The efficacy of emergency cervical cerclage in singleton and twin pregnancies: a systematic review with meta-analysis

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OBJECTIVE: An emergency (rescue) cervical cerclage can be offered to pregnant women presenting with dilatation and prolapsed membranes in the second trimester of pregnancy because of cervical insufficiency. This study aimed to investigate the effectiveness of an emergency cerclage in both singleton and twin pregnancies in the prevention of extreme premature birth.

DATA SOURCES: We performed a systematic literature search in PubMed and Embase from inception to June 2022 for transvaginal cervical emergency cerclages.

STUDY ELIGIBILITY CRITERIA: All studies on transvaginal cervical emergency cerclages with at least 5 patients and reporting survival were included.

METHODS: Included studies were assessed for quality and risk of bias with an adjusted Quality In Prognosis Studies tool. Random-effects meta-analyses and meta-regressions were performed for the primary outcome: survival.

RESULTS: Our search yielded 96 studies, incorporating 3239 women, including 14 studies with an expectant management control group, incorporating 746 women. Overall survival after cervical emergency cerclage was 74%, with a fetal survival of 88% and neonatal survival of 90%. Singleton and twin pregnancies showed similar survival, with a pregnancy prolongation of 52 and 37 days and a gestational age at delivery of 30 and 28 weeks, respectively. Meta-regression analyses indicated a significant inverse association between mean gestational age at diagnosis and pregnancy prolongation and no association between dilatation or gestational age at diagnosis and gestational age at delivery. Compared with expectant management, emergency cerclage significantly increased overall survival by 43%, fetal survival by 17% and neonatal survival by 22%, along with a significant pregnancy prolongation of 37 days and reduction in delivery at <28 weeks of gestation of 55%. These effects were more profound in singleton pregnancies than in twin pregnancies.

CONCLUSION: This systematic review indicates that, in pregnancies threatened by extreme premature birth because of cervical insufficiency, emergency cerclage leads to significantly higher survival, accompanied by significant pregnancy prolongation and reduction in delivery at <28 weeks of gestation, compared with expectant management. The mean gestational age at delivery was 30 weeks, independent of dilatation or gestational age at diagnosis. Survival was similar for singleton and twin pregnancies, implying that emergency cerclage should be considered in both.

Key words: cerclage, cervical insufficiency, emergency, examination indicated, expectant management, physical examination indicated, pregnancy, premature birth, rescue, singleton pregnancy, twin pregnancy

Introduction

Extreme prematurity is one of the main causes of perinatal mortality and morbidity and is defined as a birth below 28 weeks of gestation.[1] With every extra week of pregnancy addition, survival increases exponentially. The survival rates are as follows: 0.7% at <24 weeks, 31.0% at 24 weeks, 59.0% at 25 weeks, and 75.0% at 26 weeks.[2] However, approximately half of surviving newborns born before 25 weeks of gestation suffer from at least 1 disability, even though technical and neonatal healthcare strategies improve every year.[3] Therefore, the prevention of extreme prematurity is of utmost importance. Moreover, preventive measures vary, depending on the cause. Several pathologic pathways are thought to induce extreme prematurity, such as cervical insufficiency.

EDITOR'S CHOICE

Cervical insufficiency, incompetence, or weakness is thought to account for 15% of perinatal losses between 16 and 26 weeks of gestation and is defined as the inability of the uterine cervix to retain a pregnancy in the second trimester of pregnancy in the absence of clinical contractions or labor.[4–7] Women

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AJOG MFM at a Glance

Why was this study conducted?

This systematic review with meta-analysis and meta-regression aimed to assess pregnancy outcomes of emergency cerclages in both singleton and twin pregnancies in single-arm studies and compare the outcomes of emergency cerclages with that of expectant management.

Key findings

This systematic review indicated that, in pregnancies threatened by extreme premature birth because of cervical insufficiency, placement of an emergency cerclage leads to a significantly higher survival rate, accompanied by a significant pregnancy prolongation and reduction in delivery at <28 weeks of gestation, compared with expectant management. The survival rates were similar for singleton and twin pregnancies, implying that emergency cerclage should be considered in both singleton and twin pregnancies.

What does this add to what is known?

This systematic review includes all reported studies on emergency cerclages with ≥5 participants and provides an up-to-date overview of the efficacy of an emergency cerclage, in both singleton and twin pregnancies.

often present with hardly any symptoms, resulting in a spontaneously dilated cervix with prolapse of membranes at or beyond the external os. In this condition, fetal membranes are in direct contact with vaginal bacteria. This results in a high risk of infection leading to extreme premature birth (PMB) and associated perinatal mortality.[6,7] Therapeutic interventions may vary from expectant management to termination of pregnancy or placement of an emergency cervical cerclage.

An emergency, rescue, or physical examination−indicated cervical cerclage is a hazardous and sometimes difficult intervention. During the operation, a stitch or tape is placed around the cervix through the vaginal route to restore the original anatomic situation as much as possible, with membranes inside the uterus and a closed cervix. In this way, the cerclage offers effective structural support to the cervix and could potentially serve as a barrier to protect fetal membranes from ascending pathogens.[8,9] However, because of prolapse of membranes, there is the risk of rupture of membranes during the procedure. Furthermore, further in pregnancy, there is the risk of infection, cervical laceration, and, despite the intervention, extreme PMB. Therefore, effective placement of an emergency cerclage does not guarantee a successful pregnancy.

Studies on the efficacy of emergency cerclages present inconsistent results. Combined results of small numbers of studies in previously conducted systematic reviews have not led to a consensus as to the success of an emergency cerclage in saving singleton pregnancies.[10−14] Moreover, the success rates might be different for singleton and twin pregnancies, as the etiology, and thereby management, of the cervical insufficiency might differ.[15,16] A recent randomized controlled trial (RCT) shows that emergency cerclages reduce PMB at <28 weeks of gestation in twin pregnancies from 85% to 41% (relative risk [RR], 0.5) compared with expectant management, which is in line with previous meta-analyses.[17−19] However, data in these studies are based on very small groups concluding that sufficient evidence is lacking. Therefore, there is still no clear consensus among experts on the effectiveness of emergency cerclages in singleton and twin pregnancies.

To give an overview of all available knowledge on the effectiveness of an emergency cerclage in the prevention of extreme premature birth because of cervical insufficiency, in both singleton and twin pregnancies, we performed a systematic review with meta-analysis. Thus, we systematically searched for literature on fetal and neonatal survival after an emergency cerclage. The subanalyses were performed to compare the efficacy between emergency cerclage and expectant management in singleton and twin pregnancies. This study can be used to assist the pregnant woman, her partner, and the physician to make an evidence-based decision whether to place an emergency cerclage or not.

Materials and Methods

This systematic review was conducted following the standards conforming with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines.[20] The study protocol was registered with the International Prospective Register of Systematic Reviews (registration number: CRD42022320634).[21]

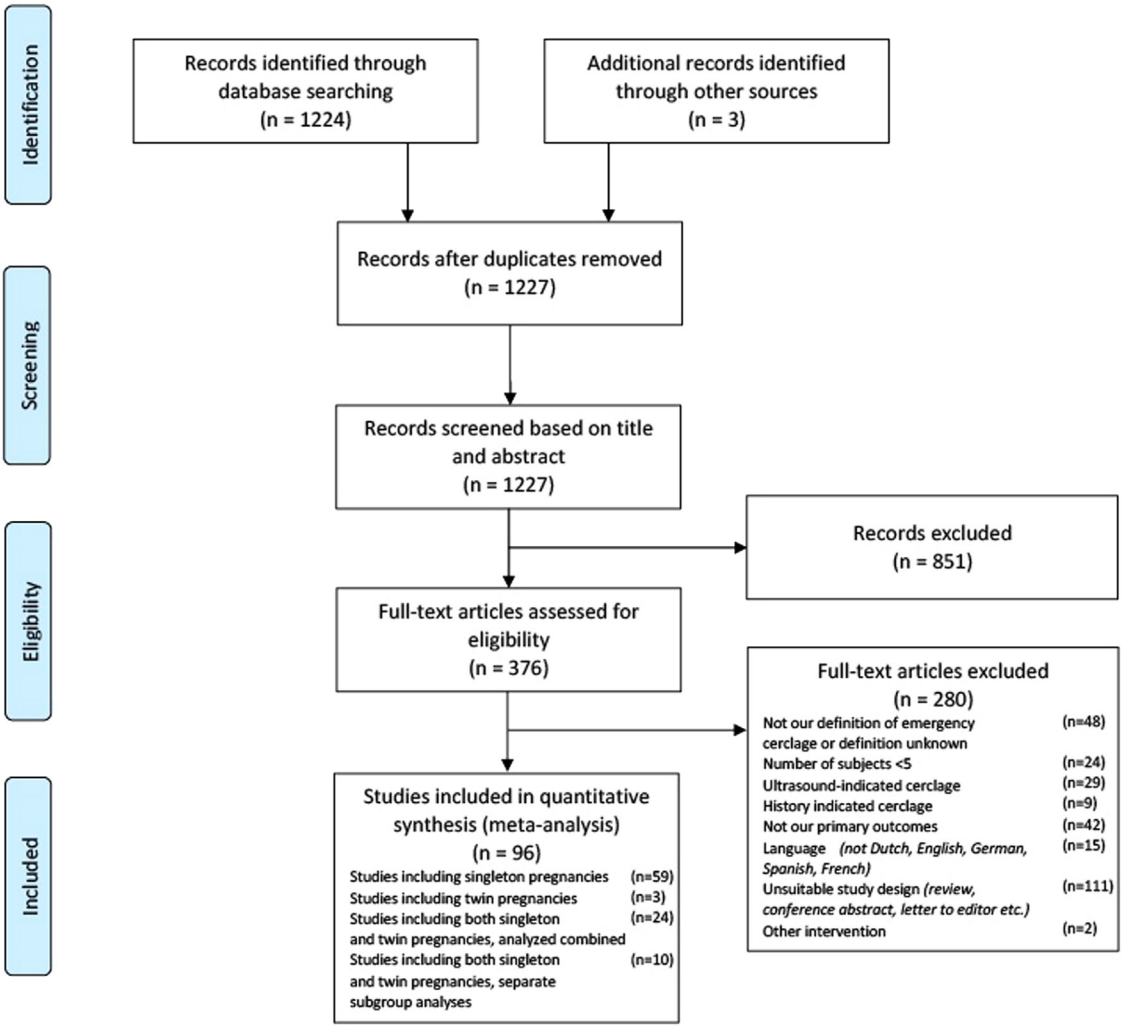
Eligibility criteria, information sources, and search strategies

We conducted a systematic search for relevant literature in which emergency (rescue) transvaginal cerclages were placed using PubMed and EMBASE. These electronic databases were searched to find literature from inception to June 2022, with the following keywords: 'cervical cerclage', 'uterine cervical insufficiency', 'cervical stitch', combined with 'emergency' or 'rescue'. There was no restriction for time or geographic location. Study selection was performed using Covidence.[22]

Study selection

To determine the eligibility of the studies, a 2-phased selection process was performed (Figure 1). Electronic search and review of the eligible studies were performed by 2 authors (C.C.H. and R.P.B.). Disagreement on potential relevance was resolved by discussion with a third author (J.V.D.). We included RCTs, cohort studies, and case series, with a minimum of 5 patients, in which women with singleton or twin pregnancies were observed for threatened (extreme) PMB based on cervical insufficiency, with or without a history of PMB before 37 weeks of gestation or second-trimester losses. Cervical insufficiency was defined as a premature cervical dilatation of

FIGURE 1
PRISMA flow diagram of the systematic review selection procedure



PRISMA, Preferred Reporting Items for Systematic Reviews and Meta-Analyses.

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≥2 cm and/or exposure of fetal membranes at or beyond the external os before 28 weeks of gestation, detected either by ultrasound examination of the cervix or by speculum or physical examination, with minimal physical complaints (vaginal discharge, bleeding, or sensation of pressure). Women had to be treated by either transvaginal cervical emergency cerclage or expectant management (which might consist of bed rest and/or progesterone treatment). Studies were only included if they reported original data on fetal and or neonatal survival. When only a subgroup of the study fitted the inclusion criteria, only this subgroup was included. When data were not reported separately for the subgroup that fitted the inclusion criteria, corresponding authors were contacted with the request of obtaining these data. Studies were excluded for the following reasons: (1) use of cerclage other than transvaginal cervical emergency cerclage; (2) use of pessary; (3) unsuitable study designs (letters to the editor, editorials, reviews [containing no original data], and poster abstracts); (4) any language other than English, Dutch, German, French, and Spanish; and (5) full text not available.

Data extraction

Characteristics and obstetrical and surgical outcomes were extracted. All included studies were independently extracted by 2 reviewers (C.C.H. and R. P.B.). Extracted information from each study consisted of study design, publication year, and sample size. When a study consisted of data in both singleton and twin pregnancies, we extracted the data separately. Subject characteristics included number of fetuses, maternal age, nulliparity, previous PMB, uterus anomaly, cervical surgical history, gestational age (GA), cervical dilatation at diagnosis, and prolapsed membranes at diagnosis, and surgical technique used.

The primary outcome of interest was the overall survival rate, defined as the number of survived neonates at 28 days

divided by the total number of fetuses. The secondary outcomes included perioperative use of antibiotics, tocolysis, or progesterone; amniocentesis; amnioreduction; chorioamnionitis; preterm premature rupture of membranes (PPROM) during or after the intervention; pregnancy prolongation; GA at delivery; delivery at <34, <32, and <28 weeks of gestation; fetal survival (defined as the number of live-born neonates divided by the number of fetuses); neonatal survival (defined as the number of survived neonates at 28 days divided by the number of live-born neonates); miscarriage at <20 weeks of gestation; stillbirth at ≥20 weeks of gestation; birthweight; neonatal intensive care unit (NICU) admission; and neonatal comorbidity.

Assessment of risk of bias

All included studies were assessed for quality and risk of bias using a modified list of items described in the Quality In Prognosis Studies (QUIPS) tool, which we made suitable for this review (Appendix A).[23] This tool is very applicable to observational studies. Studies were scored for risk of bias in 6 domains: study participation, study attrition, intervention, variable measurement, data reporting, and study design. Studies were only scored for study attrition in case of loss to follow−up. Similar to the original QUIPS tool, all domains in the adjusted tool were equally important for the total score. Studies with a positive score of ≥60% were defined as high quality, ≥30% to <60% as moderate quality, and <30% as low quality.

All studies containing an expectant management group were assessed for quality and risk of bias using the Risk Of Bias In Non-randomized Studies - of Interventions (ROBINS-I) tool.[24] Studies were scored for risk of bias on 7 domains: bias because of confounding, participant selection, intervention classification, intended intervention deviation, missing data, outcome measurement, and result selection. Risk-of-bias judgment was classified as "low," "moderate," "serious," and "critical."

Data synthesis

Overall ("total") and subgroup analyses were performed for primary and secondary outcome parameters. The subgroups consisted of data on singleton pregnancies ("singleton"), twin pregnancies ("twins"), and both singleton and twin pregnancies where study results were not presented separately ("unspecified"). We performed a separate analysis on studies that contained both an intervention group of women that received an emergency cerclage ("emergency cerclage") and a control group of women that received expectant management ("expectant management").

Weighted proportions with corresponding 95% confidence intervals (CIs) were calculated for all groups to assess the association between emergency cerclage and survival. Random intercept logistic regression models were used, with a maximum likelihood ML estimator for tau[2] and a logit transformation. For individual studies in forest plots, the Clopper-Pearson confidence intervals were presented, and a continuity correction of 0.5 in studies with zero frequencies was applied. To assess the association between emergency cerclage and continuous outcomes, weighted means with corresponding 95% CIs were calculated for all groups, using an inverse variance method with restricted ML (REML) estimator for tau.[2] The I^2 and 95% prediction intervals were used as measures of heterogeneity.[25] An I^2 value of >50% was considered suggestive of statistical heterogeneity.[26] Funnel plots in combination with the Egger regression test for funnel plot asymmetry were used to evaluate the possible presence of publication bias.[27]

Sensitivity analyses were performed to evaluate the effect of publication year and participants on the primary outcome. We evaluated the effect of publication year, as differences in neonatal healthcare over the years might affect the outcomes. Considering that neonatal healthcare has not changed substantially between the year 2000 and the present, we selected studies published since 2000. We assessed the effect of participant number, considering that small case series might introduce publication bias. We restricted the selection to studies with more than 15 participants.

Meta-regression analyses were performed to assess association between mean cervical dilatation and GA at the time of diagnosis, and pregnancy outcome in survival, pregnancy prolongation, GA at delivery. Mixed-effects meta-regression models with a linear trend were used, with REML estimator for tau.[2]

Statistical analyses were performed with the statistical software R (version 4.1.3; R Foundation for Statistical Computing, Vienna, Austria), using the *meta* (version 5.5-0) and *metafor* (version 3.4.0) packages.[28−30]

Results

Study selection

The search strategy identified 1227 unique studies eligible for this study (Figure 1). After selection, 96 studies investigating the effectivity of transvaginal emergency cerclage were found eligible for inclusion.[8,31−125] Most studies excluded used other definitions for threatened extreme premature birth because of cervical insufficiency and indication for emergency cerclage, contained only data on history- or ultrasound-indicated cerclages, did not present data of our primary outcome, or did not have a suitable study design for our review. For several studies, only existing subgroups that fitted our inclusion criteria were included.[39,41,44,51,65,67,69,70,80,97,100,109,115,116,121] For a few studies, participant data were reported or retrieved individually, and a subgroup of these studies was included, which contained only participants that fitted our inclusion criteria.[47,60,73,83,92,98,108,117,119,124,125]

Study characteristics

Singleton and twin pregnancies. Table 1 presents the baseline characteristics. The 96 studies included in the meta-analysis consisted of 3239 pregnant women. Of these studies, 10 contained 2 subgroups, which were both included in our subgroups. Of the 96 studies, 72 studies or subgroups of studies reported data on singleton pregnancies (n=2500), 9

TABLE 1
Baseline characteristics of women treated with emergency cerclage in singleton and twin pregnancies

Characteristic	Studies/ participants	Total	Singleton pregnancy (S)	Twin pregnancy (T)	Unspecified	*P* value (S-T)
Participants	3239	3239	2500	156	583	—
Demographic data						
Age (y)	76/2652	30.7 (30.2–31.2)	30.8 (30.2–31.3)	31.9 (30.3–33.5)	30.0 (28.8–31.3)	.18
Nulliparous (%)	47/1480	45.7 (38.8–52.8)	40.6 (32.5–49.3)	84.0 (53.7–96.0)	45.5 (35.9–55.4)	.01[a]
Obstetrical and surgical history						
Previous premature birth (%)	41/1502	19.2 (15.1–24.2)	23.4 (18.4–29.2)	1.7 (0.1–18.9)	17.9 (11.9–26.0)	.03[a]
Uterus anomaly (%)	11/444	3.2 (0.6–7.8)	1.5 (0.0–6.5)	2.0 (0.0–13.8)	10.7 (5.0–18.3)	.88
Previous cervical surgical procedures (%)[b]	24/645	13.4 (8.5–19.2)	12.3 (6.4–19.9)	11.6 (0.0–44.9)	16.9 (8.8–27.0)	.96
Diagnosis and surgery						
GA at diagnosis (wk)	81/2751	21.8 (21.5–22.1)	21.9 (21.5–22.2)	21.8 (21.1–22.6)	21.9 (21.5–22.3)	.92
Cervical dilatation at diagnosis (cm)	54/1768	3.3 (3.0–3.6)	3.3 (3.1–3.5)	2.5 (1.9–3.2)	3.7 (3.2–4.1)	.03[a]
Prolapsed membranes beyond the external os (%)	57/2057	97.8 (93.6–99.3)	96.6 (89.3–99.0)	92.9 (50.2–99.4)	99.8 (89.2–100.0)	.60
Surgical technique (%)	96/3239					
McDonald		64.5 (51.5–76.5)	67.0 (50.9–81.2)	61.6 (12.8–98.4)	54.7 (29.2–78.0)	.85
Shirodkar		3.2 (1.1–6.3)	3.1 (0.7–7.0)	1.7 (0.0–11.5)	4.2 (0.3–12.3)	.69
Combination or other		21.3 (11.4–33.1)	18.5 (7.7–32.6)	25.5 (0.0–77.8)	28.1 (8.4–53.7)	.77

Data are presented as proportion (95% CI) or mean (95% CI), unless otherwise specified. Unspecified indicates data that were not reported separately for singleton and twin pregnancies. In each column, not all studies are represented, depending on available data.

CI, confidence interval; *GA*, gestational age.

[a] A *P*-value of ≤ 0.05 was considered statistically significant; [b] Conization, loop excision of the transformation zone, cerclage, dilatation and evacuation, and dilatation and curettage.

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studies or subgroups of studies reported data on twin pregnancies (n=156), and 25 studies or subgroups of studies did not report data separately depending on the pregnancy type (n=583). The mean maternal age was 30.7 years, and the mean GA at diagnosis was 21.9 weeks. In the singleton group, significantly less women were nulliparous (40.6% versus 84.0%) and significantly more women had experienced previous preterm births (34.4% vs 1.7%), compared to the twin group. At the time of diagnosis, 97.8% of women had prolapsed membranes beyond the external cervical os, with a mean cervical dilatation of 3.3 cm. Mean cervical dilatation at diagnosis was significantly higher in singleton pregnancies than in twin pregnancies (3.3 vs 2.5 cm). The McDonald technique was performed most often (63.6%).

Emergency cerclage vs expectant management. Of note, 14 studies contained both intervention and control groups (Table 2), with 746 pregnant women (445 women receiving a transvaginal emergency cerclage and 301 women receiving expectant management). Thirteen studies contained data on singleton pregnancies. One study contained data on twin pregnancies, only the subgroup with ≥4 cm dilatation in this study could be included in our analysis. The mean maternal ages for the cerclage and expectant groups were 30.2 and 30.3 years, respectively. A significantly higher proportion of women had undergone previous cervical surgical procedures in the cerclage group than in the expectant management group (5.0% vs 0.0%). The mean GAs at diagnosis were 21.9 weeks for the women receiving a cerclage and 22.6 weeks for the women managed expectantly. At the time of diagnosis, 98.6% of women in the cerclage group and 98.3% of the women managed expectantly had prolapse of membranes beyond the external cervical os. The mean cervical dilatation was significantly less in the cerclage group (3.5 cm) than in the expectant management group (3.9 cm).

Risk of bias of included studies

The results of the adjusted QUIPS quality assessment of included studies are presented in Appendix A. The mean quality score of all included studies was 73% (range, 46%–93%), and most studies were classified as high quality (n=86). Of note, 9 studies were classified as moderate quality. No study was classified as low quality. The mean quality scores were 75% (range, 46%–93%) for studies

TABLE 2
Baseline characteristics of women treated with emergency cerclage vs expectant management

Characteristic	Studies/participants	Emergency cerclage	Expectant management	*P* value
Participants	14/746	445	301	—
Singleton	13/729	428	282	—
Twin	1/36	17	19	—
Demographic data				
Age (y)	11/652	30.2 (29.0–31.5)	30.3 (28.6–32.1)	.99
Nulliparous (%)	6/232	34.7 (17.4–57.2)	49.2 (38.4–60.2)	.34
Obstetrical and surgical history				
Previous premature birth (%)	4/275	26.8 (20.6–34.0)	17.7 (9.6–30.2)	.38
Previous cervical surgical procedures (%)[a]	4/143	5.0 (1.9–12.6)[b]289	0.0 (0.0–100.0)[b]	.00[b]
Diagnosis and surgery				
GA at diagnosis (wk)	13/710	21.9 (20.8–23.0)	22.6 (21.3–23.8)	.13
Cervical dilatation at diagnosis (cm)	11/652	3.5 (3.0–4.1)[b]	3.9 (3.4–4.4)[b]	.02[b]
Prolapsed membranes beyond the external os (%)	5/317	98.6 (48.1–100.0)	98.3 (39.2–100.0)	.76
Surgical technique (%)	14/746			
McDonald		40.1 (9.6–75.8)	NA	—
Shirodkar		8.3 (0.0–30.3)	NA	—
Combination or other		31.3 (3.6–70.3)	NA	—

Data are presented as proportion (95% CI) or mean (95% CI), unless otherwise specified. In each column, not all studies are represented, depending on available data.

CI, confidence interval; *GA*, gestational age; *NA*, not applicable.

[a] Conization, loop excision of the transformation zone, cerclage, dilatation and evacuation, and dilatation and curettage; [b] A *P*-value of ≤ 0.05 was considered statistically significant.

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reporting on singleton pregnancies and 72% (range, 62%–85%) for studies reporting on twin pregnancies. For studies that contained an expectant management group, the mean quality score was 80% (range, 46%–93%). Items that scored the lowest among all studies included study design, that is, the use of a prospective longitudinal or RCT study design (n = 9) or a control group (n = 14). The results of the ROBINS-I quality assessment of studies containing an expectant management group are presented in Appendix A. Of note, 1 study was classified as moderate risk of bias. Moreover, 13 studies were classified as serious risk of bias, based on a serious risk of bias because of confounding.

Interventions and complications perioperatively and during pregnancy

Singleton and twin pregnancies. Table 3 presents the interventions and complications perioperatively and during pregnancy, divided per pregnancy type. The use of perioperative antibiotics, tocolysis, and progesterone was high in the overall group (>95%). The use of progesterone significantly differed between the singleton pregnancy group (100.0%) and the twin pregnancy group (33.3%). PPROM during the procedure occurred in 6.1% of all pregnancies, and PPROM later in pregnancy occurred in 24.3% of all pregnancies. Chorioamnionitis complicated 23.1% of all pregnancies and occurred significantly less often in singleton pregnancies (20.8%) than in twin pregnancies (41.0%).

Emergency cerclage vs expectant management. Meta-analyses of the interventions and complications perioperatively and during pregnancy of the included studies that contained both intervention and expectant management groups are summarized in Table 4. The use of antibiotics, tocolysis, and progesterone was similar for both groups (>98%). Amniocentesis was performed significantly less often in the cerclage group than in the expectant management group (61.1% vs 100.0%); however, this was based on a single study with 18 participants receiving an emergency cerclage and 7 participants receiving expectant management. The rates of PPROM and chorioamnionitis did not significantly differ between the intervention and expectant management groups, possibly because of the large heterogeneity among the 5 studies.

Pregnancy outcome

Singleton and twin pregnancies. Table 5 presents the pregnancy outcomes after placement of an emergency cerclage in singleton and twin pregnancies. A total of 3496 fetuses were included in this

TABLE 3
Interventions and complications perioperatively and during pregnancy after placement of emergency cerclage in singleton and twin pregnancies

Variable	Studies/ participants	Total	Singleton pregnancy (S)	Twin pregnancy (T)	Unspecified	*P* value (S-T)
Perioperative interventions						
Antibiotics (%)	80/2739	100.0 (99.7–100.0)	100.0 (99.9–100.0)	100.0 (0.0–100.0)	99.8 (92.6–100.0)	.76
Tocolysis (%)	67/2324	99.7 (98.6–100.0)	99.8 (98.3–100.0)	99.0 (56.9–100.0)	99.7 (91.6–100.0)	.55
Progesterone (%)	17/604	95.2 (63.0–99.6)	100.0 (82.8–100.0)	33.3 (6.4–78.4)	68.8 (26.4–93.1)	.01[a]
Amniocentesis (%)	19/587	68.1 (15.1–96.2)	84.4 (12.3–99.5)	77.6 (53.3–91.3)	14.2 (0.1–97.1)	.82
Amnioreduction (%)	12/356	38.9 (8.5–81.3)	36.2 (35.6–36.9)	NA	66.7 (37.6–86.9)	NA
Perioperative complications						
PPROM during procedure (%)	25/867	6.1 (3.6–10.1)	7.6 (4.1–13.7)	NA	4.6 (2.4–8.5)	NA
Complications during pregnancy						
PPROM (%)	40/1313	24.3 (19.9–29.4)	26.1 (20.7–32.3)	34.6 (24.9–45.8)	16.7 (9.4–27.9)	.15
Chorioamnionitis (%)	49/1534	23.1 (18.0–29.1)	20.8 (15.3–27.8)	41.0 (26.4–57.4)	25.3 (15.6–38.3)	.01[a]

Data are presented as proportion (95% CI) or mean (95% CI), unless otherwise specified. Unspecified indicates data that were not reported separately for singleton and twin pregnancies. In each column, not all studies are represented, depending on available data.

CI, confidence interval; *NA*, not applicable; *PPROM*, preterm premature rupture of membranes.

[a] A *P*-value of ≤ 0.05 was considered statistically significant.

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analysis. The survival rate was 74.1%, with a viable fetal survival rate of 87.7% and a neonatal survival rate of 90.0%. No major difference in the survival rate was found between singleton (74.5%) and twin (77.7%) pregnancies. Overall pregnancy prolongation was 50.6 days and was significantly longer in singleton pregnancies than in twin pregnancies (52.4 vs 36.5 days). The mean GA at delivery was 30.0 weeks and was significantly longer in singleton pregnancies than in twin pregnancies (30.3 vs 28.3 weeks). Overall, the delivery rates by GA were 60.7% at <34 weeks, 56.9% at <32 weeks, and 38.6% at <28 weeks. The occurrence rates were less in singleton pregnancies compared with twin pregnancies. A significantly greater proportion of singleton pregnancies was affected by miscarriage at <20 weeks of gestation than twin pregnancies (3.5% vs 0.8%). Overall, neonatal birthweight was 1696.2 g, and 65.2% of born neonates were admitted to the NICU. Figure 2 depicts the forest plot of the overall survival rate, showing considerable heterogeneity among studies within the subgroups in terms of I^2, varying from 65% to 77%. This is in accordance with relatively wide 95% prediction intervals, which also reflect considerable differences across studies. Forest plots of all secondary outcomes are depicted in Appendix E, showing considerable heterogeneity. Funnel plots for the primary outcome in the combined group and in the subgroups are presented in Appendix C. The funnel plot of the survival rate in the combined analysis is slightly indicative of publication bias, statistically confirmed using the Egger regression test for funnel plot asymmetry (*P*=.04). However, separate funnel plots of subgroup analyses ("singleton," "twin," and "unspecified") are hardly indicative of publication bias.

Emergency cerclage vs expectant management. Meta-analysis results of the pregnancy outcomes of the included studies that contained both intervention and expectant groups are summarized in Table 6. A total of 782 fetuses were included in this analysis, from 710 singleton pregnancies and 36 twin pregnancies. The survival rates were all significantly higher in pregnancies of women treated with emergency cerclage than in pregnancies of women treated with expectant management: overall survival rate of 70.9% vs 27.9% (pooled ratio, 1.7), viable fetal survival rate of 81.7% vs 64.4% (pooled ratio, 1.4), and neonatal survival rate of 92.9% vs 70.7% (pooled ratio, 1.4). The survival rates were higher in studies on singleton pregnancies than in the single study on twin pregnancies, possibly partially because of the relatively advanced dilatation in twin pregnancies (≥4 cm). However, overall and fetal survival rates remained significantly higher in twin pregnancies treated with emergency cerclage than in twin pregnancies treated with expectant management. Overall, pregnancies were significantly prolonged with a mean of 51.3 days for the emergency cerclage group and 12.6 days for the expectant management group (mean difference of 36.5 days). In singleton pregnancies, the difference in prolongation between an emergency cerclage and an expectant management was 38.4 days (54.1 vs 13.8 days, respectively), whereas, in twin pregnancies, it was 19.2 days (23.7 vs 4.5 days, respectively). GA at delivery was significantly longer in the emergency cerclage group (30.1 weeks) than in the expectant management group (25.1

TABLE 4
Interventions and complications perioperatively and during pregnancy after placement of emergency cerclage vs expectant management

Variable	Studies/ participants	Emergency cerclage	Expectant management	Effect estimate[a]	*P* value
Perioperative interventions					
Antibiotics (%)	10/549	99.9 (70.5–100.0)	99.8 (70.2–100.0)		.87
Tocolysis (%)	10/562	99.0 (61.3–100.0)	98.1 (57.0–100.0)		.36
Singleton	9/526	99.3 (68.4–100.0)	96.9 (46.6–99.9)		.36
Twin	1/36	100.0 (0.0–100.0)	100.0 (0.0–100.0)		.98
Progesterone (%)	2/137	100.0 (0.0–100.0)	98.6 (87.1–99.9)		.75
Amniocentesis (%)	1/25	61.1 (35.7–82.7)	100.0 (59.0–100.0)		.01[b]
Complications during pregnancy					
PPROM (%)	5/252	30.6 (22.5–40.1)	13.2 (1.5–59.8)	1.2 (0.1–10.9)	.79
Chorioamnionitis (%)	5/170	23.4 (11.8–41.0)	35.6 (4.2–87.4)	0.6 (0.1– 3.2)	.47

Data are presented as proportion (95% CI) or mean (95% CI), unless otherwise specified. In each column, not all studies are represented, depending on available data.

CI, confidence interval; *PPROM*, preterm premature rupture of membranes.

[a] Data are presented as pooled ratio (95% CI); [b] A *P*-value of ≤ 0.05 was considered statistically significant.

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weeks), in both singleton and twin pregnancies. Placement of an emergency cerclage significantly reduced the risk of delivery <28 weeks to 38.2%, compared to 93.0% in pregnancies managed expectantly (pooled ratio 0.5). In singleton pregnancies the risk was non-significantly reduced to 30.0% after treatment with emergency cerclage, compared to 75.7% when treated expectantly (pooled ratio 0.4). In twin pregnancies treated with emergency cerclage the risk was significantly reduced to 70.6%, as to 100.0% in the expectant management group (pooled ratio 0.7). Neonates born after treatment with emergency cerclage had a significantly higher mean birthweight than neonates born after treatment with expectant management (1797.9 vs 843.9 g), in both singleton and twin pregnancies. NICU admission rates were lower after treatment with emergency cerclage than after treatment with expectant management (91.3% vs 81.4%; pooled ratio, 1.3) and were significantly lower in twin pregnancies than in singleton pregnancies (88.2% vs 31.6%; pooled ratio, 2.8). A forest plot of the survival rate is depicted in Figure 3, showing considerable heterogeneity among studies in terms of both I^2 (75%) and a relatively wide 95% prediction interval, both reflecting considerable differences across studies. Forest plots of all secondary outcomes are depicted in Appendix E, showing considerable heterogeneity. Appendix C presents the funnel plot of the primary outcome for studies that contained a control group, not indicative of publication bias and statistically confirmed using the Egger regression test for funnel plot asymmetry.

Sensitivity analyses

We performed 2 sensitivity analyses to assess the effect of publication date (studies published in or after the year 2000) and studies with more than 15 participants on the primary outcome and the heterogeneity. The results of the sensitivity analyses were similar to those of the main analysis. Forest plots of the sensitivity analyses are depicted in Appendix D.

Meta-regression analyses

We performed meta-regression analyses for the primary outcome (survival) and the secondary outcomes (pregnancy prolongation and GA at delivery). No significant association was found between mean cervical dilatation or GA at diagnosis (weeks) and survival. A significant inverse association was observed between mean GA at diagnosis and pregnancy prolongation, in both the overall (slope, −5.5; 95% CI, −9.6 to −1.5) and singleton pregnancy (slope, −6.2; 95% CI, −10.5 to −1.9) groups (Figure 4). To evaluate whether this was strongly influenced by 1 outlier study,[93] we repeated the analysis without this study and found no notable difference. Meta-regression analysis between mean GA at diagnosis and mean GA at delivery resulted in an almost horizontal regression line (slope, 0.1; 95% CI, −0.3 to 0.6) at 30 weeks of gestation, in the overall, singleton, and unspecified groups. No significant association was observed between mean cervical dilatation and pregnancy prolongation. The meta-regression analysis between mean cervical dilatation at diagnosis and GA at delivery in the overall group showed a near-complete horizontal regression line (slope, −0.3; 95% CI, −0.9 to 0.3]) at delivery at 30 weeks of gestation. A table and plots of all performed meta-regression analyses are depicted in Appendix F.

TABLE 5
Pregnancy outcomes after placement of emergency cerclage in singleton and twin pregnancies

Variable	Studies/ participants	Total	Singleton pregnancy (S)	Twin pregnancy (T)	Unspecified	*P* value (S-T)
Obstetrical outcomes						
Survival rate (% of total fetus)	89/2834	74.1 (70.1–77.7)	74.5 (69.8–78.7)	77.7 (55.0–90.8)	71.9 (62.7–79.5)	.70
Viable fetal survival rate (% viable infants of total fetus)	95/3098	87.7 (84.1–90.6)	87.4 (83.1–90.7)	90.8 (68.7–97.8)	87.7 (77.9–93.6)	.59
Neonatal survival (% survived neonates of viable infants)	87/2693	90.0 (86.85–92.4)	90.2 (86.0–93.2)	88.8 (74.6–95.5)	90.0 (82.8–94.4)	.75
Pregnancy prolongation (d)	75/2287	50.6 (45.81–55.4)	52.4 (46.4–58.4)	36.5 (20.0–52.9)	51.1 (41.0–61.2)	.03[a]
GA at delivery (wk)	79/2691	30.0 (29.3–30.6)	30.3 (29.6–31.0)	28.3 (26.4–30.1)	29.6 (28.1–31.1)	.02[a]
Delivery at a GA of ≥34 wk (% of all deliveries)	39/1254	39.3 (32.5–45.6)	46.7 (39.1–54.4)	25.2 (12.9–43.4)	32.1 (18.3–50.0)	.01[a]
Delivery at a GA of <34 wk (% of all deliveries)	39/1254	60.7 (53.6–67.3)	53.3 (46.0–60.6)	74.8 (60.6–85.2)	67.9 (52.0–80.4)	.01[a]
Delivery at a GA of <32 wk (% of all deliveries)	42/1528	56.9 (50.3–63.1)	50.4 (43.7–57.2)	68.8 (53.0–81.1)	70.6 (55.3–82.3)	.04[a]
Delivery at a GA of <28 wk (% of all deliveries)	45/1709	38.6 (32.6–45.0)	34.9 (28.0–42.4)	50.1 (38.1–62.0)	41.3 (27.0–57.3)	.03[a]
Stillbirth at a GA of ≥20 wk (% of total fetus)	52/1570	4.0 (2.1–7.0)	3.1 (1.4–6.6)	1.7 (0.0–72.3)	9.1 (4.5–17.6)	.41
Miscarriage at a GA of <20 wk (% of total fetus)	55/1725	2.3 (1.2–7.0)	3.5 (1.9–6.4)	0.8 (0.0–54.1)	1.3 (0.3–5.8)	.04[a]
Neonatal outcomes						
Birthweight (g)[b]	56/1924	1696.2 (1569.6–1822.9)	1742.8 (1581.9–1903.6)	1471.1 (1183.0–1759.1)	1686.4 (1380.0–1992.8)	.06
NICU admission (%)	37/1240	65.2 (52.1–73.3)	63.1 (34.1–82.2)	77.3 (44.8–93.4)	60.7 (34.1–82.2)	.30
Sepsis (%)	10/362	8.9 (6.0–13.1)	12.5 (3.1–38.9)	4.1 (0.0–100.0)	9.7 (4.1–21.4)	.29
Respiratory distress syndrome (%)	8/279	26.0 (10.1–52.2)	47.1 (13.3–83.8)	44.8 (0.0–100.0)	7.7 (0.7–51.0)	.90
Necrotizing enterocolitis (%)	6/116	4.6 (1.5–13.5)	10.0 (0.8–60.4)	0.0 (0.0–100.0)	3.9 (0.0–99.7)	.99
Retinopathy of prematurity (%)	5/98	17.0 (3.9–51.1)	16.7 (2.3–63.1)	3.6 (0.5–21.4)	26.5 (2.7–82.6)	.26
Intraventricular hemorrhage (%)	5/134	9.8 (2.6–30.4)	10.3 (0.0–99.6)	3.6 (0.5–21.4)	13.9 (0.0–99.9)	.34

Data are presented as proportion (95% CI) or mean (95% CI), unless otherwise specified. Unspecified indicates data that were not reported separately for singleton and twin pregnancies. In each column, not all studies are represented, depending on available data.

Quality of the evidence (GRADE): ⊕⊕⊖⊖ (observational studies).

CI, confidence interval; *GA*, gestational age; *NICU*, neonatal intensive care unit.

[a] A *P*-value of ≤ 0.05 was considered statistically significant; [b] Birthweight of all born neonates.

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Comment

Main findings

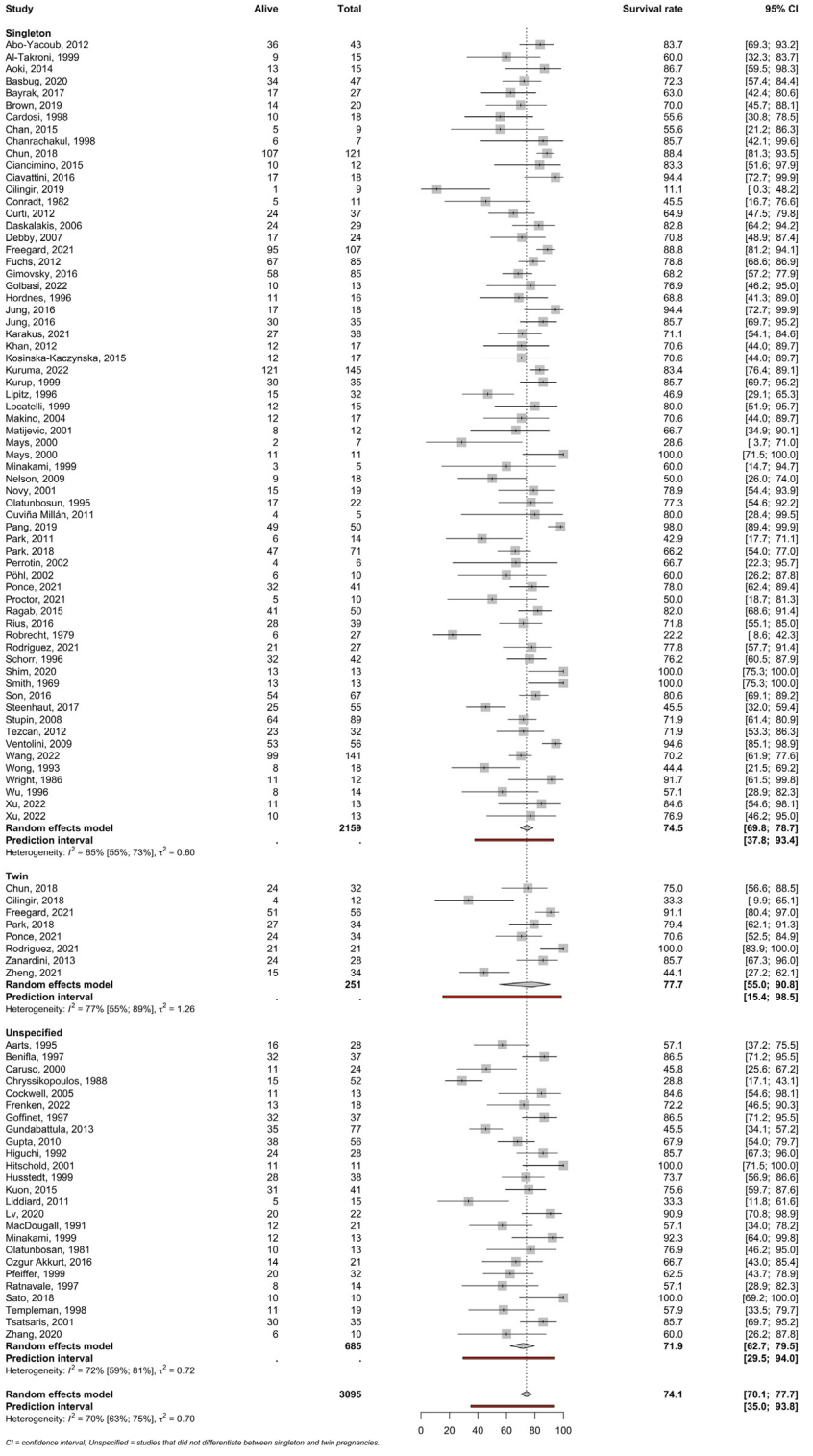
The prevention of extreme PMB is of utmost importance, as this is accompanied by high fetal mortality and morbidity. To evaluate the success of placement of a transvaginal emergency cerclage, we performed a systematic review with meta-analysis in women with threatening extreme PMB based on cervical insufficiency. We included 96 studies, consisting of 3239 pregnancies. We observed that an emergency cerclage relates to much an improved and relatively good survival of 74%, with a viable fetal survival of 88% and a neonatal survival of 90%, accompanied by both a significant pregnancy prolongation as well as reduction in deliveries at <28 weeks of pregnancy compared to expectant management. Survival was comparable between singleton and twin pregnancies, although twin pregnancies intend to deliver two weeks earlier and more often below the 34, 32 and 28 weeks of gestation than singletons. This suggests that placement of an emergency cerclage should be offered in both singleton and twin pregnancies when threatened by cervical insufficiency.

Comparison with existing literature

Cervical insufficiency impairs fetal and neonatal survival because of extreme prematurity. Previous reviews

FIGURE 2

Overall survival after placement of emergency cerclage in singleton and twin pregnancies



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concluded that emergency cerclages are associated with considerably improved outcomes.[3,11,13] We found that the survival rate in pregnancies treated with transvaginal emergency cerclage is 74%, and fetal and neonatal survival rates are similar in singleton and twin pregnancies. This is comparable to the rates reported by other systematic reviews.[11,12] Compared with expectant management, we found that emergency cerclages show a significant increase in survival rates from 28% to 71% (pooled ratio, 1.7). These results are in line with the findings of Ehsanipoor et al,[11] who observed an increase in survival rates from 43% to 72% (pooled ratio, 1.7). Therefore, it can be concluded that an emergency cerclage, as opposed to expectant management, increases fetal and neonatal survival rates tremendously.

Neonatal quality of life is strongly affected by the duration of pregnancy. We observed that an emergency cerclage prolongs the pregnancy on average by 51 days, with a mean GA at delivery of 30 weeks. This is comparable to several other systematic reviews that found a pregnancy prolongation between 47 and 56 days.[11,12,14] Compared with expectant management, we observed that an emergency cerclage significantly prolongs the pregnancy with 51 days instead of 13 days, with a mean GA at delivery of 30 weeks, as opposed to 25 weeks. We found deliveries at <28 weeks of gestation to be significantly reduced from 93% to 38% (pooled ratio, 0.5). We observed a significant inverse association between GA at diagnosis and pregnancy prolongation and no association between cervical dilatation or GA at diagnosis and GA at delivery. These findings suggest that an emergency cerclage decreases not only the mortality rate but also the morbidity rate, by prolonging the duration of pregnancy and reducing the rate of extreme PMB.

It seems plausible that the success rate of emergency cerclages depends on the pregnancy type (singleton or twin), as the etiology and, therefore,

TABLE 6

Pregnancy outcomes after placement of emergency cerclage vs expectant management

Variable	Studies/ participant	Emergency cerclage			Expectant management			Effect estimate[a]		
		Total	Singleton	Twin	Total	Singleton	Twin	Total	Singleton	Twin
Survival rate (% of total fetus)	14/746	70.9 (61.6–78.8)	73.0 (65.1–79.7)	44.1 (28.6–60.8)	27.9 (11.9–56.9)	35.5 (16.1–61.2)	2.6 (0.4–16.5)	1.7 (1.2–2.4)[b]	1.6 (1.2–2.1)[b]	16.8 (2.3–120.3)[b]
Fetal survival rate (% viable infants of total fetuses)	13/709	81.7 (67.2–90.7)	86.4 (71.8–94.1)	52.9 (36.5–68.8)	64.4 (30.5–88.2)	69.4 (33.3–91.1)	69.4 (33.3–91.1)	1.4 (1.1–1.8)[b]	1.3 (1.0–1.7)[b]	3.4 (1.5–7.5)[b]
Neonatal survival (% survived neonates of viable infants)	13/709	92.9 (84.6–96.9)	92.7 (83.9–96.9)	83.3 (59.1–94.5)	70.7 (29.5–93.3)	76.3 (32.3–95.6)	16.7 (2.3–63.1)	1.4 (1.0–1.8)[b]	1.3 (1.0–1.6)[b]	5.0 (0.8–30.3)[b]
Pregnancy prolongation (d)	13/709	51.3 (41.1–61.5)	54.1 (43.6–64.5)	23.7 (16.0–31.5)	12.6 (9.3–15.9)	13.8 (10.0–17.5)	4.5 (2.9–6.2)	36.5 (27.7–45.3)[b]	38.4 (31.3–45.6)[b]	19.2 (11.3–27.1)[b]
GA at delivery (wk)	12/677	30.1 (29.1–31.1)	30.5 (29.6–31.4)	26.0 (24.2–27.8)	25.1 (23.8–26.3)	25.4 (24.0–26.7)	22.0 (20.7–23.3)	4.7 (4.0–5.5)[b]	4.8 (3.9–5.8)[b]	4.0 (1.8–6.2)[b]
Delivery at a GA of ≥34 wk (% of all deliveries)	3/119	49.3 (9.8–89.7)	74.5 (60.2–84.9)	5.9 (0.8–32.0)	8.9 (0.2–82.6)	33.3 (2.2–91.8)	0.0 (0.0–100.0)	2.8 (0.1–101.1)	2.8 (0.0–9872626.1)	12.3 (0.0–7697.6)
Delivery at a GA of <34 wk (% of all deliveries)	3/119	50.7 (10.23–90.2)	25.5 (15.1–39.8)	94.1 (68.0–99.2)	91.1 (17.4–99.8)	66.7 (8.2–97.8)	100.0 (0.0–100.0)	0.6 (0.1–2.8)	0.4 (0.0–59.5)	0.9 (0.8–1.0)
Delivery at a GA of <32 wk (% of all deliveries)	3/106	59.9 (30.4–83.6)	45.2 (31.0–60.3)	88.2 (63.2–97.0)	96.0 (74.8–99.5)	92.9 (75.5–98.2)	100.0 (0.0–100.0)	0.6 (0.2–1.7)	0.5 (0.1–3.0)	0.9 (0.7–1.1)
Delivery at a GA of <28 wk (% of all deliveries)	4/143	38.2 (20.9–59.2)	30.0 (19.8–42.7)	70.6 (45.8–87.2)	93.0 (19.1–99.9)	75.7 (13.1–98.5)	100.0 (0.0–100.0)	0.5 (0.3–1.0)[b]	0.4 (0.2–1.3)	0.7 (0.5–1.0)[b]
Stillbirth at a GA of ≥20 wk (% of total fetus)	4/207	2.6 (0.4–15.4)	2.6 (0.4–15.4)	NA	0.5 (0.0–34.7)	0.5 (0.0–34.7)	NA	1.4 (0.0–100.0)	1.4 (0.0–100.0)	NA
Miscarriage at a GA of <20 wk (% of total fetus)	5/259	6.0 (2.4–14.6)	6.0 (2.4–14.6)	NA	7.67 (0.6–51.3)	7.67 (0.6–51.3)	NA	0.4 (0.0–3.1)	0.4 (0.0–3.1)	NA
Birthweight (g)[c]	6/291	1763.2 (1389.1;2138.1)	1851.8 (1404.8–2298.7)	1281.1 (1127.1–1435.1)	843.9 (729.1–958.7)	853.8 (705.2;1002.5)	858.3 (775.0–941.6)	807.1 (471.2–1143.0)[b]	909.5 (470.4–1348.5)[b]	422.8 (247.7–597.9)[b]
NICU admission (%)	3/182	91.3 (17.1–99.8)	96.2 (0.8–100.0)	88.2 (63.2–97.0)	81.4 (7.2–99.6)	96.6 (1.9–100.0)	31.6 (14.9–54.8)	1.3 (0.2–7.0)	1.0 (0.3–3.4)	2.8 (1.4–5.5)[b]
Sepsis (%)	1/36	11.1 (1.4–34.7)	NA	11.1 (1.4–34.7	33.3 (8.4–73.2)	NA	33.3 (8.4–73.2)	0.3 (0.1–1.9)	NA	0.3 (0.1–1.9)
Intraventricular hemorrhage (%)	1/36	11.11 (1.4–34.7)	NA	11.11 (1.38–34.7)	16.7 (0.4–64.1)	NA	16.7 (0.4–64.1)	0.7 (0.1–6.1)	NA	0.7 (0.1–6.1)

Data are presented as proportion (95% CI) or mean (95% CI), unless otherwise specified. In each column, not all studies are represented, depending on available data.

Quality of the evidence (GRADE): ⊕⊕⊖⊖ (observational studies).

CI, confidence interval; *GA*, gestational age; *NA*, not applicable; *NICU*, neonatal intensive care unit.

[a] Data are presented as pooled ratio (95% CI) for rows 2 to 5, 7 to 11, and 13 and mean difference (95% CI) for rows 1, 6, and 12; [b] Statistical significance; [c] Birthweight of all born neonates.

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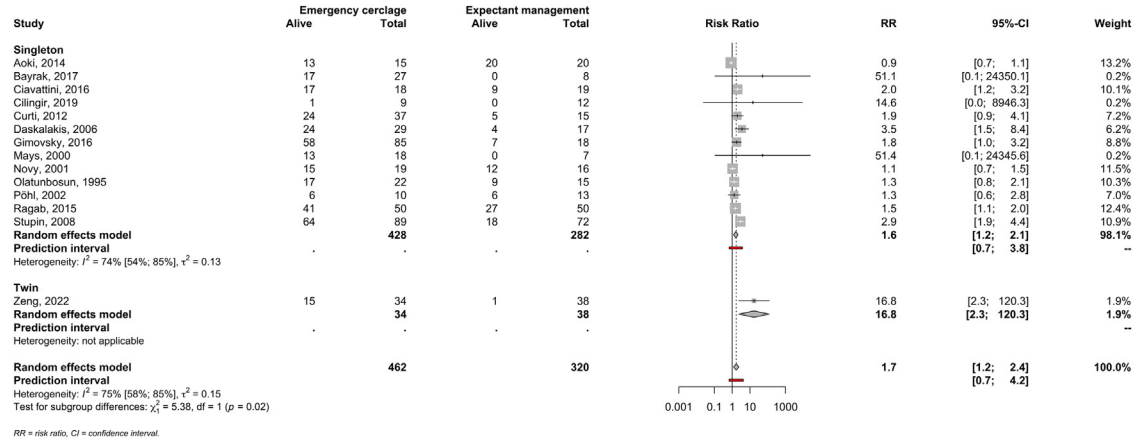
FIGURE 3
Overall survival after placement of emergency cerclage vs expectant management



CI, confidence interval; *RR*, relative risk.

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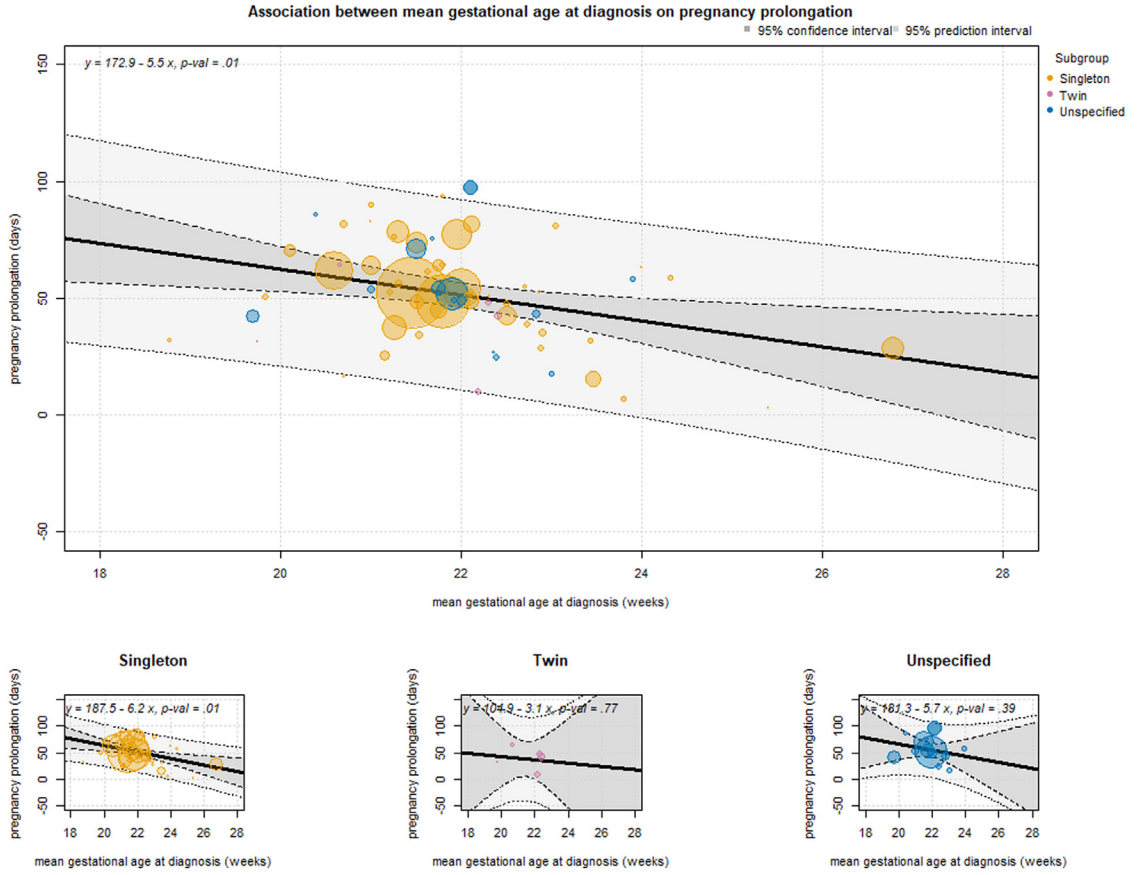
FIGURE 4
Association between pregnancy prolongation and GA at diagnosis based on meta-regression analysis



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management of cervical insufficiency in these pregnancies might differ. In our meta-analysis, we included both singleton and twin pregnancies and analyzed them separately and combined. This is in contrast to previous meta-analyses, where only singleton or twin pregnancies were studied or no distinction was made in the analyses.[11−14,17,18] We observed a survival rate of approximately 74% for both singleton and twin pregnancies. GA at delivery for singleton pregnancies was 30 weeks with a mean prolongation of 52 days, whereas the mean GA at delivery of twin pregnancies was 28 weeks with a mean prolongation of 37 days. These outcomes are in line with previous systematic reviews.[17,18] Although the quantity of studies on emergency cerclage in twin pregnancies is limited, our results suggest that survival in twin pregnancies after an emergency cerclage is similar to or slightly diminished compared with singleton pregnancies.[16,18,45] However, one has to take into account that there is only 1 study that compared emergency cerclage with expectant management in twin pregnancies, in 17 women with a ≥4-cm cervical dilatation. This has likely affected the relatively disappointing outcomes of emergency cerclage in twin pregnancies compared with expectant management in twin pregnancies. The results from this study have to be interpreted with this in mind. The above suggests that an emergency cerclage should be offered not only in singleton pregnancies but also in twin pregnancies threatened by cervical insufficiency.

Strengths and limitations

Our study has several strengths. The main strength of our review is that it combines the results of a large number of small studies and, therefore, can lead to important conclusions. In addition, we divided included pregnancies into different groups for subanalyses, which could reduce the effect of confounders. Moreover, meta-analyses on retrospective observational data are prone to heterogeneity in outcome measures. We aimed to reduce clinical heterogeneity by applying strict inclusion criteria. For instance, management and clinical consequences might depend on the degree of dilatation and/or presence of prolapsed membranes beyond the external cervical os. Therefore, we only included studies with data from women with a ≥2-cm cervical dilatation and/or prolapse of membranes at or beyond the external os. Furthermore, although the quality of study methods and reporting of these studies varied widely, the results of the meta-analyses were consistent for the primary outcome. Naturally, this review also has some potential methodological issues that need to be addressed. First, most included studies had retrospective observational and not prospective randomized control designs. This might have introduced selection and publication bias, although funnel plot analyses were hardly indicative of publication bias. Nevertheless, our results should be interpreted with some caution. Second, because of nonstandardized treatment protocols and outcome measures, our results suffered from clinical and methodological heterogeneity. This was shown in forest plots of our results, which displayed substantial heterogeneity. Sensitivity analyses on recent studies (published in or after the year 2000) and on studies with more than 15 participants confirmed the main findings concerning survival and heterogeneity. However, as our analysis depends on available data in the literature, we were not able to investigate the effect of other factors, including uterine contractions, abdominal pain, vaginal blood loss, fever, leucocyte number, and C-reactive protein. This might have contributed to selection bias and heterogeneity among included studies. Moreover, the degree of dilatation and timing of intervention at a different GA might have influenced the outcomes. This might have a considerable influence on variation in success rates. The significant difference in the degree of dilatation at diagnosis between singleton and twin pregnancies might explain the slightly higher survival rates in twin pregnancies, in contrast to most other studies where similar or slightly lower survival rates were observed. Moreover, this applies to the subanalysis on studies that contained both intervention and expectant management groups, where lower survival and pregnancy prolongation in twin pregnancies might be explained by the higher degree of dilatation in the only twin study. The analysis of individual participant data would be needed to overcome these issues. Third, some conclusions in this study might be influenced by the small number of studies available for certain subanalyses. This might have especially affected our results on emergency cerclage in twin pregnancies, as we only came across 9 studies. This might have led to imprecise outcome measures. Our findings need to be seen from this perspective. Fourth, reporting in included studies was poor for several baseline characteristics and secondary outcomes. Conclusions might be affected by the lack of this information.

Conclusions and implications

The efficacy of emergency cerclage in both singleton and twin pregnancies has generated much debate within the field of cervical insufficiency. Our systematic review with meta-analysis gives insight into available data and, although results should be interpreted with caution, indicates that, in both singleton and twin pregnancies, threatened by cervical insufficiency, an emergency cerclage should be offered. Moreover, our study underscores the need for studies that take individual patient characteristics into account and directly compare different treatment options for acute cervical insufficiency, although randomized comparison might have ethical limitations. Our data can be used in counseling couples that have a pregnancy threatened by cervical insufficiency, so they can make their decision based on the best available evidence.

Our systematic review with meta-analysis indicates that placement of an emergency cerclage in women that suffer from threatened extreme premature birth because of cervical insufficiency leads to significantly higher survival rates, accompanied by a significant pregnancy prolongation, increase in birthweight, and reduction in deliveries

at <28 weeks of gestation, in both singleton and twin pregnancies. After placement of the cerclage, the mean GA at delivery is 30 weeks, independent of dilatation and GA at diagnosis. Therefore, we propose that an emergency cerclage should be considered in both singleton and twin pregnancies for this indication. This study provides relevant results that should be used in the counseling of couples. ■

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Supplementary materials

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