

BMJ Open is committed to open peer review. As part of this commitment we make the peer review history of every article we publish publicly available.

When an article is published we post the peer reviewers' comments and the authors' responses online. We also post the versions of the paper that were used during peer review. These are the versions that the peer review comments apply to.

The versions of the paper that follow are the versions that were submitted during the peer review process. They are not the versions of record or the final published versions. They should not be cited or distributed as the published version of this manuscript.

BMJ Open is an open access journal and the full, final, typeset and author-corrected version of record of the manuscript is available on our site with no access controls, subscription charges or pay-per-view fees (http://bmjopen.bmj.com).

If you have any questions on BMJ Open's open peer review process please email <a href="mailto:info.bmjopen@bmj.com">info.bmjopen@bmj.com</a>

# **BMJ Open**

# Inequalities and stillbirth in the UK: A meta-narrative review

Journal:	BMJ Open
Manuscript ID	bmjopen-2019-029672
Article Type:	Research
Date Submitted by the Author:	04-Feb-2019
Complete List of Authors:	Kingdon, Carol; University of Central Lancashire, ReaCH group Roberts, Devender; Liverpool Womens NHS Foundation Trust, Department of Obstetrics Turner, Mark; Liverpool Women's Hospital NHS Foundation Trust, Storey, Claire; International Stillbirth Alliance, Crossland, Nicola; University of Central Lancashire, School of Health Finlayson, Kenneth; University of Central Lancashire, School of Health Downe, Soo; University of Central Lancashire, research in childbirth and health
Keywords:	stillbirth, inequalities, deprivation, ethnicity

SCHOLARONE™ Manuscripts

## Inequalities and stillbirth in the UK: A meta-narrative review

#### **Corresponding Author**

Dr Carol Kingdon, Senior Research Fellow,

School of Community Health and Midwifery, Faculty of Health and Wellbeing,

University of Central Lancashire, Preston, United Kingdom. PR1 2HE

Tel: 07935744004

ckingdon@uclan.ac.uk

ORCID ID 0000-0002-5958-9257

#### Co-authors

Dr Devender Roberts, Consultant Obstetrician in Fetal Medicine,
Liverpool Women's NHS Foundation Trust, Crown Street, Liverpool, United Kingdom. L8 7SS

<u>Devender.Roberts@lwh.nhs.uk</u>

Professor Mark Turner, Reader in Neonatology/Consultant Neonatologist,
Department of Women's and Children's Health, Institute of Translational Medicine,
Faculty of Health and Life Science, University of Liverpool, Liverpool, United Kingdom. L8 7SS

Mark.Turner@liverpool.ac.uk

Ms Claire Storey, Co-chair, International Stillbirth Alliance, 61 Kings Drive, Stoke Gifford, Bristol, United Kingdom. BS34 8RD storey.claire@yahoo.com

Dr Nicola Crossland, Research Associate,
School of Community Health and Midwifery, Faculty of Health and Wellbeing,
University of Central Lancashire, Preston, United Kingdom. PR1 2HE
<a href="mailto:ncrossland@uclan.ac.uk">ncrossland@uclan.ac.uk</a>

Mr Kenny Finlayson, Research Associate,
School of Community Health and Midwifery, Faculty of Health and Wellbeing,
University of Central Lancashire, Preston, United Kingdom. PR1 2HE
<a href="mailto:KWFinlayson1@uclan.ac.uk">KWFinlayson1@uclan.ac.uk</a>

Professor Soo Downe, Professor of Midwifery,
School of Community Health and Midwifery, Faculty of Health and Wellbeing,
University of Central Lancashire Preston, United Kingdom. PR1 2HE
sdowne@ulcan.ac.uk

Word count: 3,673

#### **Abstract**

**Objective:** To review what is known about the relationship between stillbirth and inequalities from different disciplinary perspectives to inform stillbirth prevention strategies.

**Design:** Systematic review using the meta-narrative method.

**Setting:** Studies undertaken in the UK.

**Data sources:** Scoping phase: experts in field, exploratory electronic searches and hand-searching. Systematic searches phase: Nine databases with no geographical or date restrictions. Non-English language studies were excluded.

**Study selection:** Any investigation of stillbirth and inequalities with a UK component. **Data extraction and synthesis:** Three authors extracted data and assessed study quality. Data were summarised, tabulated and presented graphically before synthesis of the unfolding storyline by research tradition; and then of the commonalities, differences and interplays between narratives into resultant summary meta-themes.

**Results:** 54 sources, from nine distinctive research traditions were included. Evidence of associations between social inequalities and stillbirth spanned 70 years. Across research traditions there was recurrent evidence of the social gradient remaining constant or increasing, fuelling repeated calls for action (*Meta-theme 1: Something must be done*). There was less evidence of an effective response to these calls. Data pertaining to socio-economic, area and ethnic disparities were routinely collected, but not consistently recorded, monitored or reported in relation to stillbirth (*Meta-theme 2: Problems of precision*). Many studies stressed the interplay of socio-economic status, deprivation or ethnicity with aggregated factors including heritable, structural, environmental, and lifestyle factors (Meta-theme 3: *Moving from associations towards intersectionality and intervention(s)*). We found no intervention studies.

**Conclusion:** Research investigating inequalities and stillbirth in the UK is underdeveloped. This is despite repeated evidence of an association between stillbirth risk and poverty, and stillbirth risk, poverty and ethnicity. A specific research forum is required to lead the development of research and policy in this area, which can harness the multiple relevant research perspectives and address the intersections between different policy areas.

Protocol registration number: CRD42017079228

**Keywords:** Stillbirth, inequalities, deprivation, social class, poverty, ethnicity, metanarrative.

#### **Article summary**

#### Strengths and limitations of this study

- Meta-narrative is a systematic and rigorous methodological approach to understand how multiple disciplines and different philosophical perspectives have researched a question over time, and is thus a powerful approach to illuminate what has been a long-standing, inherently complex problem.
- To the best of our knowledge this is the first study to use a meta-narrative approach to investigate the relationship between inequalities and stillbirth in the UK. Our findings unite research from Social Medicine; Epidemiology; Medical Sociology; Public Health; Spatial Epidemiology; Social Psychology; Audits, Reports and Confidential Enquiries; Fetal-Maternal Medicine and Nursing and Midwifery.
- We adhered to the RAMASES standards for meta-narrative review to ensure fidelity with the methodology. We used a multipronged approach to retrieving sources which included exploratory searches, systematic searches, hand searches, expert opinion, and forward and back-chaining, which gave us a broad capture of relevant documents.
- By limiting the review to UK-based studies only, we were able to focus with greater acuity on the commonalities and contestations between research traditions. However, excluding studies from other countries may have led us to miss important research on the relationship between stillbirth and inequalities from other countries, of relevance both in the UK context and globally.

**Funding statement:** This work was supported by SANDS, the Stillbirth and Neonatal Death Charity.

## INTRODUCTION

Avoidable inequalities in mortality across the life course are a global concern. [1] Ten countries account for 66% of the world's stillbirths, with most (98%) occurring in low-and middle-income countries (LMICs). [2] Inequalities exist within and between high-income countries (HICs) too. In 2011, the Lancet *Stillbirth Series* highlighted that the UK's stillbirth rate was one of the highest of all HICs. [3] In 2016 the second Lancet Series *Ending Preventable Stillbirths* reported that while overall stillbirth rates were falling in HICs, improvement was slower than expected, and significant inequalities within rates remained. [4] The UK's stillbirth rate continues to remain high in comparison to other high-income countries. [5]

The current government ambition is to halve the stillbirth rate in England by 2025, which would require the rate to fall to 2.6 per 1,000 total births. [6] In 2017, the stillbirth rate in England and Wales decreased to 4.2 per 1,000 total births. [7] Medical reasons for stillbirth are well known and strategies for prevention routine. Ongoing initiatives include the Safer Maternity Care strategic plan, [8] Saving Babies Lives Care Bundle, [9], Each Baby Counts, [10] the Perinatal Mortality Review Tool [11] and annual Perinatal Mortality Reports (MBRRACE-UK) [5]. The relationship between social determinants and stillbirth is less well understood. Clinicians acknowledge the need to do more to prevent stillbirth in women from socially disadvantaged groups. In England in 2017, the stillbirth rate in the most deprived areas was 5.5 per 1,000 total births, compared with 3.0 per 1,000 total births in the least deprived areas. [7]

The UK began to develop policies to address health inequalities in general following *The Acheson Inquiry into Inequalities in Health*. [12] The Marmot Review *Fair Society, Healthy lives*, published in 2010, progressed the UK's inequalities agenda by emphasising the importance of taking a life-course approach, starting with the early years and family building. [13] The key messages of the Marmot Review emphasised that there is a social gradient in health in the UK, whereby the lower an individual's social position the worse his or her health, which is unfair, and that this requires action across all the social determinants of health.

Public Health England's current strategy for action on inequalities *Reducing health inequalities: system, scale and sustainability* [14] is underpinned by the Dahlgren and Whitehead rainbow model of the social determinants of health. [15] This model offers a framework to explore the relative influence of these determinants on different health outcomes and the interactions between the various determinants. These are all potential mechanisms by which stillbirth risk maybe increased. What is missing from current stillbirth research agendas is an overarching synthesis of clinical and social science evidence to clarify the range of individual (including biological and behavioural), social and environmental mechanisms of increased stillbirth risk, the intersections between these mechanisms, and strategies to tackle them. This review sought to fill this knowledge gap.

The aim was to undertake an inter-disciplinary evidence synthesis (using a metanarrative approach) to understand how structural factors, lifestyle factors, and clinical factors intersect to increase stillbirth risk, and to inform future strategies to manage atrisk pregnancies. The broad research question was what is the relationship between inequality and stillbirth, how has this been studied, and with what effects?

## **METHODS**

We conducted a systematic review using the meta-narrative method, [16-18] in accordance with the RAMASES standards. [19] A PRISMA checklist is provided (Supplementary information file 1). [20] Our protocol [21] (supplementary file 2) specified four objectives:

- 1. To review the current body of knowledge of the relationship between inequalities and stillbirth across the natural and the social sciences;
- 2. To provide new insights into the interplay of biological, clinical, cultural and socioeconomic factors in increased stillbirth risk;
- 3. To explore the impact of interventions on inequalities;

4. To provide a narrative summary of this research for stakeholders tasked with reducing preventable stillbirth.

#### **Meta-narrative**

Meta-narrative review is a type of systematic review that was developed by Trisha Greenhalgh and colleagues [16-18], with meta-narrative a term for the unfolding storyline of research in a particular tradition or topic. [22]. We used this approach to make sense of evidence from heterogeneous sources in which stillbirth and inequality have been variously conceptualised and studied over time. The method is underpinned by the methodological principles of pragmatism, pluralism, historicity, contestation, reflexivity and peer-review. As a method, meta-narrative review involves six key stages:-

 Planning We registered our protocol with PROSPERO [21] and assembled a multi-disciplinary research team.

## 2. Iterative scoping searches and systematic electronic searches

Initial searches were designed to map the diversity of perspectives and approaches. We contacted experts in the field of stillbirth research and from disciplines contributing to inequalities research. Exploratory searches were conducted using the search term "stillbirth" in 13 databases in health and the humanities (Supplementary File 3). Systematic searches were conducted in November 2017 in MEDLINE, Embase, CINAHL, PsycINFO, Popline, Historical abstracts, Humanities International Complete, Race Relations Abstract, and SocIndex. See supplementary file 4: example systematic search strategy. An English language restriction was imposed, but no geographical or date restrictions. In our protocol, inclusion criteria were any study design investigating stillbirth and inequality, in a high-, middle-, or low-income setting. Following initial screening of titles and abstracts a pragmatic decision was made by the team to include only studies with a UK component. Unchanged

exclusion criteria from the protocol were: any study in non-English language; of pregnancy loss <20 weeks gestation; of perinatal loss in the neonatal period; involving participants who had assisted conception. Screening was independently undertaken by three authors (NC, KF, CK), who also assigned potential inclusions to disciplinary categories at this stage (See supplementary file 5: screening tool).

- 3. **Mapping** A data extraction form was developed based on one used in earlier reviews, [23] which was adapted for the purpose of this metanarrative review. Additional fields were added to capture data relating to how inequalities and stillbirth were conceptualised, defined and theorised. The form was piloted by extracting data from a subset of five papers (taken from across the research traditions) to test for applicability to the metanarrative, and refined. Extracted data was then summarised, tabulated and presented.
- 4. Appraisal We stated in our protocol that all articles that met the inclusion criteria would be independently assessed by three researchers to minimize bias. During the process of the review it became apparent that quality appraisal of all quantitative studies using the appropriate checklists from the Critical Appraisal Skills Programme (CASP) Toolkit was not appropriate, with quality more suitably judged by the prevailing standards in each tradition. That said it was fitting to use CASP tools [24] for some studies in the epidemiological tradition and the Walsh and Downe tool for qualitative research quality appraisal. [25]
- 5. Synthesis phase The synthesis phase built on the unfolding stories of research traditions over time developed in the mapping phase. This involved evaluating the meta-narratives to identify and compare how the different research traditions conceptualized and theorized the topic, and the methodological approaches and study designs used. Differences in findings

between the resulting meta-narratives were analysed interpretively to produce further insights. The synthesis process involved paradigm bridging (seeking commonalities in underlying conceptual and theoretical assumptions), paradigm bracketing (highlighting differences in these assumptions), interplay (exploring tensions) and meta-theorizing (exploring patterns that span conflicting understandings) to construct summary meta-themes. KF, NC and CK undertook the initial analysis and synthesis processes, with input from DR, MT, CS and SD.

 Recommendations phase We engaged with local clinical networks and the national Stillbirth and Neonatal Death Charity (SANDS) to formulate recommendations.

## Patient and public involvement

Author Claire Storey is a parent and Vice-Chair of the International Stillbirth Alliance and was involved in the design, conduct and writing up of this review.

## **RESULTS**

From electronic searches of nine databases a total of 13,610 records were identified. Following duplicate removal 4,934 records were screened (Figure 1: PRISMA diagram). We included 54 sources from nine research traditions spanning the period 1945-2017. [26-79] Table 1 provides a summary of included research traditions. Chronologically, these traditions were Social Medicine [26-31]; Epidemiology [32-48]; Medical Sociology [49-53]; Public Health [54-58]; Spatial Epidemiology [59-64]; Social Psychology [65]; Audits, Reports and Confidential Enquiries [66-74]; Fetal-Maternal Medicine [75-78] and Nursing and Midwifery [79]. Supplementary Table 6 provides details about the characteristics of included sources. With the exception of Epidemiology (n=17), most traditions generated few relevant papers. All research traditions utilised epidemiological data. We included one mixed-method study reporting qualitative data. We identified no intervention studies. Lack of studies,

heterogeneity of study design, definitions of stillbirth, and measurement of inequalities between studies, traditions and over time meant meta-analysis was not practical. Figure 2 maps the traditions contribution over time and the declining national stillbirth rate.

## Synthesis within traditions

Table 1 summarises the unfolding storylines by research tradition and their conceptualisation of inequalities.

## **Table 1: Summary of included research traditions**

Research tradition	Academic discipline	Definition and scope	Unfolding storyline 2	Inequalities conceptualised as	Included references
Social Medicine	Medicine	Social Medicine is a branch of medicine that uses epidemiological methods to establish a problem exists, determining factors and opportunities for preventative action. The tradition is distinctive in its thought on the interconnectedness between biological factors (i.e. mother's age) that have meaning whatever the social context and social factors (i.e. occupational social class) that derive their meaning from social organisation in human life emulating political economy concerns.	The Social Medicine [26-31] storyline begins with the investigation of how social and economic faces influenced the decline in stillbirths and early neonatal deaths in Scotland, England and Wales, between 1939 and 1944. Bate [26] attributed this fall to the improved nutrition of the mothers during pregnancy, a consequence of the national distribution and consumption of milk and other foods important for health during the second world war. These improvements affected every area, are group and parity. By 1949, the decline in the stillbirth rate had slowed, despite the introduction of the National Health Service (NHS). Four papers, from a series in The Lancet in 1955 [27-30] sought to understand why. The last paper concluded the independent effects of social class, region, the mother's age and parity on stillbirth risk. Illsley [31] showed how occupational class may be more than a measure of inequality simply based on environmental conditions at the time of maternity, reporting that it can also be a resider of a woman's personal characteristics (height, physique, health, intelligence and nutrition), education and social habits. Women who were intergenerationally upwardly socially mobile at marriage experienced less stillbirths.	A variety of social factors that combine with biological characteristics to increase vulnerability to stillbirth risk.	N=6 [26-31]
Epidemiology	Medicine	Epidemiology, developed out of the bio-medical model as a specific line of inquiry. Initially epidemiology focused exclusively on epidemics of communicable diseases but subsequently expanded to address endemic communicable diseases and non-communicable infectious diseases. It is the study of the distribution and determinants of health-related states (especially disease), and the application of findings to the control of diseases and other health problems.	The Epidemiology [32-48] storyline is characterised by its increasingly sophisticated use of data are the repetition of the same or similar findings over time. Of the seventeen studies aligned to this tradition, six were landmark papers, repeatedly referenced within the field. [32-37] Although most authors highlighted a significant decrease in UK stillbirth rates sing the 1960's, studies repeatedly showed that the social gradient remained constant. [36,37,41] Within overall stillbirth rates sing the 1960's, studies repeatedly showed that the social gradient remained constant. [36,37,41] Within overall stillbirth rates sing the 1960's, studies repeatedly showed that the social gradient remained constant. [36,37,41] Within overall stillbirth rates sing the 1960's, studies repeatedly showed with an increased (as measured by an individual occupation) or residing in a disadvantaged community (as measured by local area deprivation), were relatively consistent markers of increased incidence of stillbirth, when compared with more socially divantaged counterparts. An important strength is epidemiology's identification of clinical, socio-economic and lifestyle factors associated with an increased risk of stillbirth across relatively large populations. Early studies used the Registrar General's Scale of occupational social class as a measure of inequality, later studies use the socio-economic classification scheme. Other studies the term deprivation' to signify inequality. In most of the studies using deprivation as a factor the risk of stillbirth increases with increasing levels of deprivation [34,43,44] although this is not always the case. [38] Epidemiological studies looking at athnicity as a measure of inequality are a relatively recent phenomenon and do not show the same level of consistency, although the rates of stillbirth for women of African-Caribbean origin remain at twice the rate of white women. [38,45] Studies exploring the stillbirth rates of women of African-Caribbean origin remain at twice the rate of whi	A variety of factors (social class, living in an area of deprivation, occupation of partner, ethnicity, etc.) associated with an increased relative risk of stillbirth.	N=17 [32-48]
Medical Sociology	Sociology	Medical Sociology is the study of the social causes and consequences of health and illness. This tradition has positivist and interpretative, theoretical and empirical, quantitative, qualitative, and mixed-methods, and cross-disciplinary branches. The persistence of social class gradients despite the demographic and epidemiological changes associated with the transition to modernity was an important focus during the 1970's and 1980's. During the 1990's research increasingly focused on lay understandings of health and illness and lived realities.	The Medical Sociology [49-53] storyline is theoretical. Early sociological explanations for the persistence of the social gradient in stillbirth encompassed theories of capital assets (the physique, stature, nutrition of the mother), social mobility (a direct thread from Social Medicine [31]), and time-lag (whereby developments in healthcare take time to reach those most in need, benefiting those better off first). [49] After the seminal Black Report [54] more nuanced considerations of gender, and area of residence, alongside occupational class, as simultaneous and overlapping vulnerabilities, were developed. [50-52] These encompassed the broad consideration of life circumstances, behaviours, and beliefs/attitudes [50] and the precise disaggregation of the concept of 'deprivation' to reveal the complexity of materialist risks (and protections against these risks), which helps to explain the ambiguous relationship between economic deprivation and ethnicity. [52]	A set of social relations (rather than just a variable), which opens lived experience and multiplicity of factors at play (i.e. poverty, poor housing, nutrition, welfare) and relationship between structure and agency.	N=5 [49-53]
Public Health	Public Health	Public health is concerned with preventing disease, prolonging life and promoting health through organised efforts of society. From 18th and 19th century roots, during the 1980's there was a revival of public health policy. In the UK this coincided with a shift in thinking that morbidity or general health status had become the more important indicators of inequality, and increasing interest in individual behaviours and lifestyle as determinants of health.	The Public Health storyline [54-58] unites the seminal Black Report [54] (which had a major impaction research into inequalities in health in the UK), with seminal papers from the two Lancet Stillbirth Series [57-58] that were of equal significance to the stillbirth research and policy community. In the former publication [54] stillbirth is a crude cause of death of the gory, used as part of efforts to explain general trends in inequalities in health, based principally on measures of occupational social class from which artefact, natural selection, structuralist, and behaviourist explanations, (alongside the need to build on the is a of multiple causation) were developed. In the latter publications, distinguishing between different kinds of stillbirth and the imperance of making each stillbirth count, come alongside the need to build on the idea of interactions between factors that include social disadvantage. [57-58] The lack of targeted interventions for Black and Ethnic minority women in the UK, despite their complementaries of increased risk and known underutilisation of maternity services, was highlighted in the scoping review by Garcia. [56] 2016, there was an explicit recall to action to tackle inequalities and stillbirth within HICs by addressing structural factors (success poor housing, poverty) and factors, which limit women's access to antenatal care. [58]	An additional risk and considered in relation to providing targeted care to populations considered at risk.	N=5 [54-58]

			BMJ Open Jo		
			BMJ Open BMJ -2019-029		
Spatial Epidemiology	Medical Geography and Epidemiology	Spatial epidemiology is concerned with the spatial analysis of disease incidence and prevalence. It uses geographical mapping and statistical modelling to understand the spatial distribution of disease, under the assumption that this will provide indications of the environmental contributors to the disease.	The Spatial Epidemiology [59-64] storyline begins in the late 1980s and attempts to address how community deprivation and individual social class might each contribute to risk of stillbirth. Studies looking at stillbirth and inequalities have investigated the relative importance of individual level (Registrar General Social Class) versus area level (e.g. Townsend Score) measures of inequality. Studies report contradictory findings, perhaps revealing the complexity of how individual (compositional) and area (context) effects interact to affect risk, with some reporting an enduring association between area and/or individual level deprivation and stillbirth risk [59,61-63] and others reporting no association [60,64]. The storyline of UK-based search into place effects on stillbirth risk has so far conceptualised geographical areas as "containers" of people, rather than seeing place as socially constructed.	A variety of factors (social class, living in an area of deprivation, occupation of partner, ethnicity) associated with an increased relative risk of stillbirth.	N=6 [59-64]
Social Psychology	Psychology	Social psychology is the study of human social behaviour, emotion and cognition. With its focus on both the individual and society, it draws on sociological and psychological perspectives Research methods involve both quantitative and qualitative approaches, and include surveys, participant observation, laboratory experiments, field experiments, and archival and content analyses.  Experimental social psychology is underpinned by positivist assumptions, while other approaches such as critical social psychology, operate from a social constructionist stance.	The Social Psychology [65] storyline arose from the Black Report [54] and draws on theoretical expanations from the Black Report about the relationship between social inequality and ill-health. This storyline is represented by one caper from 1990 [65], which used secondary data (birth data from England Wales, 1980-1986) to develop a theoretical model of how social class may affect psychosocial mediators – emotional, social, and cognitive factors – which may in turn influence pregnancy outcome, either directly or mediated through behaviours and coping strategies. The proposed model suggests that material deprivation results in more negative life events while also reducing social support, and access to education and information. Stressful life events, unmitigated by social support, create stress, anxiety, depression, low self-esteem. Poor education, or access to information, lead to a lack of knowledge and to deleterious beliefs and attitudes. The combined emotional and cognitive effects produce coming strategies and behaviours that increase the risk of negative pregnancy outcomes (i.e. smoking). [65]	A factor influencing health Inequalities can be seen to affect health via increasing psychosocial stress, which can then directly impact on health and also induce health- limiting behaviours	N=1 [65]
Audit Reports and Confidential Enquiries	Inter- disciplinary (Epidemiology, Obstetrics, Paediatrics, Midwifery)	Audits, Reports and Confidential Enquiries provide knowledge not always thought of as research, nevertheless it usefully uses routinely collected data to examine timetrends. As a tradition it incorporates a variety of approaches including epidemiology, economics and health policy and may be further informed by qualitative data and/or expert opinion. It includes 1992-2003 CESDI (Confidential Enquiry into Stillbirths and Deaths in Infancy), 2003-2011 CMACE (Centre for Maternal and Child Enquiries), and 2011 onwards MBRACE-UK (Mother and Babies: Reducing Risk through Audits and Confidential Enquiries across the UK).	The Audit, Reports and Confidential Enquiries storyline [66-74] builds on over 50 years of local and national reporting of maternal and infant deaths. A key feature of these reports is the presentation of stillbirth rates at national, reporting of maternal and infant deaths. A key feature of these reports is the presentation of stillbirth rates at national, reporting of maternal and infant deaths. A key feature of these reports is the presentation of stillbirth rates at national, reporting on all levels and the subsequent comparisons between geographical units and benchmark averages. Over the years these processes were modified and refined into the national Confidential Enquiry scheme [66,67] and, more recently, under the banner MBRACE-UK (Mother and Babies: Reducing Risk through Audits and Confidential Enquiries across the UK) [68-70]. Although we identified more than 20 national reports only five explored the relationship between inequalities and stillbirth [66-70] with the majority focusing on 'avoidable' health system and clinical failures. Where inequalities and stillbirth were identified they were discussed in relation to lifestyle factors (smoking, excess alcohol consumption, obesity) or regional or ethnic disparities associated with ingreased stillbirth risk. Four regional reports or audits from the West Midlands [71-74] attempt to look at stillbirth and inequalities explicitly by equating higher indices of multiple deprivation (IMD's) with increased stillbirth rates. These reports were more nuamed and identify a number of social and medical risk factors that could be screened for (alone or in combination) to predict risk destillbirth (e.g. unemployment, inappropriate housing, unsupported/difficult family circumstances, emotional factors/anxiety, material age <20 yrs or > 40 yrs, obesity, smoking, consanguinity, history of mental health issues). The authors of these reports also highlight fetal growth restriction (FGR) as a potential predictor of stillbirth in deprived communities.	Regional variations in stillbirth rates with recognition of differences between areas of deprivation (high and low) and ethnicity (White and Black & Asian populations).	N=9 [66-74]
Fetal-Maternal Medicine	Medicine	Maternal-fetal medicine is a subspecialty of obstetrics. Its focus is on 'high risk' pregnancies, including women who have a pre-existing illness or a pregnancy-induced illness, and congenital abnormalities It draws on and is related to perinatal epidemiology. The clinical focus includes preterm birth prevention, screening for fetal growth restriction, and placental histopathology.	The Fetal-maternal medicine storyline [75-78] included a study reporting that women living in areas of highest deprivation (IMD 1) were more likely to experience fetal growth restriction compared to women living in the least (IMD 29). [75] Approximately 46% of these women smoked, compared to 7% in the least deprived. The study concluded that targeted antenatal management was key to stillbirth prevention amongst women living in the most deprived areas. This tradition also offered time interlinked publications, which suggested that maternal ethnicity was associated with fetal loss at different gestations White women had relatively more stillbirths (>24 weeks gestation) and Black women relatively more late intrauterine fetal deaths (20-23 week\$ gestation) [76-78]. There was a higher risk of ascending genital infection for Black mothers relative to women from other ethnic groups. This was a relatively common cause for early intrauterine fetal death, peaking at around 22 weeks, [78].	A risk factor for stillbirth and depending on the type of study, may be included as a covariate in the analysis.	N=4 [75-78]
Nursing and Midwifery	Nursing and Midwifery	Nursing and Midwifery research draws from positivist and interpretative paradigms, utilising a range of quantitative and qualitative methods. This tradition has made a significant contribution to the body of knowledge about stillbirth and bereavement care.	Only one mixed-method single-site study was identified as characteristic of this tradition. [79] It showed no statistically significant association between stillbirth and maternal ethnicity, but found more perinatal deaths in deprived at as. Qualitative interviews with White British, Pakistani, and Bangladeshi women identified health beliefs and behaviours common on all ethnic groups. These included little awareness of what to do about risk factors such as reduced fetal movements ("two days I delayed because I don't know what I need to do") and anxieties about being a burden to overstretched maternity services (they could do without me taking up a bed, taking up their time, you put yourself at a lower scale than everyone else.") Health professionals perceived they had communicated information to women about stillbirth risks and the importance of seeking prompt care. Professionals did not view any particular ethnic group to be higher risk, but were aware of how cultural norms and/or living in poverty can restrict access to timely care ("Some of them [Asian women: Pakistani and Bangladeshi] are beholden on their partners to get them there) ("It doesn't matter whether they're Asian or whatever they are They don't have transport and they don't have money they don't have access to actually get here").	An additional vulnerability, and considered in relation to the importance of providing culturally appropriate care.	N=1 [79]

## Synthesis across traditions

#### Meta-theme 1: Something must be done

Across time and research tradition the prevailing message was for action on inequalities and stillbirth. From the earliest included paper in Social Medicine that concludes 'there is still much to be done' [26] to a Public Health paper in The Lancet Ending Preventable Stillbirth Series 2016 that states 'programmes at community and country level need to improve health in disadvantaged families to address these inequalities' [58] the message is clear. The call to do something stems from persistent evidence of a social gradient coupled with perceptions of insufficient progress in diminishing stillbirth rates in the UK. In some research traditions stillbirth was used as an indicator of societal health, with references to the particularly low stillbirth rates achieved in Scandinavia commonplace. Despite the persistence of studies reporting the same or similar risk factors and the continuation of the social gradient exactly what kind of 'something should be done' is less clear. Evidence of effectiveness was absent for interventions at specific time-points, inter-generationally, at scale or targeted to social groups. The absence of stillbirths in inequalities reduction targets post-Acheson was identified as a specific barrier to action [71].

## Meta-theme 2: Problems of precision

Our meta-narrative approach highlighted how much of the challenge in seeking to act on inequalities and stillbirth lies in the lack of consensus and inherent complexities inherent to both. While there was persistent evidence of associations between stillbirth risk and poverty, and stillbirth risk, ethnicity and poverty, it was not possible to estimate the potential gain on stillbirth reduction if action was taken to reduce inequalities, because of problems with data availability and comparability. There were problems of precision in stillbirth definition and problems of precision in inequalities measurement. The traditions rooted in medical science offered the most analytic tools for defining when stillbirths happened (antepartum, intrapartum), at what gestation (early preterm, late preterm, term), and why in terms of clinical factors (classification according to ReCoDe, Wigglesworth, Aberdeen etc), but these definitions were not used consistently, and they rarely considered social inequalities.

Further problems of precision arose from how inequalities were variously conceived and measured, even when they were taken into account. In traditions informed by the social sciences, inequalities were broadly conceptualised as a set of social relations (rather than a variable/s), which opened up lived experience, multiple risk factors/ interactions between them, and consideration of the relationship between structure and agency in health and lifestyle. Further conceptual considerations arise from this, including socio-economic status/social class (an individual measure of inequality) based on occupation alone or in combination with income, education and culture (Social Medicine, Epidemiology, Medical Sociology, Public Health). The problem of how best to measure disadvantage was apparent across time. The artefact explanation for inequalities (which considers to what extent they are a construct of the measurement process) was particularly critical of the now defunct Registrar General's Scale. [31,50,51,54,55].

Deprivation (an area measure of inequality) was conceptualised according to the tool used to define it for which there was no consensus. Tools used included the Townsend deprivation index, Carstairs and Morris index, Jarman Deprivation Scores and the Index of Multiple Deprivation (IMD)). The problems associated with using crude categories to define ethnicity (i.e. white, Black, Asian) were also considered (Epidemiology, Medical Sociology, Public Health) and the complexities therein (i.e. benefits of more subtle classifications incorporating country of birth such as British Asian), including how such classifications are only proximate guides to experiences, practices, beliefs and lifestyles. In 1993, a matrix of country of birth, nationality, language group, religious affiliation, and (where appropriate) region, caste and subcaste was proposed by Andrews and Jewson to test the combining variables, as well as suggesting a more fine-grain exploration of major variables if used as part of a national dataset. [52]

# Meta-theme 3: Moving from associations to intersectionality and intervention

All the traditions included in this review report evidence of associations between living in poverty and increased risk of stillbirth. However, despite more than 70 years of

research equating inequality with increased stillbirth risk "any detailed study of why this should be so is surprisingly sparse" [50;p.393] This theme attempts to shine some 'light on the most appropriate times to provide support and the form(s) that such support should take.' [42:p11] To begin to address the need for intervention, one recent study triangulated epidemiological data with what women said (qualitative data). [79] In so doing it showed how the interactions between education level, socioeconomic status, cultural needs, language barriers, knowledge, likeliness to seek help, and assumptions by healthcare staff interact to make (or diminish) stillbirth risk in the current maternity care system. While, that study was the first study to claim an intersectionality approach, most publications across the research traditions suggest that further exploration of the interactions between risk factors, and within specific groups, is warranted.

Most of the contributory risk factors identified in this review are already well known and have been for some time. As summarised in Figure 3 risk factors for stillbirth encompass biological, clinical, behavioural, health service and social factors. Figure 3 provides a model from which to test the associations between factors, which is built on interdisciplinary evidence of the clinical causes of stillbirth, theories of natural and social selection, cultural/behavioural/lifestyle explanations, area effects, materialist/structuralist explanations and availability, access and quality of care. While some studies proposed antenatal screening for a combination of social factors (i.e. non-English speaking, unemployed household) in combination with behavioural factors (i.e. smoking) and clinical factors (i.e. previous IUGR), there was little consensus on specific factors, timing, or outcome if social conditions remain the same. [39,40,46]

## DISCUSSION

This review highlights that research investigating what might work to reduce inequalities and stillbirth in the UK is underdeveloped. We identified nine research traditions in the field but, with the exception of epidemiology, these traditions had few studies within them. Across all traditions, epidemiological data persistently suggests that membership of a lower socio-economic group (as measured by an individual's occupation), or residing in a disadvantaged community (as measured by local area deprivation) is associated with increased incidence of stillbirth when compared with more socially advantaged counterparts. However, there was a paucity of research investigating why this should be so, despite repeated calls for action. A few studies found no association between living in an area of deprivation and increased stillbirth risk. Why this was so is also unclear. This review shows that the field is not only complex, but also dynamic, with the respective components (stillbirth per se and inequalities per se) beset by conceptual and methodological challenges. In terms of advancing understanding about the complexity of the interactions between factors associated with increased stillbirth risk this review is limited. Moreover, we found no studies of interventions targeted to reduce stillbirth in specific social groups or communities. Nonetheless, what this review does add is that stillbirth is a useful marker of success in addressing inequalities. It provides a cross-disciplinary foundation from which to develop and stimulate hypotheses about the relative influence of biological, clinical, behavioural, health service and social factors on birth outcomes and the interactions between these various determinants to inform future interventions.

#### Strengths and limitations

To the best of our knowledge this is the first study to use a meta-narrative approach to investigate the relationship between inequalities and stillbirth. We adhered to the RAMASES standards for metanarrative review to ensure fidelity with the methodology. We used a multipronged approach to retrieving sources that included exploratory searches, systematic searches, hand searches, expert opinion, and forward and back chaining, which gave us a broad capture of relevant documents. By limiting the review to UK-based studies only, we were able to focus with greater acuity on the commonalities and contestations between research traditions. However, excluding

studies from other countries may have led us to miss important research on the relationship between stillbirth and inequalities of relevance both in the UK context and globally. The quality of some of the included sources in this review may also be considered an important limitation.

#### Relationship of findings to other research

The current abundance of research investigating stillbirth prevention and bereavement care in the UK is a recent development as efforts to break the silence that has traditionally surrounded stillbirth have gained momentum and international ambition to reduce stillbirth has intensified over the last decade. [3-4,6,80-83] This goes some way to explain why the field is underdeveloped in comparison to the wider health inequalities literature on mortality and social gradient. We were surprised to find no intervention studies, although there is an acknowledged paucity of evaluations of interventions to reduce inequalities in health in general. [84-87] In the international literature, public health interventions seeking to reduce stillbirth are also sparse. The few that do exist include a food supplementation programme, which was offered to low-income women in the USA, [88] and a study looking at household air pollution in India, where wood and kerosene cooking fuel, more commonly used in low-income households, is known to be associated with stillbirth. [89] However, neither of these address the underlying structural components of disadvantage.

#### Implications for clinicians and policymakers

This review suggests that addressing inequalities as a component of stillbirth prevention in the UK demands intervention at many levels. The paucity of directly relevant research to the question of stillbirth prevention means policy makers must look towards what works to reduce inequalities for other related causes of death (i.e. sudden infant death, cardio-vascular disease and cancer). Health inequalities theory advocates intervening at specific time points during the life course (i.e. pregnancy and the early years), interventions that have impact over time (i.e. intra-generational and inter-generational), interventions at scale (i.e. national policies) and interventions targeting specific groups (i.e. ethnic minorities and lower social classes). Addressing nutrition, service uptake and the wider social determinants of health may have knock on effects on many clinical outcomes, including stillbirth. [90] Scotland's *Early Years Collaborative* that encompasses cross-sector interventions at the level of individuals,

groups, organisations and society, includes a specific stillbirth reduction target. [87] In the absence of a hierarchy of causation among these complex effects stillbirth specific research is well justified, as long as it is embedded in implementation, public health and caring for and about people.

#### Unanswered questions and future research

It was not possible within or across traditions in this review to determine the potential gain of inequalities and stillbirth reduction. The field would benefit from a national consensus for routinely collected data and future research at population level. MBRRACE-UK, the RCOG and NHS England now have a high level of precision in stillbirth definition and national data capture. Since 2014, MBRRACE-UK has consistently used the Children in Low-income Families Local Measure. [5] There is also a simultaneous need for qualitative research that gets behind classificatory system labels to the lived realities of groups and communities. This review highlights there have long been important differences between communities and place that, for example, the classification Black, Asian, and Minority Ethnic (BAME), or indices of multiple deprivation (IMD) can conceal.

Most of the factors associated with inequalities and stillbirth identified in this review are already well known, and have been for some time. The findings of the review suggest that there is also likely to be advance from looking at these well-known factors anew. For example, the reasons reported as to why women delayed seeking care for reduced fetal movements in this review resonate with the findings of earlier reviews of antenatal care in general. [91-92] Similarly, studies of smoking behaviours, influence of social and community networks, the conditions in which people live, and the impact of current UK smoke-free policies that were identified on the periphery of studies included in this review, demand cross-disciplinary consideration in future strategies for stillbirth prevention. [93-95] Not least because, these particular components of antenatal care already feature as part of stillbirth reduction initiatives, but to-date, have had limited success. [83]

The role of social factors, modifiable lifestyle behaviours, and antenatal interventions in stillbirth prevention are current research priorities identified by the stillbirth community. [96] The results of this review indicate that there is little effective work

across disciplines despite the long-recognised need for it. We recommend that the UK stillbirth research community overcome this by setting up a dedicated forum to promote intervention and implementation research in this area. The forum could have three roles: 1) Define the framework for future research by identifying the ways in which disciplines should interact; 2) Develop data standards for information relating to stillbirth and inequalities; 3) Develop and promote the intervention and implementation research, policy and practice agenda relating to stillbirth and inequality.

#### Conclusion

The UK government's current ambition is to halve the national stillbirth rate by 2025. Research investigating and, critically, addressing inequalities and stillbirth in the UK is underdeveloped. This is despite repeated evidence of an association between stillbirth risk and poverty, and stillbirth risk, poverty and ethnicity. A specific research forum is required to lead the development of research and policy, which can harness multiple relevant research perspectives and address the intersections between different policy areas. This review not only unifies calls for action, by connecting multi-disciplinary insight into these complexities, challenges and opportunities, it provides a starting point for a novel transdisciplinary response.

## Figure legends

Figure 1: RAMASES-PRISMA Diagram

Figure 2: Timeline of included studies by research tradition and the stillbirth rate in

England and Wales 1945-2017

Figure 3: Factors associated with inequalities and stillbirth I

#### Supplementary files

Supplementary file 1: PRISMA Checklist

Supplementary file 2: Protocol

Supplementary file 3: Example exploratory searches

Supplementary file 4: Example systematic search strategy

Supplementary file 5: Screening tool

Supplementary file 6: Characteristics of included sources

**Author contributions:** CK, DR, MT and CS designed the review with input from SD. NC and KF conducted the searches, identification and screening with agreement by consensus of all authors on final inclusions. NC, KF and CK extracted data, and agreed initial storylines and final meta-themes with review by SD, DR, MT and CS. CK wrote the first draft of the paper. All authors read, commented and approved the final manuscript.

**Data sharing statement:** This is a review. Original research data is contained in the included studies. Data interpretation is contained in the manuscript. Further information can be obtained from the corresponding author.

Acknowledgements: Rob Rawcliffe, UCLanPrint for his design input into Figure 3.

#### References

- Commission on Social Determinants of Health (2008). Closing the gap in a generation: health equity through action on the social determinants of health. Final Report of the Commission on Social Determinants of Health. Geneva, World Health Organization.
- Blencowe H, Cousens S, Jassir FB et al. National, regional, and worldwide estimates of stillbirth rates in 2015, with trends from 2000: a systematic analysis. The Lancet Global Health. 4 (2):e98-e108 (February 2016) DOI: 10.1016/S2214-109X(15)00275-2
- 3. Mullen Z, Horton R (2011) Bringing stillbirth out of the shadows. The Lancet Stillbirth Series. DOI: http://dx.doi.org/10.1016/S0140-6736(11)60098-6
- 4. Froen JF, Friberg IK, Lawn JE et al. Stillbirths: Progress and unfinished business. The Lancet Ending Preventable Stillbirth Series. 387 (10018); p.574-586
- Draper ES, Gallimore ID, Kurinczuk JJ, Smith PW, Boby T, Smith LK, Manktelow BN, on behalf of the MBRRACE-UK Collaboration. MBRRACE-UK Perinatal Mortality Surveillance Report, UK Perinatal Deaths for Births from January to December 2016. Leicester: The Infant Mortality and Morbidity Studies, Department of Health Sciences, University of Leicester. 2018.
- 6. GOV.UK New ambition to halve rate of stillbirths and infant deaths. <a href="https://www.gov.uk/government/news/new-ambition-to-halve-rate-of-stillbirths-and-infant-deaths">https://www.gov.uk/government/news/new-ambition-to-halve-rate-of-stillbirths-and-infant-deaths</a>. Last accessed 01/02/2019

- 7. Office of National Statistics Births in England and Wales: 2017. Live births, stillbirths and the intensity of childbearing, measured by the total fertility rate. <a href="https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarri-ages/livebirths/bulletins/birthsummarytablesenglandandwales/2017">https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarri-ages/livebirths/bulletins/birthsummarytablesenglandandwales/2017</a> Last accessed 01/02/2019
- 8. Department of Health. Safer Maternity Care. The National Maternity Safety Strategy Progress and next steps. November 2017.
- NHS England. Saving Babies' Lives Care Bundle. <u>https://www.england.nhs.uk/mat-transformation/saving-babies/</u> Last accessed 01/02/2019
- 10. Royal College of Obstetricians and Gynaecologists. Each Baby Counts. <a href="https://www.rcog.org.uk/eachbabycounts">https://www.rcog.org.uk/eachbabycounts</a> Last accessed 01/02/2019
- 11. National Perinatal Epidemiology Unit. Perinatal Mortality Review Tool. <a href="https://www.npeu.ox.ac.uk/pmrt">https://www.npeu.ox.ac.uk/pmrt</a> Last accessed 01/02/2019
- 12. Acheson D. (1998) Independent inquiry into inequalities in health report. The Stationery Office: LondonAcheson 1998.
- 13. Marmot M. Fair society, healthy lives: The Marmot Review: strategic review of health inequalities in England post-2010. Institute of Health Equity; 2010.
- 14. Public Health England (2017) Reducing health inequalities: system, scale and sustainability. London.
- 15. Dahlgren G, Whitehead M. Policies and strategies to promote social equity in health. Stockholm: Institute for future studies. 1991.
- 16. Greenhalgh et al 2004; Greenhalgh T, Potts HWWW, Wong G, Bark P, Swinglehurst D. (2009) Tensions and paradoxes in electronic patient record research: A systematic literature review using the meta-narrative method. Milbank Quarterly, 87 (4), 729-88
- 17. Greenhalgh T, Robert G, Macfarlane F, Bate P, Kyriakidou O, Peacock R. Storylines of research in diffusion of innovation: a meta-narrative approach to systematic review. Soc Sci Med 2005 Jul;61(2):417-30
- 18. Greenhalgh T, Robert G, Macfarlane F, Bate P, Kyriakidou O. (2004) Diffusion of innovations in service organisations: systematic literature review and recommendations for future research. Milbank Q 2004;82(4):581-629
- 19. Wong G, Greenhalgh T, Westhorpe G et al RAMESES publication standards: meta-narrative reviews. BMC Medicine 2013: 11
- 20. Moher D, Liberati A, Tetzlaff J, et al. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. PLOS Med 2009;6:e1000097
- 21. Kingdon C, Roberts D, Turner M, Storey C, Crossland N, Finlayson K, Downe S. (2017) Inequalities and stillbirth: a meta-narrative review. PROSPERO 2017 CRD42017079228
- 22. Kuhn, T. (1962) The structure of scientific revolutions. University of Chicago University Press, Chicago.

- 23. Betrán AP, Temmerman M, Kingdon C, Mohiddin A, Opiyo N, Torloni MR et al. (2018) Interventions to reduce unnecessary caesarean sections in healthy women and babies. The Lancet. ISSN 0140-6736
- 24. Critical Appraisal Skills Programme (CASP). CASP Checklists. 2013. http://www.casp-uk.net/#!casp-tools-checklists/c18f8.
- 25. Walsh D, Downe S. Appraising the quality of qualitative research. Midwifery. 2006;22:108-119.
- 26. Baird, D. (1945) The Influence of Social and Economic Factors on Stillbirths and Neonatal Deaths. Journal of Obstetrics and Gynaecology. Vol 52 (4): 339-365
- 27. Morris JN, Heady JA (1955) Social and biological factors in infant mortality: Objects and methods. The Lancet. 268(6859):343-349
- 28. Heady, JA, Daly C, Morris JN. (1955) Social and Biological Factors in Infant Mortality. II. Variation of Mortality with mother's age and parity. The Lancet. 268(6860):395-7
- 29. Daly, C., Heady, J. A., & Morris, J. N. (1955). Social and biological factors in infant mortality: 3. The effect of mother's age and parity on social-class differences in infant mortality. The Lancet. 265 (6861): 445-448
- 30. Heady, JA, Stevens, CF, Daly, C, Morris, JN. (1955) Social and Biological Factors in Infant Mortality. The independent effects of social class, region, the mother's age and her parity. The Lancet. 265 (6862): 499-503
- 31. Illsley, R. (1955) Social class selection and class differences in relation to stillbirths and infant deaths. British Medical Journal. Dec 24; 2(4955): 1520-1524
- 32. Kincaid JC, (1965) Social Pathology of Foetal and Infant Loss. Br Med J. 1965 Apr 17;1(5441): 1057–1060.
- 33. Clarke M, Clayton DG, Mason ES, MacVicar J. 1988 Asian mothers' risk factors for perinatal death: the same or different? A 10 year review of Leicestershire perinatal deaths. BMJ: British Medical Journal, 297(6645), 384.
- 34. Guildea, Z. E. S., Fone, DL, Dunstan, FDJ, Sibert, J. R., Cartlidge, P (2001). Social deprivation and the causes of stillbirth and infant mortality. Archives of Disease in Childhood 84 (4):307-310. 10.1136/adc.84.4.307
- 35. Flenady V, Koopmans L, Middleton P, Froen JF, Smith GC, Gibbons K, et al. (2011) Major risk factors for stillbirth in high-income countries: a systematic review and meta-analysis. Lancet. 377 (9774):1331–40
- 36. Seaton SE, Field DJ, Draper ES, Manktelow BN, Smith GC, Springett A, et al. (2012) Socioeconomic inequalities in the rate of stillbirths by cause: a population-based study. BMJ Open 2(3). doi: 10.1136/bmjopen-2012-001100
- 37. Zeitlin, J Mortensen, L. Prunet, C, Macfarlane A, Hindori-Mohangoo AD, Gissler M, et al. (2016) Socio-economic inequalities in stillbirth rates in Europe: measuring the gap using routine data from the Euro-Peristat Project. BMC Pregnancy & Childbirth 16:15

- 38. Penn N, Oteng-Ntim E, Oakley LL, Doyle P. (2014) Ethnic variation in stillbirth risk and the role of maternal obesity: analysis of routine data from a London maternity unit. BMC Pregnancy and Childbirth 14:404-410
- 39. Chitty LS, Winter RM. (1989) Perinatal mortality in different ethnic groups. No evidence for increasing inequality. Archives of Disease in Childhood, 64:1036-
- 40. Gray R, Bonellie SR, Chalmers J, Greer I, Jarvis S, Kurinczuk JJ, et al. (2009) Contribution of smoking during pregnancy to inequalities in stillbirth and infant death in Scotland 1994-2003: retrospective population based study using hospital maternity records. BMJ 2009;339:b3754
- 41. Rush D, Cassano P. (1983) Relationship of cigarette smoking and social class to birth weight and perinatal mortality among all births in Britain, 5-11 April 1970. J Epidemiol Community Health. 1983 Dec;37(4):249-55.
- 42. Weightman AL, Morgan HE, Shepherd MA, Kitcher H, Roberts C, Dunstan FD. (2012) Social inequality and infant health in the UK: systematic review and meta-analyses. BMJ Open 2012;2:e000964. doi:10.1136/bmjopen-2012-000964.
- 43. Wood AM, Pasupathy D, Pell JP, Fleming M, Smith GCS. (2012) Trends in socioeconomic inequalities in risk of sudden infant death syndrome, other causes of infant mortality, and stillbirth in Scotland: population based study. BMJ 2012;344:e1552
- 44. Bambang S, SpencerNJ, Logan S, & Gill L. (2000). Cause-specific perinatal death rates, birth weight and deprivation in the West Midlands, 1991–1993. Child: Care, Health and Development, 26(1):73-82. doi:10.1046/j.1365-2214.2000.00152.
- 45. Khalil A, Rezende J, Akolekar R, Syngelaki A, & Nicolaides KH. (2013). Maternal racial origin and adverse pregnancy outcome: a cohort study. Ultrasound in Obstetrics & Gynecology, 41(3):278-285. doi:10.1002/uog.12313
- 46. Sutan R, Campbell D, Prescott GJ, Smith WCS (2010) The risk factors for unexplained antepartum stillbirths in Scotland, 1994-2003. Journal of Perinatology 30: 311–318.
- 47. Murrells TJ, Catford JC, Smith TM, & Machin D. (1985). The use of logit models to investigate social and biological factors in infant mortality. II: Stillbirths. Statistics in medicine, 4(2):189-200.
- 48. Neasham D, Dolk H, Vrijheid M, Jensen T, Best N. (2001) Stillbirth and neonatal mortality due to congenital anomalies: temporal trends and variation by small area deprivation scores in England and Wales, 1986-96. Paediatr Perinat Epidemiol. Oct;15(4):364-73.
- 49. Antonovsky A, Bernstein J. (1977) Social class and infant mortality. Soc Sci Med. 11:453-470
- 50. Macintyre S. (1986) The patterning of health by social position in contemporary Britain: Directions for sociological research. Soc Sci Med. 23:393-415

- 51. Bloor M, Samphier M, Prior L. (1987) Artefact explanations of inequalities in health: an assessment of the evidence. Sociology of Health & Illness. 9:231-264
- 52. Andrews A, Jewson N. (1993) Ethnicity and infant deaths: the implications of recent statistical evidence for materialist explanations. Sociology of Health & Illness. 15:137-156
- 53. Kingdon C. (2016) Society. In Kierans C, Bell K, Kingdon C. Social and Cultural Perspectives on Health, Technology and Medicine. Abingdon, Routledge.
- 54. Black D, Morris JN, Smith C, Townsend P (1980) The Black Report. Townsend P, Davidson N. (Eds) 1992 Edn. London, Penguin books
- 55. Whitehead M. (1988) The Health Divide. Whitehead (Ed) 1992 Edn.; London, Penguin books.
- 56. Garcia R, Ali N, Papadopoulos C, Randhawa G. 2015 Specific antenatal interventions for Black, Asian and Minority Ethnic (BAME) pregnant women at high risk of poor birth outcomes in the United Kingdom: a scoping review. BMC Pregnancy and Childbirth.;15:226. doi:10.1186/s12884-015-0657-2.
- 57. Flenady et al 2011 Stillbirths: the way forward in high-income countries. The Lancet. 377 (9778); p.1703-1717
- 58. Flenady et al 2016 Stillbirths: recall to action in high-income countries. Lancet Series. The Lancet. 387 (10019): p.691–702
- 59. Pickton, M J. (1987) A Socio-Spatial Analysis of Perinatal Mortality in Greater Leicester
- 60. Slogget and Joshi (1998) Deprivation indicators as predictors of life events 1981–1992 based on the UK ONS longitudinal study. J Epidemiol Community Health;52:228–233
- 61. Joyce, et al (1999), Social class and census-based deprivation scores: which is the best predictor of stillbirth rates? Paediatric and Perinatal Epidemiology, 13: 269-277
- 62. Dummer et al, (2000) Stillbirth risk with social class and deprivation: No evidence for increasing inequality. Journal of Clinical Epidemiology 53 (2000) 147–155
- 63. Dickinson et al (2002) Deprivation and stillbirth risk in rural and urban areas. Paediatric and Perinatal Epidemiology 2002, 16, 249–254
- 64. Pattenden et al, (2011) Geographical variation in infant mortality, stillbirth and low birth weight in Northern Ireland, 1992–2002. J Epidemiol Community Health 2011;65:1159e1165
- 65. Rutter & Quine (1990) Inequalities in pregnancy outcome: A review of psychosocial and behavioural mediators, Social Science & Medicine 30 (5): 553-568
- 66. Confidential Enquiry into Maternal and Child Health. Perinatal Mortality Surveillance, 2004: England, Wales and Northern Ireland. London: CEMACH; 2006.

- 67. Centre for Maternal and Child Enquiries (CMACE) Perinatal Mortality 2009: United Kingdom. CMACE: London, 2011.
- 68. Draper ES, Kurinczuk JJ, Kenyon S, on behalf of MBRRACE-UK. MBRRACE-UK 2015 Perinatal Confidential Enquiry: Term, singleton, normally-formed, antepartum stillbirth. The Infant Mortality and Morbidity Studies, Department of Health Sciences, University of Leicester: Leicester, 2015.
- 69. Manktelow BN, Smith LK, Evans TA, Hyman-Taylor P, Kurinczuk JJ, Field DJ, Smith PW, Mielewczyk F, Draper ES, on behalf of the MBRRACE-UK Collaboration. MBRRACE-UK Perinatal Mortality Surveillance Report, UK Perinatal Deaths for Births from January to December 2013 Supplementary Report: UK Trusts and Health Boards. The Infant Mortality and Morbidity Studies, Department of Health Sciences, University of Leicester: Leicester, 2015.
- 70. Manktelow BN, Smith LK, Seaton SE, Hyman-Taylor P, Kurinczuk JJ, Field DJ, Smith PW, Draper ES, on behalf of the MBRRACE-UK Collaboration. MBRRACE-UK Perinatal Mortality Surveillance Report, UK Perinatal Deaths for Births from January to December 2014. The Infant Mortality and Morbidity Studies, Department of Health Sciences, University of Leicester: Leicester, 2016.
- 71. Gardosi J, Francis A (2005). Perinatal Mortality and Social Deprivation: West Midlands Trends 1998-2003. West Midlands Perinatal Institute
- 72. Jason Gardosi, Nicola Beamish, Andre Francis, Mandy Williams, Manjinder Sahota, Ann Tonks, Pat McGeown, Martin Hart. (2007) Stillbirth and infant mortality, West Midlands 1997-2005: Trends, Factors, Inequalities. West Midlands Perinatal Institute.
- 73. Francis A, El Sheikh A, Gardosi J. (2009) Stillbirths, Infant Deaths and Social Deprivation West Midlands 1997-2007/8. West Midlands Perinatal Institute.
- 74. Gardosi J. (2011) Perinatal Mortality, Social Deprivation and Community Midwifery. West Midlands Perinatal Institute.
- 75. Tang et al (2008) The relation between social deprivation and stillbirth causes. Arch Dis Child Fetal Neonatal Ed 2008;93(Suppl I):Fa1–Fa14
- 76. Man et al, 2016a. Stillbirth and intrauterine fetal death: role of routinehistopathological placental findings to determine causeof death. Ultrasound Obstet Gynecol 2016; 48: 579–584
- 77. Man et al 2016b Stillbirth and intrauterine fetal death: contemporary demographic features of >1000 cases from an urban population. Ultrasound Obstet Gynecol, 48: 591-595. doi:10.1002/uog.16021
- 78. Man et al 2016c Stillbirth and intrauterine fetal death: factors affecting determination of cause of death at autopsy. Ultrasound Obstet Gynecol, 48: 566-573.
- 79. Garcia R (2017) Perinatal mortality in Pakistani, Bangladeshi and White British mothers in Luton. Unpublished PhD Thesis. University of Bedfordshire. Luton.

- 80. Heazell AEP, Weir CJ, Stock SJE, et al. Can promoting awareness of fetal movements and focusing interventions reduce fetal mortality? A stepped-wedge cluster randomised trial (AFFIRM). BMJ Open. Aug 11 2017;7(8):e014813.
- 81. Heazell AEP, Li M, Budd J, Thompson JMD, Stacey T, Cronin RS, Martin B, Roberts D, Mitchell EA, McCowan LME. Association between maternal sleep practices and late stillbirth findings from a stillbirth case-control study. BJOG 2018; 125:254–262.
- 82. Siassakos D, Jackson S, Gleeson K, Chebsey C, Ellis A, Storey C. (2018) All bereaved parents are entitled to good care after stillbirth: a mixed-methods multicentre study (INSIGHT). BJOG 125 (2): 160-170
- 83. Widdows K, Reid HE, Roberts SA, Camacho EM, Heazell AEP. Saving babies' lives project impact and results evaluation (SPiRE): a mixed methodology study. BMC pregnancy and childbirth. Jan 30 2018;18(1):43.
- 84. Macintyre S. (1997) The Black Report and beyond: What are the issues? Soc Sci Med 44(6):723-745
- 85. Bambra C, Gibson M, Sowden A, Wright K, Whitehead M, Petticrew M. (2010) Tackling the wider social determinants of health and inequalities: evidence from systematic reviews. J Epidemio Community Health 64:284-291
- 86. Smith KE & Eltanani MK. (2015) What kinds of policies to reduce health inequalities in the UK do researchers support? Journal of Public Health. 37 (1):6-17
- 87. Burns H. (2015) Health inequalities: why so little progress? Public Health. 129: 849-853
- 88. Fingar, K., Lob, S., Dove, M., Gradziel, P., & Curtis, M. (2017). Reassessing the Association between WIC and Birth Outcomes Using a Fetuses-at-Risk Approach. Maternal & Child Health Journal, 21(4), 825-835. doi:10.1007/s10995-016-2176-9
- 89. Lakshmi, P. V., Virdi, N. K., Sharma, A., Tripathy, J. P., Smith, K. R., Bates, M. N., & Kumar, R. (2013). Household air pollution and stillbirths in India: analysis of the DLHS-II National Survey. Environmental research, 121, 17-22.
- 90. World Health Organization. Equity, social determinants and public health programmes.

  <a href="https://www.who.int/social\_determinants/publications/9789241563970/en/Last accessed 01/02/2019">https://www.who.int/social\_determinants/publications/9789241563970/en/Last accessed 01/02/2019</a>
- 91. Finlayson K, Downe S (2013) Why Do Women Not Use Antenatal Services in Low- and Middle-Income Countries? A Meta-Synthesis of Qualitative Studies. PLoS Med 10(1): e1001373. doi:10.1371/journal.pmed.1001373
- 92. Downe S, Finlayson K, Walsh D, Lavender T. 'Weighing up and balancing out': a meta-synthesis of barriers to antenatal care for marginalised women in high-income countries. BJOG 2009;116:518–529.
- 93. Popay J, Bennett S, Thomas C, Williams G, Gatrell A, Bostock L. (2003) Beyond 'beer, fags, egg and chips'? Exploring lay understandings of social inequalities in health. Sociol Health Illn. 25(1):1-23.
- 94. Robinson & Holdsworth 2013; Robinson, J., & Holdsworth, C. (2013). They don't live in my house every day: How understanding lives can aid understandings of smoking. Contemporary Drug Problems, 40, 47-70.

- 95. Passey, M. E., Longman, J. M., Robinson, J., Wiggers, J., & Jones, L. L. (2016). Smoke-free homes: what are the barriers, motivators and enablers? A qualitative systematic review and thematic synthesis. BMJ OPEN, 6(3). doi:10.1136/bmjopen-2015-010260
- 96. Heazell AEP, Whitworth MK, Whitcombe SW, et al. (2015) Research priorities for stillbirth: process overview and results from UK Stillbirth Priority Setting Partnership. Ultrasound in Obstetrics and Gynaecology. Vol 46 (6). P.641-647. https://doi.org/10.1002/uog.15738



Figure 1: RAMASES-PRISMA Diagram

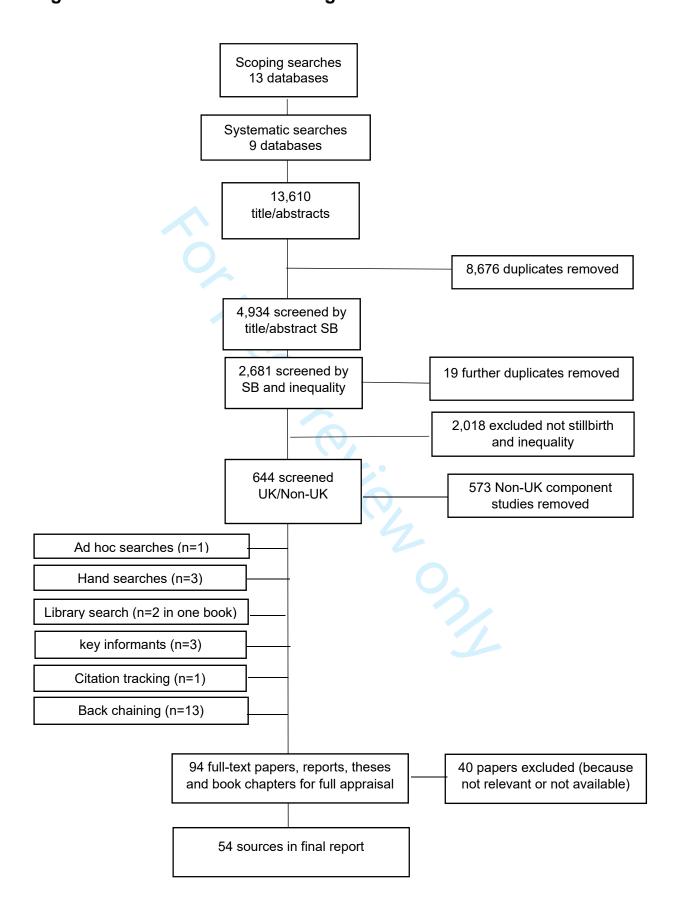


Figure 2: Timeline of included studies by research tradition and the stillbirth rate in England and Wales 1945-2017

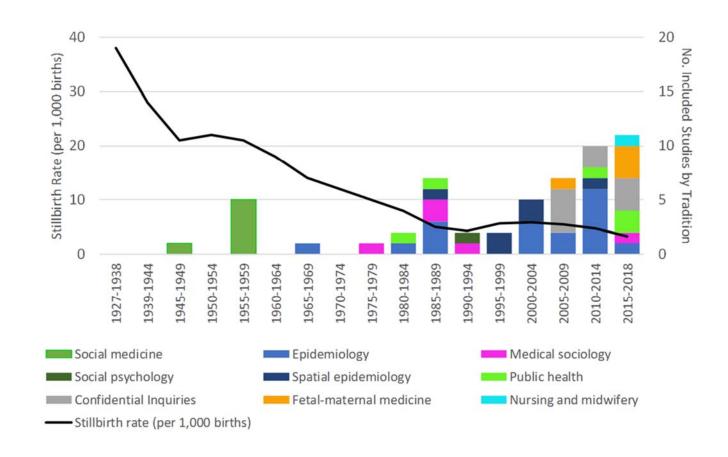
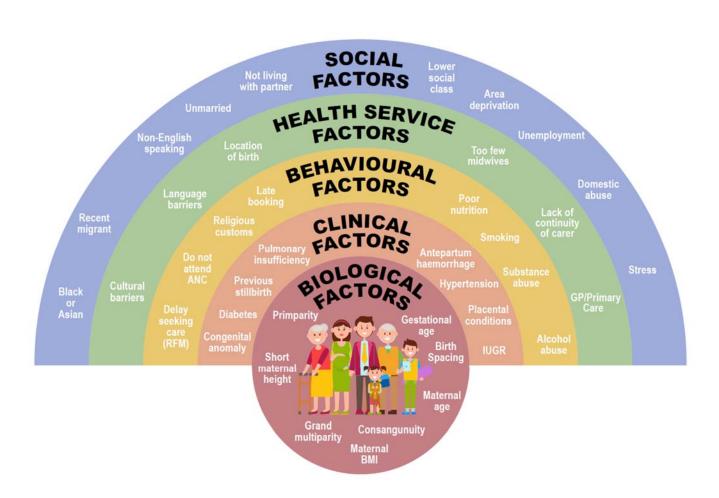


Figure 3: Factors associated with inequalities and stillbirth



Key: ANC (Antenatal Care); GP (General Practice), RFM (Reduced Fetal Movements); IUGR (Intra-uterine growth retardation also known as FGR – fetal growth restriction); BMI (Body mass index)

mjopen-2019-029672 on

## **Supplementary File 1: PRISMA Checklist**

Section/topic	#	Checklist item Se ept	Reported on page #	
TITLE		embe		
Title	1	Identify the report as a systematic review, meta-analysis, or both.	1	
ABSTRACT		19. <sub>[</sub>		
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; gata sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	2	
INTRODUCTION		rom		
Rationale	3	Describe the rationale for the review in the context of what is already known.	4-5	
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	5	
METHODS	METHODS			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	2,5	
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	6-7	
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last search.	6-7	
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	Supplementary file 4	
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	6-7	
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independedly, in duplicate) and any processes for obtaining and confirming data from investigators.	7	
		For peer review only - http://bmiopen.bmi.com/site/about/quidelines.xhtml		

		BMJ Open Bon	
		-2019-029	
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	7
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	7
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	Not applicable
Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done including measures of consistency (e.g., I²) for each meta-analysis.	7-8

Section/topic	#	Checklist item	oaded fr	Reported on page #
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (publication bias, selective reporting within studies).	€.g.,	Not applicable
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	·//bmion	Not applicable
RESULTS	•		P	
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	mi com	Figure 1
Study characteristics	18		7990 April 23 2024 hv gu	Table 1/ Supplementary File 6 as applicable to meta-narrative review methodology
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	Pst Dro	Not applicable
Results of individual studies	20	intervals, ideally with a forest plot.	ted by	Not applicable
		For neer review only - http://bmionen.hmi.com/site/about/quidelines.yhtml	copyright	

		BMJ Open ger	
		BMJ Open  BMJ Open	
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and Normal measures of consistency.	8-14 synthesis of results
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15). $\varphi$	Not applicable
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	Not applicable
DISCUSSION		er 20	
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, uses, and policy makers).	, 15
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review- (e.g., incomplete retrieval of identified research, reporting bias).	/el 15-16
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	15-18
FUNDING		tp://t	
Funding	27	Describe sources of funding for the systematic review and other support (e.g., support data); role of funders for the systematic review.	ply 3
		For peer review only - http://bmjopen.bmj.com/site/about/guidelines.xhtml	

#### **PROSPERO**

#### International prospective register of systematic reviews



#### Inequalities and stillbirth: a meta-narrative review

Carol Kingdon, Devender Roberts, Mark Turner, Claire Storey, Nicola Crossland, Kenneth Finlayson, Soo Downe

#### Citation

Carol Kingdon, Devender Roberts, Mark Turner, Claire Storey, Nicola Crossland, Kenneth Finlayson, Soo Downe. Inequalities and stillbirth: a meta-narrative review. PROSPERO 2017 CRD42017079228 Available from:

http://www.crd.york.ac.uk/PROSPERO/display\_record.php?ID=CRD42017079228

#### Review question

The aim of this review is to undertake an inter-disciplinary evidence synthesis (using a meta-narrative approach) to understand how structural factors (health system, living in poverty), lifestyle factors (smoking, obesity), and bio-clinical factors (maternal infection, non-communicable disease, fetal growth restriction) intersect to increase stillbirth risk to improve interventions to manage at-risk pregnancies.

The broad research question is: What is the relationship between inequality and stillbirth, how has this been studied, and with what effects?

The measurable objectives are:

- 1. To review the current body of knowledge of the relationship between inequalities and stillbirth across the natural and the social sciences;
- 2. To provide new insights into the interplay of biological, clinical, cultural and socioeconomic factors in increased stillbirth risk;
- 3. To explore the impact of interventions on inequalities;
- 4. To provide a narrative summary of this research for stakeholders tasked with reducing preventable stillbirth.

#### Searches

The following databases will be used to identify eligible studies for inclusion as part of the scoping searches:

MEDLINE® (Life sciences, medicine and biomedicine)

Embase® (Biomedical)

CINAHL® (Nursing and Allied Health Professionals)

PsycINFO® (Psychology and the behavioural and social sciences)

AJOL® (African Journals on-line)

Global Index Medicus

Popline (includes LILACS [Latin America and the Caribbean])

Historical abstracts

**Humanities International Complete** 

Race Relations Abstract

SocIndex

Lexis Library

Lexis Nexus

#### Types of study to be included

This is a meta-narrative review with no restrictions on type of study to be included.

#### Condition or domain being studied

Stillbirth is a profound human tragedy. The experience of stillbirth involves physical implications for the mother, together with intense grief and lasting psychological trauma for both parents and wider family.

## **PROSPERO**

#### International prospective register of systematic reviews



Studies have shown that stillbirth is associated with anxiety, depression and post-traumatic stress disorder in mothers, couples, siblings and grandparents.

The UK has the third highest rate of stillbirth among the 35 high-income countries of the world. Against a background of political, public and professional interest in halving the UK's stillbirth rate by 2030, and significant inter-disciplinary research effort to prevent or to improve maternity care services, there is longstanding evidence, across high-income countries, that the risk of stillbirth remains highest for the poorest families. Almost half (46%) of stillbirths remain of unknown cause, and inequalities in stillbirth risk demand more attention.

What is missing from current research agendas is an overarching synthesis of clinical and social science evidence to clarify the range of individual, social and biological mechanisms of increased stillbirth risk, the intersections between these mechanisms, and strategies to tackle them. There is also an urgent need to identify where interventions may decrease stillbirth risk for some, but may inadvertently increase inequalities for others. This meta-narrative review seeks to fill this gap in response to international interest in reviewing evidence from all relevant research traditions to address the unanswered questions surrounding stillbirth risk, and an NHS England Strategic Clinical Network need for this evidence to inform interventions to reduce the risk amongst women with complex social needs from vulnerable groups.

#### Participants/population

The population of interest for the purposes of our inclusion criteria is: Woman or Women or Mother or Mothers or Father or fathers or Family or Families or Husband or partner or partners or Parent or parents

Inclusion criteria

Any study design investigating stillbirth and inequality Any study in high-,middle- or low-income settings

Exclusion criteria

Any study published in non-English language Any study of pregnancy loss before 20 weeks Any study of perinatal loss in the neonatal period

Any study involving participants who had assisted conception (as identified by authors)

#### Intervention(s), exposure(s)

iflee extitioss, res socie exercimente de redeprivation or low income or poverty, or violence or abuse, or consanguinity, or ethnicity, or discrimination, or race or racism or racial, or migrant, or migration, or maternal age, or adolescence, or nutrition or obesity, or overweight or underweight, or smoking, or alcohol or drug or substance or chemical, or nonattendance, or neighborhood.

The interventions to be reviewed are:

growth chart or biomarkers or movement, or count, or support, or continuity of care or caseload.

The outcomes to be reviewed are:

stillbirth or perinatal death or pregnancy loss or miscarriage or fetal death or foetal death or feticide or foeticide or intrauterine death after 20 weeks pregnancy gestation.

#### NHS National Institute for Health Research

#### **PROSPERO**

#### International prospective register of systematic reviews

#### Comparator(s)/control

Not applicable.

#### Primary outcome(s)

Stillbirth incidence

Timing and effect measures
Effect measure exposure to inequality

#### Secondary outcome(s)

Not applicable

#### Data extraction (selection and coding)

Key results will be extracted, collated and grouped. Modified data extraction forms will be developed from existing published meta-narrative reviews and other types of review the team have been involved with in relation to stillbirth. Included studies will then be tabulated by tradition.

#### Risk of bias (quality) assessment

All articles that meet the inclusion criteria will be independently assessed by two researchers (NC,KF) and the principal investigator (CK) to minimize bias. Quality appraisal of quantitative studies will be undertaken using the appropriate checklists from the Critical Appraisal Skills Programme (CASP) Toolkit. Checklists formed from this process will be used to grade papers into categories A, B, C or D, with group A representing papers of the highest quality. Papers will be assigned to groups according to how many criteria fulfilled (i.e. Category A papers not fulfilled 0-1 of the marked criteria; Category D papers not fulfilled ?6 criteria). Qualitative appraisal will be undertaken according to the checklist described by Walsh and Downe and articles graded A – D in accordance with Downe and Simpson. A grades will be allocated to papers with no or few flaws where the study credibility, transferability, dependability, and confirmability is high; B, some flaws, unlikely to affect the credibility, transferability, dependability, and/or confirmability of the study; C, some flaws which may affect the credibility, transferability, transferability, dependability, and/or confirmability of the study; D, significant flaws which are very likely to affect the credibility, transferability, transferability, dependability, dependability. Any differences in appraisals will be discussed with the wider research team until a final decision is reached.

#### Strategy for data synthesis

The defining feature of meta-narrative reviews is their illumination of a complex topic area from multiple angles in a unified narrative synthesis. The synthesis stage involves summarizing each tradition in coherent individual accounts (including elements of data aggregation) and then comparing and contrasting the resultant meta-narratives to highlight similarities and differences. This process of contestation between the disciplines/traditions leads to higher order constructs and conclusions where recommendations can be made (i.e. in circumstances such as X, don't forget to think about y).

#### Quantitative studies

Quantitative meta-analysis will be used if the included studies are sufficiently homogeneous. In the event included quantitative studies are too heterogeneous they will be summarized narratively, separate from the qualitative evidence synthesis in the first instance, and then incorporated alongside into an overall typology of inequality and stillbirth.

#### Qualitative studies

A modified meta-ethnography approach will be used for qualitative evidence synthesis comprising 4 stages:1) Familiarization and quality appraisal; 2) Data extraction (direct participant quotations and author's thematic interpretation); 3)Coding into initial concepts; 4) Interpretative synthesis into emergent and final

#### NHS National Institute for Health Research

## PROSPERO

#### International prospective register of systematic reviews

themes to be presented in a typology.

Analysis of subgroups or subsets Not applicable

Contact details for further information

Carol Kingdon

ckingdon@uclan.ac.uk

Organisational affiliation of the review

University of Central Lancashire

Review team members and their organisational affiliations

Dr Carol Kingdon. University of Central Lancashire

Dr Devender Roberts. Liverpool Women's NHS Foundation Trust

Dr Mark Turner. University of Liverpool

Ms Claire Storey. International Stillbirth Alliance

Dr Nicola Crossland. University of Central Lancashire

Mr Kenneth Finlayson. University of Central Lancashire

Professor Soo Downe. University of Central Lancashire

Anticipated or actual start date

04 October 2017

Anticipated completion date

30 March 2018

Funding sources/sponsors

Stillbirth and Neonatal Death Society (SANDS) (Ref:RF510)

Conflicts of interest

None known

Language

English

Country

**England** 

#### PROSPERO

This information has been provided by the named contact for this review. CRD has accepted this information in good faith and registered the review in PROSPERO. CRD bears no responsibility or liability for the content of this registration record, any associated files or external websites.

# **Supplementary File 3: Example exploratory searches**

Databases	No. hits for "stillbirth"	Types of articles	Relevance to review questions
Art and Architecture source	10	poetry, feminism/women's studies, historical	Articles about impact and experiences of stillbirth rather than risk factors.
Criminal Justice Abstracts with Full Text	73	social work, women's studies, first person accounts/autoethnography	Articles about impact and experiences of stillbirth rather than risk factors.
Cochrane Library	535	Cochrane Reviews (n=83) Trials (423)	Two Cohrane reviews were tangenitaly relevant to research questions (interventions to reduce smoking in pregnancy, interventions to reduce domestic violence in pregnancy).
Humanities international complete	120	Family studies, images, religious aspects, first person accounts, responses to stillbirth, archaeology (burials), how stillbirth is conceptualised, abortion and morality/ethics	Articles about impact and experiences of stillbirth.
Race relations abstracts	6	differential medical risks by ethnicity, a couple of articles relating to social risk factors by ethnicity	Social inequalities as a risk factors
Religion and Philosophy Collection	74	grief and loss, psychosocial identity, religious aspects, social responses, attachment theory, abortion,	Articles predominantly about abortion; not relevant to research questions
SocIndex	482	psychosocial identity, psychosocial impacts, mother and family experiences, social risk factors, grief, coping,	Social inequality risk factors. Consequences, impact and coping.
Lexis Library (legal)		Professional misconduct cases. Judgements about cases where a medical intervention/decision has led to stillbirth. Employment policies. Legal frameworks about healthcare delivery. Medical law.	Tangential to research questions.
JISC Historical Texts	0 for stillbirth, 308 for stillborn	Literature (66). Medical texts (15) - midwifery manuals, descriptive works on causes of death. 1600s, 1700s.	Tangential to research questions.
Historial abstracts (via Ebsco)	56 for stillbirth.	Rates of stillbirth at different historical times and places, about perceptions of stillbirth. Lots of hits come from journal Population Studies.	Modern history – potential for understanding how stillbirth has been conceptualised.

# **Supplementary File 4: Example Systematic Search**

# **Strategy terms**

**Population**: Woman or Women or Mother or Mothers or Father or fathers or Family or Families or Husband or partner or partners or Parent or parents

#### **Exposure**

<b>Exposure type</b>	Exposure search terms
economic	Inequalit* or Depriv* or 'low income' or Poverty or socioeconomic or 'social class'
violence & abuse	Violence or Abus*
substance use	Smok* or Alcohol or Drug or Substance or Chemical
ethnicity	ethnic* or Race or Racism or Racial* or Migrant or Migration
nutrition	nutrition or undernutrition or malnutrition
age	'maternal age' or adolescen*
obesity and weight	Obes* or overweight or underweight
space and place	neighborhood or neighbourhood or residence
appointments	"Appointments and Schedules"/ or nonattendance.mp
culture	Sociocultural or cultur*
consanguinity	Consanguin*

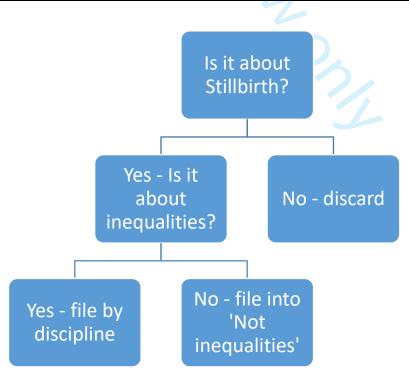
**Intervention**: growth chart' or biomarkers or movement or count or support or 'continuity of care' or caseload

**Outcome**: Stillbirth or stillborn or 'perinatal death' or 'pregnancy loss' or miscarriage or 'fetal death' or 'foetal death' or feticide or foeticide or 'intrauterine death'

**IVF-related terms** (ivf or fertil\* or infertil\* or 'assisted fertility' or inseminat\* or iui or 'embryo transfer')

# **Supplementary File 5: Screening Tool**

Answer the following questions in sequence:	Yes	No	Can't tell from abstract
1. Is it about stillbirth (defined as the death of a baby after 20 weeks in-utero until immediately before birth; excluding pregnancy loss < 20 weeks gestation and excluding neonatal deaths)?	Assess by Q2	Discard	If abstract doesn't define miscarriage, then discard.
If yes, then:	T.C	76	
<ul> <li>Is it about inequality and stillbirth?</li> <li>Working definition of inequality</li> <li>Inequality (?how circumstances of daily living disadvantage you, or structural)</li> <li>Socio-economic (as defined by authors so not necessarily NS-SEC), living in poverty, low income</li> <li>Deprivation (ditto – as defined by author - might not be IMD), neighbourhood, traveller, immigrant or refugee</li> <li>Minority ethnicity, religion, disability, young maternal age – member of marginalised group</li> <li>Risk factors (obesity, smoking, substance use, abuse etc) - may be linked to inequality but not enough on their own to include</li> </ul>	If yes, file by discipline	If no, then file as "Not about inequalities and stillbirth"	
If yes, then:			
What discipline is it from?			
3. Is it seminal?			



#### **Supplementary file 6: Characteristics of included sources**

Suppleme	entary file 6: Characteristics of included	sources	ВМЈ (	Open		mjopen-2019-029672		
Author and year	Aim and objectives	Design	Context	Population	Stillbirth definition	Stillbirth A classification (antepartum, intrapartum)	Type of inequality	Precision of inequality
Baird 1945 [26]	To examine the influence of social and economic factors on stillbirths and neonatal deaths	Descriptive	Scotland, compared with England and Wales	All maternities 1938- 1944	Undefined	Both Both Both	Socio-economic (individual)	Registrar Generals Scale of occupational class
Morris 1955 [27]	To describe the background and design of inquiry into stillbirths and infant deaths in England and Wales in 1949 and 1950	Review	England and Wales	Stillbirths and Infant deaths	Undefined		Socio-economic (individual)	Registrar Generals Scale of occupational class
Heady 1955 [28]	To describe the main features of variation of the stillbirth rate, and the neonatal and post-neonatal mortality rates, with two biological factors (age of mother and parity)	Descriptive	England and Wales	Births in 1949	Single, legitimate, stillbirth after the 28th week of pregnancy	Both Oaded	Socio-economic (individual)	Registrar Generals Scale of occupational class
Daly 1955 [29]	To consider the extent of such "biological" differences between the social classes, and the effect of these differences on the stillbirth and infant mortality rates of the social classes	Descriptive	England and Wales	Legitimate, livebirths and stillbirths born in 1949	Single, legitimate, stillbirth after the 28th week of pregnancy	Both http	Socio-economic (individual and place of residence)	Registrar Generals Scale of occupational class
Heady 1955 [30]	To investigate the "independence" of mother's age and her parity in relation to social class and region	Descriptive	England and Wales	Single, legitimate livebirths and stillbirths born in 1949	Single, legitimate, stillbirth after the 28th week of pregnancy	Both //bmjop	Socio-economic (individual)	Registrar Generals Scale of occupational class
Illsley 1955 [31]	To examine the processes of movement between social classes and the effects on stillbirths and infant death statistics	Descriptive	Aberdeen, Scotland	Married primiparae resident and delivered in Aberdeen between July, 1950, and December, 1954	Undefined	Both  Both	Socio-economic (individual)	Registrar Generals Scale of occupational class
Kincaid 1965 [32]	To explore changes in UK perinatal mortality rates over the period 1951-1961	Descriptive	Scotland, England & Wales	Birth records from national databases over periods ranging from 1948-1964	Undefined	Both On April 2	Socio-economic (individual)	Registrar Generals Scale of occupational class
Clarke 1983 [33]	(a) to measure the extent to which established risk factors, such as high parity and low social class, were risk factors in our locality; (b) to measure the extent to which other factors, such as the provision of medical services and demographic changes, might contribute to the risk of perinatal mortality; (c) to estimate the extent to which avoidable factors might be detected in the case histories of the perinatal deaths; and (d) to contribute our findings to the educational and planning processes of the local health authorities	Case-control	Leicestershire	All perinatal deaths In Leicestershire 1976- 1985	Undefined	guest.	Ethnicity	Broad categories (Asiar or European)
Guildea 2001 [34]	To investigate the relation between social deprivation and causes of stillbirth and infant mortality	Retrospective cohort	Wales	All births to women resident in Wales 1993–98	Late fetal deaths after 24 weeks gestation [stillbirth and neonatal death combined]	Protected by copyright	Socio-economic (area)	Townsend social and material deprivation score (unemployment, car ownership, owner occupation and

							mjopen-2019-029 <del>6</del> 72		
							672 on		overcrowding 1991 census)
Flenady 2011 [35]	To clearly identify important risk factors for stillbirth in high income countries	Systematic review and meta- analysis	High-income countries (including UK)	Studies from HICs	Death of a baby 20 weeks' gestation or more, or birthweight of at least 400 g	Both	12	ethnic origin, socioeconomic status, education level, young maternal age	Different in different studies
Seaton 2012 [36]	To assess time trends in socioeconomic inequalities in overall and cause-specific stillbirths in England over an 8-year period to aid understanding of each cause's impact on the deprivation gap and the overall stillbirth rate	Population-based retrospective study	England	All singleton stillbirths born to mothers resident in England between 01/01/2000 and 31/12/ 2007	Losses from the 24th weeks of gestation occurring in singleton infants	Both	September 2019. Downloaded from http://bmjopen.bmj.com/ on	Socio-economic (area)	Indices of Multiple Deprivation (IMD) 200- score (income; employment; health and disability; education, skills and training; barriers to housing; living environment and crime)
Zeitlin 2016 [37]	What is the magnitude of social inequalities in stillbirth rates in European countries?	Retrospective review of data from countries participating in the Euro-Peristat project	Europe (includes UK data)	Stillbirths and live births for the year 2010 from 29 countries	Intrauterine death after 22 weeks gestation or birthweight of 500 grams or more	Both	ed from http://	Socio-economic (individual)	Country level available data on educational level, and occupational group
Penn 2014 [38]	What is the association between clinical and socio-demographic factors and stillbirth, with a particular focus on ethnicity and obesity?	Cross-sectional	London	London population - (multi-ethnic)	24 completed weeks of pregnancy (with 28 wks (WHO definition) data in a supplementary table	Both	bmjopen.bm	Ethnicity and socio- economic (area)	Ethnicity general categories (i.e. Black, Asian), Area deprivation IMD score
Chitty 1989 [39]	To assess the contribution of lethal congenital malformations to perinatal mortality in different ethnic groups, to investigate the incidence of definite and probable autosomal recessive syndromes, and to estimate the possible effect of consanguinity.	Descriptive	North West Thames Region	All babies born between 1980-1985	Undefined	Both		Ethnicity	Ethnicity - broad categories (European, Pakistani and Indian)
Gray 2009 [40]	To study the effect of area deprivation on rates of stillbirth and infant mortality in Scotland for the 10 year period 1994-2003 and to establish whether smoking during pregnancy contributed to these gradients and, if so, to what extent.	Population based retrospective cohort study	Scotland	Live births and stillbirths 1994 - 2003.	24-44 weeks of pregnancy	Both	April 23, 2024 by	Socio-economic (area)	Deprivation according to Carstairs-Morris Index
Rush 1983 [41]	To re-explore the interrelation of smoking, social class, birth weight, and perinatal mortality in the total population of births in Britain born 5-11 April 1970.	Prospective cohort study	Britain	All livebirths and stillbirths 5th - 11th April, 1970	Undefined	Both	024 by g	Socio-economic (individual)	Registrar Generals Scale of occupational class
Weightman 2012 [42]	To assess the current evidence for the effects of social disadvantage on birth and infant outcomes for children born in the UK	Systematic review and meta- analysis	UK	UK studies	A baby born after the 24th week of pregnancy who does not show any signs of life	Unclear	Juest. Protected by copyright	Socio-economic (individual and area)	Different measures in different studies

			ВМЈ	Open			mjopen-2019-029672		
Wood 2012 [43]	To compare changes in inequalities in sudden infant death syndrome with other causes of infant mortality and stillbirth in Scotland, 1985-2008.	Retrospective cohort study	Scotland	Singleton births of infants with birth weight >500 g born at 28-43 weeks' gestation	An infant born showing no signs of life after 28 weeks gestation		on 12	Socio-economic (area)	Deprivation according to Carstairs Index (car ownership, unemployment, overcrowding, and social class)
Bambang 2000 [44]	To study the relationship between cause-specific perinatal deathrates, material deprivation and birthweight among births in 3 consecutive yearsin the West Midlands Health Region	Retrospective cohort study.	West Midlands Health Region	All live and stillbirths to mothers with addresses in the WMHR in 1991, 1992 and 1993.	Not given		September 2019	Socio-economic (area)	Townsend Deprivation Index
Khalil 2013 [45]	To examine the association between maternal-racial origin and a wide range of adverse pregnancy outcomes after adjustment for confounding factors in obstetric history and maternal characteristics	Retrospective cohort study.	London	Singleton pregnancies with a live fetus at 11 + 0 to13+ 6 weeks	Fetal deaths at or after 24 weeks	Both  Antepartun	. Downloade	Ethnicity	Ethnicity - broad categories (Caucasian, Afro-Caribbean, East Asian, South Asian, Mixed)
Sutan 2010 [46]	To determine the risk factors of unexplained antepartum stillbirth in Scotland from 1994 to 2003 and assess their value as a screening tool	Retrospective cohort study.	Scotland	All stillborn and live births in Scotland from 1994 to 2003	Not given	Antepartun	perom http://b	Socio-economic (area) and place (urban/rural)	Urban/Rural classified according to settlement size and remoteness; Deprivation according to Carstairs Morris Index
Murrells 1985 [47]	To examine changes in the data for stillbirth rates between 1949/50 and 1975	Retrospective cohort study	UK	Comparisons between data from 1949/50 and 1975	Not given	Both	mjopen.	Socio-economic (individual)	Registrar Generals Scale of occupational class
Neasham 2001 [48]	To investigate the variation of stillbirth and neonatal mortality due to congenital anomalies in relation to small-area measures of deprivation in a population-based study in England and Wales, 1986–96	Retrospective population based study	England and Wales	All births in England and Wales from 1986- 1996	On or after 24 weeks gestation	Both	bmj.com/	Socio-economic (area)	Carstairs Index
Antonovsky 1977 [49]	To examine the relationship between the components of infant mortality and social class by analysing the data available from infant mortality studies.	Review and commentary using secondary data	Western Europe and the United States	Uses data from infant mortality studies undertaken in Western Europe and the United States	Component of infant mortality - European nations require registration of stillbirths from 28 weeks gestation; most of United States from the 20th week of gestation.		rom http://bmjopen.bmj.com/ on April 23, 2024 by	Social class (individual)	Inconsistencies in reporting of class across countries discussed and complexity of occupation, and education as measures.
Macintyre 1986 [50]	To place debates (by social epidemiologists and medical sociologists among others attending to inequalities and health) in a broader and perhaps more traditional context: that of interest in observed social regularities in health, illness and death.	Review and commentary using secondary data	Britain	No specific population  - those where social patterning of health has been observed across the life-course	Unclear	Unclear	guest. Protected by c	Social class (occupational), gender, marital status, age, ethnicity, and area of residence	These six variables are t conceptualized as positions on dimensions of social differentiation which may be associated with particular patterns of life chances.

			BMJ	Open			jopen-		
							mjopen-2019-029 <b>6</b> 72		
Bloor 1987 [51]	To examine more thoroughly than was possible for Black and his colleagues, the possible role of an artefactual element in mortality data for explanations concerning health inequalities.	Review and commentary using secondary data	UK	No specific population draws on historical, theoretical and routinely collected statistical data reporting inequalities across the life-course	Unclear	Unclear	)672 on 12 September	Socio-economic	Examines social processes whereby statistics are produce
Andrews 1993 [52]	First objective, to draw attention to remarkable pattems of diversity and change revealed by recent official statistics for ethnic minority' infant deaths in England & Wales. Secondly, to suggest that these patterns represent a challenge to many orthodox explanations for inequalities in infant health offered by social scientists, not least materialist explanations. Thirdly, to propose that further development and evaluation of explanations is impeded by flaws in some of the indices and categories employed in the construction of data.	Review and commentary using secondary data	England and Wales	Uses OPCS data on ethnic inequalities in infant deaths	Unclear	Unclear	mber 2019. Downloa	Ethnicity	Argues ethnicity is as much a matter of the way in which boundaries are create and reproduced between groups as thinternal contents of ethnic cultures.
Kingdon 2016 [53]	To review the concept of society and discusses stillbirth as a social issue	Stillbirth used as a case study	Includes data from the UK	No specific population (stillbirth used as a case study)	A baby which has issued forth from its mother after the 24th week of pregnancy and which did not breathe or show any other signs of life' [UK definitionothers given]	Both	2019. Downloaded from http://bmjqpen.bmj.com/	Socio-economic, ethnic and gender	Multi-dimensional across place, time and culture
Black 1980 [54]	The first Government authorised attempt to explain trends in inequalities in health and to relate these to the policies intended to promote, as well as restore health.	Government Enquiry utilising official statistics	UK	Studies reporting inequalities based on occupational social class across the lifecourse	Not defined	Unclear	open.bmj.con	Socio-economic (individual)	Registrar Generals Scale of occupational class
Whitehead 1988 [55]	To update evidence following on from the Black Report (see above). The original aim of the Health Divide was to draw together, to summarise the wide-ranging new evidence, and to describe what had happened and could happen, in policy development.	Government Enquiry utilising official statistics	UK	Studies reporting inequalities based on occupational social class, and unemployment, income, housing, material and social deprivation in small areas, gender and ethnicity.	Undefined	Unclear	n∕ on April 23, 2024 by gi	Socio-economic	Registrar Generals Scale of occupational class
Garcia 2015 [56]	'What specific BAME maternity interventions exist for UK-based BAME women?'	Review and commentary	HICs including UK	Studies from HICs	Authors use an infant born with no life signs after 24 weeks gestation - however included studies in the review may have used different definitions	Unclear	west. Protected by	Ethnicity (BAME - Black, Asian and Minority Ethnic)	"BAME groups" - Asia Indian, Pakistani, Bangladeshi, Kashmir Black African, Black Caribbean, Arabian, Traveller [defined as a gypsy or Irish traveller

			ВМЈ	Open			mjopen-2019-029 <b>6</b> 72		
									Chinese, Mediterranean.
Flenady 2011 [57]	To present priority areas for stillbirth prevention, and interventions and research to address these priorities in high-income countries.	Review and commentary	HICs including UK	Studies from HICs	Death of a baby 500 g or more, or 22 weeks' gestation or more	Both	on 12 Septe	Socio-economic disadvantage	"Socioeconomic disadvantage" include maternal education, ethnicity
Flenady 2016 [58]	To summarise the status of stillbirths in HICs and suggest strategies to accelerate momentum in the reduction of stillbirths and to meet parents' needs when their baby is stillborn.	Review and commentary	HICs including UK	Studies from HICs	Late gestation stillbirth" as >28 weeks and "early gestation stillbirth" as occurring prior to 28 weeks gestation	Both	mber 2019. D	Socio-economic disadvantage	As above
Pickton 1987 [59]	To analyse, describe and explain the distribution of perinatal mortality within an urban area	Case-control with geographical component (statistical and cartographical)	Greater Leicester	All births in Greater Leicester in the study periods	Stillbirth (late fetal death) = death after 28 weeks gestation	Both	ownloaded from ht	Socio-economic (area and individual), ethnicity	Registrar Generals Scale of occupational class, employment housing, education level. Measures of ethnicity very broad (Asian, New Commonwealth).
Slogget 1998 [60]	To determine whether inequality in stillbirth risk between social strata has changed over time	Retrospective cohort study.	England and Wales	A random sample of more than 300 000 people enumerated at the 1981 census	Not defined	Unclear	September 2019. Downloaded from http://bmjopen.bmj.com/ or	Socio-economic (area and individual)	Deprivation was assessed by a "Townsend/Carstairs-like" index of four components – unemployment, car ownership, home ownership, lower employment status according to Registrar General.
Joyce 1999 [61]	To determine whether social class (individual level) or a census-based deprivation score (based on area of residence) is a better predictor of stillbirth rates	Unclear	South Thames (West)	All births 1993-95	Not defined	Unclear .	April 23,	Socio-economic (individual and area)	Social class using the Registrar General categories; area deprivation by Townsend score
Dummer 2000 [62]	To determine whether inequality in stillbirth risk between social strata has changed over time	Population based retrospective study	Cumbria compared with England & Wales data	For Cumbria all birth occurring between 1950-93: For E & W all births occurring between 1981-92	Two definitions given reflecting the change from 28 wks to 24wks that occurred in 1992	(	2024 by guest		
Dickinson 2002 [63]	To investigate whether stillbirth risk was higher, and the effect of deprivation on inequality in stillbirth risk more marked, in rural than in urban areas	Retrospective cohort study	Cumbria	Singleton births between 01/01/1950 – 30/09/1992 to mothers in the area now designated as Cumbria	A baby born dead after at least 28 weeks' gestation		t. Protected by copyright.	Socio-economic (individual, area and place)	Social Class - based of athers occupation Deprivation based on several scores includir IMD
	Farnas	r raviaw anly	http://bmionon	hmi com/sito/sk	oout/guidelines.x	; rhtml	copyright.		

			ВМЈ	Open		jopen-ź		
						mjopen-2019-029672		
Pattenden 2011 [64]	To analyse adverse pregnancy outcomes, concentrating on estimating the extent of geographical variation from large areas (approximately 400 000 persons population) to small areas (wards, approximately 100 times smaller)	Population based retrospective cohort study	Northern Ireland	Birth records from Northern Ireland over the years 1992-2002	24 weeks and over	Both 12 September	Socio-economic (area) and place	Deprivation according to The Noble Index (households receiving means-tested benefits Settlement size (rural areas, village, small town, medium town, large town, or city).
Rutter 1990 [65]	To review the literature on psychosocial factors in pregnancy outcome and to present a model which attempts to integrate the findings theoretically.	Theoretical	UK	UK	Not defined	201	Socio-economic (individual)	Registrar Generals Scale of occupational class
CEMCH 2006 [66]	To report on perinatal mortality statistics	Confidential Enquiry	England, Wales & Northern Ireland	England, Wales and NI Perinatal deaths during 2004	24th week of pregnancy	9. Downloa	Socio-economic (area), ethnicity	Ethnicity approximated Indices of Multiple Deprivation (IMD) scores
CMACE 2011 [67]	To report on perinatal mortality statistics	Confidential Enquiry	UK	UK Perinatal deaths in 2009	A baby delivered without signs of life after 23+6 weeks of pregnancy	Downloaded from http:	Socio-economic (area), ethnicity, and place (inferred regional and NHS Trust differences)	Ethnicity - broad categories; IMD data based on quintiles of deprivation; Employment status - simple yes/no
Draper 2015 [68]	Perinatal confidential enquiry carried out as part of the MBRRACE-UK programme of work, aim to focus on term, singleton, normally formed, antepartum stillbirths.	Confidential Enquiry	UK	Representative sample of stillbirths from those identified as eligible for review by MBRRACE-UK in April 2014	Term singleton, normally formed antepartum stillbirth (no precise definition given)	Antepartum/pmjopen.bmj.com/ on	Socio-economic (area and individual), ethnicity	Ethnicity - broad categories; IMD data based on quintiles of deprivation; Employment status - broad categories (employed/unemploye student; looking after home/family; permanently sick/disabled)
Manktelow 2015 [69]	To collect, analyse and report national surveillance data to stimulate and evaluate improvements in health care for mothers and babies.	Confidential Enquiry	UK	All births in the UK in 2013	Stillbirth: a baby delivered at or after 24+0 weeks gestational age showing no signs of life, irrespective of when the death occurred	April 23, 2024 by guest	Socio-economic (area), ethnicity, and place (inferred regional and NHS Trust differences)	Deprivation -Based on mothers' postcodes at time of delivery, using the Children in Low- Income Families Loca Measure; Ethnicity - Broad categories [white, mixed, Asian o British Asian, Black or Black British, other]
Manktelow 2016 [70]	To collect, analyse and report national surveillance data to stimulate and evaluate improvements in health care for mothers and babies.	Confidential Enquiry	UK	All births in the UK in 2014	A baby delivered at or after 24+0 weeks gestational age showing no signs of life, irrespective of when the death occurred [including	st. Protected by copyright	Socio-economic (area), ethnicity, and place (inferred regional and NHS Trust differences)	Deprivation -Based on mothers' postcodes at time of delivery, using the Children in Low- Income Families Loca Measure; Ethnicity -

			ВМЈ	Open		nijopen-zo i s-ozsorz		
					separate definitions for ante & intrapartum stillbirths]	029672 011		[white, mixed, Asian or British Asian, Black or Black British, other]
Gardosi 2005 [71]	summarise a recently completed analysis of stillbirth and infant mortality trends from 1998-2003 in the West Midlands; 2. assess the main categories contributing to these deaths; 3. analyse the trends of mortality associated with inequalities and the association with deprivation within different mortality subgroups.	Retrospective case-note audit	West Midlands	Perinatal mortality and infant mortality from the West Midlands over a 5 year period (1998 -2003)	> 24 weeks	Both V	Socio-economic	Indices of Multiple Deprivation (IMD)
Gardosi 2007 [72]	To explore trends in perinatal mortality rates (including stillbirths) in the West Midlands over the period 1997-2005	Retrospective case-note audit	West Midlands	All births in the West Midlands Health Region over the period 1997-2005	24.0 weeks of pregnancy	Rotu	Socio-economic (area), ethnicity	IMD (as above), ethnicity as recorded in notes
Gardosi 2009 [73]	To explore trends in stillbirths and infant deaths in the West Midlands over the period 1997-2007/8	Retrospective case-note audit	West Midlands	All births in the West Midlands Health Region over the period 1997-2007/8	24.0 weeks of pregnancy	Both Sign	Socio-economic (area)	IMD (as above)
Gardosi 2011 [74]	To understand the causes underlying perinatal mortality and develop strategies for prevention	Retrospective case-note audit and survey of community midwives	West Midlands	Perinatal deaths	Perinatal mortality – defined as a stillbirth or a death of a live born baby in its first week of life.	Antepartum Co.//DI.	Socio-economic (area), ethnicity	IMD (as above) Ethnicity - More subtle (Africa, African - Caribbean, Indian, Pakistani, Bangladeshi, Eastern European, Middle Eastern).
Tang 2008 [75]	To identify appropriate health targets by investigating associations between social deprivation and causes of stillbirth in Liverpool	Retrospective case-note audit	Liverpool	All stillbirths at local NHS Trust	Stillbirths from 24 weeks gestation.	Both a	Socio-economic (area)	Indices of Multiple Deprivation (IMD)
Man 2016a [76]	What is the role of placental histological examination in determining the cause of intrauterine death?	Descriptive	London	Intrauterine deaths	Stillbirths (≥ 24 weeks)	Both	Ethnicity	Ethnicity – general categories
Man 2016b [77]	To present contemporary demographic findings from a large series of > 1000 intrauterine deaths in London which underwent autopsy investigation, and to examine these features in relation to the gestational age at which fetal death occurred, including deaths across both second and third trimesters, which are not captured by traditional registry-based approaches	Descriptive	London	Intrauterine deaths	Stillbirths (≥ 24 weeks)	Both Control Both		Ethnicity – general categories
Man 2016c [78]	To examine factors relating to determination of cause of death using a large dataset extracted from an autopsy research database including cases from two specialist centers, in which observer bias was reduced as far as possible by recording objectively findings at autopsy and assigning causes and classifications of death based on predetermined criteria.	Descriptive	London	Intrauterine deaths	Stillbirths (≥ 24 weeks)	Both OZ4 by Guess	Σ	Ethnicity – general categories
Garcia 2017 [79]	How do health beliefs influence health behaviour and contribute to perinatal mortality in babies born to Pakistani, Bangladeshi and WB women living in Luton?	Mixed Methods	Luton, UK	White, Pakistani and Bangladeshi women in Luton	Delivery of an infant with 'no signs of life' between 24 and 42 weeks of gestation	Both T	Intersectional – socioeconomic (individual and area), ethnicity and immigration status	Education level, ethnicity by country of heritage. Immigration status by years in UK.

# **BMJ Open**

# Inequalities and stillbirth in the UK: A meta-narrative review

Journal:	BMJ Open
Manuscript ID	bmjopen-2019-029672.R1
Article Type:	Research
Date Submitted by the Author:	11-Jul-2019
Complete List of Authors:	Kingdon, Carol; University of Central Lancashire, ReaCH group Roberts, Devender; Liverpool Womens NHS Foundation Trust, Department of Obstetrics Turner, Mark; Liverpool Women's Hospital NHS Foundation Trust, Storey, Claire; International Stillbirth Alliance, Crossland, Nicola; University of Central Lancashire, School of Health Finlayson, Kenneth; University of Central Lancashire, School of Health Downe, Soo; University of Central Lancashire, research in childbirth and health
<b>Primary Subject Heading</b> :	Public health
Secondary Subject Heading:	Obstetrics and gynaecology, Sociology
Keywords:	stillbirth, inequalities, deprivation, ethnicity

SCHOLARONE™ Manuscripts

## Inequalities and stillbirth in the UK: A meta-narrative review

#### **Corresponding Author**

Dr Carol Kingdon, Senior Research Fellow,

School of Community Health and Midwifery, Faculty of Health and Wellbeing,

University of Central Lancashire, Preston, United Kingdom. PR1 2HE

Tel: 07935744004

ckingdon@uclan.ac.uk

ORCID ID 0000-0002-5958-9257

#### Co-authors

Dr Devender Roberts, Consultant Obstetrician in Fetal Medicine,
Liverpool Women's NHS Foundation Trust, Crown Street, Liverpool, United Kingdom. L8 7SS

<u>Devender.Roberts@lwh.nhs.uk</u>

Professor Mark Turner, Reader in Neonatology/Consultant Neonatologist,
Department of Women's and Children's Health, Institute of Translational Medicine,
Faculty of Health and Life Science, University of Liverpool, Liverpool, United Kingdom. L8 7SS
Mark.Turner@liverpool.ac.uk

Ms Claire Storey, Co-chair, International Stillbirth Alliance, 61 Kings Drive, Stoke Gifford, Bristol, United Kingdom. BS34 8RD storey.claire@yahoo.com

Dr Nicola Crossland, Research Associate,
School of Community Health and Midwifery, Faculty of Health and Wellbeing,
University of Central Lancashire, Preston, United Kingdom. PR1 2HE
ncrossland@uclan.ac.uk

Mr Kenneth William Finlayson, Research Associate,
School of Community Health and Midwifery, Faculty of Health and Wellbeing,
University of Central Lancashire, Preston, United Kingdom. PR1 2HE
<a href="mailto:KWFinlayson1@uclan.ac.uk">KWFinlayson1@uclan.ac.uk</a>

Professor Soo Downe, Professor of Midwifery,
School of Community Health and Midwifery, Faculty of Health and Wellbeing,
University of Central Lancashire Preston, United Kingdom. PR1 2HE
sdowne@ulcan.ac.uk

Word count: 3,800

#### **Abstract**

**Objective:** To review what is known about the relationship between stillbirth and inequalities from different disciplinary perspectives to inform stillbirth prevention strategies.

**Design:** Systematic review using the meta-narrative method.

**Setting:** Studies undertaken in the UK.

**Data sources:** Scoping phase: experts in field, exploratory electronic searches and hand-searching. Systematic searches phase: Nine databases with no geographical or date restrictions. Non-English language studies were excluded.

**Study selection:** Any investigation of stillbirth and inequalities with a UK component. **Data extraction and synthesis:** Three authors extracted data and assessed study quality. Data were summarised, tabulated and presented graphically before synthesis of the unfolding storyline by research tradition; and then of the commonalities, differences and interplays between narratives into resultant summary meta-themes.

**Results:** 54 sources, from nine distinctive research traditions were included. Evidence of associations between social inequalities and stillbirth spanned 70 years. Across research traditions there was recurrent evidence of the social gradient remaining constant or increasing, fuelling repeated calls for action (*Meta-theme 1: Something must be done*). There was less evidence of an effective response to these calls. Data pertaining to socio-economic, area and ethnic disparities were routinely collected, but not consistently recorded, monitored or reported in relation to stillbirth (*Meta-theme 2: Problems of precision*). Many studies stressed the interplay of socio-economic status, deprivation or ethnicity with aggregated factors including heritable, structural, environmental, and lifestyle factors (Meta-theme 3: *Moving from associations towards intersectionality and intervention(s)*). No intervention studies were identified.

**Conclusion:** Research investigating inequalities and stillbirth in the UK is underdeveloped. This is despite repeated evidence of an association between stillbirth risk and poverty, and stillbirth risk, poverty and ethnicity. A specific research forum is required to lead the development of research and policy in this area, which can harness the multiple relevant research perspectives and address the intersections between different policy areas.

Protocol registration number: CRD42017079228

**Keywords:** Stillbirth, inequalities, deprivation, social class, poverty, ethnicity, metanarrative.

#### **Article summary**

#### Strengths and limitations of this study

- Meta-narrative is a systematic methodological approach to understand how multiple disciplines and different philosophical perspectives have researched a question over time.
- To the best of our knowledge this is the first study to use a meta-narrative approach to investigate the association between inequalities and stillbirth in the UK.
- We adhered to the RAMESES standards for meta-narrative reviews to ensure fidelity with the methodology.
- We used a multipronged approach to retrieving sources that included exploratory searches, systematic searches, hand searches, expert opinion, and forward and back-chaining.

By limiting the review to UK-based studies only, we were able to focus with greater acuity on the commonalities and contestations between research traditions, but this may have led us to miss important research on the association between stillbirth and inequalities from other countries, of relevance both in the UK context and globally.

**Funding statement:** This work was supported by SANDS, the Stillbirth and Neonatal Death Charity.

### INTRODUCTION

Avoidable inequalities in mortality across the life course are a global concern. [1] Ten countries account for 66% of the world's stillbirths, with most (98%) occurring in low-and middle-income countries (LMICs). [2] Inequalities exist within and between high-income countries (HICs) too. In 2011, the Lancet *Stillbirth* Series highlighted that the UK's stillbirth rate was one of the highest of all HICs. [3] In 2016 the second Lancet Series *Ending Preventable Stillbirths* reported that while overall stillbirth rates were falling in HICs, improvement was slower than expected, and significant inequalities within rates remained. [4] The UK's stillbirth rate continues to remain high in comparison to other high-income countries. [5]

The government's ambition is to halve the stillbirth rate in England by 2025, which would require the rate to fall to 2.6 per 1,000 total births. [6] In 2017, the stillbirth rate in England and Wales was to 4.2 per 1,000 total births. [7] Medical reasons for stillbirth are well known and strategies for prevention routine. Ongoing initiatives include the Safer Maternity Care strategic plan, [8] Saving Babies Lives Care Bundle, [9], Each Baby Counts, [10] the Perinatal Mortality Review Tool [11] and annual Perinatal Mortality Reports (MBRRACE-UK) [5]. The association between social determinants and stillbirth is less well understood. Clinicians acknowledge the need to do more to prevent stillbirth in women from socially disadvantaged groups. In England in 2017, the stillbirth rate in the most deprived areas was 5.5 per 1,000 total births, compared with 3.0 per 1,000 total births in the least deprived areas. [7]

The UK began to develop policies to address health inequalities in general following *The Acheson Inquiry into Inequalities in Health*. [12] The Marmot Review *Fair Society, Healthy lives*, published in 2010, progressed the UK's inequalities agenda by emphasising the importance of taking a life-course approach, starting with the early years and family building. [13] The key messages of the Marmot Review emphasised that there is a social gradient in health in the UK, whereby the lower an individual's social position the worse his or her health, which is unfair, and that this requires action across all the social determinants of health.

Public Health England's current strategy for action on inequalities *Reducing health inequalities: system, scale and sustainability* [14] is underpinned by the Dahlgren and Whitehead rainbow model of the social determinants of health. [15] This model offers a framework to explore the relative influence of these determinants on different health outcomes and the interactions between the various determinants. These are all potential mechanisms by which stillbirth risk maybe increased. What is missing from current stillbirth research agendas is an overarching synthesis of clinical and social science evidence to clarify the range of individual (including biological and behavioural), social and environmental mechanisms of increased stillbirth risk, the intersections between these mechanisms, and strategies to tackle them. This review sought to fill this knowledge gap.

We undertook an inter-disciplinary evidence synthesis (using a meta-narrative approach) to understand how structural factors, lifestyle factors, and clinical factors intersect to increase stillbirth risk, and to inform future strategies to manage at-risk pregnancies. The broad research question was what is the relationship between inequality and stillbirth, how has this been studied, and with what effects?

# **METHODS**

We conducted a systematic review using the meta-narrative method, [16-18] in accordance with the RAMESES standards. [19] A RAMESES checklist is provided (Supplementary information file 1). [20] Our protocol [21] (supplementary file 2) specified four objectives:

- 1. To review the current body of knowledge of the relationship between inequalities and stillbirth across the natural and the social sciences;
- 2. To provide new insights into the interplay of biological, clinical, cultural and socioeconomic factors in increased stillbirth risk;
- 3. To explore the impact of interventions on inequalities;

4. To provide a narrative summary of this research for stakeholders tasked with reducing preventable stillbirth.

#### **Meta-narrative**

Meta-narrative review is a type of systematic review that was developed by Trisha Greenhalgh and colleagues. [16-18] Meta-narrative is a term for the unfolding storyline of research in a particular tradition or topic, which draws on the theoretical approach in Thomas Kuhn's writing on paradigms. [22] We used this approach to make sense of evidence from heterogeneous sources in which stillbirth and inequality have been variously conceptualised and studied over time. The method is underpinned by the methodological principles of pragmatism, pluralism, historicity, contestation, reflexivity and peer-review. As a method, meta-narrative review involves six key stages [17]:-

- Planning We registered our protocol with PROSPERO [21] and assembled a multi-disciplinary research team.
- 2. Iterative scoping searches and systematic electronic searches Initial searches were designed to map the diversity of perspectives and approaches. We contacted experts in the field of stillbirth research and from disciplines contributing to inequalities research. Exploratory searches were conducted using the search term "stillbirth" in 13 databases in health and the humanities (Supplementary File 3). Systematic searches were conducted in November 2017 in MEDLINE, Embase, CINAHL, PsycINFO, Popline, Historical abstracts, Humanities International Complete, Race Relations Abstract, and SocIndex. See supplementary file 4: example systematic search strategy. An English language restriction was imposed, but no geographical or date restrictions. In our protocol, inclusion criteria were any study design (quantitative, qualitative or mixed-methods) investigating stillbirth and inequality, in a high-, middle-, or low-income setting. Following initial screening of titles and abstracts a pragmatic decision was made by the team to include

only studies with a UK component. Unchanged exclusion criteria from the protocol were: any study in non-English language; of pregnancy loss <20 weeks gestation; of perinatal loss in the neonatal period; only involving participants who had assisted conception. Screening was independently undertaken by three authors (NC, KF, CK), who also assigned potential inclusions to disciplinary categories at this stage (See supplementary file 5: screening tool).

- 3. **Mapping** A data extraction form was developed based on one used in earlier reviews, [23] which was adapted for the purpose of this metanarrative review. Additional fields were added to capture data relating to how inequalities and stillbirth were conceptualised, defined and theorised. The form was piloted by extracting data from a subset of five papers (taken from across the research traditions) to test for applicability to the metanarrative, and refined. Extracted data was then summarised, tabulated and presented. During this phase the team had lengthy discussions about which traditions were represented, the overlap between them, and their distinctiveness. We classified traditions based on the distinctiveness of their lens (or in other words paradigm). This involved consideration of scope, historical roots, key concepts, assumptions, theoretical basis, kinds of research questions asked, and the methods used.
- 4. Appraisal We stated in our protocol that all articles that met the inclusion criteria would be independently assessed by three researchers to minimize bias. During the process of the review it became apparent that quality appraisal of all quantitative studies using the appropriate checklists from the Critical Appraisal Skills Programme (CASP) Toolkit was not appropriate, with quality more suitably judged by the prevailing standards in each tradition. That said it was fitting to use CASP tools [24] for some studies in the epidemiological tradition and the Walsh and Downe tool for qualitative research quality appraisal. [25]

- 5. Synthesis phase The identification of the meta-themes was via a two-part synthesis; (1) at the level of the traditions, which unfolded in the mapping phase and (2) at the level of data extraction from primary studies across traditions. Part one involved evaluating the meta-narratives to identify and compare how the different research traditions conceptualized and theorized the topic, and the methodological approaches and study designs used. Differences in findings between the resulting meta-narratives were analysed interpretively to produce further insights. Part two of the synthesis process involved paradigm bridging (seeking commonalities in underlying conceptual and theoretical assumptions), paradigm bracketing (highlighting differences in these assumptions), interplay (exploring tensions) and meta-theorizing (exploring patterns that span conflicting understandings) to construct summary meta-themes. KF, NC and CK undertook the initial analysis and synthesis processes, with input from DR, MT, CS and SD.
- Recommendations phase We engaged with local clinical networks and the national Stillbirth and Neonatal Death Charity (SANDS) to formulate recommendations.

# Patient and public involvement

Author Claire Storey is a parent and Vice-Chair of the International Stillbirth Alliance and was involved in the design, and conduct of the review and the writing of this paper.

# **RESULTS**

From electronic searches of nine databases a total of 13,610 records were identified. Following duplicate removal 4,934 records were screened (Figure 1: RAMESES-PRISMA diagram). We included 54 sources from nine research traditions spanning the period 1945-2017. [26-78] Table 1 provides a summary of included research traditions. Chronologically, these traditions were Social Medicine [26-31]; Epidemiology [32-48]; Medical Sociology [49-53]; Public Health [54-58]; Spatial

Epidemiology [59-64]; Social Psychology [65]; Audits, Reports and Confidential Enquiries [66-74]; Fetal-Maternal Medicine [75-78] and Nursing and Midwifery [Garcia, Perinatal mortality in Pakistani, Bangladeshi and White British mothers in Luton]. Supplementary File 6 provides details about the characteristics of included sources. With the exception of Epidemiology (n=17), most traditions generated few relevant papers. All research traditions utilised epidemiological data. We included one mixed-method study reporting qualitative data. No intervention studies were identified. Lack of studies, heterogeneity of study design, definitions of stillbirth, and measurement of inequalities between studies, traditions and over time meant meta-analysis was not practical. Figure 2 maps the traditions contribution over time and the declining national stillbirth rate.

# Synthesis within traditions

Table 1 summarises the unfolding storylines by research tradition and their conceptualisation of inequalities.

# **Table 1: Summary of included research traditions**

Research tradition	Academic discipline	Definition and scope	Unfolding storyline 2	Inequalities conceptualised as	Included references
Social Medicine	Medicine	Social Medicine is a branch of medicine that uses epidemiological methods to establish a problem exists, determining factors and opportunities for preventative action. The tradition is distinctive in its thought on the interconnectedness between biological factors (i.e. mother's age) that have meaning whatever the social context and social factors (i.e. occupational social class) that derive their meaning from social organisation in human life emulating political economy concerns.	The Social Medicine [26-31] storyline begins with the investigation of how social and economic faces influenced the decline in stillbirths and early neonatal deaths in Scotland, England and Wales, between 1939 and 1944. Bate [26] attributed this fall to the improved nutrition of the mothers during pregnancy, a consequence of the national distribution and consumption of milk and other foods important for health during the second world war. These improvements affected every area, are group and parity. By 1949, the decline in the stillbirth rate had slowed, despite the introduction of the National Health Service (NHS). Four papers, from a series in The Lancet in 1955 [27-30] sought to understand why. The last paper concluded the independent effects of social class, region, the mother's age and parity on stillbirth risk. Illsley [31] showed how occupational class may be more than a measure of inequality simply based on environmental conditions at the time of maternity, reporting that it can also be a resider of a woman's personal characteristics (height, physique, health, intelligence and nutrition), education and social habits. Women who were intergenerationally upwardly socially mobile at marriage experienced less stillbirths.	A variety of social factors that combine with biological characteristics to increase vulnerability to stillbirth risk.	N=6 [26-31]
Epidemiology	Medicine	Epidemiology, developed out of the bio-medical model as a specific line of inquiry. Initially epidemiology focused exclusively on epidemics of communicable diseases but subsequently expanded to address endemic communicable diseases and non-communicable infectious diseases. It is the study of the distribution and determinants of health-related states (especially disease), and the application of findings to the control of diseases and other health problems.	The Epidemiology [32-48] storyline is characterised by its increasingly sophisticated use of data and the repetition of the same or similar findings over time. Of the seventeen studies aligned to this tradition, six were landmark papers, repeatedly referenced within the field. [32-37] Although most authors highlighted a significant decrease in UK stillbirth rates sing the 1960's, studies repeatedly showed that the social gradient remained constant. [36,37,41] Within overall stillbirth rates sing the 1960's, studies repeatedly showed that the social gradient remained constant. [36,37,41] Within overall stillbirth rates sing the 1960's, studies repeatedly showed that the social gradient remained constant. [36,37,41] Within overall stillbirth rates sing the 1960's, studies repeatedly showed that the social gradient remained constant. [36,37,41] Within overall stillbirth rates sing the socio-economic class (as measured by an individual occupation) or residing in a disadvantaged community (as measured by local area deprivation), were relatively consistent markers of increased ricidence of stillbirth, when compared with more socially dvantaged counterparts. An important strength is epidemiology's identification of clinical, socio-economic and lifestyle factors associated with an increased risk of stillbirth across relatively large populations. Early studies used the Registrar General's Scale of occupational social class as a measure of inequality, later studies use the socio-economic classification scheme. Other studies studies the term deprivation' to signify inequality. In most of the studies using deprivation as a factor the risk of stillbirth increases with increasing levels of deprivation [34,43,44] although this is not always the case. [38] Epidemiological studies looking at athnicity as a measure of inequality are a relatively recent phenomenon and do not show the same level of consistency, although the rates of stillbirth for women of African-Caribbean origin remain at twice the rate of white women. [38,45] S	A variety of factors (social class, living in an area of deprivation, occupation of partner, ethnicity, etc.) associated with an increased relative risk of stillbirth.	N=17 [32-48]
Medical Sociology	Sociology	Medical Sociology is the study of the social causes and consequences of health and illness. This tradition has positivist and interpretative, theoretical and empirical, quantitative, qualitative, and mixed-methods, and cross-disciplinary branches. The persistence of social class gradients despite the demographic and epidemiological changes associated with the transition to modernity was an important focus during the 1970's and 1980's. During the 1990's research increasingly focused on lay understandings of health and illness and lived realities.	The Medical Sociology [49-53] storyline is theoretical. Early sociological explanations for the persistence of the social gradient in stillbirth encompassed theories of capital assets (the physique, stature, nutrition of the mother), social mobility (a direct thread from Social Medicine [31]), and time-lag (whereby developments in healthcare take time to reach those most in need, benefiting those better off first). [49] After the seminal Black Report [54] more nuanced considerations of gender, age, ethnicity and area of residence, alongside occupational class, as simultaneous and overlapping vulnerabilities, were developed. [50-52] These encompassed the broad consideration of life circumstances, behaviours, and beliefs/attitudes [50] and the precise disaggregation of the concept of 'deprivation' to reveal the complexity of materialist risks (and protections against these risks), which helps to explain the ambiguous association between economic deprivation and ethnicity. [52]	A set of social relations (rather than just a variable), which opens lived experience and multiplicity of factors at play (i.e. poverty, poor housing, nutrition, welfare) and relationship between structure and agency.	N=5 [49-53]
Public Health	Public Health	Public health is concerned with preventing disease, prolonging life and promoting health through organised efforts of society. From 18th and 19th century roots, during the 1980's there was a revival of public health policy. In the UK this coincided with a shift in thinking that morbidity or general health status had become the more important indicators of inequality, and increasing interest in individual behaviours and lifestyle as determinants of health.	The Public Health storyline [54-58] unites the seminal Black Report [54] (which had a major impaction research into inequalities in health in the UK), with seminal papers from the two Lancet Stillbirth Series [57-58] that were of equal significance to the stillbirth research and policy community. In the former publication [54] stillbirth is a crude cause of death of the grow, used as part of efforts to explain general trends in inequalities in health, based principally on measures of occupational social class from which artefact, natural selection, structuralist, and behaviourist explanations, (alongside the need to build on the ise of multiple causation) were developed. In the latter publications, distinguishing between different kinds of stillbirth and the imperance of making each stillbirth count, come alongside the need to build on the idea of interactions between factors that include social disadvantage. [57-58] The lack of targeted interventions for Black and Ethnic minority women in the UK, despite their complementaries of increased risk and known underutilisation of maternity services, was highlighted in the scoping review by Garcia. [56] 2016, there was an explicit recall to action to tackle inequalities and stillbirth within HICs by addressing structural factors (success poor housing, poverty) and factors, which limit women's access to antenatal care. [58]	An additional risk and considered in relation to providing targeted care to populations considered at risk.	N=5 [54-58]

			BMJ Open		
			BMJ Open pen-2019-029		
Spatial Epidemiology	Medical Geography and Epidemiology	Spatial epidemiology is concerned with the spatial analysis of disease incidence and prevalence. It uses geographical mapping and statistical modelling to understand the spatial distribution of disease, under the assumption that this will provide indications of the environmental contributors to the disease.	The Spatial Epidemiology [59-64] storyline begins in the late 1980s and attempts to address how esimmunity deprivation and individual social class might each contribute to risk of stillbirth. Studies looking at stillbirth and inequalities have investigated the relative importance of individual level (Registrar General Social Class) versus area level (e.g. Townsend Score) measures of inequality. Studies report contradictory findings, perhaps revealing the complexity of how individual (compositional) and area (context) effects interact to affect risk, with some reporting an enduring association between area and/or individual level deprivation and stillbirth risk [59,61-63] and others reporting no association [60,64]. The storyline of UK-based search into place effects on stillbirth risk has so far conceptualised geographical areas as "containers" of people, rather than seeing place as socially constructed.	A variety of factors (social class, living in an area of deprivation, occupation of partner, ethnicity) associated with an increased relative risk of stillbirth.	N=6 [59-64]
Social Psychology	Psychology	Social psychology is the study of human social behaviour, emotion and cognition. With its focus on both the individual and society, it draws on sociological and psychological perspectives Research methods involve both quantitative and qualitative approaches, and include surveys, participant observation, laboratory experiments, field experiments, and archival and content analyses.  Experimental social psychology is underpinned by positivist assumptions, while other approaches such as critical social psychology, operate from a social constructionist stance.	The Social Psychology [65] storyline arose from the Black Report [54] and draws on theoretical expanations from the Black Report about the association between social inequality and ill-health. This storyline is represented by one paper from 1990 [65], which used secondary data (birth data from England Wales, 1980-1986) to develop a theoretical model of how social class may affect psychosocial mediators – emotional, social, and cognitive factors – which may in turn influence pregnancy outcome, either directly or mediated through behaviours and coping strategies. The proposed model suggests that material deprivation results in more negative life events while also reducing social support, and access to education and information. Stressful by events, unmitigated by social support, create stress, anxiety, depression, low self-esteem. Poor education, or access to information, lead to a lack of knowledge and to deleterious beliefs and attitudes. The combined emotional and cognitive effects produce coming strategies and behaviours that increase the risk of negative pregnancy outcomes (i.e. smoking). [65]	A factor influencing health Inequalities can be seen to affect health via increasing psychosocial stress, which can then directly impact on health and also induce health- limiting behaviours	N=1 [65]
Audit Reports and Confidential Enquiries	Inter- disciplinary (Epidemiology, Obstetrics, Paediatrics, Midwifery)	Audits, Reports and Confidential Enquiries provide knowledge not always thought of as research, nevertheless it usefully uses routinely collected data to examine time-trends. As a tradition it incorporates a variety of approaches including epidemiology, economics and health policy and may be further informed by qualitative data and/or expert opinion. It includes 1992-2003 CESDI (Confidential Enquiry into Stillbirths and Deaths in Infancy), 2003-2011 CMACE (Centre for Maternal and Child Enquiries), and 2011 onwards MBRACE-UK (Mother and Babies: Reducing Risk through Audits and Confidential Enquiries across the UK).	The Audit, Reports and Confidential Enquiries storyline [66-74] builds on over 50 years of local and rational reporting of maternal and infant deaths. A key feature of these reports is the presentation of stillbirth rates at national, responsible to the subsequent comparisons between geographical units and benchmark averages. Over the years times processes were modified and refined into the national Confidential Enquiry scheme [66,67] and, more recently, under the banne-MBRACE-UK (Mother and Babies: Reducing Risk through Audits and Confidential Enquiries across the UK) [68-70]. Although the identified more than 20 national reports only five explored the association between inequalities and stillbirth [66-70] with the majority focusing on 'avoidable' health system and clinical failures. Where inequalities and stillbirth were identified they were discussed in relation to lifestyle factors (smoking, excess alcohol consumption, obesity) or regional or ethnic disparities associated with ingreased stillbirth risk. Four regional reports or audits from the West Midlands [71-74] attempt to look at stillbirth and inequalities explicitly by equating higher indices of multiple deprivation (IMD's) with increased stillbirth rates. These reports were more nuamed and identify a number of social and medical risk factors that could be screened for (alone or in combination) to predict risk destillbirth (e.g. unemployment, inappropriate housing, unsupported/difficult family circumstances, emotional factors/anxiety, material age <20 yrs or > 40 yrs, obesity, smoking, consanguinity, history of mental health issues). The authors of these reports also highlight fetal growth restriction (FGR) as a potential predictor of stillbirth in deprived communities.	Regional variations in stillbirth rates with recognition of differences between areas of deprivation (high and low) and ethnicity (White and Black & Asian populations).	N=9 [66-74]
Fetal-Maternal Medicine	Medicine	Maternal-fetal medicine is a subspecialty of obstetrics. Its focus is on 'high risk' pregnancies, including women who have a pre-existing illness or a pregnancy-induced illness, and congenital abnormalities it draws on and is related to perinatal epidemiology. The clinical focus includes preterm birth prevention, screening for fetal growth restriction, and placental histopathology.	The Fetal-maternal medicine storyline [75-78] included a study reporting that women living in areas of highest deprivation (IMD 1) were more likely to experience fetal growth restriction compared to women living in the least (IMD 29). [75] Approximately 46% of these women smoked, compared to 7% in the least deprived. The study concluded that targeted aptenatal management was key to stillbirth prevention amongst women living in the most deprived areas. This tradition also offered time interlinked publications, which suggested that maternal ethnicity was associated with fetal loss at different gestations which women had relatively more stillbirths (>24 weeks gestation) and Black women relatively more late intrauterine fetal deaths (20-23 weeks gestation) [76-78]. There was a higher risk of ascending genital infection for Black mothers relative to women from other ethnic groups. This was a relatively common cause for early intrauterine fetal death, peaking at around 22 weeks, [78].	A risk factor for stillbirth and depending on the type of study, may be included as a covariate in the analysis.	N=4 [75-78]
Nursing and Midwifery	Nursing and Midwifery	Nursing and Midwifery research draws from positivist and interpretative paradigms, utilising a range of quantitative and qualitative methods. This tradition has made a significant contribution to the body of knowledge about stillbirth and bereavement care.	Only one mixed-method single-site study was identified as characteristic of this tradition. Garcia, Poinatal mortality in Pakistani, Bangladeshi and White British mothers in Luton] It showed no statistically significant association between stillbirth and maternal ethnicity, but found more perinatal deaths in deprived areas. Qualitative interviews with White British, Pakistani, and Bangladeshi women identified health beliefs and behaviours common to all ethnic groups. These included little awareness of what to do about risk factors such as reduced fetal movements ("two days I delayed because I don't know what I near to do") and anxieties about being a burden to overstretched maternity services ("they could do without me taking up a bed, taking up their time, you put yourself at a lower scale than everyone else.") Health professionals perceived they had communicated information to women about stillbirth risks and the importance of seeking prompt care. Professionals did not view any particular ethnic group to be higher risk, but were aware of how cultural norms and/or living in poverty can restrict access to timely care ("Some of them [Asian women: Pakistani and Bangladeshi] are beholden on their partners to get them there) ("It doesn't matter whether there e Asian or whatever they are They don't have transport and they don't have money, they don't have access to actually get here	An additional vulnerability, and considered in relation to the importance of providing culturally appropriate care.	N=1 [Garcia, Perinatal mortality in Pakistani, Bangladeshi and White British mothers in Luton]

## Synthesis across traditions

#### Meta-theme 1: Something must be done

Across time and research tradition the prevailing message was for action on inequalities and stillbirth. From the earliest included paper in Social Medicine that concludes 'there is still much to be done' [26] to a Public Health paper in The Lancet Ending Preventable Stillbirth Series 2016 that states 'programmes at community and country level need to improve health in disadvantaged families to address these inequalities' [58] the message is clear. The call to do something stems from persistent evidence of a social gradient coupled with perceptions of insufficient progress in diminishing stillbirth rates in the UK. In some research traditions stillbirth was used as an indicator of societal health, with references to the particularly low stillbirth rates achieved in Scandinavia commonplace. Despite the persistence of studies reporting the same or similar risk factors and the continuation of the social gradient exactly what kind of 'something should be done' is less clear. Evidence of effectiveness was absent for interventions at specific time-points, inter-generationally, at scale or targeted to social groups. The absence of stillbirths in inequalities reduction targets post-Acheson was identified as a specific barrier to action [71].

## Meta-theme 2: Problems of precision

Our meta-narrative approach highlighted how much of the challenge in seeking to act on inequalities and stillbirth lies in the lack of consensus and inherent complexities inherent to both. While there was persistent evidence of associations between stillbirth risk and poverty, and stillbirth risk, ethnicity and poverty, it was not possible to estimate the potential gain on stillbirth reduction if action was taken to reduce inequalities, because of problems with data availability and comparability. There were problems of precision in stillbirth definition and problems of precision in inequalities measurement. The traditions rooted in medical science offered the most analytic tools for defining when stillbirths happened (antepartum, intrapartum), at what gestation (early preterm, late preterm, term), and why in terms of clinical factors (classification according to ReCoDe, Wigglesworth, Aberdeen etc), but these definitions were not used consistently, and they rarely considered social inequalities as underlying factors.

Further problems of precision arose from how inequalities were variously conceived and measured, even when they were taken into account. In traditions informed by the social sciences, inequalities were broadly conceptualised as a set of social relations (rather than a variable/s), which opened up lived experience, multiple risk factors/ interactions between them, and consideration of the relationship between structure and agency in health and lifestyle. Further conceptual considerations arise from this, including socio-economic status/social class (an individual measure of inequality) based on occupation alone or in combination with income, education and culture (Social Medicine, Epidemiology, Medical Sociology, Public Health). The problem of how best to measure disadvantage was apparent across time. The artefact explanation for inequalities (which considers to what extent they are a construct of the measurement process) was particularly critical of the now defunct Registrar General's Scale. [31,50,51,54,55].

Deprivation (an area measure of inequality) was conceptualised according to the tool used to define it for which there was no consensus. Tools used included the Townsend deprivation index, Carstairs and Morris index, Jarman Deprivation Scores and the Index of Multiple Deprivation (IMD). A general question for the Spatial Epidemiology tradition was whether or not individual level deprivation and area deprivation are different and how they interact. [61-62] The problems associated with using crude categories to define ethnicity (i.e. white, Black, Asian) were also considered (Epidemiology, Medical Sociology, Public Health) and the complexities therein (i.e. benefits of more subtle classifications incorporating country of birth such as British Asian), including how such classifications are only proximate guides to experiences, practices, beliefs and lifestyles. In 1993, a matrix of country of birth, nationality, language group, religious affiliation, and (where appropriate) region, caste and subcaste was proposed by Andrews and Jewson to test the combining variables, as well as suggesting a more fine-grain exploration of major variables if used as part of a national dataset. [52]

# Meta-theme 3: Moving from associations to intersectionality and intervention

All the traditions included in this review report evidence of associations between living in poverty and increased risk of stillbirth. However, despite more than 70 years of research equating inequality with increased stillbirth risk "any detailed study of why this should be so is surprisingly sparse" [50;p.393] This theme attempts to shine some 'light on the most appropriate times to provide support and the form(s) that such support should take.' [42:p11] To begin to address the need for intervention, one recent study triangulated epidemiological data with what women said (qualitative data). [Garcia, Perinatal mortality in Pakistani, Bangladeshi and White British mothers in Luton] In so doing it showed how the interactions between education level, socioeconomic status, cultural needs, language barriers, knowledge, likeliness to seek help, and assumptions by healthcare staff interact to make (or diminish) stillbirth risk in the current maternity care system. While, that study was the first study to claim an intersectionality approach, most publications across the research traditions suggest that further exploration of the interactions between risk factors, and within specific groups, is warranted.

Most of the contributory risk factors identified in this review are already well known and have been for some time. As summarised in Figure 3 risk factors for stillbirth encompass biological, clinical, behavioural, health service and social factors. Figure 3 provides a model from which to test the associations between factors, which is built on interdisciplinary evidence of the clinical causes of stillbirth, theories of natural and social selection. cultural/behavioural/lifestyle explanations, area effects. materialist/structuralist explanations and availability, access and quality of care. While some studies proposed antenatal screening for a combination of social factors (i.e. non-English speaking, unemployed household) in combination with behavioural factors (i.e. smoking) and clinical factors (i.e. previous IUGR), there was little consensus on specific factors, timing, or outcome if social conditions remain the same. [39,40,46]

### DISCUSSION

This review highlights that research investigating what might work to reduce inequalities and stillbirth in the UK is underdeveloped. We identified nine research traditions in the field but, with the exception of epidemiology, these traditions had few studies within them. Across all traditions, epidemiological data persistently suggests that membership of a lower socio-economic group (as measured by an individual's occupation), or residing in a disadvantaged community (as measured by local area deprivation) is associated with increased incidence of stillbirth when compared with more socially advantaged counterparts. However, there was a paucity of research investigating why this should be so, despite repeated calls for action. A few studies found no association between living in an area of deprivation and increased stillbirth risk. Why this was so is also unclear. This review shows that the field is not only complex, but also dynamic, with the respective components (stillbirth per se and inequalities per se) beset by conceptual and methodological challenges. In terms of advancing understanding about the complexity of the interactions between factors associated with increased stillbirth risk this review is limited. Moreover, we found no studies of interventions targeted to reduce stillbirth in specific social groups or communities. Nonetheless, what this review does add is that stillbirth is a useful marker of success in addressing inequalities. It provides a cross-disciplinary foundation from which to develop and stimulate hypotheses about the relative influence of biological, clinical, behavioural, health service and social factors on birth outcomes and the interactions between these various determinants to inform future interventions.

#### Strengths and limitations

This study used a meta-narrative approach to investigate the association between inequalities and stillbirth. We adhered to the RAMESES standards for metanarrative review to ensure fidelity with the methodology. We used a multipronged approach to retrieving sources that included exploratory searches, systematic searches, hand searches, expert opinion, and forward and back chaining, which gave us a broad capture of relevant documents. By limiting the review to UK-based studies only, we were able to focus with greater acuity on the commonalities and contestations between research traditions. However, excluding studies from other countries may have led us

to miss important research on the association between stillbirth and inequalities of relevance both in the UK context and globally. The quality of some of the included sources in this review may also be considered an important limitation with the use of pre-specified quality appraisal tools [24-25] not deemed appropriate for all traditions.

#### Relationship of findings to other research

The current abundance of research investigating stillbirth prevention and bereavement care in the UK is a recent development as efforts to break the silence that has traditionally surrounded stillbirth have gained momentum and international ambition to reduce stillbirth has intensified over the last decade. [3-4,6,79-82] This goes some way to explain why the field is underdeveloped in comparison to the wider health inequalities literature on mortality and social gradient. We were surprised to find no intervention studies, although there is an acknowledged paucity of evaluations of interventions to reduce inequalities in health in general. [83-86] In the international literature, public health interventions seeking to reduce stillbirth are also sparse. The few that do exist include a food supplementation programme, which was offered to low-income women in the USA, [87] and a study looking at household air pollution in India, where wood and kerosene cooking fuel, more commonly used in low-income households, is known to be associated with stillbirth. [88] However, neither of these address the underlying structural components of disadvantage.

#### Implications for clinicians and policymakers

This review suggests that addressing inequalities as a component of stillbirth prevention in the UK demands intervention at many levels. The paucity of directly relevant research to the question of stillbirth prevention means policy makers must look towards what works to reduce inequalities for other related causes of death (i.e. sudden infant death, cardio-vascular disease and cancer). Health inequalities theory advocates intervening at specific time points during the life course (i.e. pregnancy and the early years), interventions that have impact over time (i.e. intra-generational and inter-generational), interventions at scale (i.e. national policies) and interventions targeting specific groups (i.e. ethnic minorities and lower social classes). Addressing nutrition, service uptake and the wider social determinants of health may have knock on effects on many clinical outcomes, including stillbirth. [89] Scotland's *Early Years Collaborative* that encompasses cross-sector interventions at the level of individuals,

groups, organisations and society, includes a specific stillbirth reduction target. [86] In the absence of a hierarchy of causation among these complex effects stillbirth specific research is well justified, as long as it is embedded in implementation, public health and caring for and about people.

#### Unanswered questions and future research

It was not possible within or across traditions in this review to determine the potential gain of inequalities and stillbirth reduction. The field would benefit from a national consensus for routinely collected data and future research at population level. MBRRACE-UK, the RCOG and NHS England now have a high level of precision in stillbirth definition and national data capture. Since 2014, MBRRACE-UK has consistently used the Children in Low-income Families Local Measure. [5] There is also a simultaneous need for qualitative research that gets behind classificatory system labels to the lived realities of groups and communities. This review highlights there have long been important differences between communities and place that, for example, the classification Black, Asian, and Minority Ethnic (BAME), or indices of multiple deprivation (IMD) can conceal.

Most of the factors associated with inequalities and stillbirth identified in this review are already well known, and have been for some time. The findings of the review suggest that looking at these well-known factors afresh is likely to provide new insights. For example, the reasons reported as to why women delayed seeking care for reduced fetal movements in this review resonate with the findings of earlier reviews of antenatal care in general. [90-91] Similarly, studies of smoking behaviours, influence of social and community networks, the conditions in which people live, and the impact of current UK smoke-free policies that were identified on the periphery of studies included in this review, demand cross-disciplinary consideration in future strategies for stillbirth prevention. [92-94] Not least because, these particular components of antenatal care already feature as part of stillbirth reduction initiatives, but to-date, have had limited success. [82]

The role of social factors, modifiable lifestyle behaviours, and antenatal interventions in stillbirth prevention are current research priorities identified by the stillbirth community. [95] The results of this review indicate that there is little effective work

across disciplines despite the long-recognised need for it. We recommend that the UK stillbirth research community overcome this by setting up a dedicated forum to promote intervention and implementation research in this area. The forum could have three roles: 1) Define the framework for future research by identifying the ways in which disciplines should interact; 2) Develop data standards for information relating to stillbirth and inequalities; 3) Develop and promote the intervention and implementation research, policy and practice agenda relating to stillbirth and inequality.

#### Conclusion

The UK government's current ambition is to halve the national stillbirth rate by 2025. Research investigating and, critically, addressing inequalities and stillbirth in the UK is underdeveloped. This is despite repeated evidence of an association between stillbirth risk and poverty, and stillbirth risk, poverty and ethnicity. A specific research forum is required to lead the development of research and policy, which can harness multiple relevant research perspectives and address the intersections between different policy areas. This review not only unifies calls for action, by connecting multi-disciplinary insight into these complexities, challenges and opportunities, it provides a starting point for a novel transdisciplinary response.

## Figure legends

Figure 1: RAMESES-PRISMA Diagram

Figure 2: Timeline of included studies by research tradition and the stillbirth rate in

England and Wales 1945-2017

Figure 3: Factors associated with inequalities and stillbirth I

#### Supplementary files

Supplementary file 1: RAMESES Checklist

Supplementary file 2: Protocol

Supplementary file 3: Example exploratory searches

Supplementary file 4: Example systematic search strategy

Supplementary file 5: Screening tool

Supplementary file 6: Characteristics of included sources

**Author contributions:** CK, DR, MT and CS designed the review with input from SD. NC and KF conducted the searches, identification and screening with agreement by consensus of all authors on final inclusions. NC, KF and CK extracted data, and agreed initial storylines and final meta-themes with review by SD, DR, MT and CS. CK wrote the first draft of the paper. All authors read, commented and approved the final manuscript.

**Data sharing statement:** This is a review. Original research data is contained in the included studies. Data interpretation is contained in the manuscript. Further information can be obtained from the corresponding author.

**Conflict of interest statement**: Dr. Kingdon, Professor Downe, Dr. Crossland, Mr. Finlayson and Ms. Storey report a grant from SANDS, the Stillbirth and Neonatal Death Charity, during the conduct of the study. Ms. Storey also reports her position as Vice-Chair of the International Stillbirth Alliance (ISA). Dr. Roberts and Dr. Turner have nothing to disclose.

Acknowledgements: Rob Rawcliffe, UCLanPrint for his design input into Figure 3.

#### References

- Commission on Social Determinants of Health. Closing the gap in a generation: health equity through action on the social determinants of health. Final Report of the Commission on Social Determinants of Health 2008; Geneva, World Health Organization. <a href="https://www.who.int/social\_determinants/thecommission/finalreport/en/">https://www.who.int/social\_determinants/thecommission/finalreport/en/</a> (accessed 25th June 2019)
- Blencowe H, Cousens S, Jassir FB, et al. National, regional, and worldwide estimates of stillbirth rates in 2015, with trends from 2000: a systematic analysis. *The Lancet Global Health* 2016; 4(2):e98-e108 doi: 10.1016/S2214-109X(15)00275-2
- 3. Mullen Z, Horton R (2011) Bringing stillbirth out of the shadows. *Lancet* 2011;377:1291–2. doi: http://dx.doi.org/10.1016/S0140-6736(11)60098-6.

- 4. Froen JF, Friberg IK, Lawn JE et al. Stillbirths: Progress and unfinished business. *Lancet* 2016; 6;387(10018):574-586. doi: 10.1016/S0140-6736(15)00818-1.
- Draper ES, Gallimore ID, Kurinczuk JJ, et al. on behalf of the MBRRACE-UK Collaboration. MBRRACE-UK Perinatal Mortality Surveillance Report, UK Perinatal Deaths for Births from January to December 2016. Leicester: The Infant Mortality and Morbidity Studies, Department of Health Sciences, University of Leicester. 2018.
- 6. GOV.UK New ambition to halve rate of stillbirths and infant deaths. <a href="https://www.gov.uk/government/news/new-ambition-to-halve-rate-of-stillbirths-and-infant-deaths">https://www.gov.uk/government/news/new-ambition-to-halve-rate-of-stillbirths-and-infant-deaths</a>. (accessed 1st February 2019)
- 7. Office of National Statistics Births in England and Wales: Live births, stillbirths and the intensity of childbearing, measured by the total fertility rate. <a href="https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarri-ages/livebirths/bulletins/birthsummarytablesenglandandwales/2017">https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarri-ages/livebirths/bulletins/birthsummarytablesenglandandwales/2017</a> (accessed 1st February 2019)
- Department of Health. Safer Maternity Care. The National Maternity Safety Strategy – Progress and next steps. November 2017. <a href="https://www.gov.uk/government/publications/safer-maternity-care-progress-and-next-steps">https://www.gov.uk/government/publications/safer-maternity-care-progress-and-next-steps</a> (accessed 25th June 2019)
- NHS England. Saving Babies' Lives Care Bundle. <a href="https://www.england.nhs.uk/mat-transformation/saving-babies/">https://www.england.nhs.uk/mat-transformation/saving-babies/</a> (accessed 1<sup>st</sup> February 2019)
- Royal College of Obstetricians and Gynaecologists. Each Baby Counts. <a href="https://www.rcog.org.uk/eachbabycounts">https://www.rcog.org.uk/eachbabycounts</a> (accessed 1st February2019)
- 11. National Perinatal Epidemiology Unit. Perinatal Mortality Review Tool. <a href="https://www.npeu.ox.ac.uk/pmrt">https://www.npeu.ox.ac.uk/pmrt</a> (accessed 1st February 2019)
- 12. Acheson D. Independent inquiry into inequalities in health report. The Stationery Office: London Acheson 1998.
- 13. Marmot M. Fair society, healthy lives: The Marmot Review: strategic review of health inequalities in England post-2010. Institute of Health Equity; 2010.
- 14. Public Health England. Reducing health inequalities: system, scale and sustainability. London 2017.
  <a href="https://assets.publishing.service.gov.uk/government/uploads/system/uploads/attachment\_data/file/731682/Reducing\_health\_inequalities\_system\_scale\_an\_d\_sustainability.pdf">https://assets.publishing.service.gov.uk/government/uploads/system/uploads/attachment\_data/file/731682/Reducing\_health\_inequalities\_system\_scale\_an\_d\_sustainability.pdf</a> (accessed 25th June 2019)
- 15. Dahlgren G, Whitehead M. Policies and strategies to promote social equity in health. Stockholm: Institute for future studies. 1991.
- 16. Greenhalgh T, Potts HW, Wong G, et al. Tensions and paradoxes in electronic patient record research: A systematic literature review using the meta-narrative method. *Milbank Quarterly* 2009:87(4):729-88.
- 17. Greenhalgh T, Robert G, Macfarlane F, et al. Storylines of research in diffusion of innovation: a meta-narrative approach to systematic review. *Soc Sci Med* 2005 Jul;61(2):417-30.

- 18. Greenhalgh T, Robert G, Macfarlane F, et al Diffusion of innovations in service organisations: systematic literature review and recommendations for future research. *Milbank Quarterly* 2004;82(4):581-629.
- Wong G, Greenhalgh T, Westhorpe G et al RAMESES publication standards: meta-narrative reviews. *BMC Medicine* 2013;11(20). <a href="https://doi.org/10.1186/1741-7015-11-20">https://doi.org/10.1186/1741-7015-11-20</a>
- 20. Moher D, Liberati A, Tetzlaff J, et al. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *PLOS Med* 2009;6:e1000097.
- 21. Kingdon C, Roberts D, Turner M, et al Inequalities and stillbirth: a metanarrative review. PROSPERO 2017 CRD42017079228.
- 22. Kuhn, T. The structure of scientific revolutions. University of Chicago University Press, Chicago 1962.
- 23. Betrán AP, Temmerman M, Kingdon C, et al. Interventions to reduce unnecessary caesarean sections in healthy women and babies. *Lancet* 2018; 392(10155):1358-1368. doi:https://doi.org/10.1016/S0140-6736(18)31927-5
- 24. Critical Appraisal Skills Programme (CASP). CASP Checklists. 2013. <a href="http://www.casp-uk.net/#!casp-tools-checklists/c18f8">http://www.casp-uk.net/#!casp-tools-checklists/c18f8</a>. (accessed 25<sup>th</sup> June 2019)
- 25. Walsh D, Downe S. Appraising the quality of qualitative research. *Midwifery*. 2006;22:108-119.
- 26. Baird, D. The Influence of Social and Economic Factors on Stillbirths and Neonatal Deaths. *Journal of Obstetrics and Gynaecology*. 1945;52(4): 339-365
- 27. Morris JN, Heady JA. Social and biological factors in infant mortality: Objects and methods. *Lancet* 1955; 268(6859):343-349.
- 28. Heady, JA, Daly C, Morris JN. Social and Biological Factors in Infant Mortality. II. Variation of Mortality with mother's age and parity. *Lancet* 1955; 268(6860):395-7
- Daly C, Heady JA, Morris JN. Social and biological factors in infant mortality:
   The effect of mother's age and parity on social-class differences in infant mortality. *Lancet* 1955; 265 (6861):445-448
- 30. Heady JA, Stevens CF, Daly C, et al. Social and Biological Factors in Infant Mortality. The independent effects of social class, region, the mother's age and her parity. *Lancet* 1955; 265(6862):499-503.
- 31. Illsley R. Social class selection and class differences in relation to stillbirths and infant deaths. *British Medical Journal* 1955; 24;2(4955):1520-1524
- 32. Kincaid JC. Social Pathology of Foetal and Infant Loss. *British Medical Journal* 1965;17;1(5441):1057–1060.
- 33. Clarke M, Clayton DG, Mason ES, et al. Asian mothers' risk factors for perinatal death: the same or different? A 10 year review of Leicestershire perinatal deaths. *British Medical Journal* 1988;297(6645):384-87.

- 34. Guildea ZES, Fone DL, Dunstan FDJ, et al. Social deprivation and the causes of stillbirth and infant mortality. *Archives of Disease in Childhood* 2001;84 (4):307-310. doi 10.1136/adc.84.4.307
- 35. Flenady V, Koopmans L, Middleton P, et al. Major risk factors for stillbirth in high-income countries: a systematic review and meta-analysis. *Lancet* 2011; 377 (9774):1331–40.
- 36. Seaton SE, Field DJ, Draper ES, et al. Socioeconomic inequalities in the rate of stillbirths by cause: a population-based study. BMJ Open 2012;2(3). doi: 10.1136/bmjopen-2012-001100
- 37. Zeitlin, J Mortensen, L. Prunet, C, et al. Socio-economic inequalities in stillbirth rates in Europe: measuring the gap using routine data from the Euro-Peristat Project. *BMC Pregnancy & Childbirth* 2016;16:15. https://doi.org/10.1186/s12884-016-0804-4
- 38. Penn N, Oteng-Ntim E, Oakley LL, et al. Ethnic variation in stillbirth risk and the role of maternal obesity: analysis of routine data from a London maternity unit. *BMC Pregnancy and Childbirth* 2014;14:404-410
- 39. Chitty LS, Winter RM. Perinatal mortality in different ethnic groups. No evidence for increasing inequality *Archives of Disease in Childhood* 1989; 64:1036-1041
- 40. Gray R, Bonellie SR, Chalmers J, et al. Contribution of smoking during pregnancy to inequalities in stillbirth and infant death in Scotland 1994-2003: retrospective population based study using hospital maternity records. *British Medical Journal* 2009;339:b3754
- 41. Rush D, Cassano P. Relationship of cigarette smoking and social class to birth weight and perinatal mortality among all births in Britain, 5-11 April 1970. *J Epidemiol Community Health* 1983 Dec;37(4):249-55.
- 42. Weightman AL, Morgan HE, Shepherd MA, et al. Social inequality and infant health in the UK: systematic review and meta-analyses. *BMJ Open* 2012;2:e000964. doi:10.1136/bmjopen-2012-000964.
- 43. Wood AM, Pasupathy D, Pell JP, et al. Trends in socioeconomic inequalities in risk of sudden infant death syndrome, other causes of infant mortality, and stillbirth in Scotland: population based study. *British Medical Journal* 2012;344:e1552
- 44. Bambang S, SpencerNJ, Logan S, et al. Cause-specific perinatal death rates, birth weight and deprivation in the West Midlands, 1991–1993. *Child: Care, Health and Development* 2000;26(1):73-82. doi:10.1046/j.1365-2214.2000.00152.
- 45. Khalil A, Rezende J, Akolekar R, et al. Maternal racial origin and adverse pregnancy outcome: a cohort study. *Ultrasound in Obstetrics & Gynecology* 2013;41(3):278-285. doi:10.1002/uog.12313
- 46. Sutan R, Campbell D, Prescott GJ, et al. The risk factors for unexplained antepartum stillbirths in Scotland, 1994-2003. *Journal of Perinatology* 2010;30: 311–318.

- 47. Murrells TJ, Catford JC, Smith TM et al. The use of logit models to investigate social and biological factors in infant mortality. II: Stillbirths. *Statistics in medicine* 1985;4(2):189-200.
- 48. Neasham D, Dolk H, Vrijheid M et al. Stillbirth and neonatal mortality due to congenital anomalies: temporal trends and variation by small area deprivation scores in England and Wales, 1986-96. *Paediatr Perinat Epidemiol* 2001; 15(4):364-73.
- 49. Antonovsky A, Bernstein J. Social class and infant mortality. *Soc Sci Med* 1977; 11:453-470
- 50. Macintyre S. The patterning of health by social position in contemporary Britain: Directions for sociological research. *Soc Sci Med* 1986;23:393-415
- 51. Bloor M, Samphier M, Prior L. Artefact explanations of inequalities in health: an assessment of the evidence. *Sociology of Health & Illness* 1987;9:231-264
- 52. Andrews A, Jewson N. Ethnicity and infant deaths: the implications of recent statistical evidence for materialist explanations. *Sociology of Health & Illness* 1993;15:137-156.
- 53. Kingdon C. Society. In Kierans C, Bell K, Kingdon C. Social and Cultural Perspectives on Health, Technology and Medicine. Abingdon, Routledge 2016.
- 54. Black D, Morris JN, Smith C, Townsend P. The Black Report. Townsend P, Davidson N. (Eds) 1992 Edn. London, Penguin books 1980.
- 55. Whitehead M. The Health Divide. Whitehead (Ed) 1992 Edn.; London, Penguin books 1988.
- 56. Garcia R, Ali N, Papadopoulos C, et al Specific antenatal interventions for Black, Asian and Minority Ethnic (BAME) pregnant women at high risk of poor birth outcomes in the United Kingdom: a scoping review. *BMC Pregnancy and Childbirth* 2015;15:226. doi:10.1186/s12884-015-0657-2.
- 57. Flenady V, Middleton P, Smith GC et al. Stillbirths: the way forward in high-income countries. *Lancet* 2011;377 (9778):703-1717
- 58. Flenady V, Wojcieszek AM, Middleton P, et al. Stillbirths: recall to action in high-income countries. Lancet Series. *Lancet* 2016;387(10019):691–702.
- 59. Pickton MJ. A Socio-Spatial Analysis of Perinatal Mortality in Greater Leicester [PhD doctoral thesis]. University of Leicester 1987. http://hdl.handle.net/2381/9240.
- 60. Sloggett A, Joshi H Deprivation indicators as predictors of life events 1981-1992 based on the UK ONS Longitudinal Study. Journal of Epidemiology & Community Health 1998;52:228-233.
- 61. Joyce R, Webb R, Peacock J. Social class and census-based deprivation scores: which is the best predictor of stillbirth rates? *Paediatric and Perinatal Epidemiology* 1999;13: 269-277
- 62. Dummer TJ, Dickinson HO, Pearce MS, et al. Stillbirth risk with social class and deprivation: No evidence for increasing inequality. *Journal of Clinical Epidemiology* 2000; 53(2):147–155

- 63. Dickinson HO, Hutton JL, Greaves LH,et al Deprivation and stillbirth risk in rural and urban areas. *Paediatric and Perinatal Epidemiology* 2002;16(3): 249–254
- 64. Pattenden S, Casson K, Cook S et al. Geographical variation in infant mortality, stillbirth and low birth weight in Northern Ireland, 1992–2002. *J Epidemiol Community Health* 2011;65(12):1159-65
- 65. Rutter DR, Quine L. Inequalities in pregnancy outcome: A review of psychosocial and behavioural mediators. *Soc Sci Med* 1990;30(5):553-568.
- 66. Confidential Enquiry into Maternal and Child Health. Perinatal Mortality Surveillance, 2004: England, Wales and Northern Ireland. London: CEMACH; 2006.
- 67. Centre for Maternal and Child Enquiries (CMACE) Perinatal Mortality 2009: United Kingdom. CMACE: London, 2011.
- 68. Draper ES, Kurinczuk JJ, Kenyon S et al. on behalf of MBRRACE-UK. MBRRACE-UK 2015 Perinatal Confidential Enquiry: Term, singleton, normally-formed, antepartum stillbirth. The Infant Mortality and Morbidity Studies, Department of Health Sciences, University of Leicester: Leicester, 2015.
- 69. Manktelow BN, Smith LK, Evans TA, et al. on behalf of the MBRRACE-UK Collaboration. MBRRACE-UK Perinatal Mortality Surveillance Report, UK Perinatal Deaths for Births from January to December 2013 - Supplementary Report: UK Trusts and Health Boards. The Infant Mortality and Morbidity Studies, Department of Health Sciences, University of Leicester: Leicester, 2015.
- 70. Manktelow BN, Smith LK, Seaton SE, et al. on behalf of the MBRRACE-UK Collaboration. MBRRACE-UK Perinatal Mortality Surveillance Report, UK Perinatal Deaths for Births from January to December 2014. The Infant Mortality and Morbidity Studies, Department of Health Sciences, University of Leicester: Leicester, 2016.
- 71. Gardosi J, Francis A. Key Health Data for the West Midlands. Chapter Five: Perinatal Mortality and Social Deprivation: West Midlands Trends 1998-2003. West Midlands Perinatal Institute 2005. <a href="http://medweb4.bham.ac.uk/websites/key\_health\_data/2004/ch\_05.htm">http://medweb4.bham.ac.uk/websites/key\_health\_data/2004/ch\_05.htm</a> (accessed 25<sup>th</sup> June 2019)
- 72. Gardosi J, Beamish N, Francis A, et al. Stillbirth and infant mortality, West Midlands 1997-2005: Trends, Factors, Inequalities. West Midlands Perinatal Institute 2007.
  <a href="http://medweb4.bham.ac.uk/websites/key\_health\_data/2006/pdf\_Files/Additional%20Report%20from%20WMPI.pdf">http://medweb4.bham.ac.uk/websites/key\_health\_data/2006/pdf\_Files/Additional%20Report%20from%20WMPI.pdf</a> (accessed 25th June 2019)
- 73. Francis A, El Sheikh A, Gardosi J. Key Health Data for the West Midlands. Chapter Thirteen: Stillbirths, Infant Deaths and Social Deprivation, West Midlands 1997-2007/8. West Midlands Perinatal Institute 2009. <a href="http://medweb4.bham.ac.uk/websites/key\_health\_data/2008/pdf\_Files/West%">http://medweb4.bham.ac.uk/websites/key\_health\_data/2008/pdf\_Files/West%</a>

- 20Midlands%20Key%20Health%20Data%202008-09.pdf (accessed 25<sup>th</sup> June 2019)
- 74. Gardosi J. Perinatal Mortality, Social Deprivation and Community Midwifery. West Midlands Perinatal Institute 2011.

  <a href="http://www.perinatal.nhs.uk/pnm/clinicaloutcomereviews/Report\_on\_perinatal\_mortality\_deprivation\_community\_midwifery\_2008-9.pdf">http://www.perinatal.nhs.uk/pnm/clinicaloutcomereviews/Report\_on\_perinatal\_mortality\_deprivation\_community\_midwifery\_2008-9.pdf</a> (accessed 28th October 2018)
- 75. Tang A, Whitworth M, Roberts D. The relation between social deprivation and stillbirth causes. *Arch Dis Child Fetal Neonatal Ed* 2008;93(Suppl I):Fa1–Fa14
- 76. Man J, Hutchinson JC, Heazell AE et al. Stillbirth and intrauterine fetal death: role of routine histopathological placental findings to determine cause of death. *Ultrasound Obstet Gynecol* 2016; 48(5): 579–584. doi: 10.1002/uog.16019.
- 77. Man J, Hutchinson JC, Ashworth M, et al. Stillbirth and intrauterine fetal death: contemporary demographic features of >1000 cases from an urban population. *Ultrasound Obstet Gynecol* 2016; 48(5): 591-595. doi:10.1002/uog.16021.
- 78. Man J, Hutchinson JC, Heazell AE et al. Stillbirth and intrauterine fetal death: factors affecting determination of cause of death at autopsy. *Ultrasound Obstet Gynecol* 2016; 48(5): 566-573. doi: 10.1002/uog.16016.
- 79. Heazell AE, Weir CJ, Stock SJE, et al. Can promoting awareness of fetal movements and focusing interventions reduce fetal mortality? A stepped-wedge cluster randomised trial (AFFIRM). *BMJ Open*. 2017;7(8):e014813.
- 80. Heazell AEP, Li M, Budd JT et al. Association between maternal sleep practices and late stillbirth findings from a stillbirth case-control study. *BJOG* 2018: 125:254–262.
- 81. Siassakos D, Jackson S, Gleeson K, et al. All bereaved parents are entitled to good care after stillbirth: a mixed-methods multicentre study (INSIGHT). BJOG 2018;125 (2): 160-170
- 82. Widdows K, Reid HE, Roberts SA, et al. Saving babies' lives project impact and results evaluation (SPiRE): a mixed methodology study. BMC Pregnancy and Childbirth 2018;18(1):43. doi: 10.1186/s12884-018-1672-x.
- 83. Macintyre S. The Black Report and beyond: What are the issues? *Soc Sci Med* 1997;44(6):723-745.
- 84. Bambra C, Gibson M, Sowden A, et al. Tackling the wider social determinants of health and inequalities: evidence from systematic reviews. *Journal of Epidemiology and Community Health* 2010;64:284-291.
- 85. Smith KE, Eltanani MK. What kinds of policies to reduce health inequalities in the UK do researchers support? *Journal of Public Health* 2015; 37(1):6-17
- 86. Burns H. Health inequalities: why so little progress? *Public Health* 2015;129: 849-853.
- 87. Fingar K, Lob S, Dove M, et al. Reassessing the Association between WIC and Birth Outcomes Using a Fetuses-at-Risk Approach. *Maternal & Child Health Journal* 2017; 21(4):825-835. doi:10.1007/s10995-016-2176-9
- 88. Lakshmi PV, Virdi NK, Sharma A, et al. Household air pollution and stillbirths in India: analysis of the DLHS-II National Survey. *Environmental Research* 2013; 121:17-22. doi: 10.1016/j.envres.2012.12.004

- 89.WHO. Equity, social determinants and public health programmes. World Health Organization, Geneva, Switzerland 2010.

  <a href="https://www.who.int/social\_determinants/publications/9789241563970/en/">https://www.who.int/social\_determinants/publications/9789241563970/en/</a>
  (accessed 1st February 2019)
- 90. Finlayson K, Downe S. Why Do Women Not Use Antenatal Services in Lowand Middle-Income Countries? A Meta-Synthesis of Qualitative Studies. *PLoS Med* 10(1): e1001373. doi:10.1371/journal.pmed.1001373
- 91. Downe S, Finlayson K, Walsh D, et al. 'Weighing up and balancing out': a meta-synthesis of barriers to antenatal care for marginalised women in high-income countries. *BJOG* 2009;116:518–529.
- 92. Popay J, Bennett S, Thomas C, et al. Beyond 'beer, fags, egg and chips'? Exploring lay understandings of social inequalities in health. *Sociol Health Illn* 2003;25(1):1-23.
- 93. Robinson J, Holdsworth C. They don't live in my house every day: How understanding lives can aid understandings of smoking. *Contemporary Drug Problems* 2013;40: 47-70.
- 94. Passey ME, Longman JM, Robinson J, et al (2016). Smoke-free homes: what are the barriers, motivators and enablers? A qualitative systematic review and thematic synthesis. *BMJ Open* 2016; 6(3). doi:10.1136/bmjopen-2015-010260
- 95. Heazell AEP, Whitworth MK, Whitcombe SW, et al. Research priorities for stillbirth: process overview and results from UK Stillbirth Priority Setting Partnership. *Ultrasound in Obstetrics and Gynaecology* 2015; 46(6):641-647. https://doi.org/10.1002/uog.15738

Figure 1: RAMESES-PRISMA Diagram

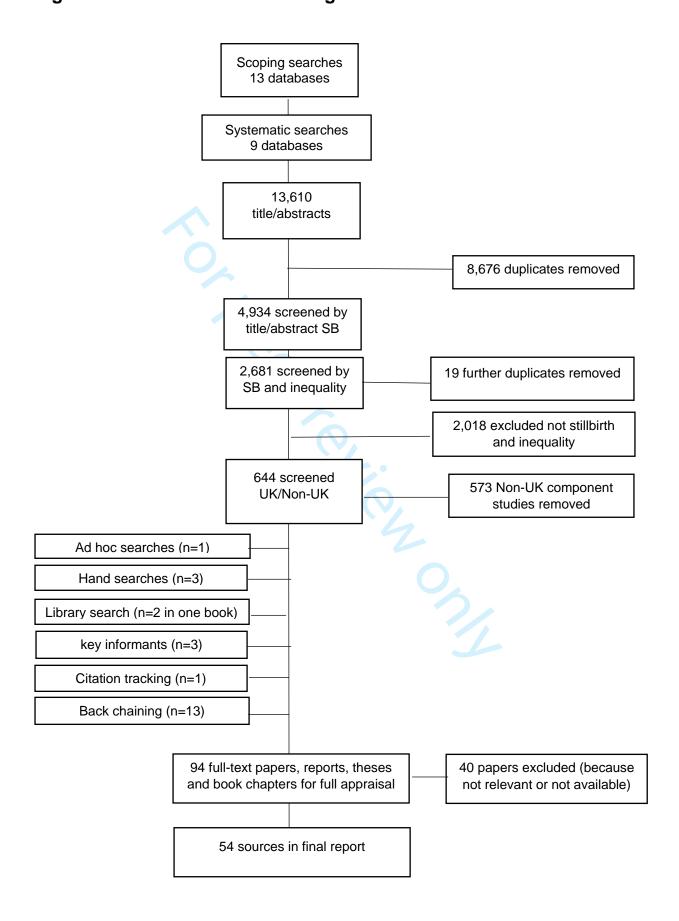


Figure 2: Timeline of included studies by research tradition and the stillbirth rate in England and Wales 1945-2017

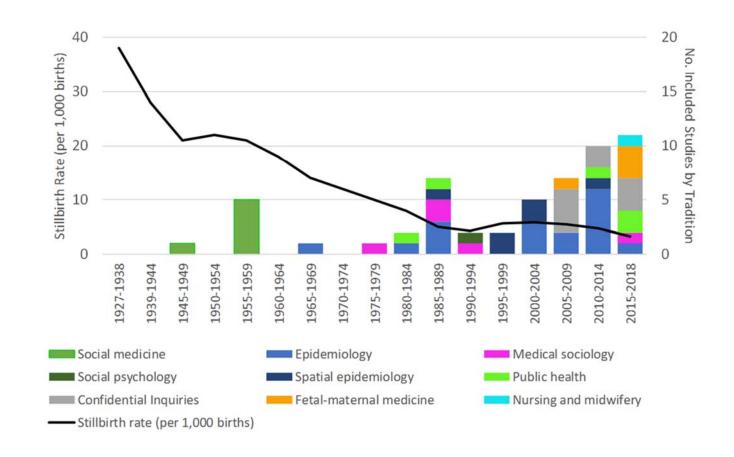
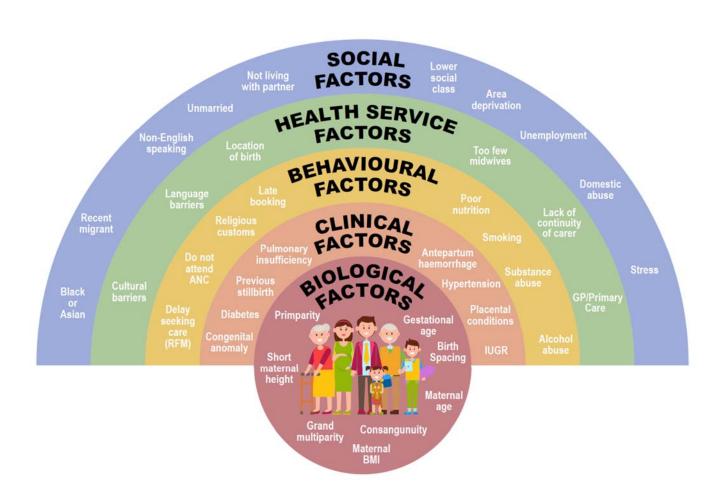


Figure 3: Factors associated with inequalities and stillbirth



Key: ANC (Antenatal Care); GP (General Practice), RFM (Reduced Fetal Movements); IUGR (Intra-uterine growth retardation also known as FGR – fetal growth restriction); BMI (Body mass index)

# **Supplementary File 1: RAMESES Checklist**

TITLE Inequalities and stillbirth in the UK: A meta-narrative review							
1	In the title, identify the document as a meta-narrative review or synthesis	1					
ABSTRACT							
2	While acknowledging publication requirements and house style, abstracts should ideally contain brief details of: the study's background, review question or objectives; search strategy; methods of selection, appraisal, analysis and synthesis of sources; main results; and implications for practice.	2					
INTRODUCTION	CV;						
3 Rationale for review	Explain why the review is needed and what it is likely to contribute to existing understanding of the topic area.	4-5					
4 Objectives and focus of review	State the objective(s) of the review and/or the review question(s). Define and provide a rationale for the focus of the review.	5					
METHODS							
5 Changes in the review process	Any changes made to the review process that was initially planned should be briefly described and justified.	6-7					

TITLE Inequalities	TITLE Inequalities and stillbirth in the UK: A meta-narrative review									
6 Rationale for using meta-narrative review	Explain why meta-narrative review was considered the most appropriate method to use.									
7 Evidence of adherence to guiding principles of metanarrative review	Where appropriate show how each of the six guiding principles (pragmatism, pluralism, historicity, contestation, reflexivity and peer review) have been followed.	6								
8 Scoping the literature	Describe and justify the initial process of exploratory scoping of literature.	6								
9 Searching processes	While considering specific requirements of the journal or other publication outlet, state and provide a rationale for how the iterative searching was done. Provide details on all the sources accessed for information in the review. Where searching in electronic databases has taken place, the details should include (for example) name of database, search terms, dates of coverage and date last searched. If individuals familiar with the relevant literature and/or topic area were contacted, indicate how they were identified and selected.	6-7								
10 Selection and appraisal of documents	Explain how judgements were made about including and excluding data from documents, and justify these.	6-7								
11 Data extraction	Describe and explain which data or information were extracted from the included documents and justify this selection.	7								
12 Analysis and synthesis processes	Describe the analysis and synthesis processes in detail. This section should include information on the constructs analysed and describe the analytic process.	8								

TITLE Inequalities	FITLE Inequalities and stillbirth in the UK: A meta-narrative review							
RESULTS								
13 Document flow diagram	Provide details on the number of documents assessed for eligibility and included in the review with reasons for exclusion at each stage as well as an indication of their source of origin (for example, from searching databases, reference lists and so on). You may consider using the example templates (which are likely to need modification to suit the data) that are provided.	9						
14 Document characteristics	Provide information on the characteristics of the documents included in the review.	9						
15 Main findings	Present the key findings with a specific focus on theory building and testing.	9-14						
DISCUSSION	Ch.							
<b>16</b> Summary of findings	Summarise the main findings, taking into account the review's objective(s), research question(s), focus and intended audience(s).	15						
17 Strengths, limitations and future research	Discuss both the strengths of the review and its limitations. These should include (but need not be restricted to) (a) consideration of all the steps in the review process and (b) comment on the overall strength of evidence supporting the explanatory insights which emerged.  The limitations identified may point to areas where further work is needed.	15-16						
18 Comparison with existing literature	Where applicable, compare and contrast the review's findings with the existing literature (for example, other reviews) on the same topic.	16						

TITLE Inequalities	s and stillbirth in the UK: A meta-narrative review	PAGE/LIN Number
<b>19</b> Conclusion and Recommendations	List the main implications of the findings and place these in the context of other relevant literature. If appropriate, offer recommendations for policy and practice.	16-17
20 Funding	Provide details of funding source (if any) for the review, the role played by the funder (if any) and any conflicts of interests of the reviewers.	3
	Trotac deals of laterily for the revent, the fore played by the laterily did any continues of the revents.	

#### **PROSPERO**

## International prospective register of systematic reviews



#### Inequalities and stillbirth: a meta-narrative review

Carol Kingdon, Devender Roberts, Mark Turner, Claire Storey, Nicola Crossland, Kenneth Finlayson, Soo Downe

#### Citation

Carol Kingdon, Devender Roberts, Mark Turner, Claire Storey, Nicola Crossland, Kenneth Finlayson, Soo Downe. Inequalities and stillbirth: a meta-narrative review. PROSPERO 2017 CRD42017079228 Available from:

http://www.crd.york.ac.uk/PROSPERO/display\_record.php?ID=CRD42017079228

#### Review question

The aim of this review is to undertake an inter-disciplinary evidence synthesis (using a meta-narrative approach) to understand how structural factors (health system, living in poverty), lifestyle factors (smoking, obesity), and bio-clinical factors (maternal infection, non-communicable disease, fetal growth restriction) intersect to increase stillbirth risk to improve interventions to manage at-risk pregnancies.

The broad research question is: What is the relationship between inequality and stillbirth, how has this been studied, and with what effects?

The measurable objectives are:

- 1. To review the current body of knowledge of the relationship between inequalities and stillbirth across the natural and the social sciences;
- 2. To provide new insights into the interplay of biological, clinical, cultural and socioeconomic factors in increased stillbirth risk;
- 3. To explore the impact of interventions on inequalities;
- 4. To provide a narrative summary of this research for stakeholders tasked with reducing preventable stillbirth.

#### Searches

The following databases will be used to identify eligible studies for inclusion as part of the scoping searches:

MEDLINE® (Life sciences, medicine and biomedicine)

Embase® (Biomedical)

CINAHL® (Nursing and Allied Health Professionals)

PsycINFO® (Psychology and the behavioural and social sciences)

AJOL® (African Journals on-line)

Global Index Medicus

Popline (includes LILACS [Latin America and the Caribbean])

Historical abstracts

**Humanities International Complete** 

Race Relations Abstract

SocIndex

Lexis Library

Lexis Nexus

#### Types of study to be included

This is a meta-narrative review with no restrictions on type of study to be included.

#### Condition or domain being studied

Stillbirth is a profound human tragedy. The experience of stillbirth involves physical implications for the mother, together with intense grief and lasting psychological trauma for both parents and wider family.



#### **PROSPERO**

#### International prospective register of systematic reviews

Studies have shown that stillbirth is associated with anxiety, depression and post-traumatic stress disorder in mothers, couples, siblings and grandparents.

The UK has the third highest rate of stillbirth among the 35 high-income countries of the world. Against a background of political, public and professional interest in halving the UK's stillbirth rate by 2030, and significant inter-disciplinary research effort to prevent or to improve maternity care services, there is longstanding evidence, across high-income countries, that the risk of stillbirth remains highest for the poorest families. Almost half (46%) of stillbirths remain of unknown cause, and inequalities in stillbirth risk demand more attention.

What is missing from current research agendas is an overarching synthesis of clinical and social science evidence to clarify the range of individual, social and biological mechanisms of increased stillbirth risk, the intersections between these mechanisms, and strategies to tackle them. There is also an urgent need to identify where interventions may decrease stillbirth risk for some, but may inadvertently increase inequalities for others. This meta-narrative review seeks to fill this gap in response to international interest in reviewing evidence from all relevant research traditions to address the unanswered questions surrounding stillbirth risk, and an NHS England Strategic Clinical Network need for this evidence to inform interventions to reduce the risk amongst women with complex social needs from vulnerable groups.

## Participants/population

The population of interest for the purposes of our inclusion criteria is: Woman or Women or Mother or Mothers or Father or fathers or Family or Families or Husband or partner or partners or Parent or parents

Inclusion criteria

Any study design investigating stillbirth and inequality Any study in high-,middle- or low-income settings

Exclusion criteria

Any study published in non-English language Any study of pregnancy loss before 20 weeks Any study of perinatal loss in the neonatal period

Any study involving participants who had assisted conception (as identified by authors)

## Intervention(s), exposure(s)

iflee extitioss, res socie exercimente de redeprivation or low income or poverty, or violence or abuse, or consanguinity, or ethnicity, or discrimination, or race or racism or racial, or migrant, or migration, or maternal age, or adolescence, or nutrition or obesity, or overweight or underweight, or smoking, or alcohol or drug or substance or chemical, or nonattendance, or neighborhood.

The interventions to be reviewed are:

growth chart or biomarkers or movement, or count, or support, or continuity of care or caseload.

The outcomes to be reviewed are:

stillbirth or perinatal death or pregnancy loss or miscarriage or fetal death or foetal death or feticide or foeticide or intrauterine death after 20 weeks pregnancy gestation.

#### **PROSPERO**

# International prospective register of systematic reviews



## Comparator(s)/control

Not applicable.

#### Primary outcome(s)

Stillbirth incidence

Timing and effect measures
Effect measure exposure to inequality

## Secondary outcome(s)

Not applicable

## Data extraction (selection and coding)

Key results will be extracted, collated and grouped. Modified data extraction forms will be developed from existing published meta-narrative reviews and other types of review the team have been involved with in relation to stillbirth. Included studies will then be tabulated by tradition.

#### Risk of bias (quality) assessment

All articles that meet the inclusion criteria will be independently assessed by two researchers (NC,KF) and the principal investigator (CK) to minimize bias. Quality appraisal of quantitative studies will be undertaken using the appropriate checklists from the Critical Appraisal Skills Programme (CASP) Toolkit. Checklists formed from this process will be used to grade papers into categories A, B, C or D, with group A representing papers of the highest quality. Papers will be assigned to groups according to how many criteria fulfilled (i.e. Category A papers not fulfilled 0-1 of the marked criteria; Category D papers not fulfilled ?6 criteria). Qualitative appraisal will be undertaken according to the checklist described by Walsh and Downe and articles graded A – D in accordance with Downe and Simpson. A grades will be allocated to papers with no or few flaws where the study credibility, transferability, dependability, and confirmability is high; B, some flaws, unlikely to affect the credibility, transferability, dependability, and/or confirmability of the study; C, some flaws which may affect the credibility, transferability, dependability, and/or confirmability of the study; D, significant flaws which are very likely to affect the credibility, transferability, transferability, dependability. Any differences in appraisals will be discussed with the wider research team until a final decision is reached.

#### Strategy for data synthesis

The defining feature of meta-narrative reviews is their illumination of a complex topic area from multiple angles in a unified narrative synthesis. The synthesis stage involves summarizing each tradition in coherent individual accounts (including elements of data aggregation) and then comparing and contrasting the resultant meta-narratives to highlight similarities and differences. This process of contestation between the disciplines/traditions leads to higher order constructs and conclusions where recommendations can be made (i.e. in circumstances such as X, don't forget to think about y).

#### Quantitative studies

Quantitative meta-analysis will be used if the included studies are sufficiently homogeneous. In the event included quantitative studies are too heterogeneous they will be summarized narratively, separate from the qualitative evidence synthesis in the first instance, and then incorporated alongside into an overall typology of inequality and stillbirth.

#### Qualitative studies

A modified meta-ethnography approach will be used for qualitative evidence synthesis comprising 4 stages:1) Familiarization and quality appraisal; 2) Data extraction (direct participant quotations and author's thematic interpretation); 3)Coding into initial concepts; 4) Interpretative synthesis into emergent and final



# PROSPERO

#### International prospective register of systematic reviews

themes to be presented in a typology.

Analysis of subgroups or subsets Not applicable

Contact details for further information

Carol Kingdon

ckingdon@uclan.ac.uk

Organisational affiliation of the review

University of Central Lancashire

Review team members and their organisational affiliations

Dr Carol Kingdon. University of Central Lancashire

Dr Devender Roberts. Liverpool Women's NHS Foundation Trust

Dr Mark Turner. University of Liverpool

Ms Claire Storey. International Stillbirth Alliance

Dr Nicola Crossland. University of Central Lancashire

Mr Kenneth Finlayson. University of Central Lancashire

Professor Soo Downe. University of Central Lancashire

Anticipated or actual start date

04 October 2017

Anticipated completion date

30 March 2018

Funding sources/sponsors

Stillbirth and Neonatal Death Society (SANDS) (Ref:RF510)

Conflicts of interest

None known

Language

English

Country

**England** 

#### PROSPERO

This information has been provided by the named contact for this review. CRD has accepted this information in good faith and registered the review in PROSPERO. CRD bears no responsibility or liability for the content of this registration record, any associated files or external websites.

# **Supplementary File 3: Example exploratory searches**

Databases	No. hits for "stillbirth"	Types of articles	Relevance to review questions
Art and Architecture source	10	poetry, feminism/women's studies, historical	Articles about impact and experiences of stillbirth rather than risk factors.
Criminal Justice Abstracts with Full Text	73	social work, women's studies, first person accounts/autoethnography	Articles about impact and experiences of stillbirth rather than risk factors.
Cochrane Library	535	Cochrane Reviews (n=83) Trials (423)	Two Cohrane reviews were tangenitaly relevant to research questions (interventions to reduce smoking in pregnancy, interventions to reduce domestic violence in pregnancy).
Humanities international complete	120	Family studies, images, religious aspects, first person accounts, responses to stillbirth, archaeology (burials), how stillbirth is conceptualised, abortion and morality/ethics	Articles about impact and experiences of stillbirth.
Race relations abstracts	6	differential medical risks by ethnicity, a couple of articles relating to social risk factors by ethnicity	Social inequalities as a risk factors
Religion and Philosophy Collection	74	grief and loss, psychosocial identity, religious aspects, social responses, attachment theory, abortion,	Articles predominantly about abortion; not relevant to research questions
SocIndex	482	psychosocial identity, psychosocial impacts, mother and family experiences, social risk factors, grief, coping,	Social inequality risk factors. Consequences, impact and coping.
Lexis Library (legal)		Professional misconduct cases. Judgements about cases where a medical intervention/decision has led to stillbirth. Employment policies. Legal frameworks about healthcare delivery. Medical law.	Tangential to research questions.
JISC Historical Texts	0 for stillbirth, 308 for stillborn	Literature (66). Medical texts (15) - midwifery manuals, descriptive works on causes of death. 1600s, 1700s.	Tangential to research questions.
Historial abstracts (via Ebsco)	56 for stillbirth.	Rates of stillbirth at different historical times and places, about perceptions of stillbirth. Lots of hits come from journal Population Studies.	Modern history – potential for understanding how stillbirth has been conceptualised.

# **Supplementary File 4: Example Systematic Search**

# **Strategy terms**

**Population**: Woman or Women or Mother or Mothers or Father or fathers or Family or Families or Husband or partner or partners or Parent or parents

#### **Exposure**

<b>Exposure type</b>	Exposure search terms
economic	Inequalit* or Depriv* or 'low income' or Poverty or socioeconomic or 'social class'
violence & abuse	Violence or Abus*
substance use	Smok* or Alcohol or Drug or Substance or Chemical
ethnicity	ethnic* or Race or Racism or Racial* or Migrant or Migration
nutrition	nutrition or undernutrition or malnutrition
age	'maternal age' or adolescen*
obesity and weight	Obes* or overweight or underweight
space and place	neighborhood or neighbourhood or residence
appointments	"Appointments and Schedules"/ or nonattendance.mp
culture	Sociocultural or cultur*
consanguinity	Consanguin*

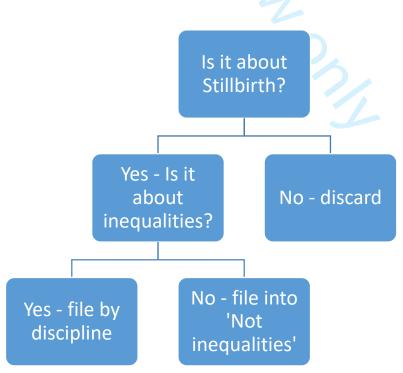
**Intervention**: growth chart' or biomarkers or movement or count or support or 'continuity of care' or caseload

**Outcome**: Stillbirth or stillborn or 'perinatal death' or 'pregnancy loss' or miscarriage or 'fetal death' or 'foetal death' or feticide or foeticide or 'intrauterine death'

**IVF-related terms** (ivf or fertil\* or infertil\* or 'assisted fertility' or inseminat\* or iui or 'embryo transfer')

# **Supplementary File 5: Screening Tool**

Answer the following questions in sequence:	Yes	No	Can't tell from abstract
1. Is it about stillbirth (defined as the death of a baby after 20 weeks in-utero until immediately before birth; excluding pregnancy loss < 20 weeks gestation and excluding neonatal deaths)?	Assess by Q2	Discard	If abstract doesn't define miscarriage, then discard.
If yes, then:			
<ul> <li>2. Is it about inequality and stillbirth?</li> <li>Working definition of inequality</li> <li>Inequality (?how circumstances of daily living disadvantage you, or structural)</li> <li>Socio-economic (as defined by authors so not necessarily NS-SEC), living in poverty, low income</li> <li>Deprivation (ditto – as defined by author - might not be IMD), neighbourhood, traveller, immigrant or refugee</li> <li>Minority ethnicity, religion, disability, young maternal age – member of marginalised group</li> <li>Risk factors (obesity, smoking, substance use, abuse etc) - may be linked to inequality but not enough on their own to include</li> </ul>	If yes, file by discipline	If no, then file as "Not about inequalities and stillbirth"	
If yes, then:			
What discipline is it from?			
3. Is it seminal?			



## **Supplementary file 6: Characteristics of included sources**

Sunnleme	entary file 6: Characteristics of included	sources	BMJ Open					
Author and year	Aim and objectives	Design	Context	Population	Stillbirth definition	mjopen-2019-029672 om 1200 Stillbirth classification (anteparture) intraparturer	Type of inequality	Precision of inequality
Baird 1945 [26]	To examine the influence of social and economic factors on stillbirths and neonatal deaths	Descriptive	Scotland, compared with England and Wales	All maternities 1938- 1944	Undefined	Both ber 2019.	Socio-economic (individual)	Registrar Generals Scale of occupational class
Morris 1955 [27]	To describe the background and design of inquiry into stillbirths and infant deaths in England and Wales in 1949 and 1950	Review	England and Wales	Stillbirths and Infant deaths	Undefined	Both 9. Down	Socio-economic (individual)	Registrar Generals Scale of occupational class
Heady 1955 [28]	To describe the main features of variation of the stillbirth rate, and the neonatal and post-neonatal mortality rates, with two biological factors (age of mother and parity)	Descriptive	England and Wales	Births in 1949	Single, legitimate, stillbirth after the 28th week of pregnancy	Both Oaded	Socio-economic (individual)	Registrar Generals Scale of occupational class
Daly 1955 [29]	To consider the extent of such "biological" differences between the social classes, and the effect of these differences on the stillbirth and infant mortality rates of the social classes	Descriptive	England and Wales	Legitimate, livebirths and stillbirths born in 1949	Single, legitimate, stillbirth after the 28th week of pregnancy	Both Om http	Socio-economic (individual and place of residence)	Registrar Generals Scale of occupational class
Heady 1955 [30]	To investigate the "independence" of mother's age and her parity in relation to social class and region	Descriptive	England and Wales	Single, legitimate livebirths and stillbirths born in 1949	Single, legitimate, stillbirth after the 28th week of pregnancy	Both //bmjop	Socio-economic (individual)	Registrar Generals Scale of occupational class
Illsley 1955 [31]	To examine the processes of movement between social classes and the effects on stillbirths and infant death statistics	Descriptive	Aberdeen, Scotland	Married primiparae resident and delivered in Aberdeen between July, 1950, and December, 1954	Undefined	Both  Both  Both  Both  Both  Both  Both	Socio-economic (individual)	Registrar Generals Scale of occupational class
Kincaid 1965 [32]	To explore changes in UK perinatal mortality rates over the period 1951-1961	Descriptive	Scotland, England & Wales	Birth records from national databases over periods ranging from 1948-1964	Undefined	on April 23	Socio-economic (individual)	Registrar Generals Scale of occupational class
Clarke 1983 [33]	(a) to measure the extent to which established risk factors, such as high parity and low social class, were risk factors in our locality; (b) to measure the extent to which other factors, such as the provision of medical services and demographic changes, might contribute to the risk of perinatal mortality; (c) to estimate the extent to which avoidable factors might be detected in the case histories of the perinatal deaths; and (d) to contribute our findings to the educational and planning processes of the local health authorities	Case-control	Leicestershire	All perinatal deaths In Leicestershire 1976- 1985	Undefined	April 23, 2024 by guest. Pro	Ethnicity	Broad categories (Asiar or European)
Guildea 2001 [34]	To investigate the relation between social deprivation and causes of stillbirth and infant mortality	Retrospective cohort	Wales	All births to women resident in Wales 1993–98	Late fetal deaths after 24 weeks gestation [stillbirth and neonatal death combined]	Protected by o	Socio-economic (area)	Townsend social and material deprivation score (unemployment, car ownership, owner occupation and

	BMJ Open								
							029 <b>6</b> 72 on		overcrowding 1991 census)
Flenady 2011 [35]	To clearly identify important risk factors for stillbirth in high income countries	Systematic review and meta- analysis	High-income countries (including UK)	Studies from HICs	Death of a baby 20 weeks' gestation or more, or birthweight of at least 400 g	Both	12	ethnic origin, socioeconomic status, education level, young maternal age	Different in different studies
Seaton 2012 [36]	To assess time trends in socioeconomic inequalities in overall and cause-specific stillbirths in England over an 8-year period to aid understanding of each cause's impact on the deprivation gap and the overall stillbirth rate	Population-based retrospective study	England	All singleton stillbirths born to mothers resident in England between 01/01/2000 and 31/12/ 2007	Losses from the 24th weeks of gestation occurring in singleton infants	Both	September 2019. Downloaded from http://bmjopen.bmj.com/ on	Socio-economic (area)	Indices of Multiple Deprivation (IMD) 200 score (income; employment; health an disability; education, skills and training; barriers to housing; living environment and crime)
Zeitlin 2016 [37]	What is the magnitude of social inequalities in stillbirth rates in European countries?	Retrospective review of data from countries participating in the Euro-Peristat project	Europe (includes UK data)	Stillbirths and live births for the year 2010 from 29 countries	Intrauterine death after 22 weeks gestation or birthweight of 500 grams or more	Both	ed from http://	Socio-economic (individual)	Country level available data on educational level, and occupational group
Penn 2014 [38]	What is the association between clinical and socio-demographic factors and stillbirth, with a particular focus on ethnicity and obesity?	Cross-sectional	London	London population - (multi-ethnic)	24 completed weeks of pregnancy (with 28 wks (WHO definition) data in a supplementary table	Both	dmjopen.bm	Ethnicity and socio- economic (area)	Ethnicity general categories (i.e. Black, Asian), Area deprivation IMD score
Chitty 1989 [39]	To assess the contribution of lethal congenital malformations to perinatal mortality in different ethnic groups, to investigate the incidence of definite and probable autosomal recessive syndromes, and to estimate the possible effect of consanguinity.	Descriptive	North West Thames Region	All babies born between 1980-1985	Undefined	Both		Ethnicity	Ethnicity - broad categories (European, Pakistani and Indian)
Gray 2009 [40]	To study the effect of area deprivation on rates of stillbirth and infant mortality in Scotland for the 10 year period 1994-2003 and to establish whether smoking during pregnancy contributed to these gradients and, if so, to what extent.	Population based retrospective cohort study	Scotland	Live births and stillbirths 1994 - 2003.	24-44 weeks of pregnancy	Both	April 23, 2	Socio-economic (area)	Deprivation according to Carstairs-Morris Index
Rush 1983 [41]	To re-explore the interrelation of smoking, social class, birth weight, and perinatal mortality in the total population of births in Britain born 5-11 April 1970.	Prospective cohort study	Britain	All livebirths and stillbirths 5th - 11th April, 1970	Undefined	Both	2024 by g	Socio-economic (individual)	Registrar Generals Scale of occupational class
Weightman 2012 [42]	To assess the current evidence for the effects of social disadvantage on birth and infant outcomes for children born in the UK	Systematic review and meta- analysis	UK	UK studies	A baby born after the 24th week of pregnancy who does not show any signs of life	Unclear	west.	Socio-economic (individual and area)	Different measures in different studies
2012 [42]	disadvantage on birth and infant outcomes for children born in the UK	review and meta- analysis			24th week of pregnancy who does not show any signs of			sst. Protected by copyright.	(individual and area)  (individual and area)  (individual and area)

			ВМЈ	Open			mjopen-2019-029 <b>6</b> 72		
Wood 2012 [43]	To compare changes in inequalities in sudden infant death syndrome with other causes of infant mortality and stillbirth in Scotland, 1985-2008.	Retrospective cohort study	Scotland	Singleton births of infants with birth weight >500 g born at 28-43 weeks' gestation	An infant born showing no signs of life after 28 weeks gestation	Both	on 12	Socio-economic (area)	Deprivation according to Carstairs Index (car ownership, unemployment, overcrowding, and social class)
Bambang 2000 [44]	To study the relationship between cause-specific perinatal deathrates, material deprivation and birthweight among births in 3 consecutive yearsin the West Midlands Health Region	Retrospective cohort study.	West Midlands Health Region	All live and stillbirths to mothers with addresses in the WMHR in 1991, 1992 and 1993.	Not given	Both	September 2019	Socio-economic (area)	Townsend Deprivation Index
Khalil 2013 [45]	To examine the association between maternal-racial origin and a wide range of adverse pregnancy outcomes after adjustment for confounding factors in obstetric history and maternal characteristics	Retrospective cohort study.	London	Singleton pregnancies with a live fetus at 11 + 0 to13+ 6 weeks	Fetal deaths at or after 24 weeks	Both	. Downloade	Ethnicity	Ethnicity - broad categories (Caucasian, Afro-Caribbean, East Asian, South Asian, Mixed)
Sutan 2010 [46]	To determine the risk factors of unexplained antepartum stillbirth in Scotland from 1994 to 2003 and assess their value as a screening tool	Retrospective cohort study.	Scotland	All stillborn and live births in Scotland from 1994 to 2003	Not given	Antepartui	ægrom http://bmjopen.bmj.com/ an April 23,	Socio-economic (area) and place (urban/rural)	Urban/Rural classified according to settlement size and remoteness; Deprivation according to Carstairs Morris Index
Murrells 1985 [47]	To examine changes in the data for stillbirth rates between 1949/50 and 1975	Retrospective cohort study	UK	Comparisons between data from 1949/50 and 1975	Not given	Both	mjopen.	Socio-economic (individual)	Registrar Generals Scale of occupational class
Neasham 2001 [48]	To investigate the variation of stillbirth and neonatal mortality due to congenital anomalies in relation to small-area measures of deprivation in a population-based study in England and Wales, 1986–96	Retrospective population based study	England and Wales	All births in England and Wales from 1986- 1996	On or after 24 weeks gestation	Both	bmj.com/ c	Socio-economic (area)	Carstairs Index
Antonovsky 1977 [49]	To examine the relationship between the components of infant mortality and social class by analysing the data available from infant mortality studies.	Review and commentary using secondary data	Western Europe and the United States	Uses data from infant mortality studies undertaken in Western Europe and the United States	Component of infant mortality - European nations require registration of stillbirths from 28 weeks gestation; most of United States from the 20th week of gestation.	Unclear	2024 by	Social class (individual)	Inconsistencies in reporting of class across countries discussed and complexity of occupation, and education as measures.
Macintyre 1986 [50]	To place debates (by social epidemiologists and medical sociologists among others attending to inequalities and health) in a broader and perhaps more traditional context: that of interest in observed social regularities in health, illness and death.	Review and commentary using secondary data	Britain	No specific population  - those where social patterning of health has been observed across the life-course	Unclear	Unclear	guest. Protected by c	Social class (occupational), gender, marital status, age, ethnicity, and area of residence	These six variables are t conceptualized as positions on dimensions of social differentiation which may be associated with particular patterns of life chances.

Bloor 1987 [51]	To examine more thoroughly than was possible for Black and his colleagues, the possible role of an artefactual element in mortality data for explanations concerning health inequalities.	Review and commentary using secondary data	UK	No specific population draws on historical, theoretical and routinely collected	Unclear	Unclear	29 <b>6</b> 72 on `	Socio-economic	Examines social processes whereby statistics are produced.
		data		statistical data reporting inequalities across the life-course			12 Septe		
Andrews 1993 [52]	First objective, to draw attention to remarkable patterns of diversity and change revealed by recent official statistics for ethnic minority infant deaths in England & Wales. Secondly, to suggest that these patterns represent a challenge to many orthodox explanations for inequalities in infant health offered by social scientists, not least materialist explanations. Thirdly, to propose that further development and evaluation of explanations is impeded by flaws in some of the indices and categories employed in the construction of data.	Review and commentary using secondary data	England and Wales	Uses OPCS data on ethnic inequalities in infant deaths	Unclear	Unclear	mber 2019. Downloa	Ethnicity	Argues ethnicity is as much a matter of the way in which boundaries are created and reproduced between groups as the internal contents of ethnic cultures.
Kingdon 2016 [53]	To review the concept of society and discusses stillbirth as a social issue	Stillbirth used as a case study	Includes data from the UK	No specific population (stillbirth used as a case study)	A baby which has issued forth from its mother after the 24th week of pregnancy and which did not breathe or show any other signs of life' [UK definitionothers given]	Both	mber 2019. Downloaded from http://bmjo	Socio-economic, ethnic and gender	Multi-dimensional across place, time and culture
Black 1980 [54]	The first Government authorised attempt to explain trends in inequalities in health and to relate these to the policies intended to promote, as well as restore health.	Government Enquiry utilising official statistics	UK	Studies reporting inequalities based on occupational social class across the lifecourse	Not defined	Unclear	pen.bmj.com	Socio-economic (individual)	Registrar Generals Scale of occupational class
Whitehead 1988 [55]	To update evidence following on from the Black Report (see above). The original aim of the Health Divide was to draw together, to summarise the wide-ranging new evidence, and to describe what had happened and could happen, in policy development.	Government Enquiry utilising official statistics	UK	Studies reporting inequalities based on occupational social class, and unemployment, income, housing, material and social deprivation in small areas, gender and ethnicity.	Undefined	Unclear	on April 23, 2024 by g	Socio-economic	Registrar Generals Scale of occupational class
Garcia 2015 [56]	'What specific BAME maternity interventions exist for UK-based BAME women?'	Review and commentary	HICs including UK	Studies from HICs	Authors use an infant born with no life signs after 24 weeks gestation - however included studies in the review may have used different definitions	Unclear	luest. Protected by	Ethnicity (BAME - Black, Asian and Minority Ethnic)	"BAME groups" - Asian Indian, Pakistani, Bangladeshi, Kashmiri, Black African, Black Caribbean, Arabian, Traveller [defined as a gypsy or Irish traveller],

			ВМЈ	Open			mjopen-2019-029 <b>6</b> 72		
							-029672 on		Chinese, Mediterranean.
Flenady 2011 [57]	To present priority areas for stillbirth prevention, and interventions and research to address these priorities in high-income countries.	Review and commentary	HICs including UK	Studies from HICs	Death of a baby 500 g or more, or 22 weeks' gestation or more	Both	12	Socio-economic disadvantage	"Socioeconomic disadvantage" includes maternal education, ethnicity
Flenady 2016 [58]	To summarise the status of stillbirths in HICs and suggest strategies to accelerate momentum in the reduction of stillbirths and to meet parents' needs when their baby is stillborn.	Review and commentary	HICs including UK	Studies from HICs	Late gestation stillbirth" as >28 weeks and "early gestation stillbirth" as occurring prior to 28 weeks gestation	Both	mber 2019. D	Socio-economic disadvantage	As above
Pickton 1987 [59]	To analyse, describe and explain the distribution of perinatal mortality within an urban area	Case-control with geographical component (statistical and cartographical)	Greater Leicester	All births in Greater Leicester in the study periods	Stillbirth (late fetal death) = death after 28 weeks gestation	Both	dwnloaded from ht	Socio-economic (area and individual), ethnicity	Registrar Generals Scale of occupational class, employment housing, education level. Measures of ethnicity very broad (Asian, New Commonwealth).
Slogget 1998 [60]	To determine whether inequality in stillbirth risk between social strata has changed over time	Retrospective cohort study.	England and Wales	A random sample of more than 300 000 people enumerated at the 1981 census	Not defined	Unclear	September 2019. Downloaded from http://bmjopen.bmj.com/ or	Socio-economic (area and individual)	Deprivation was assessed by a "Townsend/Carstairs-like" index of four components unemployment, car ownership, home ownership, lower employment status according to Registrar General.
Joyce 1999 [61]	To determine whether social class (individual level) or a census- based deprivation score (based on area of residence) is a better predictor of stillbirth rates	Unclear	South Thames (West)	All births 1993-95	Not defined	Unclear	April 23,	Socio-economic (individual and area)	Social class using the Registrar General categories; area deprivation by Townsend score
Dummer 2000 [62]	To determine whether inequality in stillbirth risk between social strata has changed over time	Population based retrospective study	Cumbria compared with England & Wales data	For Cumbria all birth occurring between 1950-93: For E & W all births occurring between 1981-92	Two definitions given reflecting the change from 28 wks to 24wks that occurred in 1992	Both	2024 by guest		
Dickinson 2002 [63]	To investigate whether stillbirth risk was higher, and the effect of deprivation on inequality in stillbirth risk more marked, in rural than in urban areas	Retrospective cohort study	Cumbria	Singleton births between 01/01/1950 – 30/09/1992 to mothers in the area now designated as Cumbria	A baby born dead after at least 28 weeks' gestation	Unclear	: Protected by copyright.	Socio-economic (individual, area and place)	Social Class - based on fathers occupation Deprivation based on several scores including IMD
	For nee	r review only	- http://hmionen	ı.bmj.com/site/ak	oout/quidelines v	html	copyright.		

			ВМЈ	Open			mjopen-2019-029 <b>6</b> 72		
							19-029		
Pattenden 2011 [64]	To analyse adverse pregnancy outcomes, concentrating on estimating the extent of geographical variation from large areas (approximately 400 000 persons population) to small areas (wards, approximately 100 times smaller)	Population based retrospective cohort study	Northern Ireland	Birth records from Northern Ireland over the years 1992-2002	24 weeks and over		672 on 12 September	Socio-economic (area) and place	Deprivation according to The Noble Index (households receiving means-tested benefits Settlement size (rural areas, village, small town, medium town, large town, or city).
Rutter 1990 [65]	To review the literature on psychosocial factors in pregnancy outcome and to present a model which attempts to integrate the findings theoretically.	Theoretical	UK	UK	Not defined	Unclear	ber 2019	Socio-economic (individual)	Registrar Generals Scale of occupational class
CEMCH 2006 [66]	To report on perinatal mortality statistics	Confidential Enquiry	England, Wales & Northern Ireland	England, Wales and NI Perinatal deaths during 2004	24th week of pregnancy			Socio-economic (area), ethnicity	Ethnicity approximate Indices of Multiple Deprivation (IMD) scores
CMACE 2011 [67]	To report on perinatal mortality statistics	Confidential Enquiry	UK	UK Perinatal deaths in 2009	A baby delivered without signs of life after 23+6 weeks of pregnancy	Both	aded from http:	Socio-economic (area), ethnicity, and place (inferred regional and NHS Trust differences)	Ethnicity - broad categories; IMD data based on quintiles of deprivation; Employment status - simple yes/no
Draper 2015 [68]	Perinatal confidential enquiry carried out as part of the MBRRACE-UK programme of work, aim to focus on term, singleton, normally formed, antepartum stillbirths.	Confidential Enquiry	UK	Representative sample of stillbirths from those identified as eligible for review by MBRRACE-UK in April 2014	Term singleton, normally formed antepartum stillbirth (no precise definition given)	Both  Both  Antepartum	://bmjopen.bmj.com/ on /	Socio-economic (area and individual), ethnicity	Ethnicity - broad categories; IMD data based on quintiles of deprivation; Employment status - broad categories (employed/unemploys student; looking after home/family; permanently sick/disabled)
Manktelow 2015 [69]	To collect, analyse and report national surveillance data to stimulate and evaluate improvements in health care for mothers and babies.	Confidential Enquiry	UK	All births in the UK in 2013	Stillbirth: a baby delivered at or after 24+0 weeks gestational age showing no signs of life, irrespective of when the death occurred		April 23, 2024 by guest	Socio-economic (area), ethnicity, and place (inferred regional and NHS Trust differences)	Deprivation -Based or mothers' postcodes a time of delivery, using the Children in Low- Income Famillies Loca Mosmer Famillies Loca Mosmer Famillies Loca Mosmer Famillies Loca Broad categories [white, mixed, Asian of British Asian, Black of Black British, other]
Manktelow 2016 [70]	To collect, analyse and report national surveillance data to stimulate and evaluate improvements in health care for mothers and babies.	Confidential Enquiry	UK	All births in the UK in 2014	A baby delivered at or after 24+0 weeks gestational age showing no signs of life, irrespective of when the death occurred [including		t, Protected by copyright.	Socio-economic (area), ethnicity, and place (inferred regional and NHS Trust differences)	Deprivation -Based or mothers' postcodes at time of delivery, using the Children in Low- Income Families Loca Measure; Ethnicity - Broad categories
				n.bmj.com/site/ak			copyright.		

			ВМЈ	Open		mjopen-2019-029672		
					separate definitions for ante & intrapartum stillbirths]	)29672 on		[white, mixed, Asian or British Asian, Black or Black British, other]
Gardosi 2005 [71]	summarise a recently completed analysis of stillbirth and infant mortality trends from 1998-2003 in the West Midlands; 2. assess the main categories contributing to these deaths; 3. analyse the trends of mortality associated with inequalities and the association with deprivation within different mortality subgroups.	Retrospective case-note audit	West Midlands	Perinatal mortality and infant mortality from the West Midlands over a 5 year period (1998 -2003)	> 24 weeks	Both September	Socio-economic (area)	Indices of Multiple Deprivation (IMD)
Gardosi 2007 [72]	To explore trends in perinatal mortality rates (including stillbirths) in the West Midlands over the period 1997-2005	Retrospective case-note audit	West Midlands	All births in the West Midlands Health Region over the period 1997-2005	24.0 weeks of pregnancy	2019.	Socio-economic (area), ethnicity	IMD (as above), ethnicity as recorded in notes
Gardosi 2009 [73]	To explore trends in stillbirths and infant deaths in the West Midlands over the period 1997-2007/8	Retrospective case-note audit	West Midlands	All births in the West Midlands Health Region over the period 1997-2007/8	24.0 weeks of pregnancy	Both Both	Socio-economic (area)	IMD (as above)
Gardosi 2011 [74]	To understand the causes underlying perinatal mortality and develop strategies for prevention	Retrospective case-note audit and survey of community midwives	West Midlands	Perinatal deaths	Perinatal mortality – defined as a stillbirth or a death of a live born baby in its first week of life.	Antepartum http://bmjopen.bm	Socio-economic (area), ethnicity	IMD (as above) Ethnicity - More subtle (Africa, African - Caribbean, Indian, Pakistani, Bangladeshi, Eastern European, Middle Eastern).
Tang 2008 [75]	To identify appropriate health targets by investigating associations between social deprivation and causes of stillbirth in Liverpool	Retrospective case-note audit	Liverpool	All stillbirths at local NHS Trust	Stillbirths from 24 weeks gestation.	Both en.b	Socio-economic (area)	Indices of Multiple Deprivation (IMD)
Man 2016a [76]	What is the role of placental histological examination in determining the cause of intrauterine death?	Descriptive	London	Intrauterine deaths	Stillbirths (≥ 24 weeks)	Both J. CO	Ethnicity	Ethnicity – general categories
Man 2016b [77]	To present contemporary demographic findings from a large series of > 1000 intrauterine deaths in London which underwent autopsy investigation, and to examine these features in relation to the gestational age at which fetal death occurred, including deaths across both second and third trimesters, which are not captured by traditional registry-based approaches	Descriptive	London	Intrauterine deaths	Stillbirths (≥ 24 weeks)	Both Both Both 23,	Ethnicity	Ethnicity – general categories
Man 2016c [78]	To examine factors relating to determination of cause of death using a large dataset extracted from an autopsy research database including cases from two specialist centers, in which observer bias was reduced as far as possible by recording objectively findings at autopsy and assigning causes and classifications of death based on predetermined criteria.	Descriptive	London	Intrauterine deaths	Stillbirths (≥ 24 weeks)	2024 by guest.	Ethnicity	Ethnicity – general categories
Garcia 2017 [Unpublished PhD Thesis]	How do health beliefs influence health behaviour and contribute to perinatal mortality in babies born to Pakistani, Bangladeshi and WB women living in Luton?	Mixed Methods	Luton, UK	White, Pakistani and Bangladeshi women in Luton	Delivery of an infant with 'no signs of life' between 24 and 42 weeks of gestation	Both Both	Intersectional – socioeconomic (individual and area), ethnicity and immigration status	Education level, ethnicity by country of heritage. Immigration status by years in UK.

# **BMJ Open**

# Inequalities and stillbirth in the UK: A meta-narrative review

Journal:	BMJ Open
Manuscript ID	bmjopen-2019-029672.R2
Article Type:	Research
Date Submitted by the Author:	25-Jul-2019
Complete List of Authors:	Kingdon, Carol; University of Central Lancashire, ReaCH group Roberts, Devender; Liverpool Womens NHS Foundation Trust, Department of Obstetrics Turner, Mark; Liverpool Women's Hospital NHS Foundation Trust, Storey, Claire; International Stillbirth Alliance, Crossland, Nicola; University of Central Lancashire, School of Health Finlayson, Kenneth; University of Central Lancashire, School of Health Downe, Soo; University of Central Lancashire, research in childbirth and health
<b>Primary Subject Heading</b> :	Public health
Secondary Subject Heading:	Obstetrics and gynaecology, Sociology
Keywords:	stillbirth, inequalities, deprivation, ethnicity

SCHOLARONE™ Manuscripts

# Inequalities and stillbirth in the UK: A meta-narrative review

## **Corresponding Author**

Dr Carol Kingdon, Senior Research Fellow,

School of Community Health and Midwifery, Faculty of Health and Wellbeing,

University of Central Lancashire, Preston, United Kingdom. PR1 2HE

Tel: 07935744004

ckingdon@uclan.ac.uk

ORCID ID 0000-0002-5958-9257

#### Co-authors

Dr Devender Roberts, Consultant Obstetrician in Fetal Medicine,
Liverpool Women's NHS Foundation Trust, Crown Street, Liverpool, United Kingdom. L8 7SS

<u>Devender.Roberts@lwh.nhs.uk</u>

Professor Mark Turner, Reader in Neonatology/Consultant Neonatologist,
Department of Women's and Children's Health, Institute of Translational Medicine,
Faculty of Health and Life Science, University of Liverpool, Liverpool, United Kingdom. L8 7SS
Mark.Turner@liverpool.ac.uk

Ms Claire Storey, Co-chair, International Stillbirth Alliance, 61 Kings Drive, Stoke Gifford, Bristol, United Kingdom. BS34 8RD storey.claire@yahoo.com

Dr Nicola Crossland, Research Associate,
School of Community Health and Midwifery, Faculty of Health and Wellbeing,
University of Central Lancashire, Preston, United Kingdom. PR1 2HE
<a href="mailto:ncrossland@uclan.ac.uk">ncrossland@uclan.ac.uk</a>

Mr Kenneth William Finlayson, Research Associate,
School of Community Health and Midwifery, Faculty of Health and Wellbeing,
University of Central Lancashire, Preston, United Kingdom. PR1 2HE
<a href="mailto:KWFinlayson1@uclan.ac.uk">KWFinlayson1@uclan.ac.uk</a>

Professor Soo Downe, Professor of Midwifery,
School of Community Health and Midwifery, Faculty of Health and Wellbeing,
University of Central Lancashire Preston, United Kingdom. PR1 2HE
sdowne@ulcan.ac.uk

Word count: 4,006

## **Abstract**

**Objective:** To review what is known about the relationship between stillbirth and inequalities from different disciplinary perspectives to inform stillbirth prevention strategies.

**Design:** Systematic review using the meta-narrative method.

**Setting:** Studies undertaken in the UK.

**Data sources:** Scoping phase: experts in field, exploratory electronic searches and hand-searching. Systematic searches phase: Nine databases with no geographical or date restrictions. Non-English language studies were excluded.

**Study selection:** Any investigation of stillbirth and inequalities with a UK component. **Data extraction and synthesis:** Three authors extracted data and assessed study quality. Data were summarised, tabulated and presented graphically before synthesis of the unfolding storyline by research tradition; and then of the commonalities, differences and interplays between narratives into resultant summary meta-themes.

**Results:** 54 sources, from nine distinctive research traditions were included. Evidence of associations between social inequalities and stillbirth spanned 70 years. Across research traditions there was recurrent evidence of the social gradient remaining constant or increasing, fuelling repeated calls for action (*Meta-theme 1: Something must be done*). There was less evidence of an effective response to these calls. Data pertaining to socio-economic, area and ethnic disparities were routinely collected, but not consistently recorded, monitored or reported in relation to stillbirth (*Meta-theme 2: Problems of precision*). Many studies stressed the interplay of socio-economic status, deprivation or ethnicity with aggregated factors including heritable, structural, environmental, and lifestyle factors (Meta-theme 3: *Moving from associations towards intersectionality and intervention(s)*). No intervention studies were identified.

**Conclusion:** Research investigating inequalities and stillbirth in the UK is underdeveloped. This is despite repeated evidence of an association between stillbirth risk and poverty, and stillbirth risk, poverty and ethnicity. A specific research forum is required to lead the development of research and policy in this area, which can harness the multiple relevant research perspectives and address the intersections between different policy areas.

Protocol registration number: CRD42017079228

**Keywords:** Stillbirth, inequalities, deprivation, social class, poverty, ethnicity, metanarrative.

# **Article summary**

## Strengths and limitations of this study

- Meta-narrative is a systematic methodological approach to understand how multiple disciplines and different philosophical perspectives have researched a question over time.
- This study used a meta-narrative approach to investigate the association between inequalities and stillbirth in the UK.
- We adhered to the RAMESES standards for meta-narrative reviews to ensure fidelity with the methodology.
- We used a multipronged approach to retrieving sources that included exploratory searches, systematic searches, hand searches, expert opinion, and forward and back-chaining.
- By limiting the review to UK-based studies only, we were able to focus with greater acuity on the commonalities and contestations between research traditions, but this may have led us to miss important research on the association between stillbirth and inequalities from other countries, of relevance both in the UK context and globally.

**Funding statement:** This work was supported by SANDS, the Stillbirth and Neonatal Death Charity.

## INTRODUCTION

Avoidable inequalities in mortality across the life course are a global concern. [1] Ten countries account for 66% of the world's stillbirths, with most (98%) occurring in low-and middle-income countries (LMICs). [2] Inequalities exist within and between high-income countries (HICs) too. In 2011, the Lancet *Stillbirth* Series highlighted that the UK's stillbirth rate was one of the highest of all HICs. [3] In 2016 the second Lancet Series *Ending Preventable Stillbirths* reported that while overall stillbirth rates were falling in HICs, improvement was slower than expected, and significant inequalities within rates remained. [4] The UK's stillbirth rate continues to remain high in comparison to other high-income countries. [5]

The government's ambition is to halve the stillbirth rate in England by 2025, which would require the rate to fall to 2.6 per 1,000 total births. [6] In 2017, the stillbirth rate in England and Wales was to 4.2 per 1,000 total births. [7] Medical reasons for stillbirth are well known and strategies for prevention routine. Ongoing initiatives include the Safer Maternity Care strategic plan, [8] Saving Babies Lives Care Bundle, [9], Each Baby Counts, [10] the Perinatal Mortality Review Tool [11] and annual Perinatal Mortality Reports (MBRRACE-UK) [5]. The association between social determinants and stillbirth is less well understood. Clinicians acknowledge the need to do more to prevent stillbirth in women from socially disadvantaged groups. In England in 2017, the stillbirth rate in the most deprived areas was 5.5 per 1,000 total births, compared with 3.0 per 1,000 total births in the least deprived areas. [7]

The UK began to develop policies to address health inequalities in general following *The Acheson Inquiry into Inequalities in Health*. [12] The Marmot Review *Fair Society, Healthy lives*, published in 2010, progressed the UK's inequalities agenda by emphasising the importance of taking a life-course approach, starting with the early years and family building. [13] The key messages of the Marmot Review emphasised that there is a social gradient in health in the UK, whereby the lower an individual's social position the worse his or her health, which is unfair, and that this requires action across all the social determinants of health.

Public Health England's current strategy for action on inequalities *Reducing health inequalities: system, scale and sustainability* [14] is underpinned by the Dahlgren and Whitehead rainbow model of the social determinants of health. [15] This model offers a framework to explore the relative influence of these determinants on different health outcomes and the interactions between the various determinants. These are all potential mechanisms by which stillbirth risk maybe increased. What is missing from current stillbirth research agendas is an overarching synthesis of clinical and social science evidence to clarify the range of individual (including biological and behavioural), social and environmental mechanisms of increased stillbirth risk, the intersections between these mechanisms, and strategies to tackle them. This review sought to fill this knowledge gap.

We undertook an inter-disciplinary evidence synthesis (using a meta-narrative approach) to understand how structural factors, lifestyle factors, and clinical factors intersect to increase stillbirth risk, and to inform future strategies to manage at-risk pregnancies. The broad research question was what is the relationship between inequality and stillbirth, how has this been studied, and with what effects?

# **METHODS**

We conducted a systematic review using the meta-narrative method, [16-18] in accordance with the RAMESES standards. [19] A RAMESES checklist is provided (Supplementary information file 1). [20] Our protocol [21] (supplementary file 2) specified four objectives:

- 1. To review the current body of knowledge of the relationship between inequalities and stillbirth across the natural and the social sciences;
- 2. To provide new insights into the interplay of biological, clinical, cultural and socioeconomic factors in increased stillbirth risk;
- 3. To explore the impact of interventions on inequalities;

4. To provide a narrative summary of this research for stakeholders tasked with reducing preventable stillbirth.

## **Meta-narrative**

Meta-narrative review is a type of systematic review that was developed by Trisha Greenhalgh and colleagues. [16-18] Meta-narrative is a term for the unfolding storyline of research in a particular tradition or topic, which draws on the theoretical approach in Thomas Kuhn's writing on paradigms. [22] We used this approach to make sense of evidence from heterogeneous sources in which stillbirth and inequality have been variously conceptualised and studied over time. The method is underpinned by the methodological principles of pragmatism, pluralism, historicity, contestation, reflexivity and peer-review. As a method, meta-narrative review involves six key stages [17]:-

- Planning We registered our protocol with PROSPERO [21] and assembled a multi-disciplinary research team.
- 2. Iterative scoping searches and systematic electronic searches Initial searches were designed to map the diversity of perspectives and approaches. We contacted experts in the field of stillbirth research and from disciplines contributing to inequalities research. Exploratory searches were conducted using the search term "stillbirth" in 13 databases in health and the humanities (Supplementary File 3). Systematic searches were conducted in November 2017 in MEDLINE, Embase, CINAHL, PsycINFO, Popline, Historical abstracts, Humanities International Complete, Race Relations Abstract, and SocIndex. See supplementary file 4: example systematic search strategy. An English language restriction was imposed, but no geographical or date restrictions. In our protocol, inclusion criteria were any study design (quantitative, qualitative or mixed-methods) investigating stillbirth and inequality, in a high-, middle-, or low-income setting. Following initial screening of titles and abstracts a pragmatic decision was made by the team to include

only studies with a UK component. Unchanged exclusion criteria from the protocol were: any study in non-English language; of pregnancy loss <20 weeks gestation; of perinatal loss in the neonatal period; only involving participants who had assisted conception. The decision to exclude studies involving participants who had assisted conception was based on evidence of increased risk of stillbirth in pregnancies following IVF/ICSI. We applied this criteria during the database searches where it was possible to exclude studies focusing specifically on assisted conception. Screening was independently undertaken by three authors (NC, KF, CK), who also assigned potential inclusions to disciplinary categories at this stage (See supplementary file 5: screening tool).

- 3. **Mapping** A data extraction form was developed based on one used in earlier reviews, [23] which was adapted for the purpose of this metanarrative review. Additional fields were added to capture data relating to how inequalities and stillbirth were conceptualised, defined and theorised. The form was piloted by extracting data from a subset of five papers (taken from across the research traditions) to test for applicability to the metanarrative, and refined. Extracted data was then summarised, tabulated and presented. During this phase the team had lengthy discussions about which traditions were represented, the overlap between them, and their distinctiveness. We classified traditions based on the distinctiveness of their lens (or in other words paradigm). This involved consideration of scope, historical roots, key concepts, assumptions, theoretical basis, kinds of research questions asked, and the methods used.
- 4. Appraisal We stated in our protocol that all articles that met the inclusion criteria would be independently assessed by three researchers to minimize bias. During the process of the review it became apparent that quality appraisal of all quantitative studies using the appropriate checklists from the Critical Appraisal Skills Programme (CASP) Toolkit was not appropriate, with quality more suitably judged by the prevailing standards in each tradition. That said it was fitting to use CASP tools [24] for some studies in the epidemiological

tradition and the Walsh and Downe tool for qualitative research quality appraisal. [25]

- 5. Synthesis phase The identification of the meta-themes was via a two-part synthesis; (1) at the level of the traditions, which unfolded in the mapping phase and (2) at the level of data extraction from primary studies across traditions. Part one involved evaluating the meta-narratives to identify and compare how the different research traditions conceptualized and theorized the topic, and the methodological approaches and study designs used. Differences in findings between the resulting meta-narratives were analysed interpretively to produce further insights. Part two of the synthesis process involved paradigm bridging (seeking commonalities in underlying conceptual and theoretical assumptions), paradigm bracketing (highlighting differences in these assumptions), interplay (exploring tensions) and meta-theorizing (exploring patterns that span conflicting understandings) to construct summary meta-themes. KF, NC and CK undertook the initial analysis and synthesis processes, with input from DR, MT, CS and SD.
- 6. **Recommendations phase** We engaged with local clinical networks and the national Stillbirth and Neonatal Death Charity (SANDS) to formulate recommendations.

# Patient and public involvement

Author Claire Storey is a parent and Vice-Chair of the International Stillbirth Alliance and was involved in the design, and conduct of the review and the writing of this paper.

# **RESULTS**

From electronic searches of nine databases a total of 13,610 records were identified. Following duplicate removal 4,934 records were screened (Figure 1: RAMESES-

PRISMA diagram). We included 54 sources from nine research traditions spanning the period 1945-2017. [26-78] Table 1 provides a summary of included research Chronologically, these traditions were Social Medicine traditions. Epidemiology [32-48]; Medical Sociology [49-53]; Public Health [54-58]; Spatial Epidemiology [59-64]; Social Psychology [65]; Audits, Reports and Confidential Enquiries [66-74]; Fetal-Maternal Medicine [75-78] and Nursing and Midwifery [Garcia, Perinatal mortality in Pakistani, Bangladeshi and White British mothers in Luton]. Supplementary File 6 provides details about the characteristics of included sources. With the exception of Epidemiology (n=17), most traditions generated few relevant papers. All research traditions utilised epidemiological data. We included one mixedmethod study reporting qualitative data. No intervention studies were identified. Lack of studies, heterogeneity of study design, definitions of stillbirth, and measurement of inequalities between studies, traditions and over time meant meta-analysis was not practical. Figure 2 maps the traditions contribution over time and the declining national stillbirth rate.

# Synthesis within traditions

Table 1 summarises the unfolding storylines by research tradition and their conceptualisation of inequalities.

# **Table 1: Summary of included research traditions**

Research tradition	Academic discipline	Definition and scope	Unfolding storyline $\frac{1}{N}$	Inequalities conceptualised as	Included references
Social Medicine	Medicine	Social Medicine is a branch of medicine that uses epidemiological methods to establish a problem exists, determining factors and opportunities for preventative action. The tradition is distinctive in its thought on the interconnectedness between biological factors (i.e. mother's age) that have meaning whatever the social context and social factors (i.e. occupational social class) that derive their meaning from social organisation in human life emulating political economy concerns.	The Social Medicine [26-31] storyline begins with the investigation of how social and economic faces influenced the decline in stillbirths and early neonatal deaths in Scotland, England and Wales, between 1939 and 1944. Bare [26] attributed this fall to the improved nutrition of the mothers during pregnancy, a consequence of the national distribution and consumption of milk and other foods important for health during the second world war. These improvements affected every area, see group and parity. By 1949, the decline in the stillbirth rate had slowed, despite the introduction of the National Health Service (MIS). Four papers, from a series in The Lancet in 1955 [27-30] sought to understand why. The last paper concluded the independent effects of social class, region, the mother's age and parity on stillbirth risk. Illsley [31] showed how occupational class may be more than a measure of inequality simply based on environmental conditions at the time of maternity, reporting that it can also be a marker of a woman's personal characteristics (height, physique, health, intelligence and nutrition), education and social habits. Women who were intergenerationally upwardly socially mobile at marriage experienced less stillbirths.	A variety of social factors that combine with biological characteristics to increase vulnerability to stillbirth risk.	N=6 [26-31]
Epidemiology	Medicine	Epidemiology, developed out of the bio-medical model as a specific line of inquiry. Initially epidemiology focused exclusively on epidemics of communicable diseases but subsequently expanded to address endemic communicable diseases and non-communicable infectious diseases. It is the study of the distribution and determinants of health-related states (especially disease), and the application of findings to the control of diseases and other health problems.	The Epidemiology [32-48] storyline is characterised by its increasingly sophisticated use of data and the repetition of the same or similar findings over time. Of the seventeen studies aligned to this tradition, six were landmark papers, repeatedly referenced within the field. [32-37] Although most authors highlighted a significant decrease in UK stillbirth rates sing the 1960's, studies repeatedly showed that the social gradient remained constant. [36,37,41] Within overall stillbirth rates, being the 1960's, studies repeatedly showed that the social gradient remained constant. [36,37,41] Within overall stillbirth rates sing the 1960's, studies repeatedly showed that the social gradient remained constant. [36,37,41] Within overall stillbirth rates sing the 1960's, studies repeatedly showed that the social gradient remained constant. [36,37,41] Within overall stillbirth rates sing the 1960's, studies repeatedly showed social gradient remained to the season of stillbirth across relatively later studies of increased risk of stillbirth across relatively large populations. Early studies used the Registrar General's Scale of occupational social class as a measure of inequality, later studies used the socio-economic classification scheme. Other studies still utilised the term 'deprivation' to signify inequality. In most of the studies using deprivation as a factor the risk of stillbirth increases with increasing levels of deprivation [34,43,44] although this is not always the case. [38] Epidemiological studies looking at athinicity as a measure of inequality are a relatively recent phenomenon and do not show the same level of consistency, although the rates of stillbirth for women of African-Caribbean origin remain at twice the rate of white women. [38,45] Studies explosing the stillbirth rates of women of Asian origin show a degree of variance with some authors highlighting an increased rate – equivalent to women of African-Caribbean origin temain at twice the rate of white women. [38,45] Studies explosing the stillb	A variety of factors (social class, living in an area of deprivation, occupation of partner, ethnicity, etc.) associated with an increased relative risk of stillbirth.	N=17 [32-48]
Medical Sociology	Sociology	Medical Sociology is the study of the social causes and consequences of health and illness. This tradition has positivist and interpretative, theoretical and empirical, quantitative, qualitative, and mixed-methods, and cross-disciplinary branches. The persistence of social class gradients despite the demographic and epidemiological changes associated with the transition to modernity was an important focus during the 1970's and 1980's. During the 1990's research increasingly focused on lay understandings of health and illness and lived realities.	The Medical Sociology [49-53] storyline is theoretical. Early sociological explanations for the persistence of the social gradient in stillbirth encompassed theories of capital assets (the physique, stature, nutrition of the mother), social mobility (a direct thread from Social Medicine [31]), and time-lag (whereby developments in healthcare take time to reach those most in need, benefiting those better off first). [49] After the seminal Black Report [54] more nuanced considerations of gender, age, ethnicity and area of residence, alongside occupational class, as simultaneous and overlapping vulnerabilities, were developed. [50-52] These encompassed the broad consideration of life circumstances, behaviours, and beliefs/attitudes [50] and the precise disaggregation of the concept of 'deprivation' to reveal the complexity of materialist risks (and protections against these risks), which helps to explain the ambiguous association between economic deprivation and ethnicity. [52]	A set of social relations (rather than just a variable), which opens lived experience and multiplicity of factors at play (i.e. poverty, poor housing, nutrition, welfare) and relationship between structure and agency.	N=5 [49-53]
Public Health	Public Health	Public health is concerned with preventing disease, prolonging life and promoting health through organised efforts of society. From 18th and 19th century roots, during the 1980's there was a revival of public health policy. In the UK this coincided with a shift in thinking that morbidity or general health status had become the more important indicators of inequality, and increasing interest in individual behaviours and lifestyle as determinants of health.	The Public Health storyline [54-58] unites the seminal Black Report [54] (which had a major impactor research into inequalities in health in the UK), with seminal papers from the two Lancet Stillbirth Series [57-58] that were of equal significance to the stillbirth research and policy community. In the former publication [54] stillbirth is a crude cause of death category, used as part of efforts to explain general trends in inequalities in health, based principally on measures of occupational social class from which artefact, natural selection, structuralist, and behaviourist explanations, (alongside the need to build on the ist and unit of multiple causation) were developed. In the latter publications, distinguishing between different kinds of stillbirth and the imperance of making each stillbirth count, come alongside the need to build on the idea of interactions between factors that includes ald disadvantage. [57-58] The lack of targeted interventions for Black and Ethnic minority women in the UK, despite their complementaries of increased risk and known underutilisation of maternity services, was highlighted in the scoping review by Garcia. [56] 2016, there was an explicit recall to action to tackle inequalities and stillbirth within HICs by addressing structural factors (success poor housing, poverty) and factors, which limit women's access to antenatal care. [58]	An additional risk and considered in relation to providing targeted care to populations considered at risk.	N=5 [54-58]

			BMJ Open		
			BMJ Open pen-2019-029		
Spatial Epidemiology	Medical Geography and Epidemiology	Spatial epidemiology is concerned with the spatial analysis of disease incidence and prevalence. It uses geographical mapping and statistical modelling to understand the spatial distribution of disease, under the assumption that this will provide indications of the environmental contributors to the disease.	The Spatial Epidemiology [59-64] storyline begins in the late 1980s and attempts to address how community deprivation and individual social class might each contribute to risk of stillbirth. Studies looking at stillbirth and inequalities have investigated the relative importance of individual level (Registrar General Social Class) versus area level (e.g. Townsend Score) measures of inequality. Studies report contradictory findings, perhaps revealing the complexity of how individual (compositional) and area (context) effects interact to affect risk, with some reporting an enduring association between area and/or individual level deprivation and stillbirth risk [59,61-63] and others reporting no association [60,64]. The storyline of UK-based search into place effects on stillbirth risk has so far conceptualised geographical areas as "containers" of people, rather than seeing place as socially constructed.	A variety of factors (social class, living in an area of deprivation, occupation of partner, ethnicity) associated with an increased relative risk of stillbirth.	N=6 [59-64]
Social Psychology	Psychology	Social psychology is the study of human social behaviour, emotion and cognition. With its focus on both the individual and society, it draws on sociological and psychological perspectives Research methods involve both quantitative and qualitative approaches, and include surveys, participant observation, laboratory experiments, field experiments, and archival and content analyses. Experimental social psychology is underpinned by positivist assumptions, while other approaches such as critical social psychology, operate from a social constructionist stance.	The Social Psychology [65] storyline arose from the Black Report [54] and draws on theoretical expanations from the Black Report about the association between social inequality and ill-health. This storyline is represented by one paper from 1990 [65], which used secondary data (birth data from England Wales, 1980-1986) to develop a theoretical model of how social class may affect psychosocial mediators – emotional, social, and cognitive factors – which may in turn influence pregnancy outcome, either directly or mediated through behaviours and coping strategies. The proposed model suggests that material deprivation results in more negative life events while also reducing social support, and access to education and information. Stressful be events, unmitigated by social support, create stress, anxiety, depression, low self-esteem. Poor education, or access to information, lead to a lack of knowledge and to deleterious beliefs and attitudes. The combined emotional and cognitive effects produce coefficients and behaviours that increase the risk of negative pregnancy outcomes (i.e. smoking). [65]	A factor influencing health Inequalities can be seen to affect health via increasing psychosocial stress, which can then directly impact on health and also induce health- limiting behaviours	N=1 [65]
Audit Reports and Confidential Enquiries	Inter- disciplinary (Epidemiology, Obstetrics, Paediatrics, Midwifery)	Audits, Reports and Confidential Enquiries provide knowledge not always thought of as research, nevertheless it usefully uses routinely collected data to examine time-trends. As a tradition it incorporates a variety of approaches including epidemiology, economics and health policy and may be further informed by qualitative data and/or expert opinion. It includes 1992-2003 CESDI (Confidential Enquiry into Stillbirths and Deaths in Infancy), 2003-2011 CMACE (Centre for Maternal and Child Enquiries), and 2011 onwards MBRACE-UK (Mother and Babies: Reducing Risk through Audits and Confidential Enquiries across the UK).	The Audit, Reports and Confidential Enquiries storyline [66-74] builds on over 50 years of local and antional reporting of maternal and infant deaths. A key feature of these reports is the presentation of stillbirth rates at national, responsible to the subsequent comparisons between geographical units and benchmark averages. Over the years tirgs processes were modified and refined into the national Confidential Enquiry scheme [66,67] and, more recently, under the banner MBRACE-UK (Mother and Babies: Reducing Risk through Audits and Confidential Enquiries across the UK) [68-70]. Although we identified more than 20 national reports only five explored the association between inequalities and stillbirth [66-70] with the majority focusing on 'avoidable' health system and clinical failures. Where inequalities and stillbirth were identified they were discussed in relation to lifestyle factors (smoking, excess alcohol consumption, obesity) or regional or ethnic disparities associated with ingreased stillbirth risk. Four regional reports or audits from the West Midlands [71-74] attempt to look at stillbirth and inequalities explicitly by equating higher indices of multiple deprivation (IMD's) with increased stillbirth rates. These reports were more nuamed and identify a number of social and medical risk factors that could be screened for (alone or in combination) to predict risk distillbirth (e.g. unemployment, inappropriate housing, unsupported/difficult family circumstances, emotional factors/anxiety, maternal age <20 yrs or > 40 yrs, obesity, smoking, consanguinity, history of mental health issues). The authors of these reports also highlight fetal growth restriction (FGR) as a potential predictor of stillbirth in deprived communities.	Regional variations in stillbirth rates with recognition of differences between areas of deprivation (high and low) and ethnicity (White and Black & Asian populations).	N=9 [66-74]
Fetal-Maternal Medicine	Medicine	Maternal-fetal medicine is a subspecialty of obstetrics. Its focus is on 'high risk' pregnancies, including women who have a pre-existing illness or a pregnancy-induced illness, and congenital abnormalities It draws on and is related to perinatal epidemiology. The clinical focus includes preterm birth prevention, screening for fetal growth restriction, and placental histopathology.	The Fetal-maternal medicine storyline [75-78] included a study reporting that women living in areas of highest deprivation (IMD 1) were more likely to experience fetal growth restriction compared to women living in the least (IMD 29). [75] Approximately 46% of these women smoked, compared to 7% in the least deprived. The study concluded that targeted aptenatal management was key to stillbirth prevention amongst women living in the most deprived areas. This tradition also offered troe interlinked publications, which suggested that maternal ethnicity was associated with fetal loss at different gestations White women are latively more stillbirths (>24 weeks gestation) and Black women relatively more late intrauterine fetal deaths (20-23 weeks gestation) [76-78]. There was a higher risk of ascending genital infection for Black mothers relative to women from other ethnic greups. This was a relatively common cause for early intrauterine fetal death, peaking at around 22 weeks, [78].	A risk factor for stillbirth and depending on the type of study, may be included as a covariate in the analysis.	N=4 [75-78]
Nursing and Midwifery	Nursing and Midwifery	Nursing and Midwifery research draws from positivist and interpretative paradigms, utilising a range of quantitative and qualitative methods. This tradition has made a significant contribution to the body of knowledge about stillbirth and bereavement care.	Only one mixed-method single-site study was identified as characteristic of this tradition. Garcia, Reinatal mortality in Pakistani, Bangladeshi and White British mothers in Luton] It showed no statistically significant association between stillbirth and maternal ethnicity, but found more perinatal deaths in deprived areas. Qualitative interviews with White British, Pakistani, and Bangladeshi women identified health beliefs and behaviours common to all ethnic groups. These included little wareness of what to do about risk factors such as reduced fetal movements ("two days I delayed because I don't know what I near to do") and anxieties about being a burden to overstretched maternity services ("they could do without me taking up a bed, taking up their time, you put yourself at a lower scale than everyone else.") Health professionals perceived they had communicated information to women about stillbirth risks and the importance of seeking prompt care. Professionals did not view any particular of them [Asian women: Pakistani and Bangladeshi] are beholden on their partners to get them there) ("It doesn't matter whether there a Asian or whatever they are They don't have transport and they don't have money, they don't have access to actually get here	An additional vulnerability, and considered in relation to the importance of providing culturally appropriate care.	N=1 [Garcia, Perinatal mortality in Pakistani, Bangladesl and White British mothers in Luton]

## Synthesis across traditions

#### Meta-theme 1: Something must be done

Across time and research tradition the prevailing message was for action on inequalities and stillbirth. From the earliest included paper in Social Medicine that concludes 'there is still much to be done' [26] to a Public Health paper in The Lancet Ending Preventable Stillbirth Series 2016 that states 'programmes at community and country level need to improve health in disadvantaged families to address these inequalities' [58] the message is clear. The call to do something stems from persistent evidence of a social gradient coupled with perceptions of insufficient progress in diminishing stillbirth rates in the UK. In some research traditions stillbirth was used as an indicator of societal health, with references to the particularly low stillbirth rates achieved in Scandinavia commonplace. Despite the persistence of studies reporting the same or similar risk factors and the continuation of the social gradient exactly what kind of 'something should be done' is less clear. Evidence of effectiveness was absent for interventions at specific time-points, inter-generationally, at scale or targeted to social groups. The absence of stillbirths in inequalities reduction targets post-Acheson was identified as a specific barrier to action [71].

## Meta-theme 2: Problems of precision

Our meta-narrative approach highlighted how much of the challenge in seeking to act on inequalities and stillbirth lies in the lack of consensus and inherent complexities inherent to both. While there was persistent evidence of associations between stillbirth risk and poverty, and stillbirth risk, ethnicity and poverty, it was not possible to estimate the potential gain on stillbirth reduction if action was taken to reduce inequalities, because of problems with data availability and comparability. There were problems of precision in stillbirth definition and problems of precision in inequalities measurement. The traditions rooted in medical science offered the most analytic tools for defining when stillbirths happened (antepartum, intrapartum), at what gestation (early preterm, late preterm, term), and why in terms of clinical factors (classification according to ReCoDe, Wigglesworth, Aberdeen etc), but these definitions were not used consistently, and they rarely considered social inequalities as underlying factors.

Further problems of precision arose from how inequalities were variously conceived and measured, even when they were taken into account. In traditions informed by the social sciences, inequalities were broadly conceptualised as a set of social relations (rather than a variable/s), which opened up lived experience, multiple risk factors/ interactions between them, and consideration of the relationship between structure and agency in health and lifestyle. Further conceptual considerations arise from this, including socio-economic status/social class (an individual measure of inequality) based on occupation alone or in combination with income, education and culture (Social Medicine, Epidemiology, Medical Sociology, Public Health). The problem of how best to measure disadvantage was apparent across time. The artefact explanation for inequalities (which considers to what extent they are a construct of the measurement process) was particularly critical of the now defunct Registrar General's Scale. [31,50,51,54,55].

Deprivation (an area measure of inequality) was conceptualised according to the tool used to define it for which there was no consensus. Tools used included the Townsend deprivation index, Carstairs and Morris index, Jarman Deprivation Scores and the Index of Multiple Deprivation (IMD). A general question for the Spatial Epidemiology tradition was whether individual level deprivation and area deprivation are different and how they interact. [61-62] The problems associated with using crude categories to define ethnicity (i.e. white, Black, Asian) were also considered (Epidemiology, Medical Sociology, Public Health) and the complexities therein (i.e. benefits of more subtle classifications incorporating country of birth such as British Asian), including how such classifications are only proximate guides to experiences, practices, beliefs and lifestyles. In 1993, a matrix of country of birth, nationality, language group, religious affiliation, and (where appropriate) region, caste and sub-caste was proposed by Andrews and Jewson to test the combining variables, as well as suggesting a more fine-grain exploration of major variables if used as part of a national dataset. [52]

# Meta-theme 3: Moving from associations to intersectionality and intervention

All the traditions included in this review report evidence of associations between living in poverty and increased risk of stillbirth. However, despite more than 70 years of research equating inequality with increased stillbirth risk "any detailed study of why this should be so is surprisingly sparse" [50;p.393] This theme attempts to shine some 'light on the most appropriate times to provide support and the form(s) that such support should take.' [42:p11] To begin to address the need for intervention, one recent study triangulated epidemiological data with what women said (qualitative data). [Garcia, Perinatal mortality in Pakistani, Bangladeshi and White British mothers in Luton] In so doing it showed how the interactions between education level, socioeconomic status, cultural needs, language barriers, knowledge, likeliness to seek help, and assumptions by healthcare staff interact to make (or diminish) stillbirth risk in the current maternity care system. While, that study was the first study to claim an intersectionality approach, most publications across the research traditions suggest that further exploration of the interactions between risk factors, and within specific groups, is warranted.

Most of the contributory risk factors identified in this review are already well known and have been for some time. As summarised in Figure 3 risk factors for stillbirth encompass biological, clinical, behavioural, health service and social factors. Figure 3 provides a model from which to test the associations between factors, which is built on interdisciplinary evidence of the clinical causes of stillbirth, theories of natural and social selection. cultural/behavioural/lifestyle explanations, area effects. materialist/structuralist explanations and availability, access and quality of care. While some studies proposed antenatal screening for a combination of social factors (i.e. non-English speaking, unemployed household) in combination with behavioural factors (i.e. smoking) and clinical factors (i.e. previous IUGR), there was little consensus on specific factors, timing, or outcome if social conditions remain the same. [39,40,46]

#### DISCUSSION

This review highlights that research investigating what might work to reduce inequalities and stillbirth in the UK is underdeveloped. We identified nine research traditions in the field but, with the exception of epidemiology, these traditions had few studies within them. Across all traditions, epidemiological data persistently suggests that membership of a lower socio-economic group (as measured by an individual's occupation), or residing in a disadvantaged community (as measured by local area deprivation) is associated with increased incidence of stillbirth when compared with more socially advantaged counterparts. However, there was a paucity of research investigating why this should be so, despite repeated calls for action. A few studies found no association between living in an area of deprivation and increased stillbirth risk. Why this was so is also unclear. This review shows that the field is not only complex, but also dynamic, with the respective components (stillbirth per se and inequalities per se) beset by conceptual and methodological challenges. In terms of advancing understanding about the complexity of the interactions between factors associated with increased stillbirth risk this review is limited. Moreover, we found no studies of interventions targeted to reduce stillbirth in specific social groups or communities. Nonetheless, what this review does add is that stillbirth is a useful marker of success in addressing inequalities. It provides a cross-disciplinary foundation from which to develop and stimulate hypotheses about the relative influence of biological, clinical, behavioural, health service and social factors on birth outcomes and the interactions between these various determinants to inform future interventions.

#### Strengths and limitations

This study used a meta-narrative approach to investigate the association between inequalities and stillbirth. We adhered to the RAMESES standards for metanarrative review to ensure fidelity with the methodology. We used a multipronged approach to retrieving sources that included exploratory searches, systematic searches, hand searches, expert opinion, and forward and back chaining, which gave us a broad capture of relevant documents. By limiting the review to UK-based studies only, we were able to focus with greater acuity on the commonalities and contestations between research traditions. However, excluding studies from other countries may have led us

to miss important research on the association between stillbirth and inequalities of relevance both in the UK context and globally. The quality of some of the included sources in this review may also be considered an important limitation with the use of pre-specified quality appraisal tools [24-25] not deemed appropriate for all traditions. The interpretive nature of meta-narrative review means another team, outside of the UK, may classify the traditions differently. If, for example, Social Medicine and Medical Sociology were grouped together this would change the number and chronology of included traditions, although the interpretive synthesis across traditions is likely to remain intact.

#### Relationship of findings to other research

The current abundance of research investigating stillbirth prevention and bereavement care in the UK is a recent development as efforts to break the silence that has traditionally surrounded stillbirth have gained momentum and international ambition to reduce stillbirth has intensified over the last decade. [3-4,6,79-82] This goes some way to explain why the field is underdeveloped in comparison to the wider health inequalities literature on mortality and social gradient. We were surprised to find no intervention studies, although there is an acknowledged paucity of evaluations of interventions to reduce inequalities in health in general. [83-86] In the international literature, public health interventions seeking to reduce stillbirth are also sparse. The few that do exist include a food supplementation programme, which was offered to low-income women in the USA, [87] and a study looking at household air pollution in India, where wood and kerosene cooking fuel, more commonly used in low-income households, is known to be associated with stillbirth. [88] However, neither of these address the underlying structural components of disadvantage.

#### Implications for clinicians and policymakers

This review suggests that addressing inequalities as a component of stillbirth prevention in the UK demands intervention at many levels. The paucity of directly relevant research to the question of stillbirth prevention means policy makers must look towards what works to reduce inequalities for other related causes of death (i.e. sudden infant death, cardio-vascular disease and cancer). Health inequalities theory advocates intervening at specific time points during the life course (i.e. pregnancy and the early years), interventions that have impact over time (i.e. intra-generational and

inter-generational), interventions at scale (i.e. national policies) and interventions targeting specific groups (i.e. ethnic minorities and lower social classes). Addressing nutrition, service uptake and the wider social determinants of health may have knock on effects on many clinical outcomes, including stillbirth. [89] Scotland's *Early Years Collaborative* that encompasses cross-sector interventions at the level of individuals, groups, organisations and society, includes a specific stillbirth reduction target. [86] In the absence of a hierarchy of causation among these complex effects stillbirth specific research is well justified, as long as it is embedded in implementation, public health and caring for and about people.

In the global health community, remediable differences between and within countries are increasingly being addressed by agendas for health equity [1, 89]. The equity in health agenda is distinct in its focus on unnecessary and avoidable differences in health that are considered unfair and unjust. However, in the UK, inequalities is a term that has endured. [13-14, 86, 90] Future research in the field of inequalities and stillbirth would benefit from a more precise definition of the term inequalities that takes into account the concurrent global agenda for equity in health.

#### Unanswered questions and future research

It was not possible within or across traditions in this review to determine the potential gain of inequalities and stillbirth reduction. The field would benefit from a national consensus for routinely collected data and future research at population level. MBRRACE-UK, the RCOG and NHS England now have a high level of precision in stillbirth definition and national data capture. Since 2014, MBRRACE-UK has consistently used the Children in Low-income Families Local Measure. [5] There is also a simultaneous need for qualitative research that gets behind classificatory system labels to the lived realities of groups and communities. This review highlights there have long been important differences between communities and place that, for example, the classification Black, Asian, and Minority Ethnic (BAME), or indices of multiple deprivation (IMD) can conceal. Most of the factors associated with inequalities and stillbirth identified in this review are already well known, and have been for some time. The findings of the review suggest that looking at these well-known factors afresh is likely to provide new insights. For example, the reasons reported as to why women delayed seeking care for reduced fetal movements in this review resonate with the

findings of earlier reviews of antenatal care in general. [91-92] Similarly, studies of smoking behaviours, influence of social and community networks, the conditions in which people live, and the impact of current UK smoke-free policies that were identified on the periphery of studies included in this review, demand cross-disciplinary consideration in future strategies for stillbirth prevention. [93-95] Not least because, these particular components of antenatal care already feature as part of stillbirth reduction initiatives, but to-date, have had limited success. [82]

The role of social factors, modifiable lifestyle behaviours, and antenatal interventions in stillbirth prevention are current research priorities identified by the stillbirth community. [96] The results of this review indicate that there is little effective work across disciplines despite the long-recognised need for it. We recommend that the UK stillbirth research community overcome this by setting up a dedicated forum to promote intervention and implementation research in this area. The forum could have three roles: 1) Define the framework for future research by identifying the ways in which disciplines should interact; 2) Develop data standards for information relating to stillbirth and inequalities; 3) Develop and promote the intervention and implementation research, policy and practice agenda relating to stillbirth and inequality.

#### Conclusion

The UK government's current ambition is to halve the national stillbirth rate by 2025. Research investigating and, critically, addressing inequalities and stillbirth in the UK is underdeveloped. This is despite repeated evidence of an association between stillbirth risk and poverty, and stillbirth risk, poverty and ethnicity. A specific research forum is required to lead the development of research and policy, which can harness multiple relevant research perspectives and address the intersections between different policy areas. This review not only unifies calls for action, by connecting multi-disciplinary insight into these complexities, challenges and opportunities, it provides a starting point for a novel transdisciplinary response.

#### Figure legends

Figure 1: RAMESES-PRISMA Diagram

Figure 2: Timeline of included studies by research tradition and the stillbirth rate in

England and Wales 1945-2017

Figure 3: Factors associated with inequalities and stillbirth I

#### Supplementary files

Supplementary file 1: RAMESES Checklist

Supplementary file 2: Protocol

Supplementary file 3: Example exploratory searches

Supplementary file 4: Example systematic search strategy

Supplementary file 5: Screening tool

Supplementary file 6: Characteristics of included sources

**Author contributions:** CK, DR, MT and CS designed the review with input from SD. NC and KF conducted the searches, identification and screening with agreement by consensus of all authors on final inclusions. NC, KF and CK extracted data, and agreed initial storylines and final meta-themes with review by SD, DR, MT and CS. CK wrote the first draft of the paper. All authors read, commented and approved the final manuscript.

**Data sharing statement:** This is a review. Original research data is contained in the included studies. Data interpretation is contained in the manuscript. Further information can be obtained from the corresponding author.

**Conflict of interest statement**: Dr. Kingdon, Professor Downe, Dr. Crossland, Mr. Finlayson and Ms. Storey report a grant from SANDS, the Stillbirth and Neonatal Death Charity, during the conduct of the study. Ms. Storey also reports her position as Vice-Chair of the International Stillbirth Alliance (ISA). Dr. Roberts and Dr. Turner have nothing to disclose.

**Acknowledgements:** Rob Rawcliffe, UCLanPrint for his design input into Figure 3.

#### References

- Commission on Social Determinants of Health. Closing the gap in a generation: health equity through action on the social determinants of health. Final Report of the Commission on Social Determinants of Health 2008; Geneva, World Health Organization. <a href="https://www.who.int/social\_determinants/thecommission/finalreport/en/">https://www.who.int/social\_determinants/thecommission/finalreport/en/</a>. (accessed 25th June 2019).
- 2. Blencowe H, Cousens S, Jassir FB, et al. National, regional, and worldwide estimates of stillbirth rates in 2015, with trends from 2000: a systematic analysis. *The Lancet Global Health* 2016; 4(2):e98-e108 doi: 10.1016/S2214-109X(15)00275-2.
- 3. Mullen Z, Horton R (2011) Bringing stillbirth out of the shadows. *Lancet* 2011;377:1291–2. doi: http://dx.doi.org/10.1016/S0140-6736(11)60098-6.
- 4. Froen JF, Friberg IK, Lawn JE et al. Stillbirths: Progress and unfinished business. *Lancet* 2016; 6;387(10018):574-586. doi: 10.1016/S0140-6736(15)00818-1.
- Draper ES, Gallimore ID, Kurinczuk JJ, et al. on behalf of the MBRRACE-UK Collaboration. MBRRACE-UK Perinatal Mortality Surveillance Report, UK Perinatal Deaths for Births from January to December 2016. Leicester: The Infant Mortality and Morbidity Studies, Department of Health Sciences, University of Leicester. 2018.
- 6. GOV.UK New ambition to halve rate of stillbirths and infant deaths. <a href="https://www.gov.uk/government/news/new-ambition-to-halve-rate-of-stillbirths-and-infant-deaths">https://www.gov.uk/government/news/new-ambition-to-halve-rate-of-stillbirths-and-infant-deaths</a>. (accessed 1st February 2019).
- 7. Office of National Statistics Births in England and Wales: Live births, stillbirths and the intensity of childbearing, measured by the total fertility rate. <a href="https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarriages/livebirths/bulletins/birthsummarytablesenglandandwales/2017">https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarriages/livebirths/bulletins/birthsummarytablesenglandandwales/2017</a>. (accessed 1st February 2019).
- 8. Department of Health. Safer Maternity Care. The National Maternity Safety Strategy Progress and next steps. November 2017. <a href="https://www.gov.uk/government/publications/safer-maternity-care-progress-and-next-steps">https://www.gov.uk/government/publications/safer-maternity-care-progress-and-next-steps</a>. (accessed 25th June 2019).
- NHS England. Saving Babies' Lives Care Bundle. <a href="https://www.england.nhs.uk/mat-transformation/saving-babies/">https://www.england.nhs.uk/mat-transformation/saving-babies/</a>. (accessed 1<sup>st</sup> February 2019).
- 10. Royal College of Obstetricians and Gynaecologists. Each Baby Counts. <a href="https://www.rcog.org.uk/eachbabycounts">https://www.rcog.org.uk/eachbabycounts</a>. (accessed 1st February2019).
- 11. National Perinatal Epidemiology Unit. Perinatal Mortality Review Tool. <a href="https://www.npeu.ox.ac.uk/pmrt">https://www.npeu.ox.ac.uk/pmrt</a>. (accessed 1st February 2019).
- 12. Acheson D. Independent inquiry into inequalities in health report. The Stationery Office: London Acheson 1998.

- 13. Marmot M. Fair society, healthy lives: The Marmot Review: strategic review of health inequalities in England post-2010. Institute of Health Equity; 2010.
- 14. Public Health England. Reducing health inequalities: system, scale and sustainability. London 2017. <a href="https://assets.publishing.service.gov.uk/government/uploads/system/uploads/attachment\_data/file/731682/Reducing\_health\_inequalities\_system\_scale\_and\_sustainability.pdf">https://assets.publishing.service.gov.uk/government/uploads/system/uploads/attachment\_data/file/731682/Reducing\_health\_inequalities\_system\_scale\_and\_sustainability.pdf</a>. (accessed 25th June 2019).
- 15. Dahlgren G, Whitehead M. Policies and strategies to promote social equity in health. Stockholm: Institute for future studies. 1991.
- 16. Greenhalgh T, Potts HW, Wong G, et al. Tensions and paradoxes in electronic patient record research: A systematic literature review using the meta-narrative method. *Milbank Quarterly* 2009:87(4):729-88.
- 17. Greenhalgh T, Robert G, Macfarlane F, et al. Storylines of research in diffusion of innovation: a meta-narrative approach to systematic review. *Soc Sci Med* 2005 Jul;61(2):417-30.
- 18. Greenhalgh T, Robert G, Macfarlane F, et al Diffusion of innovations in service organisations: systematic literature review and recommendations for future research. *Milbank Quarterly* 2004;82(4):581-629.
- 19. Wong G, Greenhalgh T, Westhorpe G et al RAMESES publication standards: meta-narrative reviews. *BMC Medicine* 2013;11(20). https://doi.org/10.1186/1741-7015-11-20
- 20. Moher D, Liberati A, Tetzlaff J, et al. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *PLOS Med* 2009:6:e1000097.
- 21. Kingdon C, Roberts D, Turner M, et al Inequalities and stillbirth: a metanarrative review. PROSPERO 2017 CRD42017079228.
- 22. Kuhn, T. The structure of scientific revolutions. University of Chicago University Press, Chicago, 1962.
- 23. Betrán AP, Temmerman M, Kingdon C, et al. Interventions to reduce unnecessary caesarean sections in healthy women and babies. *Lancet* 2018; 392(10155):1358-1368. doi:https://doi.org/10.1016/S0140-6736(18)31927-5.
- 24. Critical Appraisal Skills Programme (CASP). CASP Checklists. 2013. <a href="http://www.casp-uk.net/#!casp-tools-checklists/c18f8">http://www.casp-uk.net/#!casp-tools-checklists/c18f8</a>. (accessed 25<sup>th</sup> June 2019).
- 25. Walsh D, Downe S. Appraising the quality of qualitative research. *Midwifery*. 2006;22:108-119.
- 26. Baird, D. The Influence of Social and Economic Factors on Stillbirths and Neonatal Deaths. *Journal of Obstetrics and Gynaecology*. 1945;52(4): 339-365.
- 27. Morris JN, Heady JA. Social and biological factors in infant mortality: Objects and methods. *Lancet* 1955; 268(6859):343-349.

- 28. Heady, JA, Daly C, Morris JN. Social and Biological Factors in Infant Mortality. II. Variation of Mortality with mother's age and parity. *Lancet* 1955; 268(6860):395-7.
- 29. Daly C, Heady JA, Morris JN. Social and biological factors in infant mortality: III. The effect of mother's age and parity on social-class differences in infant mortality. *Lancet* 1955; 265 (6861):445-448.
- 30. Heady JA, Stevens CF, Daly C, et al. Social and Biological Factors in Infant Mortality. The independent effects of social class, region, the mother's age and her parity. *Lancet* 1955; 265(6862):499-503.
- 31. Illsley R. Social class selection and class differences in relation to stillbirths and infant deaths. *British Medical Journal* 1955; 24;2(4955):1520-1524
- 32. Kincaid JC. Social Pathology of Foetal and Infant Loss. *British Medical Journal* 1965;17;1(5441):1057–1060.
- 33. Clarke M, Clayton DG, Mason ES, et al. Asian mothers' risk factors for perinatal death: the same or different? A 10 year review of Leicestershire perinatal deaths. *British Medical Journal* 1988;297(6645):384-87.
- 34. Guildea ZES, Fone DL, Dunstan FDJ, et al. Social deprivation and the causes of stillbirth and infant mortality. *Archives of Disease in Childhood* 2001;84 (4):307-310. doi 10.1136/adc.84.4.307.
- 35. Flenady V, Koopmans L, Middleton P, et al. Major risk factors for stillbirth in high-income countries: a systematic review and meta-analysis. *Lancet* 2011; 377 (9774):1331–40.
- 36. Seaton SE, Field DJ, Draper ES, et al. Socioeconomic inequalities in the rate of stillbirths by cause: a population-based study. BMJ Open 2012;2(3). doi: 10.1136/bmjopen-2012-001100.
- 37. Zeitlin, J Mortensen, L. Prunet, C, et al. Socio-economic inequalities in stillbirth rates in Europe: measuring the gap using routine data from the Euro-Peristat Project. *BMC Pregnancy & Childbirth* 2016;16:15. https://doi.org/10.1186/s12884-016-0804-4
- 38. Penn N, Oteng-Ntim E, Oakley LL, et al. Ethnic variation in stillbirth risk and the role of maternal obesity: analysis of routine data from a London maternity unit. *BMC Pregnancy and Childbirth* 2014;14:404-410.
- 39. Chitty LS, Winter RM. Perinatal mortality in different ethnic groups. No evidence for increasing inequality *Archives of Disease in Childhood* 1989; 64:1036-1041.
- 40. Gray R, Bonellie SR, Chalmers J, et al. Contribution of smoking during pregnancy to inequalities in stillbirth and infant death in Scotland 1994-2003: retrospective population based study using hospital maternity records. *British Medical Journal* 2009;339:b3754.
- 41. Rush D, Cassano P. Relationship of cigarette smoking and social class to birth weight and perinatal mortality among all births in Britain, 5-11 April 1970. *J Epidemiol Community Health* 1983 Dec;37(4):249-55.

- 42. Weightman AL, Morgan HE, Shepherd MA, et al. Social inequality and infant health in the UK: systematic review and meta-analyses. *BMJ Open* 2012;2:e000964. doi:10.1136/bmjopen-2012-000964.
- 43. Wood AM, Pasupathy D, Pell JP, et al. Trends in socioeconomic inequalities in risk of sudden infant death syndrome, other causes of infant mortality, and stillbirth in Scotland: population based study. *British Medical Journal* 2012;344:e1552.
- 44. Bambang S, SpencerNJ, Logan S, et al. Cause-specific perinatal death rates, birth weight and deprivation in the West Midlands, 1991–1993. *Child: Care, Health and Development* 2000;26(1):73-82. doi:10.1046/j.1365-2214.2000.00152.
- 45. Khalil A, Rezende J, Akolekar R, et al. Maternal racial origin and adverse pregnancy outcome: a cohort study. *Ultrasound in Obstetrics & Gynecology* 2013;41(3):278-285. doi:10.1002/uog.12313.
- 46. Sutan R, Campbell D, Prescott GJ, et al. The risk factors for unexplained antepartum stillbirths in Scotland, 1994-2003. *Journal of Perinatology* 2010;30: 311–318.
- 47. Murrells TJ, Catford JC, Smith TM et al. The use of logit models to investigate social and biological factors in infant mortality. II: Stillbirths. *Statistics in medicine* 1985;4(2):189-200.
- 48. Neasham D, Dolk H, Vrijheid M et al. Stillbirth and neonatal mortality due to congenital anomalies: temporal trends and variation by small area deprivation scores in England and Wales, 1986-96. *Paediatr Perinat Epidemiol* 2001; 15(4):364-73.
- 49. Antonovsky A, Bernstein J. Social class and infant mortality. *Soc Sci Med* 1977; 11:453-470.
- 50. Macintyre S. The patterning of health by social position in contemporary Britain: Directions for sociological research. *Soc Sci Med* 1986;23:393-415.
- 51. Bloor M, Samphier M, Prior L. Artefact explanations of inequalities in health: an assessment of the evidence. *Sociology of Health & Illness* 1987;9:231-264.
- 52. Andrews A, Jewson N. Ethnicity and infant deaths: the implications of recent statistical evidence for materialist explanations. *Sociology of Health & Illness* 1993;15:137-156.
- 53. Kingdon C. Society. In Kierans C, Bell K, Kingdon C. Social and Cultural Perspectives on Health, Technology and Medicine. Abingdon, Routledge 2016.
- 54. Black D, Morris JN, Smith C, Townsend P. The Black Report. Townsend P, Davidson N. (Eds) 1992 Edn. London, Penguin books 1980.
- 55. Whitehead M. The Health Divide. Whitehead (Ed) 1992 Edn.; London, Penguin books 1988.
- 56. Garcia R, Ali N, Papadopoulos C, et al Specific antenatal interventions for Black, Asian and Minority Ethnic (BAME) pregnant women at high risk of poor birth outcomes in the United Kingdom: a scoping review. *BMC Pregnancy and Childbirth* 2015;15:226. doi:10.1186/s12884-015-0657-2.

- 57. Flenady V, Middleton P, Smith GC et al. Stillbirths: the way forward in high-income countries. *Lancet* 2011;377 (9778):703-1717.
- 58. Flenady V, Wojcieszek AM, Middleton P, et al. Stillbirths: recall to action in high-income countries. Lancet Series. *Lancet* 2016;387(10019):691–702.
- 59. Pickton MJ. A Socio-Spatial Analysis of Perinatal Mortality in Greater Leicester [PhD doctoral thesis]. University of Leicester 1987. <a href="http://hdl.handle.net/2381/9240">http://hdl.handle.net/2381/9240</a>.
- 60. Sloggett A, Joshi H. Deprivation indicators as predictors of life events 1981-1992 based on the UK ONS Longitudinal Study. Journal of Epidemiology & Community Health 1998;52:228-233.
- 61. Joyce R, Webb R, Peacock J. Social class and census-based deprivation scores: which is the best predictor of stillbirth rates? *Paediatric and Perinatal Epidemiology* 1999;13: 269-277.
- 62. Dummer TJ, Dickinson HO, Pearce MS, et al. Stillbirth risk with social class and deprivation: No evidence for increasing inequality. *Journal of Clinical Epidemiology* 2000; 53(2):147–155.
- 63. Dickinson HO, Hutton JL, Greaves LH,et al Deprivation and stillbirth risk in rural and urban areas. *Paediatric and Perinatal Epidemiology* 2002;16(3): 249–254.
- 64. Pattenden S, Casson K, Cook S et al. Geographical variation in infant mortality, stillbirth and low birth weight in Northern Ireland, 1992–2002. *J Epidemiol Community Health* 2011;65(12):1159-65.
- 65. Rutter DR, Quine L. Inequalities in pregnancy outcome: A review of psychosocial and behavioural mediators. *Soc Sci Med* 1990;30(5):553-568.
- 66. Confidential Enquiry into Maternal and Child Health. Perinatal Mortality Surveillance, 2004: England, Wales and Northern Ireland. London: CEMACH; 2006.
- 67. Centre for Maternal and Child Enquiries (CMACE) Perinatal Mortality 2009: United Kingdom. CMACE: London, 2011.
- 68. Draper ES, Kurinczuk JJ, Kenyon S et al. on behalf of MBRRACE-UK. MBRRACE-UK 2015 Perinatal Confidential Enquiry: Term, singleton, normally-formed, antepartum stillbirth. The Infant Mortality and Morbidity Studies, Department of Health Sciences, University of Leicester: Leicester, 2015.
- 69. Manktelow BN, Smith LK, Evans TA, et al. on behalf of the MBRRACE-UK Collaboration. MBRRACE-UK Perinatal Mortality Surveillance Report, UK Perinatal Deaths for Births from January to December 2013 - Supplementary Report: UK Trusts and Health Boards. The Infant Mortality and Morbidity Studies, Department of Health Sciences, University of Leicester: Leicester, 2015.
- 70. Manktelow BN, Smith LK, Seaton SE, et al. on behalf of the MBRRACE-UK Collaboration. MBRRACE-UK Perinatal Mortality Surveillance Report, UK Perinatal Deaths for Births from January to December 2014. The Infant

- Mortality and Morbidity Studies, Department of Health Sciences, University of Leicester: Leicester, 2016.
- 71. Gardosi J, Francis A. Key Health Data for the West Midlands. Chapter Five: Perinatal Mortality and Social Deprivation: West Midlands Trends 1998-2003. West Midlands Perinatal Institute 2005. <a href="http://medweb4.bham.ac.uk/websites/key\_health\_data/2004/ch\_05.htm">http://medweb4.bham.ac.uk/websites/key\_health\_data/2004/ch\_05.htm</a>. (accessed 25<sup>th</sup> June 2019).
- 72. Gardosi J, Beamish N, Francis A, et al. Stillbirth and infant mortality, West Midlands 1997-2005: Trends, Factors, Inequalities. West Midlands Perinatal Institute 2007.
  <a href="http://medweb4.bham.ac.uk/websites/key\_health\_data/2006/pdf\_Files/Additional%20Report%20from%20WMPI.pdf">http://medweb4.bham.ac.uk/websites/key\_health\_data/2006/pdf\_Files/Additional%20Report%20from%20WMPI.pdf</a>. (accessed 25th June 2019).
- 73. Francis A, El Sheikh A, Gardosi J. Key Health Data for the West Midlands. Chapter Thirteen: Stillbirths, Infant Deaths and Social Deprivation, West Midlands 1997-2007/8. West Midlands Perinatal Institute 2009. <a href="http://medweb4.bham.ac.uk/websites/key\_health\_data/2008/pdf\_Files/West%20Midlands%20Key%20Health%20Data%202008-09.pdf">http://medweb4.bham.ac.uk/websites/key\_health\_data/2008/pdf\_Files/West%20Midlands%20Key%20Health%20Data%202008-09.pdf</a>. (accessed 25th June 2019).
- 74. Gardosi J. Perinatal Mortality, Social Deprivation and Community Midwifery. West Midlands Perinatal Institute 2011.

  <a href="http://www.perinatal.nhs.uk/pnm/clinicaloutcomereviews/Report\_on\_perinatal\_mortality\_deprivation\_community\_midwifery\_2008-9.pdf">http://www.perinatal.nhs.uk/pnm/clinicaloutcomereviews/Report\_on\_perinatal\_mortality\_deprivation\_community\_midwifery\_2008-9.pdf</a>. (accessed 28th October 2018).
- 75. Tang A, Whitworth M, Roberts D. The relation between social deprivation and stillbirth causes. *Arch Dis Child Fetal Neonatal Ed* 2008;93(Suppl I):Fa1–Fa14.
- 76. Man J, Hutchinson JC, Heazell AE et al. Stillbirth and intrauterine fetal death: role of routine histopathological placental findings to determine cause of death. *Ultrasound Obstet Gynecol* 2016; 48(5): 579–584. doi: 10.1002/uog.16019.
- 77. Man J, Hutchinson JC, Ashworth M, et al. Stillbirth and intrauterine fetal death: contemporary demographic features of >1000 cases from an urban population. *Ultrasound Obstet Gynecol* 2016; 48(5): 591-595. doi:10.1002/uog.16021.
- 78. Man J, Hutchinson JC, Heazell AE et al. Stillbirth and intrauterine fetal death: factors affecting determination of cause of death at autopsy. *Ultrasound Obstet Gynecol* 2016; 48(5): 566-573. doi: 10.1002/uog.16016.
- 79. Heazell AE, Weir CJ, Stock SJE, et al. Can promoting awareness of fetal movements and focusing interventions reduce fetal mortality? A stepped-wedge cluster randomised trial (AFFIRM). *BMJ Open*. 2017;7(8):e014813.
- 80. Heazell AEP, Li M, Budd JT et al. Association between maternal sleep practices and late stillbirth findings from a stillbirth case-control study. *BJOG* 2018; 125:254–262.

- 81. Siassakos D, Jackson S, Gleeson K, et al. All bereaved parents are entitled to good care after stillbirth: a mixed-methods multicentre study (INSIGHT). *BJOG* 2018;125 (2): 160-170.
- 82. Widdows K, Reid HE, Roberts SA, et al. Saving babies' lives project impact and results evaluation (SPiRE): a mixed methodology study. BMC Pregnancy and Childbirth 2018;18(1):43. doi: 10.1186/s12884-018-1672-x.
- 83. Macintyre S. The Black Report and beyond: What are the issues? *Soc Sci Med* 1997;44(6):723-745.
- 84. Bambra C, Gibson M, Sowden A, et al. Tackling the wider social determinants of health and inequalities: evidence from systematic reviews. *Journal of Epidemiology and Community Health* 2010;64:284-291.
- 85. Smith KE, Eltanani MK. What kinds of policies to reduce health inequalities in the UK do researchers support? *Journal of Public Health* 2015; 37(1):6-17
- 86. Burns H. Health inequalities: why so little progress? *Public Health* 2015;129: 849-853.
- 87. Fingar K, Lob S, Dove M, et al. Reassessing the Association between WIC and Birth Outcomes Using a Fetuses-at-Risk Approach. *Maternal & Child Health Journal* 2017; 21(4):825-835. doi:10.1007/s10995-016-2176-9.
- 88. Lakshmi PV, Virdi NK, Sharma A, et al. Household air pollution and stillbirths in India: analysis of the DLHS-II National Survey. *Environmental Research* 2013; 121:17-22. doi: 10.1016/j.envres.2012.12.004.
- 89. WHO. Equity, social determinants and public health programmes. World Health Organization, Geneva, Switzerland 2010. <a href="https://www.who.int/social\_determinants/publications/9789241563970/en/">https://www.who.int/social\_determinants/publications/9789241563970/en/</a>. (accessed 1st February 2019).
- 90. National Perinatal Epidemiology Unit. Inequalities in Infant Mortality Work Programme. https://www.npeu.ox.ac.uk/infant-mortality (accessed 24<sup>th</sup> July 2019)
- 91. Finlayson K, Downe S. Why Do Women Not Use Antenatal Services in Lowand Middle-Income Countries? A Meta-Synthesis of Qualitative Studies. *PLoS Med* 10(1): e1001373. doi:10.1371/journal.pmed.1001373.
- 92. Downe S, Finlayson K, Walsh D, et al. 'Weighing up and balancing out': a meta-synthesis of barriers to antenatal care for marginalised women in high-income countries. *BJOG* 2009;116:518–529.
- 93. Popay J, Bennett S, Thomas C, et al. Beyond 'beer, fags, egg and chips'? Exploring lay understandings of social inequalities in health. *Sociol Health Illn* 2003;25(1):1-23.
- 94. Robinson J, Holdsworth C. They don't live in my house every day: How understanding lives can aid understandings of smoking. *Contemporary Drug Problems* 2013;40: 47-70.
- 95. Passey ME, Longman JM, Robinson J, et al (2016). Smoke-free homes: what are the barriers, motivators and enablers? A qualitative systematic review and thematic synthesis. *BMJ Open* 2016; 6(3). doi:10.1136/bmjopen-2015-010260.
- 96. Heazell AEP, Whitworth MK, Whitcombe SW, et al. Research priorities for stillbirth: process overview and results from UK Stillbirth Priority Setting

Partnership. *Ultrasound in Obstetrics and Gynaecology* 2015; 46(6):641-647. https://doi.org/10.1002/uog.15738.

Figure 1: RAMESES-PRISMA Diagram

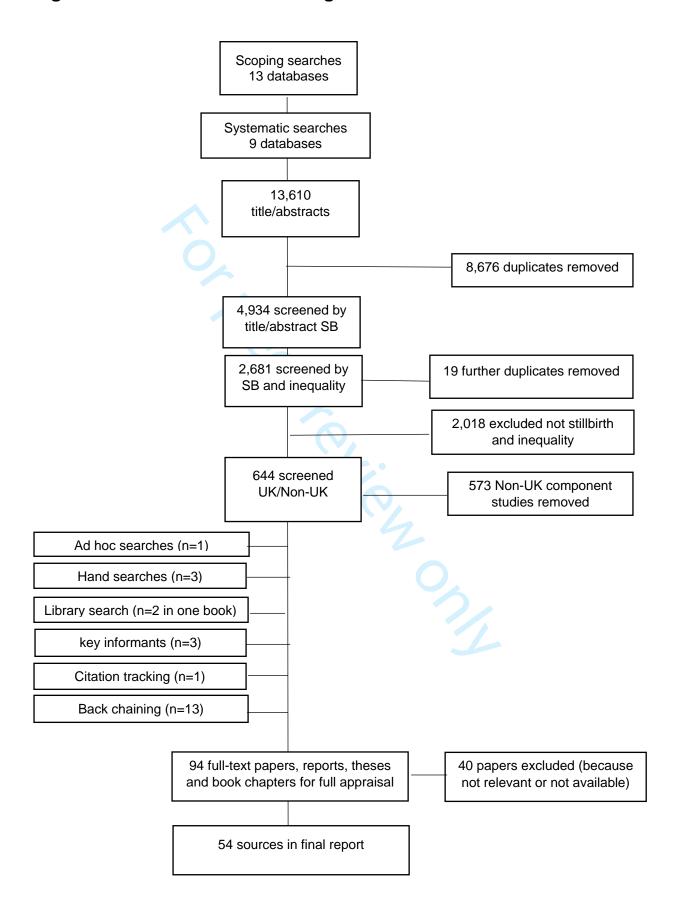


Figure 2: Timeline of included studies by research tradition and the stillbirth rate in England and Wales 1945-2017

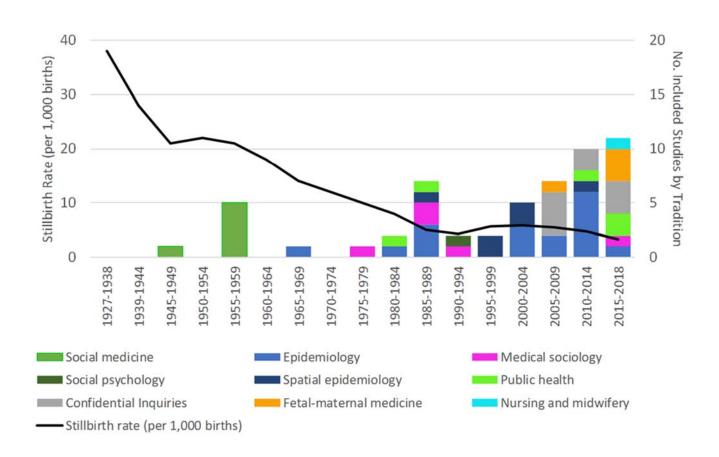
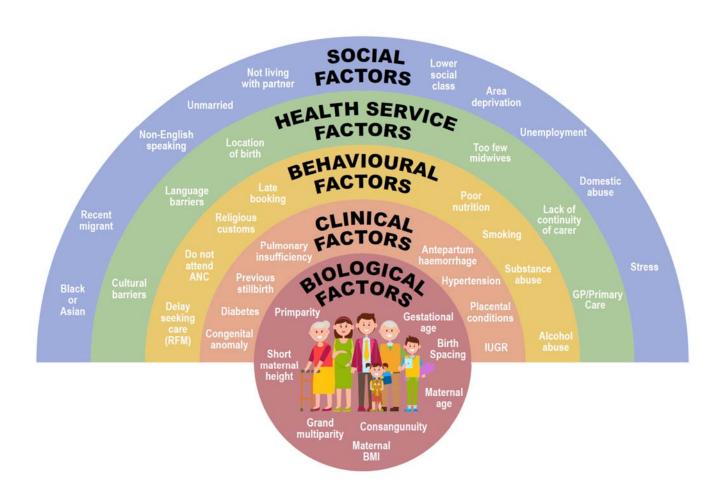


Figure 3: Factors associated with inequalities and stillbirth



Key: ANC (Antenatal Care); GP (General Practice), RFM (Reduced Fetal Movements); IUGR (Intra-uterine growth retardation also known as FGR – fetal growth restriction); BMI (Body mass index)

#### **Supplementary File 1: RAMESES Checklist**

TITLE Inequalities	and stillbirth in the UK: A meta-narrative review	PAGE/LINE Number
1	In the title, identify the document as a meta-narrative review or synthesis	1
ABSTRACT	0/	
2	While acknowledging publication requirements and house style, abstracts should ideally contain brief details of: the study's background, review question or objectives; search strategy; methods of selection, appraisal, analysis and synthesis of sources; main results; and implications for practice.	2
INTRODUCTION	CV:	
3 Rationale for review	Explain why the review is needed and what it is likely to contribute to existing understanding of the topic area.	4-5
4 Objectives and focus of review	State the objective(s) of the review and/or the review question(s). Define and provide a rationale for the focus of the review.	5
METHODS		
5 Changes in the review process	Any changes made to the review process that was initially planned should be briefly described and justified.	6-7

TITLE Inequalities	and stillbirth in the UK: A meta-narrative review	PAGE/LINE Number
6 Rationale for using meta-narrative review	Explain why meta-narrative review was considered the most appropriate method to use.	6
7 Evidence of adherence to guiding principles of meta- narrative review	Where appropriate show how each of the six guiding principles (pragmatism, pluralism, historicity, contestation, reflexivity and peer review) have been followed.	6
8 Scoping the literature	Describe and justify the initial process of exploratory scoping of literature.	6
9 Searching processes	While considering specific requirements of the journal or other publication outlet, state and provide a rationale for how the iterative searching was done. Provide details on all the sources accessed for information in the review. Where searching in electronic databases has taken place, the details should include (for example) name of database, search terms, dates of coverage and date last searched. If individuals familiar with the relevant literature and/or topic area were contacted, indicate how they were identified and selected.	6-7
10 Selection and appraisal of documents	Explain how judgements were made about including and excluding data from documents, and justify these.	6-7
11 Data extraction	Describe and explain which data or information were extracted from the included documents and justify this selection.	7
12 Analysis and synthesis processes	Describe the analysis and synthesis processes in detail. This section should include information on the constructs analysed and describe the analytic process.	8

TITLE Inequalities	and stillbirth in the UK: A meta-narrative review	PAGE/LINE Number
RESULTS		
13 Document flow diagram	Provide details on the number of documents assessed for eligibility and included in the review with reasons for exclusion at each stage as well as an indication of their source of origin (for example, from searching databases, reference lists and so on). You may consider using the example templates (which are likely to need modification to suit the data) that are provided.	9
14 Document characteristics	Provide information on the characteristics of the documents included in the review.	9
15 Main findings	Present the key findings with a specific focus on theory building and testing.	9-14
DISCUSSION	- Ch-	
<b>16</b> Summary of findings	Summarise the main findings, taking into account the review's objective(s), research question(s), focus and intended audience(s).	15
17 Strengths, limitations and future research	Discuss both the strengths of the review and its limitations. These should include (but need not be restricted to) (a) consideration of all the steps in the review process and (b) comment on the overall strength of evidence supporting the explanatory insights which emerged.  The limitations identified may point to areas where further work is needed.	15-16
<b>18</b> Comparison with existing literature	Where applicable, compare and contrast the review's findings with the existing literature (for example, other reviews) on the same topic.	16

TITLE Inequalities and stillbirth in the UK: A meta-narrative review

19 Conclusion and Recommendations

List the main implications of the findings and place these in the context of other relevant literature. If appropriate, offer recommendations for policy and practice.

16-17

20 Funding

Provide details of funding source (if any) for the review, the role played by the funder (if any) and any conflicts of interests of the reviewers.

Theer review only

BMJ Open

Page 34 of 48

#### **PROSPERO**

#### International prospective register of systematic reviews



#### Inequalities and stillbirth: a meta-narrative review

Carol Kingdon, Devender Roberts, Mark Turner, Claire Storey, Nicola Crossland, Kenneth Finlayson, Soo Downe

#### Citation

Carol Kingdon, Devender Roberts, Mark Turner, Claire Storey, Nicola Crossland, Kenneth Finlayson, Soo Downe. Inequalities and stillbirth: a meta-narrative review. PROSPERO 2017 CRD42017079228 Available from:

http://www.crd.york.ac.uk/PROSPERO/display\_record.php?ID=CRD42017079228

#### Review question

The aim of this review is to undertake an inter-disciplinary evidence synthesis (using a meta-narrative approach) to understand how structural factors (health system, living in poverty), lifestyle factors (smoking, obesity), and bio-clinical factors (maternal infection, non-communicable disease, fetal growth restriction) intersect to increase stillbirth risk to improve interventions to manage at-risk pregnancies.

The broad research question is: What is the relationship between inequality and stillbirth, how has this been studied, and with what effects?

The measurable objectives are:

- 1. To review the current body of knowledge of the relationship between inequalities and stillbirth across the natural and the social sciences;
- 2. To provide new insights into the interplay of biological, clinical, cultural and socioeconomic factors in increased stillbirth risk;
- 3. To explore the impact of interventions on inequalities;
- 4. To provide a narrative summary of this research for stakeholders tasked with reducing preventable stillbirth.

#### Searches

The following databases will be used to identify eligible studies for inclusion as part of the scoping searches:

MEDLINE® (Life sciences, medicine and biomedicine)

Embase® (Biomedical)

CINAHL® (Nursing and Allied Health Professionals)

PsycINFO® (Psychology and the behavioural and social sciences)

AJOL® (African Journals on-line)

Global Index Medicus

Popline (includes LILACS [Latin America and the Caribbean])

Historical abstracts

**Humanities International Complete** 

Race Relations Abstract

SocIndex

Lexis Library

Lexis Nexus

#### Types of study to be included

This is a meta-narrative review with no restrictions on type of study to be included.

#### Condition or domain being studied

Stillbirth is a profound human tragedy. The experience of stillbirth involves physical implications for the mother, together with intense grief and lasting psychological trauma for both parents and wider family.

#### **PROSPERO**



#### International prospective register of systematic reviews

Studies have shown that stillbirth is associated with anxiety, depression and post-traumatic stress disorder in mothers, couples, siblings and grandparents.

The UK has the third highest rate of stillbirth among the 35 high-income countries of the world. Against a background of political, public and professional interest in halving the UK's stillbirth rate by 2030, and significant inter-disciplinary research effort to prevent or to improve maternity care services, there is longstanding evidence, across high-income countries, that the risk of stillbirth remains highest for the poorest families. Almost half (46%) of stillbirths remain of unknown cause, and inequalities in stillbirth risk demand more attention.

What is missing from current research agendas is an overarching synthesis of clinical and social science evidence to clarify the range of individual, social and biological mechanisms of increased stillbirth risk, the intersections between these mechanisms, and strategies to tackle them. There is also an urgent need to identify where interventions may decrease stillbirth risk for some, but may inadvertently increase inequalities for others. This meta-narrative review seeks to fill this gap in response to international interest in reviewing evidence from all relevant research traditions to address the unanswered questions surrounding stillbirth risk, and an NHS England Strategic Clinical Network need for this evidence to inform interventions to reduce the risk amongst women with complex social needs from vulnerable groups.

#### Participants/population

The population of interest for the purposes of our inclusion criteria is: Woman or Women or Mother or Mothers or Father or fathers or Family or Families or Husband or partner or partners or Parent or parents

Inclusion criteria

Any study design investigating stillbirth and inequality Any study in high-,middle- or low-income settings

Exclusion criteria

Any study published in non-English language Any study of pregnancy loss before 20 weeks Any study of perinatal loss in the neonatal period

Any study involving participants who had assisted conception (as identified by authors)

#### Intervention(s), exposure(s)

if heap explicitions are so discrimination or low income or poverty, or violence or abuse, or consanguinity, or ethnicity, or discrimination, or race or racism or racial, or migrant, or migration, or maternal age, or adolescence, or nutrition or obesity, or overweight or underweight, or smoking, or alcohol or drug or substance or chemical, or nonattendance, or neighborhood.

The interventions to be reviewed are:

growth chart or biomarkers or movement, or count, or support, or continuity of care or caseload.

The outcomes to be reviewed are:

stillbirth or perinatal death or pregnancy loss or miscarriage or fetal death or foetal death or feticide or foeticide or intrauterine death after 20 weeks pregnancy gestation.

#### NHS National Institute for Health Research

## PROSPERO International prospective register of systematic reviews

#### Comparator(s)/control

Not applicable.

#### Primary outcome(s)

Stillbirth incidence

Timing and effect measures
Effect measure exposure to inequality

#### Secondary outcome(s)

Not applicable

#### Data extraction (selection and coding)

Key results will be extracted, collated and grouped. Modified data extraction forms will be developed from existing published meta-narrative reviews and other types of review the team have been involved with in relation to stillbirth. Included studies will then be tabulated by tradition.

#### Risk of bias (quality) assessment

All articles that meet the inclusion criteria will be independently assessed by two researchers (NC,KF) and the principal investigator (CK) to minimize bias. Quality appraisal of quantitative studies will be undertaken using the appropriate checklists from the Critical Appraisal Skills Programme (CASP) Toolkit. Checklists formed from this process will be used to grade papers into categories A, B, C or D, with group A representing papers of the highest quality. Papers will be assigned to groups according to how many criteria fulfilled (i.e. Category A papers not fulfilled 0-1 of the marked criteria; Category D papers not fulfilled ?6 criteria). Qualitative appraisal will be undertaken according to the checklist described by Walsh and Downe and articles graded A – D in accordance with Downe and Simpson. A grades will be allocated to papers with no or few flaws where the study credibility, transferability, dependability, and confirmability is high; B, some flaws, unlikely to affect the credibility, transferability, dependability, and/or confirmability of the study; C, some flaws which may affect the credibility, transferability, transferability, dependability, and/or confirmability of the study; D, significant flaws which are very likely to affect the credibility, transferability, dependability, dependability. Any differences in appraisals will be discussed with the wider research team until a final decision is reached.

#### Strategy for data synthesis

The defining feature of meta-narrative reviews is their illumination of a complex topic area from multiple angles in a unified narrative synthesis. The synthesis stage involves summarizing each tradition in coherent individual accounts (including elements of data aggregation) and then comparing and contrasting the resultant meta-narratives to highlight similarities and differences. This process of contestation between the disciplines/traditions leads to higher order constructs and conclusions where recommendations can be made (i.e. in circumstances such as X, don't forget to think about y).

#### Quantitative studies

Quantitative meta-analysis will be used if the included studies are sufficiently homogeneous. In the event included quantitative studies are too heterogeneous they will be summarized narratively, separate from the qualitative evidence synthesis in the first instance, and then incorporated alongside into an overall typology of inequality and stillbirth.

#### Qualitative studies

A modified meta-ethnography approach will be used for qualitative evidence synthesis comprising 4 stages:1) Familiarization and quality appraisal; 2) Data extraction (direct participant quotations and author's thematic interpretation); 3)Coding into initial concepts; 4) Interpretative synthesis into emergent and final

## National Institute for Health Research

## PROSPERO

#### International prospective register of systematic reviews

themes to be presented in a typology.

Analysis of subgroups or subsets Not applicable

Contact details for further information

Carol Kingdon

ckingdon@uclan.ac.uk

Organisational affiliation of the review

University of Central Lancashire

Review team members and their organisational affiliations

Dr Carol Kingdon. University of Central Lancashire

Dr Devender Roberts. Liverpool Women's NHS Foundation Trust

Dr Mark Turner. University of Liverpool

Ms Claire Storey. International Stillbirth Alliance

Dr Nicola Crossland. University of Central Lancashire

Mr Kenneth Finlayson. University of Central Lancashire

Professor Soo Downe. University of Central Lancashire

Anticipated or actual start date

04 October 2017

Anticipated completion date

30 March 2018

Funding sources/sponsors

Stillbirth and Neonatal Death Society (SANDS) (Ref:RF510)

Conflicts of interest

None known

Language

English

Country

**England** 

#### PROSPERO

This information has been provided by the named contact for this review. CRD has accepted this information in good faith and registered the review in PROSPERO. CRD bears no responsibility or liability for the content of this registration record, any associated files or external websites.

## **Supplementary File 3: Example exploratory searches**

Databases	No. hits for "stillbirth"	Types of articles	Relevance to review questions
Art and Architecture source	10	poetry, feminism/women's studies, historical	Articles about impact and experiences of stillbirth rather than risk factors.
Criminal Justice Abstracts with Full Text	73	social work, women's studies, first person accounts/autoethnography	Articles about impact and experiences of stillbirth rather than risk factors.
Cochrane Library	535	Cochrane Reviews (n=83) Trials (423)	Two Cohrane reviews were tangenitaly relevant to research questions (interventions to reduce smoking in pregnancy, interventions to reduce domestic violence in pregnancy).
Humanities international complete	120	Family studies, images, religious aspects, first person accounts, responses to stillbirth, archaeology (burials), how stillbirth is conceptualised, abortion and morality/ethics	Articles about impact and experiences of stillbirth.
Race relations abstracts	6	differential medical risks by ethnicity, a couple of articles relating to social risk factors by ethnicity	Social inequalities as a risk factors
Religion and Philosophy Collection	74	grief and loss, psychosocial identity, religious aspects, social responses, attachment theory, abortion,	Articles predominantly about abortion; not relevant to research questions
SocIndex	482	psychosocial identity, psychosocial impacts, mother and family experiences, social risk factors, grief, coping,	Social inequality risk factors. Consequences, impact and coping.
Lexis Library (legal)		Professional misconduct cases. Judgements about cases where a medical intervention/decision has led to stillbirth. Employment policies. Legal frameworks about healthcare delivery. Medical law.	Tangential to research questions.
JISC Historical Texts	0 for stillbirth, 308 for stillborn	Literature (66). Medical texts (15) - midwifery manuals, descriptive works on causes of death. 1600s, 1700s.	Tangential to research questions.
Historial abstracts (via Ebsco)	56 for stillbirth.	Rates of stillbirth at different historical times and places, about perceptions of stillbirth. Lots of hits come from journal Population Studies.	Modern history – potential for understanding how stillbirth has been conceptualised.

## **Supplementary File 4: Example Systematic Search**

## **Strategy terms**

**Population**: Woman or Women or Mother or Mothers or Father or fathers or Family or Families or Husband or partner or partners or Parent or parents

#### **Exposure**

<b>Exposure type</b>	Exposure search terms
economic	Inequalit* or Depriv* or 'low income' or Poverty or socioeconomic or 'social class'
violence & abuse	Violence or Abus*
substance use	Smok* or Alcohol or Drug or Substance or Chemical
ethnicity	ethnic* or Race or Racism or Racial* or Migrant or Migration
nutrition	nutrition or undernutrition or malnutrition
age	'maternal age' or adolescen*
obesity and weight	Obes* or overweight or underweight
space and place	neighborhood or neighbourhood or residence
appointments	"Appointments and Schedules"/ or nonattendance.mp
culture	Sociocultural or cultur*
consanguinity	Consanguin*

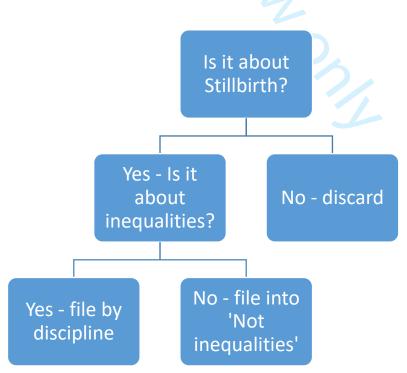
**Intervention**: growth chart' or biomarkers or movement or count or support or 'continuity of care' or caseload

**Outcome**: Stillbirth or stillborn or 'perinatal death' or 'pregnancy loss' or miscarriage or 'fetal death' or 'foetal death' or feticide or foeticide or 'intrauterine death'

**IVF-related terms** (ivf or fertil\* or infertil\* or 'assisted fertility' or inseminat\* or iui or 'embryo transfer')

## **Supplementary File 5: Screening Tool**

Answer the following questions in sequence:	Yes	No	Can't tell from abstract
1. Is it about stillbirth (defined as the death of a baby after 20 weeks in-utero until immediately before birth; excluding pregnancy loss < 20 weeks gestation and excluding neonatal deaths)?	Assess by Q2	Discard	If abstract doesn't define miscarriage, then discard.
If yes, then:			
<ul> <li>2. Is it about inequality and stillbirth?</li> <li>Working definition of inequality</li> <li>Inequality (?how circumstances of daily living disadvantage you, or structural)</li> <li>Socio-economic (as defined by authors so not necessarily NS-SEC), living in poverty, low income</li> <li>Deprivation (ditto – as defined by author - might not be IMD), neighbourhood, traveller, immigrant or refugee</li> <li>Minority ethnicity, religion, disability, young maternal age – member of marginalised group</li> <li>Risk factors (obesity, smoking, substance use, abuse etc) - may be linked to inequality but not enough on their own to include</li> </ul>	If yes, file by discipline	If no, then file as "Not about inequalities and stillbirth"	
If yes, then:			
What discipline is it from?			
3. Is it seminal?			



Page 42 of 48

#### **Supplementary file 6: Characteristics of included sources**

Author and year	Aim and objectives	Design	Context	Population	Stillbirth definition	Stillbirth :: classification (antepartum, intrapartum)	Type of inequality	Precision of inequality
Baird 1945 [26]	To examine the influence of social and economic factors on stillbirths and neonatal deaths	Descriptive	Scotland, compared with England and Wales	All maternities 1938- 1944	Undefined	Both er 2019.	Socio-economic (individual)	Registrar Generals Scale of occupational class
Morris 1955 [27]	To describe the background and design of inquiry into stillbirths and infant deaths in England and Wales in 1949 and 1950	Review	England and Wales	Stillbirths and Infant deaths	Undefined	Both Down	Socio-economic (individual)	Registrar Generals Scale of occupational class
Heady 1955 [28]	To describe the main features of variation of the stillbirth rate, and the neonatal and post-neonatal mortality rates, with two biological factors (age of mother and parity)	Descriptive	England and Wales	Births in 1949	Single, legitimate, stillbirth after the 28th week of pregnancy	Both Both Both	Socio-economic (individual)	Registrar Generals Scale of occupational class
Daly 1955 [29]	To consider the extent of such "biological" differences between the social classes, and the effect of these differences on the stillbirth and infant mortality rates of the social classes	Descriptive	England and Wales	Legitimate, livebirths and stillbirths born in 1949	Single, legitimate, stillbirth after the 28th week of pregnancy	Both Om http	Socio-economic (individual and place of residence)	Registrar Generals Scale of occupational class
Heady 1955 [30]	To investigate the "independence" of mother's age and her parity in relation to social class and region	Descriptive	England and Wales	Single, legitimate livebirths and stillbirths born in 1949	Single, legitimate, stillbirth after the 28th week of pregnancy	Both //bmjop	Socio-economic (individual)	Registrar Generals Scale of occupational class
Illsley 1955 [31]	To examine the processes of movement between social classes and the effects on stillbirths and infant death statistics	Descriptive	Aberdeen, Scotland	Married primiparae resident and delivered in Aberdeen between July, 1950, and December, 1954	Undefined	//bmjopen.bmj.com/ on April 23	Socio-economic (individual)	Registrar Generals Scale of occupational class
Kincaid 1965 [32]	To explore changes in UK perinatal mortality rates over the period 1951-1961	Descriptive	Scotland, England & Wales	Birth records from national databases over periods ranging from 1948-1964	Undefined	on April 23	Socio-economic (individual)	Registrar Generals Scale of occupational class
Clarke 1983 [33]	(a) to measure the extent to which established risk factors, such as high parity and low social class, were risk factors in our locality; (b) to measure the extent to which other factors, such as the provision of medical services and demographic changes, might contribute to the risk of perinatal mortality; (c) to estimate the extent to which avoidable factors might be detected in the case histories of the perinatal deaths; and (d) to contribute our findings to the educational and planning processes of the local health authorities	Case-control	Leicestershire	All perinatal deaths In Leicestershire 1976- 1985	Undefined	8 Both Both Both Both Both Both Both Both	Ethnicity	Broad categories (Asian or European)
Guildea 2001 [34]	To investigate the relation between social deprivation and causes of stillbirth and infant mortality	Retrospective cohort	Wales	All births to women resident in Wales 1993–98	Late fetal deaths after 24 weeks gestation [stillbirth and neonatal death combined]	Both dected by co	Socio-economic (area)	Townsend social and material deprivation score (unemployment, car ownership, owner occupation and

			mjopen-2019-029 <mark>6</mark> 72						
							-029672		overcrowding 1991 census)
Flenady 2011 [35]	To clearly identify important risk factors for stillbirth in high income countries	Systematic review and meta- analysis	High-income countries (including UK)	Studies from HICs	Death of a baby 20 weeks' gestation or more, or birthweight of at least 400 g	Both	on 12 Septem	ethnic origin, socioeconomic status, education level, young maternal age	Different in different studies
Seaton 2012 [36]	To assess time trends in socioeconomic inequalities in overall and cause-specific stillbirths in England over an 8-year period to aid understanding of each cause's impact on the deprivation gap and the overall stillbirth rate	Population-based retrospective study	England	All singleton stillbirths born to mothers resident in England between 01/01/2000 and 31/12/ 2007	Losses from the 24th weeks of gestation occurring in singleton infants	Both	September 2019. Downloaded from http://b	Socio-economic (area)	Indices of Multiple Deprivation (IMD) 2004 score (income; employment; health and disability; education, skills and training; barriers to housing; living environment and crime)
Zeitlin 2016 [37]	What is the magnitude of social inequalities in stillbirth rates in European countries?	Retrospective review of data from countries participating in the Euro-Peristat project	Europe (includes UK data)	Stillbirths and live births for the year 2010 from 29 countries	Intrauterine death after 22 weeks gestation or birthweight of 500 grams or more	Both	ed from http://k	Socio-economic (individual)	Country level available data on educational level, and occupational group
Penn 2014 [38]	What is the association between clinical and socio-demographic factors and stillbirth, with a particular focus on ethnicity and obesity?	Cross-sectional	London	London population - (multi-ethnic)	24 completed weeks of pregnancy (with 28 wks (WHO definition) data in a supplementary table	Both	omjopen.bmj	Ethnicity and socio- economic (area)	Ethnicity general categories (i.e. Black, Asian), Area deprivation IMD score
Chitty 1989 [39]	To assess the contribution of lethal congenital malformations to perinatal mortality in different ethnic groups, to investigate the incidence of definite and probable autosomal recessive syndromes, and to estimate the possible effect of consanguinity.	Descriptive	North West Thames Region	All babies born between 1980-1985	Undefined	Both	.com/ on	Ethnicity	Ethnicity - broad categories (European, Pakistani and Indian)
Gray 2009 [40]	To study the effect of area deprivation on rates of stillbirth and infant mortality in Scotland for the 10 year period 1994-2003 and to establish whether smoking during pregnancy contributed to these gradients and, if so, to what extent.	Population based retrospective cohort study	Scotland	Live births and stillbirths 1994 - 2003.	24-44 weeks of pregnancy	Both	April 23, 2	Socio-economic (area)	Deprivation according to Carstairs-Morris Index
Rush 1983 [41]	To re-explore the interrelation of smoking, social class, birth weight, and perinatal mortality in the total population of births in Britain born 5-11 April 1970.	Prospective cohort study	Britain	All livebirths and stillbirths 5th - 11th April, 1970	Undefined	Both	2024 by g	Socio-economic (individual)	Registrar Generals Scale of occupational class
Weightman 2012 [42]	To assess the current evidence for the effects of social disadvantage on birth and infant outcomes for children born in the UK	Systematic review and meta- analysis	UK	UK studies	A baby born after the 24th week of pregnancy who does not show any signs of life	Unclear	gwest. Protected	Socio-economic (individual and area)	Different measures in different studies

			ВМЈ	Open			mjopen-2019-029 <b>6</b> 72		
Wood 2012 [43]	To compare changes in inequalities in sudden infant death syndrome with other causes of infant mortality and stillbirth in Scotland, 1985-2008.	Retrospective cohort study	Scotland	Singleton births of infants with birth weight >500 g born at 28-43 weeks' gestation	An infant born showing no signs of life after 28 weeks gestation	Both	on 12	Socio-economic (area)	Deprivation according to Carstairs Index (car ownership, unemployment, overcrowding, and social class)
Bambang 2000 [44]	To study the relationship between cause-specific perinatal deathrates, material deprivation and birthweight among births in 3 consecutive yearsin the West Midlands Health Region	Retrospective cohort study.	West Midlands Health Region	All live and stillbirths to mothers with addresses in the WMHR in 1991, 1992 and 1993.	Not given	Both	September 2019	Socio-economic (area)	Townsend Deprivation Index
Khalil 2013 [45]	To examine the association between maternal-racial origin and a wide range of adverse pregnancy outcomes after adjustment for confounding factors in obstetric history and maternal characteristics	Retrospective cohort study.	London	Singleton pregnancies with a live fetus at 11 + 0 to13+ 6 weeks	Fetal deaths at or after 24 weeks	Both	. Downloade	Ethnicity	Ethnicity - broad categories (Caucasian, Afro-Caribbean, East Asian, South Asian, Mixed)
Sutan 2010 [46]	To determine the risk factors of unexplained antepartum stillbirth in Scotland from 1994 to 2003 and assess their value as a screening tool	Retrospective cohort study.	Scotland	All stillborn and live births in Scotland from 1994 to 2003	Not given	Antepartur	n <u>on</u> ly	Socio-economic (area) and place (urban/rural)	Urban/Rural classified according to settlement size and remoteness; Deprivation according to Carstairs Morris Index
Murrells 1985 [47]	To examine changes in the data for stillbirth rates between 1949/50 and 1975	Retrospective cohort study	UK	Comparisons between data from 1949/50 and 1975	Not given	Both	mjopen.	Socio-economic (individual)	Registrar Generals Scale of occupational class
Neasham 2001 [48]	To investigate the variation of stillbirth and neonatal mortality due to congenital anomalies in relation to small-area measures of deprivation in a population-based study in England and Wales, 1986–96	Retrospective population based study	England and Wales	All births in England and Wales from 1986- 1996	On or after 24 weeks gestation	Both	bmj.com/	Socio-economic (area)	Carstairs Index
Antonovsky 1977 [49]	To examine the relationship between the components of infant mortality and social class by analysing the data available from infant mortality studies.	Review and commentary using secondary data	Western Europe and the United States	Uses data from infant mortality studies undertaken in Western Europe and the United States	Component of infant mortality - European nations require registration of stillbirths from 28 weeks gestation; most of United States from the 20th week of gestation.	Unclear	rom http://bmjopen.bmj.com/ qn April 23, 2024 by .	Social class (individual)	Inconsistencies in reporting of class across countries discussed and complexity of occupation, and education as measures.
Macintyre 1986 [50]	To place debates (by social epidemiologists and medical sociologists among others attending to inequalities and health) in a broader and perhaps more traditional context: that of interest in observed social regularities in health, illness and death.	Review and commentary using secondary data	Britain	No specific population  - those where social patterning of health has been observed across the life-course	Unclear	Unclear	guest. Protected by c	Social class (occupational), gender, marital status, age, ethnicity, and area of residence	These six variables are t conceptualized as positions on dimensions of social differentiation which may be associated with particular patterns of life chances.

			mjopen-2019-029 <del>6</del> 72						
Bloor 1987 [51]	To examine more thoroughly than was possible for Black and his colleagues, the possible role of an artefactual element in mortality data for explanations concerning health inequalities.	Review and commentary using secondary data	UK	No specific population draws on historical, theoretical and routinely collected statistical data reporting inequalities across the life-course	Unclear	Unclear	-029672 on 12 Septe	Socio-economic	Examines social processes whereby statistics are produced.
Andrews 1993 [52]	First objective, to draw attention to remarkable pattems of diversity and change revealed by recent official statistics for ethnic minority' infant deaths in England & Wales. Secondly, to suggest that these patterns represent a challenge to many orthodox explanations for inequalities in infant health offered by social scientists, not least materialist explanations. Thirdly, to propose that further development and evaluation of explanations is impeded by flaws in some of the indices and categories employed in the construction of data.	Review and commentary using secondary data	England and Wales	Uses OPCS data on ethnic inequalities in infant deaths	Unclear	Unclear	ember 2019. Downloaded	Ethnicity	Argues ethnicity is as much a matter of the way in which boundaries are created and reproduced between groups as the internal contents of ethnic cultures.
Kingdon 2016 [53]	To review the concept of society and discusses stillbirth as a social issue	Stillbirth used as a case study	Includes data from the UK	No specific population (stillbirth used as a case study)	A baby which has issued forth from its mother after the 24th week of pregnancy and which did not breathe or show any other signs of life' [UK definitionothers given]	Both	ded from http://bmjqpen.bmj.com	Socio-economic, ethnic and gender	Multi-dimensional across place, time and culture
Black 1980 [54]	The first Government authorised attempt to explain trends in inequalities in health and to relate these to the policies intended to promote, as well as restore health.	Government Enquiry utilising official statistics	UK	Studies reporting inequalities based on occupational social class across the lifecourse	Not defined	Unclear	open.bmj.con	Socio-economic (individual)	Registrar Generals Scale of occupational class
Whitehead 1988 [55]	To update evidence following on from the Black Report (see above). The original aim of the Health Divide was to draw together, to summarise the wide-ranging new evidence, and to describe what had happened and could happen, in policy development.	Government Enquiry utilising official statistics	UK	Studies reporting inequalities based on occupational social class, and unemployment, income, housing, material and social deprivation in small areas, gender and ethnicity.	Undefined	Unclear	n∕ on April 23, 2024 by g	Socio-economic	Registrar Generals Scale of occupational class
Garcia 2015 [56]	'What specific BAME maternity interventions exist for UK-based BAME women?'	Review and commentary	HICs including UK	Studies from HICs	Authors use an infant born with no life signs after 24 weeks gestation - however included studies in the review may have used different definitions	Unclear	luest. Protected by	Ethnicity (BAME - Black, Asian and Minority Ethnic)	"BAME groups" - Asian, Indian, Pakistani, Bangladeshi, Kashmiri, Black African, Black Caribbean, Arabian, Traveller [defined as a gypsy or Irish traveller],

		mjopen-2019-029 <b>6</b> 72							
									Chinese, Mediterranean.
Flenady 2011 [57]	To present priority areas for stillbirth prevention, and interventions and research to address these priorities in high-income countries.	Review and commentary	HICs including UK	Studies from HICs	Death of a baby 500 g or more, or 22 weeks' gestation or more	Both	on 12 Septe	Socio-economic disadvantage	"Socioeconomic disadvantage" includes maternal education, ethnicity
Flenady 2016 [58]	To summarise the status of stillbirths in HICs and suggest strategies to accelerate momentum in the reduction of stillbirths and to meet parents' needs when their baby is stillborn.	Review and commentary	HICs including UK	Studies from HICs	Late gestation stillbirth" as >28 weeks and "early gestation stillbirth" as occurring prior to 28 weeks gestation	Both	mber 2019. Do	Socio-economic disadvantage	As above
Pickton 1987 [59]	To analyse, describe and explain the distribution of perinatal mortality within an urban area	Case-control with geographical component (statistical and cartographical)	Greater Leicester	All births in Greater Leicester in the study periods	Stillbirth (late fetal death) = death after 28 weeks gestation	Both	ownloaded from ht	Socio-economic (area and individual), ethnicity	Registrar Generals Scale of occupational class, employment housing, education level. Measures of ethnicity very broad (Asian, New Commonwealth).
Slogget 1998 [60]	To determine whether inequality in stillbirth risk between social strata has changed over time	Retrospective cohort study.	England and Wales	A random sample of more than 300 000 people enumerated at the 1981 census	Not defined	Unclear	September 2019. Downloaded from http://bmjopen.bmj.com/ or	Socio-economic (area and individual)	Deprivation was assessed by a "Townsend/Carstairs-like" index of four components — unemployment, car ownership, home ownership, lower employment status according to Registrar General.
Joyce 1999 [61]	To determine whether social class (individual level) or a census- based deprivation score (based on area of residence) is a better predictor of stillbirth rates	Unclear	South Thames (West)	All births 1993-95	Not defined	Unclear	April 23,	Socio-economic (individual and area)	Social class using the Registrar General categories; area deprivation by Townsend score
Dummer 2000 [62]	To determine whether inequality in stillbirth risk between social strata has changed over time	Population based retrospective study	Cumbria compared with England & Wales data	For Cumbria all birth occurring between 1950-93: For E & W all births occurring between 1981-92	Two definitions given reflecting the change from 28 wks to 24wks that occurred in 1992	Both	2024 by guest		
Dickinson 2002 [63]	To investigate whether stillbirth risk was higher, and the effect of deprivation on inequality in stillbirth risk more marked, in rural than in urban areas	Retrospective cohort study	Cumbria	Singleton births between 01/01/1950 – 30/09/1992 to mothers in the area now designated as Cumbria	A baby born dead after at least 28 weeks' gestation	Unclear	: Protected by cop	Socio-economic (individual, area and place)	Social Class - based on fathers occupation Deprivation based on several scores including IMD

mjopen-2019-0

Pattenden 2011 [64]	To analyse adverse pregnancy outcomes, concentrating on estimating the extent of geographical variation from large areas (approximately 400 000 persons population) to small areas (wards, approximately 100 times smaller)	Population based retrospective cohort study	Northern Ireland	Birth records from Northern Ireland over the years 1992-2002	24 weeks and over	029672 on 12 Septem	Socio-economic (area) and place	Deprivation according to The Noble Index (households receiving means-tested benefits Settlement size (rural areas, village, small town, medium town,
Rutter 1990	To review the literature on psychosocial factors in pregnancy	Theoretical	UK	UK	Not defined	Unclear e	Socio-economic	large town, or city).  Registrar Generals
[65]	outcome and to present a model which attempts to integrate the findings theoretically.					2019	(individual)	Scale of occupational class
CEMCH 2006 [66]	To report on perinatal mortality statistics	Confidential Enquiry	England, Wales & Northern Ireland	England, Wales and NI Perinatal deaths during 2004	24th week of pregnancy	. Downloa	Socio-economic (area), ethnicity	Ethnicity approximated Indices of Multiple Deprivation (IMD) scores
CMACE 2011 [67]	To report on perinatal mortality statistics	Confidential Enquiry	UK	UK Perinatal deaths in 2009	A baby delivered without signs of life after 23+6 weeks of pregnancy	Both Both Both	Socio-economic (area), ethnicity, and place (inferred regional and NHS Trust differences)	Ethnicity - broad categories; IMD data based on quintiles of deprivation; Employment status - simple yes/no
Draper 2015 [68]	Perinatal confidential enquiry carried out as part of the MBRRACE-UK programme of work, aim to focus on term, singleton, normally formed, antepartum stillbirths.	Confidential Enquiry	UK	Representative sample of stillbirths from those identified as eligible for review by MBRRACE-UK in April 2014	Term singleton, normally formed antepartum stillbirth (no precise definition given)	Antepartum/bmjopen.bmj.com/ on	Socio-economic (area and individual), ethnicity	Ethnicity - broad categories; IMD data based on quintiles of deprivation; Employment status - broad categories (employed/unemployer student; looking after home/family; permanently sick/disabled)
Manktelow 2015 [69]	To collect, analyse and report national surveillance data to stimulate and evaluate improvements in health care for mothers and babies.	Confidential Enquiry	UK	All births in the UK in 2013	Stillbirth: a baby delivered at or after 24+0 weeks gestational age showing no signs of life, irrespective of when the death occurred	April 23, 2024 by guest	Socio-economic (area), ethnicity, and place (inferred regional and NHS Trust differences)	Deprivation -Based on mothers' postcodes at time of delivery, using the Children in Low- Income Families Loca Measure; Ethnicity - Broad categories [white, mixed, Asian o British Asian, Black or Black British, other]
Manktelow 2016 [70]	To collect, analyse and report national surveillance data to stimulate and evaluate improvements in health care for mothers and babies.	Confidential Enquiry	UK	All births in the UK in 2014	A baby delivered at or after 24+0 weeks gestational age showing no signs of life, irrespective of when the death occurred [including	Protected by copyright.	Socio-economic (area), ethnicity, and place (inferred regional and NHS Trust differences)	Deprivation -Based on mothers' postcodes at time of delivery, using the Children in Low- Income Families Local Measure; Ethnicity - Broad categories
						copyrigh		

		BMJ Open  BMJ Open  Separate definitions for ante & intrapartum  Separate definitions for ante & intrapartum						
					separate definitions for ante & intrapartum stillbirths	-029672 on		[white, mixed, Asian or British Asian, Black or Black British, other]
Gardosi 2005 [71]	summarise a recently completed analysis of stillbirth and infant mortality trends from 1998-2003 in the West Midlands; 2. assess the main categories contributing to these deaths; 3. analyse the trends of mortality associated with inequalities and the association with deprivation within different mortality subgroups.	Retrospective case-note audit	West Midlands	Perinatal mortality and infant mortality from the West Midlands over a 5 year period (1998 -2003)	> 24 weeks	Both September	Socio-economic (area)	Indices of Multiple Deprivation (IMD)
Gardosi 2007 [72]	To explore trends in perinatal mortality rates (including stillbirths) in the West Midlands over the period 1997-2005	Retrospective case-note audit	West Midlands	All births in the West Midlands Health Region over the period 1997-2005	24.0 weeks of pregnancy	2019.	Socio-economic (area), ethnicity	IMD (as above), ethnicity as recorded in notes
Gardosi 2009 [73]	To explore trends in stillbirths and infant deaths in the West Midlands over the period 1997-2007/8	Retrospective case-note audit	West Midlands	All births in the West Midlands Health Region over the period 1997-2007/8	24.0 weeks of pregnancy	Both Wnloaded	Socio-economic (area)	IMD (as above)
Gardosi 2011 [74]	To understand the causes underlying perinatal mortality and develop strategies for prevention	Retrospective case-note audit and survey of community midwives	West Midlands	Perinatal deaths	Perinatal mortality – defined as a stillbirth or a death of a live born baby in its first week of life.	Antepartum http://bmjopen.bm	Socio-economic (area), ethnicity	IMD (as above) Ethnicity - More subtle (Africa, African - Caribbean, Indian, Pakistani, Bangladeshi, Eastern European, Middle Eastern).
Tang 2008 [75]	To identify appropriate health targets by investigating associations between social deprivation and causes of stillbirth in Liverpool	Retrospective case-note audit	Liverpool	All stillbirths at local NHS Trust	Stillbirths from 24 weeks gestation.	Both 00	Socio-economic (area)	Indices of Multiple Deprivation (IMD)
Man 2016a [76]	What is the role of placental histological examination in determining the cause of intrauterine death?	Descriptive	London	Intrauterine deaths	Stillbirths (≥ 24 weeks)	Both	Ethnicity	Ethnicity – general categories
Man 2016b [77]	To present contemporary demographic findings from a large series of > 1000 intrauterine deaths in London which underwent autopsy investigation, and to examine these features in relation to the gestational age at which fetal death occurred, including deaths across both second and third trimesters, which are not captured by traditional registry-based approaches	Descriptive	London	Intrauterine deaths	Stillbirths (≥ 24 weeks)	Both nj.com/ on April 23,	Ethnicity	Ethnicity – general categories
Man 2016c [78]	To examine factors relating to determination of cause of death using a large dataset extracted from an autopsy research database including cases from two specialist centers, in which observer bias was reduced as far as possible by recording objectively findings at autopsy and assigning causes and classifications of death based on predetermined criteria.	Descriptive	London	Intrauterine deaths	Stillbirths (≥ 24 weeks)	2024 by guest.	Ethnicity	Ethnicity – general categories
Garcia 2017 [Unpublished PhD Thesis]	How do health beliefs influence health behaviour and contribute to perinatal mortality in babies born to Pakistani, Bangladeshi and WB women living in Luton?	Mixed Methods	Luton, UK	White, Pakistani and Bangladeshi women in Luton	Delivery of an infant with 'no signs of life' between 24 and 42 weeks of gestation	. Protected by	Intersectional – socioeconomic (individual and area), ethnicity and immigration status	Education level, ethnicity by country of heritage. Immigration status by years in UK.