



Global prevalence of stillbirth among fetuses from twin pregnancies: a systematic review and meta-analysis

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Abstract

Background Evidence shows that twin pregnancy is associated with increased adverse outcomes, including stillbirth. The present study aimed to investigate the global prevalence of stillbirth among fetuses resulting from twin pregnancies through a systematic review and meta-analysis.

Methods To identify relevant studies, we adhered to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines. We searched databases including PubMed, Web of Science, Google Scholar, Scopus, Embase, and Science Direct using keywords such as “stillbirth,” “twin pregnancy,” “fetus,” and “prevalence” without applying any time constraints to the search. The quality of the articles was evaluated using the STROBE checklist. To ensure the reliability of our findings, we employed a random-effects model for analysis, and the heterogeneity of the studies was assessed using the I^2 index. Data analysis was conducted using Version 2 of Comprehensive Meta-Analysis software.

Results In a review of 10 studies with a sample size of 627,797 people, our meta-analysis revealed a global prevalence of stillbirth among fetuses resulting from twin pregnancies, reported to be 1.4% (95% CI 0.9–2.1). Our study also showed that with increasing sample size and years of study, the global prevalence of stillbirth among fetuses resulting from twin pregnancies decreased ($p < 0.05$). The highest prevalence of stillbirth, 3.5 (95% CI 2.2–5.4), was reported in the monochorionic type.

Conclusion The occurrence of stillbirth in twin pregnancies is a significant health concern for countries worldwide. Identifying its prevalence and contributing factors can aid health planning and policy development.

Keywords Stillbirth · Twin pregnancy · Fetus · Meta-analysis

Abbreviations

PRISMA Preferred Reporting Items for Systematic reviews and Meta-Analyses
STROBE Strengthening the reporting of observational studies in epidemiology

Background

Stillbirth, as an indicator of the quality of prenatal care, has not yet declined in developing countries. The World Health Organization defines stillbirth as the death of a fetus weighing 500 g or more, birth after 22 weeks of gestation, or birth with a head-to-heel difference of 25 cm or more. [1] It is estimated that 2.7 million stillbirths occur worldwide each year, most of which are thought to be due to preventable causes [2]. Ninety-eight percent of all stillbirths, a global concern, occur in low- and middle-income countries and 77% in South Asia and sub-Saharan Africa [3]. The stillbirth rate in developed countries is 3.4 per 1000 live births. In 2014, the World Health Organization (WHO) launched Every Newborn Action Plan (ENAP) to reduce stillbirths to less than 12 per 1000 live births by 2030 [4].

The rate of twin births varies considerably worldwide, depending on the country's development, from less than 1% of all births in South and Southeast Asia to more than 3% in

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the United States and France [5]. The increasing maternal age following the advent of new contraceptive methods and infertility treatments is a significant reason for the increase in twin pregnancy rates [6]. The issue of stillbirth in twin pregnancies is an important public health concern, given the increasing rate of multiple births due to factors such as assisted reproductive technologies and maternal age [7]. Despite advances in prenatal care, the incidence of stillbirth is significantly higher in twin pregnancies than in singleton pregnancies. Twin pregnancies inherently carry a higher risk of adverse outcomes, including stillbirth [8]. Research suggests that the incidence of stillbirth in twin pregnancies is approximately 6.9% [9]. Twin pregnancies are more likely to experience FGR and SGA, both of which are important risk factors for stillbirth. Studies suggest that more twins than singletons are diagnosed with these conditions, mainly when assessed using twin-specific growth charts [10]. The research findings on the risk of stillbirth associated with FGR in twins, especially those born before 32 weeks of gestation. While this risk decreases with increasing gestational age, it remains a significant factor compared with singletons. For DC twins born after 34 weeks, the prospective risk of stillbirth is consistent with the risks of neonatal death, indicating a balance in outcomes at this stage. However, for MC twins, there is a trend indicating an increase in stillbirth rates as term approaches, particularly after 36 weeks [11]. On average, premature infant mortality is seven times higher among twins. Studies in Cameroon and Ethiopia have shown that multiple pregnancy is a risk factor for stillbirth [12]. Women with multiples are at increased risk of preterm labor, placental problems, gestational hypertension, gestational diabetes, preeclampsia, postpartum hemorrhage, and anemia [13]. Notably, the risk of stillbirth is significantly higher for women with a previous history of stillbirth compared with women without a history of stillbirth, underscoring the need for targeted interventions. Apart from the immediate distress of losing a baby, stillbirth has severe psychosocial consequences for parents, including a wide range of impacts such as anxiety, long-term depression, post-traumatic stress disorder, and stigma. These effects include depression, anxiety, sleep disturbances, high levels of sadness, avoidance of social activities, substance abuse, and marital problems, which can have an impact on later life [2].

Different studies conducted in other countries have reported different prevalences of stillbirth among fetuses resulting from twin pregnancies. In a study conducted in the United States, the prevalence was 3.9% [17], in a study conducted in the United Kingdom, the prevalence was 1.03 [15], in a study conducted in Italy, the prevalence was 5.8% [5], and in a study conducted in Brazil, the prevalence was 1.1% [19], which indicates the heterogeneity of the prevalence's reported in the world. Overall, the prevalence of stillbirth among fetuses resulting from twin pregnancies varies

in different countries, a significant issue that our study aims to address. This issue can impose a tremendous social, economic, and psychological burden on other societies and lead to numerous unfortunate consequences. Therefore, given that the reported prevalence's around the world are heterogeneous and different, and also considering the importance of this condition in pregnant mothers, the aim of the present study is to investigate the global prevalence of stillbirth among fetuses resulting from twin pregnancies through a systematic review and meta-analysis.

Methods

The initial search for the present study was conducted in October 2024. For this purpose, six databases, including "PubMed," "Web of Science," "Scopus," "Embase," "Science Direct," and "Google Scholar," were searched using the keywords Prevalence, Stillbirths, and Twin Pregnancy suicides. No restrictions were applied to the year of publication of the articles. The obtained information was then transferred to the information management software (Endnote). In addition, the list of references used in the relevant articles was manually evaluated to maximize the number of articles. To ensure no relevant articles were missed, the searches were repeated in October 2024.

Inclusion and exclusion criteria.

Study inclusion criteria were:

1. Studies that reported the prevalence of stillbirth among fetuses resulting from twin pregnancies.
2. Studies with access to their full text.
3. Studies that provided sufficient data such as sample size and prevalence.
4. Studies that were in English.

Study exclusion criteria were:

1. Case report and case series studies.
2. Interventional studies.
3. Review studies.
4. Duplicate studies.
5. Studies with insufficient data (lack of information on prevalence and sample size).
6. Studies that were not in English.

Study selection

The selection of studies was carried out following the PRISMA guidelines. Initially, duplicate studies were eliminated from various databases. The titles and abstracts of the articles were reviewed based on the inclusion and exclusion criteria to carry out the initial selection and review of the

articles. Subsequently, the full text of the selected articles was reviewed based on the inclusion and exclusion criteria, and appropriate articles were selected. In order to prevent bias, the steps mentioned above were performed by two researchers independently of each other. Whenever there was a disagreement between the two researchers regarding an article, a third person resolved the article through a robust evaluation process.

Quality evaluation

The validation and qualitative assessment of the articles was carried out based on the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) checklist, ensuring the inclusivity of our research. This checklist includes six scales: title, abstract, introduction, methods, results, and discussion. It is based on a 32-item guideline such as title, problem statement, study objectives, type of study, study population, sampling method, determination of appropriate sample size, definition of variables and procedures, study data collection tools, statistical analysis methods, and findings. In general, articles that obtain good and moderate scores in terms of methodological quality are retained and otherwise are excluded from the study, ensuring that all relevant articles are considered in our research.

Data extraction

Two researchers extracted data based on a checklist that included the first author's name, year of publication, study location, study type, number of stillbirths, sample size, age group, prevalence of stillbirths, type of twin birth, and study instrument.

Statistical analysis

Data extracted from articles were entered into the Comprehensive Meta-Analysis (Version 2) software, and the I^2 test was used to examine the heterogeneity of studies. The Egger test was at a significance level of 0.05, and the Funnel plot was used to investigate publication bias.

Results

In this study, a systematic review and meta-analysis of data from studies have shown the prevalence of stillbirth among fetuses resulting from twin pregnancies. The studies were systematically evaluated according to the PRISMA guidelines. Through a search of databases, 895 potentially relevant articles were identified and transferred to the information management software (Endnote). 795 articles were excluded due to duplication. In the screening stage, the title

and abstract of the studies were evaluated, and 43 articles were excluded based on the inclusion and exclusion criteria. In the eligibility assessment stage, 43 articles were excluded through a full-text study based on the inclusion and exclusion criteria. In the qualitative assessment stage, through a full-text study of the articles and based on the score obtained from the STROBE (Strengthening the Reporting of Observational Studies in Epidemiology) checklist, four studies with poor methodological quality were excluded. Finally, ten studies were included in the final evaluation. The information from these ten studies is reported in Fig. 1 and Table 1.

In this study, based on Table 1, the highest prevalence of stillbirth in twin pregnancies was reported in the survey by Russo et al. in 2013 in Italy, which was 5.84% of 426 participants aged 18–35 years, which was determined using the participants' medical history [5]. In addition, the lowest prevalence of stillbirth in twin pregnancies was reported in the study by Sun Ko et al. in 2017 in South Korea, which was 0.69% of a substantial sample of 2598 participants aged 18–35 years, determined using data from the South Korean Statistics Center [14].

In a review of 10 studies with a sample size of 627,797 people, the I^2 heterogeneity test showed high heterogeneity (I^2 : 99.2), and accordingly, the random-effects method was used to analyze the results. Therefore, based on the meta-analysis, the global prevalence of stillbirth among fetuses resulting from twin pregnancies was reported to be 1.4% (95% CI 0.9–2.1) (Fig. 2). In addition, the examination of publication bias in studies through the Egger test showed the absence of publication bias in studies (p : 0.538) (Fig. 3).

Our examination of the factors affecting the heterogeneity of studies and the effect of sample size on this heterogeneity has led to findings that could have a significant impact. We report that with increasing sample size, the global prevalence of stillbirth among fetuses resulting from twin pregnancies decreases ($p < 0.05$) (Fig. 4). Furthermore, with each passing year of study, the global prevalence of stillbirth among fetuses resulting from twin pregnancies also decreases ($p < 0.05$) (Fig. 5).

According to the results presented in Table 2, which details the global prevalence of stillbirth among fetuses from twin pregnancies categorized by type of twinning, the monochorionic type had the highest prevalence, reported at 3.5% (95% CI 2.2–5.4) (Table 2).

Discussion

The present study, aimed at investigating the global prevalence of stillbirth among fetuses resulting from twin pregnancies, has yielded significant findings. The global prevalence of stillbirth among fetuses resulting from twin pregnancies was reported to be 1.4, a crucial piece of

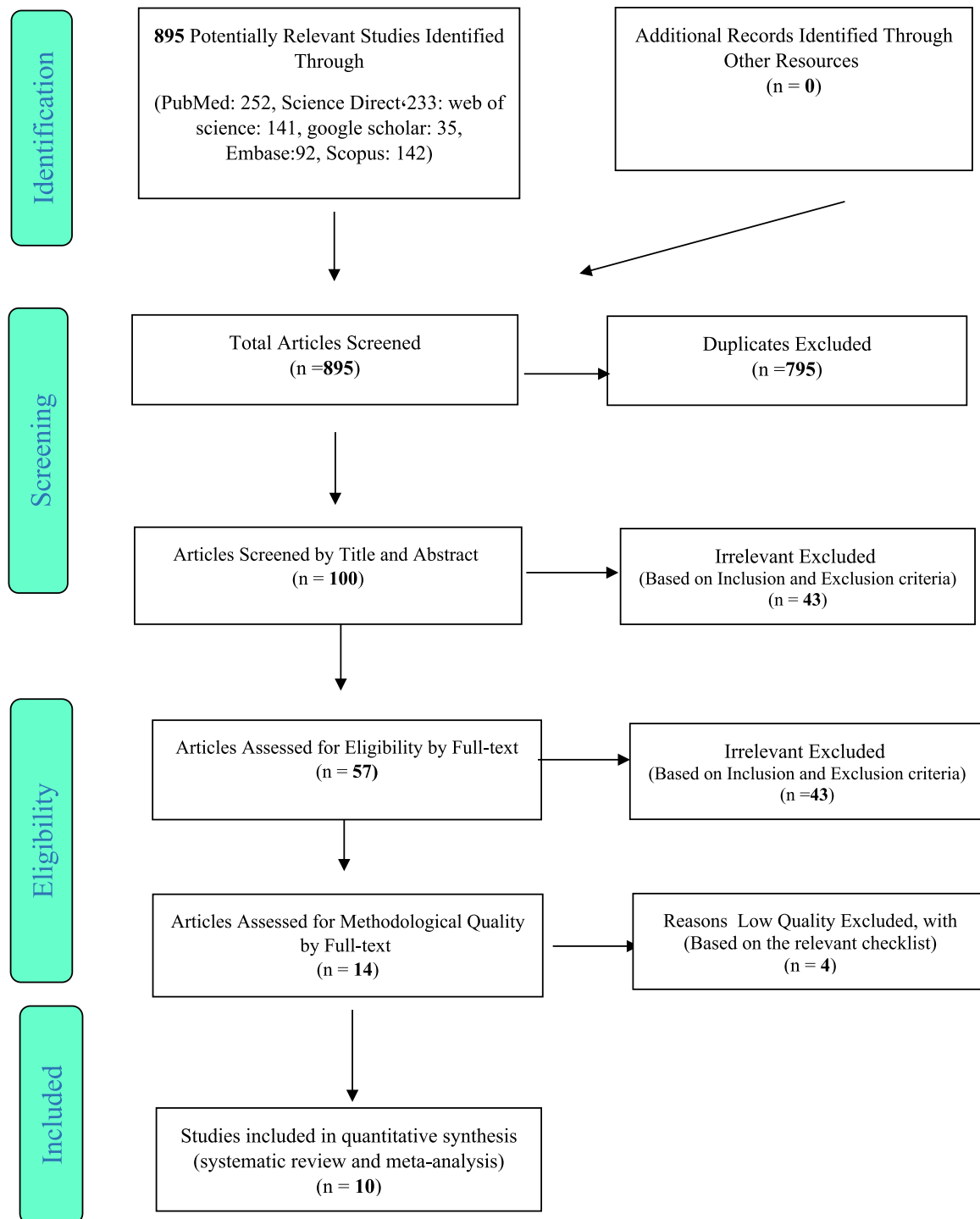


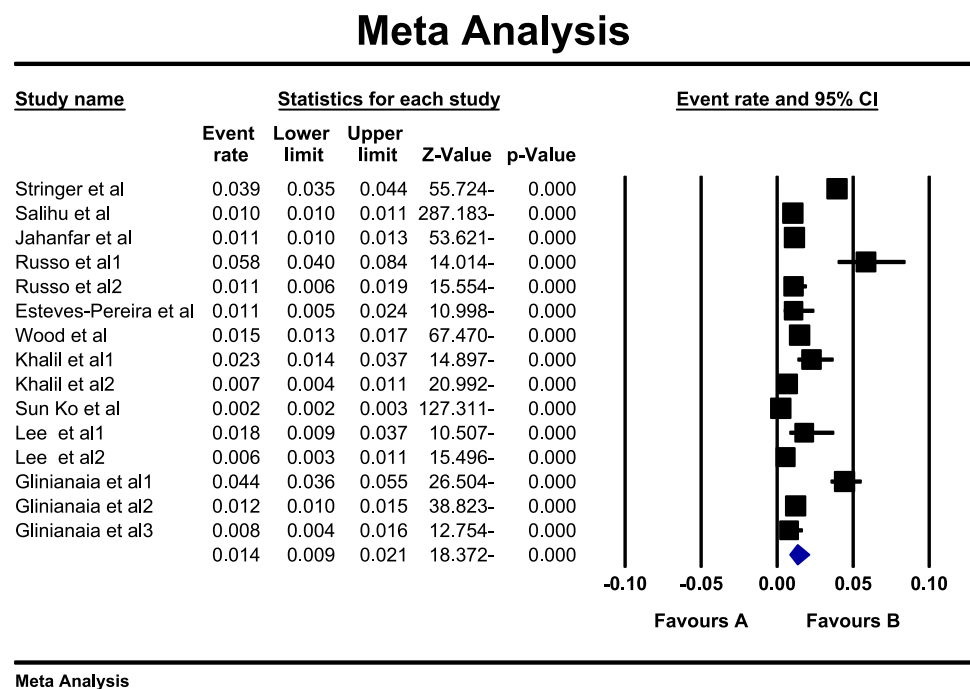
Fig. 1 The flowchart on the stages of including the studies in the systematic review and meta-analysis (PRISMA 2009)

information for our understanding of maternal and fetal health. Furthermore, the study revealed that with increasing sample size and years of study, the global prevalence of stillbirth among fetuses resulting from twin pregnancies decreases, shedding light on potential areas for further research and intervention. Notably, the results showed

that the prevalence of stillbirth among fetuses resulting from monochorionic twin pregnancies is higher than that of fetuses resulting from dichorionic twin pregnancies. In this study, the highest prevalence of stillbirth in twin pregnancies was reported in the survey by Russo et al. in 2013 in Italy [5], and the lowest prevalence of stillbirth in twin

Table 1 Summary of characteristics of included studies of the prevalence of stillbirth among fetuses from twin pregnancies

Author	Year	Country	Sample size	Instrument	Type of chorion	Prevalence of stillbirth in twins
Stringer et al. [17]	2015	USA	8042	ZEPRS	NR	3.9%
Salihu et al. [15]	2004	UK	387,419	“Matched multiple birth file	NR	1.03%
Jahanfar et al. [12]	2019	USA	12,814	PSBC	NR	1.1%
Russo et al. [5]	2013	Italy	462	ReCoDe, Medical History	Monochorionic	5.84%
Russo et al. [5]	2013	Italy	1108	ReCoDe, Medical History	Dichorionic	1.08%
Esteves-Pereira et al. [19]	2021	Brazil	554	Birth in Brazil	NR	1.1%
Wood et al. [20]	2014	Canada	17,724	App	NR	1.48%
Khalil et al. [18]	2013	UK	708	Electronic maternity records	Monochorionic	2.26%
Khalil et al. [18]	2013	UK	2598	Electronic maternity records	Dichorionic	0.69%
Sun Ko et al. [14]	2017	Korea	185,238	Statistics Korean Vital	NR	0.25%
Lee et al. [21]	2008	USA	392	departmental perinatal database	Monochorionic	1.78%
Lee et al. [21]	2008	USA	1608	departmental perinatal database	Dichorionic	0.56%
Glinianaia et al. [9]	2011	UK	1758	NorSTAMP	Monochorionic	4.44%
Glinianaia et al. [9]	2011	UK	6480	NorSTAMP	Dichorionic	1.22%
Glinianaia et al. [9]	2011	UK	892	NorSTAMP	Unknown chorionicity	0.78%

Fig. 2 Forest plot of global prevalence of stillbirth among fetuses from twin pregnancies based on random-effects method

pregnancies was reported in the study by Sun Ko et al. in 2017 in South Korea [14]. The difference in the results obtained in different countries can be attributed to factors such as the level of access to the prenatal health care system, the level and manner of self-care, and follow-up of high-risk pregnancies by mothers and families. However, it is crucial to note that genetic and environmental factors also play a significant role in stillbirth rates. According to the results of a large part of the studies included in this

study, congenital anomalies were the main determining factor of stillbirth in twin pregnancies [14–16].

Our research has uncovered several important risk factors that significantly increase the incidence of stillbirth in twin pregnancies. These factors, including obesity, maternal age, smoking, pre-pregnancy diabetes, high blood pressure, and timing of delivery, have been consistently reported in numerous studies [4, 17, 18]. A particularly significant finding of our research is the higher incidence of stillbirth among

Fig. 3 Funnel plot examining publication bias in the reviewed studies

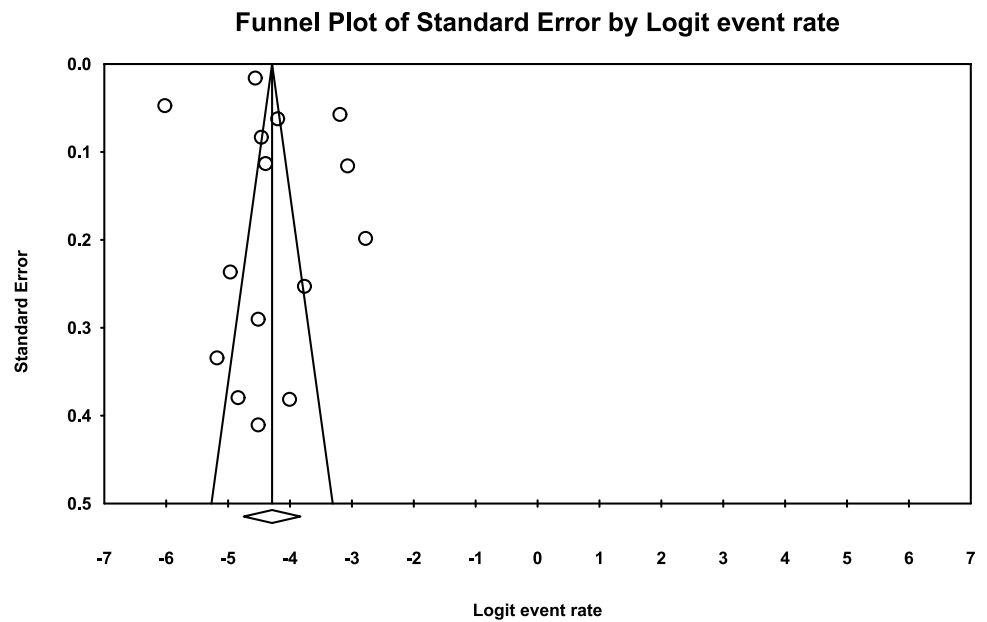


Fig. 4 Meta-regression of the effect of sample size on the global prevalence of stillbirth among fetuses from twin pregnancies

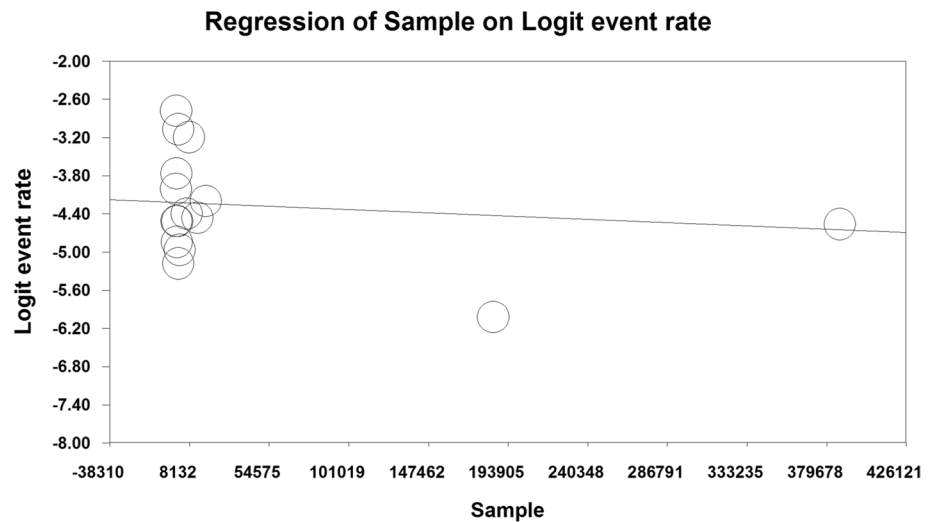


Fig. 5 Meta-regression of the effect of study year on the global prevalence of stillbirth among fetuses from twin pregnancies

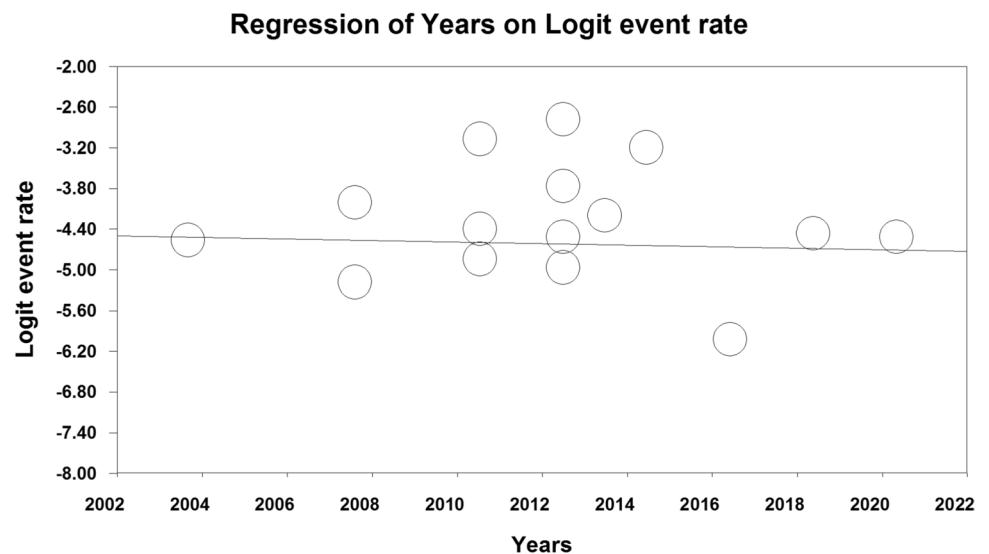


Table 2 Global prevalence of stillbirth among fetuses resulting from twin pregnancies by type of twinning

Prevalence (95% CI)	Egger test	I^2	Sample size	N	Type
3.5 (95% CI 2.2–5.4)	0.326	79.9	3320	4	Monochorionic
0.9 (95% CI 0.6–1.3)	0.182	64.8	11,794	4	Dichorionic

fetuses from monochorionic twin pregnancies compared to those from dichorionic twin pregnancies. This difference may be due to factors such as blood transfusion syndrome in fetuses from monochorionic twin pregnancies, placental angioarchitecture being a significant factor for weight inconsistency between twins in monochorionic fetuses, resulting in poor blood supply and fetal distress, which can subsequently cause fetal death [12].

In a study by Annelise Skeie et al., it was reported that the epidemiology of twin pregnancies complicated by the stillbirth of one or both fetuses is an area that has rarely been studied. While the risk of perinatal death in twin pregnancies is two- to fivefold higher than in singletons, this study reports that the risk of stillbirth is increased with monochorionic, non-Western origin, and assisted reproductive techniques (ART), and that identification of monochorionic pregnancy should be performed early in pregnancy to determine the level of risk [11]. Fiona Cheong-See's and et al., study emphasized the importance of the risk of stillbirth and neonatal complications according to gestational age in uncomplicated monochorionic and dichorionic twin pregnancies and reported that to minimize perinatal mortality, delivery should be considered at 37 weeks of gestation in uncomplicated twin pregnancies. In monochorionic pregnancies, delivery should be considered at 36 weeks [1].

Rydstroem et al. also reported that the main risk factor for fetal death appears to be same-sex pregnancies with monochorionic twins. About half of the pregnancies with available data did not have more than 9 days between ultrasound examination, confirmation of two viable fetuses, and birth. The majority of these twins showed no evidence of growth retardation, but birth weight discrepancy may be a risk factor for fetal death (22).

Limitations

The main limitation of this meta-analysis is that the included studies were limited to those published in English, potentially overlooking studies in other languages. In addition, the number of studies reporting the prevalence of stillbirth by type of twin birth was unexpectedly low. It is important to note that some studies were excluded due to poor quality,

such as not reporting prevalence or having a small sample size. This underscores the importance of considering the quality of studies in a meta-analysis, as it ensures the rigor and reliability of the research.

Conclusion

The results of this study indicate that there are a limited number of studies in this field and that more extensive studies are needed in this field. This study emphasizes the importance of knowing the prevalence of stillbirth among fetuses resulting from twin pregnancies to examine global estimates of this health problem and plan to mitigate it.

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Author contributions NS, MB and MM contributed to the design, MM statistical analysis, and participated in most of the study steps. MM, RA and MH prepared the manuscript. MM, KH, PH and SHSH assisted in designing the study, and helped in the, interpretation of the study. All the authors have read and approved the content of the manuscript.

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Data availability Datasets are available through the corresponding author upon reasonable request.

Declarations

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

Ethical approval and consent to participate Not applicable.

Consent for publication Not applicable.

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