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Article type : Systematic review

Worldwide prevalence of tocophobia in pregnant women: systematic review and meta-analysis

Short title: Worldwide prevalence of tocophobia

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Abstract

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Introduction: Tocophobia is defined as a severe fear of pregnancy and childbirth. There is increasing evidence that tocophobia may have short and long-term adverse effects on mother and baby. We performed a systematic review and meta-analysis to determine the global prevalence of tocophobia in pregnancy. *Material and methods:* Relevant articles were identified through searching six relevant databases: MEDLINE, CINAHL, Pubmed, PsycINFO, Maternity & Infant Care and Scopus between 1946 and April 2016. We used search terms for tocophobia prevalence in pregnant women which we agreed with a medical librarian. There were no language restrictions. Two review authors independently assessed data for inclusion, extracted data and assessed quality using a standardized appraisal tool. Meta-analysis was performed to determine the overall pooled-prevalence of tocophobia. Several subgroup and sensitivity analysis were conducted. *Results:* Thirty-three studies were included in the systematic review from 18 countries of which data from 29 studies were used in the meta-analysis of 853,988 pregnant women. Definition of tocophobia varied, while prevalence rates ranged between 3.7% and 43%. The overall pooled prevalence of tocophobia, using a random-effects model, was 14% (95% CI; 0.12-0.16). Significant heterogeneity was observed *(I^2=99.25%, p=0.00*) which was not explained in subgroup analyses including tocophobia definition used, screening trimester and parity. *Conclusion:* The prevalence of tocophobia is estimated at 14% and appears to have increased in recent years (2000 onwards). Considerable heterogeneity (99.25%) was noted which may be attributed to lack of consensus on the definition of tocophobia therefore our results should be interpreted with caution.

Keywords

Tocophobia, pregnancy, systematic review, epidemiology, fear of childbirth, W-DEQ A

Abbreviations

FOC fear of childbirth

CI: confidence interval

FOBS: Fear of Birth Scale

CAQ Childbirth Attitudes Questionnaire

W-DEQ A: Wijma Delivery Experience Questionnaire Part A

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Key message

Definitions of tocophobia vary widely. This meta-analysis estimated a global pooled-prevalence of 14%, however this should be interpreted with caution due to significant heterogeneity. This is the first systematic review of the prevalence of tocophobia which affects a significant minority of women.

Introduction

Over the last 30 years, there has been increasing interest in tocophobia (severe fear of childbirth) both in empirical research and clinical practice (1-5). Tocophobia has been defined as "an unreasoning dread of childbirth", and further classified into primary (affecting nulliparous women) and secondary (affecting parous women usually after a previous birth experience) tocophobia (6, 7). There is however, no one agreed definition of tocophobia and much of the published literature to date refers to tocophobia as a severe "Fear of Childbirth (FOC)" rather than "an unreasoning dread of childbirth" (8, 9). Factors including anxious personality types, previous sexual abuse, past traumatic birth or any traumatic experience in health care, previous miscarriages, long duration of infertility, smoking, low social supports and poor partner relationships have been associated with primary and secondary tocophobia (9-13).

While there are no standard criteria for defining tocophobia, the Wijma Delivery Expectancy Questionnaire Part A (W-DEQ A) is the most commonly used tool for assessment and diagnosis (14, 15). Other tools include the Fear of Birth Scale (FOBS) and Childbirth Attitudes Questionnaire (14, 16, 17). The FOBS is a Visual Analogue Scale consisting of two questions, developed to encourage compliance in completion of the questionnaire due to the length of the W-DEQ A (consisting of 33 questions). The prevalence of tocophobia has also been reported by analysis of the International Classification of Diseases 10th Revision codes, assigned to women who attended tocophobia clinics in countries where care pathways are well established (9, 18).

It is reported that 6-10% of pregnant women suffer with FOC that affects everyday life (1, 18-20). However, lack of consistency in defining tocophobia has led to variation in prevalence reports (21, 22). Therefore, the aims of this systematic review were to:
1) Assess how 'tocophobia' is defined in the literature and 2) provide the first quantitative pooled estimate of the prevalence of tocophobia in pregnant women by synthesizing the data from eligible studies (where feasible) in a meta-analysis.

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Material and methods

The review adheres to the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) guidelines (23) and has been registered on the International prospective register of systematic reviews [PROSPERO ID: CRD42015017443] (24).

Sources

Six electronic databases (PubMed, CINAHL, PsycINFO, Maternity & Infant Care, Scopus and MEDLINE) were searched for all published literature up until April 11th 2016 using a detailed search strategy and without date or language restrictions (Supporting Information Appendix S1). Medical subject headings or keyword terms for tocophobia during pregnancy were combined according to the principles of Boolean logic including: "*tocophobia", "fear of childbirth", "fear of labour", "fear of birth", "childbirth related fear", "childbirth related anxiety", "fear in pregnancy", "antenatal" and "childbirth"*.

Study Selection

Published observational studies including pregnant women of any age and origin and reporting the prevalence of tocophobia (or sufficient data in order for us to compute this estimate) were eligible for inclusion. Two researchers (MOC and SMON) independently reviewed study titles and abstracts applying the inclusion/exclusion criteria. Full-text studies were obtained where required and where consensus was not reached, a third reviewer (PLW) ensured agreement. Reference lists of eligible studies were hand searched for further potentially eligible studies. The following data were abstracted from each study using a standardized form by two reviewers (MOC, SMON): Author, year, study location (country), study design, scale used, sample size, and prevalence. If it was considered that a study had collected data on the prevalence of tocophobia but had not reported it, the authors were contacted for this information.

Quality assessment

Quality assessment of each study was independently evaluated by two reviewers (MOC and SMON) using a standardized tool including eight questions to assess bias (25) (Supporting Information Appendix S2) pertaining to the following criteria: target population, sampling ascertainment methods, response rate, information on non-responders, if the sample was representative, data collection methods, use of a validated tool for tocophobia, and prevalence

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with 95% CIs. Each study received a score of between 0 and 8 points, based on meeting the prescribed criteria as agreed by the reviewers. High quality studies were defined as those receiving a score of 5 or more out of 8.

Statistical analyses

Search results were compiled in EndNote Reference Manager Version X7 (Clarivate Analytics, New York, NY, USA). Characteristics of the included studies were summarized and presented in Table 1. For the meta-analysis, an overall pooled prevalence was calculated using the sample size and the proportion of women with tocophobia and the fixed or random-effects model as appropriate. Using the *metaprop* command, we generated pooled proportions and an overall pooled estimate with inverse variance weights derived from a random-effects model (26) in STATA software Version 13.1 (StataCorp, Collega Station, TX, USA,).

Subgroup and sensitivity analyses

A priori sensitivity analyses included: studies with a W-DEQ A ≥85 for tocophobia, by parity (nulliparous women only, multiparous women only), by screening trimester (first trimester, second trimester, third trimester). *A priori* subgroup analyses included: by study quality (high versus low), by region (Scandinavia versus Rest of Europe versus Australia versus America versus Asia), and by time period (1980s versus 1990s versus 2000-2009 versus 2010-2016).

Heterogeneity assessment

Heterogeneity between studies was assessed by examining the study characteristics presented in Table 1. In addition, the I^2 statistic was used to determine statistical heterogeneity according to the Cochrane Handbook for Systematic Reviews threshold recommendations (27) . For this meta-analysis, where heterogeneity was greater than 50% the random-effects model was used (28).

Results

Results of the systematic search are presented in Figure 1, which yield 33 studies eligible for inclusion in the systematic review (29). Twenty-four high quality studies and five low quality studies were included in the meta-analysis. Where there was more than one publication on a cohort of patients (i.e. the same population), data on the prevalence of tocophobia were taken from those that described the total population rather than a subset.

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Study characteristics

Study characteristics are presented in Table 1. One study was published in 1981 (30), one study in the late 1990s (31), fourteen studies between 2000 and 2009 (15, 18, 19, 21, 32-41) and seventeen studies between 2010 and April 2016 (1, 8, 9, 16, 20, 42-53). Study settings included the following: USA (32), Canada (37), Australia (47, 54, 55), Sweden (15, 16, 19, 30, 31, 33, 38, 40, 41, 55-57), Norway (20, 35, 44, 46), Finland (9, 21), Switzerland (34), Denmark (18, 19), Italy (1), Turkey (8), Iran (52), China (49), Japan (53), South India (51) and the Netherlands (45). One study was conducted across six countries- Belgium, Iceland, Denmark, Estonia, Norway and Sweden (43). Study population sizes ranged from 105 to 788,317 (9, 45). One study included multiparous women (8), seven studies included nulliparous women and 25 studies were not restricted by parity.

Definition of tocophobia

Tocophobia was defined using a variety of measures and cut-offs. The majority (21/33 studies (1, 2, 8, 15, 19, 20, 31, 33, 35-39, 41, 43-45, 53, 58-60)) used the W-DEQ Part A to assess tocophobia [of which three studies (20, 44, 60) used the same cohort], meaning that 19 different cohorts in this review used the W-DEQ Part A as a tool to assess tocophobia. While the majority of included studies used W-DEQ Part A, only a minority of the total study population (21,619/ 853,988) were assessed with this tool. Other methods used to define tocophobia included the FOBS [3 studies (16, 55, 59)], Childbirth Attitudes Questionnaire (CAQ) [3 studies (17, 32, 49)] and International Statistical Classification of Diseases and Health Related problems 10th Revision [1 study (9)] (Table 1). A Finnish study comprised the largest study population (n=788,317) which reported the prevalence of tocophobia based on an International Statistical Classification of Diseases and Health Related problems 10th Revision Code allocated to all women who attended tocophobia clinics during the period of the study (9). In addition, tocophobia was measured using phone interviews with pre-defined standardized questions, face–to-face interviews using standardized questions or self-reported questionnaires completed in the clinic or returned via post (18, 30, 40, 51, 52). Sampling was done in different languages, and in the case of standardized instruments (W-DEQ A, FOBS, CAQ) the studies ensured correct translation of the questionnaires in the following ways which varied according to study: the questionnaire was translated into the most commonly spoken languages of the study area (forward translation); the various language versions of the questionnaire were translated by both lay and professional translators (expert back translation); draft versions of the translated questionnaire were assessed for accuracy and

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validated by professionals who were fluent in one or more of the languages (pre-testing) (46, 51, 61, 62). One study (53) was the first to use the W-DEQ A in the Japanese language and thus needs to be validated in further studies.

Of the 21 studies that used the W-DEQ Part A, two used ≥100 as a cut-off for tocophobia (35, 59), one used ≥95 (35), one used ≥85.8 (41), 12 used ≥85 (8, 19, 20, 33, 38, 39, 43, 44, 46, 47, 56, 63), one used ≥84 (31), one used ≥71 (36) and two used ≥66 (15, 64). Studies that used the FOBS estimated a much higher prevalence estimate (double the other prevalence estimates) than the other studies included in the review. Regarding screening trimester, four studies questioned women in all trimesters (9, 30, 43, 56), twelve studies recruited women in the second trimester (12-27 weeks) (2, 16, 18, 30, 34, 35, 37, 40, 43, 50, 52, 58) and 17 studies recruited women in the third trimester (28-41 weeks) (1, 8, 15, 18-20, 30-33, 36, 37, 39, 41, 43, 51, 63). Of these studies, one recruited in both the second and third trimesters (18). Data on the prevalence of tocophobia were available for two population-based (9, 18) and 31 hospital-based cohorts of pregnant women.

Quality assessment

Study quality was assessed independently by two reviewers (MOC, SMON). While there was variation in the quality of the studies, overall quality was considered high [26/33 studies with a score of 5 or more out of 8] (Table 1). Seven studies were considered low quality (a score of ≤4 out of 8) due to the following: the target population was not clearly defined, the response rate was not reported, information on non-responders was not provided or the sample selection was unclear or not reported or did not use validated tools for tocophobia.

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Prevalence of tocophobia - meta-analysis

Of the 33 studies included in the systematic review, data from 28 studies were included in the meta-analysis. One study (50) included two cohorts from Australia and Sweden which we split into two studies for the purpose of the meta-analysis, (Haines 2011a, and Haines 2011b), resulting in 29 studies in total. A fixed-effects model yielded a 4% (95% CI; 0.04-0.04) prevalence of tocophobia in pregnant women. Due to significant heterogeneity *(I^2=99.5%, p<0.0001)*, a random-effects model was used and a pooled prevalence of 14% (95% CI; 0.12-0.16) for tocophobia, with considerable heterogeneity *(I^2 = 99.25%)* (Fig. 2) was obtained.

Sensitivity Analyses

W-DEQ A ≥85

The twelve studies which used a W-DEQ A score of ≥85 as the definition of tocophobia detected a pooled prevalence of 12% (95% CI; 0.09-0.14) and significant heterogeneity *(I^2 = 95.41%, p=0.00)* using the random-effects model (Fig. 3).

Parity

Studies including nulliparous women (Fig. 4), yielded a pooled prevalence of 16% (95%CI; 0.14-0.19) with significant heterogeneity *(I^2=99.42%, p=0.00). S*tudies including multiparous women (Fig. 5), resulted in a pooled prevalence of 12% (95% CI; 0.10-0.14) and significant heterogeneity *(I^2=97.81%, p=0.00).*

Screening Trimester

In one study women were screened in the first trimester of pregnancy and was not included in a sensitivity analysis (61). Studies which screened women in the second trimester (Fig. 6), yielded a pooled prevalence of 14% (95% CI; 0.12-0.16) and significant heterogeneity remained *(I^2=98.1%, p=0.00).*Studies which screened in the third trimester yielded (Supporting Information Figure S1), a pooled prevalence of 12% (95% CI; 0.10-0.14), with significant heterogeneity *(I^2=97.78%, p=0.00).*

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Sub-group analyses

Study Quality

The prevalence of tocophobia in the high quality studies was 13% (95% CI; 0.11-0.15) *(I^2 = 99.3%, p=0.00)* compared to 19% (95% CI; 0.08-0.30) *(I^2 = 97.96%, p=0.00)* in the low quality studies (Supporting Information Figure S2).

By Region

The prevalence of tocophobia found in Scandinavia was 12% (95% CI; 0.09-0.15) *(I^2 = 99.51%, p=0.00)* (Supporting Information Figure S3). In the Rest of Europe the prevalence was 8% (95% CI; 0.04-0.13) *(I^2 = 99.51%, p=0.00),* in Australian studies the prevalence was 23% (95%CI; 0.07-0.39) *(I^2 = 98.63%, p=0.00)*, in American studies the prevalence was 11% (95% CI; 0.03-0.20) *(I^2 = 92.97%, p=0.00)* and in Asian studies the prevalence was 25% (95% CI; 0.11-0.40) *(I^2 = 97.69%, p=0.00).*

By Time Period

One study looked at the prevalence of tocophobia in the 1980s, which was 6% (95% CI; 0.03-0.12) (Supporting Information Figure S4). Prevalence of tocophobia was reported by one study in the 1990s at 10% (95% CI; 0.09-0.11). Fourteen studies between 2000 and 2009 examined the prevalence of tocophobia which was 12% (95% CI; 0.10-0.15) *(I^2=98.18%, p=0.00)*, and 13 studies conducted between 2010 and 2016 resulted in a pooled prevalence of 17% (95% CI; 0.13-0.21) *(I^2=98.98%, p=0.00).* Overall heterogeneity was highly significant *(I^2= 99.26%, p=0.00).*

Studies not eligible for inclusion in the meta-analysis

Three studies (17, 49, 53) did not include data that could be included in the meta-analysis and two studies (44, 46) included the same population as a third study (20). A brief summary of the studies not included in the meta-analysis are presented in Table 2.

Discussion

To our knowledge, this is the first systematic review and meta-analysis of the prevalence of tocophobia in pregnant women. Overall, the pooled prevalence of tocophobia was 14%. Subgroup analyses according to region showed a significant difference in the prevalence of tocophobia. For example in Scandinavia the prevalence was 12% compared to 8% in the rest of Europe and 23% in Australia. Furthermore when we looked at the prevalence of tocophobia by time period, it was lower in the earlier years (1980s, 1990s) but increased in the more recent years (2000 onwards). However, our findings need to be interpreted with

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caution since significant heterogeneity was found *(I^2=99.25%, p=0.00)*. Extensive pre-specified subgroup and sensitivity analyses did not explain the significant heterogeneity in the meta-analysis. Differences in the way studies were conducted and information collected and recorded as well as variations in the social and cultural characteristics of women included in these studies may explain the heterogeneity (28).

There has been conflicting evidence as to the prevalence of tocophobia in nulliparous and multiparous women (61, 65). We carried out a subgroup analysis which identified that tocophobia was more prevalent in nulliparous women (who have never experienced childbirth before), this is similar to the findings of nine previous studies (1, 2, 15, 16, 20, 36, 37, 43, 50).

The results of our study are clinically relevant in the following respects. Firstly, we identified there is a lack of a clear operational definition for tocophobia. Although tocophobia has become a term commonly used to describe severe FOC, a clear, consistent operational definition is lacking (9, 66). This was reflected in the literature where several tools were used to assess FOC and tocophobia (Table 1). The W-DEQ A questionnaire was employed in nineteen studies, and although there is a recommended cut-off point for the definition of tocophobia (≥85), some studies used different cut-off points (1, 15, 31, 67). Terms used included 'high childbirth related fear', 'intense fear', 'high childbirth fear', 'severe childbirth fear' or 'severe FOC' (8, 16, 30, 34, 45, 49, 52). It is important to recognise that it may be normal for pregnant women to have worries (34, 36, 42, 68) (recurrent but unspecific thoughts) since birth is unpredictable, however fears can be strong, specific and continuous (68). It has been suggested that when a woman expresses FOC during pregnancy and requests support, this could be in itself a definition (66). Secondly, this is the first time a pooled-prevalence has been calculated for tocophobia giving an indication of the overall burden for public health. Moreover, our study revealed an apparent increase in the prevalence of tocophobia over the last thirty years. Therefore, our results highlight the need for clinicians and the healthcare service to be aware of and encourage women to express FOC since identifying women with tocophobia early in pregnancy may provide an opportunity to support maternal mental health (3, 22, 55). This is important as there is growing evidence linking tocophobia with increased maternal cortisol levels as well as the exacerbation of other mental health issues, which may lead to serious and long-term consequences for mother and baby (65).

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Tocophobia is difficult to quantify. Currently, the W-DEQ A is used as the 'gold standard' for assessment and 'on the spot' diagnosis (14, 69). As mentioned, we found a variation in the cut-off point used for the W-DEQ A. A criticism of this tool has been that it may exclude some women who could benefit from support, therefore some studies used a slightly lower cut-off point (66 or 71 rather than 85) (15, 36), resulting in more referrals for intervention. Moreover, an in-depth psychometric analysis of the W-DEQ A advised that calculating a total score and using a cut-off to define tocophobia may not be appropriate as this is based on the premise that the W-DEQ A is uni-dimensional (36, 70-73). The use of subscales has been advocated to determine specific reasons behind the woman's fear and identify risk factors which might make a woman more vulnerable such as lack of social support (70). In addition to the issues outlined above, the W-DEQ A is lengthy and impractical for clinical use therefore researchers are striving to establish more practical tools (14, 22).

The FOBS (a two question Visual Analogue Scale) is deemed a feasible tool used to prompt referral in clinical practice (22, 68) and has been validated in samples of Swedish and Australian populations (sensitivity (89%) and specificity (79%)) (22, 55). It is argued there is likely to be high compliance as it is easily understood (22). Screening for FOC is suggested in order to offer appropriate referral as there is evidence that women may benefit if offered timely antenatal support (2, 3, 22, 74, 75). However, similarly to the introduction of other screening assessments to the antenatal booking appointment, it may be envisaged that time constraints in the clinic and lack of clear referral pathways may be barriers to the effectiveness of this tool (76-78).

There is considerable evidence endorsing the need for improved perinatal psychological support in maternity services (3-5, 65, 79, 80). Reasons for tocophobia may be complex (3, 33, 81) and include lack of trust in or worries about unfriendly staff (36), being left alone in labour, appearing silly and lack of involvement in decision-making (13, 32, 40) as well as trauma and previous sexual abuse. In addition, FOC often coincides with depressive and compulsive personalities predisposing women to postnatal depression and Post Traumatic Stress Disorder (3, 35, 79, 82). Thus, various strategies have been proposed to help women cope with FOC i.e. psycho-education, birth preparation (2, 68). There is evidence that continuity of care, developing meaningful, trusting relationships, involving women fully in decision-making and working in partnership to provide woman-centered care can improve outcomes (75, 83-85) but there is no standardized care pathway for women with tocophobia in pregnancy (80). Future researchers could strive to develop appropriate

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interventions aimed at identifying pregnant women at risk of tocophobia, such as decision aids which are increasingly being used in healthcare settings (86).

This comprehensive systematic review was based on a detailed search carried out on six relevant databases with no language or date restrictions and is based on a protocol which is registered on the International prospective register of systematic reviews database (24). This protocol was available on the National Institute for Health Research website and subsequently, the systematic review followed standardized reporting guidelines (24, 87). The strength in our review lies in the large number of studies which allowed extensive sensitivity and subgroup analysis to be conducted.

The main limitation in this study was the very high statistical heterogeneity evident from the I^2 estimates in the meta-analyses. It was not possible to carry out a subgroup analysis on maternal age, social supports and existing mental health due to lack of such data in the included studies. These factors are reported to be associated with tocophobia (9, 18, 40, 65). When we conducted a subgroup analysis including only studies that used the W-DEQ A to define tocophobia, significant heterogeneity remained suggesting that this issue is more complex than simply being explained by variation in the definition used. The authors acknowledge that the prevalence of tocophobia depends on several factors including various personality characteristics, previous birth experiences and cultural determinants including local obstetric norms, personal and religious beliefs (42, 66, 81). Furthermore many of the studies included in the systematic review were of a cross sectional design which only capture FOC at one point in time during pregnancy (See Table 1).

It is possible that questionnaires may not be applicable in different countries and in other cultural contexts (even in the same language) since psychometric aspects of the tool may be lost (14) thus tools should be specifically validated for use in each country (14, 53). This is a limitation of our study as we included studies that used various questionnaires administered in different languages (53). Of note, a high literacy level is required to complete the W-DEQ A (14). However, some studies used the three step approach to minimize any potential foreign language misinterpretation (46, 51, 61, 62). We acknowledge that the variety of different measurements for tocophobia both validated and non-validated used by the studies included in this systematic review may introduce possible bias including responder bias, language barrier bias, and reporter bias.

Despite these limitations, the information from this review provides important findings for use in future research and clinical practice. We identified that there are variations

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in the definition of tocophobia and that the prevalence of tocophobia appears to be increasing over time.

Conclusion

This systematic review and meta-analysis of the prevalence of tocophobia in pregnant women found a prevalence of 14%. However, these findings should be interpreted with caution due to significant heterogeneity which was not explained by extensive subgroup and sensitivity analysis. We ascertained that a clear operational definition for tocophobia is lacking in the literature. More research is required to gain a better understanding of FOC and how women with tocophobia may be given optimum support in clinical practice to achieve positive birth experiences. Despite limitations, these findings add to our limited understanding of tocophobia.

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Supporting Information Legends

Figure S1. Sensitivity analysis: Forest plot of the pooled prevalence of tocophobia using studies that screened women in the third trimester (27-42 weeks gestation) only.

Figure S2. Subgroup analysis: Forest plot of the pooled prevalence of tocophobia in high and low quality studies as determined by the quality assessment score. High quality studies were studies that scored 5 or more out of a maximum of 8.

Figure S3. Subgroup analysis: Forest plot of the pooled prevalence of tocophobia by study region.

Figure S4. Subgroup analysis: Forest plot of the pooled prevalence of tocophobia according to the time period in which the studies were conducted.

Appendix S1. Search Strategy.

Appendix S2. Quality Assessment Tool.

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Table and Figure legends

Table 1. Characteristics and quality assessment of studies included in the systematic review.

Table 2. Studies not included in the meta-analysis.

Figure 1. Flow chart of systematic search.

Figure 2. Forest plot of the pooled prevalence of tocophobia for all studies included in the meta-analysis.

Figure 3. Sensitivity analysis: Forest plot of the pooled prevalence of tocophobia including studies which used W-DEQ A≥85 as the definition for tocophobia.

Figure 4. Sensitivity analysis: Forest plot of the prevalence of tocophobia for studies which included nulliparous women only.

Figure 5. Sensitivity analysis: Forest plot of the pooled prevalence of tocophobia for studies which included multiparous women only.

Figure 6. Sensitivity analysis: Forest plot of the pooled prevalence of tocophobia using studies that screened women in the second trimester (13-27 weeks gestation) only.

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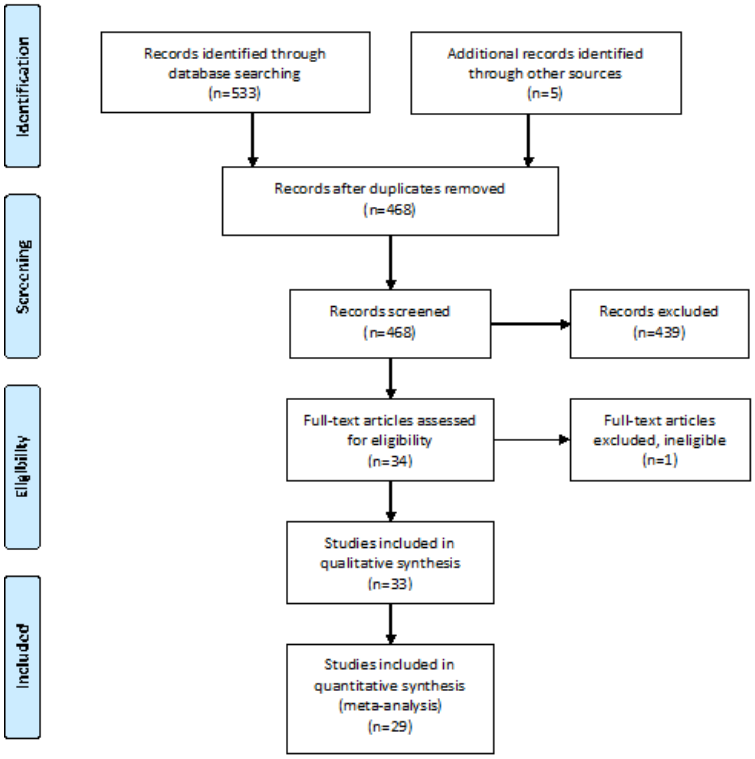

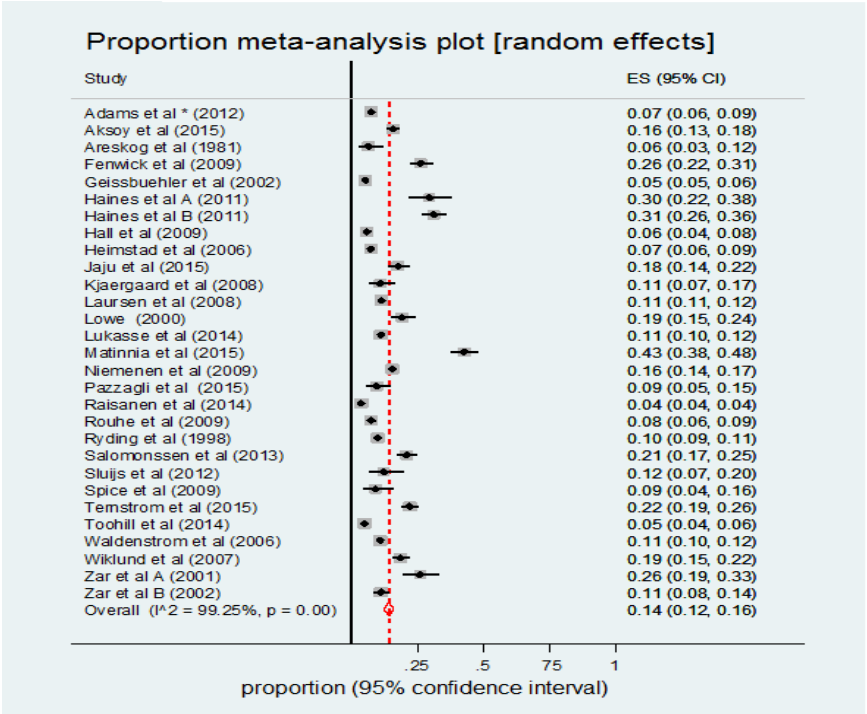

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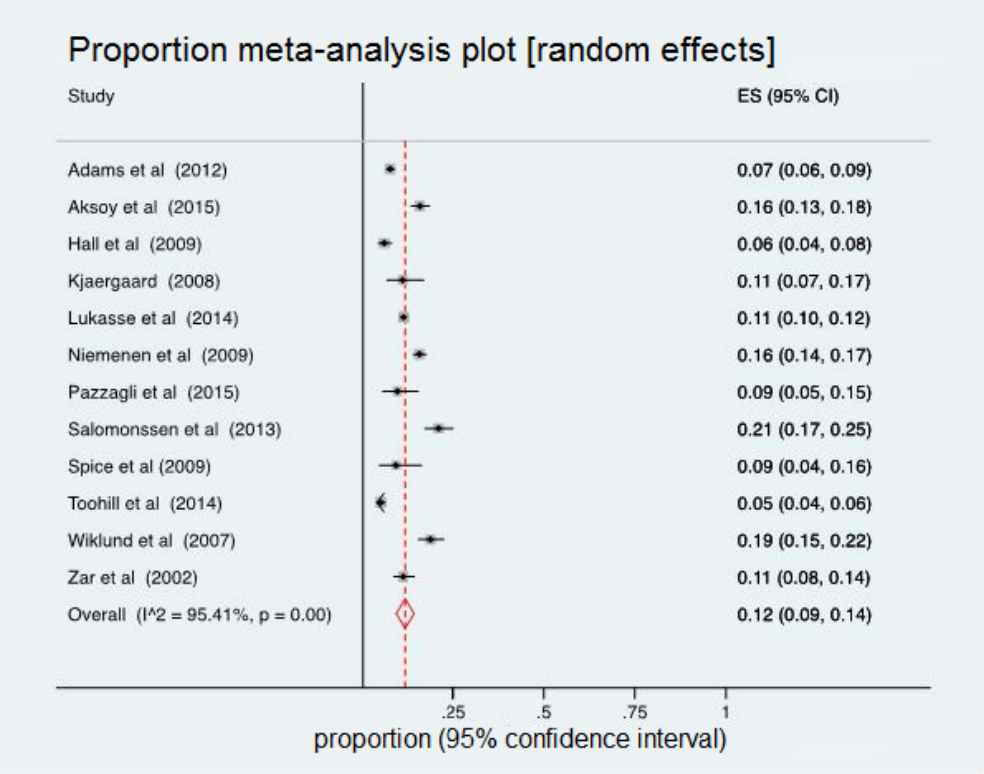

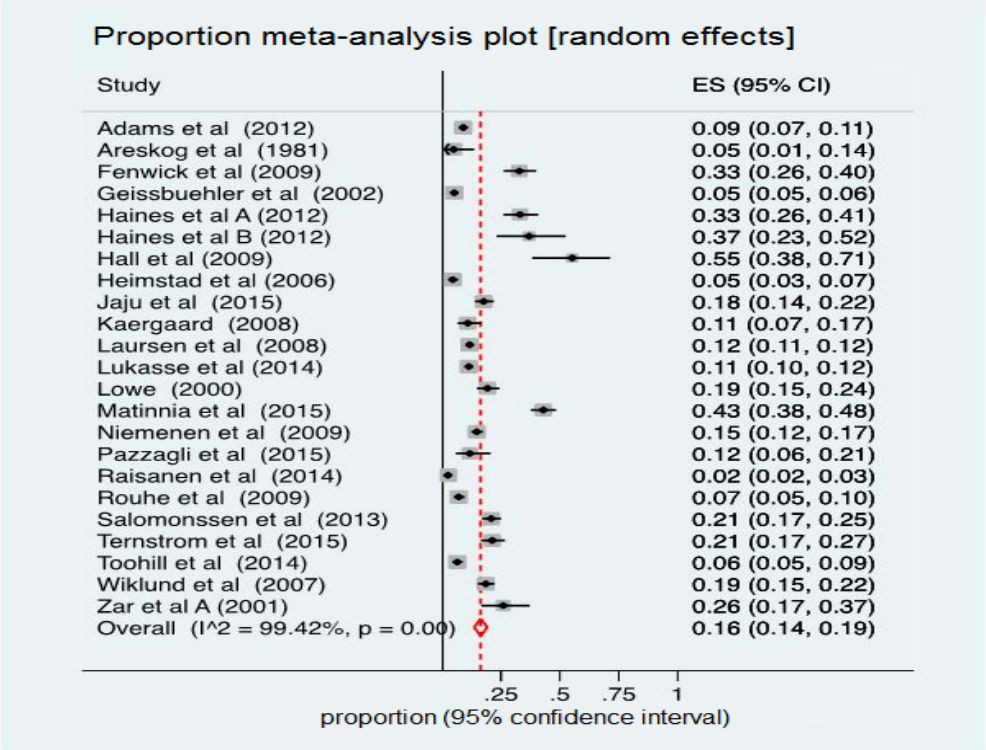
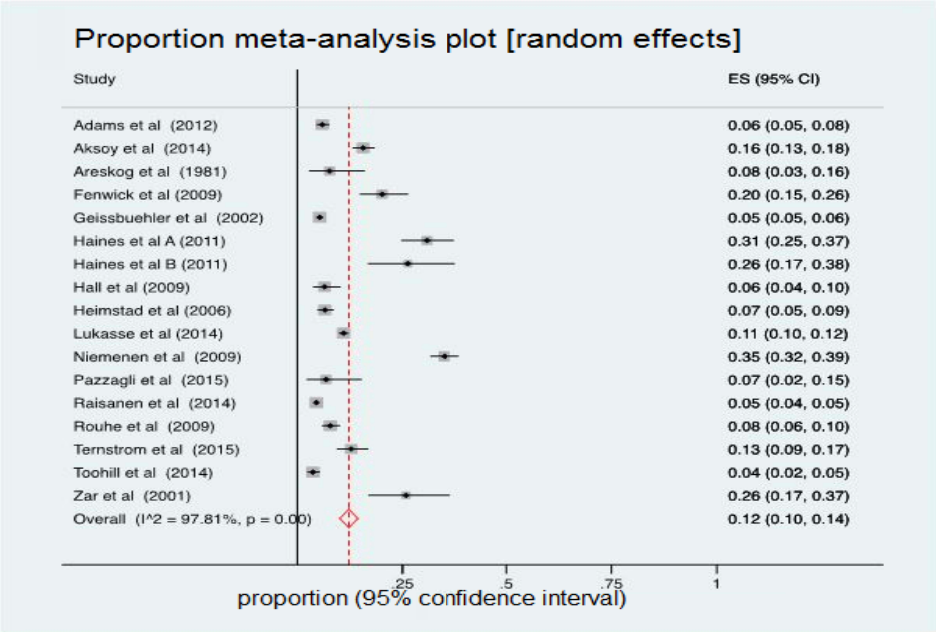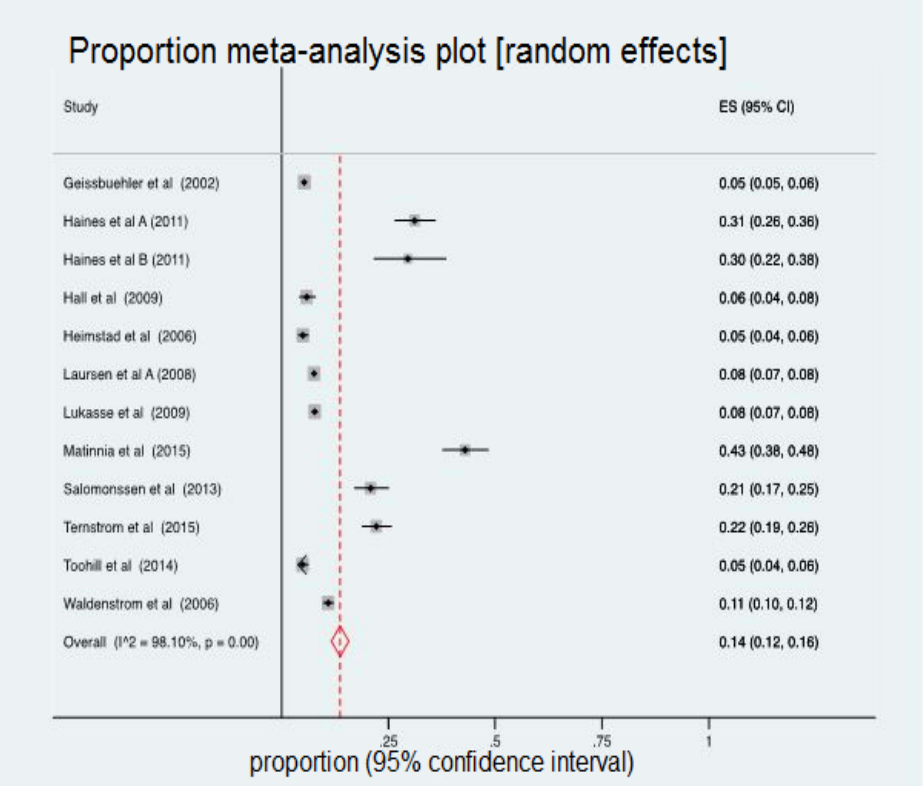
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