

# BMJ Open Alterations in maternally perceived fetal movement and their association with late stillbirth: findings from the Midland and North of England stillbirth case-control study

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## ABSTRACT

**Objective** To report perception of fetal movements in women who experienced a stillbirth compared with controls at a similar gestation with a live birth.

**Design** Case-control study.

**Setting** 41 maternity units in the UK.

**Participants** Cases were women who had a late stillbirth  $\geq 28$  weeks gestation ( $n=291$ ) and controls were women with an ongoing pregnancy at the time of the interview ( $n=733$ ). Controls were frequency matched to cases by obstetric unit and gestational age.

**Methods** Data were collected using an interviewer-administered questionnaire which included questions on maternal perception of fetal movement (frequency, strength, increased and decreased movements and hiccups) in the 2 weeks before the interview/stillbirth. Five fetal movement patterns were identified incorporating the changes in strength and frequency in the last 2 weeks by combining groups of similar pattern and risk. Multivariable analysis adjusted for known confounders.

**Primary outcome measure** Association of maternally perceived fetal movements in relation to late stillbirth.

**Results** In multivariable analyses, women who reported increased strength of movements in the last 2 weeks had decreased risk of late stillbirth compared with those whose movements were unchanged (adjusted OR (aOR) 0.18, 95% CI 0.13 to 0.26). Women with decreased frequency (without increase in strength) of fetal movements were at increased risk (aOR 4.51, 95% CI 2.38 to 8.55). Daily perception of fetal hiccups was protective (aOR 0.31, 95% CI 0.17 to 0.56).

**Conclusions** Increased strength of fetal movements and fetal hiccups is associated with decreased risk of stillbirth. Alterations in frequency of fetal movements are important in identifying pregnancies at increased risk of stillbirth, with the greatest risk in women noting a reduction in fetal activity. Clinical guidance should be updated to reflect that increase in strength and frequency of fetal movements is associated with the lowest risk of stillbirth, and that decreased fetal movements are associated with stillbirth.

**Trial registration number** NCT02025530.

## Strengths and limitations of this study

- This is the largest case-control study reporting detailed information about maternal perception of fetal movements in relation to stillbirth.
- This study addressed different aspects of fetal activity including frequency and strength and hiccups.
- Attempts were made to reduce recall bias including: using a structured questionnaire and no explicit hypotheses communicated to participants.
- The recruitment rate was lower than initially expected, 44.1% for cases and 25.9% for controls, which may introduce selection bias.
- The multivariate model included a prioritised variable that was partially derived from the data obtained in the study, this approach could introduce bias, thus, these observations require replication in further independent data sets.

## INTRODUCTION

Maternal perception of fetal activity is an accepted marker of fetal well-being. Conversely, maternal perception of changes in activity can indicate fetal compromise; the most commonly reported change is a reduction in fetal movement.<sup>1 2</sup> Maternal perception of reduced fetal movements (RFM) is associated with adverse pregnancy outcomes including fetal growth restriction,<sup>3</sup> oligohydramnios<sup>4</sup> and stillbirth.<sup>3</sup> These conditions are associated with placental dysfunction, which is observed in women with RFM.<sup>5 6</sup> Despite the known association between RFM and stillbirth, two Confidential Enquiries into antepartum stillbirth in the UK conducted 15 years apart highlighted suboptimal care in terms of the information given to mothers about fetal movements and clinical

management when mothers attend with RFM as factors contributing to stillbirth.<sup>7,8</sup>

In comparison to a reduction in frequency of fetal movements, little is known about other aspects of maternally perceived fetal activity, such as: strength of movements, an episode of vigorous movement and fetal hiccups and how these relate to risk of stillbirth. Data from two case-control studies and a large international cohort study have both suggested that any significant deviation from a mother's usual pattern of fetal movement is a risk factor for stillbirth.<sup>9</sup> Importantly, existing data suggest that an increase in both strength and frequency of fetal movements in late pregnancy was reported significantly less frequently by women who had a stillbirth.<sup>9-11</sup> Due to this paucity of data, it is important to better understand maternal perception of altered fetal activity and whether these perceptions can be used to identify fetuses at high risk of antepartum stillbirth. Furthermore, women report receiving mixed messages about the importance of fetal movements and the significance of RFM, indicating the need for clear information regarding these symptoms.<sup>12</sup> To address these needs, we conducted a case-control study to explore modifiable risk factors associated with late stillbirth. The objective of this manuscript is to report maternally perceived fetal movements in women who experienced a recent stillbirth compared with a control group of women at similar gestation who had a live baby.

## METHODS

The Midlands and North of England Stillbirth Study (MiNESS) was conducted in 41 maternity units in the UK. The study was registered on [www.clinicaltrials.gov](http://www.clinicaltrials.gov) (NCT02025530) and the study protocol was published.<sup>13</sup> Participants were recruited between April 2014 and March 2016. The study methodology has been described in detail elsewhere.<sup>14</sup> Cases were included if the stillbirth occurred at or after 28 weeks gestation and the fetus did not have a congenital anomaly. The cause of stillbirth was assigned using the ReCoDe classification system.<sup>15</sup> Controls were women with an ongoing pregnancy. To ensure that controls would be at a similar gestation to cases, the gestation at interview was frequency matched to the expected distribution of stillbirths based on the prior 4 years of data from that unit. Potential controls were randomly selected from the booking lists and the gestation for interview calculated from the expected date of delivery. Women with multiple pregnancies, maternal age less than 16 years and inability to give consent were excluded from the study.

The primary outcome reported here was the association of maternal perception of fetal movements with late stillbirth. Maternal perception of fetal movements was classified as increased, reduced or stayed the same. Data specific to this analysis relates to questions asked about fetal movements and more specifically about changes in strength and frequency in the last 2 weeks (before the baby died for cases and last 2 weeks before interview for

controls). Additional information was collected on fetal hiccups. Data on uterine contractions were also collected as it has been argued elsewhere that women may interpret uterine contractions as fetal movements.<sup>16</sup> All questions as asked are reported in [table 1](#) (the questionnaire is included as online supplementary file 1).

## Statistical methods

Univariable analyses were carried out using logistic regression to estimate the effect of each variable. Due to likely relationships between the variables, bivariate models were fitted between each pair of movement variables to assess (by changes in effect size) which variables were able to be placed in multivariable analyses together. There was a strong association between reduced movements after 26 weeks gestation and the variables for strength and frequency of movements in the last 2 weeks, meaning these variables could not be included in the same multivariable model. Additionally, the question relating to RFM since 26 weeks is complicated by the fact that the time frame relating to this question varies by subjects, that is, 2 weeks for a women at 28 weeks gestation and 15 weeks at 41 weeks gestation.

Although the strength and frequency variables were also associated with each other, they measure different aspects of movement. However, as using 16 potential combinations (derived from four different levels of both strength and frequency) independently would greatly reduce the statistical power, a combined strength/frequency variable was developed to describe the relationship between the changes in strength and frequency of movements in the last 2 weeks before the interview/stillbirth. The variable was prioritised based on the prevalence of the perception in controls and the magnitude of the associated risk. Groups of similar pattern and risk were then combined (see online supplementary table 1). Thus, the prioritised strength/frequency variable used the following rules:

1. Increase in strength of movements.
2. Increase in frequency but not strength of movements.
3. Decrease in frequency of movements.
4. Unsure of change in strength or frequency.
5. No change in strength or frequency (reference category based on current guidelines).

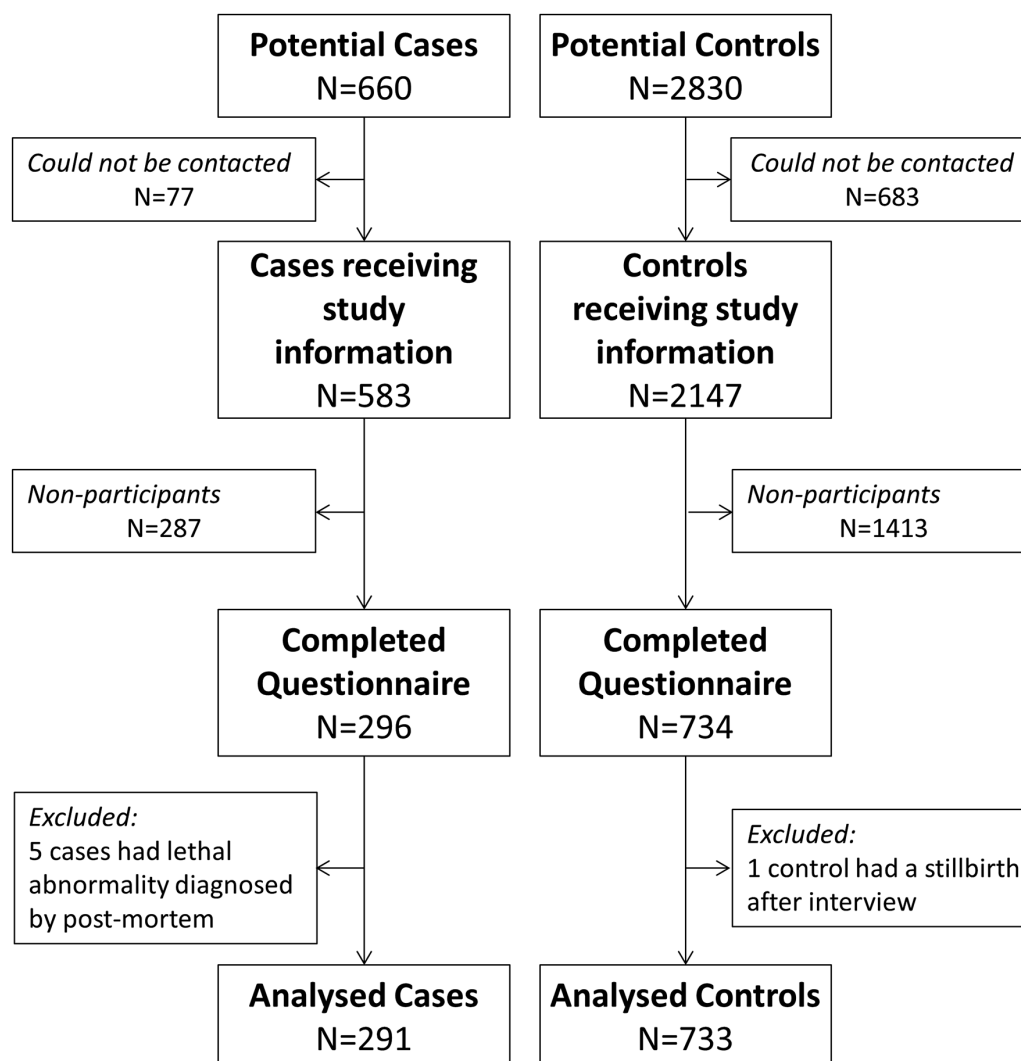
Multivariable analyses were carried out by adding the variables identified as not showing significant collinearity (prioritised strength/frequency variable, frequency of increased fetal movements and frequency of feeling hiccups) to the model previously developed in relation to the risk of stillbirth in this study (maternal: age, ethnicity, parity, education, smoking in pregnancy, marital status, customised birthweight centile,<sup>14</sup> sleep factors on the last night before stillbirth/interview (position went to sleep in, sleep duration, number of times got up to toilet), naps in the daytime, gestation and study centre). All analyses were carried out using the logistic procedure in SAS V.9.4 (SAS Institute).

**Table 1** Univariable risks associated with perception of fetal movements and late stillbirth risk

	Cases, n (%)	Controls, n (%)	Odds ratio (95% Confidence Interval)* $\chi^2$ , p values
Was there any time from 26 weeks of pregnancy that your baby's movements were less than usual?			
No	112 (38.7)	469 (64.2)	Reference: $\chi^2=66.69$ , $p<0.0001$
Once	88 (30.5)	156 (21.3)	2.36 (1.69 to 3.30)
Two times	39 (13.5)	65 (6.9)	2.51 (1.61 to 3.93)
Three or more times	50 (17.3)	41 (5.6)	5.11 (3.22 to 8.10)
In the last 2 weeks did the strength of your baby's movements			
Increase	53 (18.3)	455 (62.8)	0.15 (0.11 to 0.22)
Decrease	62 (21.4)	50 (6.9)	1.61 (1.05 to 2.46)
Stay the same	153 (52.8)	198 (27.3)	Reference: $\chi^2=169.96$ , $p<0.0001$
Unsure	22 (7.6)	22 (3.0)	1.29 (0.69 to 2.42)
In the last 2 weeks did the frequency of your baby's movements			
Increase	37 (12.7)	254 (34.8)	0.38 (0.26 to 0.56)
Decrease	86 (29.6)	63 (8.6)	3.54 (2.44 to 5.15)
Stay the same	153 (52.6)	397 (54.3)	Reference: $\chi^2=103.49$ , $p<0.0001$
Unsure	15 (5.2)	17 (2.3)	2.29 (1.12 to 4.70)
During the last 2 weeks did you notice anytime that your baby was more vigorous than usual?			
No	182 (62.5)	326 (44.7)	Reference: $\chi^2=57.39$ , $p<0.0001$
Once	41 (14.1)	50 (6.9)	1.47 (0.94 to 2.31)
More than once	68 (23.4)	354 (48.5)	0.34 (0.25 to 0.47)
During the last 2 weeks did you feel you baby having hiccups?			
Yes	126 (43.5)	460 (62.9)	0.41 (0.30 to 0.54)
No	141 (48.6)	209 (28.6)	Reference: $\chi^2=38.10$ , $p<0.0001$
Unsure	23 (7.9)	62 (8.5)	0.55 (0.33 to 0.93)
How often did you feel hiccups in the last 2 weeks?			
Not felt hiccups	141 (48.5)	209 (28.6)	Reference: $\chi^2=42.01$ , $p<0.0001$
Unsure if felt	23 (7.9)	62 (8.5)	0.73 (0.39 to 1.35)
Once	17 (5.8)	36 (4.9)	0.69 (0.37 to 1.27)
Occasionally	69 (23.7)	235 (32.2)	0.44 (0.32 to 0.62)
Daily	38 (13.1)	177 (24.3)	0.32 (0.21 to 0.48)
Unsure of frequency	3 (1.0)	11 (1.5)	0.37 (0.17 to 0.80)
During the last 2 weeks did you feel uterine contractions (tightenings/prelabour contractions/Braxton Hicks contractions/false labour) for longer than an hour			
Yes	94 (32.3)	241 (33.0)	0.97 (0.72 to 1.29)
No	191 (65.6)	473 (64.7)	Reference: $\chi^2=0.12$ , $p=0.94$
Unsure	6 (2.1)	17 (2.3)	0.87 (0.34 to 2.25)
Combination of strength and frequency changes in the last 2 weeks (prioritised variable)†			
Increased strength	53 (18.2)	455 (62.1)	0.18 (0.13 to 0.26)
Increased frequency but not strength	8 (2.8)	22 (3.0)	0.57 (0.25 to 1.32)
Decreased frequency	79 (27.2)	36 (4.9)	3.45 (2.20 to 5.43)
Unsure strength or frequency	22 (7.6)	17 (2.3)	2.04 (1.04 to 3.98)
Same	129 (44.3)	203 (27.7)	Reference: $\chi^2=205.34$ , $p<0.0001$

\* $\chi^2$  and associated p values are given for the overall effect of each variable.

†See online supplementary table 1 for detailed description of category's included in prioritised variable categories.



**Figure 1** Flow diagram reporting the numbers of women eligible for the study, women who did not participate and those included in the final analysis.

### Patient and public involvement

MiNESS was developed in response to research questions prioritised in the Stillbirth Priority Setting Partnership according to methodology developed by the James Lind Alliance.<sup>17</sup> These included: 'Do modifiable 'lifestyle' factors (eg, diet, vitamin deficiency, obesity, sleep position, sleep apnoea, lifting and bending) cause or contribute to stillbirth?' and 'Would empowering women to know about relevant evidence-based signs and symptoms and raise them with healthcare professionals reduce stillbirth?' Study design and participant materials were designed in conjunction with the Maternal and Fetal Health Research Centre Patient and Public Involvement Group. Participants were not involved in recruitment to or conduct of the study.

### RESULTS

In total, 3490 women were identified as potentially eligible participants (660 cases and 2830 controls, [figure 1](#)). Seven hundred and sixty women could not be contacted

(77 cases and 683 controls) and 1700 women did not consent to participate in the study (287 cases and 1413 controls). Six cases were excluded after data collection (five stillbirths had previously unidentified congenital abnormalities detected on postmortem and one control participant had a stillbirth). Cases were more likely to participate than controls ( $p<0.0001$ ), 291 cases (44.1%) and 733 controls (25.9%) were included in the analysis ([figure 1](#)).

The demographic characteristics of the study population have been presented in detail previously.<sup>14</sup> Briefly, the majority of participants were from white ethnic background (80.4% of cases and 81.0% of controls), with a significant proportion of participants from South Asian (13.4% of cases and 13.0% controls) and black ethnic groups (4.1% of cases and 4.0% of controls). Participants' ages were distributed across the reproductive lifespan, with the largest group between 30 and 34 years of age in both groups (29.6% cases, 36.6% controls). There was no difference in mean body mass index (cases 26.9 kg/m<sup>2</sup>,



controls 26.0 kg/m<sup>2</sup>). The median gestation at interview was 36 weeks 3 days for controls (IQR 32 weeks 6 days to 38 weeks 5 days). In cases, the median gestation at diagnosis of stillbirth was 37 weeks 4 days (IQR 33 weeks 4 days to 39 weeks 5 days,  $p=0.003$  compared with controls). The median interval between the presumed date of death in utero and diagnosis was 0 days (IQR 0–1) and the median time between the diagnosis of stillbirth and interview was 25 days (IQR 17–35). The most frequent factors associated with stillbirth were fetal growth restriction (45.2%), placental insufficiency (16.4%), placental abruption (6.5%) and acute infection (4.5%).

The prevalence of each variable relating to fetal movements and their univariable ORs associated with stillbirth are presented in table 1. Women who reported RFM any time after 26 weeks gestation were at increased risk of having a stillbirth with the risk increasing with the number of times that they reported that decreased movements had occurred ranging from an OR of 2.36 (95% CI 1.69 to 3.30) for one episode to an OR of 5.11 (95% CI 3.22 to 8.10) for three or more episodes. Similarly, women who reported a decrease in either strength (OR 1.61, 95% CI 1.05 to 2.42) or even more so frequency (OR 3.54, 95% CI 2.44 to 5.15) of fetal movements in the last 2 weeks were at increased risk of having a stillbirth compared with those who reported no change.

Conversely, increasing strength (OR 0.15, 95% CI 0.11 to 0.22) or frequency (OR 0.38, 95% CI 0.26 to 0.56) of fetal movement was associated with reduced stillbirth risk compared with those who reported no change. There was

a significantly decreased stillbirth risk in those reporting more than one episode of vigorous movement (OR 0.34, 95% CI 0.25 to 0.47) and a trend towards increased stillbirth risk in women who felt a single episode of vigorous movement (OR 1.47, 95% CI 0.94 to 2.31) compared with those who never perceived movements to be more vigorous than usual. The combined variable derived from the strength and frequency variables showed that compared with no change in frequency or strength of movement, those who reported increased strength in the last 2 weeks had a decreased risk of stillbirth (OR 0.18, 95% CI 0.13 to 0.26), which was the most commonly reported scenario in controls (62%), while those reporting decreased frequency of movements were at increased risk (OR 3.45, 95% CI 2.20 to 5.43).

Maternal perception of fetal hiccups in the last 2 weeks was associated with a decreased risk of stillbirth (OR 0.41, 95% CI 0.30 to 0.54). The magnitude of this reduced risk increased as the frequency of feeling hiccups increased, with the lowest risk for daily perception of hiccups (OR 0.32, 95% CI 0.21 to 0.48). There was no association between feeling contractions in the last 2 weeks and stillbirth (OR 0.97, 95% CI 0.72 to 1.29).

The multivariable model (table 2) showed that a decrease in frequency of fetal movements remained associated with increased risk of stillbirth (adjusted OR (aOR) 4.51, 95% CI 2.38 to 8.55) and increasing strength of fetal movements was still associated with decreased risk of stillbirth (aOR 0.14, 95% CI 0.08 to 0.24) compared with no change in perception of frequency or strength of

**Table 2** Univariable and multivariable odds of late stillbirth associated with perceptions of fetal movements

	Univariable OR (95% CI)	Multivariable* OR (95% CI), p values
Combination of strength and frequency changes in the last 2 weeks (prioritised variable)		
Increased strength	0.18 (0.13 to 0.26)	0.14 (0.08 to 0.24)
Increased frequency but not strength	0.57 (0.25 to 1.32)	0.86 (0.30 to 2.52)
Decreased frequency	3.45 (2.20 to 5.43)	4.51 (2.38 to 8.55)
Unsure strength or frequency	2.04 (1.04 to 3.98)	2.02 (0.77 to 5.25)
Same	Reference	Reference: $\chi^2=104.90$ , $p<0.0001$
During the last 2 weeks, did you notice anytime that your baby was more vigorous than usual		
No	Reference	Reference: $\chi^2=12.43$ , $p=0.002$
Once	1.47 (0.94 to 2.31)	2.10 (1.06 to 4.17)
More than once	0.34 (0.25 to 0.47)	0.59 (0.37 to 0.96)
How often did you feel your baby having hiccups in the last 2 weeks		
Never	Reference	Reference: $\chi^2=5.95$ , $p=0.007$
Unsure if felt	0.73 (0.39 to 1.35)	0.90 (0.42 to 1.93)
Once	0.69 (0.37 to 1.27)	0.85 (0.35 to 2.05)
Occasionally	0.44 (0.32 to 0.62)	0.75 (0.45 to 1.26)
Daily	0.32 (0.21 to 0.48)	0.31 (0.17 to 0.56)
Unsure of frequency	0.37 (0.17 to 0.80)	0.50 (0.07 to 3.44)

\*Controls for age, ethnicity, parity, education, marital status, smoking in pregnancy, customised birthweight centile, going-to-sleep position, sleep duration, got up to toilet in the night, naps in the daytime, gestation and study centre.

movement in the last 2 weeks. The decreased risk associated with feeling vigorous movements on more than one occasion in the last 2 weeks remained statistically significant (aOR 0.59, 95% CI 0.37 to 0.96), but the association between a single episode of vigorous movement in the preceding 2 weeks and stillbirth became statistically significant (aOR 2.10, 95% CI 1.06 to 4.17). Compared with not feeling hiccups in the last 2 weeks, feeling hiccups daily was associated with a significant reduction in risk (aOR 0.31, 95% CI 0.17 to 0.56).

When baby's movements were reported as less than usual in the preceding 2 weeks, mothers of cases were significantly more likely to have spoken to a health professional (79% vs 70%,  $p=0.02$ ) and tended to have attended hospital due to RFM more frequently than controls although this did not achieve statistical significance (68% vs 60%,  $p=0.07$ ).

## DISCUSSION

### Main findings

This study shows that the majority of women with a live birth after 28 weeks gestation perceive an increase in strength of fetal movements and feel fetal hiccups in the previous 2 weeks; perception of these patterns of fetal movements is associated with a substantial reduction in the risk of late stillbirth (aORs 0.14 and 0.31, respectively). Conversely, a decrease in the strength or frequency of fetal movements is associated with an increased risk of late stillbirth particularly if this is a recurrent phenomenon (OR 2.36 rising to 5.11). A single episode of vigorous fetal activity is also associated with an increased risk of stillbirth.

### Strengths and limitations

This study is the largest case-control study that has reported detailed information about maternal perception of fetal movements in relation to the risk of late stillbirth. A comparatively novel feature of this study is that assessment of analysis of fetal activity was not restricted to the frequency of fetal movements, but also addressed changes in strength of fetal activity as well as fetal hiccups. Apart from the Auckland Stillbirth Study (TASS),<sup>9</sup> most recent publications evaluating the significance of fetal movements have comprised small cohort studies of women with reduced frequency of fetal movements in centres where intervention may be employed to prevent stillbirth.<sup>18 19</sup> By including a broader description of fetal activity, this study has been able to report the frequency and strength of fetal movements in ongoing pregnancy and describe changes that are related to late stillbirth.

A case-control design was considered the most practical means to identify late stillbirth, as a prospective cohort study is not feasible, requiring almost 108 000 women to identify 291 late stillbirths (at the current frequency of 2.9 per 1000 births in the UK). However, it is important to consider the potential influence of recall bias, a limitation of case-control studies. This study attempted to minimise recall bias in several ways. Firstly, all participants

were asked the same series of questions about fetal movements embedded in a questionnaire about many different factors (eg, smoking, diet, stress, social situation, sleep position and fetal movements). Secondly, women who experienced a stillbirth were interviewed within a median of 25 days, a time when events surrounding the death of a baby are likely to be clearly recalled.<sup>20</sup> As women in the control group were pregnant at the time they completed the survey, their experiences or concerns could not have been biased by knowledge of the outcome of their pregnancy. Finally, this study described novel findings of vigorous fetal movements and fetal hiccups, which are rarely addressed in prior studies, reducing the possibility that respondents may have read about these symptoms in advance of the questionnaire. While recall bias cannot be completely discounted, responses from participants who had a stillbirth do not universally show a deviation from controls, for example, there was no difference in maternal perception of uterine contractions between the two groups.

The possibility of selection bias was minimised by recruiting controls who were frequency matched to cases over the duration of the study period which resulted in similar ages and ethnicities in both groups. The recruitment rate of MiNESS was lower than that of the TASS case-control study (cases 45.3% vs 72%; controls 26.2% vs 72%, respectively).<sup>21</sup> This may have resulted in part from random selection of controls from booking lists which meant that some of these women could not be contacted and others were approached with no prior knowledge of the study, which given the sensitive subject matter, may have been reduced participation rates. A qualitative substudy was undertaken to further understand the barriers and facilitators to participation in this study for both cases and controls, this will be reported separately.

Another potential bias in the multivariate model is the inclusion of a prioritised variable that was partially derived from the data obtained in the study. This approach was taken to reduce the number of combinations of strength and frequency by placing them in like groups (eg, increased strength, increased frequency) which had similar risk estimates. This approach could introduce bias and consequently requires replication in further independent data sets, which is currently under way using data from the multicentre stillbirth study from New Zealand.<sup>22</sup>

### Interpretation

Data regarding the pattern of fetal movements in late pregnancy are limited. Previous literature has suggested that the frequency of fetal movements increases until the 32 weeks of pregnancy and then plateaus.<sup>23 24</sup> Studies also note that the type and quality of fetal movements change with advancing gestation.<sup>23-26</sup> In this study, the majority of controls reported that the frequency of fetal movements stayed the same (54.3%) but that there was increased strength of fetal movements in most controls (62.8%) in the preceding 2 weeks. Interestingly, an increase in strength of fetal movements had a greater protective

effect for stillbirth than increase in frequency (OR 0.15 vs 0.38). These findings are similar to those reported in TASS.<sup>9</sup> As ultrasound studies suggest that mothers are more likely to feel larger movements of trunk and limbs,<sup>27</sup> an increase in strength may also be perceived as an increase in frequency. Critically, for a reduction in frequency of fetal movements can only be judged in retrospect, whereas, increased strength may be easier to judge in real time which could prompt more rapid reporting of maternal concerns. We were not able to stratify levels of fetal activity by gestation due to insufficient sample size, this will be addressed in a planned individual participant data (IPD) meta-analysis.<sup>28</sup> The other studies within the IPD can also be used to determine whether the interaction between strength and frequency is similar, and has similar association with late stillbirth.

Although regular vigorous movements are important and protective, a one-off episode of excessive fetal activity may be a warning sign of fetal compromise although the effect size in this study was less than in TASS (aOR 2.10 vs 6.81).<sup>9</sup> The repeated identification of this association here strengthens the relationship between a single episode of excessive fetal activity and stillbirth. However, practical application of this association is challenging as a woman cannot know at the time whether an episode of vigorous movement is isolated or will become a part of regular fetal activity. Furthermore, the origin of the excessive movement is unclear.<sup>29</sup> Therefore, this association requires further investigation in our planned IPD meta-analysis to establish whether it is consistently observed,<sup>28</sup> and whether there are any clues to the aetiology of this symptom.

In agreement with many studies since the mid-1970s, we have confirmed that decreased frequency of fetal movements is a major risk factor for late stillbirth.<sup>30</sup> Furthermore, this study agrees with data from other UK units that recurrent presentation with RFM is associated with an even greater risk of adverse outcome.<sup>31 32</sup> This link is biologically plausible as RFM is associated with abnormal placental structure and function which may deteriorate as pregnancy progresses.<sup>5 6</sup> Notably, mothers with recurrent episodes of RFM have been shown to have an increased likelihood of abnormal uterine artery Doppler waveforms in the second trimester and delivery of a small for gestational age infant,<sup>31</sup> both of which are associated with abnormal placental morphology.<sup>33 34</sup> This study did not have sufficient power to determine whether maternal perception of RFM was related to stillbirths associated with a specific cause (eg, placental dysfunction) but this will be addressed in the IPD meta-analysis.

Our data regarding the protective effect of fetal hiccups are an important observation which is consistent with findings from TASS.<sup>9</sup> This finding contrasts with a single case report which proposed that hiccups are linked to umbilical cord complications.<sup>35</sup> Fetal hiccups appear on ultrasound to be interspersed with normal breaths and are considered physiological.<sup>36 37</sup> Mothers are aware of fetal hiccups throughout pregnancy, one study of 45 women

suggested that they were perceived more frequently prior to 26 weeks gestation and remained constant after that with an average of 0.4 episodes per hour.<sup>38</sup> Although another study found 36.6% of women perceived hiccups in pregnancy, and this increased with gestational age.<sup>39</sup> Fetal hiccups do not appear to relate to other aspects of fetal movement, although they are associated with active fetal behavioural states.<sup>38</sup>

Women in this study with RFM who went on to have a stillbirth were more likely to have spoken to a healthcare professional about the symptom but only 68% attended hospital because of RFM, indicating that contacting a health professional does not appear to prevent stillbirth. This may be because a significant proportion of women do not attend hospital, management of RFM is variable,<sup>40 41</sup> or that the baby was already dead at the time of presentation.<sup>42</sup> Critically, management is presently not informed by high-quality evidence as there are insufficient data from randomised trials to guide practice.<sup>43</sup> It is anticipated that the AFFIRM study (NCT01777022), a multicentre stepped-wedge cluster randomised trial will address whether standardised information for women and a standardised management strategy (employing antepartum cardiotocography and ultrasound for fetal biometry and liquor volume) following attendance with RFM will reduce stillbirth.<sup>44</sup>

## CONCLUSION

This study demonstrates that maternal perception of increased strength of fetal movements in late pregnancy is protective of late stillbirth. Decreased frequency of fetal movements is associated with risk of stillbirth as is decreased strength. Clinical guidelines and health promotion information currently suggest that fetal movements tend to increase until the 32nd week of pregnancy and then plateau.<sup>45</sup> However, data from this study and TASS show that an increase in strength and frequency of fetal movements is associated with the lowest risk of stillbirth suggesting that guidance should be altered to indicate that maternal perception of fetal movement normally increases throughout pregnancy. This study adds to the evidence base that when fetal movements are reduced, there is an increased risk of late stillbirth. Thus, women should contact their maternity care provider and be managed according to current clinical guidance.<sup>46</sup> Importantly, development of an effective strategy for the investigation and management of RFM in late pregnancy has the potential to reduce the incidence of late stillbirth.

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**Data sharing statement** No additional data from the MiNESS study are available from a repository. Anonymised data are available on request to the corresponding author.

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