



# Cesarean delivery to prevent anal incontinence: a systematic review and meta-analysis

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## Abstract

**Background** Cesarean delivery (CD), is increasingly recommended as a mode of delivery that prevents the anal incontinence (AI) that arises in some women after vaginal delivery (VD). The assessment of the efficacy of CD in this regard was the subject of this systematic review.

**Methods** Searches were conducted in Medline, EMBASE and the Cochrane Library. Both randomized (RCTs) and non-randomized trials (NRTs) comparing the risk of sustained fecal and/or flatus incontinence after VD or CD were sought from 1966 to 1 January, 2019. Studies were eligible if they assessed AI more than 6 months after birth, and had statistical adjustment for at least one of the three major confounders for AI: age, maternal weight or parity. In addition, each study was required to contain more than 250 participants, more than 50 CDs and more than 25 cases of AI. Data after screening and selection were abstracted and entered into Revman for meta-analysis. Analyses were done for combined fecal and flatus incontinence (comAI), fecal incontinence (FI), gas incontinence (GI), CD before or during labor, time trend of incontinence after delivery, assessment of both statistical and clinical heterogeneity, parity and late incident AI.

**Results** Out of the 2526 titles and abstracts found, 24 eligible studies were analyzed, 23 NRTs and one RCT. These included women with 29,597 VDs and women with 6821 CDs. Among the primary outcomes, VD was found not to be a significant predictor of postpartum comAI compared to CD in 6 studies, incorporating 18,951 deliveries (OR = 0.74; 0.54–1.02). VD was also not a significant predictor of FI in 14 studies, incorporating 29,367 deliveries, (OR = 0.89; 0.76–1.05). VD was not a significant predictor of GI in six studies, incorporating 6724 deliveries (OR = 0.96; 0.79–1.18). The strength of the grading of recommendations, assessment, development and evaluations (GRADE) evidence for each of these was low for comAI and moderate for FI and GI (upgrade for lack of expected effect). Time trend FI showed incontinence at 3 months often resolved at 1 year. Other secondary analyses assessing parity, delayed incidence of FI, clinical and statistical heterogeneity, spontaneous VD only, late risk of incidence of AI, and CD in or prior to labor all had similar results as in the primary outcomes.

**Conclusions** There are three components of pelvic floor dysfunction that are thought to be caused by VD and hopefully prevented by CD: AI, urinary incontinence and pelvic floor prolapse. Of these, AI was not found to be reliably prevented by CD in this review.

**Keywords** Female · Humans · Pregnancy · Cesarean section · Delivery · Obstetric · Delivery · Obstetric/adverse effects · Fecal incontinence · Fecal incontinence/prevention and control · Flatulence · Flatulence/prevention and control

## Introduction

Anal incontinence (AI) may occur in women during the immediate postpartum period and persist throughout adult life [1]. The impact of AI on the mother is dependence upon protective undergarments, social isolation, and, as age advances, nursing home residence [2, 3]. Because of the multiple facets of AI, ascertainment of prevalence has been complex compared to urinary incontinence (wet or not

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wet), leading to a large variance in AI prevalence figures in published reports [2], varying from 1.4% to over 11% in healthy ambulatory populations and as much as 50% of nursing home residents [3].

The only intervention currently employed to prevent AI is Cesarean delivery (CD). CD may preserve maternal pelvic floor function by avoiding direct vaginal, sphincter and distal pelvic nerve trauma that might occur during vaginal delivery (VD). To assess the effectiveness of CD in this regard, studies in which women were either randomized to CD or VD [randomized controlled trials (RCTs)], or studies which compared cohorts or cross-sectional surveys of women having either CD or VD [non-randomized controlled trials (NRTs)], and in which they were investigated for the outcome 6 months or more after pregnancy of AI, were sought. When possible, emergency CD was separated from elective CD in the analyses, spontaneous vaginal delivery (SVD) from operative vaginal delivery (OVD; forceps or suction extraction), prima-parous pregnancies separated from multiparous women, and CD women with prior or subsequent VD separated from women with only prior CD.

## Materials and methods

Participants were women with a history of pregnancy and delivery of a live infant by VD (SVD or OVD) or CD (either electively or in labor as an emergency). Twin pregnancies were included in this review. Maternal postpartum gas incontinence (GI), fecal incontinence, (solid or liquid (FI), and combined GI and FI (comAI) are the primary outcomes. “AI” in this report refers to any one or all of these. The timing of the measurement of the outcome was limited to studies in which this assessment was done exclusively or predominantly more than 6 months after the index birth. This is because the outcome to be measured in this review is sustained incontinence rather than early dysfunction.

Further eligibility criteria for studies in this review include adjustment in the analyses for at least one of the known confounders of AI: maternal age, maternal obesity, and parity. There was a lower limit on the size of the cohorts in NRTs to 250 participants, at least 50 of whom had CD and at least 25 of whom had AI.

Medline, EMBASE and the Cochrane library central register of controlled trials were searched from 1966 to January 1, 2019. The search strategy is presented in the “Appendix”.

Reference lists and authors of relevant publications were also screened for potential studies. Data collection and analyses were conducted in accordance with the Cochrane Handbook for Systematic Reviews of Interventions [4]. Titles and abstracts of all articles retrieved by the search were scanned by three authors. The full texts of all potentially eligible studies were retrieved. Again three authors examined the

full text articles for compliance with the inclusion criteria and studies were selected for inclusion in the review. Study investigators were contacted if required to clarify study eligibility or data. Disagreements as to study eligibility were resolved by discussion and consensus of the whole group. Data were extracted from eligible studies by three authors using a data extraction form designed by the authors and once again disagreements resolved by group discussion.

Included studies, all but one being NRTs, were assessed for risk of bias, including the following:

- 1 a validated AI instrument was used to ascertain incontinence,
- 2 did AI occur before delivery or during pregnancy,
- 3 adjustment for maternal age,
- 4 adjustment for maternal obesity.
- 5 adjustment for parity,
- 6 analysis of emergency and elective CD separately (i.e., mothers in labor or not),
- 7 timing of the assessment of outcomes after more than 6 months post-partum, to assure that sustained incontinence was being assessed.
- 8 VD before or after a CD within the CD group
- 9 All VD or VD without OVD compared to CD

In each included study, the odds ratio for incontinence was obtained from logistic regression and converted to the natural log of the odds ratios and confidence intervals to calculate the standard error, using the VD as the reference. These values were entered into Revman and meta-analyses were performed to obtain a combined effect for each comparison along with calculation of heterogeneity. Heterogeneity significance is defined as a Chi-square  $p \leq 0.10$  and/or  $I^2 > 50\%$ .

When significant heterogeneity was found, it was explored for that outcome in subgroup analyses and clinical review. If the primary outcome of sustained AI was not assessed, this study was excluded. If a three-way comparison was done between VD, CD and nulliparous women, and the reference in the logistic regression was for nulliparous women, this was also excluded [5], because direct comparison of VD and CD alone was not possible. There was one study that fulfilled every inclusion criterion, however, the participants had a known obstetrical anal sphincter injuries (OASIS) secondary to a VD before the index pregnancy. That study was excluded [6].

Random effects analyses were done for all comparisons due to clinical and/or statistical heterogeneity except in the comparison of VD vs. CD before or during labor, wherein neither applied, in which case a fixed effect analysis was done.

The following outcomes were assessed in comparisons of VD to CD:

- 1 *comAI*: Combined anal incontinence. This is a subgroup analysis of eligible studies, as are all the remaining comparisons. This comparison includes studies that reported sustained combined FI and/or GI.
- 2 *FI*, the involuntary loss of solid and liquid stool.
- 3 *GI*, the involuntary loss of gas.
- 4 VD vs. CD, when the CD was done in women either *before labor*, or *after labor* had commenced.
- 5 *Time trend* of FI in three different cohorts which were reported first early in the postpartum period and then again later on several occasions after delivery. Only a qualitative forest plot comparison is shown without quantitative meta-analysis.
- 6 VD vs. CD after just *a single pregnancy*.
- 7 VD vs. CD *stratified by parity*.
- 8 Investigation of *heterogeneity*.
- 9 *Exclusion of SVD* from analyses to examine the effect of All VD (including SVD and OVD) vs. CD.
- 10 Exclusion of studies in which the postpartum period before incontinence was assessed was variable, and may have been less than 6 months in a minority of the women.
- 11 Late incidence (not prevalence) of AI.

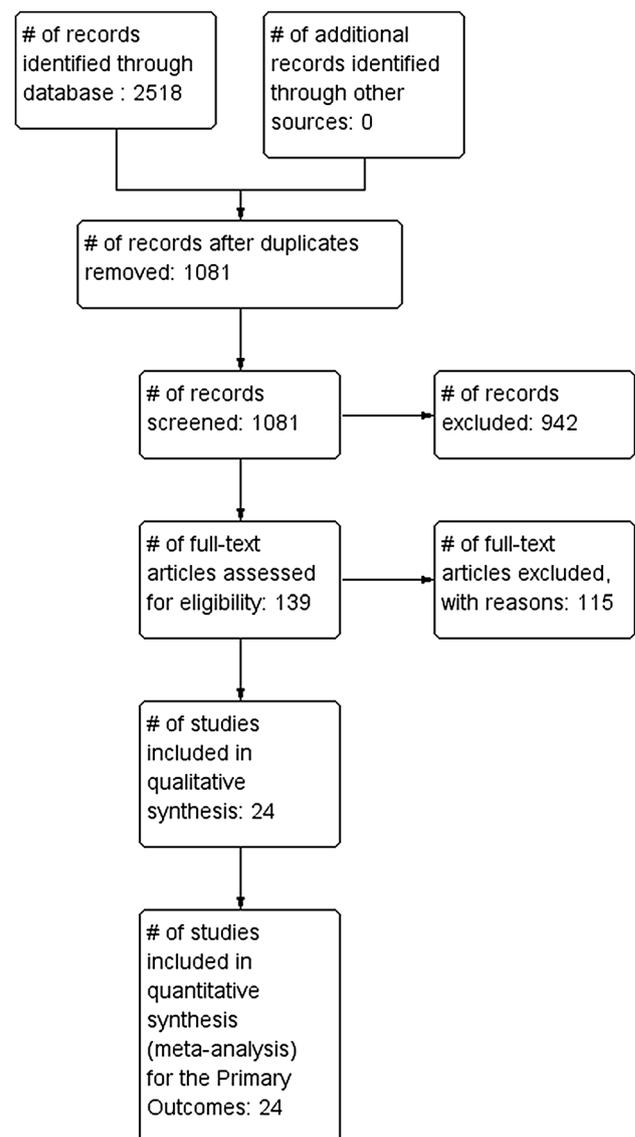
**GRADE:** The grade approach was used to classify the quality of evidence. GRADE can be upgraded from low in NRTs to moderate or high if there is an extreme effect ( $OR > 2.0$ , or  $< 0.5$  for one step, and  $OR > 5.0$  or  $< 0.2$  for two steps), a dose response effect (not relevant in this review), or when residual confounding is expected to reduce a demonstrated effect. Upgrading can also be done if a significant effect is anticipated and when no significant effect is observed [7].

## Results

The searches resulted in 2526 titles and abstracts. After removal of duplicates and screening of abstracts, 138 titles were obtained in full text and reviewed for eligibility (Fig. 1). Of these, 24 fulfilled the eligibility requirements [8–31]. Eligible studies included 29,597 VD women and 6281 CD women.

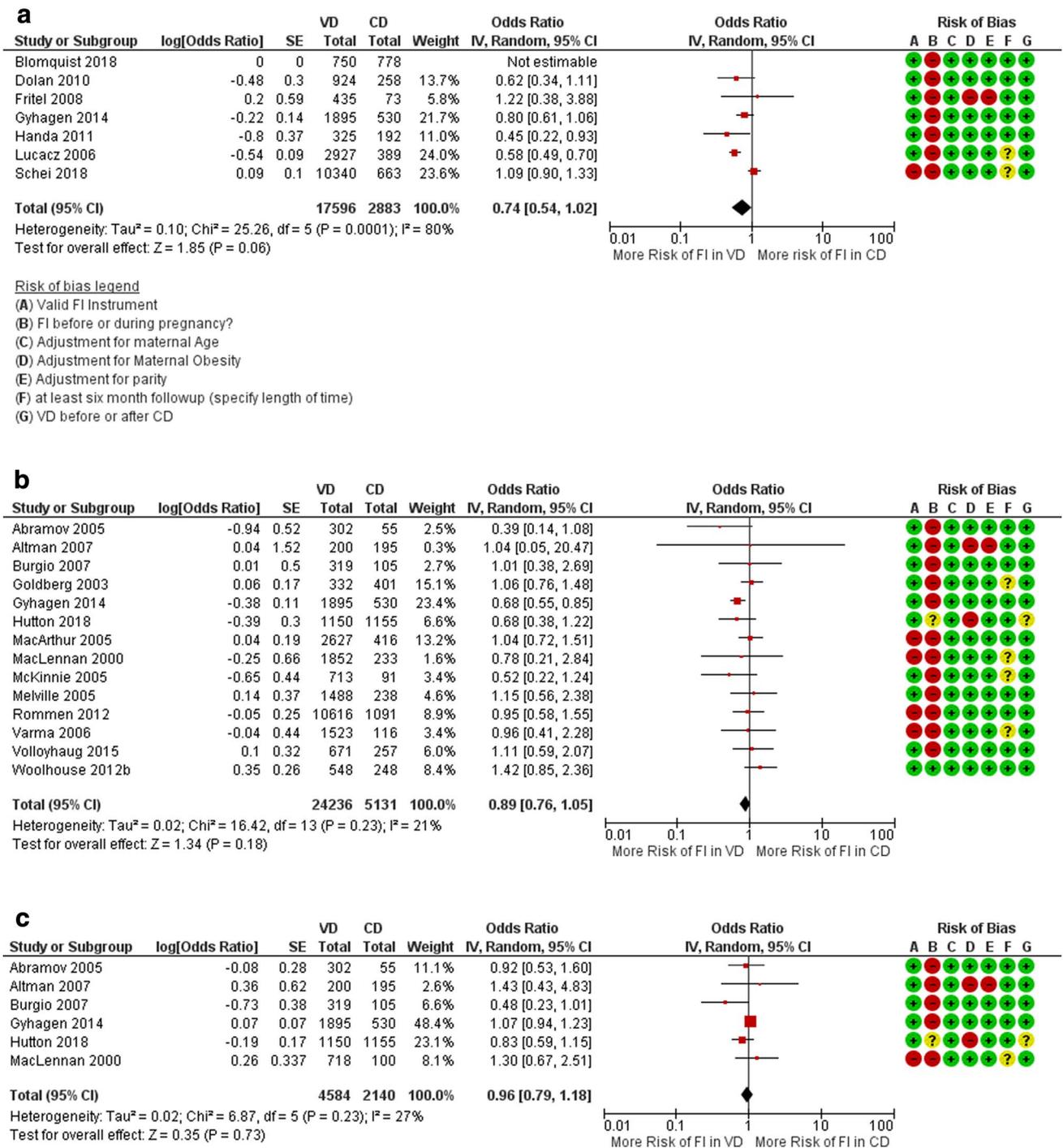
## Primary Outcomes

VD was found not to be a significant risk factor for postpartum *comAI* compared to CD in six studies [9–14], incorporating 18,951 deliveries ( $OR = 0.74$ ;  $0.54–1.02$ ) (Fig. 2a) (for Blomquist, see below [8]). VD was also not



**Fig. 1** Preferred reporting items for systematic reviews and meta-analyses (PRISMA) flow diagram

a significant risk for FI in 14 studies [15, 16, 18, 20, 21, 23, 25–31], incorporating 29,367 deliveries ( $OR = 0.89$ ;  $0.76–1.05$ ) (Fig. 2b). VD was not a significant risk for GI in 6 studies, incorporating 6724 deliveries ( $OR = 0.96$ ;  $0.79–1.18$ ) (Fig. 2c) [11, 15, 16, 18, 21, 25]. The forest plots show the natural log of the OR ( $\log[OR]$ ) with the standard error (SE) on the left and the OR and 95% confidence intervals (CI) on the right of the graph. Study characteristics are summarized in Table 1. If there was more than one name or citation in Column 1, that cohort was presented in more than one publication with a new first author or with varying duration since the index pregnancy or different outcomes reported.



**Fig. 2** Primary outcomes **a** combined fecal incontinence and gas incontinence (comAI) see text for Bloomquist [8]. **b** Fecal incontinence, **c** gas incontinence. VD vaginal delivery, CD cesarian delivery

**Further analyses**

There was no difference in 3 studies with 6836 deliveries comparing risk of FI of VD vs. CD in labor or CD done before the onset of labor (subgroup differences  $p = 0.97$ ,  $I^2 = 0$ ) [12, 17, 24].

Three cohorts presented follow-up assessment of FI at various times. In each case, the first comparison of VD to CD was 3 months postpartum. Assessment was then done again at 6 and 12 years in Mac [22–24], 1 and 4 years in Woolhouse [19, 31], and 2 years in Hannah (not an included study in the meta-analyses) [32, 33]. The 3-month

**Table 1** Characteristics of included studies

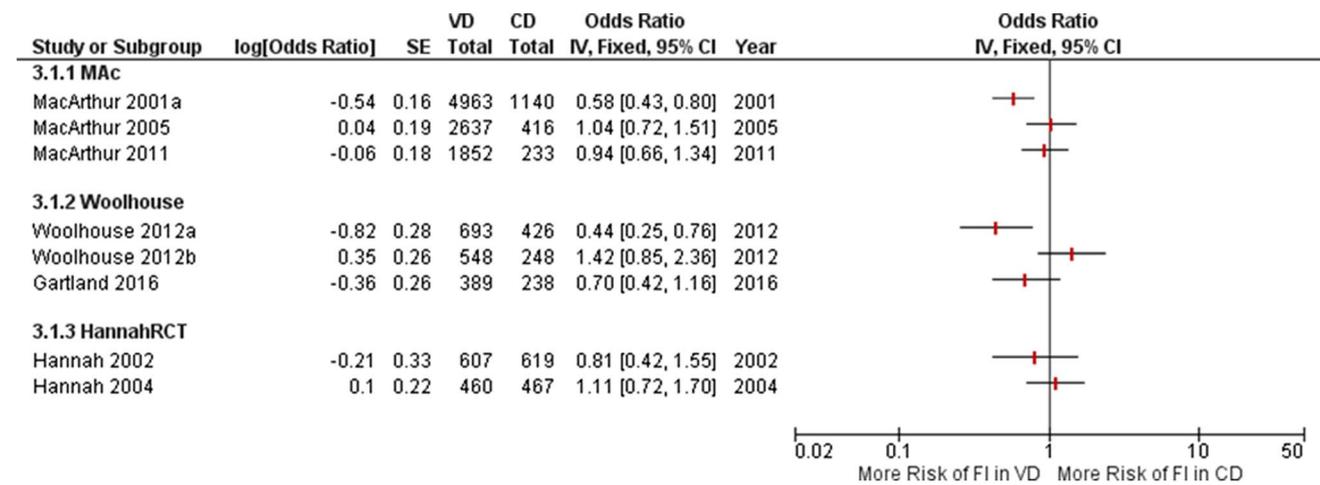
Author name and citation	Participant contact method	Fecal incontinence ascertained how long after birth?	Did any members FOF the cd group have prior or later VD births	Was CD compared to all VD or spontaneously only VD
Abramov [15]	Twin convention Twin sisters study	Variable, mean age 47 years	Primip at index pregnancy	All
Altman [16]	Postal survey	10 years	Primip at index pregnancy	All
Blomquist [8]	Clinic visit	Continuous, enrolled 5–10 years postpartum, followed 9 years. Time and hazard ratios. Mean age at enrollment 38.3 years.	CD Only	SVD ref. and OVD
Burgio [18]	Telephone	6 months	Primip at index pregnancy	All
Dolan [9]	Post	20 years	Primip at index pregnancy	SVD
Fritel [10]	Post	4 years	Primip at index pregnancy	SVD
Goldberg [20]	Twin convention Mothers of twins study	Variable, median age 37 years. Mean 7.6 years since twin delivery.	CD group: CD only by history in later pregnancies	All
Gyhagen [11]	Post	20 years	Only one delivery	All
Handa [12]	Clinic	5–10 years	CD only	All
Hutton [21]	Clinic or home visit randomized trial to twin pregnancy	2 years	Not stated	All; RCT, randomized to VD
Lucasz [13]	Post	Variable, mean age 57 years	CD only	All
MacArthur [22–24]	Post	3 months, 6 years and 12 years	CD only	SVD
MacLennan [25]	Home visit	Variable, mean age 44.8 years	CD only	SVD
McKinney [26]	Not stated? Gynecology clinic?	Variable, mean age 42.7 years	CD only	All
Melville [27]	Post	Variable, mean age 52.9 years, ages 30–90 years	CD only	SVD
Schei, Rommen [14, 28]	Not stated (HUNT 3 health survey cohort)	Variable, ages 30–80 years. Assessment lasted 2 years after recruitment	CD only	All minus OASIS
Varma [29]	Not stated	Variable, All age > 40 years	CD only	All
Volleyhaug [30]	Post	Variable, mean age 47.3 years	CD only	SVD
Woolhouse (Brown and Gartland) [17, 19, 31]	Post	3, 6, 12, 18 months and 4 years	Primip at index pregnancy	SVD

VD vaginal delivery, CD cesarean delivery, SVD spontaneous vaginal delivery, RCT randomized controlled trial, OASIS overt obstetrical anal sphincter injuries, *Primip* first baby

assessment was outside the eligibility requirements for inclusion in this review. Similarly, the 2-year assessment was not eligible for quantitative meta-analysis in Hannah, since there was no adjustment made for confounders in that study. Therefore, only a qualitative demonstration of the change in risk of incontinence over time since delivery comparing VD to CD is shown in Fig. 3 without quantitative meta-analysis.

There was no significant difference in FI in studies of mothers having only a single baby, at 6 months postpartum [18], 20 years postpartum [11], a variable length of time to incontinence assessment in [28] and 1 year postpartum [31] (OR = 0.91; 0.63–1.32).

There was no difference in FI detected in women in a Norwegian cohort of 11,071 women, the HUNT 3 cohort, with parity varying from 1, 2, or  $\geq 3$  deliveries (subgroup differences;  $p = 0.56$ ,  $I^2 = 0$ ) [28].



**Fig. 3** Time trend for patients with postpartum FI. The date after each dated author's name from top to bottom of each cohort shows the duration of follow-up for that cohort, so 12 years for MacArthur,

4 years for Woolhouse and 2 years for Hannah. Movement to the right indicates improving AI after 1 year. *VD* vaginal delivery, *CD* cesarian delivery

## Heterogeneity

Significant statistical heterogeneity was detected in two meta-analyses: comAI (Fig. 2a) and FI assessment after women having only one baby. Exclusion of Schei et al. [14], from Fig. 2a resolved that heterogeneity, from  $p = 0.0001$ ,  $I^2 = 80\%$  to  $p = 0.21$ ,  $I^2 = 31\%$ . It is the largest study in this review. There were no methodological flaws found. The assessment of comAI was thorough but no validation of their questionnaire was cited. The protective effect of VD also became more apparent with this exclusion, from  $OR = 0.74$ ;  $0.54–1.02$  to  $OR = 0.65$ ;  $0.53–0.81$ . In comparison of women with only one pregnancy, exclusion of either Gyhagen [11] or Woolhouse [31] resolved the heterogeneity with opposite effects on the summary risk of FI.

This review has much clinical heterogeneity, from mothers of twins [20, 21] to large variations in age groups [14, 18] assessed and variations within exposure groups, in some cases separating CD in and before labor [12, 17, 24], and in some cases separating SVD from OVD. There is one study that stood out. This is a cohort that had incontinence assessment 20 years after a first pregnancy with no subsequent pregnancies [11]. A question is raised as to whether events in that only pregnancy resulted in a decision to not get pregnant again and how this clinical heterogeneity might have influenced results. This study is heavily weighted in the comparisons of which it is part, not because of its size, but because of very narrow confidence intervals (Fig. 2a–c, and single delivery). Exclusion of this study had little effect on analyses Fig. 2a, c. However, compared to Fig. 2b, excluding [11], the mode of birth showed some difference in FI ( $OR = 0.98$ ,  $0.83–1.16$ ). A similar difference is seen comparing single deliveries with the exclusion of [11], ( $OR = 1.10$ ;

$0.79–1.54$ ). There is only this clinical difference reported and no methodological issues that separate this from others in the review.

## Late incident FI

One publication looked at this problem from a different perspective [8]. It describes incident cases of AI, urinary incontinence and pelvic organ prolapse, beginning with enrolling 1528 participants from a single institution, 750 with CD, 565 with SVD and 185 with OVD, 5–10 years after the index delivery of a first baby. Enrollees were matched for CD and VD and age at first delivery. Women with SVD were the reference with comparisons to comAI and OVD. This cohort was followed annually for 9 years after enrollment, with a fair amount of attrition during that time. Instead of ORs, the incident comAI risk is presented as time and hazard ratios, and so this study cannot be included in the above meta-analyses. Adjustment was made for the three major confounders. The adjusted hazard ratio (HR) for AI =  $0.77$ ;  $0.55–1.08$ , and for OVD =  $1.70$ ;  $1.11–2.59$ . Parity, as in [28] was not found to be a significant risk for comAI comparing VD to CD.

## Risk of bias

The risk of bias in each comparison is shown for each primary outcome with the forest plots (Fig. 2). A key for the color coding is present in Fig. 2a. In general, the risk of bias for all comparisons was low, due to the eligibility criteria. The one factor for which risk of bias is significant for all but 2 studies was assessment of AI during pregnancy. This is a significant predictor of AI in the postpartum period [34]. No study adjusted for this in the logistic regression. In the

studies for which a validated continence instrument was not used, the assessment of FI appeared to be thorough.

Assessment of AI more than 6 months after the index birth was both an eligibility criterion and a risk of bias factor. That is because it was decided to include certain studies which had unspecified and variable times of assessment [13–15, 20, 25–27, 29, 30]. There was evidence that the vast majority of these assessments were not carried out within 6 months of birth. This is found from the age range and mean age of the participants, and other factors in Table 1.

Exclusion of these studies with variable AI assessment intervals in the meta-analyses shifted the odds ratio for comAI from 0.74 to 0.73, for FI from 0.89 to 0.93, and for GI from 0.90 to 0.91. The small shift of the ORs to the right for FI and GI may imply that a few assessments might have been done less than 6 months postpartum.

Seventeen studies grouped all types of VD (SVD and OVD) together, while 7 (besides [8]) separated SVD from OVD [9, 10, 23, 25, 27, 30, 31]. Exclusion of the SVD studies in the primary outcomes had very little effect on the summary statistic: for Fig. 2a, OR from 0.74 to 0.73, for Fig. 2b, OR 0.86 to 0.78, and for Fig. 2c, OR from 1.02 to 1.01.

## Grade

Risk of bias as stated above was not serious in any comparison. Both statistical and clinical heterogeneity were detected in the review. The causes were investigated. Exclusion of studies found to be responsible resolved the heterogeneity and in most cases with minor alterations in the summary statistic of efficacy of CD. Indirectness was not a problem in these studies, nor was imprecision. A funnel plot shows no clear evidence of publication bias or small studies effect (since there were size thresholds as eligibility requirements). Therefore, for each of the primary outcomes, the GRADE was Low for comAI (down one for significant heterogeneity and up one for lack of expected effect), and Moderate for FI and GI, with upgrading due to the lack of the expected effect that CD had on AI risk.

## Discussion

A meta-analysis of published reports [1] estimated that one-quarter (after 1 VD) to one-third (after multiple VDs) of women will have occult sphincter injury detected by ultrasound, though many did not have symptomatic AI. Of women with FI, in the Bayesian analysis it was found that 77% (primip) to 89% (multip) had sphincter defects. The strength of these associations would seem to support sphincter injury as a causative mechanism for FI. For certain types of sphincter injuries that is not controversial such as an open

tear requiring immediate repair (OASIS) or OVD [8]. However, three things are implied by this conclusion. The first is that women having VD, with this injury added to other risk factors, should have a much higher prevalence of AI than those in the population that have never had VD. There is scant epidemiological evidence that this is the case [3].

Second, it is implied that sphincter repair of an occult anal sphincter injury would be effective treatment for anal incontinence for many women. Yet repair of a disrupted sphincter has less than a perfect track record. Even more importantly, there is a reported rapid decay in function after repair that is far too great to be explained by age alone [33–42].

Third, if direct trauma to the anal sphincter and its distal enervation (and not intra-pelvic nerves) were the major cause of AI, then CD should be highly effective in preventing incontinence. Based upon the first sentence of this discussion, the protection provided by CD over VD should be at least 50% and probably very much more. That is not approached in any of the comparisons in this review. One study with large weighting in the analyses had a statistically significant benefit for CD over VD for FI, and also possibly an important clinical heterogeneity [11]. In this study the OR (0.68) was still not in the extreme effect range.

CD is a life-saving procedure for many infants and mothers. However, over the past decades there has been a lively debate concerning the health benefits of elective primary CD [43–53], in some cases known as Cesarean delivery on maternal request (CDMR) [54], or no indicated risk CD [55]. The diversity of opinion even among those most expert in the field is provided by a survey in London, wherein 31% of female obstetricians stated that they would elect to have a CDMR [51] although in Holland the figure among female obstetricians was only 1.4%. [56].

From 1994 to 2014, the rate of CD has more than doubled worldwide [57].

Many reasons are given for a woman choosing to have a CDMR. In Taiwan, timing of delivery to certain days of the week is associated with lifelong good fortune for the baby [58]. Women in Brazil believe that CD, a more aggressive and sophisticated intervention, is better for the health of the baby, and that it is better medicine [59]. Socio-economic status also correlates with CDMR rates in Brazil and Chile, where CD occurs more than twice as often in private patients, exceeding 70% of all deliveries in wealthier classes [59–61]. However, the most common reason stated for CDMR is preservation of maternal pelvic floor function: continence of urine, feces, flatus, and prevention of pelvic floor prolapse [50, 53].

However, CD is also associated with a number of health risks when compared to VD, both to mother and infant. For the mother, the risk is that of abdominal surgery in general. A laparotomy would be repeated with each pregnancy, since most CDs are followed only with CDs. Bleeding, infection,

adhesions, prolonged hospital stay, transfusion, wound infection, bowel obstruction can all occur with abdominal surgery and are more likely with repeat operations. Also, repeated trauma to the uterus might cause rupture. For the child, problems may persist for years after CD, including asthma, obesity, food allergy and risk of many types of infection [62, 63]. It is not clear whether the risk of these problems singly or cumulatively would approach the risk of pelvic floor disorders with VD, and, for each type of disorder, how much CD alters that risk. The cost of CD has been increasing in part due to these added risks [64].

## Other systematic reviews

Other reviews have been published, but not with the eligibility limitations of this review [62, 63, 65–67]. Studies that have focused on urinary incontinence and pelvic floor prolapse have found that CD does have a significant protective effect over VD [9, 62, 68]. However, none of these systematic reviews have had inclusion of RCTs of average risk pregnancies, much less the exclusive inclusion of such studies, to do their meta-analyses. This includes this current review.

## Randomized-controlled trials

Mode of delivery is a very significant public health issue. CD rates in some countries exceed 50 per cent [57, 69]. That a randomized trial of average risk pregnancies has not yet been done suggests that such trials may not be done soon, if ever. Several reports have explored why this is the case [70–73], all suggesting that the clinical situation is too complex and that preconceived preferences and biases among both patients and doctors would make randomization nearly impossible. Yet 2 large RCTs have been done [21, 33] in even more complex clinical situations: breech presentation and twin pregnancy. Both were large multicenter international studies centered in Canada. The first [33], the Term Breech Trial, randomized 2088 women and had an anticipated very large 56% crossover in delivery mode after randomization. No adjustment was made for confounders of AI in their analyses. It could, therefore, only be analyzed for maternal AI as treated in a previous systematic review [65]. In [21], the Twin Trial, the same authors recruited and randomized 2305 mothers of twins. The crossover rate was halved compared to the Term Breech Trial and adjustment was made for the three major confounders of AI. Therefore, it was included in this systematic review, despite being a methodological outlier. As the only RCT (deliveries were analyzed as randomized, not as treated), it was weighted only 6.6% in the meta-analyses. If it is excluded from the

analysis of FI because of methodological heterogeneity, the shift is very small towards no protective effect of CD in Fig. 2b (OR = 0.91; 0.71–1.09). There are also 4 very small RCTs of women in preterm labor randomized to CD or expectant VD reported in a Cochrane review [74]. All 4 studies were stopped because of difficulty in recruitment. Only 116 participants were analyzed. The recruitment for the Term Breech and Twin Trials is remarkable, some consents even being done during labor.

## Conclusions

The last study described in the Results is one of the most interesting both in its conception and results. Bloomquist [8] specifically and uniquely looked at late incident AI beyond child bearing years. The other studies included in this review focused instead on prevalence of AI again mostly in women beyond child bearing years (Table 1).

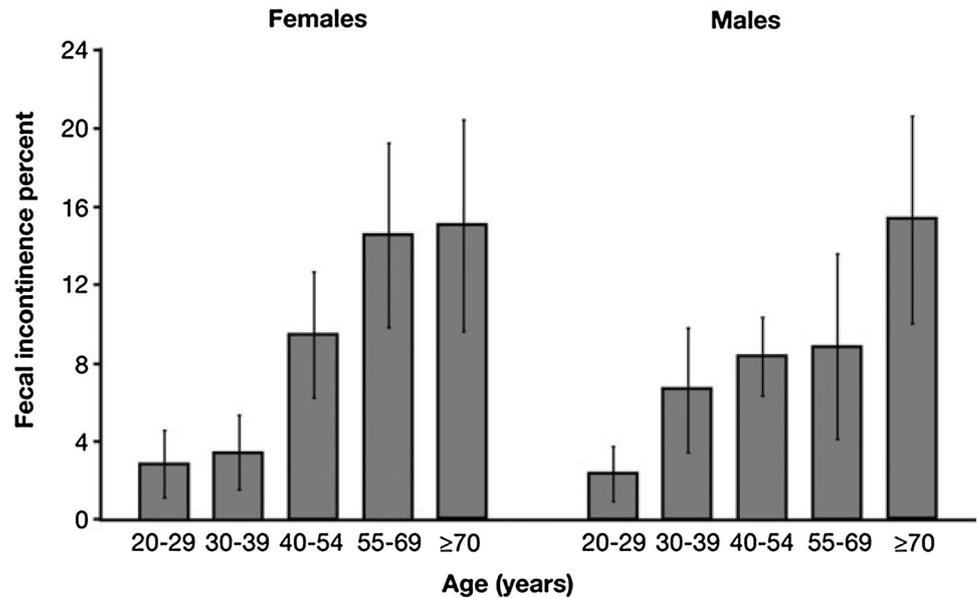
One of the conundra of the demographics of AI in women is that AI is commonly thought to be associated with VD, yet there is no spike in prevalence of AI in the child bearing years (Fig. 4). Rather the spike occurs, especially when women are compared to men, in middle age: 40–69. The survey in Fig. 4 [75], The National Health And Nutrition Examination Survey 2005–2006 (NHANES) measured FI as any involuntary loss of solid or liquid stool within the last 30 days prior to their interview.

So who is at a relative increase in risk for AI? Certainly women with VD have a greater risk than women who have never been pregnant. But so do women who have had only had CD, when compared to nulliparous women [5, 25, 28], at a rate that is not significantly different from VD. Women who have AI during the third trimester are at increased risk of postpartum AI [34]. This has not been adequately investigated nor has adjustment for this been done in studies that had adjusted for other confounders. And women beyond their childbearing years are at increased risk when compared with men, but this difference is no longer apparent in the over 70 age group (Fig. 4).

The ORs and CIs of the analyses in this review do show a trend towards increased risk of AI after VD compared to CD, but again, not nearly of the magnitude anticipated by the sphincter injury literature. The HR for newly incident FI when comparing VD and CD was similar to those found in the meta-analyses [8]. Sphincter injury cannot be ignored, but other mechanisms must be considered such as trauma to pelvic nerves in the third trimester.

RCTs of women with average risk pregnancies and with adequate follow-up for AI would provide stronger evidence of these relationships. The term breech trial and twin trial both show that such RCTs are feasible [21, 32].

**Fig. 4** Prevalence of fecal incontinence (not gas) in the USA 2005–2006 NHANES [75]



### Compliance with ethical standards

**Conflict of interest** The authors declare that they have no conflict of interest.

**Ethical approval** This article does not contain any studies with human participants or animals performed by any of the authors.

**Informed consent** For this type of study, formal consent is not required.

### Appendix

#### Search strategy for NEL103: cesarean delivery for the prevention of anal incontinence

Medline (Ovid) 1/1/2019

#### Search history

1. exp Cesarean Section/
2. exp Delivery, obstetric/
3. [((cesarean or caesarean) adj section) or delivery or c? section or c-section].m\_titl.
4. 1 or 2 or 3
5. exp Fecal incontinence/
6. exp Urinary incontinence/
7. [(anal or fecal or urin\*) adj incontinence].m\_titl.
8. 5 or 6 or 7
9. 4 and 8
10. randomized controlled trial.pt.
11. controlled clinical trial.pt.
12. randomized.ab.
13. placebo.ab.

14. clinical trial.sh.
15. randomly.ab.
16. trial.ti.
17. 10 or 11 or 12 or 13 or 14 or 15 or 16
18. humans.sh.
19. 17 and 18
20. exp epidemiology/or exp epidemiologic studies/or exp case–control studies/or exp cohort studies/or exp longitudinal studies/or exp retrospective studies/or exp cross-sectional studies/or intervention studies/
21. (epidemiolog\* or incidence or prevalence or case–control or cohort\* longitudinal or retrospectiv\* cross-sectional or intervention).mp.
22. 20 or 21
23. 19 or 22
24. 9 and 23

Embase (Ovid) 1/1/2019

#### Search history

1. exp cesarean section/
2. exp delivery/
3. (((cesarean or caesarean) adj section) or delivery or c? section or c-section).m\_titl.
4. 1 or 2 or 3
5. exp feces incontinence/
6. exp urine incontinence/
7. ((anal or fecal or urin\*) adj incontinence).m\_titl.
8. 5 or 6 or 7
9. 4 and 8
10. randomized controlled trial/
11. randomization/

12. controlled study/
13. multicenter study/
14. phase 3 clinical trial/
15. phase 4 clinical trial/
16. double blind procedure/
17. single blind procedure/
18. ((singl\* or doubl\* or trebl\* or tripl\*) adj (blind\* or mask\*)).ti,ab.
19. (random\* or cross\* over\* or factorial\* or placebo\* or volunteer\*).ti,ab
20. 15 or 12 or 16 or 18 or 11 or 17 or 13 or 10 or 19 or 14
21. "human\*".ti,ab.
22. (animal\* or nonhuman\*).ti,ab.
23. 22 and 21
24. 22 not 23
25. 20 not 24
26. exp epidemiology/
27. (epidemiolog\* or incidence or prevalence or case-control or cohort\* longitudinal or retrospectiv\* cross-sectional or intervention).mp.
28. 26 or 27
29. 25 or 28
30. 9 and 29

### Cochrane library 1.1.19

#1 MeSH descriptor Cesarean Section explode all trees #2 MeSH descriptor Delivery, Obstetric explode all trees #3 (((cesarean or caecarean) near section) or delivery or c? section or c-section):ti #4 (#1 OR #2 OR #3) #5 MeSH descriptor Fecal Incontinence explode all trees #6 MeSH descriptor Urinary Incontinence explode all trees #7 ((anal or fecal or urin\*) near incontinence):ti #8 (#5 OR #6 OR #7) #9 (#4 AND #8)

### References

1. Oberwalder M, Connor J, Wexner SD (2003) Meta-analysis to determine the incidence of obstetric anal sphincter damage. *Br J Surg* 90:1333–1337
2. Nelson RL (2004) Epidemiology of fecal incontinence. *Gastroenterol* 126:s3–s7
3. Nelson RL, Furner S, Jesudason V (1998) Fecal incontinence in Wisconsin nursing homes: prevalence and Associations. *Dis Colon Rectum* 41:1226–1229
4. Higgins JPT, Green S (editors) (2008) *Cochrane Handbook for Systematic Reviews of Interventions Version 5.0.0* [www.cochrane-handbook.org](http://www.cochrane-handbook.org) The Cochrane Collaboration
5. Kepenekci I, Keskinkilic B, Akinsu F, Cakir P, Elhan AH, Erkek AB, Kuzu MA (2011) Prevalence of pelvic floor disorders in the female population and the impact of age, mode of delivery and parity. *Dis Colon Rectum* 54:85–94
6. Jangö H, Langhoff-Roos J, Rosthøj S, Sakse A (2016) Mode of delivery after obstetric anal sphincter injury and the risk of long-term anal incontinence. *Am J Obstet Gynecol* 214(6):733
7. Schünemann H, Brożek J, Guyatt G, Oxman A (eds) (2013) *The GRADE Working Group. GRADE Handbook for Grading Quality of Evidence and Strength of Recommendations*. Available from <https://gdt.guidelinedevelopment.org/app/handbook/handbook.html>. Updated October 2013
8. Blomquist JL, Muñoz A, Carroll M, Handa VL (2018) Association of delivery mode with pelvic floor disorders after childbirth. *JAMA* 320(23):2438–2447
9. Dolan LM, Hilton P (2010) Obstetric risk factors and pelvic floor dysfunction 20 years after first delivery. *Int Urogynecol J* 21(5):535–544
10. Fritel X, Schaal JP, Fauconnier A, Bertrand V et al (2008) Pelvic floor disorders 4 years after first delivery: a comparative study of restrictive versus systematic episiotomy. *BJOG* 115(2):247–252
11. Gyhagen M, Bullarbo M, Nielsen TF, Milsom I (2018) Faecal incontinence 20 years after one birth: a comparison between vaginal delivery and caesarean section. *Int Urogynecol J* 25(10):1411–1418
12. Handa VL, Blomquist JL, Knoepp LR, Hoskey KA, McDermott KC, Munoz A (2011) Pelvic floor disorders 5–10 years after vaginal or cesarean childbirth. *Obstet Gynecol* 118(4):777–784
13. Lucasz ES, Lawrence JM, Contreras R, Nager CW, Lubner KM (2006) Parity, mode of delivery and pelvic floor disorders. *Obstet Gynecol* 107:1253–1260
14. Schei B, Johannessen HH, Rydning A, Sultan A, Mørkved S (2019) Anal incontinence after vaginal delivery or cesarean section. *Acta Obstet Gynecol Scand* 98(1):51–60
15. Abramov Y, Sand PK, Botros SM, Goldberg RP et al (2005) Risk factors for female anal incontinence: new insight through the Evanston-Northwestern twin sisters study. *Obstet Gynecol* 106:726–732
16. Altman D, Ekstrom A, Frogren C, Nordenstam J, Zetterstrom J (2007) Symptoms of anal and urinary incontinence following cesarean section or spontaneous vaginal delivery. *Am J Obstet Gynecol* 197:512.e1–e7
17. Brown SJ, Gartland D, Donath S, MacArthur C (2012) Fecal incontinence during the first 12 months postpartum: complex causal pathways and implications for clinical practice. *Obstet Gynecol* 119(2 Pt 1):240–249
18. Burgio KL, Borello-France D, Richter HE, Fitzgeralds MP et al (2007) Risk factors for fecal and urinary incontinence after childbirth: the childbirth and pelvic symptoms study. *Am J Gastro* 102:1998–2004
19. Gartland D, MacArthur C, Woolhouse H, McDonald E, Brown SJ (2016) Frequency, severity and risk factors for urinary and faecal incontinence at 4 years postpartum: a prospective cohort. *BJOG Int J Obstet Gynaecol* 123(7):1203–1211
20. Goldberg RP, Kwon C, Gandhi S, Atkuru LV et al (2003) Prevalence of anal incontinence among mothers of multiples and analysis of risk factors. *Am J Obstet Gynecol* 189:1627–1631
21. Hutton EK, Hannah ME, Willan AR, Ross S, Allen AC, Armson BA et al (2018) Urinary stress incontinence and other maternal outcomes two years after Caesarean or vaginal birth for twin pregnancy: a multicentre randomized trial. *BJOG* 125(13):1682–1690
22. MacArthur C, Glazener CM, Wilson PD, Herbison GP, Gee H, Lang GD et al (2001) Obstetric practice and faecal incontinence three months after delivery. *BJOG* 108(7):678–683
23. MacArthur C, Glazener CMA, Lancashire R, Herbison P et al (2005) Faecal incontinence and mode of first and subsequent delivery: a six-year longitudinal study. *BJOG* 112:1075–1082
24. MacArthur C, Glazener C, Lancashire R, Herbison P, Wilson D (2011) Exclusive caesarean section delivery and subsequent

- urinary and faecal incontinence: a 12-year longitudinal study. *BJOG* 118(8):1001–1007
25. MacLennan AH, Taylor AW, Wilson DH, Wilson D (2000) The prevalence of pelvic floor disorders and their relationship to gender, age, parity and mode of delivery. *J Obstet Gynecol* 107:1460–1470
  26. McKinnie V, Swift S, Wang W, Woodman P, O'Boyle A, Kahn M, Valley M, Bland D, Shaffer J (2005) The effect of pregnancy and mode of delivery on the prevalence of urinary and fecal incontinence. *Am J Obstet Gynecol* 103(2):512–518
  27. Melville JL, Fan M-Y, Newton K, Fenner D (2005) Fecal incontinence in US women: a population-based study. *Obstet Gynecol* 193:2071–2076
  28. Rommen K, Schei B, Rydning A, Daltveit AK, Sultan A, Morkved S (2011) Prevalence of fecal incontinence after vaginal deliveries versus caesarean sections. *Female Pelvic Med Reconstr Surg* 17(5):767–768
  29. Varma MG, Brown JS, Creasman JM, Thom DH, van den Eeden SK, Beattie MS, Subak LL (2006) Fecal incontinence in females older than 40 years: who is at risk? *Dis Colon Rectum* 49(6):841–851
  30. Volloyhaug I, Morkved S, Salvesen O, Salvesen K (2015) Pelvic organ prolapse and incontinence 15–23 years after first delivery: a cross-sectional study. *BJOG* 122(7):964–971
  31. Woolhouse H, Perlen S, Gartland D, Brown SJ (2012) Physical health and recovery in the first 18 months postpartum: does cesarean section reduce long-term morbidity? *Birth (Berkeley, Calif.)* 39(3):221–229
  32. Hannah ME, Hannah WJ, Hodnett ED et al (2002) Outcomes at 3 months after planned Cesarean vs. planned vaginal delivery for breech presentation at term. *JAMA* 287:1822–1831
  33. Hannah ME, Whyte H, Hannah WJ, Hewson S, Amankwah K, Cheng M, Gafni A et al (2004) Maternal outcomes at 2 years after planned cesarean section versus planned vaginal birth for breech presentation at term: the international randomized term breech trial. *Am J Obstet Gynecol* 191:917–927
  34. Johannessen HH, Wibe A, Stordahl A, Sandvik L, Backe B, Morkved S (2019) Prevalence and predictors of anal incontinence during pregnancy and 1 year after delivery: a prospective cohort study. *Neurol Urodyn* 38(1):310–319
  35. Goffeng AR, Andersch B, Andersson M, Berndtsson I, Hulten L, Oreslan T (1998) Objective methods cannot predict anal incontinence after primary repair of extensive anal tears. *Acta Obstet Gynecol Scand* 77:439–443
  36. Guttierz AB, Madoff RD, Lowry AC, Parker SC, Buie WD, Baxter NN (2004) Long term results of anterior sphincteroplasty. *Dis Colon Rectum* 47:727–732
  37. Halverson AN, Hull TL (2002) Long-term outcome of overlapping anal sphincter repair. *Dis Colon Rectum* 45:345–348
  38. Karoui S, Leroi AM, Koning E, Menar JF, Michot F, Dens P (2000) Results of sphincteroplasty in 86 patients with anal incontinence. *Dis Colon Rectum* 43:813–820
  39. Malouf AJ, Norton CS, Engel AF, Nicholls RJ, Kamm MA (2000) Long-term results of overlapping anterior anal sphincter repair for obstetrical trauma. *Lancet* 355:260–265
  40. Pinta T, Kyaanpaa ML, Salmi T, Jaarvinen HJ, Luukkonen P (2003) Delayed sphincter repair for obstetric ruptures: analysis of failure. *Colorectal Dis* 5:73–78
  41. Rothbart J, Bemelman WA, Meijerink WJ (2000) Long term results of anterior anal sphincter repair for rectal incontinence due to obstetric injury. *Dig Surg* 17:390–394
  42. Vaizey CJ, Norton C, Thornton MJ, Nicholls RJ, Kamm MA (2004) Long term results of repeat anterior anal sphincter repair. *Dis Colon Rectum* 47:858–863
  43. Amu O, Rajendran S, Bolaji II (1998) Should doctors perform an elective cesarean section on request? maternal choice alone should not determine method of delivery. *BMJ* 317:463–465
  44. Abramowitz I, Sobhani I, Ganansia R (2000) Are sphincter defects the cause of anal incontinence after vaginal delivery? results of a prospective survey. *Dis Colon Rectum* 43:590–596
  45. Farrell SA (2002) Cesarean section versus forceps-assisted vaginal birth: it's time to include pelvic injury in the risk benefit equation. *Can Med Assoc J* 155:117–118
  46. Faridi FA, Willis S, Schelzig P, Siggelkow W, Schumpelick V, Rath W (2002) Anal sphincter injury during vaginal delivery—an argument for cesarean section on request? *Perinat. Med* 30:379–387
  47. Guise JM, McDonough MS, Osterwell P, Nygren P, Chan KS, Helfand M (2004) Systematic review of the incidence and consequences of uterine rupture in women with previous cesarean section. *BMJ* 329:1–7
  48. Idama TO, Lindow SW (1999) Safest option is still to aim for vaginal delivery. *BMJ* 318:121
  49. Zetterstrom J, Lopez A, Anzen B, Dolk A, Norman M, Mellgren A (1999) Anal incontinence after vaginal delivery: a prospective study in primiparous women. *Obstet Gynecol* 106:324–330
  50. Nygaard IE, Cruickshank FA (2003) Should all women be offered elective cesarean section. *Obstet Gynecol* 102:217–219
  51. Paterson-Brown S (1998) Should doctors perform an elective cesarean section on request? yes, as long as the woman is fully informed. *BMJ* 317:462–463
  52. Lockwood CJ (2004) Cesarean delivery: is it time to embrace elective procedures? *Contemp Obstet Gynecol* 12:8–10
  53. Minkoff H, Chervenak FA (2003) Elective primary cesarean delivery. *N Eng J Med* 348:946–950
  54. NIH Consensus Conference. Cesarean Delivery on Maternal Request (2006) NIH consensus and state of the science statements; [http://consensus.nih.gov/2006/CesareanStatement\\_Final\\_053106.pdf](http://consensus.nih.gov/2006/CesareanStatement_Final_053106.pdf) 23(1):1–29
  55. Declercq E, Menacker F, MacDorman M (2005) Rise on “no indicated risk” primary caesareans in the United States, 1991–2001: cross sectional analysis. *BMJ* 330:71–73
  56. van Roosmalen J (1999) Unnecessary caesarean sections should be avoided. *BMJ* 318:121
  57. Betrán AP, Ye J, Moller AB, Zhang J, Gülmezoglu AM, Torloni MR (2016) The increasing trend in caesarean section rates: global, regional and national estimates: 1990–2014. *PLoS One* 11(2):e0148343
  58. Hsu KH, Liao PJ, Hwang CJ (2008) Factors affecting Taiwanese women's choice of Cesarean section. *Soc Sci Med* 66(1):201–209
  59. Behague DP, Victoria CG, Barros FC (2002) Consumer demand for caesarean sections in Brazil; informed decision making, patient choice or social inequality? a population based birth cohort study linking ethnographic and epidemiological methods. *BMJ* 321:1–6
  60. Potter JE, Berquo E, Perpetuo IHO, Leal OF, Hopkins K, Souza MR, de Carvalho Formiga MC (2001) Unwanted caesarean sections among public and private patients in Brazil: prospective study. *BMJ* 323:1155–1158
  61. Murray S (2000) Relation between private health insurance and high rates of caesarean section in Chile: qualitative and quantitative study. *BMJ* 321:1501–1505
  62. Keag OE, Norman JE, Stock SJ (2018) Long-term risks and benefits associated with cesarean delivery for mother, baby, and subsequent pregnancies: systematic review and meta-analysis. *PLoS Med* 15(1):e1002494
  63. Sandall J, Tribe RM, Avery L, Mola G, Visser GH, Homer CS, Gibbons D, Kelly NM, Kennedy HP, Kidanto H, Taylor P, Temmerman M (2018) Short-term and long-term effects of

- caesarean section on the health of women and children. *Lancet* 392(10155):1349–1357
64. The Childbirth Connection. The cost of Having a Baby. <http://transform.childbirthconnection.org/reports/cost/>
  65. Nelson RL, Furner SE, Westercamp M, Farquhar C (2010) Cesarean delivery for the prevention of anal incontinence. *Cochrane Database Syst Rev* 2:CD006756
  66. Pretlove SJ, Thompson PJ, Toozs-Hobson PM, Radley S, Khan KS (2008) Does the mode of delivery predispose women to anal incontinence in the first year postpartum? A comparative systematic review. *BJOG* 115(4):421–434
  67. Rørtveit G, Hannestad YS (2014) Association between mode of delivery and pelvic floor dysfunction. *Tidsskr Nor Laegeforen* 134(19):1848–1852
  68. Tähtinen RM, Cartwright R, Tsui JF, Aaltonen RL, Aoki Y, Cárdenas JL, El Dib R, Joronen KM, Al Juaid S, Kalantan S, Kochana M, Kopec M, Lopes LC, Mirza E, Oksjoki SM, Pesonen JS, Valpas A, Wang L, Zhang Y, Heels-Ansdell D, Guyatt GH, Tikkinen KAO (2016) Long-term impact of mode of delivery on stress urinary incontinence and urgency urinary incontinence: a systematic review and meta-analysis. *Eur Urol* 70(1):148–158
  69. Shorten A (2007) Maternal and neonatal effects of caesarean section; more accurate estimates of benefits and harms are needed to support informed childbirth choices. *BMJ* 335:1003–1004
  70. Tilbrook H (2009) Patients' preferences within randomized trial: systematic review and patient level meta-analysis. *BMJ* 338:85–88
  71. Turner CE, Young JM, Solomon MJ, Ludlow Bennes C, Phipps H (2008) Willingness of pregnant women and clinicians to participate in a hypothetical randomized controlled trial comparing vaginal delivery and elective caesarean section. *Aust New Zeal J Obstet Gynecol* 48:542–546
  72. Lavender T, Kingdon C, Hart A, Gyte G, Gabbay M, Neilson JP (2005) Could a randomized trial answer the controversy relating to elective caesarean section? National survey of consultant obstetricians and heads of midwifery. *BMJ* 331:490–491
  73. Kotaska A (2004) Inappropriate use of randomized trials to evaluate complex phenomena: a case study. *BMJ* 329:1039–1042
  74. Alfirevic Z, Milan SJ, Livio S (2013) Caesarean section versus vaginal delivery for preterm birth in singletons. *Cochrane Database Syst Rev*. <https://doi.org/10.1002/14651858.CD000078.pub3>
  75. Whitehead WE, Borrud L, Goode PS, Meikle S, Mueller ER, Tuteja A, Weidner A, Weinstein M, Ye W (2009) Pelvic floor disorders network. Fecal incontinence in US adults: epidemiology and risk factors. *Gastroenterology* 137(2):512–517

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