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[PII_REPLACE]

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[A: We have edited your paper to avoid repetition, enhance readability, reduce length, and achieve consistency with *Lancet* style]

[A: we now have a Comment commissioned for the Series on the return on investment case for stillbirths. A different title for this paper might be warranted. Would "Stillbirth: economic and psychosocial consequences" work?]

Ending preventable stillbirths 3

Stillbirth: Why invest?

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Despite the frequency of stillbirths, the subsequent implications are overlooked and underappreciated. We present findings from comprehensive, systematic literature reviews, and new analyses of published and unpublished data, to establish the effect of stillbirth on parents, families, health-care providers, and societies worldwide. Data for direct costs of this event are sparse but suggest that a stillbirth needs more resources than a livebirth, both in the perinatal period and in additional surveillance during subsequent pregnancies. Indirect and intangible costs of stillbirth are extensive and are usually met by families alone. This issue is particularly onerous for those with few resources. Negative effects, particularly on parental mental health, might be moderated by empathic attitudes of care providers and tailored interventions. The value of the baby, as well as the associated costs for parents, families, care providers, communities, and society, should be considered to prevent stillbirths and reduce associated morbidity.

Introduction

Despite the 2·7 million stillbirths worldwide,¹ the costs of stillbirth are largely unknown and therefore 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 for the loss of life or the loss to parents and families. Most economic analyses have focused on the cost of stillbirth prevention.^{4,6,7} In low-income and middle-income countries (LMICs), costs vary from US\$4781 to \$10 571 per stillbirth averted (in 2013 prices).^{4,6} In high-income countries (HICs) with lower stillbirth rates, prevention costs are greater than are in LMICs, for example smoking cessation costs \$125 961 per stillbirth averted.⁸ If stillbirths are included in analyses of the effect of antenatal and intrapartum care on maternal and newborn deaths, the cost per death averted reduces substantially from \$27 551 to \$2143 (panel 1).⁴ However, to accurately assess whether these programmes are cost-effective, a better appreciation of the costs of stillbirth is needed and so far, no comprehensive estimates have been made.

In this Series 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 effects of bereaved parents and families,⁹ and [A: overall effects?] effects on health professionals. We identify these costs and outcomes through systematic reviews and new analyses of published and unpublished data (panel 2). We also evaluate interventions to reduce negative effects [A: such as?]. To address the cost-effectiveness of these interventions and those to prevent stillbirth, we consider the effects of different methods used to value the loss of fetal life. [A: please add a line in this paragraph about data being mostly from HICs and MICs/LMICs]

Direct financial costs of stillbirth

Three studies described direct costs, including investigations into the cause of death, ranged from \$1450,¹⁰ and £1951¹¹ to \$8067.¹² Care costs for stillbirths were 10–70% greater than with a livebirth.^{11,12} Direct costs of health-care provision were typically met by government or insurance companies, although in some cases this expenditure was passed on to parents; 14% of respondents from HICs and 32% from middle-income countries (MICs) had medical

Key messages

- Stillbirth is associated with substantial direct, indirect, and intangible costs to women, their partners and families, health-care providers, the government, and the wider society. Appreciation of the costs of stillbirth is essential to evaluate the cost-effectiveness of interventions to prevent stillbirth or ameliorate negative effects of stillbirth.
- Data for the cost of stillbirth in high-burden countries are inadequate. In addition to collection of data for the number of stillbirths, data should also be collected for the resource implications.
- Adverse experiences including stigma, social isolation, and disenfranchised grief are widespread among parents whose baby is stillborn and need to be addressed through focused interventions and supportive activities including parents, communities, care providers, and relevant stakeholders.
- Empathic behaviours during every encounter between bereaved parents and caregivers are essential to minimise additional emotional and psychological burdens in the short, medium, and long term.
- Caring for families during and after stillbirth places a substantial personal and professional burden on staff. Negative effects on staff could be addressed by education, training, and provision of formal and informal support.



This is the third in a Series of five papers on ending preventable stillbirths

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Panel 1: Modelled scenario—the effect and cost of 90% coverage for quality antenatal and intrapartum care

We used Lives Saved Tool (LiST) (version 5.28) to model the results 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 [A: please reference to specify these countries, eg in the appendix] (which account for 99% of all deaths) [A: Please provide a reference]. For each of the 75 Countdown countries, baseline scenarios were created that represent the most up-to-date details about the health status of these countries, including mortality, cause of death structure, and present 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 and protein supplementation, folic acid supplementation or fortification, and micronutrient supplementation (various micronutrients, including iron and folic acid).
- Basic antenatal care: prevention of malaria with insecticide-treated bednets or intermittent preventive treatment with antimalarial drugs, syphilis detection and treatment, and tetanus toxoid immunisation. Intermittent preventive treatment was only scaled up in countries where malaria is endemic and the effect would only apply to the proportion of women exposed to malaria.
- Advanced antenatal care: detection and management of hypertensive disorders 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 at 41 weeks of gestation or more.
- Child birth care: skilled birth attendance, antenatal steroids for preterm labour, antibiotics for preterm premature rupture of the membranes, active management of the third stage of labour, neonatal resuscitation, immediate assessment, and stimulation of the newborn.

For costing, we used the LiST costing submodule to assess the running costs of the interventions for which we used an ingredients-based approach, identifying and valuing every resource [A: added a qualifier of ingredients-based approach, ok?]. The costing submodule draws its assumptions about staffing, drugs, and need for services from the UN's OneHealth Tool database [A: please provide a ref that will be added in as a margin link]. 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 (figure [A: figure does not estimate these figures, are you referring to something in the appendix?]) at an additional annual running cost of US\$4.6 billion or \$2143 for each life saved (including stillbirth, maternal, and neonatal deaths; table [A: table does not relate directly to this sentence, please explain or okay to delete table citation?]). 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 \$1370 [A: lower than what?] to save each life. This analysis reaffirms previous estimates that not only is prevention of stillbirths possible but prevention can be achieved at a reasonable cost of \$2143 for each life saved.

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centers to meet during and after the birth. Where reported, parents paid between \$197–3093 for investigations to identify the cause of stillbirth and \$118–20 000 in hospital fees for additional medical care (appendix p 75, 76).

1 No direct reports of the cost of care in subsequent pregnancy exist, although three papers,^{13–15} all from HICs, recommended additional monitoring [A: of these costs?]. By use of these recommendations to derive models of care, we estimated costs from £3499 [A: the change in currency makes comparison within the text difficult, please convert GBP to US dollars] after a stillbirth of a non-recurrent cause to £4057 for a stillbirth of unknown cause.¹¹ A pregnancy after stillbirth costs £558–1735 more than if the previous pregnancy ended in an uncomplicated livebirth. Additionally, if care included more intensive surveillance with cardiotocography, costs rose to £4654–5616.^{16,17} Thus, the costs of subsequent pregnancy care add to the health-care costs associated with stillbirths in HICs; this situation will extend to MICs as these countries scale-up more intensive antenatal monitoring and care.

Indirect financial costs of stillbirth

20 The most frequent indirect costs for parents after stillbirth were for the funeral and burial or cremation of their baby (appendix). For some, this cost was mitigated by health insurance, government payments, or grants. Parents' free mobile phone text responses in the International Stillbirth Alliance (ISA) survey [A: reference available?] show the substantial financial burden of this group, magnifying the effect of these parents' loss (panel 3). Although some parents did not have to pay, others reported costs for funerals ranged from \$469–\$11 719, extending to \$1179–11 605 for burial plots and \$1410–4605 for memorials (appendix p 75, 76). The theme that occurred most frequently in the free-text responses was the long-term financial effect on families. For many parents, stillbirth was associated with reduced earnings from employment or an inability to return to paid employment. Meeting the continuing costs of counselling and medical care in further pregnancies was also mentioned.

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 (panel 3). Even after parents return 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 [A: please provide a reference] showed that only 12 of 170 countries with maternity benefit policies included specific provision for stillbirths; [A: an average?] 11 days off for mothers [A: paid or unpaid?] (28–84 days leave [A: confidence interval?]) and [A: an average?] 1 day off for fathers (5 days leave [A: what does this represent?]). Even in the few countries with this leave provision, bereaved parents seem to have little option to delay their return to work. Policies relating to stillbirth or miscarriage were identified from five (9.8%) of XX [A: please add] African countries, five (17.9%) of XX [A: please add] countries in

Asia, three (6·4%) of XX [A: please add] countries in 1
Europe, and four (11·8%) of XX [A: please add] in the
Americas (appendix p 78–81). Governments might incur
costs in countries that extend maternity rights to the
parents of a stillborn child. 5

Psychological and social effects of stillbirth

The period after stillbirth has extensive consequences for
parents and their families. Much of the effect is non-
monetary, suggesting the negative results of grief, 10
anxiety, fear, and suffering. These emotional factors have
been described as intangible costs.¹⁸ Almost all parents
report negative psychological symptoms after a stillbirth.
In the Listening to Parents study¹⁹ in the UK (n=473),
68% of mothers and 44% of partners reported four or 15
more psychological [A: correct?] symptoms at 10 days,
reducing to 35% of mothers and 13% of partners at
9 months. This situation is over three times greater than
after a livebirth, when 8–13% of mothers and 3% of
fathers report depressive symptoms at about 9 months 20
after the birth of their baby.^{20–22}

Family was the most frequently cited source of support
for parents after a stillbirth, although family input was
not universally positive (panel 3). This need for support
between parents and the wider family could strain 25
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 issues with other family members
(12% of mothers and 4% of partners).¹⁹ In the TEARS 30
cohort²³ in USA (n=216), the mean Family Assessment
Device score of respondents was 3·2 (range 0·5–4·0), in
which a score of 4 indicates significant [A: statistically?]
Or do you mean substantial?] dysfunction in family
relationships. Ultimately, this tension might lead to 35
relationship breakdown, which some studies report as
more frequent in parents who have a stillborn child
compared with a livebirth (odds ratio 1·40, 95% CI
1·10–1·79).²⁴ In other studies,²⁵ [A: only 1 study cited,
should there be more as you refer to “studies?”] the 40
proportion of families that divorce is unchanged, but
perceived relationship quality changed between married
(improved) and single [A: do you mean unmarried rather
than single?] women (deteriorated).

Systematic searching located 1082 relevant data points 45
from 144 studies of the psychological effect of stillbirth
(appendix p 31–50). These data were summarised into
23 themes and thematic sentences of the effect on
parents [A: correct?] with variable frequency effect sizes
(table). The most frequently reported experiences after 50
stillbirth were negative psychological symptoms,
including high rates of depressive symptoms, anxiety,
post-traumatic stress, suicidal ideation, panic, and
phobias.^{26,27} Although most studies evaluated these
symptoms subjectively rather than with a formal clinical
diagnosis, 60–70% of grieving mothers in HICs had
significant [A: statistically? or do you mean substantial?]

Panel 2: Methods

To extend the knowledge base with respect to direct, indirect, and intangible costs of
stillbirth on parents, families, and health-care providers, a series of systematic reviews
were implemented and meta-syntheses were undertaken with established methods.^{11,96,97}
A further systematic review and meta-synthesis identified interventions or systems that
might reduce the negative effects of stillbirth. Detailed methods of search strategies and
PRISMA diagrams were included (appendix). 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.
After screening, studies were identified by whether they met the inclusion criteria and
reported relevant information, three studies reported information on direct costs,
144 studies reported on the psychological and social effect on parents, 20 studies
reported psychological effect on professionals, and 42 studies were included in the
analysis of interventions to maximise wellbeing for bereaved parents.

To supplement data from published medical literature, we extracted data from three
questionnaire studies (the Listening to Parents study,¹⁹ the TEARS study,²⁹ and the
International Stillbirth Alliance [A: ref for this study?] survey) including a total of
5358 parents from HICs and MICs. We searched for data to match ten themes to identify
intangible costs of stillbirth identified by systematic review and meta-ethnography.⁹⁸
Data addressed eight of these themes, including: negative psychological impacts 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
or counselling for any negative effects associated with the stillbirth. These data were
supplemented with data extracted from a questionnaire survey of the experiences of care
providers in HICs and LMICs distributed by the International Stillbirth Alliance. For the
surveys, quantitative data were analysed with descriptive statistics, and free mobile
phone text responses were analysed by thematic analysis. Where financial costs were
reported by parents, the costs were converted to US\$ and shown in 2013 prices. Published
cost estimates are reported in their original currency but shown in 2013 prices.^{99,100}

grief-related depressive symptoms 1 year after their
babies death.^{23,28} These symptoms endured for at least
4 years after the loss in about half of cases. If these
figures are extrapolated to the 2·7 million women who
had a stillbirth each year,¹ an estimated 4·2 million
women are living with depressive symptoms after
stillbirth. Many [A: specific value available?] parents
reported persistent feelings of remorse or guilt for not
being able to save their baby. Nearly 40% of grieving
mothers in a convenience-sample survey²⁹ in the USA
were prescribed psychiatric drugs despite an absence of
evidence for the efficacy of these drugs in this population.
Parents responding to the ISA survey reported accessing
internet forums (more than 85%), support groups (about
30%), or consulting with religious leaders (about 30%)
or health-care professionals (about 55%) to address their
psychological symptoms. Little difference was noted in
the support [A: types of support and frequency?] sought
by parents from HICs and MICs (appendix, p 69).

Psychological distress persisted into subsequent
pregnancies when parents reported differing emotions
(eg, relief and worry, hopeful optimism, and panic attacks
or depressive symptoms).³⁰ Women tended to report

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confirm if any authors are full
professors and that the
affiliations are correct])

Panel 3: Parents' experiences of the direct, indirect, and intangible cost of stillbirth in high-income countries [A: why only high income countries?]

Direct and indirect costs [A: please indicate direct quotes by adding quotation marks]

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 [the tests] if my child lived, but having to pay for them after he died was difficult and a constant reminder as the bills kept coming ([A: is this parent number?]) #3903, Australia).

I could not properly bury my child because I lacked the financial means; that hurts today, because I have no grave (#19 342, 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 (#11 707, 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 (#26 496, 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

[The costs of stillbirth were] 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 (#19 795, Spain).

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See Online for appendix

volatile emotional states, whereas fathers tended to report suppression of their feelings. Parents were afraid to prepare for the birth of their subsequent baby and avoided general antenatal classes because they felt, as parents, they were outside the boundaries of normality. Some [A: specific values available?] women struggled to differentiate their dead baby's identity from their subsequently-born live baby.

The capacity to express and integrate grief reactions was a crucial 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.^{31–33} This issue 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.^{34–36} In these contexts, mothers' accounts suggested that they suppressed grief in public, instead choosing to deal with the emotions privately and alone.^{37,38} These accounts are supported by responses to the ISA survey of care providers (LMIC n=117, HIC n=2020). Fewer care providers from LMICs agreed that a death before birth is the same as the death of a child (19% LMIC vs 33% HIC) and more care providers attributed stillbirth to a mother's fault (4% LMIC vs 0·5% HIC [A: correct? Changed order of numbers from original to match text]) compared with HICs. Respondents from LMICs more frequently agreed that parents should forget about their stillborn baby and have another child (26% LMIC vs 3% HIC) and parents should not talk about their stillborn baby (12% LMIC vs 4% HIC) compared with HICs (appendix).

Fathers reported feeling unacknowledged as a legitimately grieving parent. The burden of these men keeping feelings to themselves increased the risk of chronic grief.³⁹ Differences in the grieving process between parents can lead to incongruent grief,^{40,41} which was reported to cause serious relationship issues, from conflicts about sexual intercourse to marital breakdown.^{24,42} Although family and friends were often essential for effective support,⁴³ respondents to some studies reported that family members had unrealistic, unhelpful expectations of recovery after stillbirth.

Many studies described the adverse effects of stillbirth on siblings, a surviving twin, and subsequent children, including issues with parent-child relationships, which could affect siblings' physical and mental health in the longer term.^{44,45} Some parents described anxiety with respect to their interactions with [A: correct?] children of other parents.⁴⁶ Stillbirth was reported to have adversely affected the emotional wellbeing of grandparents and other family members.⁴⁷

For some mothers, stillbirth affected their approach to life and death, self-esteem, and their own identity.^{33,48,49} Some mothers reported losing their sense of control, including during subsequent pregnancies, and their confidence in parenthood and child-rearing. Some women avoided contact with babies, creating social isolation and worsening depressive symptoms.²⁶ Some mothers were hesitant to meet neighbours or those who had known them when they were pregnant. Many women 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.⁵⁰ In reports from some LMICs, [A: refs 35,36,50–52?] women reported being substantially less valued by partners, families, and society. In extreme circumstances, this situation has led to spousal abuse, enforced divorce, and rejection by family and society, partly based on beliefs that women who have stillbirths are possessed by evil spirits or have procured abortions.^{35,36,50–52}

	Frequency effect size*	Example quotes (country) [A: I have noticed some sentences were direct copies from the cited papers without quote marks, to avoid plagiarism, please paraphrase these sentences]
Psychological effect of stillbirth on parents		
Stillbirth has been associated with a number of emotional and psychological symptoms	77%	"I am depressed, saddened, hurt, empty, guilty and lonely. I cry every day. I will mourn him forever." ²⁷ (Australia) A number of mothers recalled suicidal thoughts because of their desire to be with their baby ²⁶ (USA)
Parental grief following stillbirth may not be legitimised by health professionals, family, and society (disenfranchised grief)	31%	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." ³¹ (USA) This perceived lack of social understanding left these mothers alone and un comforted. 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 ³² (Australia) "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." ³² (USA)
Stillbirth might have a positive or negative effect on relationships, for example through different grief reactions (incongruent)	29%	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 ⁴¹ (Taiwan) 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 ⁴⁰ (Sweden)
In subsequent pregnancy some parents may experience psychological distress	27%	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. ⁶⁰ (USA) "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..." ³¹ (USA)
Stillbirth may change parents approach to life and death, self-esteem, own identity, and sense of control in subsequent pregnancy, parenthood, and childrearing	26%	The thoughts expressed by parents in our study consisted of being more humble and more grateful toward life itself and taking nothing for granted ⁴⁹ (Sweden) The men in the study also questioned their identity as fathers, uncertain as to their right to the term father ⁶ (UK) [A: should this be Ireland?] 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 ³³ (Australia)
Stillbirth can have an adverse impact on siblings, including the surviving twin, and subsequent children	24%	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 ⁴⁴ (Sweden) Infants next-born after a stillbirth were significantly more likely to be classified as disorganised in their attachment behaviour with their mothers than controls, this was strongly predicted by unresolved mourning in the mothers ⁴⁵ (UK)
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%	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" ⁶⁰ (USA) Men looked at sex as a tension reliever and attributed a therapeutic value to it ⁴² (Norway) 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 ²⁶ (USA) "I cry when I talk to a real person so it was easier to talk to someone online, less emotional" ³² (USA)
Some parents feel the need to suppress outward grief, including during subsequent pregnancy	18%	Fathers felt they denied their own emotional reactions in order to protect and support and care for their partners ³⁹ (USA) According to Taiwan's culture talking about death is a taboo subject and these mothers often dealt with their grief privately and alone ³⁸ (Taiwan) "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" ³⁷ (USA)
Stillbirth may lead to avoidance of activities that remind them of the pregnancy and the baby	13%	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 ⁵⁶ (USA)
Parents report stigmatisation, rejection, and spousal abuse	13%	"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 luck" ⁵⁰ (UK) "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." ³⁵ (Taiwan) "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." ³⁶ (Ethiopia)
Parents may have mixed feelings towards the decisions they made, for example post mortem or seeing and holding their baby	13%	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 ⁵⁵ (Japan)
Parents might have 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 ²⁶ (USA) 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 ⁵⁴ (Benin)
Bereaved parents might become hypervigilant with siblings and subsequent children, and anxious about other people's children	8%	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 child ⁴⁶ (Australia)

(Table continues on next page)

	Frequency effect size*	Example quotes (country) [A: I have noticed some sentences were direct copies from the cited papers without quote marks, to avoid plagiarism, please paraphrase these sentences]
(Continued from previous page)		
Bereaved parents might increase or decrease their use of health-care services; and in subsequent pregnancy, fathers might express a desire to be more included in care	7%	Mothers with a history of prior perinatal loss may attempt to cope with their anxiety in pregnancy and depression in early post partum with requests for additional health-care resources ⁶¹ (USA) Fathers felt the need to take more interest or active involvement in the subsequent pregnancies ³⁹ (USA)
Chronic pain and fatigue can occur after stillbirth	7%	3 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 ⁴⁰ (Sweden)
Some parents described parental pride after the birth of their stillborn baby	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." ⁷² (Australia) 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 ³⁷ (Sweden) There were parents who described a surging feeling of love from the moment they saw their child ³⁵ (Japan)
Employment difficulties and financial debt are potential effects of stillbirth	6%	Together with sustained difficulties in paying off hospital bills, this strained relations with family members from whom funds had been borrowed ⁵⁴ (Benin)
Stillbirth can motivate parents to engage with health-care improvement, including public awareness	4%	"I deal with it in a way that you know, to crusade, to campaign, to make sure things change, to try and take the positives as much as you can out of the whole situation" ⁵⁰ (UK)
Increased substance use has been reported for some parents	4%	They always were social drinkers but after Ricky died Mom increased her drinking ⁵⁵ (USA)
Women might develop a complex emotional response to body image	4%	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 ⁴² (Norway) 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 ⁶³ (USA)
Stillbirth has an adverse effect on the wider family	3%	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 ⁴⁷ (USA)
For some parents, quality of life might be affected in the long term	2%	Women with histories of fetal death seem to have poorer quality of life ⁹⁵ (Brazil)
Some couples experience competing emotional reactions to sexual relationships	1%	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 ⁴² (Norway) Women more frequently reported disturbing images, thoughts and feelings that interfered with sex than did men ⁴² (Norway)
Psychological effect of stillbirth on professionals		
Stillbirth has a powerful psychological effect	95%	"I think it's possible to experience too much grief in this work." ⁸⁵ (Ireland)
Emotional response or distancing	40%	"It is a mixture of everything, anxiety, rage, oppression, impotence..." ⁸⁴ (Spain)
Trauma	42%	"... I had to cut off my emotions to just get through it" ³⁷ (USA)
Guilt	35%	"It shook me to my core." ³⁷ (USA)
Anger	30%	"... you've got anger, huge anger, especially where a mistake has been made or something has been missed" ⁸⁵ (Ireland)
Fear	30%	"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" ⁹⁶ (Australia)
Stress	30%	
Anxiety	25%	
Blame	20%	
Depression	20%	
Frustration	15%	
Sadness	15%	
Powerlessness	10%	
Challenge to faith	5%	
Humiliation	5%	
Stillbirth has a professional effect	65%	"Is this the one that is going to blame you?" ⁸² (USA)
Effect of litigation	30%	"If you...lose a mother or a baby, you will lose your license, your income, your work" ⁸⁶ (USA)
Fear of disciplinary action	10%	
Fear of public censure	5%	
Exposure	5%	
Professionals need support	65%	"I think what would be helpful...is having that debriefing time after it's over and not being directly assigned" ⁸⁴ (Canada)
Education	30%	"...they do not teach you the necessary strategies to provide support in these situations" ⁸⁴ (Spain)
Peer support	5%	"...we need to support each other and not tear each other down." ³⁸ (USA)
Institutional support	5%	

(Table continues on next page)

	Frequency effect size*	Example quotes (country) [A: I have noticed some sentences were direct copies from the cited papers without quote marks, to avoid plagiarism, please paraphrase these sentences]
(Continued from previous page)		
Stillbirth can have a positive effect	30%	"I think having that experience, I've grown as a person." ³⁴ (Canada)
Benefit of experience	20%	"I feel like I make a difference, and if I can ease their pain I am happy" ³⁹ (USA)
Sense of honour	10%	
Privilege	5%	
Special bond with parents	5%	
Making a difference	5%	

Frequency effect sizes and representative quotes are shown for each theme. *Frequency effect size is the proportion of included reports containing a theme.

Table: Thematic sentences derived from meta-synthesis of studies assessing psychological effect of stillbirth on parents and on health-care professionals.

In the period shortly after the stillbirth, changed body image was important.^{42,53} Some women reported being embarrassed by their body after pregnancy. Others wanted to keep a pregnant body shape, maintaining a connection with their baby. Some women linked the grief to their physical body through pain [A: do you mean self-harm?] 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.^{26,54} Chronic pain and fatigue, increased substance use, employment difficulties, and financial debt were also reported. Some studies^{40,54,55} described a long-lasting negative effect on quality of life.

The consequences of a stillbirth were not exclusively negative. Some couples reported feeling closer.⁴⁰ Parental pride was reported by some parents after contact with their baby.^{27,56,57} For some, deciding to see or hold their baby brought a sense of finality that contributed to the grieving process.⁵⁸ Some parents engaged in therapeutic activities; seeking solitude, changing their uptake of religious practice, and changing their approach [A: in what sense?] to sexual intercourse or engagement with health promoting activities, work, and social media.^{26,32,42,59,60} Some parents campaigned for, and contributed to, health service improvements to help other families. Many parents changed the way they accessed health-care services, especially in subsequent pregnancies when fathers became more involved.^{39,61}

Effect of stillbirth on professionals

All 20 studies included in the systematic review of the effect on professionals undertaken for this paper (appendix p 51–53) documented a substantial personal and professional burden for staff involved with caring for families during and after stillbirth [A: data from HICs and LMICs?]. Four themes emerged from the data for staff: psychological effects, professional effects, need for support, and positive effects (table)[A: please note references will be renumbered after author revisions to house style]. The psychological effect was most frequently reported as somatic, including symptoms of trauma, diminished emotional availability, stress, and affective

states such as guilt, anger, blame, anxiety, and sadness.^{47,62–64} The professional effect of stillbirth was characterised by fear of litigation and disciplinary action. In one study [A: please provide a reference], data from LMICs suggested that professionals attending to a woman who has had a stillbirth could result in loss of livelihood and public humiliation.

Most studies (n=13) emphasised the need for further education and professional support for staff, especially in terms of the psychosocial care and communication skills needed after a stillbirth. Many studies ([A: n=xx?]) suggested that peer support was valuable, even though this guidance was usually informal. However, an absence of structured institutional and peer support was stressed. Quantitative studies ([A: n=xx?]) showed the risk of vicarious traumatic stress, and depressive and psychological symptoms such as guilt, self-blame, self-doubt, and grief. Importantly, those health workers who felt they had received adequate training in stillbirth care were less likely to report guilt and fear of litigation.

In six studies [A: refs please, 65-67 is only 3 studies], staff also reported feeling some positive gains, such as a sense of honour or privilege at being able to support parents experiencing the death of their baby.^{65–67} Some staff cited personal growth and the development of a special bond with parents and staff. In four studies [A: refs please], staff reported more confidence and comfort, with fewer negative effects, when they had more direct clinical experience with stillbirth.

These findings suggest that, although mothers, partners, and their families endure most of the effects of stillbirth, the event also has a substantial effect on health-care providers. The negative effects could be addressed by education, training, and provision of formal and informal support during and after stillbirth, and encouragement of positive experiences of caring for parents after stillbirth.

Interventions to maximise wellbeing for bereaved parents and families

What works?

43 studies provided evidence on what works to reduce the negative effects of stillbirths (appendix, p 56–67). Two systematic reviews^{68,69} of randomised controlled

trials (one on social support and the other on autopsy) did not locate any studies **[A: Do you mean identify any studies for reducing the negative effects of stillbirth?]**. No other randomised controlled trials were identified in the 43 studies. No intervention studies were identified for Africa, Asia, or the Middle East. Of the 16 studies that directly assessed interventions, ten included mothers only, one had fathers only, one had parents and care providers, and the remaining four studies included parents or the wider family, or both.

Effective interventions (in HIC settings) included: families seeing and holding the baby, social support and support groups, families making and sharing memories, autopsy, psychological interventions, and interventions with various components.^{70–75} 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 **[A: than those without this support?]**.⁷² A specific psychological intervention⁷⁶ in Brazil was associated with a range of positive effects, finding that inclusion of family members in the intervention reinforced network support. A US study⁷² reported that support groups were associated with **[A: statistically?]** significant improvement in scores on the Impact of Events Scale-Revised. Programmes with many components generally increased parents' satisfaction, with those more satisfied reporting less grief.^{72–74,77} Where measured longitudinally, this effect was maintained for up to 2 years. Finnish fathers receiving an intervention with various components, reported stronger personal growth and less blame and anger **[A: than those Finnish fathers who did not receive the intervention?]**.⁷⁷

The key findings of all included studies (qualitative and quantitative) were mapped to Sarafino's taxonomy of social support. This system comprises five support elements: tangible, emotional, esteem, informational, and network and belonging (appendix, p 56–67).⁷⁸

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

Data pertaining to specific groups of people were only reported from HICs. This showed 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 or co-mother, sister or brother, and grandparent, even after the death of the baby that created these social roles.

Variation in access to what works by cultural context

Access to support groups or services is not equitable. In the three surveys **[A: refs please]** on parents that were analysed in this paper, 54–93% of parents in HICs were given information about support groups or services compared with 12% of parents in MICs. Information about grief and psychological symptoms (16% in MICs vs 52% in HICs) was given less frequently in MICs than for physical symptoms (28% in MICs vs 47%), but this was not the case in HICs. The perceived effectiveness of support groups varied, but 77% of respondents to the ISA survey who used a group, reported benefit. Lower amounts of support available for parents in MICs might account for a greater proportion of parents rating their follow-up care as poor compared with HICs (60% vs 38%; appendix, p 69).

In the systematic review of what works for mitigating the negative consequences of stillbirths, eight of ten studies in LMICs included only women. The only positive factors reported by respondents from Malawi were basic physical care and brief information giving from nurses **[A: edit correct?]**, which were seen as surprising but welcome occurrences.⁷⁹ Studies in Tanzania,³⁴ Ethiopia,³⁶ India,³¹ and Taiwan³² **[A: correct addition of Taiwan and placement of refs?]** suggested that having a stillborn baby can lead to maternal abuse, social abandonment, and divorce. Despite feelings of grief and loss, mourning in these countries was actively discouraged and suppressed, and interventions such as families seeing and holding the baby and taking mementoes, were not culturally acceptable. This situation was echoed in care providers' responses to the ISA survey (LMIC n=117, HIC n=2020), which reported that parents in LMICs were less likely compared with those in HICs to be offered contact with their baby (35% in LMICs vs 94% in HICs), the opportunity to see and hold their baby (42% in LMICs vs 95% HICs), make memories (35% in LMICs vs 87% in HICs), and name their baby (39% in LMICs vs 83% in HICs) after a stillbirth.

The main support mechanisms reported in the included LMIC studies were family and local religious communities, rather than health-care professionals and wider society **[A: as noted in HICs?]**. In these contexts, interventions designed to improve emotional and informational support might depend on enhancement of community esteem for those who have had a stillbirth, especially through key religious groups. Networking and belonging support interventions could be primary mechanisms for improving women's wellbeing after a stillbirth in LMICs.



Figure: The effect of stillbirth originating with the death of the baby, affecting mother, family, health services, society, and government. Widespread themes of direct, indirect, and intangible costs are shown.

Summary of what works

On the basis of these data, the key element of what works to reduce the results of stillbirth on bereaved parents and families can be summarised as “seeing through the eyes of those affected” [A: is this a quote or a saying]. This includes staff who understand what different parents and families need and when they need it; communities that acknowledge grief and loss and do not stigmatise those who have had stillbirths; employers who provide effective leave arrangements; and governments that provide tangible support, such as funeral costs, and paid leave from work commitments.

The consequences of stillbirth

Stillbirth is associated with substantial direct, indirect, psychological, and social costs to women, and to their families, society, and government (figure). These include: medical care and investigations at the time of stillbirth and in subsequent pregnancies; funeral costs; grief and negative psychological effects; reduced social functioning; family and relationship disruption and breakdown; and negative effects on employment. The effect of stillbirth is enduring, and can persist for years. Similar issues, particularly direct health care and funeral costs⁸⁰ and the lasting effects on family function have been described for maternal death.^{81–84} In addition to families, the effects on staff and subsequent implications for staff wellbeing and future service quality and delivery must be considered. Depending on the setting, costs might be met by the government, insurance companies, or individuals and their families. Before this Series paper, these various costs of stillbirth have not been considered together. We argue that this situation has led to an underestimation of the economic, social, emotional, and psychological burden of stillbirth.

The worldwide effect of stillbirth: how to address research gaps

Our systematic approach has shown large gaps in available data with respect to costs and interventions that might reduce the burden of stillbirth by preventing these events or their negative consequences. Few studies established the direct costs of stillbirth in the perinatal period or subsequent pregnancies; all studies were from HICs. Studies that reported on the psychological and social costs of stillbirth or practices that might reduce the subsequent negative effects are concentrated in HICs (n=177), which have a low-burden of stillbirth, with little or no data available from high-burden LMICs (n=26; appendix, p 68). As most components of effective care were identified from studies in HICs, the data obtained are similar to a review restricted to only HICs.⁸⁵ Although some themes are consistent between HICs and LMICs, other factors, such as stigma and social isolation, seem to be particularly relevant in LMICs.^{36,38,54,86–88} Therefore, to appreciate the full cost of stillbirth, tailored research is urgently needed to establish direct, psychological, and social costs of stillbirth, particularly in LMICs and in marginalised women and their families.

In all settings, very little information is available about what works for fathers or 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 seems 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.

Little emphasis is given in intervention studies to networking and belonging support, and almost none to esteem support. In all settings, but particularly in LMICs, these components can form a basis to address 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. By contrast, where local family and social (notably religious) networks were supportive, mothers, in particular, reported positive benefits. Likewise, some parents and staff (in both LMICs and HICs) believed that they had grown spiritually, and had gained substantial coping skills as a result of their experience. Acknowledgment of the personal and professional cost of stillbirth on staff is essential, for their personal wellbeing and to enable health workers to deliver effective care to bereaved parents.

In LMICs, an intervention that addresses stillbirth at a health care, societal, and community level could make two major gains. The first could be the adoption of preventive measures, including improved health messaging [A: Do you mean communication of health information? From and to whom?], monitoring, support and care for women pre-pregnancy, antenatally, and during delivery, improving the health of the mother and her baby. The second could be destigmatisation of stillbirth, thereby reducing the negative consequences, especially for women.

Interpretation of the cost of stillbirth

In view of the research gaps identified, comprehensive estimates of the costs of stillbirth cannot be derived at present to inform cost-effectiveness analyses. Data for the financial costs of the sequelae of stillbirth are 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 to be meaningful. Data for the psychological and social costs are also scarce, particularly with regard to LMIC settings, fathers, the wider family, and health-care providers. Finally, any cost-effectiveness analysis must include a decision on how the loss of life to the baby is to be measured. Consequently, any attempt to assign a worldwide cost to stillbirth—in monetary terms or with summary measures of health such as QALYs and DALYs—would be misleading at present.

Despite the substantial costs of stillbirth set out in this Series paper, the extent of the total loss associated with stillbirth is substantially affected by whether the stillbirth is also counted as a loss in its own right (ie, 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.

Women's rights and values must be respected, including access to safe termination of pregnancy; however, recognition must also be given to the fact that most women who have had stillbirths had wanted pregnancies [A: edit OK?]. Similar evaluations of interventions to reduce neonatal mortality typically show results based on the time-discounted life expectancy of surviving infants.⁸⁹ To avoid undervaluation of interventions that prevent stillbirth, these controls should be assessed in this same way.^{90,91}

The use of QALYs in guidance by National Institute for Health and Clinical Excellence and earlier [A: do you mean previously mentioned?] iterations of DALYs apply discounting techniques to accommodate time-discounting of future benefits [A: what is meant here by future benefits?], giving 25 QALYs lost or 32 DALYs associated with stillbirth.^{92,93} 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. Alternatively, Jamison and colleagues [A: reference please] suggest that deaths before age 2 years should be adjusted according to extent of cognitive development or so-called acquired life potential. With time-discounting, this adjustment gives stillbirth DALY values of between 5 and 9 years, without time-discounting this figure would give DALY values of between 14 years and 26 years. Thus, proposals for how to value the life of a stillborn baby vary greatly. How these babies are valued can make a difference of orders of magnitude to the overall loss attributable to stillbirth (appendix, p 26). For example, a study⁹³ of the cost-effectiveness of a syphilis screening programme 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 stillbirths were included as a loss to the deceased.

Conclusion

Despite the gaps in the evidence, the findings in this Series paper suggest that the burden of stillbirths is substantial yet greatly underappreciated. This undervaluation might contribute to the slow pace of change to address stillbirths on national and international platforms, as identified by Frøen and colleagues.⁹⁴ [A: citation for ref 95 is missing, please advise on placement or deletion] Crucially, although the costs of stillbirth prevention might seem substantial in LMICs and HICs, the combined direct, indirect, and intangible costs of stillbirth are almost certainly greater still. We call on the global community to recognise the enduring effect of stillbirth on parents, families, staff, societies, and health and social care systems; to develop strategies to collect data for the cost of stillbirths and to use that information to invest in strategies, local services, and practices to prevent stillbirth and to invest in interventions to reduce the negative effects of stillbirth.

The Lancet Ending Preventable Stillbirths Series Study Group

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Contributors

AEPH was responsible for overall coordination and oversight of the Series paper and the writing process. JM and TR modelled values assigned to stillbirth. AEPH and TR were responsible for the systematic review of economic studies, direct costs, and costs in subsequent pregnancies. VF and AMW were responsible for the design and analysis of the international questionnaire. AEPH and MR analysed published questionnaire data. HB searched the International Labour Organization databases. DS and CS were responsible for the systematic review of psychological effects on parents. JC, KJG, DN, and KO'D were responsible for the systematic review of psychological effects on professionals. SD and OKM were responsible for systematic review of interventions to ameliorate effects on parents. ND and HETS helped put the paper into international context. JD, AR, and ZAB were responsible for using the Lives Saved Tool analysis. All named authors contributed to the conceptualisation, development, writing, and finalisation of the paper. AH is the overall guarantor.

Declaration of interests

HB received grants from Save the Children/Saving Newborn Lives. DS received grants from Stillbirth and Neonatal Death charity (Sands), is a members of International Stillbirth Alliance, and on the executive committee of the Stillbirth Clinical Study Group, Department of Health Stillbirth task-and-finish groups, and PRACTICAL Obstetric Multi-Professional Training maternity foundation. AEPH received grants from Tommy's during the conduct of the paper and grants from Sands and Holly Martin Stillbirth Research Fund outside the paper. AEPH is chair of the board International Stillbirth Alliance and on the executive committee of the Stillbirth Clinical Study Group, and Department of Health Stillbirth task-and-finish groups. CS received grants from Sands during the conduct of the study. [A: please confirm whether all other authors have no competing interests?]

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