89 women with intact perineum Second degree not included; because not all of them had been repaired	Dyspareunia and urinary incontinence
Leeman, USA, English, 2007 [20]	Cohort study, prospective, consecutive	Primiparous and multiparous women, included in a RCT previously, having SVB	Incidence of PP-related intact perineum and degree of perineal trauma or episiotomy or 3rd/4th-degree perineal injury	131 women intact perineum 1st and 2nd degree (grouped sutured and unsutured trauma) and episiotomy not included because no mention material and technique repair	Perineal pain, use of analgesia, anal incontinence and urine incontinence
McDonald, Australia, English, 2015 [11]	Cohort study, prospective, consecutive	Primiparous women, having birth	Incidence of dysp-related intact perineum and degree of perineal trauma or episiotomy	PP: 129 women intact perineum Dysp: 123 women intact perineum Any degree of perineal trauma or episiotomy not included because IVD has been included in same data	Dyspareunia and postnatal depression
Persico, Italy, English, 2013 [18]	Cohort study, prospective, consecutive	Primiparous and multiparous women, having birth	Incidence of PP and dysp-related intact perineum and degree of perineal trauma or episiotomy	169 women intact perineum No degree of perineal trauma or episiotomy included because IVD has been included and material and technique repair not mentioned	Perineal pain, use of analgesia and dyspareunia
Safarnejad, Iran, English, 2009 [19]	Cohort study, prospective, consecutive	Primiparous women, having birth	Incidence of dysp-related intact perineum and degree of perineal trauma or episiotomy	9 women intact perineum	Dyspareunia
					Perineal pain

Table 1 (continued)

Soares, Brazil, English, 2013 [42]	Cohort study, prospective, consecutive	Primiparous and multiparous women, having SVB	Incidence of PP-related intact perineum and degree of perineal trauma or episiotomy	Not included any degree of perineal trauma or episiotomy because material and technique repair not mentioned
<p>Study characteristics of all RCTs included in the Systematic Review of Perineal pain and Dyspareunia (N = 8)</p> <p>Author, country, language, year, reference</p>	<p>Was a validated measurement tool used? Name (if given), setting, mode of interview</p>	<p>Study 'data period', timing of when survey(s) undertaken</p>	<p>Extracted findings for perineal pain after SVB: incidence</p>	<p>Extracted findings for perineal pain or dyspareunia after SVB: intensity of pain (total women %)</p> <p>Use of analgesia</p> <p>Use of analgesia: All continuous technique vs only vagina continuous tech: - at day 1: 18 (58%) vs 19 (63.3%) - at day 4: 9 (29%) vs 11 (36.6%) - at 6 weeks: 11 (61%) vs 17 (56.6%)</p> <p>All continuous technique: - at 24 h: mild: 26 (38%), moderate/severe: 5 (7%) - at day 10: mild: 11 (16%), moderate/severe: 1 (1%)</p> <p>At day 1: slight: 40 (40%), moderate: 31 (31%) severe: 16(16%) - at day 2: slight: 43 (43%), moderate: 27(27%), severe: 7(7%) - at day 3: slight: 54 (54%), moderate: 10 (10%), severe: 0 (0%)</p> <p>Use of analgesia: Only subcuticular continuous tech: - at day 1: 51 (51%) - at day 2: 38 (38%) - at day 3: 21 (21%)</p> <p>At least subcuticular continuous tech: PP: - at day 3: 1: 18 (34.6%) 2: 25 (48%) 3: 6 (1.5%)</p> <p>Dysp: - at 2 months: 1: 1(20%), 2: 1(20%) 3: 0 (0%)</p>
Almeida, Brazil, Spanish, 2008 [21]	PP: numerical rating scale (0–10): validated Dysp: yes/no. Not validated Maternity ward and out-patient clinic, verbal interview	2001–2002, at 1 h, day 4 and 6 weeks	All continuous technique vs only vagina continuous technique: PP: - at 1 h: 3 (10%) vs 3 (10%) - at day 4: 15 (48%) vs 18 (60%) Dysp: - at 6 weeks: 5 (42%) vs 5 (45%)	
Aslam, Pakistan, English, 2015 [22]	VAS: validated 1–3: mild, 4–10: moderate/severe Maternity ward and out-patient clinic, verbal interview	Study data period: not reported, at 24 h and day 10	All continuous technique: - at 24 h: 31 (45%) - at day 10: 12 (17%)	
Beard, UK, English, 1974 [35]	Pain severe, moderate, slight and none: not validated Maternity ward, verbal interview	Study data period: not reported, at day 1, day 2 and day 3	At day 1: 87 (87%) - at day 2: 77 (77%) - at day 3: 64 (64%)	
Graczyk, Poland, Polish, 1998 [36]	0–3: 0 no pain–3 worst pain. Not validated. Maternity ward, verbal interview	1995, at day 3 and 2 months	At least subcuticular continuous tech: PP: - at day 3: 50 (77%) Dysp: - at 2 months: 3 (12%)	
McElhinney, UK, English, 2000 [23]	VAS: validated Maternity ward, verbal interview	1996, at 12 weeks	Vicryl or Vicryl rapid: - at 12 weeks: 20 (17%)	

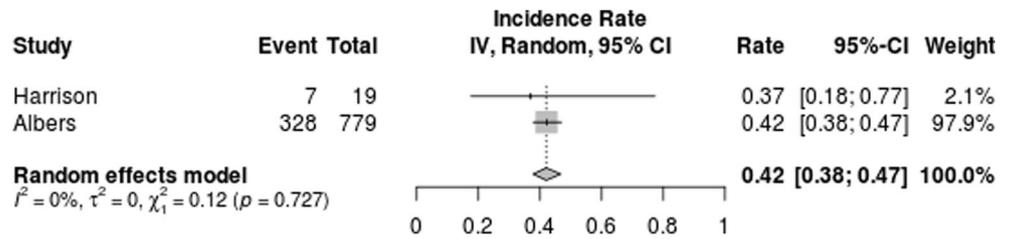
Table 1 (continued)

Morano, Italy, English, 2006 [24]	PP: VAS: validated Dysp: yes/no: not validated Self-administrated questionnaire and telephone	2003, at day 2 and day 10	All continuous techn vs only vagina cont tech: PP: - at day 2: 56 (52%) vs 89 (83%) - at day 10: 32 (32%) vs 58 (60%) Dysp: - at 3 months: 18 (21%) vs 18 (23%) All continuous technique vs only vagina continuous technique: PP: -at day 2: 9 (18%) vs 7 (14%) at day 10: 4 (8%) vs 6 (12%) at 6 weeks: 2 (4%) vs 3 (6%) Dysp: at 3 months: 3 (6%) vs 3 (6%)	Use of analgesia: All continuous tech vs only vagina cont tech: - at day 2: 36 (33.6%) vs 58 (54.2%)
Perveen, Pakistan, English, 2009 [37]	PP: yes/no: not validated Dysp: yes/no: not validated Setting: unreported, verbal interview	Study data period: not reported, at day 2, day 10 and 6 weeks	All continuous technique vs only vagina continuous technique: PP: - at day 2: 109 (49%) vs 113 (51%) - at day 10: 42 (19%) vs 41 (19%) - at 6 weeks: 6 (3%) vs 4 (2%) Dysp: - Resuming intercourse: 109 (51%) vs 110 (53%) - At 3 months: 78 (36%) vs 76 (37%)	All continuous technique vs only vagina continuous technique: - at day 2: slight: 80 (36%) vs 89(40%) moderate/severe: 29(13%) vs 24(10%) - at day 10: slight: 37 (17%) vs 34 (15%) moderate/severe: 5 (2%) vs 7(3%) - at 3 months: slight: 6 (2%) vs 4 (1%) moderate/severe: 0 (0%) vs 0(0%)
Valenzuela, Spain, Spanish, 2008 [25]	PP: analogous visual scale (0–10): No pain (0), slight pain (1–3) and moderate/severe pain (4–10) Dysp: yes/no. Not validated Maternity ward and telephone, verbal interview	2005–2007, at day 2, day 10 and 6 weeks, at resuming intercourse and 3 months	Extracted findings for perineal pain and dyspareunia after SVB: incidence (total women %) Intact perineum: PP: - at 2 days: 328 (41%) - at 10 days: 83 (10%) - at 3 months: 22 (3%) First degree (only outer vagina or labial tear): PP: - at 2 days: 566 (64.3%) - at 10 days: 222 (25.5%) - at 3 months: 45 (5.5%)	Extracted findings for perineal pain and dyspareunia after SVB: intensity of pain (total women %)
Study characteristics of all NRSs included in the Systematic Review of Perineal Pain (PP) and Dyspareunia (Dysp) (N = 10) Authors, country, language, year, reference	Was a validated measurement tool used, name (if given)? Setting Yes/no: no validated Setting: unreported, self-administrated questionnaire.	Study 'data period', timing of when survey(s) undertaken 1994–1996, at 2 days, 10 days and 3 months		
Albers, UK, English, 1999 [38]				
Harrison, Ireland, English, 1984 [39]	Scale 0–4. Not validated Verbal interview, maternity ward	July–August 1982, at day 1, day 2, day 3, day 4	Intact perineum: - at day 1: 12(63%) - at day 2: 7 (37%) - at day 3: 5 (26%) - at day 4: 2 (10%)	Intact perineum: - at day 1: mild: 8(42%), moderate 4(21%), severe 0 (0%), very severe 0 (0%) - at day 2: mild: 4(21%), moderate 2(11%), severe 1 (5%), very severe 0 (0%) - at day 3: mild: 5(26%), moderate 0(0%), severe 0 (0%), very severe 0 (0%) - at day 4: mild: 1(5%), moderate 0(0%), severe 0 (0%)

Table 1 (continued)

Kalis, Czech Republic, English, 2011 [40]	PP: verbal rating score (0–3) Validated Dysp: verbal rating score (0–3) Validated Out-patient clinic: Verbal interview and telephone PP: McGill Pain Scale 6-point scale ranging from 0 'no pain' to 6 'excruciating': Validated Dysp: 3-point scale. Not validated Out-patient clinic: verbal interview Yes/no: not validated Self-administrated questionnaire, setting unreported	2008–2009, at 6 months 1988–1990; at 3 months 1992–1999; < 6 months after SVB 2002–2005; at 6 months	Epiototomy: PP: - at 6 months: 0 (0%), dysp: at 6 months: 7 (14%) Intact perineum: PP: - at 3 months: 33 (30%) Dysp: at resuming intercourse: 68 (67%) Intact perineum: - up to 6 months: 8 (25%) Data not included in MA because did not mention date: from resuming intercourse to 6 months Intact perineum: - at discharge: not reported - at 6 weeks–3 months: 8 (9%) Not included in MA because did not mention day of discharge and data provided were for a period of time: from 6 weeks to 3 months Intact perineum: - at 6 months: 44 weeks (33.6%) - at 12 months: 30 weeks (22.7%) Intact perineum: PP: - at day 1: 93(72%) - at 6 months: 1(0.8%) Dysp: - at 7 weeks: 20 (25.6%) - at 6 months: 6 (4.9%) Intact perineum: - at resuming intercourse: 107(63.3%) - at 3 months: 47 (27.8%) - at 6 months: 31 (18.3%) - at 12 months: 19 (11.2%) Intact perineum: - at day 1: 6 (67%) - at 8 weeks: 0 (0%)	very severe 0 (0%) At 3 months: 17 women (all interviewed women) None-mild discomfort: 17 Distress-horrible/excruciating: 0
Layton, UK, English, 2004 [41]	Visual analogue scale and present pain intensity Validated Out-patient clinic: verbal interview and telephone	2003–2005, from 6 different hospitals, at 3 months, 6 months, 12 months and 18 months		
Leeman, USA, English, 2007 [20]	Postnatal sexual health. Not validated Australian longitudinal women's health study Validated Self-administrated questionnaire, unreported	2009–2010, at day 1, day 7 and 6 months		
Persico, Italy, English, 2013 [18]	PP: VNS (0–10) and VRS (none, mild, moderate, strong, very strong). Validated Dysp: VNS (0–10) and VRS (none, mild, moderate, strong, very strong). Validated Maternity ward and telephone: verbal interview Female Sexual Function Index (FSFI) Validated Setting: not reported, verbal interview	2005–2006, at resuming intercourse, 6 months and 12 months 2010–2011, at day 1 and 8 weeks		
Safarinejad, Iran, English, 2009 [19]	Numerical pain scale, pain-related self-statements Scale Catastrophising subscale Validated Maternity ward and telephone: verbal interview			
Soares, Brazil, English, 2013 [42]				

Fig. 2 Intact perineum. Perineal pain at day 2



respectively. However, although this was a large study of 5471 primiparous and multiparous women, the authors chose to include only labial tears and lower vaginal trauma into a sub-group of ‘first-degree perineal trauma’ and all ‘other types of first-degree perineal trauma’ were grouped and analysed with any second-degree perineal trauma.

Perineal pain after 2nd-degree perineal trauma or episiotomy repaired with the continuous technique and absorbable material

Meta-analysis of three studies [24, 25, 37] showed an incidence of perineal pain at day 2 postpartum of 39% (379 women; 95% CI 0.21 to 0.70; $I^2 = 78\%$) for women with a second-degree tear or episiotomy following SVB with all layers of trauma sutured by the continuous technique. A further meta-analysis of five studies [21, 22, 24, 25, 37] showed that at day 4–10 postpartum, the incidence of perineal pain had reduced to 23% (465 women; 95% CI 0.14 to 0.38; $I^2 = 78\%$). However, the I^2 values for both of these meta-analyses was 78%, showing high heterogeneity.

Unlike all of the other studies included in the two meta-analyses, Perveen et al. (2009) [37] did not use a validated questionnaire in their study and the total score for study quality assessment was very low. Repeat meta-analyses were performed with this study excluded that showed a higher rate of perineal pain at day 2 postpartum of 50% (329 women; 95% CI 0.43 to 0.58; $I^2 = 0\%$) (Fig. 4) and a slightly higher rate of perineal pain at 4–10 days postpartum of 27% (415 women; 95% CI 0.17 to 0.42; $I^2 = 77\%$) (Fig. 5). However, the I^2 value for 4–10 days postpartum remained high.

Regarding the intensity of pain for women with a second-degree tear or episiotomy following SVB with all layers of trauma sutured by the continuous technique, Aslam et al. (2015) [22] reported that 7% of the cohort of 69 primiparous women showed moderate or severe perineal pain at day 2,

decreasing to only 1% at day 10. Similarly, in the cohort of 216 primiparous and multiparous women in the study by Valenzuela et al. (2009) [25], 13% at day 2 and 2% at day 10 felt moderate or severe pain.

From all of the studies included in this systematic review, there were not enough extracted data suitable to undertake a meta-analysis for perineal pain at 6 weeks, 3 or 6 months.

Perineal pain after the second perineal trauma or episiotomy where only the vaginal tissue was repaired with the continuous technique

Meta-analysis of three studies [24, 25, 37] showed an incidence of perineal pain for women with a second-degree tear or episiotomy following SVB with only the vaginal mucosa sutured by the continuous technique of 41% (378 women; 95% CI 0.15 to 1.0; $I^2 = 93\%$) at 2 days postpartum. A further meta-analysis of four studies [21, 24, 25, 37] showed that at 4–10 days the postpartum rate of perineal pain was 31% (393 women; 95% CI 0.14 to 0.68%; $I^2 = 93\%$). Because of the considerable methodological differences between Perveen et al. [37] and the other studies [21, 24, 25], the meta-analysis was repeated with the exclusion of this study. Despite the fact that the rate of perineal pain observed at both day 2 and 4–10 days postpartum was higher than previously, 65% (329 women; 95% CI 0.40 to 1.0; $I^2 = 92\%$) and 41% (343 women; 95% CI 0.19 to 0.87; $I^2 = 94\%$), respectively, the heterogeneity still remained very high.

Perineal pain after episiotomy when subcutaneous tissue was repaired with the continuous technique

Meta-analysis of two RCTs [35, 36], showed an incidence of perineal pain of 69% (165 women; 95% CI 0.58 to 0.83; $I^2 = 0\%$) at 3 days postpartum for women who had episiotomy

Fig. 3 Intact perineum. Perineal pain at 4–10 days

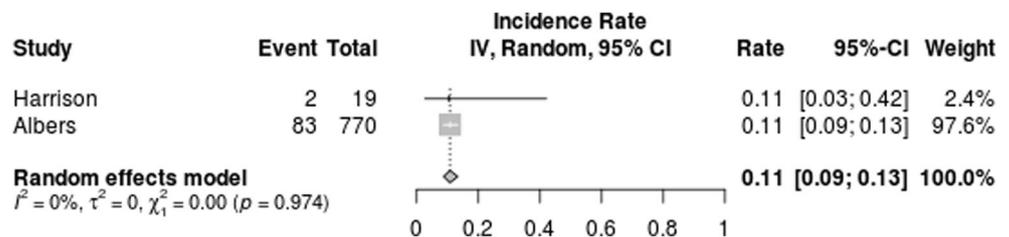
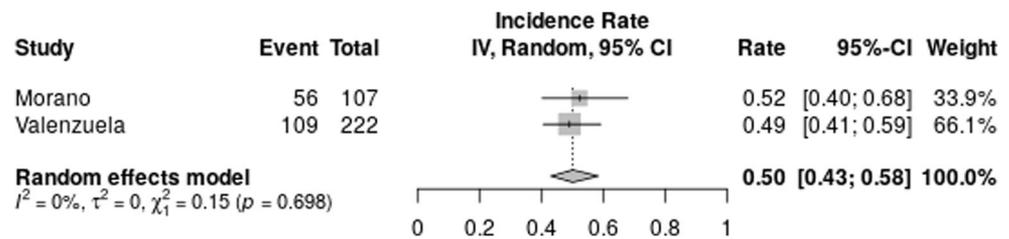


Fig. 4 Second-degree perineal trauma or episiotomy repaired with the continuous technique throughout all layers. Perineal pain at day 2, excluding Perveen



during SVB and at least subcutaneous tissue was sutured with the continuous technique (Fig. 6).

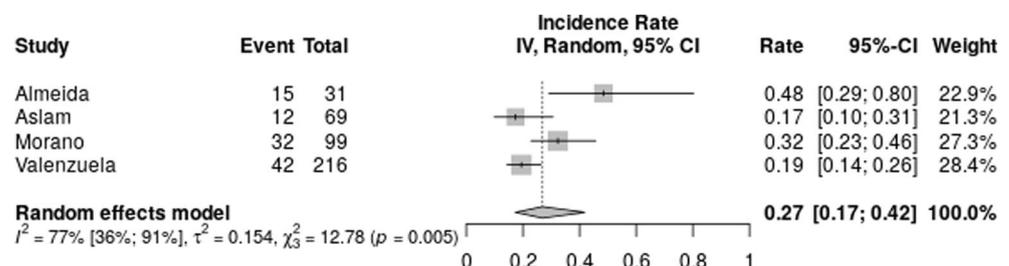
Regarding pain intensity, the cohort of 200 primiparous and multiparous women in the study by Beard et al. (1974) [35] stated that moderate pain was reported by 10% at day 3, whereas in a cohort of 117 primiparous women in the study by Graczyk et al. (1998) [36], 49.5% reported that the perineal pain was moderate or severe.

Dyspareunia after SVB

Dyspareunia after intact perineum

Meta-analysis of two studies [17, 19] showed an incidence of reported dyspareunia upon resumption of sexual intercourse following SVB with an intact perineum of 65% (270 women; CI 0.56 to 0.75; $I^2 = 0\%$) (Fig. 7). Three studies [11, 18, 19] provided data on dyspareunia for women with an intact perineum following SVB at 6 months postpartum and meta-analysis of these gave a rate of 15% (423 women; 95% CI 0.05 to 0.44; $I^2 = 91\%$). Two of the studies [11, 19] included in this meta-analysis involved women who had undergone their first SVB. However, the study by Persico et al. (2013) [18] used a cohort of 123 nulliparous and multiparous women. The meta-analysis was therefore re-run with Persico et al. (2013) excluded and showed a slightly higher incidence of 25% but not much improvement in I^2 (0.14 to 0.45; $I^2 = 85\%$). Interestingly, the presence of dyspareunia for women with an intact perineum following SVB remained similar at 12 months postpartum, with data pooled from two studies [11, 19] showing an incidence of 16% (301 women; 95% CI 0.08 to 0.32; $I^2 = 83\%$) (Fig. 8). However, the high heterogeneity among these studies must be considered.

Fig. 5 Second-degree perineal trauma or episiotomy repaired with the continuous technique throughout all layers. Perineal pain at day 4–10 days, excluding Perveen



Dyspareunia after first-degree perineal trauma

No studies that met the inclusion criteria for SR reported the incidence of dyspareunia following first-degree perineal trauma at the time of SVB.

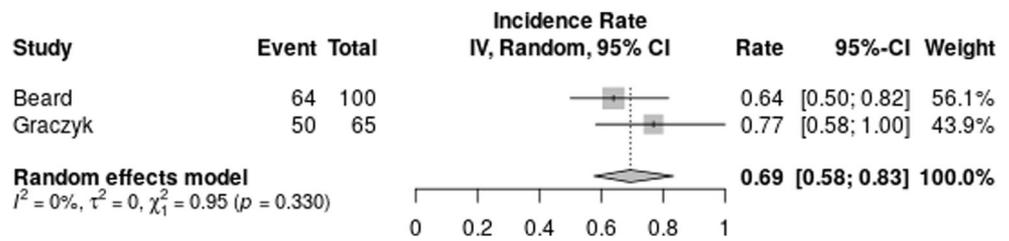
Dyspareunia after second-degree perineal trauma or episiotomy repaired with the continuous technique and absorbable material

There were not enough data suitable for meta-analysis of the incidence of dyspareunia at resuming intercourse. In a study by Valenzuela et al. (2009) [25] of 198 mixed parity women, 109 (55%) acknowledged dyspareunia the first time they resumed intercourse.

Meta-analysis of two studies [21, 37] showed an incidence of dyspareunia for women with a second-degree tear or episiotomy following SVB with all layers of trauma sutured by the continuous technique of 16% (95% CI 0.02 to 1.0; $I^2 = 86\%$) at 6–7 weeks postpartum. However, the high I^2 value is probably due to the very small number of women ($n = 62$) and low-quality assessment scores of the included studies, so this finding must be interpreted with caution.

Regarding longer term dyspareunia, meta-analysis of three studies [23–25] demonstrated an incidence of 19% (434 women; 95% CI 0.13 to 0.28%; $I^2 = 63\%$) for dyspareunia at 3 months postpartum. The study by McElhinney et al. (2000) had a much lower quality score, which may have contributed to the moderate I^2 value of 63%. The meta-analysis was redone with this study excluded; the rate of dyspareunia for women with a second-degree tear or episiotomy following SVB with all layers of trauma sutured by the continuous technique was slightly higher at 23% (281 women; 95% CI 0.18 to 0.30; $I^2 = 0\%$), with improved homogeneity (Fig. 9).

Fig. 6 Episiotomy repaired with the continuous technique at least in the subcuticular tissue. Perineal pain at 3 days



The incidence of dyspareunia at 6 months postpartum was only reported in one study where Kalis et al. (2011) [40], in a small study of 51 nulliparous women with episiotomy and continuous repair technique to all layers, reported an incidence of 14%.

Dyspareunia after second-degree perineal trauma or episiotomy where only the vaginal tissue was repaired with the continuous technique

There were not enough data suitable for meta-analysing the incidence of dyspareunia at resuming intercourse for this group. However, in the study by Valenzuela et al. (2009), 59% of women (110/186) with either a second-degree tear or episiotomy following SVB with only the vagina mucosa repaired using the continuous technique reported dyspareunia the first time they resumed sexual intercourse. At 3 months postpartum, a meta-analysis of two RCTs [24, 25] showed an incidence of 28% (263 women; 95% CI 0.22 to 0.35; $I^2 = 0\%$) for women with a second-degree tear or episiotomy following SVB with only the vaginal mucosa repaired using a continuous suture technique.

Dyspareunia after episiotomy where the skin was closed using a subcutaneous continuous technique

The incidence and intensity of dyspareunia were only reported in one small cohort study by Graczyk et al. (1998) [36] who found 12% (3/24) of the women reported pain as mild or moderate during sexual intercourse at 2 months postpartum.

Discussion

Regarding perineal pain, meta-analysis demonstrated that women with no perineal trauma still reported pain at 2 days, 10 days and 3 months postpartum. Although at 2 days the rate

of pain is similar to that reported by the women who sustained perineal trauma, it is more likely for pain to be reported for longer in case of a second-degree tear or episiotomy. Women with an episiotomy reported the highest rate of perineal pain after SVB compared with other degrees of perineal trauma. However, the incidence is lower when a continuous repair technique has been used. The intensity of perineal pain and use of analgesia in the postpartum period were not always adequately reported in the included studies limiting our ability to understand the natural history of this symptom.

Dyspareunia is frequently reported by women following SVB, even without perineal trauma, and sometimes this persists into the longer term. Our metaanalysis has also shown that dyspareunia is frequently experienced by women following a second-degree perineal tear or episiotomy at the time of SVB. However, this seems to be less likely to be reported if the trauma has been repaired with the continuous technique for all layers compared with when the vaginal epithelium is the only layer repaired using this technique. Due to confounding factor of the hypoestrogenic state when breastfeeding on postnatal dyspareunia, we attempted to extract data on breastfeeding and lubrication disorders. Safarinejad et al. (2009) [19] was the only study that reported data on both aspects. Although lubrication disorders were reported at 3, 6 and 12 months postpartum, being slightly higher with episiotomy, the study did not find any correlation between dyspareunia and perineal trauma for women who were breastfeeding.

Several published studies have reported relatively high rates of pre-conception dyspareunia or dyspareunia after undergoing an elective caesarean section [11, 43, 44]. It is therefore plausible that, for some women, postnatal dyspareunia represents a persistence of a problem that preceded the birth. Nevertheless, our findings highlight the importance of giving attention to this symptom even in women who did not sustain any degree of trauma and the need for further research into the aetiology and natural history of dyspareunia.

Fig. 7 Intact perineum. Dyspareunia at resuming intercourse

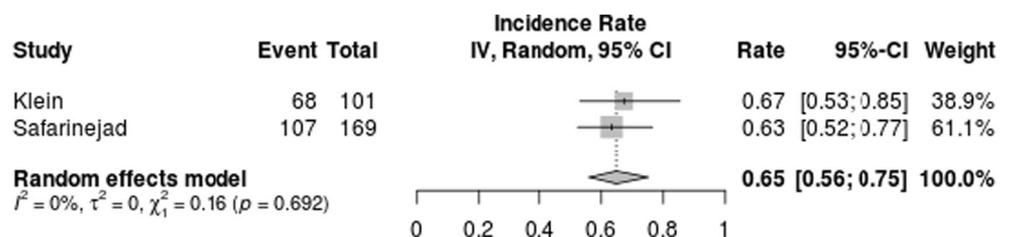
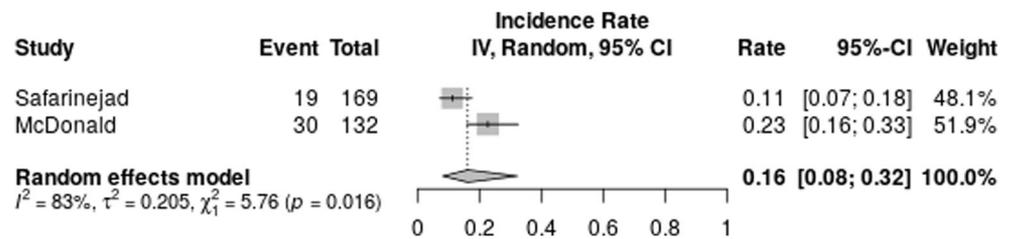


Fig. 8 Intact perineum. Dyspareunia at 12 months postpartum



Vaginal childbirth is generally accepted as a main risk factor for pelvic floor dysfunction, especially when the levator ani muscle (LAM) has been damaged [45]. One of our initial aims was to extract data on perineal pain and dyspareunia when an injury on LAM occurred during the SVB. However, none of the included studies reported on LAM injuries.

Strengths and weaknesses

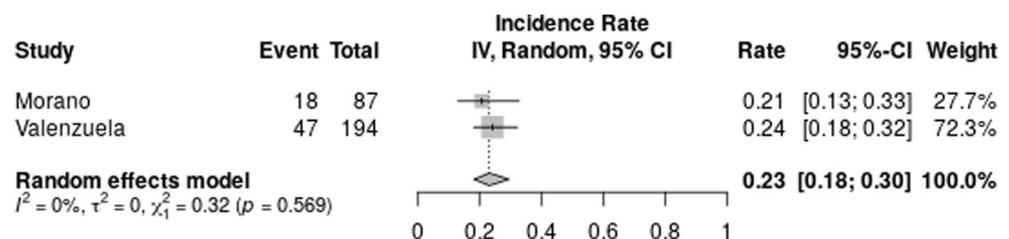
The main strength of this systematic review lies in its methodological aspects, namely, a rigorous search strategy as well as the study selection, quality appraisal and data extraction by independent reviewers and analysis following standardised protocols. The terms ‘anal sphincter’ or ‘OASIS’ or ‘instrumental vaginal birth’ were not included in the search to reduce the risk of limiting access to all possible relevant articles. We also searched the literature irrespective of study design and hence were able to include data from both randomised or non-randomised studies. However, we appreciate that the main limitation of our review arises from the quality and the heterogeneity of included primary studies. Of a total of 266 potentially eligible papers, 103 studies (41.5%) were not included because the suture material and/or method of repair had not been reported. However, a decision to exclude such studies was made a priori in view of the strong confounding effect of the methods and materials used for the repair on our primary outcomes of interest [13, 14, 46]. Moreover, the relatively short follow-up periods, insufficient information on pain intensity, variability in measurement tools and time points and small sample size of some studies were, at times, a limitation to our analyses. Nevertheless, we were still able to achieve the primary aims of the review. One of the main issues that our review has identified and, sometimes, limited our analysis was the inconsistency in the definition of the “perineum” among studies. The ‘Terminologia Anatomica’

report [47] highlights the term ‘perineum’ is often used in different ways. Within maternity care it is often used in its most restricted sense, equating to the perineal body [16], while anatomists refer to it in its widest sense to include all the structures within the urogenital and anal triangles [7]. We believe that the anatomical definition of the perineum is more inclusive and hence should be standardised and adopted by all disciplines. Additionally, although studies tended to follow the classification of trauma as proposed in the NICE Clinical Guidelines [48], authors of these studies did not always report results categorised by each degree of trauma and opted to present in other categories, for example, whether the trauma was sutured or not. Finally, some of the authors [49, 50] suggest that second-degree tears vary in depth and complexity and hence require some form of sub-categorisation or re-grouping to improve our understanding of trauma in relation to perineal pain and dyspareunia after birth. However, until such a sub-classification is clearly defined and standardised, outcomes related to this degree of trauma should be reported under the relevant category and not combined with first-degree tears or OASIs depending on whether they are deemed superficial or deep respectively.

Conclusion and implications

This systematic review highlights the current gap in our understanding about the size and, consequently, the potential impact of perineal pain and dyspareunia following childbirth on women’s health. Maternity-related healthcare professionals must appreciate that such morbidities are not only limited to women who experience perineal trauma. Further studies on the incidence of both perineal pain and dyspareunia related to intact perineum and perineal trauma after SVB are needed with particular attention to exploring the severity of these symptoms, use of analgesia and impact on the quality of life of the woman and her family at the short and longer term.

Fig. 9 Second-degree perineal trauma or episiotomy repaired with the continuous technique throughout all layers. Dyspareunia at 3 months, excluding McElhinney



Although not one of our initial aims, it became apparent that the definition of “the perineum” requires clarification and standardisation.

Compliance with ethical standards

Conflicts of interest None.

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