# RESEARCH



# Child and maternal benefits and risks of caseload midwifery – a systematic review and meta-analysis



# Abstract

**Background** It has been reported that caseload midwifery, which implies continuity of midwifery care during pregnancy, childbirth, and the postnatal period, improves the outcomes for the mother and child. The aim of this study was to review benefits and risks of caseload midwifery, compared with standard care comparable to the Swedish setting where the same midwife usually provides antenatal care and the checkup postnatally, but does not assist during birth and the first week postpartum.

**Methods** Medline, Embase, Cinahl, and the Cochrane Library were searched (Nov 4th, 2021) for randomized controlled trials (RCTs). Retrieved articles were assessed and pooled risk ratios calculated when possible, using random-effects meta-analyses. Certainty of evidence was assessed according to GRADE.

**Results** In all, 7,594 patients in eight RCTs were included, whereof five RCTs without major risk of bias, including 5,583 patients, formed the basis for the conclusions. There was moderate certainty of evidence for little or no difference regarding the risk of Apgar ≤ 7 at 5 min, instrumental birth, and preterm birth. There was low certainty of evidence for little or no difference regarding the risk of perinatal mortality, neonatal intensive care, perineal tear, bleeding, and acute caesarean section. Caseload midwifery may reduce the overall risk of caesarean section. Regarding breastfeeding after hospital discharge, maternal mortality, maternal morbidity, health-related quality of life, postpartum depression, health care experience/satisfaction and confidence, available studies did not allow conclusions (very low certainty of evidence). For severe child morbidity and Apgar ≤ 4 at 5 min, there was no literature available.

**Conclusions** When caseload midwifery was compared with models of care that resembles the Swedish one, little or no difference was found for several critical and important child and maternal outcomes with low-moderate certainty of evidence, but the risk of caesarean section may be reduced. For several outcomes, including critical and important ones, studies were lacking, or the certainty of evidence was very low. RCTs in relevant settings are therefore required.

Keywords Caseload midwifery, GRADE, Meta-analysis, Care model, Systematic review

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# Introduction

Midwife-led continuity models, compared with other models of care for childbearing women, are reportedly favorable regarding several mother and child outcomes, including a reduced risk of instrumental vaginal birth, a reduced risk of preterm birth (<37 weeks), and a reduced risk of fetal loss before and after 24 weeks including neonatal death [1]. Midwife-led continuity models include caseload midwifery, a model in which a primary midwife, within a team of midwives provides care to a load of cases during antenatal, intrapartum, and postpartum care. Caseload midwifery has been established in several countries, including Denmark [2], the Netherlands [3], England [4], and Australia. [5].

In Sweden, about 115,000 children are born each year. In standard care, midwives provide antenatal, intrapartum and postpartum care. When complications arise, other professionals, e.g. physicians, are consulted. When possible, the same midwife provides care during pregnancy and postnatal follow-up after the first week, whereas hospital-employed midwives provide care during childbirth and the first week postpartum. Almost all childbirths in Sweden take place in hospitals, and midwives assist the women during vaginal birth. According to the Swedish Pregnancy Register [6], the prevalence of caesarean section was 18.5% in 2021, and perinatal death 0.35%.

As there is a shortage of midwives in Swedish labor wards, caseload midwifery has gained increased interest as an alternative model of care. Indeed, an integrative literature review revealed several factors contributing to job satisfaction for midwives in caseload models of care, including the ability to build relationships with women, the flexibility and control, as well as the professional autonomy and identity [7]. The midwife-woman relationship has also been explored to be of value for the woman, including themes of personalized care, trust, and empowerment [8]. As far as we are aware, however, the scientific literature regarding effects for mother and child in the caseload model of care, compared with models comparable to Swedish standard care, has not previously been reviewed. Therefore, this systematic review was performed to evaluate benefits and risks, for the mother and the child, of caseload midwifery compared with other models of care where the same midwife generally provides ante- and postnatal care but does not assist during childbirth and the first week postpartum.

# Methods

This systematic review was performed according to the established routines at the regional health technology assessment (HTA) center (HTA-centrum) in Region Västra Götaland, Sweden, and reported according to the PRISMA guidelines [9]. The aim was defined in a PICO (Participants, Intervention, Comparison, Outcome). Participants (P) were pregnant women, without planned home birth, and their child/ren. The intervention (I) was caseload midwifery. For scheduling reasons, we considered it reasonable that caseload midwifery could include quite large caseload teams. Therefore, we did not exclude studies based on the size of the team. The comparison (C) was standard care similar to the Swedish model, i.e. with one maternal care midwife during antenatal and postnatal care, and hospital care midwives during birth and the week afterwards. As we did not expect to identify studies exactly matching the Swedish model, we decided to include studies in which midwives performed a considerable part of the antenatal care. When not clearly reported, we decided to include rather than to exclude, and to handle this uncertainty in the directness assessments. Outcomes (O) included child outcomes: perinatal mortality, severe morbidity (e.g. brain injuries, body injuries, or severe infection), Apgar  $\leq 4$  at 5 min, Apgar  $\leq 7$ at 5 min, neonatal intensive care, and breastfeeding after discharge, as well as maternal outcomes: mortality, intensive care, health-related quality of life (HRQL), perineal tear (grade I-IV and III-IV), bleeding, caesarean section (total and acute), instrumental birth, postpartum depression, preterm birth, and health care experience/satisfaction/confidence. Perinatal mortality and morbidity, as well as maternal mortality and intensive care were considered critical for decision-making. At the other end, breastfeeding and health care experience/satisfaction/ confidence were considered useful, and the remaining outcomes important for decision-making. Regarding preterm birth, this outcome may not be directly affected by adding continuity of carer during intrapartum care when ante- and postnatal care is already performed by the same midwife. However, as this outcome was included in a previous systematic review within the field [1], and as caseload models may have indirect effects, it was considered an important outcome. Publications were restricted to randomized controlled trials (RCTs) and languages to English, Swedish, Danish, and Norwegian. The preparatory work for this review was performed within an HTA [10].

#### Literature search and study selection

On November 4th, 2021, two medical librarians performed systematic searches in Medline, Embase, Cinahl, and the Cochrane Library. Reference lists of relevant articles were scrutinized for additional references. To identify ongoing studies, we performed a search in Clinicaltrials.gov and WHO International Clinical Trials Registry Platform (ICTRP, March 28th, 2022). Search strategies are provided in Additional file 1.

Two authors screened identified abstracts and those that clearly did not meet the PICO criteria were excluded

in a consensus discussion. When there were uncertainties regarding inclusion/exclusion, the full text was retrieved and assessed independently by at least two authors. Inclusion/exclusion according to the PICO was then decided in a consensus meeting. As rostered midwives may provide ante- and postnatal care in Sweden, although continuity of carer is intended, we included studies where the circumstances in the control group in consensus discussions were considered sufficiently similar to the Swedish model, and, as described previously, handled potential uncertainties in the assessments of directness. For articles excluded in consensus, after fulltext reading, reasons for exclusion were recorded. The remaining studies were included in the systematic review.

#### Data extraction and study assessments

Data were extracted from the studies by two authors independently and were subsequently checked by the other authors. Data extraction included the participants studied, the number and characteristics of individuals in the intervention and control groups, and the results regarding the outcomes selected in the PICO.

Each study was critically and independently appraised by at least two authors, focusing on the domains directness and risk of bias according to the checklist for assessing RCTs used by HTA-centrum [11]. Regarding directness, we assessed to what extent the studied population, intervention, comparison, and outcome measures corresponded to the question at issue. Regarding risk of bias, we focused on selection bias (random sequence generation and concealed allocation, respectively), performance bias, detection bias, attrition bias, reporting bias, and other bias. Subsequently, all authors discussed the assessments and categorized, in consensus, each study overall as having no or minor problems (+), some problems (?), or major problems (-) in the domains directness and risk of bias. The certainty of evidence was assessed using the Grading of Recommendations Assessment, Development and Evaluation (GRADE) in which potential issues regarding risk of bias (study limitations), consistency, directness, and precision are considered across studies, to appraise the overall quality of evidence [12].

#### Statistics

When three or more RCTs provided data regarding a specific outcome, we performed random-effects metaanalyses using the software Review Manager (Rev-Man) version 5.4.1 (The Nordic Cochrane Centre, The Cochrane Collaboration, Copenhagen, Denmark) to obtain risk ratios with 95% confidence intervals (CIs). When merely two RCTs provided data regarding a specific outcome, we assessed if these were sufficiently clinically homogenous to allow pooling. We consistently used the number of randomized individuals as denominator. When the pooled result was statistically significant, i.e., the 95% CI of the risk ratio did not cover 1, we calculated the risk difference with 95% CI to gain knowledge about the magnitude of the effect. According to the predefined analysis plan, RCTs without major risk of bias were compiled and formed the basis for the conclusions. Beforehand, we also planned subgroup meta-analyses including pregnant women with fear of childbirth; crude analyses in a small non-randomized study suggest that women with fear may benefit from a continuity model of care [13]. When relevant, we also performed sensitivity meta-analyses to investigate the robustness of the results. Heterogeneity was assessed with I<sup>2</sup>.

# Results

After removal of duplicates, the literature search identified 2,575 unique publications, and 12 publications, based on eight RCTs, were included in this systematic review (Fig. 1) [14–25]. Publications excluded after fulltext reading by the authors, as well as the reasons for excluding them, are presented in Supplemental Table 1.

#### **Study characteristics**

In all, seven RCTs with individual randomization [14, 17, 19–21, 23, 25] and one RCT with cluster-randomization [24] fulfilled the PICO of this review, all non-blinded and including a total of 7,594 women, (Table 1). Four RCTs were performed in Australia [17, 19, 21, 25], three in England [14, 20, 24], and one in New Zealand [23]. Three RCTs were assessed to have major risk of bias [19, 23, 24], and the remaining five, with associated publications, formed the basis for the conclusions [14, 17, 20, 21, 25]. Reasons underlying the directness and risk of bias assessments are described in Supplemental Table 2.

#### **Child outcomes**

Results for each RCT presenting data for the studied outcomes are presented in Table 2, and forest plots in Fig. 2. Regarding perinatal mortality, four RCTs without major risk of bias, including 5,465 women, were pooled resulting in a risk ratio of 0.93 (95% CI: 0.41 to 2.08). No data were available regarding severe morbidity and Apgar  $\leq 4$ at 5 min. Regarding Apgar ≤7 at 5 min, two RCTs without major risk of bias contributed data. They were not pooled as they were considered clinically heterogeneous; one study included women at increased risk of preterm birth [14], and the other women of any risk without planned caesarean section [25]. As one additional study reported Apgar <7 instead of  $\leq$ 7 at 5 min [21], we performed a sensitivity meta-analysis of the three trials, resulting in a risk ratio of 0.92 (95% CI: 0.62 to 1.30; I<sup>2</sup>: 0%). Regarding neonatal intensive care, two RCTs provided unpoolable data [14, 21], none reporting statistically significant



Fig. 1 PRISMA flowchart

differences and none separating routine surveillance from care due to child health issues.

One RCT reported breastfeeding six weeks and six months after discharge [25], with no significant differences between the groups. There were many dropouts in the intervention and comparison groups, 35% versus 50% at six weeks, and 37% versus 55% at six months respectively. Pooling results from two RCTs without major risk of bias regarding breastfeeding at discharge [14, 25] resulted in a risk ratio of 1.11 (95% CI: 0.94 to 1.31; I<sup>2</sup>: 71%).

Reasons for downgrading in the GRADE process are described in Supplemental Table 3. Comparing caseload midwifery with standard care, there may be little or no difference in perinatal mortality ( $\oplus \oplus OO$ ). Concerning child morbidity, no data were available regarding the risk of severe morbidity and Apgar  $\leq 4$  at 5 min. Further, there was probably little or no difference regarding the risk of Apgar  $\leq 7$  at 5 min ( $\oplus \oplus \oplus O$ ) and there may be little or no difference in the risk of admission to neonatal intensive care ( $\oplus \oplus OO$ ). Regarding breastfeeding after discharge,

the certainty of evidence was assessed as very low, not allowing conclusions ( $\oplus OOO$ ).

### Maternal outcomes

Results for each study presenting data for the studied maternal outcomes are presented in Table 2, and forest plots in Fig. 3. Regarding mortality, five RCTs reported that no deaths occurred [14, 19, 21, 24, 25]. Regarding intensive care, one event, due to sickle cell crisis, was reported in one RCT [14]. HRQL was reported in one RCT, with no significant differences between the comparison groups [14].

Regarding perineal tears grade I-IV, two RCTs without major risk of bias reported results. They were not pooled as they were considered clinically heterogeneous. In one RCT [25], but not the other [14], a statistically significant difference favoring standard care was found. Regarding grade III-IV perineal tears, three RCTs without major risk of bias were pooled resulting in a risk ratio of 1.13 (95% CI: 0.81 to 1.59). In a sensitivity meta-analysis restricted to women with vaginal birth, the risk ratio was 1.08 (95% CI: 0.77 to 1.51; I<sup>2</sup>: 0%). Episiotomy was

First author Year	Country	Participants		Caseload midwifery	Standard care	
	(Acronym)	Characteristics	l vs. C	-		
			n			
			age			
Fernandez Turienzo	England	Women at increased	169 vs. 165	One team	Rostering midwife, no	
2020 [14]	(POPPIE)	risk of preterm birth	32 vs. 32 yrs.	Six midwives	planned continuity	
Fernandez Turienzo 2021 [15]				35 women/midwife/year		
Forster 2016 [16]	See McLachlar	2012 ו				
Homer 2001 [17]	Australia	Women without	550 vs. 539	One team	Hospital-based antenatal	
Homer 2002 [18]	(STOMP)	significant disease	28 vs. 28 yrs.	Six midwives	care, midwives and/or	
				50 women/midwife/year	obstetricians/GPs	
Homer 2021 [19]	Australia	Women with one	108 vs. 110	NR	Midwife team antenatally,	
		previous CS, low-risk	31 vs. 31 yrs.		rostering midwives at	
		pregnancy			birth	
Marks 2003 [20]	England	Women with $\geq 1$	44 vs. 43	One team	Midwives or GPs	
		episode of major	32 vs. 32 yrs.	Six midwives		
		depressive disorder		Number of women/midwite/year NR		
McLachlan 2012 [21]	Australia	Women with low-	1156 vs. 1158	One team	Antenatally: midwives	
McLachlan 2016 [22]	(COSMOS)	risk pregnancy	32 vs. 32 yrs.	12 midwives	(/8%), GPs (15%), obstet-	
				45 women/midwife/year	ric trainee (2%), other (5%)	
Morrison 2002 [23]	New Zealand	Women with	140 vs. 144	One team	Antenatally: One of two	
		diabetes	33 vs. 32 yrs.	Three midwives	dedicated diabetic clinic	
				Number of women/midwife/year NR	midwives	
North Staffordshire	England	No exclusions <sup>1</sup>	770 vs. 735	Three caseload areas, 26 midwives	Shared care: GP/midwife	
2000 [24]			28 vs. 28 yrs.	Two-three midwives/team		
				35–40 women/midwife/year		
Tracy 2013 [25]	Australia	Women of any risk,	871 vs. 877	Number of teams NR	Shared care: GP/midwife	
	(M@NGO)	without a planned	32 vs. 32 yrs.	Four midwives/team		
		elective CS		40 women/midwife/year		

# Table 1 Characteristics of included RCTs

<sup>1</sup>cluster-randomized

C=comparison (standard care), COSMOS=COmparing Standard Maternity care with One-to-one midwifery Support, CS=caesarean section, I=intervention (caseload midwifery), GP=general practitioner, M@NGO=Midwives @ New Group practice Options, NR=not reported, POPPIE=Pilot study Of midwifery Practice in Preterm birth Including women's Experiences, RCT=randomized controlled trial, STOMP=St George Outreach Maternity Project

reported in four RCTs, with frequencies in standard care varying between 9% and 29% [14, 21, 24, 25]. Regarding bleeding>1000 ml, three RCTs without major risk of bias [14, 21, 25] could be pooled resulting in a risk ratio of 0.76 (95% CI: 0.58 to 1.01). No RCT reported bleedings requiring transfusion.

Caesarean section was reported in seven RCTs, four of which without major risk of bias [14, 17, 21, 25]. Pooling caesarean section overall, the risk ratio was 0.84 (95% CI: 0.75 to 0.94), with absolute risks in standard care varying between 18% and 31% and a pooled risk difference of -4.0 (95% CI: -6.1 to -1.9) percentage units. Three RCTs without major risk of bias reported acute caesarean sections [17, 21, 25], with a pooled risk ratio of 0.86 (95% CI: 0.70 to 1.06).

Instrumental birth was reported in seven RCTs, four of which without major risk of bias [14, 17, 21, 25]. Pooling these RCTs resulted in a risk ratio of 0.98 (95% CI: 0.87 to 1.10). Postpartum depression was reported in one small RCT, with similar event rates in the comparison groups [20]. Preterm birth was reported in four RCTs, three of

which without major risk of bias [14, 21, 25]. Pooling these RCTs resulted in a risk ratio of 1.01 (95% CI: 0.69 to 1.50).

Parent experience/satisfaction was reported in four publications [15, 16, 22, 23], based on three RCTs [14, 21, 23]. All four publications were assessed to have major risk of bias. Experience/satisfaction did not differ between the groups in two RCTs [14, 23], but was reported to favor caseload midwifery in one [16]. Confidence, measured as trust in midwife, was reported in one RCT with major risk of bias, favouring the caseload model of care [15].

Reasons for downgrading the certainty of evidence in the GRADE process are described in Supplemental Table 3. The certainty of evidence regarding maternal mortality maternal intensive care, HRQL, and postpartum depression was assessed as very low, not allowing conclusions ( $\oplus$ OOO). There may be little or no difference regarding perineal tears (I-IV and III-IV, respectively) and bleedings (all  $\oplus \oplus$ OO). Caseload midwifery may decrease the risk of caesarean section overall, but there may be little or no difference regarding acute

First Child outcomes author I vs. C		Maternal outcomes I vs. C	Overall assessment <sup>1</sup>		
Year	n/n with available data (%)	n/n with available data (%)	Directness	Risk of bias	
Fernandez Turienzo 2020 [14]	Perinatal mortality: 0/168     (0) vs. 1/163 (0.6)     Apgar ≤ 7 at 5 min: 5/160     (3.1) vs. 7/159 (4.4)     NICU, mean days±SD:     7.1±14.7 vs. 1.1±3.4     Breastfeeding, initiation:     133/161 (81) vs. 118/158     (75); at discharge: 112/161     (70) vs. 89/158 (57)	<ul> <li>Mortality: 0/168 vs. 1/163</li> <li>Intensive care: 1/168 (0.6) vs. 0/163 (0)</li> <li>Perineal tear, I-IV: 36/162 (22) vs. 45/160 (28); III-IV: 2/162 (1.2) vs. 3/160 (1.9)</li> <li>Bleeding, &gt; 1000 ml: 2/168 (1.2) vs. 1/163 (0.6)</li> <li>CS, total: 51/162 (31) vs. 49/160 (31); acute NR</li> <li>Instrumental birth: 14/162 (9) vs. 12/160 (8)</li> <li>Preterm birth: 31/168 (18) vs. 19/163 (12)</li> <li>Feasibility, assisted at delivery by primary caseload midwife: 95/168 (57); a midwife in the caseload team: 136/168 (81)</li> </ul>	?	?	
Fernandez Turienzo 2021 [15]	-	<ul> <li>HRQL, physical health<sup>2</sup>: 50.43 vs. 15.80, P=0.36; mental health<sup>2</sup>: 14.65 vs. 14.76, P=0.85</li> <li>Health care experience/satisfaction, LAS<sup>3</sup>, mean ± SD: 52.12 ± 13.09 vs. 50.69 ± 13.13, NS</li> <li>Health care confidence, TNS<sup>4</sup>, mean ± SD: 28.89 ± 2.01 vs. 24.68 ± 5.68, MD (95% Cl): -4.21 (-5.44; -2.97)</li> </ul>	?	-	
Forster 2016 [16]		<ul> <li>Feasibility, assisted at delivery by primary caseload midwife: 573/981 (58); a midwife in the caseload team 889/981 (91)</li> <li>Health care experience/satisfaction, proportional OR (95% Cl)<sup>5,</sup> pregnancy: 3.35 (2.79; 4.03); labor/birth: 2.13 (1.78; 2.56); postnatally: 3.19 (2.64; 3.85)</li> </ul>	?	-	
Homer 2001 [17]	• Perinatal mortality: 4/550 (0.7) vs. 4/539 (0.7)	<ul> <li>Bleeding, non-specified postpartum hemorrhage: 31/550 (6) vs. 26/539 (5)</li> <li>CS, total: 73/550 (13) vs. 96/539 (18); acute: 52/550 (9) vs. 62/539 (12)</li> <li>Instrumental birth: 71/550 (13) vs. 63/539 (12)</li> </ul>	?/-	?	
Homer 2002 [18]	-	Feasibility, assisted at delivery by a midwife in the caseload team: 435/550 (79)	?	?	
Homer 2021 [19]	<ul> <li>Perinatal mortality: 0/108</li> <li>(0) vs. 0/110 (0)</li> <li>Breastfeeding at discharge: 123/134 (88) vs.</li> <li>122/138 (88)</li> </ul>	<ul> <li>Mortality: 0/108 vs. 0/110</li> <li>CS, total: 78/108 (72) vs. 74/110 (67), acute NR</li> <li>Instrumental birth: 12/108 (11) vs. 16/110 (15)</li> </ul>	?/-	-	
Marks 2003 [ <mark>20</mark> ]	-	Postpartum depression: 10/44 (23) vs. 10/43 (23) (any psychiatric illness)	-	?	
McLachlan 2012 [21]	<ul> <li>Perinatal mortality:</li> <li>4/1146 (0.3) vs. 4/1151</li> <li>(0.3)</li> <li>Apgar &lt; 7 at 5 min:</li> <li>15/1112 (1.4) vs. 20/1080</li> <li>(1.9)</li> <li>NICU: 15/1139 (1.3) vs.</li> <li>20/1137 (1.9)</li> </ul>	<ul> <li>Mortality: 0/1142 vs. 0/1144</li> <li>Perineal tear, I-IV: 343/696 (49) vs. 301/679 (44); III-IV: 26/693 (4) vs. 20/679 (3)</li> <li>Bleeding, &gt; 1000 ml: 53/1142 (5) vs. 65/1144 (6)</li> <li>CS, total: 221/1142 (19) vs. 285/1144 (25); acute: 186/1142 (16) vs. 245/1144 (21)</li> <li>Instrumental birth: 202/1142 (18) vs. 222/1144 (19)</li> <li>Preterm birth: 42/1111 (4) vs. 45/1086 (4)</li> <li>Feasibility, assisted at delivery by primary caseload midwife: 650/1142 (57); a midwife in the caseload team 1016/1142 (89)</li> </ul>	?	+	
McLachlan 2016 [22]	-	Health care experience/satisfaction <sup>6</sup> : 697/979 (71) vs. 516/824 (63)	?	-	
Morrison 2002 [23]	Perinatal mortality: 2/134     (1.5) vs. 0/138 (0)     Breastfeeding at dis- charge: 123/134 (92) vs. 122/138 (88)	<ul> <li>CS, total: 47/134 (35) vs. 49/138 (35); acute: 40/134 (30) vs. 38/138 (28)</li> <li>Instrumental birth: 20/134 (15) vs. 15/138 (11)</li> <li>Health care experience/satisfaction<sup>7</sup>: 111/126 (88) vs. 103/127 (81), NS</li> </ul>	-	-	

#### Table 2 (continued)

First author	Child outcomes I vs. C	Maternal outcomes I vs. C	Overall assessment <sup>1</sup>		
Year	n/n with available data (%)	n/n with available data (%)	Directness	Risk of bias	
North Staf- fordshire 2000 [24]	• Perinatal mortality: 6/770 (0.7) vs. 11/735 (1.5)	<ul> <li>Mortality: 0/770 vs. 0/735</li> <li>Perineal tear, I-IV: 248/770 (32) vs. 221/735 (30); III-IV NR</li> <li>CS, total: 137/770 (18) vs. 128/735 (17); acute: 62/770 (8) vs. 76/735 (10)</li> <li>Instrumental birth: 74/770 (10) vs. 84/735 (11)</li> <li>Feasibility, assisted at delivery by a midwife in the caseload team: 696/770 (95)</li> </ul>	-	-	
Tracy 2013 [25]	<ul> <li>Perinatal mortality: 3/871</li> <li>(0.3) vs. 3/877 (0.3)</li> <li>Apgar ≤ 7 at 5 min: 38/871 (4) vs. 36/877 (4)</li> <li>Breastfeeding at discharge: 776/871 (89) vs. 747/877 (85); after</li> <li>weeks: 509/567 (90) vs. 388/440 (88); after 6 months: 396/546 (73) vs. 279/398 (70)</li> </ul>	<ul> <li>Mortality: 0/871 vs. 0/877</li> <li>Perineal tear, I-IV: 343/696 (49) vs. 301/679 (44); III-IV: 26/693 (4) vs. 20/679 (3)</li> <li>Bleeding, &gt; 1000 ml: 28/820 (3) vs. 43/791 (5)</li> <li>CS, total: 183/871 (21) vs. 204/877 (23); acute: 114/871 (13) vs.</li> <li>110/877 (13)</li> <li>Instrumental birth: 172/871 (20) vs. 171/877 (19)</li> <li>Preterm birth: 39/871 (4) vs. 51/877 (6)</li> </ul>	?	?	

 $^{1}$  + = no or minor problems; ?=some problems; – = major problems; for directness and risk of bias issues, see Table S2

<sup>2</sup>PROMIS-10 instrument: mean: 50 for a United States general population, higher scores indicate better outcome

<sup>3</sup>LAS instrument: ten questions, experience of control during delivery, from 1: never or almost never, to 7: almost all of the time

<sup>4</sup>TNS instrument, adapted for midwives: five questions on confidence in midwife over pregnancy and delivery, from 1 = never, to 5 = always

<sup>5</sup>Response to one question "Overall, how would you describe your care in...", from 1 = very poor, to 7 = very good

<sup>6</sup>Response to one question "Overall experience of childbirth", from 1 = very negative, to 7 = very positive, proportion responding 6 or 7 presented

<sup>7</sup>Proportion "very satisfied with care"

C=comparison (standard care), CI=confidence interval, CS=caesarean section, HRQL=health-related quality of life, I=intervention (caseload midwifery), LAS=Labour Agentry Scale, MD=mean difference, NICU=neonatal intensive care unit, NR=not reported, NS=non-significant, OR=odds ratio, PROMIS=Patient reported outcomes measurement information system, RCT=randomized controlled trial, SD=standard deviation, TNS=Trust in Nurses Scale

#### A Perinatal mortality

Caseload midwifery		Standard care			<b>Risk Ratio</b>	Risk Ratio		
Study or Subgroup	Events	Total	Events	Total	Weight	M-H, Random, 95% CI	M-H, Random, 95% CI	
Fernandez Turienzo 2020 (POPPIE)	0	169	1	165	6.4%	0.33 [0.01, 7.93]		
Homer 2001 (STOMP)	4	550	4	539	34.1%	0.98 [0.25, 3.90]		
McLachlan 2012 (COSMOS)	4	1156	4	1158	34.0%	1.00 [0.25, 4.00]		
Tracy 2013 (M@NGO)	3	871	3	877	25.5%	1.01 [0.20, 4.97]	·	
Total (95% CI)		2746		2739	100.0%	0.93 [0.41, 2.08]	-	
Total events	11		12					
Heterogeneity: Tau <sup>2</sup> = 0.00; Chi <sup>2</sup> = 0.44, df = 3 (P = 0.93); l <sup>2</sup> = 09			6					100
Test for overall effect: Z = 0.19 (P = 0.85)							Favours caseload Favours standard care	900

## B Apgar ≤7 at 5 minutes

Caseload midwifery		Standard care		Risk Ratio		Risk	Ratio		
Study or Subgroup	Events	Total	Events	Total	M-H, Random, 95% Cl		M-H, Rand	om, 95% CI	
Fernandez Turienzo 2020 (POPPIE)	5	169	7	165	0.70 [0.23, 2.15]				
Tracy 2013 (M@NGO)	38	871	36	877	1.06 [0.68, 1.66]			+	
						0.2	0.5 Favours caseload	1 2 Favours stan	dard care

Fig. 2 Forest plots of RCTs reporting child outcomes, summarized in meta-analyses if not deemed clinically heterogeneous. A: perinatal mortality (absolute risk in standard care ranged from 0.3–0.7%). A: Apgar ≤ 7 at 5 min (absolute risk in standard care 4% in both studies)

# A Perineal tear grade I-IV

Study or Subgroup	Caseload m	nidwifery	Standar	d care	Weight	Risk Ratio	Risk Ratio
Fernandez Turienzo 2020 (POPPIE	) 36	169	events 45	165	weight	0.78 [0.53, 1.14]	m-n, kaldom, 95% Cl
Fracy 2013 (M@NGO)	343	871	301	877		1.15 [1.01, 1.30]	
							Favours caseload Favours standard care
B Perineal tear grade III-IV							
Study or Subgroup	Caseload m Events	nidwifery Total	Standar Events	d care Total	Weight	Risk Ratio M-H, Random, 95% Cl	Risk Ratio M-H, Random, 95% Cl
emandez Turienzo 2020 (POPPIE	2	169	3	165	3.7%	0.65 [0.11, 3.85]	· · · · · · · · · · · · · · · · · · ·
Fracy 2013 (M@NGO)	26	871	20	877	34.9%	1.31 [0.74, 2.33]	
Fotal (95% CI)		2196		2200	100.0%	1.13 [0.81, 1.59]	
Fotal events	69	2150	61	2200	100.070	1110 [0.01, 1.00]	
Heterogeneity: Tau" = 0.00; Chi" = 0 Test for overall effect: Z = 0.73 (P =	0.66, df = 2 (P = 0 0.47)	0.72); I <sup>e</sup> = 09	6				0.5 0.7 1 1.5 2 Favours caseload Favours standard care
<b>C</b> Bleeding >1000 ml							
Study or Subgroup	Caseload n	nidwifery Total	Standar	d care	Weight	Risk Ratio	Risk Ratio
Fernandez Turienzo 2020 (POPPIE	E) 2	168	1	165	1.4%	1.96 [0.18, 21.45]	
McLachlan 2012 (COSMOS)	53	1156	65	1158	62.7%	0.82 [0.57, 1.16]	
11acy 2013 (M@/460)	28	871	43	877	30.0%	0.00 [0.41, 1.05]	-
Total (95% CI) Total events	00	2195	100	2200	100.0%	0.76 [0.58, 1.01]	
Heterogeneity: Tau <sup>2</sup> = 0.00; Chi <sup>2</sup> =	83 1.15, df = 2 (P = 1	0.56); l² = 0*	109 %				
Test for overall effect: Z = 1.89 (P =	0.06)						0.2 0.5 1 2 Favours caseload Favours standard care
Caesarean section, total							
Study or Subgroup	Caseload m	idwifery Total	Standar	d care Total	Weight	Risk Difference M-H, Random, 95% Cl	Risk Difference M-H, Random, 95% Cl
ernandez Turienzo 2020 (POPPIE	) 51	169	49	165	4.7%	0.00 [-0.09, 0.10]	
Iomer 2001 (STOMP)	73	550	96	539	24.7%	-0.05 [-0.09, -0.00]	
racy 2013 (M@NGO)	183	871	285	877	40.4%	-0.05 [-0.09, -0.02]	
otal (05% CI)		3740		2720	100.05	0.0110.0	
otal (95% CI) Total events	528	2/40	634	2139	100.0%	-0.04 [-0.06, -0.02]	
E Caesarean section, acute	asoload midwit	ary Sta	ndard ca	70		Disk Patio	Risk Patio
Study or Subgroup	Events	Total Eve	ents T	otal We	eight M	H, Random, 95% CI	M-H, Random, 95% CI
Homer 2001 (STOMP) McLachlan 2012 (COSMOS)	52 186	550	62	539 22 158 43	2.7%	0.82 [0.58, 1.17]	
Tracy 2013 (M@NGO)	114	871	110	877 33	3.6%	1.04 [0.82, 1.33]	
Total (05% CI)		2577	2	574 10	0.0%	0.86 [0.70, 1.06]	
Fotal events	352	2511	417	514 10	0.074	0.00 [0.10, 1.00]	
Heterogeneity: Tau <sup>2</sup> = 0.02; Chi <sup>2</sup> =	= 4.32, df = 2 (P =	= 0.12); I <sup>z</sup> =	54%			-	0.5 0.7 1 1.5
Test for overall effect: Z = 1.41 (P	= 0.16)						Favours caseload Favours standard care
F Instrumental birth							
	Caseload m	idwifery	Standard	l care		Risk Ratio	Risk Ratio
amonder Turienzo 2020 (PODDIC)	Events	Total	Events	Total	Weight	M-H, Random, 95% Cl	M-H, Random, 95% Cl
lomer 2001 (STOMP)	71	550	63	539	14.1%	1.10 [0.80, 1.52]	
IcLachlan 2012 (COSMOS) racy 2013 (M@NGO)	187 172	1156 871	207 171	1158 877	43.8% 39.5%	0.90 [0.76, 1.08] 1.01 [0.84, 1.22]	
otal (95% CI)		2746		2739	100.0%	0.98 [0.87, 1.10]	+
otal events leterogeneity: Tau <sup>2</sup> = 0.00; Chi <sup>2</sup> = 1 'est for overall effect: Z = 0.35 (P =	444 .57, df = 3 (P = 0 0.73)	1.67); I² = 0%	453				0.5 0.7 1 1.5 2 Favours caseload Favours standard care
C Drotorn birth							
a Preterm birth	Casalaad	iduiforu	Standar	d care		Risk Ratio	Risk Ratio
	1 (1)		Junudi	Tetal	Weight	M-H Random 95% Cl	M.H. Random 95% CI
Study or Subgroup	Events	Total	Events	Total		man, nundoni, oow or	min, rundoni, oow or
Study or Subgroup Fernandez Turienzo 2020 (POPPIE	Events 31	Total 169	Events 19	165	28.4%	1.59 [0.94, 2.70]	
Study or Subgroup Fernandez Turienzo 2020 (POPPIE AcLachlan 2012 (COSMOS) Fracy 2013 (M@NGO)	Events ) 31 42 39	Total 169 1156 871	Events 19 45 51	165 1158 877	28.4% 35.6% 36.0%	1.59 [0.94, 2.70] 0.93 [0.62, 1.41] 0.77 [0.51, 1.16]	
Study or Subgroup Fernandez Turienzo 2020 (POPPIE AcLachian 2012 (COSMOS) racy 2013 (M@NGO)	Events Events 31 42 39	Total 169 1156 871	Events 19 45 51	165 1158 877	28.4% 35.6% 36.0%	1.59 [0.94, 2.70] 0.93 [0.62, 1.41] 0.77 [0.51, 1.16]	
itudy or Subgroup fernandez Turienzo 2020 (POPPIE IdLachian 2012 (COSMOS) fracy 2013 (M@NOO) fotal (95% CI) fotal events	Events ) 31 42 39 112	Total 169 1156 871 2196	Events 19 45 51	165 1158 877 2200	28.4% 35.6% 36.0% 100.0%	1.59 [0.94, 2.70] 0.93 [0.62, 1.41] 0.77 [0.51, 1.16] 1.01 [0.69, 1.50]	
Study or Subgroup Fernandez Turienzo 2020 (POPPIE McLachian 2012 (COSMOS) Tracy 2013 (M@NGO) Total (95% CI) Total events Heterogeneiby: Tau <sup>2</sup> = 0.07; Chi <sup>2</sup> = J	Events Events 31 42 39 4.64, df = 2 (P = 0	<u>Total</u> 169 1156 871 <b>21</b> 96 0.10); <b>P</b> = 57	Events 19 45 51 115 %	165 1158 877 2200	28.4% 35.6% 36.0% 100.0%	1.59 [0.94, 2.70] 0.93 [0.62, 1.41] 0.77 [0.51, 1.16] 1.01 [0.69, 1.50]	

Fig. 3 Forest plots of RCTs reporting maternal outcomes, summarized in meta-analyses if not deemed clinically heterogeneous. A; perineal tear grade I-IV (absolute risk in standard care: 27% and 34%). B: perineal tear grade III-IV (absolute risk in standard care ranged from 1.8–3.3%). C: bleeding > 1000 ml (absolute risk in standard care ranged from 0.6–5.6%). D: total caesarean section (absolute risk in standard care ranged from 18–31%). E: acute caesarean section (absolute risk in standard care ranged from 12–21%). F: instrumental birth (absolute risk in standard care ranged from 7–19%). G: preterm birth (absolute risk in standard care ranged from 4–12%) caesarean section (both  $\oplus \oplus OO$ ). The risk of instrumental birth and preterm delivery is probably not affected (both  $\oplus \oplus \oplus O$ ).

Regarding feasibility, RCTs without major risk of bias reported that 56% [14] or 57% [21] of the women were assisted by their primary caseload midwife during birth, and 79% [18], 81% [14], and 89% [21], respectively, by a midwife in the caseload team. No studies reported results separately for women with fear of birth.

#### **Ongoing studies**

Out of 115 trials identified in Clinical Trials.gov and 198 in ICTRP, no one fulfilled our PICO.

# Discussion

This quantitative evidence synthesis shows, in general, little or no difference between caseload midwifery and standard care comparable to the Swedish setting, where the same midwife generally provides antenatal care and postnatal checkup, but other midwives assist at birth and the first week postpartum. For child outcomes, there may be little or no difference regarding perinatal mortality and neonatal morbidity. No RCTs reported severe neonatal morbidity, and available evidence did not allow conclusions regarding breastfeeding after hospital discharge. Regarding maternal outcomes, the present review shows that the risk of preterm birth, as well as instrumental birth, is probably not affected. There may be little or no difference in the risk of perineal tear and bleeding. Caseload midwifery may, however, reduce the incidence of caesarean section. No conclusions could be drawn regarding maternal mortality, intensive care, HRQL, postpartum depression, and health care experience/ satisfaction/confidence.

Our results differ in several ways from the Cochrane review that was also based on RCTs [1]. In contrast to their findings, our systematic review and meta-analyses do not support favorable effects of caseload midwifery regarding the risk of preterm birth or the risk of instrumental birth. Furthermore, the Cochrane review reported favorable effects of the intervention regarding their primary mortality outcome, which in addition to perinatal mortality also included fetal loss before 24 weeks [1]. Nevertheless, when perinatal mortality was included only, our results were consistent with the Cochrane review, showing no difference between the comparison groups. Methodological aspects may explain the partly divergent findings. In our meta-analyses, for instance, we only included RCTs without major risk of bias whereas the Cochrane review included all RCTs [1]. Furthermore, data used from one of the RCTs [21], in the meta-analysis of preterm birth in the Cochrane review, seem to be wrongly extracted. When the correct data are used instead (data not shown), the results regarding preterm birth are consistent with ours, i.e. no statistically significant difference. Finally, our comparison, where the same midwife usually provides antenatal care and checkup postnatally, also represents continuity to some extent.

Synthesizing available literature, we found that the only difference between caseload midwifery and standard care concerned the risk of caesarean section. This result was primarily based on studies performed in countries with a high incidence, illustrated, for instance, with the estimate of 25% caesarean section in the low-risk population included in the largest study [21]. With an overall incidence below 20% in 2021 [6], caesarean section is in general less common in Sweden than the worldwide average of 21% in 2018, and considerably lower than 25%, the average for countries in Northern Europe [26]. This may have implications for the applicability of the results. Indeed, the summarized absolute risk reduction of 4% units may neither be reasonable nor desirable in settings with a low incidence of caesarean sections; this intervention is also performed for medical reasons. Interestingly, our results of a significantly reduced incidence of caesarean section differ from the prior Cochrane review, where no difference was reported for midwife-led continuity models versus other models of care, with high certainty confidence in the evidence, and similar results in the subgroup analysis specifically focusing on caseload midwifery [1]. As elaborated upon above, differences in comparison groups as well as methodology may contribute to the divergent results.

Regarding perineal tear and bleeding, our meta-analyses show that there may be no difference between caseload midwifery and standard care. Regarding perineal tear irrespective of grade, however, it may be worth noting that there were only two RCTs available, too heterogeneous to be pooled, and that one of them significantly favored standard care [25].

Notably, the evidence regarding breastfeeding after discharge was inconclusive. Although one study provided data, the risk of bias was conspicuous due to very large loss-to-follow-up, with a skewed distribution between groups [25]. Future RCTs would be required to gain knowledge regarding the potential effect of the caseload model of care on breastfeeding.

For the important maternal outcomes HRQL and postpartum depression, no conclusions could be drawn from available literature. Neither could any conclusions be drawn regarding potential effects of caseload midwifery on health care experience/satisfaction and confidence. Furthermore, no study focused specifically on pregnant women with fear of birth, a subgroup that could benefit from further attention in future RCTs investigating the effects of caseload midwifery. Another subgroup that may deserve further attention, partly overlapping with fear of birth, could be women with previous trauma, including birth trauma.

Regarding feasibility, available literature shows that more than every second woman in a caseload model of care has their primary caseload midwife present at birth. With a caseload team of 12, nine in 10 women could be expected to have any of the team midwives present at birth, and the corresponding numbers with a team of six was eight in 10. These results could be of value for women entering the model, for informed expectations.

An important strength of this systematic review is that it, with a quantitative approach, provides a synthesis of currently available evidence regarding child and maternal outcomes for caseload midwifery compared with standard care, the latter defined as the same midwife usually providing care during pregnancy and postnatally but not during birth. Indeed, the fact that several studies were excluded as physicians provided much of the care illustrates the diversity of models of care worldwide. This may also have implications for the generalizability; our results may primarily be applicable in settings where midwives are the main care providers, and physicians if medical issues arise. Another strength of this review is the performance, being guided by an established HTA process and including thorough and transparent assessments. It could also be considered a strength that studies with major risk of bias were not included in our meta-analyses; this approach may increase the certainty of evidence.

Limitations of the present review include that few studies fulfilled our PICO. Furthermore, no RCTs at all could be identified for some outcomes, and for others, the certainty of evidence was very low and inconclusive. Another limitation is that we focused on quantifiable outcomes; an evidence synthesis of qualitative research could provide additional insights. The subgroup of women with fear of birth and/or previous birth trauma could be a subgroup of particular interest in such an evidence synthesis.

#### Conclusions

This systematic review shows that there may be little or no difference between caseload midwifery and standard care regarding perinatal mortality and neonatal morbidity. Furthermore, the risk of preterm birth, as well as instrumental birth, is probably not affected. Regarding perineal tear, there may be no difference between the comparison groups. Caseload midwifery may reduce the incidence of caesarean section. No evidence is available regarding severe neonatal morbidity, and no conclusions can be drawn regarding breastfeeding after hospital discharge, maternal mortality, intensive care, HRQL, postpartum depression, and health care experience/ satisfaction/confidence. As evidence regarding some critical and important child and maternal outcomes is lacking, and the certainty of evidence for others is very low or low, additional RCTs in relevant settings could add further insights regarding the potential of caseload midwifery model of care.

#### Supplementary Information

The online version contains supplementary material available at https://doi.org/10.1186/s12884-023-05967-x.

Supplementary Material 1	
Supplementary Material 2	
Supplementary Material 3	
Supplementary Material 4	
Supplementary Material 5	

#### Acknowledgements

The authors are grateful to Maud Eriksson, retired medical librarian at Sahlgrenska University Hospital, who contributed to the literature search and in the inclusion process.

#### Authors' contributions

L Wassén, A Hagman, M Lindroth, C Rubertsson, and A Wessberg conceived the study, and B Borgström Bolmsjö, S Frantz, A Strandell, and SM Wallerstedt designed it. T Svanberg performed the literature searches. All authors took part in the process of including and excluding articles. S Frantz and SM Wallerstedt extracted data from the included studies, which were checked by L Wassén, B Borgström Bolmsjö, A Hagman, M Lindroth, C Rubertsson, A Strandell, and A Wessberg. L Wassén, B Borgström Bolmsjö, A Hagman, M Lindroth, C Rubertsson, A Strandell, A Wessberg, and SM Wallerstedt assessed the study quality. SM Wallerstedt performed the meta-analyses. L Wassén and SM Wallerstedt drafted the manuscript and all authors revised if for intellectual content. All authors approved the final version of the article.

#### Funding Not applicable

Open access funding provided by University of Gothenburg.

#### **Data Availability**

The datasets supporting the conclusions of this article are included within the article and its additional file.

## Declarations

#### **Ethics approval and consent to participate** Not applicable.

**Consent for publication** 

Not applicable.

#### **Competing interests**

The authors declare no competing interests.

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# Received: 21 October 2022 / Accepted: 31 August 2023 Published online: 15 September 2023

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