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# **BMJ Open** Inequalities and stillbirth in the UK: a meta-narrative review

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#### 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 handsearching. 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** Fifty-four sources from nine distinctive research traditions were included. The 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 socioeconomic, 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 socioeconomic 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.

PROSPERO registration number CRD42017079228.

### 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 me-ta-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.

### INTRODUCTION

Avoidable inequalities in mortality across the life course are a global concern.<sup>1</sup> Ten countries account for 66% of the world's stillbirths, with most (98%) occurring in low-income and middle-income countries.<sup>2</sup> 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.<sup>3</sup> 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.<sup>4</sup> The UK's stillbirth rate continues to remain high in comparison to other HICs.<sup>5</sup>

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 1000 total births.<sup>6</sup> In 2017, the stillbirth rate in England and Wales was to 4.2 per 1000 total births.<sup>7</sup> Medical reasons for stillbirth

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are well known and strategies for prevention routine. Ongoing initiatives include the Safer Maternity Care strategic plan,<sup>8</sup> Saving Babies Lives Care Bundle,<sup>9</sup> Each Baby Counts,<sup>10</sup> the Perinatal Mortality Review Tool<sup>11</sup> and annual Perinatal Mortality Reports (MBRRACE-UK).<sup>5</sup> 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 1000 total births, compared with 3.0 per 1000 total births in the least deprived areas.<sup>7</sup>

The UK began to develop policies to address health inequalities in general following *The Acheson Inquiry into Inequalities in Health.*<sup>12</sup> 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.<sup>13</sup> 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<sup>14</sup> is underpinned by the Dahlgren and Whitehead rainbow model of the social determinants of health.<sup>15</sup> 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 interdisciplinary 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,<sup>16-18</sup> in accordance with the Realist And MEta-narrative Evidence Syntheses: Evolving Standards (RAMESES) standards.<sup>19</sup> A RAMESES checklist is provided (online supplementary file 1).<sup>20</sup> Our protocol<sup>21</sup> (online 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.

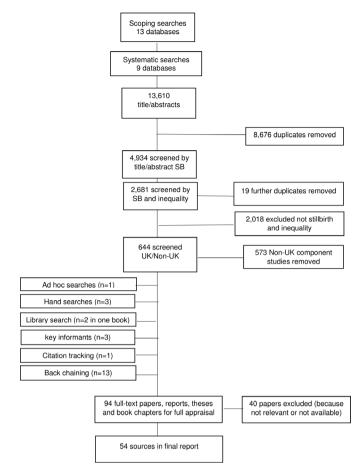
#### **Meta-narrative**

Meta-narrative review is a type of systematic review that was developed by Greenhalgh *et al.*<sup>16–18</sup> Meta-narrative is a term for the unfolding storyline of research in a particular tradition or topic, which draws on the theoretical approach in Kuhn's writing on paradigms.<sup>22</sup> 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<sup>17</sup>:

- 1. Planning: We registered our protocol with PROSPERO<sup>21</sup> and assembled a multidisciplinary 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 (online 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 online 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-income, middle-income 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 In vitro fertilisation/Intracytoplasmic sperminjection (IVF/ICSI). We applied these 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, KWF and CK), who also assigned potential inclusions to disciplinary categories at this stage (see online supplementary file 5: screening tool).

- 3. Mapping: A data extraction form was developed based on one used in earlier reviews,<sup>23</sup> 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 were 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 minimise 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<sup>24</sup> for some studies in the epidemiological tradition and the Walsh and Downe tool for qualitative research quality appraisal.<sup>25</sup>
- 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 1 involved evaluating the meta-narratives to identify and compare how the different research traditions conceptualised and theorised 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 2 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-theorising (exploring patterns that span conflicting understandings) to construct summary meta-themes. KWF, NC and CK undertook the initial analysis and synthesis processes, with input from DR, MAT, CS and SD.
- 6. Recommendations phase: We engaged with local clinical networks and the national Stillbirth and Neonatal Death Charity to formulate recommendations.



**Figure 1** RAMESES-PRISMA diagram. RAMESES, Realist And MEta-narrative Evidence Syntheses: Evolving Standards; PRISMA, Preferred Reporting Items for Systematic Reviews and Meta-Analyses.

#### Patient and public involvement

Author CS 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 13610 records were identified. Following duplicate removal 4934 records were screened (figure 1). We included 54 sources from nine research traditions spanning the period 1945-2017.<sup>26-78</sup> Table 1 provides a summary of included research traditions. Chronologically, these traditions were Social Medicine<sup>26–31</sup>; Epidemiology<sup>32–48</sup>; Medical Sociology<sup>49-53</sup>; Public Health<sup>54-58</sup>; Spatial Epidemiology<sup>59-64</sup>; Social Psychology<sup>65</sup>; Audits, Reports and Confidential Enquiries<sup>66–74</sup>; Fetal-Maternal Medicine<sup>75–78</sup> and Nursing and Midwifery (Garcia, Perinatal mortality in Pakistani, Bangladeshi and White British mothers in Luton). Online 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 used epidemiological data. We included one mixed-method

Research	Academic			Inequalities	Included
tradition	discipline	Definition and scope	Unfolding storyline	conceptualised as	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 (ie, mother's age) that have meaning whatever the social context and social factors (ie, occupational social class) that derive their meaning from social organisation in human life emulating political economy concerns.	The social medicine <sup>26-31</sup> storyline begins with the investigation of how social and economic factors influenced the decline in stillbirths and early neonatal deaths in Scotland, England and Wales, between 1939 and 1944. Baird <sup>26</sup> 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, age group and parity. By 1949, the decline in the stillbirth rate had slowed, despite the introduction of the National Health Service. Four papers, from a series in The Lancet in 1955 <sup>27-30</sup> 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 <sup>31</sup> 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 fewer stillbirths.	A variety of social factors that combine with biological characteristics to increase vulnerability to stillbirth risk.	n=6 ( <sup>26-31</sup> )
Epidemiology	Medicine	Epidemiology, developed out of the biomedical model as a specific line of inquiry. Initially epidemiology focused exclusively on epidemics of communicable diseases but subsequently expanded to address endemic 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 <sup>32-48</sup> storyline is characterised by its increasingly sophisticated use of data and the repetition of the same or similar findings over time. Of the 17 studies aligned to this tradition, six were landmark papers, repeatedly referenced within the field. <sup>32-37</sup> Although most authors highlighted a significant decrease in UK stillbirth rates since the 1960s, studies repeatedly showed that the social gradient remained constant. <sup>36 37 41</sup> Within overall stillbirth rates, being in a lower socioeconomic 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 incidence of stillbirth, when compared with more socially advantaged counterparts. An important strength is epidemiology's identification of clinical, socioeconomic 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 socioeconomic classification scheme. Other studies still used 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 <sup>34 43 44</sup> although this is not always the case. <sup>36</sup> Epidemiological studies looking at ethnicity 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. <sup>38</sup> <sup>45</sup> Studies exploring 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 <sup>38</sup> ; while other studies indicate a much lower rate—similar	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 ( <sup>32-48</sup> )

Continued

# 6

Table 1 Co	ontinued				
Research tradition	Academic discipline	Definition and scope	Unfolding storyline	Inequalities conceptualised as	Included references
Medical sociology	Sociology	despite the demographic and epidemiological changes associated with the transition to modernity was an important focus during the	The medical sociology <sup>49-53</sup> 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 <sup>31</sup> , and time lag (whereby developments in healthcare take time to reach those most in need, benefiting those better off first). <sup>49</sup> After the seminal Black report <sup>54</sup> more nuanced considerations of gender, age, ethnicity and area of residence, alongside occupational class, as simultaneous and overlapping vulnerabilities, were developed. <sup>50-52</sup> These encompassed the broad consideration of life circumstances, behaviours and beliefs/attitudes <sup>50</sup> and the precise disaggregation of the concept of 'deprivation' to reveal the complexity of materialist risks (and protections against those risks), which helps to explain the ambiguous association between economic deprivation and ethnicity. <sup>52</sup>	A set of social relations (rather than just a variable), which opens lived experience and multiplicity of factors at play (ie, poverty, poor housing, nutrition, welfare) and relationship between structure and agency.	n=5 ( <sup>49-53</sup> )
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 1980s, 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 <sup>54-58</sup> unites the seminal Black report <sup>54</sup> (which had a major impact on research into inequalities in health in the UK), with seminal papers from the two Lancet Stillbirth Series <sup>57,58</sup> that were of equal significance to the stillbirth research and policy community. In the former publication <sup>54</sup> , 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 idea of multiple causation) were developed. In the latter publications, distinguishing between different kinds of stillbirth and the importance of making each stillbirth count, come alongside the need to build on the idea (so father the interventions for black and ethnic minority women in the UK, despite their complex patterns of increased risk and known underutilisation of maternity services, was highlighted in the scoping review by Garcia et al. <sup>56</sup> In 2016, there was an explicit recall to action to tackle inequalities and stillbirth within HICs by addressing structural factors (such as poor housing, poverty) and factors, which limit women's access to antenatal care. <sup>58</sup>	An additional risk and considered in relation to providing targeted care to populations considered at risk.	n=5 ( <sup>54-58</sup> )
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 <sup>59-64</sup> 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 (eg, 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 <sup>59</sup> 61-63 and others reporting no association <sup>60</sup> <sup>64</sup> . The storyline of UK- based research 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 ( <sup>99-64</sup> )

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Continued

### **Open access**

Table 1 Continued   Research Academic   Inequalities Included						
tradition	discipline	Definition and scope	Unfolding storyline	conceptualised as	references	
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 <sup>65</sup> storyline arose from the Black report <sup>54</sup> and draws on theoretical explanations from the black report about the association between social inequality and ill health. This storyline is represented by one paper from 1990 <sup>65</sup> , 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 leads to a lack of knowledge and to deleterious beliefs and attitudes. The combined emotional and cognitive effects produce coping strategies and behaviours that increase the risk of negative pregnancy outcomes (ie, smoking). <sup>66</sup>	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 ( <sup>65</sup> )	
Audit reports and confidential enquiries	Interdisciplinary (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 Confidential Enquiry into Stillbirths and Deaths in Infancy, 2003–2011 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 <sup>66-74</sup> 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, regional and local 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 <sup>66 67</sup> and, more recently, under the banner MBRACE-UK <sup>66-70</sup> . Although we identified more than 20 national reports, only five explored the association between inequalities and stillbirth <sup>66-70</sup> 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 increased stillbirth risk. Four regional reports or audits from the West Midlands <sup>71-74</sup> attempt to look at stillbirth increased stillbirth rates. These reports were more nuanced and dentify a number of social and medical risk factors that could be screened for (alone or in combination) to predict risk of stillbirth (eg, unemployment, inappropriate housing, unsupported/difficult family circumstances, emotional factors/anxiety, maternal age <20 years or >40 years, obesity, smoking, consanguinity, history of mental health issues). The authors of these reports also highlight fetal growth restriction 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 and Asian populations).	n=9 ( <sup>66-74</sup> )	

6

Continued

Table 1 Continued						
Research tradition	Academic discipline	Definition and scope	Unfolding storyline	Inequalities conceptualised as	Included references	
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 <sup>75–78</sup> included a study reporting that women living in areas of highest deprivation (IMD 1) were more likely to experience fetal growth restriction compared with women living in the least (IMD 3–9). <sup>75</sup> Approximately 46% of these women smoked, compared with 7% in the least deprived. The study concluded that targeted antenatal management was key to stillbirth prevention among women living in the most deprived areas. This tradition also offered three 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 weeks gestation) <sup>76–78</sup> . 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. <sup>78</sup> .	A risk factor for stillbirth and depending on the type of study, may be included as a covariate in the analysis.	n=4 ( <sup>75-78</sup> )	
Nursing and midwifery	Nursing and midwifery	Nursing and midwifery research draws from positivist and interpretative paradigms, using 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, perinatal 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 ('2 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 (Garcia, perinatal mortality in Pakistani, Bangladeshi and white British mothers in Luton).	

HIC, high-income country.

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 were not practical. Figure 2 maps the traditions contribution over time and the declining national stillbirth rate.

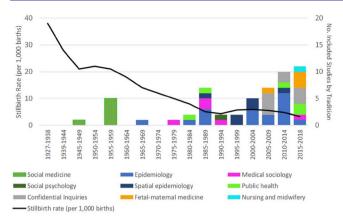
#### Synthesis within traditions

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

#### 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'<sup>26</sup> 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'<sup>58</sup> 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



**Figure 2** Timeline of included studies by research tradition and the stillbirth rate in England and Wales 1945–2017.

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, intergenerationally, 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.<sup>71</sup>

#### 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 and intrapartum), at what gestation (early preterm, late preterm and 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 socioeconomic 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 and 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.<sup>3150515455</sup>

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.<sup>61</sup> <sup>62</sup> The problems associated with using crude categories to define ethnicity (ie, white, black, Asian) were also considered (Epidemiology, Medical Sociology and Public Health) and the complexities therein (ie, 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 finegrain 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'. (Macintyre, p.393)<sup>50</sup> This theme attempts to shine some 'light on the most appropriate times to provide support and the form(s) that such support should take.' (Weightman, p11)<sup>42</sup> 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

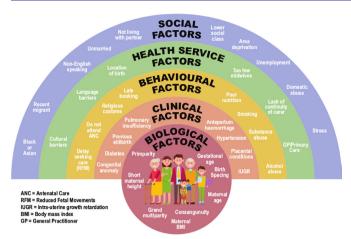


Figure 3 Factors associated with inequalities and stillbirth.

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 (ie, non-English speaking, unemployed household) in combination with behavioural factors (ie, smoking) and clinical factors (ie, previous intrauterine growth retardation), there was little consensus on specific factors, timing, or outcome if social conditions remain the same.<sup>39 40 46</sup>

#### 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 suggest that membership of a lower socioeconomic 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 complex, and 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 meta-narrative 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 prespecified quality appraisal tools<sup>2425</sup> 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.<sup>3 4 6 79–82</sup> 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.<sup>83–86</sup> 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,<sup>87</sup> 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.<sup>88</sup> However, neither of these address the underlying structural components of disadvantage.

#### Implications for clinicians and policy-makers

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 (ie, sudden infant death, cardiovascular disease and cancer). Health inequalities theory advocates intervening at specific time points during the life course (ie, pregnancy and the early years), interventions that have impact over time (ie, intragenerational and intergenerational), interventions at scale (ie, national policies) and interventions targeting specific groups (ie, 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.<sup>89</sup> 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.<sup>86</sup> 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.<sup>1 89</sup> 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, inequality is a term that has endured.<sup>13 14 86 90</sup> 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 Royal College of Obstetricians and Gynaecologists (RCOG) and National Health Service 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.<sup>5</sup> 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 or 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.<sup>91 92</sup> 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.<sup>93–95</sup> Not least because, these particular components of antenatal care already feature as part of stillbirth reduction initiatives, but to date, have had limited success.<sup>82</sup>

The role of social factors, modifiable lifestyle behaviours and antenatal interventions in stillbirth prevention are current research priorities identified by the stillbirth community.<sup>96</sup> 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 and (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 multidisciplinary insight into these complexities, challenges and opportunities, it provides a starting point for a novel transdisciplinary response.

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# 6

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