Stillbirths

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Stillbirth: Why Invest?

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Abstract

Despite its frequency, the implications of stillbirth are overlooked and underappreciated. We present findings from comprehensive, systematic literature reviews, and new analyses of published and unpublished data, to establish the impact of stillbirth on parents, families, healthcare providers, and societies around the world. Data on direct costs of stillbirth are sparse, but indicate that stillbirth requires more resources than a live birth, both in the perinatal period and in additional surveillance during subsequent pregnancies. Indirect and intangible costs of stillbirth are far-reaching and are usually met by families. This is particularly onerous for those with fewer resources. Negative effects, particularly on parental mental health, may be moderated by empathic attitudes of care providers and tailored interventions. Efforts to prevent stillbirths and reduce associated morbidity should consider the value of the baby, as well as the associated costs and resource implications for parents, families, care providers and communities.

Key Messages

- Stillbirth is associated with significant direct, indirect and intangible costs to women, their partners and families, health care providers, government and wider society. Appreciation of the costs of stillbirth is essential to evaluate the cost-effectiveness of interventions to prevent stillbirth or ameliorate negative impact of stillbirth.

- Data on the cost of stillbirth in high-burden countries are inadequate. In addition to collecting data on the number of stillbirths, data should be collected on the resource implications.

- Adverse experiences including stigma, social isolation and disenfranchised grief are common amongst parents whose baby is stillborn and need to be addressed through focussed interventions and supportive activities involving parents, communities, care-providers and relevant stakeholders.

- Empathic behaviours during every encounter between bereaved parents and care givers are essential to minimise additional emotional and psychological burdens in the short, medium and longer term.

- Caring for families during and following stillbirth places a substantial personal and professional burden on staff. Negative impacts on staff could be addressed by education, training, and provision of formal and informal support.

Introduction

Despite the 2.7 million stillbirths globally,[1] the costs of stillbirth are largely unknown and consequently unappreciated in contrast to other adverse pregnancy outcomes.[2-5] For the most part, health metrics, such as quality-adjusted life years (QALYs) and disability-adjusted life years (DALYs) have neglected stillbirth. No value is generally given to the loss of life or the loss to parents and families. To date the majority of economic analyses have focussed on the cost of stillbirth prevention.[4, 6, 7] In low and middle income countries (LMICs) costs vary from US$ 4,781-10,571 per stillbirth averted (in 2013 prices).[4, 6] In high-income countries (HICs) with lower stillbirth rates, prevention costs are greater, for example smoking cessation costs $125,961 per stillbirth averted.[8] If stillbirths are included in analyses of the impact of antenatal and intrapartum care on maternal and newborn deaths, the cost per death averted falls considerably from $27,551 to $2,143 (Panel 1).[4] However, to accurately assess whether these programmes are cost-effective, a better appreciation of the costs of stillbirth is required and to date there have been no comprehensive estimates.

In this paper, the costs associated with stillbirths are described as direct (including the cost of medical care) or indirect financial costs (such as welfare payments). Outcomes are divided into psychological and social impacts on bereaved parents and families [9] and impacts on health professionals. We determine these costs and outcomes through a series of systematic reviews (see Panel 2) and new analyses of published and unpublished data. We also evaluate interventions to prevent stillbirths and to reduce negative impacts. To address the cost-effectiveness of these interventions we consider the effects of different methods for valuing the loss of fetal life.

Direct financial costs of stillbirth

Direct costs, including investigations of the cause of death, ranged from $1,450 [10] and £1,951 [11] to $8,067.[12] In comparison to a live birth, care costs for stillbirth were 10-70% greater.[11, 12] The direct costs of healthcare provision were typically met by government or insurance companies, although in some cases they were passed on to parents. Analysis of the international questionnaire undertaken

for this Series, including responses from 3,503 bereaved parents from HICs and 680 from MICs, indicates that 14% of respondents from HICs and 32% from MICs had medical costs to meet during and after the birth (see supplementary information) No data were recorded from LICs. Where reported, parents paid between $197 - $3,093 for investigations to determine the cause of stillbirth; and $118 - $20,000 in hospital fees for additional medical care (see supplementary information).

Three papers, all from HICs, recommended additional monitoring in subsequent pregnancies following stillbirth.[13-15] Estimated costs of care in subsequent pregnancies varied from £3,499 after a stillbirth of non-recurrent cause to £4,057 for a stillbirth of unknown cause.[11] A pregnancy after stillbirth costs £558 - £1,735 more than if the prior pregnancy ended in an uncomplicated live birth. If care included more intensive surveillance with cardiotocography, costs rose to £4,654 - £5,616.[16, 17] Thus, the costs of subsequent pregnancy care add to healthcare costs associated with stillbirths in HICs which will extend to MICs as they scale-up more intensive antenatal monitoring and care.

Indirect financial costs of stillbirth

The most frequently incurred indirect costs for parents after stillbirth were for the funeral and burial or cremation of their baby (see supplementary information). For some, this was mitigated by insurance, government payments or grants. Parents' free text responses in the International Stillbirth Alliance (ISA) survey reveal the significant financial burden placed upon them, magnifying the impact of their loss (see Panel 3). Although some parents did not have to pay, others reported costs for funerals ranged from $469 - $11,719, extending to $1,179 - $11,605 for burial plots and $1,410 - $4,605 for memorials. The theme that occurred most frequently in the free-text responses was the long-term financial impact on families. For many parents, stillbirth was associated with reduced earnings from employment or an inability to return to paid employment. Meeting the on-going costs of counselling and medical care in further pregnancies was also cited.

The experience of stillbirth also affected parents' employment, with 10% of bereaved parents remaining off work for 6 months and 38% of mothers and 21% of partners reducing their working hours (see Panel 3). Even after returning to work, productivity was greatly reduced, with estimates of 26% of normal work after 30 days, increasing to 63% after 6 months. Searches of the International Labour Organization database revealed that only 12 of the 170 countries with maternity benefit policies included specific provision for stillbirth; 11 for mothers (28-84 days leave) and one for fathers (5 days leave). Even in the few countries with such leave provision, bereaved parents seem to have little option to delay their return to work. Policies relating to stillbirth or miscarriage were found from five African countries (9.8% of countries in the region), five in Asia (17.9%), three in Europe (6.4%), and four in the Americas (11.8%, see Figure 5, supplementary information). In addition to the specific rights identified here, costs may also be incurred by governments where countries extend maternity rights to the parents of a stillborn child.

Psychological and Social impact of stillbirth

The period after stillbirth has wide ranging consequences for parents and their families. Much of the impact may be non-monetary, reflecting the negative impact of grief, anxiety, fear and suffering. These have been described as 'intangible' costs.[18] From our systematic review and meta-ethnography we identified ten themes in this area (see supplementary information).[19] Data were extracted to address eight of these themes from three questionnaire studies (the Listening to Parents study, [20] the TEARS study [21] and the International Stillbirth Alliance (ISA) survey) including a total of 5,358 parents from HICs and MICs. These themes included: negative psychological feelings after stillbirth; effects on relationships with others; the duration of these effects; how soon after the stillbirth parents returned to their previous routine and on returning to work how soon parents returned to a full productive capacity; the nature, adequacy and effectiveness of any supportive measures and whether parents sought medical treatment for any negative effects connected with the stillbirth.

Almost all parents in these studies report negative psychological symptoms following a stillbirth. In the Listening to Parents study (UK, n=473) 68% of mothers and 44% of partners reported four or more symptoms at 10 days, falling to 35% of mothers and 13% of partners at 9 months.[20] This is over three times greater than following a live birth, when 8-13% of mothers and 3% of fathers report depressive symptoms around 9 months postnatal.[22-24]

Family was the most frequently cited source of support for parents following a stillbirth, although family input was not universally positive (Panel 3). This need for support between parents and the wider family could place strain on relationships. In the Listening to Parents study, 9% of mothers and 5% of partners reported difficulties in their relationship 9 months after the event, and a similar proportion reported problems with other family members (12% of mothers and 4% of partners).[20] In the TEARS cohort (USA, n=216), the mean Family Assessment Device score of respondents was 3.2 (range 0.5-4.0) where a score of 4 indicates significant dysfunction in family relationships.[25] Ultimately, this may lead to relationship breakdown, which some studies report as more frequent in parents who have a stillborn child compared to a live birth (odds ratio 1.40, 95% confidence interval: 1.10-1.79).[26] In other studies divorce is unchanged, but relationship quality with their partner altered between married and single women.[27]

Systematic searching located 1,082 relevant data points from 144 studies of the psychological impact of stillbirth (see supplementary information for references). These were summarised into 23 themes (*shown in italics*) and thematic sentences with variable frequency effect sizes (Table 1). The most frequently reported experiences following stillbirth were *negative psychological symptoms* with high rates of depressive symptoms, anxiety, post-traumatic stress, suicidal ideation, panic and phobias.[28, 29] Although the majority of studies evaluated these symptoms subjectively rather than with a formal clinical diagnosis, 60-70% of grieving mothers in HICs experience significant grief-related depressive symptoms one year after the death.[25, 30] These symptoms endured for at least four years post-loss in about half of cases. If these figures are extrapolated to the 2.7 million women

who experience a stillbirth per year,[1] the estimated number of affected women living with depressive symptoms after stillbirth is 4.2 million. Many parents showed persistent feelings of remorse or guilt for not being able to save their child. Nearly 40% of grieving mothers in a convenience-sample survey in the USA were prescribed psychiatric medication despite a lack of evidence for its efficacy in this population.[21] Parents responding to the ISA survey reported accessing internet forums (>85%), support groups (~30%), or consultation with religious leaders (~30%) or health care professionals (~55%) to address their psychological symptoms. There was little difference between parents responding from HICs and MICs (see supplementary information).

Psychological distress *persisted into subsequent pregnancy* when parents reported conflicting emotions (relief and worry, hopeful optimism and panic attacks or depressive symptoms).[31] Women tended to report volatile emotional states, whereas fathers tended to report suppression of their feelings. Both parents were afraid to prepare for the birth of their subsequent baby, and avoided general antenatal classes as they felt they were outside the boundaries of 'normality'. Some women even struggled to differentiate their dead baby's identity from their subsequently-born live baby.

The capacity to express and integrate grief reactions was a critical part of parents' psychological responses. Many studies described *disenfranchised grief*, when parents felt their grief was not legitimised or accepted by health professionals, family or society.[32-34] This was particularly evident in LMICs, in cultures where talking about death is taboo, and where the dead baby was not yet deemed to be a person.[35-37] In these contexts, mothers' accounts indicated that they *suppress grief in public,* instead dealing with it privately and alone.[38, 39] These accounts are supported by responses to the ISA survey from care providers (LMIC n=117, HIC n=2,020). Fewer care providers from LMICs agreed that a death prior to birth is the same as the death of a child (19% vs 33%) and more attributed it to a mother's fault (0.5% vs 4%) compared to HICs. Respondents from LMICs more frequently agreed that parents should forget about their stillborn baby and have another child (26% vs. 3%) and should not talk about their stillborn child (12% vs 4%) compared to HICs (see supplementary information).

Fathers reported feeling unacknowledged as a legitimately grieving parent. The burden of keeping feelings to themselves increased the risk of chronic grief.[40] Differences in the grieving process between parents can lead to *incongruent grief,*[41, 42] which was reported to cause serious relationship issues, from conflicts around sexual intercourse to marital breakdown.[26, 43] As highlighted above, while family and friends were often essential for effective support,[44] respondents to some studies reported that family members had unrealistic, unhelpful expectations of recovery following stillbirth.

Many studies described the *adverse impact of stillbirth on siblings, a surviving twin and subsequent children*, including problems with parent-child relationships which could affect siblings' physical and mental health in the longer term.[45, 46] Some parents described anxiety regarding children of other parents.[47] Stillbirth was reported to have adversely affected the emotional wellbeing of grandparents and other family members.[48]

For some mothers, stillbirth affected their *approach to life and death, self-esteem, and their own identity*.[34, 49, 50] Some reported losing their sense of control, including in subsequent pregnancies, and their confidence in parenthood and child-rearing. Some women *avoided* contact with babies, creating social isolation and worsening depressive symptoms.[28] They were hesitant to meet their neighbours or those who had known them when they were pregnant. Many stopped going out, leading to voluntary social isolation. Social isolation could also be involuntary, with parents reporting *stigmatisation*, resulting in them feeling less valued as members of society.[51] In reports from some LMICs, women reported being significantly less valued by partners, families and society. In extreme circumstances, this led to spousal abuse, enforced divorce and rejection by family and society, partly based on beliefs that women who experience stillbirth are possessed by evil spirits, or have procured abortions.[36, 37, 51-53]

In the period shortly after the stillbirth, *altered body image* was important.[43, 54] Some women reported being embarrassed by their post-pregnant body. In contrast, others wanted to keep a pregnant form, maintaining a connection with their baby. Others linked the grief to their physical body through pain and by developing an image of themselves as unattractive and ugly. Such negative self-perceptions decreased sexual activity and pleasure. Women reported *pressures to delay or prioritise conception* originating from themselves or from family and society.[28, 55]

Chronic pain and fatigue, increased substance use, employment difficulties, and *financial debt* were also reported. Some studies described a long-lasting negative impact on quality of life.[41, 55, 56]

The consequences of a stillbirth were not exclusively negative. Some couples reported becoming closer.[41] Parental pride was reported by some parents after contact with their baby.[29, 57, 58] For others, deciding to see or hold their baby brought a sense of finality which contributed to the grieving process.[59] Some parents engaged in *therapeutic activities*. They sought solitude, changed their uptake of religious practice, and there were changes in their approach to sexual intercourse or engagement with health promoting activities, work and social media.[28, 33, 43, 60, 61] Some parents campaigned for, and contributed to, health service improvements to help other families and many parents changed the way they accessed *healthcare services*, especially in subsequent pregnancies when fathers became more involved.[40, 62]

Impact of stillbirth on professionals

All of the 20 studies included in the systematic review of the impact on professionals undertaken for this paper (see supplementary information) documented a substantial personal and professional burden for staff involved with caring for families during and following stillbirth. Four themes emerged from the data: *psychological impact, professional impact, need for support,* and *positive effects for staff* (Table 1). Psychological impact was most frequently reported as somatic, including trauma symptomatology, diminished emotional availability, stress, and affective states such as guilt,

anger, blame, anxiety and sadness. The professional impact of stillbirth was characterized by fear of litigation and of disciplinary action. In one study, data from LMICs suggested that attending a woman who experiences a stillbirth could result in 'loss of livelihood' and 'public humiliation'.

The majority of studies (n=13) highlighted the need for further education and professional support for staff, especially in terms of the psychosocial care and communication skills required following stillbirth. Many studies suggested that peer support was valuable when it occurred, even though it was usually informal. The lack of structured institutional and peer support was highlighted. Quantitative studies revealed the risk of vicarious traumatic stress, and depressive and psychological symptoms such as guilt, self-blame, self-doubt and grief. Importantly, those who felt they had received adequate training in stillbirth care were less likely to report guilt and fear of litigation.

In six studies, staff also reported experiencing some positive gains , such as feeling a sense of 'honour' or 'privilege' at being able to support parents experiencing the death of their baby.[63-68] Some staff also cited 'personal growth' and the development of a 'special bond' with parents and staff. In four studies staff reported more confidence and comfort, with fewer negative effects, when they had more direct clinical experience with stillbirth.

These findings indicate that, while mothers, partners, and their families bear the brunt of the effects of stillbirth, it also has a considerable impact on healthcare providers. The negative effects could be addressed by education, training, and provision of formal and informal support during and after such an event, and by enabling positive experiences of caring for parents after stillbirth.

Interventions to maximise wellbeing for bereaved parents and families – What works?

Forty-three studies provided evidence on what works to reduce the negative impact of stillbirths (see supplementary information). Two systematic reviews of randomised controlled trials (one on social support and the other on autopsy) did not locate any studies.[69, 70] No other RCTs were located. No intervention studies were found for Africa, Asia, or the Middle East. Of the 16 studies that

directly assessed interventions, ten included mothers only, one fathers only, one parents and care providers, and the remaining four included parents and/or the wider family.

Effective interventions (HIC settings) included: seeing and holding the baby, social support/support groups, making and sharing memories, autopsy, psychological interventions, and interventions with multiple components (see supplementary information). [71-76] Professional support to enable parents to share their experiences with others, and social support from family and local social networks were both associated with lower rates of depression and better mental health.[73] A specific psychological intervention in Brazil was associated with a range of positive effects, finding that including family members reinforced network support.[77] Support groups in a US study was associated with significant improvement in scores on the Impact of Events Scale (IES-R).[73] Programmes with multiple components generally increased parents' satisfaction, with those more satisfied reporting less grief.[73-75, 78] Where measured longitudinally, this effect was maintained for up to 2 years. Finnish fathers receiving a multiple component intervention reported stronger personal growth and less blame and anger.[78]

The key findings of all included studies (qualitative and quantitative) were mapped to Sarafino's taxonomy of social support. This comprises five support elements: Tangible, Emotional, Esteem, Informational and Network/belonging (see Supplementary information).[79]

All effective interventions, and all qualitative studies of interventions with positive participant responses, included emotional support. Nine included informational support, and ten addressed tangible support. Usually, this was help from staff to see and hold the baby after birth (14 studies, including HICs and LMICs). Two studies included esteem support, such as help with reclaiming a lost sense of motherhood or fatherhood. Eight involved networking/belonging. Positive staff attitude was universally appreciated.

Data pertaining to specific groups were only available from HICs. This demonstrated that fathers, siblings and female partners need to be acknowledged and included in interventions, to mitigate their experiences of the negative effects of stillbirth. Interventions for siblings need to be tailored for their age and maturity. The need for esteem support for family members was particularly apparent, including recognition of continuing status as father, co-mother, sister, grandparent or brother, even after the death of the baby that created these social roles.

Variation in access to what works by cultural context

Importantly, access to support groups or services is not equitable. In the three parents' surveys that analysed in this paper, 54-93% of parents in HICs were given information about support groups or services compared to 12% of parents in MICs. Information about grief and psychological symptoms was given less frequently than for physical symptoms in MICs (16% vs. 28% respectively) but not HICs (52% vs. 47%). The perceived effectiveness of support groups varied, but 77% of respondents to the ISA Survey who used a group reported benefit, with comments such as "s*upport group was key in our grief journey.....identifying with other parents that had experienced it was the only way we knew how to cope with it "* (#2852, USA). Lower amounts of support available for parents in MICs might account in a greater proportion rating their follow-up care as poor compared to HICs (60 vs. 38%, see supplementary information).

In the systematic review of what works, all except two of 10 studies conducted in LMICs included only women. The only positive factors reported by respondents from Malawi were basic physical nursing care and brief information-giving, which were seen as surprising but welcome occurrences.[80] Indeed, as noted above, studies in Ethiopia, Tanzania, and India indicated that having a stillborn baby can lead to maternal abuse, social abandonment, and divorce.[35, 37, 52, 53] Despite feelings of grief and loss, mourning was actively discouraged and suppressed, and interventions such as seeing and holding, and taking of mementoes, were not culturally acceptable. This was echoed in care providers' responses to the ISA survey (LMIC n = 117, HIC n=2,020) who reported that parents in

LMICs were less likely to be offered contact with their baby (35% vs 94%), the opportunity to see and hold their baby (42% vs 95%), make memories (35% vs 87%), and name their baby (39% vs 83%) after a stillbirth compared to parents in HICs.

The main support mechanisms in the included LMIC studies in the review data were reported to be family and local religious communities, rather than health care professionals and wider society. In these contexts, interventions designed to improve emotional and informational support may depend on enhancement of community esteem for those who have experienced stillbirth, especially through key religious groups. Networking and belonging support interventions could be primary mechanisms for improving women's wellbeing following stillbirth in LMICs.

Summary of what works

Based on these data, the key element of 'what works' in reducing the impact of stillbirth on bereaved parents and families can be summarised as '*seeing through the eyes of those affected*'. This includes staff who understand what different parents and families need and when they need it; communities that acknowledge grief and loss, and that does not stigmatise those who experience stillbirth; employers who provide effective leave arrangements; and governments that provide tangible support, such as funeral costs, and paid leave from work commitments.

Understanding the Impact of Stillbirth

Stillbirth is associated with significant direct, indirect and intangible costs to women, and to their families, society and government (Figure 1). These include: medical care and investigations at the time of stillbirth and in subsequent pregnancies; funeral costs; grief and negative psychological impact; reduced social functioning; family and relationship disruption and breakdown; and negative effects on employment (Figure 1). The impact of stillbirth is enduring, with consequences persisting for years. Similar issues, particularly direct healthcare and funeral costs [81] and the enduring impact on family function have been described for maternal death.[82-85] In addition to families, the impact

upon staff and consequent implications for staff wellbeing and future service quality and delivery must be considered. Depending on the setting, costs may be met by government, insurance companies, or individuals and their families. Prior to the reviews and analyses presented here, these various costs of stillbirth have not been considered together. We argue that this has led to an underestimation of the economic, social, emotional, and psychological burden of stillbirth.

The global impact of stillbirth: addressing research gaps

Our systematic approach has demonstrated large gaps in available data regarding costs and interventions which may reduce the burden of stillbirth by preventing stillbirths or their negative consequences. Few studies determined the direct costs of stillbirth in the perinatal period or subsequent pregnancy; all were from HICs. Studies that report on the intangible costs of stillbirth or practices that might reduce its' negative consequences are concentrated in fewer low-burden HICs (n=177), with little or no data available from LMICs with high-burden of stillbirth (n=26, Figure 3 in Supplementary File). As most of the components of effective care were identified from studies conducted in HICs, the data obtained are similar to a recent review restricted to HICs.[86] While some of the themes identified are consistent between HICs and LMICs, others, such as stigma and social isolation, appear to be particularly relevant in LMICs.[37, 39, 55, 87-90] Therefore, to appreciate the full cost of stillbirth tailored research is urgently needed to establish direct and intangible costs of stillbirth, particularly in LMICs and amongst marginalised women and their families.

In all settings, there is very little information about what works for fathers/partners and other family members. Substantial comparative research on effective interventions to mitigate the effects of stillbirth is missing in all contexts. Where evidence does exist, effective care appears to include emotional, informational and, to an extent, tangible support, in terms of practical or financial help, at and around the time of diagnosis and birth. Based on questionnaire data, parents greatly valued support to help with direct financial costs (such as funeral arrangements) when it was provided by governments or insurance schemes.

There has also been less emphasis in intervention studies on networking and belonging support, and almost none on esteem support. In all settings, but particularly in LMICs, these components may form a basis for addressing stigma, taboos and social rejection for bereaved mothers. Fear of loss of esteem and of exclusion from social networks has the potential to stifle attempts to allow women to express and to deal with their grief, potentially leading to long-term costs. In contrast, where local family and social (notably religious) networks were supportive, mothers, in particular, reported positive benefits. Indeed, some parents, and some staff (in both LMICs and HICs) believed that they had grown spiritually, and had gained significant coping skills as a consequence of their experience. Acknowledgement of the personal and professional cost of stillbirth on staff is essential, both for their personal wellbeing, and to enable them to deliver what works to bereaved parents.

In LMICs, which have the greatest burden of stillbirth, an intervention that addresses stillbirth at a healthcare, societal and community level could make two major gains. The first could be the adoption of preventative measures, including improved health messaging, monitoring, support and care for women pre-pregnancy, antenatally and during delivery, improving the health of both mother and her baby. The second could be destigmatisation of stillbirth, thereby reducing the negative consequences, especially for women.

Interpreting the Value of Stillbirth

Given the gaps identified, comprehensive estimates of the costs of stillbirth cannot currently be derived to inform cost-effectiveness analyses. Data on the financial costs of the sequelae of stillbirth is not routinely collected in any country. Wide variation in monetary and opportunity costs between different countries, such as those relevant to health care provision or lost labour productivity, mean that such data must be local if it is to be meaningful. Data on the intangible costs is also limited, particularly with regard to LMIC settings, fathers, the wider family, and healthcare providers. Finally, any cost-effectiveness analysis must include a decision on how the loss of life to the baby is to be

valued. Consequently, any attempt to assign a global cost to stillbirth—in monetary terms or using summary measures of health like QALYs and DALYs — would presently be misleading.

Despite the substantial costs of stillbirth set out in this manuscript, the magnitude of the total loss associated with stillbirth is substantially affected by whether the stillbirth is also counted as a loss in its own right (i.e. as a loss to the baby). Economic evaluations of interventions to prevent stillbirths have to make the critical decision of whether and how to count this loss. Comparable evaluations of interventions to reduce neonatal mortality typically present results based on the time-discounted life expectancy of surviving infants.[91] To avoid interventions to prevent stillbirth from being undervalued they should be assessed in the same way.[92, 93] It should be emphasized that this does not entail that women's rights over their bodies, including rights to terminate pregnancy, should be restricted

The use of QALYs in NICE guidance and earlier iterations of DALYs apply discounting techniques to accommodate time-discounting of future benefits, giving, 25 QALYs lost or 32.4 DALYs associated with stillbirth.[94, 95] The appropriateness of time-discounting of health benefits is the subject of debate. Without discounting stillbirth would be associated with 86 DALYs on account of the loss to the baby. Other approaches to estimating the value of early lives lost include Jamison et al.'s suggestion that deaths before age 2 should be adjusted according to degree of cognitive development or "acquired life potential". With time-discounting this gives DALY values of between 5 and 9, without time-discounting this would give DALY values of between 14 and 26. Thus, proposals for how to value the life of a stillborn baby vary dramatically. How it is valued can make a difference of several orders of magnitude to the overall loss attributable to the stillbirth (see Figure 2, supplementary file). For example, a study of the cost-effectiveness of a syphilis screening program for pregnant women in Mwanza City, Tanzania estimated a cost of $92.56 per DALY averted without including stillbirths and $8.88 per DALY averted if they were included as a loss to the deceased.[95]

Despite the gaps in the evidence, the findings in this paper suggest that the burden of stillbirths is substantial yet greatly underappreciated. This undervaluation may contribute to the slow pace of change to address stillbirth on national and international platforms, as identified by Frøen et al. [96] Critically, while the costs of preventing stillbirth may appear considerable in both LMICs and HICs, the combined direct, indirect and intangible costs of stillbirth are almost certainly greater still. We call upon the global community to recognise the enduring impact of stillbirth on parents, families, staff, societies, and health and social care systems: to develop strategies to collect data regarding the cost of stillbirth; and to use that information to invest in strategies and local services and practices to prevent stillbirth and interventions to reduce the negative impact of stillbirth.

Authorship Statement

AH was responsible for overall coordination and oversight of the review and writing process; JM and TR for modelling regarding value(s) assigned to stillbirth. AH and TR for the systematic review of economic studies, direct costs and costs in subsequent pregnancies. VF and AW for the design and analysis of international questionnaire. AH and MR for analysis of published questionnaire data. HB for searches of ILO databases. DS, CS for systematic review of psychological impact upon parents. JC, KG, DN, KO'D for systematic review of psychological impact on professionals. SD and OM for systematic review of interventions to ameliorate impact on parents. ND and TS for international context; JD, AR, ZAB for LiST analysis. All named authors contributed to the conceptualisation, development, writing, and finalisation of the paper. AH is the overall guarantor.

Conflict of Interest

Dr. Blencowe reports grants from Save the Children/ Saving Newborn Lives during the conduct of the study; Dr. Siassakos reports grants from Stillbirth and Neonatal Death charity (Sands) during the conduct of the study; grants from Sands and membership of International Stillbirth Alliance and the executive committee of the Stillbirth Clinical Study Group, UK (RCOG & Sands), Department of Health Stillbirth task-and-finish groups and PROMPT maternity foundation. Dr. Heazell reports grants from Tommy's during the conduct of the study and grants from Sands, Holly Martin Stillbirth Research Fund outside the study. Dr. Heazell is currently chair of the board International Stillbirth Alliance and the executive committee of the Stillbirth Clinical Study Group, UK (RCOG & Sands), Department of Health Stillbirth task-and-finish groups. Ms Storey reports grants from Sands during the conduct of the study.

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Table 1 – Thematic sentences derived from meta-synthesis of studies assessing psychological impact of stillbirth on parents and on healthcare professionals. Frequency effect sizes and representative quotes are shown for each theme. Frequency effect size = the proportion of included reports containing a theme.

Thematic sentences derived from meta-synthesis of studies assessing psychological impact of stillbirth on parents				
THEMATIC SENTENCE	**Frequency Effect Size %**	**Example Quotes**	**Reference**	**Country**
Stillbirth has been associated with a number of emotional/psychological symptoms.	77.1	'I am depressed, saddened, hurt, empty, guilty and lonely. I cry every day. I will mourn him forever.' A number of mothers recalled suicidal thoughts because of their desire to be with their baby.	Lee 2012 [29] Kavanaugh 2005[28]	Australia USA
Parental grief following stillbirth may not legitimised by health professionals, family & society (disenfranchised grief).	31.2	Women shared their distress that their motherhood of their dead babies was denied by others. One participant recounted that when she told her sister she was not sure she was ready for Mother's Day rituals, her sister replied "Well, you're not a mother – you have to have your baby first." This perceived lack of social understanding left these mothers alone and uncomforted. Added to this, the silence was aggravated by the failure of friends and family to acknowledge the loss and grief as real. They experienced people avoiding them, or treating them as though they had never been a mother. 'Women who have not gone through stillbirth don't want to hear about my birth, or what my daughter looked like, or anything about my experience.'	Cote-Arsenault 2004 [32] St John 2006 [34] Gold 2012 [33]	USA Australia USA

Stillbirth may have a positive or negative impact on relationships, for example through different grief reactions (incongruent).	28.5	Some women felt their husbands did not show any sadness and were impatient with them, they felt their relationship had changed; stillbirth had created a distance between them. Mothers and fathers stated that they became closer after the loss, and that the feeling deepened over the course of the following year. They had something in common; going through the loss together – a sense of experiencing a special unifying bond.	Tseng 2014, [42] Avelin 2013 [41]	Taiwan Sweden
In subsequent pregnancy some parents may experience psychological distress.	27.1	…fathers exhibited great emotion as they shared the burden of worry over what was going on at home. They had difficulty concentrating at work and called home frequently, asking the mother to validate fetal movements. 'You're happy that you are [pregnant] but you can't be that innocent… Am I confident? No. Will I relax? No. There is not a point that I will relax until they are out and breathing...'	O'Leary 2006 [61] Cote-Arsenault 2004 [32]	USA USA
Stillbirth may change parents approach to life and death, self-esteem, own identity, and sense of control in subsequent pregnancy, parenthood and childrearing.	25.7	…The thoughts expressed by parents in our study consisted of being more humble and more grateful toward life itself and taking nothing for granted. The men in the study also questioned their identity as fathers, uncertain as to their right to the term 'father'. Each woman struggled with her sense of identity. Although each felt she was a mother, she was a mother without a child, and did not have tangible evidence of her motherhood.	Saflund 2004 [50] McCreight 2004 [49] St John 2006 [34]	Sweden UK Australia
Stillbirth can have an adverse impact on siblings, including the surviving twin, and subsequent children.	23.6	Older siblings from the ages of 7-12 years were described as being worried, nervous, tense and silent. They were worried about life and their parents' health. Infants next-born after a stillbirth were significantly more likely to be classified as disorganised in their attachment	Erlandsson 2010 [45] Hughes 2001 [46]	Sweden UK

		behaviour with their mothers than controls, this was strongly predicted by unresolved mourning in the mothers.		
After stillbirth some parents may seek isolation, can change their uptake of religious practice, approach to sexual intercourse, engagement with health promoting activities, work and social media and this may continue into subsequent pregnancies.	20.1	The fathers in this study were exhausted, physically and emotionally. When asked to say more about how they managed, a common response was 'I keep myself busy'. Men looked at sex as a tension reliever and attributed a therapeutic value to it. Many parents relied on their spirituality to deal with their loss. For some parents this was in the form of praying; for others, it was going to church. 'I cry when I talk to a real person so it was easier to talk to someone online, less emotional'.	O'Leary 2006 [61] Dyregrov 2011 [43] Kavanaugh 2005 [28] Gold 2012 [33]	USA Norway USA USA
Some parents feel the need to suppress outward grief, including during subsequent pregnancy.	18.1	Fathers felt they denied their own emotional reactions in order to protect and support and care for their partners. According to Taiwan's culture talking about death is a taboo subject and these mothers often dealt with their grief privately and alone. 'I think I genuinely suppressed a lot of my anxiety because of my [desire to protect my] family. Yes, I wanted to stay strong for my husband and myself. Outward I was strong but inside I was a mess'.	Armstrong 2001 [40] Sun 2011 [39] Cote-Arsenault 2011 [38]	USA Taiwan USA
Stillbirth may lead to avoidance of activities that remind them of the pregnancy and the baby.	13.2	Most mothers found it very difficult to be in situations that reminded them of "what could have been." Examples of these situations were being around pregnant women or infants, attending baby showers, and celebrating holidays.	Kavanaugh 2005 [28]	USA
Parents report stigmatisation, rejection, and spousal abuse.	13.2	'There were a few people at work who just never spoke to me again…I mean I definitely got the feeling… like I was bad	Murphy 2012 [51]	UK

		luck'. 'Every time I walked into the living room, my in-laws lowered their voices. Mostly, they stopped talking. I disappointed them because I didn't give them a descendent like every daughter-in-law should do. I felt unwomanly, since I failed to have a baby.' 'I know a girl who was in school and married off by her parents. After the marriage, she repeatedly lost her new-borns and was divorced. Not to face the humiliation in the village she ran away to a city and now she is a commercial sex worker.'	Hsu 2004 [36] Sisay 2014 [37]	Taiwan Ethiopia
Parents may have mixed feelings towards the decisions they made e.g. post mortem or seeing/holding their baby.	12.5	In the limited time available for mothers to meet the child, mothers did not know how to spend time with their child, and had multiple hesitations due to their child being dead, and regretted this later on.	Akiko 2009 [57]	Japan
Parents may experience external or internal pressures to prioritise or delay conception.	9	Some mothers did not plan on a subsequent pregnancy because of their concern about their ability to deal with another perinatal loss. Perinatal loss signalled a potential underlying health problem, which in turn accentuated anxieties relating to both future reproductive abilities and investment of limited resources on another potentially unsuccessful pregnancy. Such women described feeling pressure to prove their reproductive capabilities as soon as possible.	Kavanaugh 2005 [28] Fottrell 2010 [55]	USA Benin
Bereaved parents may become hypervigilant with siblings and subsequent children, and anxious about other people's children.	7.6	All mothers shared stories of feeling out of control, especially when faced with 'normal' or 'common' childhood events, such as tonsillitis, middle ear infection or being stung by a bee. These events were enough to cause them to feel hysteria and intense fear they were about to lose another	Warland 2011 [47]	Australia

		child.		
Bereaved parents may increase or decrease their use of health care services; and in subsequent pregnancy fathers express a desire to be more included in care.	6.9	Mothers with a history of prior perinatal loss may attempt to cope with their anxiety in pregnancy and depression in early postpartum with requests for additional healthcare resources. Fathers felt the need to take more interest or active involvement in the subsequent pregnancies.	Hutti 2011 [62] Armstrong 2001 [40]	USA USA
Chronic pain & fatigue can follow stillbirth.	6.9	Three months after the loss both mothers and fathers responded to grief most usually with tears; men also reacted with anger, irritation, silence, and one mother reacted most frequently with physical pain.	Avelin 2013 [41]	Sweden
Some parents described parental pride after the birth of their stillborn baby.	5.6	'Even though it wasn't the outcome I wanted, I loved giving birth to my son. It was a beautiful experience and how I wanted it.' Virtually every mother in this study felt tenderness and warmth when they held their baby… this supports the belief that the mother attaches to her new-born even if the baby is dead. There were parents who described a surging feeling of love from the moment they saw their child.	Lee 2012 [29] Radestad 2009 [58] Akiko 2009 [57]	Australia Sweden Japan
Potential impact includes employment difficulties and financial debt.	5.6	Together with sustained difficulties in paying off hospital bills, this strained relations with family members from whom funds had been borrowed.	Fottrell 2010 [55]	Benin
Stillbirth can motivate parents to engage with healthcare	4.2	'I deal with it in a way that you know, to crusade, to campaign, to make sure things change, to try and take the	Murphy 2012 [51]	UK

improvement including public awareness.		positives as much as you can out of the whole situation'.		
Increased substance use has been reported for some parents.	4.2	They always were social drinkers but after Ricky died Mom increased her drinking	O'Leary 2013 [56]	USA
Women may develop a complex emotional response to body image.	3.5	Some women also found that their own body reminded them of their loss… their body was a bearer of both pain and memories. They could feel intense pain in their body, feel physically exhausted and sense that their body was against them. Women were embarrassed/guilty of their post pregnant body as they did not have a baby, conversely some women wanted to keep their body in a pregnant shape to not let go of the baby.	Dyregrov 2011 [43] Huberty 2014 [54]	Norway USA
Stillbirth has an adverse effect on the wider family.	2.8	This sense of constrained grieving caused by social discomfort and taboo extended to husbands and grandparents, who were not expected to grieve the loss of a stillborn baby beyond feeling some transient disappointment or sadness for their wife or daughter.	Kelley 2012 [48]	USA
For some parents quality of life might be affected in the long term.	2.1	Women with histories of fetal death seem to have poorer quality of life.	Couto 2009 [97]	Brazil
Some couples experience conflicting emotional reactions to sexual relationships.	1.4	Women also wrote somewhat more often than men about increased activity in order to have another child, as well as sex being used for comfort closeness and tension reduction. Women more frequently reported disturbing images, thoughts and feelings that interfered with sex than did men.	Dyregrov 2011 [43] Dyregrov 2011 [43]	Norway Norway

Themes and effect sizes derived from analysis of studies assessing psychological impact of stillbirth on professionals

THEME	Frequency Effect Size %	Example Quotes	Reference	Country
Stillbirth has powerful psychological impacts on professionals.	95			
Emotional response/ distancing	*40*	"I think it's possible to experience too much grief in this work"	Nuzum 2014[85]	Ireland
Trauma	*42*			
Guilt	*35*			
Anger	*30*	"It is a mixture of everything, anxiety, rage, oppression, impotence…"	Pastor Montero 2011[84]	Spain
Fear	*30*			
Stress	*30*			
Anxiety	*25*	"… I had to cut off my emotions to just get through it	Puia 2011[37]	USA
Blame	*20*			
Depression	*20*	"It shook me to my core"	Puia 2011[37]	USA
Frustration	*15*	"… you've got anger, huge anger, especially where a mistake has been made or something has been missed"	Nuzum 2014[85]	Ireland
Sadness	*15*			
Powerlessness	*10*			
Challenge to faith	*5*	"It sort of haunted me for a couple of days. …I had some issues falling asleep that night and getting the images out of my head"	McKenna 2011[36]	Australia
Humiliation	*5*			
Stillbirth has a professional impact.	65			
Effect of litigation	*30*	"Is this the one that is going to blame you?"	Kelley 2012[82]	USA
Fear of disciplinary action	*10*			
Fear of public censure	*5*	"If you … lose a mother or a baby, you will lose your license, your income, your work"	McCool 2009[86]	USA
Exposure	*5*			

Professionals need support *Education* *Peer support* *Institutional support*	65 *30* *5* *5*	"I think what would be helpful … is having that debriefing time after it's over and not being directly assigned" "… they do not teach you the necessary strategies to provide support in these situations" "… we need to support each other and not tear each other down"	Jonas Simpson 2010[34] Pastor Montero 2011[84] Roehrs 2008[38]	Canada Spain USA
Stillbirth can have a positive impact on professionals *Benefit of experience* *Sense of honour* *Privilege* *Special bond with parents* *Making a difference*	30 *20* *10* *5* *5* *5*	"I think having that experience, I've grown as a person" "I feel like I make a difference, and if I can ease their pain I am happy"	Jonas Simpson 2010[34] Steen 2015[39]	Canada USA

Figure Legends

Figure 1 – Schematic figure representing the impact of stillbirth originating with the death of the baby, affecting mother, parents, family, health services, society and government. Common themes of direct, indirect and intangible costs are shown.

Panel 1 – *Modelled Scenario - The impact and cost of 90% coverage for quality antenatal and intrapartum care*

We used Lives saved tool (LiST) (version 5.28) to model the impact of effective proven interventions on stillbirths and maternal and neonatal deaths. We modelled the potential effect of introducing selected interventions within health systems of the 75 high-burden Countdown countries (which account for 99% of all deaths). For each of the 75 Countdown countries, baseline scenarios were created that represent the most up-to-date details about health status, including mortality rates, cause of death structure and current coverage of interventions. The base year was set as 2015 and coverage of selected interventions was scaled up linearly to reach 90% by 2030. The modelled interventions were grouped into four packages along the continuum of care.

- Preconception nutrition care: Balanced energy protein supplementation, folic acid supplementation/fortification, and micronutrient supplementation [multiple micronutrients, including iron and folic acid].

- Basic antenatal care: Prevention of malaria with insecticide-treated bed nets or intermittent preventive treatment (IPTp) with antimalarial drugs, syphilis detection and treatment and tetanus toxoid immunization. IPTp was only scaled up in countries where malaria is endemic and the effect would only apply to the percentage of women exposed to malaria.

- Advanced antenatal care: Detection and management of hypertensive disease of pregnancy including treatment with magnesium sulphate and hospital care or caesarean section if needed, detection and management of diabetes in pregnancy, detection and management of fetal growth restriction, identification and induction of mothers with 41 weeks of gestation or more.

- Child birth care: Skilled birth attendance, antenatal steroids for preterm labour, antibiotics for pPROM, active management of the third stage of labour, neonatal resuscitation, immediate assessment and stimulation of the newborn.

For costing, we used the LiST costing sub-module to assess the running costs of the interventions for which we used an ingredients-based approach. The costing sub-module draws its assumptions about staffing, drugs, and need for services from the United Nation's OneHealth Tool database. We have only included running costs and that was divided in four components: capital costs, drug and supply costs, labour costs and other recurrent costs.

The results suggest that scaling up these proven antenatal and intrapartum interventions in the 75 high burden countries can prevent 823,000 stillbirths, 1,145,000 neonatal deaths and 166,000 maternal deaths annually by the year 2030 (see figure) at an additional annual running cost of 4.6 billion US$ or US$ 2,143 for each live saved (including stillbirth, maternal and neonatal deaths) (see table). The analysis suggests that interventions in the preconception, basic and advanced antenatal care packages are crucial, but most of the deaths including stillbirths and neonatal and maternal deaths are prevented by intervening in the intrapartum period alone and with a lower estimated cost of US$ 1,370 to save each life. This analysis reaffirms previous estimates that not only prevention of stillbirths is possible but can be achieved at a reasonable cost of US$ 2,143 for each life saved.

<End of Panel 1>

Panel 2 – Search Strategy

To extend the knowledge base regarding direct, indirect and intangible costs of stillbirth a series of systematic reviews were performed and meta-syntheses were undertaken using established methods and search strategies.[11, 98, 99] A further systematic review and meta-synthesis was undertaken to identify interventions or systems that may reduce the negative impact of stillbirth (detailed methods can be found in supplementary information). The search strategy was designed to capture the whole field of studies worldwide; no language restrictions were imposed and searches were carried out in CINAHL, AJOL, LILACS, MEDLINE, PsycINFO, Cochrane and PubMed. These data were supplemented with data extracted from recent studies regarding the impact of perinatal death and two questionnaire surveys developed for the current Lancet Stillbirth series which contained specific questions regarding costs experienced by parents and families in HICs and MICs and experiences of care providers in HICs and LMICs. These surveys were distributed by the International Stillbirth Alliance. For the surveys, quantitative data were analysed using descriptive statistics, and free text responses were analysed by thematic analysis. Published cost estimates are reported in their original currency but presented in 2013 prices.[100, 101]

<END OF PANEL 2>

Panel 3 – Parents' experiences of the direct, indirect and intangible cost of stillbirth in HICs

Direct and Indirect Costs

It's difficult as I had already purchased all the baby items and then had additional tests to pay for. I wouldn't have minded them if my child lived, but having to pay for them after he died was difficult and a constant reminder as the bills kept coming (#3903, Australia).

I could not properly bury my child because I lacked the financial means; that hurts today, because I have no grave (#19342, Germany).

The higher cost, in financial terms, was the long process of psychotherapy that I followed in the next three years and more examinations that I had privately before and throughout the course of the next pregnancy (#11707, Italy).

Employment

The loss of income when you can't bring yourself to go back to work is substantial and many work places don't understand the pain (#7358, Australia).

Because neither I nor my husband was able to start work after the birth, we had no income. We could not get compensation from the social insurance because we were not sick we were just grieving (#26496, Sweden).

Financial support from family, friends and others

I never thought anything like this would happen, so I was unprepared! Glad I had family and friends to help give her a beautiful burial service which I could not afford on my own (#5582, UK).

The funeral home did not charge us for our daughter's cremation or vessel for her ashes. They told us that "we had already paid too much." We will always remember their kindness and compassion (#2295, USA).

Support from family

"My family and my friends were a great help to us. They were always there to listen and offer support when I needed it. They got me through a lot of the time" (#4583, Australia).

"My family was supportive at first. After a while they seemed to think I should get over the death of my twin; that I had grieved long enough" (#3159, USA).

Financial support from government

This is all paid for by the state. I am very happy for this. It is devastating enough losing your baby, without getting debt because of it, or having to consider if you can afford the help you need, or can afford a funeral, an autopsy etc. (#8516, Norway).

All medical expenses were covered by social security and burial expenses by insurance. The only expense was the grave (#19795, Spain).

<END OF PANEL 3>