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Safer Baby Bundle: study protocol for the economic evaluation of a quality improvement initiative to reduce stillbirths

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ABSTRACT

Introduction Stillbirth continues to be a public health concern in high-income countries, and with mixed results from several stillbirth prevention interventions worldwide the need for an effective prevention method is ever present. The Safer Baby Bundle (SBB) proposes five evidence-based care packages shown to reduce stillbirth when implemented individually, and therefore are anticipated to produce significantly better outcomes if grouped together. This protocol describes the planned economic evaluation of the SBB quality improvement initiative in Australia.

Methods and analysis The implementation of the SBB will occur over three state-based health jurisdictions in Australia—New South Wales, Queensland and Victoria, from July 2019 onwards. The intervention is being applied at the state level, with sites opting to participate or not, and no individual woman recruitment. The economic evaluation will be based on a whole-of-population linked administrative dataset, which will include the data of all mothers, and their resultant children, who gave birth between 1 January 2016 and 31 December 2023 in these states, covering the preimplementation and postimplementation time period. The primary health outcome for this economic evaluation is late gestation stillbirths, with the secondary outcomes including but not limited to neonatal death, gestation at birth, mode of birth, admission to special care nursery and neonatal intensive care unit, and physical and mental health conditions for mother and child. Costs associated with all healthcare use from birth to 5 years post partum will be included for all women and children. A cost-effectiveness analysis will be undertaken using a difference-in-difference analysis approach to compare the primary outcome (late gestation stillbirth) and total costs for women before and after the implementation of the bundle.

Ethics and dissemination Ethics approval for the SBB project was provided by the Royal Brisbane & Women's Hospital Human Research Ethics Committee (approval number: HREC/2019/QRBW/47709). Approval for the extraction of data to be used for the economic evaluation was granted by the New South Wales Population and Health Services Research Ethics Committee (approval number: 2020/ETH00684/2020.11), Australian Institute of Health and Welfare Human Research Ethics Committee (approval number: EO2020/4/1167), and Public Health Approval (approval number: PHA 20.00684) was also granted. Dissemination will occur via publication in peer reviewed journals, presentation at clinical and policy-focused conferences and meetings, and through the authors' clinical and policy networks.

This study will provide evidence around the cost effectiveness of a quality improvement initiative to prevent stillbirth, identifying the impact on health service use during pregnancy and long-term health service use of children.

STRENGTHS AND LIMITATIONS OF THIS STUDY

⇒ Use of linked administrative data to capture the real-world costs and outcomes of the Safer Baby Bundle.
⇒ Follow-up to 12 months post partum to potentially capture any unintended consequences of the Safer Baby Bundle.
⇒ Inclusion of both in-hospital and out-of-hospital health service use.
⇒ Lack of randomised controlled environment to identify the consequences of the Safer Baby Bundle on health service utilisation.
⇒ Limited capture of some health services such as over the counter medications, some allied health services, and non-government funded services; and time period of the intervention aligns with COVID-19 pandemic.

INTRODUCTION

Reducing the rate of stillbirth has become a global priority, driven by consecutive calls for action over the last decade,[1] in recognition of the frequency of the occurrence[2] and suboptimal performance of many countries in reducing this often preventable tragedy.[3] Globally, an estimated 2 million late gestation (28 weeks or more) stillbirth occur each year[4] with over half potentially preventable with known effective interventions; an avoidable loss of human life with far reaching social and economic consequences.[5] If the trends of stillbirth observed between 2000 and 2019 continue, it is projected that 20 million babies will be stillborn within the next decade.[6] Among this 20 million, 2.9 million stillbirths could potentially be prevented by accelerating

progress to meet Every Newborn Action Plan target in the 56 countries at risk to miss the goal.[7]

A number of stillbirth prevention activities have been implemented in high-income countries—notably the awareness of fetal movements and care (AFFIRM) package in the UK,[8] the Saving Babies' Lives Bundle in England[4] and the My Baby's Movement (MBM) maternal fetal movement monitoring app and education package in Australia.[9] The AFFIRM and MBM studies did not show a significant reduction in stillbirth rates, but the Saving Babies' Lives Bundle did reduce stillbirth risk. Within high-income countries, stillbirth has been noted to be associated with modifiable factors such as undetected fetal growth restriction, smoking during pregnancy, maternal supine sleeping position and decreased fetal movements,[10–13] pointing to the need for ongoing improvement of the provision of maternity care around such factors. In recognition of this the Safer Baby Bundle (SBB) has been proposed for the Australian context[14 15]—this consists of five evidence-based elements, each shown to reduce likelihood of stillbirth individually, which grouped together should result in better outcomes.

The potential for stillbirth reduction interventions to increase the use of caesarean section and induction of labour rates, and birth at earlier gestation, including early term births, has been raised by findings from the AFFIRM trial[8] and the Saving Babies' Lives Bundle.[4] This may not be surprising per se, as delivering all babies early would in effect end all late gestation stillbirth.[16] However, these results are significant as there is concurrent effort to stem the global increase in use of early caesarean section[17] and induction of labour[18] due to concern of long-term effects of child health and development outcomes such as allergies, asthma, diabetes, gastroenteritis, autism, attention deficit/hyperactivity disorder and poorer child cognitive and developmental outcomes.[18–27] In addition to the health impacts, these health outcomes also have important economic consequences associated with long-term health service needs.[28 29] Previous economic evaluation of stillbirth interventions has identified varying levels of cost effectiveness, and costs to healthcare funders.[4 30]

The evaluation of the SBB thus needs to consider the effect on reduction in stillbirth rate, as well as the costs implications—through any effect on background health services use such as caesarean section, induction of labour and preterm birth—independent of background trends in this area. This protocol describes the planned economic evaluation of SBB quality improvement initiative in Australia.

METHODS AND ANALYSIS

Intervention

This protocol pertains to the economic evaluation of the implementation of a quality improvement initiative to reduce stillbirth, the 'Safer Baby Bundle' (referred to hereafter as 'SBB'), which consists of five care elements:

1. supporting women to stop smoking in pregnancy,
2. improving detection and management of fetal growth restriction,
3. raising awareness and improving care for women with decreased fetal movements,
4. improving awareness of maternal safe going-to-sleep position in late pregnancy,
5. improving decision-making about the timing of birth for women with risk factors for stillbirth.

Each of these elements is described in detail in the overall study protocol.[14] Briefly, the SBB consists of an education programme and resources around each of these five elements developed by Queensland, New South Wales and Victorian state Departments of Health. In partnership with the Centre of Research Excellence in Stillbirth, each state Department of Health is responsible for the roll-out and embedding of the SBB—with the evidence-based education programme and resources in the five core elements. The SBB is being promoted across the entirety of the participating states—and were freely available from October 2019 to all hospitals and healthcare professionals. The decision of whether to implement the SBB at the service level was left to individual hospitals within each state, and was dependent on each hospital committing formally to be part of the state-based initiative.

The overall evaluation of the SBB will quantify the population-level effect on late gestation stillbirth (primary outcome), clinical intervention and preterm birth; identifying variations in the provision of care on outcomes for vulnerable (Aboriginal and Torres Strait Islander, migrant and refugee and rural and remote) women; evaluating the coverage, acceptability, feasibility, fidelity and sustainability of the SBB; exploring the indirect process and contextual factors that influenced the provision of the SBB in light of the COVID-19 pandemic; exploring the views of women and maternity health professions on the bundle; and estimating the cost-effectiveness of the SBB compared with standard care. For the purposes of this protocol, we focus exclusively on this last element—the economic evaluation of the SBB. The design of the economic evaluation is independent of other components of the overall evaluation.

The design of the economic evaluation, as described in this protocol, follows the recommendations of the International Society for Pharmacoeconomics and Outcomes Research Randomised Controlled Trial Cost-Effectiveness Analysis (ISPOR RCT-CEA) Task Force[31] and ISPOR Consolidated Health Economic Evaluation Reporting Standards.[32] This economic evaluation is being conducted alongside an observational study, rather a clinical trial; however, the ISPOR RCT-CEA recommendations are the most aligned with the study design.

Study design

The economic evaluation will be undertaken using a difference-in-difference (DDD) analysis approach alongside an observational study to compare the outcomes and costs at the population level across the three participating

states—Queensland, New South Wales and Victoria—before and after the implementation of the SBB. This will use a within-study analysis, as opposed to a modelled economic evaluation, with all inputs into the evaluation coming from observation study data as opposed to being drawn from the literature. A 5-year time horizon will be used, which is considered sufficient to capture key longer term health impacts of the SBB, without introducing unnecessary uncertainty; however, a lifetime horizon will also be used in a sensitivity analysis. A health funder perspective will be used, as is preferred by Australian funding decision-makers[33] as it is intended that this analysis will be used to inform ongoing funding decision-making around the SBB.

Patient and public involvement

No patient involvement in the economic evaluation design.

Participants and recruitment—implementation time period

Individual participants will not be recruited, rather the implementation of the SBB will occur over three state-based health jurisdictions (the delivery of maternity care is largely provided by hospitals within Australia, with each state within Australia being responsible for delivering hospital care) in Australia—New South Wales, Queensland and Victoria, and thus this is a population-level analysis. Victoria commenced implementation in June 2019; New South Wales in February 2020; and Queensland in October 2020. The wash-in implementation period, for the purposes of the evaluation will be for 2.5 years stretching from July 2019 to December 2021. The postimplementation maintenance phase will cover 2 years, stretching from January 2022 to December 2023. A list of formally participating hospitals in each state will be recorded as a part of the evaluation.

Based on the annual number of births in 2018 in Victoria (n=77 700), New South Wales (n=94 200), and Queensland (n=59 600) it is expected that around 463 000 births will be exposed over the 2-year maintenance phase. Currently, 23 hospitals in Victoria, 25 hospitals in New South Wales and 43 hospitals in Queensland are participating, which together provide care for over half of the population giving birth annually. The evaluation will consider the effect of the intervention over the entire state, with the analysis considering the effect on both the 'targeted' implementing sites, and the sites not formally participating ('non-targeted' implementors).

Control—preimplementation

The control group will represent standard care prior to implementation. The preimplementation time period for the control time period will be prior to June 2019 for Victoria, February 2020 for New South Wales, and October 2020 for Queensland. All births across New South Wales, Queensland and Victoria prior to these dates will be included for the control group.

Data source

The analysis will be based on a whole-of-population linked administrative dataset, which will include the data of all mothers, and their resultant children, who gave birth between 1 January 2016 and 31 December 2023 in Victoria, New South Wales and Queensland. The linkage will use the Perinatal Data Collection (PDC) in each state for the state-based linkage units to identify mothers and their children for inclusion. The PDC records all reported births within each state. It contains information on maternal demographics, clinical characteristics of mothers, medical interventions preformed in pregnancy and childbirth and infant outcomes. This will contain an estimated 1.8 million and 2.1 million children. The PDC within each state will then be linked to the

- Admitted Patient Data Collection: contains details of all inpatient admissions to public and private hospitals, and day surgery units. Covers diagnosis and procedure (International Classification of Diseases 10th Revision, ICD-10), and diagnosis-related group (Australian Refined Diagnosis Related Group) codes for each admission.
- Non-Admitted Patient Data Collection: contains details of all services provided through public hospital outpatient clinics, including of service, and investigations performed.
- Emergency Department Information System/Data Collection: contains details of all emergency department presentations in public hospitals. Covers triage category and diagnosis.

These datasets will then be linked by the Australian Institute of Health and Welfare (AIHW), a part of the Federal Department of Health, to:

- Medicare Benefits Schedule (MBS) claims records: contains details of all healthcare services provided under the MBS, such as general practitioner, specialist and some allied health attendances, and diagnostic tests, plus serviced assessed in private hospitals. Contains the MBS item number describing the type of service assessed, fee charged, amount covered by Medicare and the out-of-pocket fee charged to the patient.
- Pharmaceutical Benefits Scheme (PBS) claims records: contains details of any medication dispensed that is listed on the PBS. Contains the medicine item code, benefit paid and patient out of pocket fee.

Outcome measures

The primary outcome for the economic evaluation is late gestation stillbirths. This is defined as a stillbirth that occurs after 28 weeks gestation in singleton pregnancies without lethal fetal congenital anomalies.

Secondary outcomes will be:

- *Neonatal death*: all-cause mortality for child within 28 days of birth (including stillbirth).
- *Child admitted to special care nursery (SCN) or neonatal intensive care unit (NICU):* whether the child was

admitted to SCN or NICU at time of birth, before discharge.

- *Physical or mental health condition for the child from birth to 5 years post partum*: (1) a hospital admission or emergency department presentation/admission for infections, cancer, metabolic, mental health, gastrointestinal, neurological, musculoskeletal, cardiovascular, renal, or reproductive system conditions, injury, or pregnancy, as classified by ICD-10 code; (2) access to autism, pervasive development disorder and disability service MBS items (item numbers 135, 137, 139, 82015, 82020, 82025, 82035).
- *Mental health condition for mother within 12 months of birth:* onset of a mental condition for the mother identified from (1) a hospital admission or emergency department presentation for mental health as classified by ICD-10 code; (2) presentation for mental health outpatient care, based on diagnosis; (3) access to psychology support or psychiatric services through the MBS (item numbers 2700, 2701, 2712, 2713, 2715, 2717, 2725, 10956, 10968, 293, 296, 297, 359, 361, 348, 350, 352, 319); prescription of antidepressant or antipsychotics through the PBS.

Imputing chronic health conditions for children with missing data: the linked administrative dataset contains up to 7 years of follow-up health service use for children. However, for children born after December 2018 less than 5 years of follow-up data will be available, and any physical or mental health conditions for the child that developed up to the age of five will be imputed. A series of machine-learning informed risk prediction models will be constructed to predict the likelihood of developing infections, cancer, metabolic, mental health, gastrointestinal, neurological, musculoskeletal, cardiovascular, renal, or reproductive system conditions, injury or pregnancy, as classified by hospital admission and ICD-10 code, or autism based on MBS service use (as defined above).

The data will be randomly split into a 70% sample to be used to construct the models and a 30% sample to be used for validating the models. Stepwise probit models will be constructed whereby all available variables will be used to predict the conditions of interest. Backward selection stepwise estimation will be used to remove any variables that were not significantly related to the outcome of interest, or that are perfectly correlated, with a cut-off of $p<0.20$. The final predictive risk model will include variables that can predict each of the conditions of interest.

The 30% validation sample will be used to assess the model's performance in correctly identifying women who gave birth in private hospitals. Model performance will be assessed with area under the receiver operating characteristic curve and the 95% CI. These models will then be applied to the dataset for children with missing follow-up health data, with imputation commencing after last follow-up time point (rather than from birth). The relationship between birth characteristics and later onset of chronic health conditions is not thought to be affected by the SBB, although the SBB may affect the birth characteristics.

Costs

We will use a health funder perspective to quantify the direct costs of the interventions and all background health service use of the postimplementation and preimplementation groups from onset of antenatal care to 5 years post partum. All health services will be included in the analysis, including (but not limited to) antenatal appointments, scans, pathology tests, birth interventions, postnatal and early childhood appointments. The cost of all services accessed will be identified (described in table 1 below), and then summed to provide a cost per woman and child dyad. Costs will also be disaggregated to present costs to government funders, private health insurers and individual of pocket costs of health service use. Sunk costs, such as those related to the design and development of the intervention will not be considered. Study data will be used to estimate ongoing implementation costs such as educational resource development, and training costs. As the time horizon is 5 years, we will discount all future costs at a rate of 5%. All costs will be presented in 2023/2024 Australian dollars.

Analysis

Differences in costs between the postimplementation and preimplementation groups will be calculated on a cost-per-birth basis. Based on these data, we will estimate the DDD estimator using an ordinary least square linear regression with robust standard errors. Due to the skewed nature of cost data, the costs will be log-transformed prior to analysis. This model will include a dummy variable for the implementation time period (preimplementation, wash-in, post implementation), a time (calendar year) variable, and an implementation/time period interaction term (the coefficient of which will be the DDD estimator). A dummy variable for sites formally participating in the quality improvement initiative will also be included, and costs will also be adjusted for mother's age, body mass index, if assisted reproductive technology was used for conception, mother's pre-existing health conditions deemed to influence pregnancy, parity and plurality. Changes in primary outcome measure between groups will be assessed as a linear probability model with a DDD approach, similar to the above.

Cost-effectiveness analysis

The cost-effectiveness of the *Safe Baby Bundle* initiative will be conducted. An incremental cost-effectiveness ratio (ICER) will be calculated by comparing the *Safe Baby Bundle* initiative to the usual care group using the following formula:

$$ICER = \frac{(Implementation\ group\ total\ costs) - (Pre-implementation\ group\ total\ costs)}{(Implementation\ group\ number\ of\ stillbirths) - (Pre-implementationgroup\ number\ of\ stillbirths)}$$

To assess uncertainty around the ICER, we will use bootstrapping (running 1000 iterations) to estimate ICER CIs.[34 35] This will be graphically presented in the form

Table 1 Key sources of unit costs for the *Safer Baby Bundle* initiative

Health service unit	Description	How unit cost will be assigned
Ongoing implementation resources	Per participant, future (prospective) costs associated with the maintenance of the *Safer Baby Bundle* initiative.	The following will be costed from study data, or estimated based on best assumptions: 1. Salary costs (and on-costs) for personnel involved in maintaining and updating the education and training components; 2. Salary costs (and on-costs) for personnel providing the education and training; 3. Salary costs (and on-costs) for clinician time associated with attending education and training sessions.
Health service utilisation—pregnancy and birth	Per birth costs associated with total health service utilisation from onset of pregnancy to discharge from hospital post birth.	1. Each public hospital occasion of service (inpatient, outpatient and emergency department); out of public-hospital occasion of service; private hospital impatient and service; and prescription pharmaceutical dispensed from onset of pregnancy to discharge from hospital post birth will be identified from the linked administrative data. This will cover all health resources accessed by the mother during this time period, plus any resources accessed by the child from birth. 2. The cost of each public hospital occasion of service will be assigned from the Independent Hospital Pricing Authority's National Hospital Cost Data Collection (NHCDC) and the Australian Refined Diagnosis Related Group, Tier-2 or Urgency Related Group code given to each inpatient, outpatient and emergency department occasion of service respectively. The most recent NHCDC round will be utilised. The cost of health service use outside of public hospitals, or for outpatient use covered under the MBS, is contained on the MBS claims records. Cost of prescription medications covered under the PBS is listed in the PBS claims records. Costs will include those paid by individuals, government and other sources, and be disaggregated into the following sources: a. Public hospitals—paid by Federal and state governments via public hospital funding agreements; b. Medicare—paid by the Federal Government; c. PBS—paid by the Federal Government; d. Private health insurers; e. Individuals.
Health service utilisation—birth to 5 years post partum	Per birth costs associated with total health service utilisation from discharge from hospital post birth to 5 years post partum.	Health service use and costs will be identified per previous row; health service use of mother and baby until the time the baby is aged five will be included.

MBS, Medicare Benefits Schedule; PBS, Pharmaceutical Benefits Scheme.

of a cost-effectiveness plane against various willingness-to-pay thresholds to support decision-making. One-way sensitivity analysis will be undertaken to investigate the robustness of the ICER,[36] including alternative ongoing maintenance costs associated with the SBB, and with and without costs after birth.

Ethics and dissemination

Ethics approval for the SBB project was provided by the Royal Brisbane & Women's Hospital Human Research Ethics Committee (approval number: HREC/2019/QRBW/47709). Approval for the extraction of data to be utilised for the economic evaluation was granted by the New South Wales Population and Health Services Research Ethics Committee (approval number: 2020/ETH00684/2020.11), Australian Institute of Health and Welfare Human Research Ethics Committee (approval number: EO2020/4/1167), and Public Health Approval (approval number: PHA 20.00684) was also granted.

Data storage

Data from this project will be stored and accessed in Secure Unified Research Environment (SURE), which is operated by the SAX Institute and monitored by independent AIHW personnel via a curated gateway. The data never leave the SURE virtual workspace, and therefore, data access and storage compliance will be in line with SURE requirements. Only researchers with ethics and Public Health Act approval will be able to view or access the data within SURE, and researchers must only access the dataset in SURE while physically located in Australia. Data will be retained in SURE for the life of the project, which is set to be completed by 22 December 2025, after this time all electronic data will be destroyed by the operators of SURE. No paper-based versions will be made.

DISCUSSION

The aim of this study is to investigate the cost-effectiveness of the SBB. This will be achieved by comparing the costs and outcomes associated with SBB, including all background health service use of women during pregnancy and birth, as well as ongoing health service of the surviving children. This will also capture any unintended consequences of the SBB such as increase in induction of labour, caesarean section, birth at earlier gestation.

The key challenge in designing the economic evaluation lies in the lack of randomised controlled environment to assess the impact of the intervention. Maternity care in Australia, as in other high-income countries is constantly changing, with intervention rates that have been steadily increasing over the previous decades,[37 38] and the impact of COVID-19, which resulted in an increase in the use of telehealth, and a disruption in the provision of antenatal care delivery. A key challenge in this evaluation is to identify the true impact of the SBB, independent of background trends in service provision. The study design proposed in this protocol attempts to address this issue using a DDD approach, and the inclusion of both sites

targeted to implementing the SBB and those not formally a part of it.

Stillbirth rates have remained near constant within Australia over the previous decades, and Australia performs well below other comparable countries.[1] Addressing this unacceptably high rate is a national priority and the Federal Government has shown leadership in attempting to address this area.[39 40] The SBB offers a unique opportunity to address stillbirth rates by implementing a quality improvement package of strategies known to prevent stillbirth.[14] However, the full cost-effectiveness of the initiative will need to be determined to identify value that the ongoing provision of the initiative would offer.

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Contributors EJC designed the study and drafted the initial manuscript. All authors contributed to the preparation of the final manuscript.

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Patient and public involvement Patients and/or the public were not involved in the design, or conduct, or reporting, or dissemination plans of this research.

Patient consent for publication Not applicable.

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