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EVIDENCE BASED MIDWIFERY

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Evidence Based Midwifery

Those were the days of ploughing the research landscape, sowing the seeds of knowledge and now is the time to write research

Reflections on the development of the RCM's Evidence Based Midwifery journal

Keywords: Royal College of Midwives, Evidence Based Midwifery, reflections

It has been my privilege to be the founder and editor of the RCM's *Evidence Based Midwifery* (EBM) journal and to see it become an internationally recognised research journal, formally inducted into the Nursing Journal Hall of Fame (INANE) in 2020 (Nicoll 2020). The award is given for excellence and it was warmly welcomed by the RCM, myself and the membership.

The aim of EBM was to provide midwives (and others) with a robust platform for the publication of high-quality midwifery research when we launched it in 2003 (Silverton 2003). The quality of the research paper was a major factor for all of us as we set out to develop, nurture and support our profession to grow top-quality midwifery researchers.

The challenge resulted in the establishment of a dedicated and committed editorial team and a small number of trusted and reliable reviewers. The administrative processes were minimal but sufficiently functional and fit for purpose. At the beginning, we received a mixture of papers written in a variety of styles: some were full theses, others full reports.

In those days, we spent much of our time advising and revising and pruning papers as they went backwards and forwards many times before acceptance. It is important to remember the era and the context in which not all midwives were graduates and the majority were used to writing short notes for the records, not for public consumption and critical assessment.

Furthermore, the number of midwives with PhDs or MScs was small and this led to the formation of the Doctoral Midwifery Research Society (DMRS), founded in 2007 and supported by a fund from the (then) Northern Ireland Research and Development Office. EBM is the official journal for publication of papers presented at DMRS conferences.

Today, I can say with confidence, the RCM has indeed grown a strong and fruitful tree of knowledge for producing high-quality research and should glow with pride. The memories of those early days are bubbles of sheer joy as midwives floated with delight when they received their first publication in EBM. I can still see several of my PhD midwives swirling around the place animated with enthusiasm for research and for the EBM opportunity to publish, with RCM support. The distinguished wine and silver journal came with six free copies for those who had published. This was a great gift as hard copies were precious then and holding the journal in your hand was a necessary proof of publication. However, in this new era, everything is available online: hard copies are becoming extinct and a rebrand is necessary.

Those were indeed the days of ploughing the land and sowing the seeds of knowledge and now we are gathering the evidence. Looking back, there were times when the work seemed to be endless and recognition of the journal impact factor seemed an elusive goal. This all changed when the guidance for the REF2014 made it clear that the impact factor of a journal was not to be taken into consideration by the assessment panels (Higher Education Funding Council for England (HEFCE) 2014). This was a major boost for EBM as it made its name known as a serious research journal for showcasing midwifery's contribution to research.

I gleamed with pride when I saw papers published in the journal submitted for assessment in the REF and was even more delighted when they received high scores. We made the grade and this was a major indicator for me that we were achieving the necessary recognition for quality research without having the Thomson Reuter Impact Factor.

In this new era, with so many online journals popping up every day, it is essential to tread carefully: I would strongly advise any midwife planning to publish to select the publication journal with great care. It is worth searching for predator journals by accessing *Beall's List* (Beall 2021) and to be aware of the

Retraction Watch database (Retraction Watch 2021a) which exposes publication misdemeanours and publishes an evidence trail of papers that have been withdrawn due to plagiarism or misrepresentation of data. A recent post on the database demonstrates how a PhD thesis was ripped off by another author and subsequently exposed, leading to it being withdrawn (Retraction Watch 2021b).

Writing research for publication requires ethical and professional knowledge in addition to the expected academic skills. I would therefore also advise authors to visit the Committee on Publication Ethics (COPE) (2021) website to refresh themselves on good ethical behaviour in publication practices and policies.

Google searches on writing for publication produce pages of names of experts who will write papers for you — and charge a range of fees. However, many journals also produce top tips for successful publishing; I was fascinated by a really good short piece in which a selection of authors shared their tips, with catchy headings such as '*Prune that purple prose*' (Nature Careers 2018). In essence the same messages — about audience engagement, good titles, organised content and getting to the point — are all in the prose.

The easy papers to write are the straightforward reporting of randomised controlled trials and cohort studies as they follow a pre-defined script (Consolidated Standards of Reporting Trials (CONSORT) 2021) Writing qualitative papers is a much bigger challenge as they tend to be more complex and information-dense, requiring considerable pruning and refinement.

More recently in EBM we have introduced the opportunity for midwives to publish their research protocol or their literature review protocol. This is important to facilitate the development of publication profiles for midwives who are planning a research career. They are also important outputs for funders and institutions supporting research.

Time passes noiselessly: one hardly notices the amazing developments taking place in the world of midwifery researchers — and looking back is a valuable exercise. Today, I see some of the midwives who published in the early EBM days established in major research roles. I often wonder if they look back and remember their publication experience with the EBM team.

There is a time for everything and now the new EBM in MIDIRS does not require that same level of input and has a large support team with great experience and vision for the future. Our profession is now a graduate midwifery workforce with highly skilled midwives who have the necessary knowledge and skills to produce the highest quality clinical midwifery research, in partnership with the women we serve.

The new era for the journal is just beginning and I am delighted to see it become integrated into the MIDIRS portfolio where a new style of support will facilitate new and much needed growth.

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Evidence Based Midwifery

Women's experiences of planning a home birth with maternity care providers in middle- to high-income countries: a systematic review protocol

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ABSTRACT

Background: A woman's choice of birthplace does not only influence her birth experience, but also impacts on maternal and neonatal outcomes. For healthy women who have had a straightforward pregnancy, a planned home birth supported by midwives and other maternity care providers, is now a recognised choice within many individual countries' health care systems. However, there is limited evidence on women's actual experiences of engaging with maternity care providers to plan for a home birth, especially within the context of middle- to high-income countries where there is integration of maternity care services. Therefore, this systematic review will synthesise findings from previous studies, which have reported on women's experiences of planning a home birth in consultation with maternity care providers, in middle- to high-income countries. We anticipate that primarily qualitative studies will be located, as the focus of the review is on experiences of women.

Methods: Using a systematic approach, we will develop a search strategy to identify relevant research studies on women's experiences of planning a home birth, with the support of their maternity care providers.

Search terms will be iteratively developed using text words derived from the review aim, database-indexed terms and the Population, Intervention, Comparison and Outcome (PICO) framework:

- **P**opulation: women who planned or are planning a home birth within the context of a middle or high-income country
- **I**ntervention: planning home birth with maternity care providers
- **C**omparison: none applicable
- **O**utcome: experiences.

Searches will be undertaken on seven bibliographic databases: MEDLINE, Embase, PsycInfo, CINAHL Plus, Scopus, ProQuest and Cochrane (Central and Library). Supplementary searches will also be undertaken to identify additional articles, including grey literature. At least two reviewers will do the screening, quality appraisal, data extraction and analysis. Included studies will be appraised using a quality appraisal tool suited to the study design. Data will be analysed depending on the methodological design of the studies included (that is, if all qualitative studies are included a thematic synthesis will be undertaken).

Expected outcome: Review findings will provide useful recommendations to improve care and support provided for women when planning a home birth. We will publish review findings in a peer-reviewed journal and present it at relevant conferences while also sharing summaries with maternity care providers and service users via social media fora.

Systematic review registration: *PROSPERO* CRD42018095042 (updated 28 September 2020).

Keywords: home birth, women, planned, childbirth, maternity care provider, experiences, Evidence Based Midwifery

Background

A woman's chosen place of birth impacts not only the type of birth, but also the number of unnecessary interventions that the mother and baby are exposed to during their labour and birth (Brocklehurst et al 2011, de Jonge et al 2015, Scarf et al 2018).

Women who give birth in a midwife-led unit or at home, rather than an obstetric unit, experience lower rates of unnecessary interventions. These include amniotomy, augmentation of labour, instrumental vaginal birth, caesarean section, and opiate or regional analgesia (Brocklehurst et al 2011, Halfdansdottir et al 2015, Hutton et al 2016, Reitsma et al 2020).

There is also evidence that the outcomes for both multiparous and nulliparous women and the babies of multiparous women who have birthed at home are equal to, if not better than those in other birth settings: for example, women are less likely to experience third- or fourth-degree perineal trauma, maternal infection, or postpartum haemorrhage (Brocklehurst et al 2011, Hollowell et al 2011, National Institute for Health and Care Excellence (NICE) 2014, Reitsma et al 2020).

The Birthplace in England study also reported an increased incidence of adverse outcomes for the babies of nulliparous women who had a planned home birth (Brocklehurst et al 2011). However, cohort studies on home birth have identified perinatal outcomes from home birth as low and not significantly different for infants of nulliparous women (van der Kooy et al 2011, de Jonge et al 2015).

NICE (2014) CG190 was updated in 2017 and continues to support a policy of offering all women with straightforward pregnancies a choice of birth settings including home, midwifery units (both alongside and freestanding) or obstetric unit.

Coxon et al (2017) conducted a qualitative synthesis of women's decision making for a birthplace preference and choice. The review identified that women's choice of birthplace was influenced by how informed they were about available options, their right to choose, experiences of previous births, risk perceptions, safety concerns and their care-givers' views (including family, friends and health care professionals). Planning birth at home can be enabled by following an evidence-based guideline and co-produced resources for women and their partners (Regulation and Quality Improvement Authority (RQIA) 2019).

A position statement on home birth by the International Confederation of Midwives (ICM) states that '*women have a right to home birth as a valid and safe option*' (ICM 2017:1). It also states that women have a right to make an informed decision to give birth at home supported by a midwife within their own country's health care system.

A recent joint statement by the Royal College of Midwives (RCM) and Royal College of Obstetricians and Gynaecologists (RCOG) (2020) asserts that healthy women with low-risk pregnancy may benefit from giving birth at home during the evolving COVID-19 pandemic (RCM & RCOG 2020).

In a principle-based concept analysis, Beecher et al (2019) propose a theoretical definition of 'Women's experiences of their maternity care' as:

'*… a complex concept referring to women's interpretation of their care encounters within the maternity services. It is subjective in nature and evolves throughout the course of pregnancy, childbirth and the postpartum period. It is dependent upon woman's individual needs and expectations, shaped by their personal circumstances and influenced by how their care is organised and delivered.*' (Beecher et al 2019:4).

In their systematic review and meta-analysis, Rietsma et al (2020) identified that women who plan to birth at home may hold different values around birth outcomes. However, they also recognised that those who plan a home birth are less likely to experience unnecessary interventions and adverse birth outcomes.

Little is known regarding women's actual experiences of engaging with maternity care providers to plan for a home birth. The dynamics of the woman–health care provider relationship in planning for a home birth within the context of a middle- to high-income country, where women have access to an integrated community and hospital maternity care system, is worthy of investigation.

Previous reviews have looked at maternal and neonatal outcomes (Catling-Paull et al 2013, Elder et al 2016, Kobayashi et al 2017, Scarf et al 2018) and comparison between planned hospital and planned home births (Olsen & Clausen 2012, Rossi & Prefumo 2018). Others have examined post-partum issues (Pantoja et al, 2016), model of care for childbearing women (Sandall et al 2016), integration of home birth into a health care system (Comeau et al 2018) and scope of hospital transfers during homebirth (Blix et al 2014, Vedam et al 2014). A recent review by Hill (2020) looked at women's experiences of planned home birth. This review used the Sample, Phenomenon of Interest, Design, Evaluation, Research (SPIDER) framework (Cooke et al 2012) and included only four papers. The review was not focused on the experience of planning home birth with maternity care providers.

A systematic review of studies on women's experiences of planning a home birth is needed to provide an in-depth understanding of what matters to women, including their information and support needs. Insights gleaned from this proposed systematic review could potentially help to enhance woman–health care provider interactions in planning for a

home birth and inform future service provision to maximise positive experiences for women, planning to birth their babies at home. Therefore, the aim of this systematic review is to synthesise findings from previous studies which have reported on women's experiences of planning a home birth in consultation with maternity care providers in middle- to high-income countries. The review question is: 'What are women's experiences of planning a home birth in consultation with maternity care providers in middle- to high-income countries?'

Methods

An important starting point for any review is operational definitions of the concepts under review. Given the rise in literature reporting on unassisted or free birthing, on Babies Born before Arrival (BBA's) to hospital, and the increased visibility of birthing supported by unregistered attendants, operational definitions of planned home birth and maternity care providers are central.

Operational definition of terms

We use the following definitions:

Planned home birth: an informed decision by women to birth their baby at home with the support of maternity care providers.

Maternity care providers: health care providers involved in supporting women to plan their birth at home. These will include midwives, obstetricians, general practitioners, (GPs) anaesthetists, paediatricians, and paramedics.

Country classifications for middle- and high-income countries

The organisation of health care differs between countries and between low- and middle-income countries and middle- and high-income countries. The focus of this review is on middle- and high-income countries. The classifications used are provided below.

Country classifications

According to the World Bank classification, high-income countries (also known as developed countries) are countries with per capita gross national income (GNI) of at least US$12,476 as of 2018 (World Bank 2020). For example, Argentina, Australia, Barbados, Canada, Chile, Croatia, Denmark, New Zealand, France, Germany, Finland, Portugal, Spain, Sweden, Switzerland, United Kingdom, United States of America.

Middle-income countries have per capita GNI of between US$1025 and US$12,476 as of 2018. For example, Angola, Bangladesh, China, Cameroon, Ghana, India, Kenya, Indonesia, Nigeria, Pakistan, Philippines, Sri Lanka, Republic of the Sudan, Tunisia, Vietnam, Zambia.

Low-income countries are those with GNI per capita of US$1025 or less as of 2018. For example, Afghanistan, Benin, Burkina Faso, Burundi, Central African Republic, Chad, Democratic Republic of Congo, Eritrea, Ethiopia, Gambia, Guinea, Guinea Bissau, Haiti, Korea, Liberia, Madagascar, Malawi, Mali, Mozambique, Nepal, Niger, Rwanda, Sierra Leone, Somalia, Republic of South Sudan, Tajikistan, Syria, Tanzania, Togo, Uganda, Republic of Yemen.

Search strategy

Using a systematic approach (Centre for Reviews and Dissemination 2009), we will develop a search strategy to identify relevant research studies on women's experiences of planning a home birth, with the support of their maternity care providers.

Search terms will be iteratively developed using text words derived from the review aim, the PICO framework (Thomas et al 2019) (see Table 1), and database-indexed terms. Broadly, search terms will be words related to: (home birth OR childbirth) AND plan AND experience (see Appendix 1 for a sample draft of the MEDLINE search).

Table 1. PICO framework

Population	**Inclusion criteria**
	Women who planned, or are planning, a home birth within the context of a middle- or high-income country in consultation with maternity care providers. **Exclusion criteria** • Women who had an unplanned or unassisted/free home birth • Women planning a home birth without consulting a professional maternity care provider • Women who planned, or are planning, a home birth within the context of a low-income country (low-income countries are excluded because their health care provision or context is different to that of middle- to high-income countries).
Intervention/ exposure	Primary studies which: • Focus on the planning phase of the home birth experience for women (planned home birth as defined above) and • Report on women's experiences of planning their home birth with their maternity care providers
Comparison	Not applicable
Outcome(s)	(i) Women's experiences of planning a home birth (ii) Women's perceptions of their consultation with maternity care providers to plan a home birth

We will test and refine the search strategy for accuracy on MEDLINE prior to running it on other databases, as appropriate. The refined search strategy will be utilised on seven bibliographic databases: MEDLINE, Embase, PsycInfo, CINAHL Plus, Scopus, ProQuest and Cochrane (Central and Library) from January 2015.

January 2015 was chosen as our cut-off point for the searches as the publication of the NICE clinical guideline (CG190) *Intrapartum care for healthy women and babies*, which advocated for home birth as a choice of place of birth for women, was December 2014 (NICE 2014). Evidence previous to January 2015 would, therefore, have been utilised to develop the guideline.

In line with the Peer Review of Electronic Search Strategies (PRESS) guidelines, we will develop the search strategy in consultation with an experienced subject librarian, which will be checked by at least two authors.

We will tailor the refined search terms to each database's indexing requirement. Boolean operators 'AND' and 'OR' will be used to combine search terms as appropriate. We will also use quotation (") and truncation (*) marks to capture possible variations of the search terms on each database. We will further conduct supplementary searches to identify additional articles, which we may have missed during the electronic database searches. This will include back-chain referencing of included papers (hand searching of reference lists), consultation with members of the *Planning to birth at home in Northern Ireland* (RQIA 2019) guideline development group, professional networks and grey literature search (for example, OpenGrey).

We will run the searches again on the selected databases prior to the final analysis to identify any newly published articles. We will manage search results with the bibliographic databases Endnote, Refworks and Covidence. Deduplication of retrieved articles will be undertaken on Endnote and Covidence using a systematic method (Bramer et al 2016).

The review is registered on the International Prospective Register of Systematic Reviews (PROSPERO CRD42018095042, updated 28 September 2020).

Identification and selection of studies

Studies will be identified and selected based on the following inclusion and exclusion criteria:

Inclusion criteria

Primary studies, which investigated women's experiences of planning a home birth within the context of middle- and high-income countries, reported in the English language and published from January 2015 will be included. Studies that report on women's experience and/or perceptions of their consultation with maternity care providers when planning a home birth will also be included.

Exclusion criteria

We will exclude grey literature which lacks a clear methodology (for example, editorials and books) and conference abstracts if full papers cannot be accessed and PhD and MSc dissertations. We will exclude studies focused on health care professionals' or partners' views on home birth planning. We will exclude home birth studies that lack clear separate data on women's experiences of the planning phase of the home birth, and studies conducted in low-income countries.

Screening

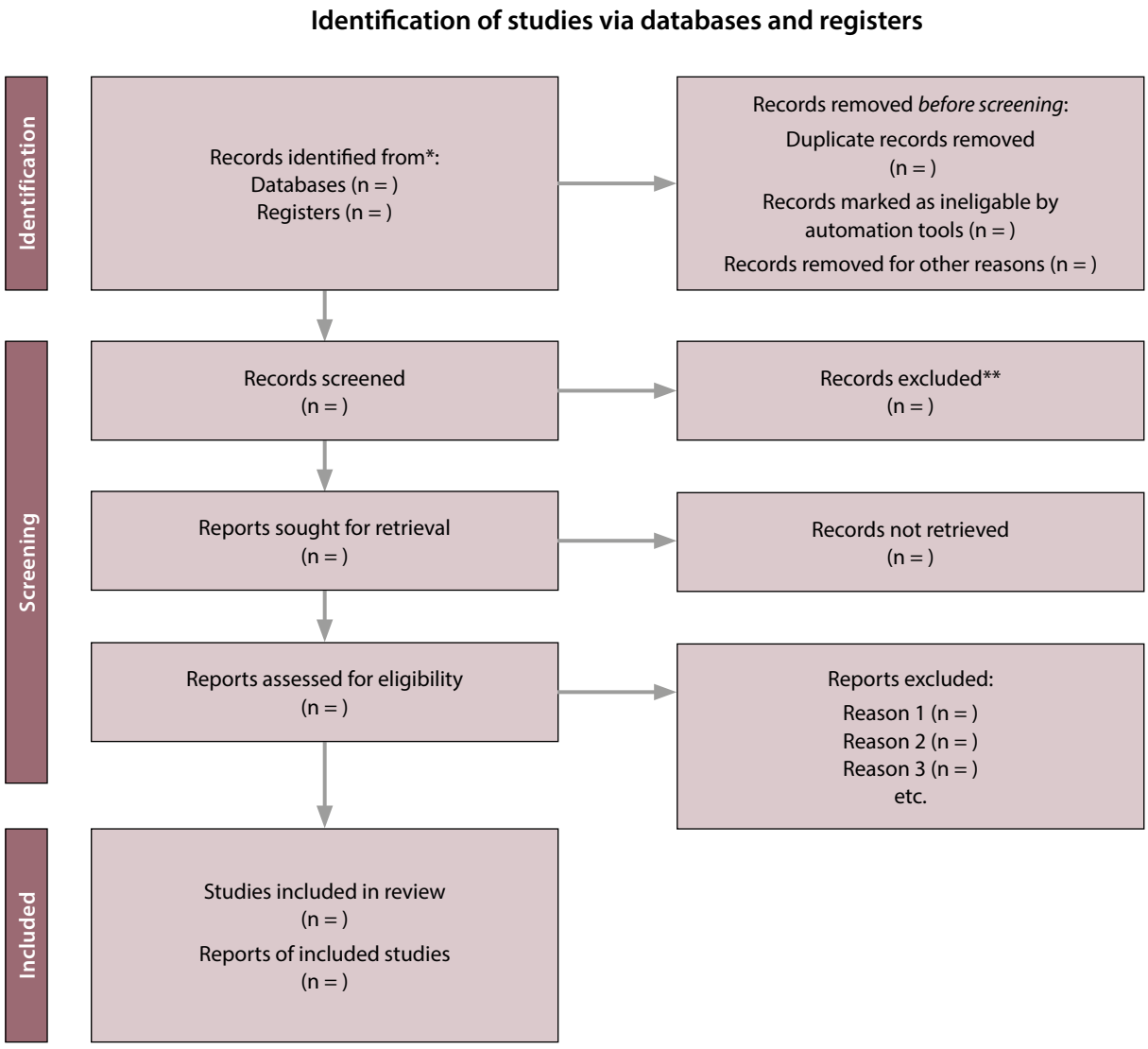
Following deduplication on Endnote, we will upload the remaining articles into Cochrane's systematic review management software to manage the screening process in a rigorous and transparent approach in line with the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) guidelines (Page et al 2021).

At least two reviewers (PG, MH or OB) will independently screen the titles and abstracts of retrieved studies to remove irrelevant articles. Two authors will resolve any conflicts and, if not possible, a third author will review and then all three authors will reach agreement. Two authors (PG, MH or OB) will then screen the full text of potentially relevant articles against the review's inclusion and exclusion criteria. We will resolve differences in opinion through discussion (by a minimum of two authors) to reach a mutual agreement. We will report the study selection process on a Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) diagram (see Figure 1).

Quality appraisal

At least two reviewers (PG, MH, or OB) will independently appraise the quality of the included studies using an appraisal tool relevant to each study's methodological design. We will appraise studies using the Critical Appraisal Skills Programme tool suited to each study's design (CASP 2020). For example, qualitative studies will be assessed using the CASP tool for qualitative studies. We will appraise randomised controlled trial (RCT) studies (if included), using the CASP tool for RCTs, although we do not expect to find any RCTs due to the nature of the review question.

We will assess other quantitative studies (non-RCTs), using the Grading of Recommendations Assessment, Development and Evaluation (GRADE) tool (Guyatt et al 2008). Mixed methods studies will be assessed using the Mixed Methods Appraisal (MMAT) tool (Hong et al 2018). We will assess risk of bias in



*Consider, if feasible to do so, reporting the number of records identified from each database or register searched (rather than the total number across all databases/registers).

**If automation tools were used, indicate how many records were excluded by a human and how many were excluded by automation tools.

Source: Page et al (2021).

Figure 1. PRISMA 2020 flow diagram for new systematic reviews

RCTs (if included) using the Cochrane risk of bias tool (Higgins et al 2021) and the Confidence in the Evidence for Reviews of Qualitative Research (CERQual) tool for qualitative studies (Lewin et al 2018).

Data extraction

At least two reviewers will extract data using a standardised form on MS Excel or MS Word. Conflicts will be resolved through discussion. We will systematically extract data on outcomes related to women's experiences on planning a home birth with their maternity care providers. We will extract data on the study title, author(s) and year of publication, study setting, methodology, population, key findings, quality appraisal score and key conclusions. Where possible, we will attempt to retrieve missing data in relevant studies by contacting the corresponding author.

Data analysis

At least two reviewers will analyse aggregate data from the final included studies and resolve any conflict through discussion. The approach for data analysis will be determined by the methodological design of the included studies. If the included studies

are quantitative and qualitative, we will integrate the findings, however, if all the studies are qualitative a thematic synthesis will be undertaken according to Thomas & Harden (2008). NViVo 12 software will be used to manage the data analysis process where appropriate.

Discussion

Findings will be discussed in relation to existing research. Review findings will provide useful recommendations to improve the experiences of women planning a home birth. We will publish the review findings in a peer-reviewed journal, and present at relevant conferences while also sharing summaries with maternity care providers and service users via social media fora.

Acknowledgments

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Conflicts of interest

The authors declare they have no competing interests.

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The funders were not involved in collection, analysis and interpretation of the data.

Ethical approval

This is a protocol for a systematic review which utilises published data therefore ethical approval was not required.

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Dr Patricia Gillen, Head of Research & Development for Nurses Midwives and AHPs/Reader in Nursing and Midwifery Research and Development, Southern Health Social Care Trust/Institute of Nursing and Health Research, Ulster University.

Contributions

PG and MH conceived and designed the review. All authors (MH, OB and PG) contributed to the writing of the protocol and will contribute to the collection, analysis and interpretation of the data. PG is the guarantor of the review.

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Appendix 1. Sample draft of search strategy on MEDLINE.

1. (home adj3 birth$).mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
2. home childbirth$.mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
3. homebirth$.mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
4. (place adj3 birth$).mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
5. home delivery/
6. (home adj3 deliver$).mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
7. 1 or 2 or 3 or 4 or 5 or 6
8. plan$.mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
9. 7 and 8
10. limit 9 to yr="2015 -Current"
11. limit 10 to English language
12. exp Communication/
13. experience*.mp.
14. "Referral and Consultation"/
15. consultation.mp.
16. Social Perception/
17. perception*.mp.
18. 12 or 13 or 14 or 15 or 16 or 17
19. 11 and 18

What is known about the experiences of women receiving hands-on perineal interventions in the second stage of labour – a scoping review

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ABSTRACT

Background: Perineal damage affects 77 per cent of Australian women birthing vaginally with many women perceiving this as an expected outcome of vaginal birth. Evidence regarding effective interventions in the second stage of labour for reducing perineal damage is lacking. There is also some disagreement about appropriate methods of providing perineal care in the second stage of labour. The quantitative nature of current evidence regarding perineal care in labour undervalues women's psychosocial health which may have negative impacts on women's experiences.

Aim: To determine the scope and nature of evidence regarding perineal care in the second stage of labour.

Methods: A scoping review was conducted using Arksey and O'Malley's (2005) five-step framework whereby the review question was identified, relevant studies sourced and selected and data charted and synthesised. Systematic and replicable searches of the Cochrane Database of Systematic Reviews, CINAHL Complete, MEDLINE, PsycInfo, MIDIRS Maternity & Infant Care (MIC) database, Google Scholar, Google and NICE guidance were performed using combinations of key words related to the review aim in combination with Boolean operators AND and OR. A scoping review methodology allowed for the inclusion of both published and grey literature, including clinical guidelines.

Findings: The initial search resulted in 344 hits, with 341 accessed and found not to meet inclusion criteria. Google provided a further 14 hits. The literature search resulted in four articles consistent with the review criteria. As this is a scoping review, formal critical appraisal was not conducted, however a PRISMA charting exercise was performed to ensure pertinent information in each data source was accurately identified.

Australian and international literature were included, giving a global perspective, and both qualitative and quantitative data were included. Analysis of data from sources retained for review highlights a maternity care culture which values the physiological outcomes of birth, while paying little attention to the potential psychosocial implications of hands-on perineal interventions, such as manual perineal support and the use of warm compresses.

Conclusions: Future research investigating women's views about, experiences of, and preferences regarding intimate aspects of birth care is required to better inform clinical practice guidelines. Further education pertaining to the provision of trauma and violence informed care (TVIC) may be of benefit to maternity care providers.

Keywords: perineal pain, perineal interventions, perineal damage, labour, women's experiences, trauma and violence informed care, birth experiences, Evidence Based Midwifery

Introduction

Perineal injury is a common outcome for women giving birth vaginally (Australian Institute of Health and Welfare (AIHW) 2020). The perineum comprises the tissues between the vagina and anus, including the muscles of the pelvic floor (see Table 1) (Tortora & Derrickson 2012). Damage occurs either spontaneously as the tissues stretch during birth, or due to episiotomy.

In Australia, in 2018, almost half of women experiencing a vaginal birth sustained first- or second-degree tears, with a further three per cent suffering more serious third- or fourth-degree tears (AIHW 2020). These can cause significant morbidity including urinary and anal incontinence, and sexual dysfunction (Dahlen & Priddis 2019).

Table 1. Classification of perineal tears.

First-degree	Damage to perineal skin *and/or* vaginal mucosa
Second-degree	Damage to perineal muscles, *not* involving the anal sphincter
Third-degree	Damage to perineum *and* anal sphincter complex
3a	*Less* than 50% of external anal sphincter (EAS) damaged
3b	*More* than 50% of EAS damaged
3c	*Both* EAS and internal anal sphincter (IAS) damaged
Fourth-degree	Damage to perineum including EAS, IAS *and* anal mucosa

Source: Royal College of Obstetricians and Gynaecologists (RCOG) 2015

As perineal damage carries the potential for negative short- and long-term outcomes (Dahlen & Priddis 2019), maternity care providers attempt to prevent its occurrence in a variety of ways. This may involve manually supporting the perineum during crowning and birth of the fetal presenting part, and/or warm compresses applied to perineal tissues (Dahlen 2012, Kopas 2014, Bulchandani et al 2015, Aasheim et al 2017, Newman 2017).

The aim of these interventions is to reduce perineal trauma and increase maternal comfort (Dahlen et al 2007, Aasheim et al 2017), however, their use is inconsistent amongst midwives, as are the techniques themselves (Begley et al 2019, Healy et al 2020).

Clinical guidance related to intrapartum perineal management, both in Australia and internationally, is provided in a recent Cochrane review which states the interventions described earlier as being supported by moderate quality evidence (Aasheim et al 2017, Queensland Health (QH) 2018, World Health Organization (WHO) 2018).

These interventions are supported and encouraged, however, the guidance fails to recognise their potential impacts on maternal satisfaction and mental health (Lewis et al 2016, Reed et al 2017, Maimburg & De Vries 2019). This, together with the fact that, anecdotally, explicit consent to apply these interventions is not always gained from women is concerning. Having one's perineum touched without express permission has the potential to psychologically traumatise women, or to re-traumatise those with a history of being sexually violated.

Informed consent from women prior to any intervention is understood as foundational to midwifery practice (International Confederation of Midwives (ICM) 2014) and is particularly pertinent given one in six women over the age of 15 has experienced sexual violence (AIHW 2018). The re-traumatisation of women with a lived experience of sexual violence during their pregnancy, birth and the postpartum period is often inadvertently triggered by care providers and has adverse maternal and neonatal impacts, including impaired bonding and attachment (Montgomery 2013, Sperlich et al 2017, Sobel et al 2018). Thus, it is critical that maternity care providers acknowledge the inherently intimate nature of routine interactions (Montgomery et al 2015).

The review of literature aimed to explore what is known about the experiences of women receiving hands-on perineal interventions in the second stage of labour, which was not addressed in the recently published systematic review by Aasheim et al (2017). This information is included in the methods section.

Methods

An approach drawn from the works of Arksey & O'Malley (2005), Levac et al (2010), and Peters et al (2020) was used for this review of literature in which the following framework was followed:

- Identify the research question
- Identify relevant studies
- Study selection
- Charting data
- Collating, summarising and reporting results

Search strategy

The search strategy was adapted from that described by Douma et al (2020) in order to obtain both published research and grey literature, including clinical practice guidelines. A pilot search was not carried out, however the author worked with a specialist librarian to create the search terms.

The search terms outlined in Table 2 were derived from the population (women) and concept (experiences of perineal care in second-stage labour) in focus for this review and were used to search the eight databases listed below. Sources of evidence retrieved from the initial database searches with titles reflecting the review topic were retained and

Table 2. Search terms

OR	Concept 1	Concept 2	Concept 3	Concept 4	Concept 5
	Wom*n OR	"Hands on" OR	Birth* OR	"Heat pack" OR	Interven* OR
	"Wom*ns experienc*" OR	Support* OR	Crown* OR	"Warm compress*" OR	Technique* OR
	"Wom*ns thoughts" OR	"Manual support" OR	"Physiological birth*" OR	"Hot pack" OR	Manage* OR
AND	"Wom*ns think*" OR	Care* OR	"Vaginal birth*" OR	Compress* OR	Prevent*
	"Wom*ns feel*" OR	"Pressure app*" OR	"Natural birth*" OR	Heat*	
	"Matern*comfort" OR	"Guard*perine*" OR	Vagin* OR	"warm pack*"	
	"Wom*ns satisf*" OR	"Control* birth*"	"Second stage labou*" OR		
	"Matern* satisf*" OR		Pushing OR		
	"Wom*n cent*" OR		"2nd stage labo*" OR		
	"Wom*ns views"		Labour OR		
			Labor OR		
			"Preserv* perine*" OR		
			Deliver* OR		
			Childbirth OR		
			Perine* OR "perine* outcomes"		

the full text read to assess relevance for inclusion. The inclusion of grey literature ensured applicable evidence which may not be available through databases was also included.

A systematic search was completed in the Cochrane Database of Systematic Reviews, CINAHL Complete, MEDLINE, PsycInfo, MIC and Google Scholar. A grey literature search was performed in Google and NICE guidance online. All search terms including keywords, Boolean operators and truncation are outlined in Table 2.

Sources of information on the topic of interest written in English and published from 1995–2020 were sought. These date parameters were selected to capture the seminal 'Hands On or Poised' (HOOP) trial (McCandlish et al 1998). No geographical limitations were imposed to ensure a global perspective. Both qualitative and quantitative data were pursued, including those included in systematic reviews to ensure as broad a range of information as possible was included. Forward and backward citation chaining and pearl growing were utilised as these techniques have been shown to be effective complementary literature search strategies (Booth 2008).

Articles were included for review if they were written in English, published from 1995–2020, and were concerned with the topic of interest after title, abstract and full text were reviewed. Articles that did not meet these criteria were excluded at either the title review, abstract review or cursory full text review stage.

As this is a scoping review, formal critical appraisal of the included evidence was not conducted (Levac et al 2010), however, a charting exercise (see Table 3) was performed to capture pertinent information in each data source.

Results/findings

The total number of data sources identified through the database searches as potentially relevant for inclusion, based on their titles, was 344. Complementary search strategies provided one further publication.

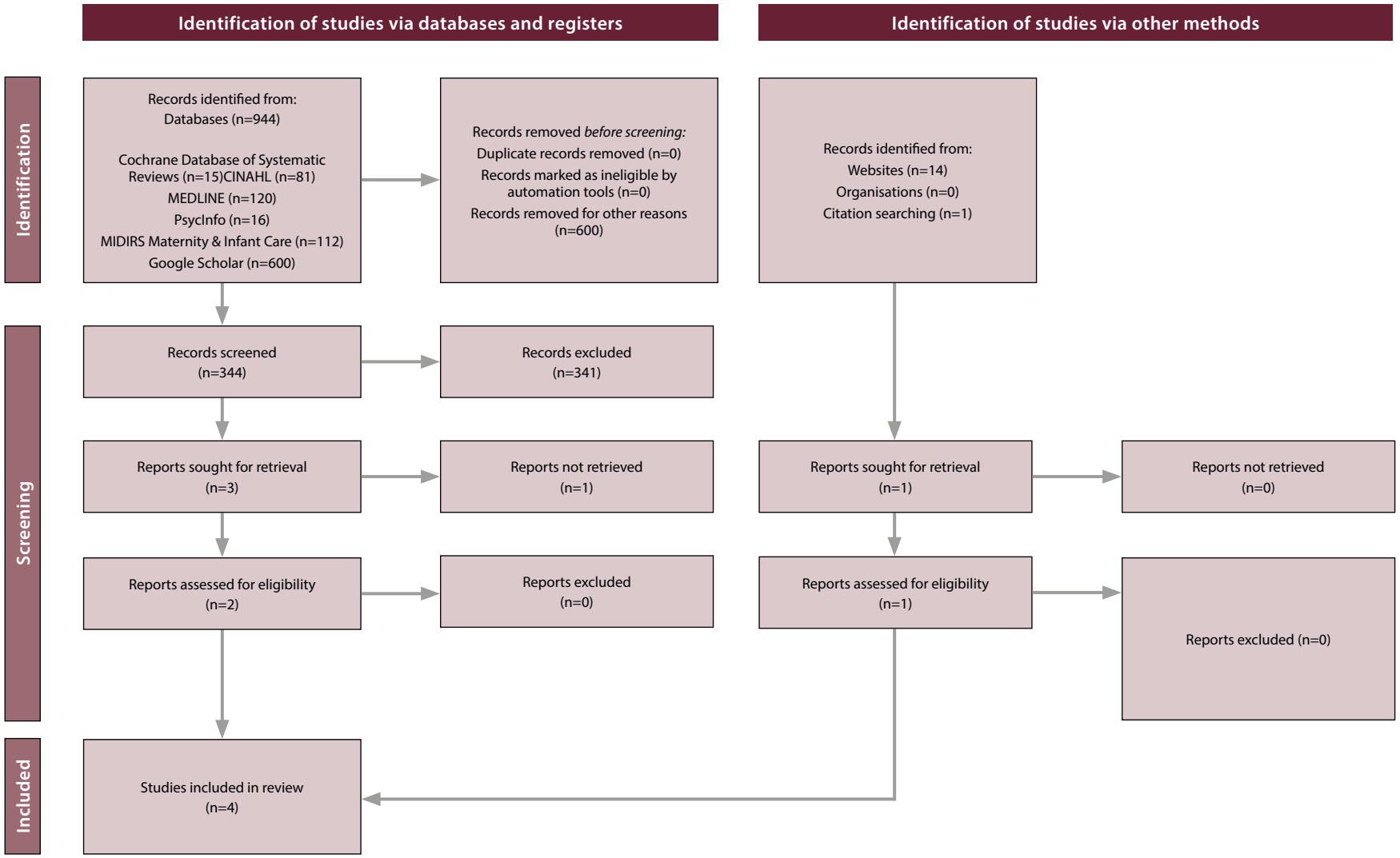
After a cursory full read of each source, 341 were found not to meet the inclusion criteria, leaving four articles for review. One of these was unavailable, and emails to the author were unreturned, resulting in three articles for inclusion. The Cochrane review by Aasheim et al (2017) was included. This inclusion brought the total to four.

Google produced 14 relevant hits, including current clinical guidance from RCOG (2015), WHO (2018) and QH (2018) among others.

Figure 1, the PRISMA flow diagram, shows the data sources identified and included. Tables 4 and 5 detail the full search strategy and results (see Supplementary information).

Evidence sources were included for review if they were written in English, were published between 1995–2020 and investigated the use of perineal interventions in the second stage of labour. Quantitative data via a systematic review were included as these inform current practice guidelines both locally and globally.

The systematic review by Aasheim et al (2017) reports reductions in third- and fourth-degree tears with the use of warm compresses, but is inconclusive regarding manual perineal support, even when reporting increases in episiotomy rates in included studies. Of the remaining three studies, one was a randomised controlled trial (RCT), one was a non-RCT, and one was a qualitative study in conjunction with the included RCT. Overviews of included studies are provided in the charting exercise table (Table 3).



Source: Page MJ et al (2021). The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. *BMJ* 372:n71. For more information: http://www.prisma-statement.org/

Figure 1. PRISMA 2020 flow diagram

Table 3. Charting exercise

Author & year	Aim	Setting & sample	Design	Findings/results
Dahlen et al, 2007	Determine the effects of perineal warm compresses in second stage labour on perineal trauma and maternal comfort	Two maternity hospitals in Sydney, Australia with extensive culturally and linguistically diverse populations. 717 women randomised	Randomised controlled trial, hospitals provided education on intervention procedure, intrapartum and postpartum data collected by midwives, perineal trauma assessed, pain scale and questionnaire completed	No difference between groups in need of suturing (78.6% intervention/79.9% standard care) or episiotomy. Significant difference between groups for severe perineal damage (50% more likely in standard care group). Intervention group less likely to report pain at birth and postpartum
Dahlen et al, 2009	Obtain experiences of women and midwives using perineal warm compresses in second stage labour	Two maternity hospitals in Sydney, Australia. 717 women randomised	As part of the above RCT, women and midwives were asked to complete questionnaires about the effects of perineal warm compresses on pain, comfort, trauma sustained, feelings of satisfaction and control	Perineal warm packs were highly acceptable to women in relieving pain in late second stage labour. 79.7% thought warm packs reduced pain and 89.1% reported receiving comfort from the intervention. 56% reported the intervention helped them feel in control. 1.9% disliked the intervention 'a lot', 6.8% disliked it a little'. 5.3% were unsure if they disliked the warm packs
Essa & Ismail, 2016	Determine the effect of second stage perineal warm compresses on perineal outcomes and pain in primiparous women	Labour and delivery unit of National Medical Institution, Egypt, 160 women included	Non-randomised clinical control trial. Numerical pain rating score and modified behavioural pain scale used. Perineal outcomes documented	Significant reductions in 'severe' pain reported in the intervention group, down from 62.5% to 10% post-intervention
Aasheim et al, 2017	Assess the effect of perineal techniques in second stage labour on perineal outcomes	22 trials eligible for inclusion. 20 trials including 15,181 women total	Systematic review	Moderate quality evidence supports warm compresses in reducing 3rd and 4th degree perineal trauma. Poor quality evidence suggests hands off may reduce episiotomy

In the study by Essa & Ismail (2016) conducted in Egypt, a numerical pain scale, and modified behavioural pain scale were used to determine women's comfort during the application of warm compresses to the perineum. This behavioural scale saw scores assigned to women's behaviour during labour based on the clinician's perception of the amount of noise and grimacing displayed, muscle tension and restlessness, so a true indication of women's experiences is not represented. These authors did not consider the views of women during the intervention, however reductions in pain are reported with use of warm compresses in this study.

Almost 10 years prior to Essa & Ismail's (2016) study, Australian researchers Dahlen et al (2007) conducted a randomised controlled trial to determine the effect of warm compresses applied to the perineum in the second stage of labour on perineal outcomes and maternal comfort. Women were asked to complete a pain scale immediately after giving birth to rate their pain during the intervention, followed by a more in-depth questionnaire within the first day postpartum regarding their experience of pain reduction and comfort and thoughts on acceptability of the intervention. Findings were reported in Dahlen et al (2007) and a subsequent paper published by the same

team (Dahlen et al 2009). Dahlen et al's intervention was associated with a reduction in pain and increased maternal comfort and the results, unlike those reported by Essa & Ismail (2016), were obtained from data collected directly from women.

Discussion

The majority of literature included in this review focuses on physiological outcomes, including degree of perineal damage and suturing requirements, with only one study reporting data directly obtained from women. Furthermore, clinical guidance supported by the Cochrane systematic review conducted by Aasheim et al (2017) encourages the use of these now routine interventions, proposing it is '*therefore likely that women will value any technique that may limit perineal trauma*' (WHO 2018:142).

This focus on anatomy and physiology further highlights a birth culture which does not take into consideration the experiences of women. Limited high-quality evidence exists to support the use of manual perineal support as a means of protecting the perineum, and it is acknowledged this intervention may cause more damage to multiparous women and increase episiotomy rates (Aasheim et al 2017).

Warm compresses are supported by moderate quality evidence in reducing third- and fourth-degree tears, and some women may experience decreased pain and increased comfort with their application (Dahlen et al 2009), however their preparation and use is not homogenous across maternity settings. It has been suggested that coaching a slow birth and appreciating the mechanisms by which birth occurs may be a more appropriate way to reduce perineal trauma while respecting women's autonomy (Maimburg & De Vries 2019).

Conclusion

The aim of this review was to determine the scope and nature of evidence regarding perineal care in the second stage of labour. Limitations include the omission of online discussion forums as a data source and not completing a pilot search.

It is evident from this review that insufficient research exists about women's experiences of perineal interventions during the second stage of labour. This is concerning given the prevalence of perineal interventions in clinical practice, and the prevalence of women who have experienced sexual violence (AIHW 2018, Maimburg & De Vries 2019). Maternity care providers are tasked with providing evidence-based care aimed at improving outcomes for women (ICM 2014). However, many barriers to implementation exist, including lack of organisational support or individual attitudes of clinicians (Greenhalgh et al 2016).

Maternity care providers are accountable to women and have a responsibility to implement evidence-based practice, individualise care, and uphold women's autonomy in order to engender positive maternity care outcomes (ICM 2014). Embracing a TVIC approach may promote this by facilitating the provision of sensitive and equitable maternity care for all women (Sperlich et al 2017).

Although maternity care providers may view an intervention as sufficiently evidence-based to be considered 'routine', the individual needs and preferences of women must be respected (Maimburg & De Vries 2019). There is, at the time of reporting, a dearth of information about women's own views and experiences of perineal interventions. Studies examining the voices of women are essential to making recommendations for care practices and providing clinical guidance, thus further research is needed to understand the wants and needs of women during the second stage of labour.

Studies examining women's experiences of current care practices may assist in the formation of updated guidelines as, although these guidelines ask care providers to seek consent and individualise care, this often does not occur (Reed et al 2017). A paradigm shift in maternity services which incorporates TVIC principles may support this process. TVIC acts to protect vulnerable women with a history of sexual trauma from re-enactment (Montgomery et al 2015), though it may also enhance the provision of sensitive, respectful maternity care for *all* women.

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Dunshore YM (2021). What is known about the experiences of women receiving hands-on perineal interventions in the second stage of labour – a scoping review. *Evidence Based Midwifery* 19(4):13-23.

Supplementary information

Table 4. Database searches

Database: Cochrane Database of Systematic Reviews **Date range:** 1995-2020 Pregnancy and childbirth		
#	**Searches**	**Results**
1	Interven* OR Technique* OR Manage* OR Prevent*	6784
2	"Hands on" OR Support* OR "Manual support" OR Care* OR "Pressure app*" OR "Guard*perine*" OR "Control* birth*"	4739
3	Birth* OR Crown* OR "Physiological birth*" OR "Vaginal birth*" OR "Natural birth*" OR Vagin* OR "Second stage labou*" OR Pushing OR "2nd stage labo*" OR Labour OR Labor OR "Preserv* perine*" OR Deliver* OR Childbirth OR Perine*	2236
4	"Heat pack" OR "Warm compress*" OR "Hot pack" OR Compress* OR Heat* OR "Warm pack*"	297
5	1 AND 2 AND 3 AND 4	53
6	Limiter Pregnancy and Childbirth, Jan 1995-Aug 2020	15
Aasheim, 2017: *Systematic review-perineal techniques during the second stage of labour for reducing perineal trauma*		
Database: CINAHL Complete **Date range:** 1995-2020 TI/AB		
#	**Searches**	**Results**
1	Wom*n OR "Wom*ns experienc*" OR "Wom*ns thoughts" OR "Wom*ns think*" OR "Wom*ns feel*" OR "Matern*comfort" OR "Wom*ns satisf*" OR "Matern* satisf*" OR "Wom*n cent*" OR "Wom*ns views"	397,935
2	"Hands on" OR Support* OR "Manual support" OR Care* OR "Pressure app*" OR "Guard*perine*" OR "Control* birth*"	1,262,604
3	(MH childbirth+)	30,296
4	S2 OR S3	1,285,210
5	Birth* OR Crown* OR "Physiological birth*" OR "Vaginal birth*" OR "Natural birth*" OR Vagin* OR "Second stage labou*" OR Pushing OR "2nd stage labo*" OR Labour OR Labor OR "Preserv* perine*" OR Deliver* OR Childbirth OR Perine*	310,262
6	(MH "Pushing (Childbirth)") OR (MH "Labor Stage, Second") OR (MH "Prepared Childbirth") OR (MH "Vaginal Birth")	4953
7	S5 OR S6	311,298
8	"Heat pack" OR "Warm compress*" OR "Hot pack" OR Compress* OR Heat* OR "Warm pack*"	48,305
9	Interven* OR Technique* OR Manage* OR Prevent*	1,262,906
10	S1 AND S4 AND S7 AND S8 AND S9	84
11	Limiters Jan 1995-Aug 2020, English	81
Dahlen, 2009: *'Soothing the ring of fire': Australian women's and midwives' experiences of using perineal warm packs in the second stage of labour.*		
Database: MEDLINE **Date range:** 1995-2020 TI/AB		
#	**Searches**	**Results**
1	Wom*n OR "Wom*ns experienc*" OR "Wom*ns thoughts" OR "Wom*ns think*" OR "Wom*ns feel*" OR "Matern*comfort" OR "Wom*ns satisf*" OR "Matern* satisf*" OR "Wom*n cent*" OR "Wom*ns views"	1,161,495
2	"Hands on" OR Support* OR "Manual support" OR Care* OR "Pressure app*" OR "Guard*perine*" OR "Control* birth*"	3,061,179
3	(MH "natural childbirth")	2422
4	S2 OR S3	3,063,289
5	Birth* OR Crown* OR "Physiological birth*" OR "Vaginal birth*" OR "Natural birth*" OR Vagin* OR "Second stage labou*" OR Pushing OR "2nd stage labo*" OR Labour OR Labor OR "Preserv* perine*" OR Deliver* OR Childbirth OR Perine*	1,159,303
6	(MH "Labor Stage, Second")	1403
7	S5 OR S6	1,159,435
8	"Heat pack" OR "Warm compress*" OR "Hot pack" OR Compress* OR Heat* OR "Warm pack*"	435,766
9	Interven* OR Technique* OR Manage* OR Prevent*	4,610,838
10	S1 AND S4 AND S7 AND S8 AND S9	180
11	Limiters Jan 1995-Aug 2020, English, human, female	120
	Aasheim, 2017: *DUPLICATE*	

Database: PsycInfo **Date range:** 1995-2020 TI/AB		
#	**Searches**	**Results**
1	Wom*n OR "Wom*ns experienc*" OR "Wom*ns thoughts" OR "Wom*ns think*" OR "Wom*ns feel*" OR "Matern*comfort" OR "Wom*ns satisf*" OR "Matern* satisf*" OR "Wom*n cent*" OR "Wom*ns views"	296,166
2	((DE "Clients" OR DE "Client Characteristics") OR (DE "Mothers")) OR (MM "Physical Comfort")	70,756
3	S1 OR S2	353,894
4	"Hands on" OR Support* OR "Manual support" OR Care* OR "Pressure app*" OR "Guard*perine*" OR "Control* birth*"	1,040,456
5	(DE "Natural Childbirth" OR DE "Birth" OR DE "Labor (Childbirth)" OR DE "Natural Childbirth")	8557
6	S4 OR S5	1,045,973
7	Birth* OR Crown* OR "Physiological birth*" OR "Vaginal birth*" OR "Natural birth*" OR Vagin* OR "Second stage labou*" OR Pushing OR "2nd stage labo*" OR Labour OR Labor OR "Preserv* perine*" OR Deliver* OR Childbirth OR Perine*	202,088
8	(DE "Labor (Childbirth)") AND (DE "Labor (Childbirth)" OR DE "Natural Childbirth")	1078
9	S7 OR S8	202,123
10	"Heat pack" OR "Warm compress*" OR "Hot pack" OR Compress* OR Heat* OR "Warm pack*"	17,994
11	Interven* OR Technique* OR Manage* OR Prevent*	927,350
12	S3 AND S6 AND S9 AND S10 AND S11	20
13	Limiters Jan 1995-Aug 2020, English, human, female	16
ZERO relevant studies		

Database: MIDIRS Maternity & Infant Care **Date range:** 1995-2020 TI/AB		
#	**Searches**	**Results**
1	Wom*n OR Wom*ns experience OR Wom*ns thoughts OR Wom*ns think* OR Wom*ns feel* OR Matern*comfort OR Wom*ns satisf* OR Matern* satisf* OR Wom*n cent* OR Wom*ns views	106,790
2	Hands on OR Support* OR Manual support OR Care* OR Pressure app* OR Guard*perine* OR Control* birth*	89,511
3	(Labour or Childbirth).de.	102,475
4	S2 OR S3	
5	Birth* OR Crown* OR Physiological birth* OR Vaginal birth* OR Natural birth* OR Vagin* OR Second stage labo* OR Pushing OR 2nd stage labo* OR Labour OR Labor OR Preserv* perine* OR Deliver* OR Childbirth OR Perine*	126,289
6	(Labour or Childbirth).de.	18,395
7	S5 OR S6	129,058
8	Heat pack OR Warm compress* OR Hot pack OR Compress* OR Heat* OR Warm pack*	1773
9	Interven* OR Technique* OR Manage* OR Prevent*	74,367
10	S1 AND S4 AND S7 AND S8 AND S9	112
11	Limiters 1995 – 2020	112
Dahlen, 2007: *Perineal outcomes and maternal comfort related to the application of perineal warm packs in the second stage of labor: a randomized controlled study*		

Google Scholar **Date range:** 1995-2020		
	Link to source	**Notes**
	Search 1: perineum ~146,000 results 200 viewed **O included**	
1	https://obgyn.onlinelibrary.wiley.com/doi/full/10.1111/j.1471-0528.1998.tb10004.x	RCT – intervention of either hands on OR hand off during 2nd stage
1	https://pubmed.ncbi.nlm.nih.gov/12092017/	Hands on as "traditional method" V hands poised as "innovative method"
1	https://bmjopen.bmj.com/content/2/5/e001649.short	Discusses incidence of OASI after implementation of training bundle to protect the perineum
1	https://link.springer.com/article/10.1007/s00192-011-1454-8	Estimate of no. of midwives using hands off or hands on in England
1	Manual protection of the perineum reduces the risk of obstetric anal sphincter ruptures	Intervention to reduce OASIS

1	Getting through birth in one piece: protecting the perineum	Discusses 'protective factors' against perineal trauma
1	https://www.sciencedirect.com/science/article/abs/pii/S0266613897900632	Views and practices of Australian midwives
1	https://www.sciencedirect.com/science/article/abs/pii/S1871519217305978	Qualitative midwives' views
1	https://www.sciencedirect.com/science/article/abs/pii/S026661381400179X	Preparation and practice in managing perineum, Australia (Monash) Drs and MWs
	Search 2: perineal support ~120,000 results 100 viewed **2 relevant (DUPLICATES)**	
2	https://obgyn.onlinelibrary.wiley.com/doi/full/10.1111/1471-0528.13431	SR and meta-analysis on 'routine' hands on V hands off/poised/no support
2	https://link.springer.com/content/pdf/10.1007/s00192-016-3176-4.pdf	Perineal support to reduce OASI > increased labial tears and reduction only in 3/4th degree
2	https://www.sciencedirect.com/science/article/abs/pii/S0029784499005608	SR from 2000: Preventing perineal trauma – no mention of compresses
2	https://obgyn.onlinelibrary.wiley.com/doi/full/10.1111/aogs.12547	Hands on recommended by panel of 20 experts
2	https://europepmc.org/article/med/22139117	Suggests support has become the norm in Norwegian settings
2	https://onlinelibrary.wiley.com/doi/abs/10.1016/0091-2182(94)00086-L	Nurse-midwives practice
2	https://www.cochranelibrary.com/cdsr/doi/10.1002/14651858.CD006672.pub3/abstract	**Aasheim, 2017 DUPLICATE** Latest Cochrane SR
2	https://obgyn-onlinelibrary-wiley-com.ezproxy2.acu.edu.au/doi/full/10.1111/aogs.13781	Talks about Finnish and Viennese methods
2	https://www.sciencedirect.com/science/article/abs/pii/S1526952305005714	Describes as 'expectant' V 'interventionist'
2	https://www.sciencedirect.com/science/article/abs/pii/S0301211518309874	Perineal support program in Dutch hospital
2	https://obgyn.onlinelibrary.wiley.com/doi/full/10.1111/ajo.12330	NSW study on hands poised V hands on in midwives
2	https://www.ncbi.nlm.nih.gov/pmc/articles/PMC6205860/	Breaks down each element of a perineal bundle
2	https://www.sciencedirect.com/science/article/abs/pii/S002074891830083X	Australian study to 2016 perineal injury and hand position
2	https://link.springer.com/article/10.1007/s00192-014-2425-7	Role of thumb and finger placement in perineal protection
2	https://www.ncbi.nlm.nih.gov/pmc/articles/PMC3263081/	Hands on vs. hands off + perineal trauma and outcome – better for hands off and less PPH?
2	https://pubmed.ncbi.nlm.nih.gov/18031878/	**Dahlen, 2009 DUPLICATE** – 'Soothing the ring of fire'
	Search 3: perineal compress ~17,800 results 100 viewed 2 relevant **1 included** **1 duplicate**	
3	Effect of second stage perineal warm compresses on perineal pain and outcome among primiparae	**Essa, 2016** Uses VAS pain scale to claim positive impact of intervention
3	Effect of warm compresses versus lubricated massage during the second stage of labor on perineal outcomes among primiparous women	Similar to above – Egypt-based but no pain score
3	https://ebn.bmj.com/content/15/4/103	Dahlen, 2012, reduces 3rd/4th degree and should be part of 2nd stage
3	Perineal outcomes and maternal comfort related to the application of perineal warm packs in the second stage of labor: a randomized controlled study	**Dahlen, 2007 DUPLICATE** talks about maternal comfort and acceptability of intervention with warm compress

Search 4: perineal hot pack ~2460 results 100 viewed	
https://opus.lib.uts.edu.au/handle/10453/29212	Australian thesis – perineal outcomes in Asian women in Australia
Search 5: hands on perineum ~29,600 results 100 viewed **0 included**	
https://link.springer.com/article/10.1007/s00192-011-1454-8	Hands on or off- survey of care in 2nd stage
https://pubmed.ncbi.nlm.nih.gov/12092017/	Traditional care of the perineum
https://pubmed.ncbi.nlm.nih.gov/12092017/	Effect of perineal control
https://pubmed.ncbi.nlm.nih.gov/12092017/	Have we been duped by HOOP?
https://pubmed.ncbi.nlm.nih.gov/12092017/	Promoting normality in second stage
Link not working	Coaching a slow birth may be less harmful than routine hands on

Table 5. Grey literature search strategy

Internet search engine strategy - Google				
	Terms	**#**	**Inc.**	**Links**
1	Perineal support	~ 6,240,000	10	RCOG https://www.rcog.org.uk/en/patients/tears/reducing-risk/ RANZCOG https://ranzcog.edu.au/RANZCOG_SITE/media/RANZCOG-MEDIA/Women%27s%20Health/Statement%20and%20guidelines/Clinical-Obstetrics/RCOG-Management-of-Third-and-Fourth-Degrees-Perineal-Tears.pdf?ext=.pdf WHO https://extranet.who.int/rhl/topics/preconception-pregnancy-childbirth-and-postpartum-care/care-during-childbirth/care-during-labour-2nd-stage/who-recommendation-techniques-preventing-perineal-trauma-during-labour QLD Health CPG https://www.health.qld.gov.au/__data/assets/pdf_file/0022/142384/g-pericare.pdf WHA Collaborative https://women.wcha.asn.au/sites/default/files/docs/wha_national_collaborative_how_to_guide_8.8.2019_0.pdf SA Health CPG https://www.sahealth.sa.gov.au/wps/wcm/connect/76d1590047ff0344affeff21d1663cdf/Perineal+Care_PPG_v1_1_03052018.pdf?MOD=AJPERES&CACHEID=ROOTWORKSPACE-76d1590047ff0344affeff21d1663cdf-n5j4l9d ACSQH lit RV https://www.safetyandquality.gov.au/sites/default/files/migrated/D19-2045-Perineal-tears-lit-review-including-Commission-cover-for-external-publications_Jan-2019.pdf SCV https://www.bettersafercare.vic.gov.au/our-work/clinical-improvement-and-innovation/reducing-perineal-tears Midwife Thinking-Rachel Reed Perineal Protectors https://midwifethinking.com/2016/01/13/perineal-protectors/ AIHW Data Mothers and babies https://www.aihw.gov.au/getmedia/aa54e74a-bda7-4497-93ce-e0010cb66231/aihw-per-108.pdf.aspx?inline=true Sara Wickham perineal support archives https://www.sarawickham.com/tag/perineal-support/
2	Perineal warm packs	~ 745,000		Hannah Dahlen video on application of perineal compress https://www.youtube.com/watch?v=_Up-GKAj_fI&feature=share&app=desktop Clinical Trials.gov https://clinicaltrials.gov/ct2/show/NCT02588508 Hannah Dahlen thesis https://opus.lib.uts.edu.au/bitstream/10453/49236/1/01Front.pdf

Grey literature database search		
Database	**Search terms**	**Link**
NICE guidelines	Perineal care	https://www.nice.org.uk/guidance/cg190/chapter/Recommendations#second-stage-of-labour

Hand searching results		
Author/year	**Keywords**	**Notes**
Musgrove – emailed with no reply	RCT perineal preservation	Perineal preservation and heat application during the second stage of labour

Female childhood cancer survivors and the impact of flank, abdominal or pelvic radiotherapy on live birth rates: a systematic review and meta-analysis

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ABSTRACT

Population: Adult female survivors of childhood, adolescent, and young adult cancers (childhood cancer survivors (CCS)) have a survival rate of up to 84 per cent, with many now able to maintain fertility into adulthood.

Intervention: This review critically appraised and synthesised evidence for live birth rates and adverse pregnancy outcomes of female CCS (aged 0–24 years at diagnosis), treated with radiotherapy to the abdomen, flank, or pelvis.

MEDLINE, PubMed, CINAHL, Google Scholar, Trip, Scopus, and ProQuest were searched on 30 September 2017 and on 11 June 2020. Studies were subjected to inclusion and exclusion criteria with secondary independent review. Patient and Public Involvement and Engagement (PPIE) was used to assist in the selection of outcomes.

Data were analysed using *EPPI-Reviewer 4* software. Risk of bias was assessed using the Newcastle Ottawa scale (NOS). Meta-analysis used a random effects model (DerSimonian & Laird 1986) with parameters of heterogeneity set at I^2 of >50% and a chi-squared p value of <0.05 using *RevMan 5* software. The review adhered to PRISMA (Moher et al 2009) reporting guidelines and flow chart.

Comparator: Sibling control groups and/or general population controls with no history of treatment for childhood cancer, where available.

Outcomes: The database search yielded 1495 studies; 1289 screened for title and abstract; 26 screened as full text; eight used for meta-analysis. Upon aggregation (11 June 2020), one study was added and used in the meta-analysis (total n=9 used for meta-analysis).

Female CCS who received radiotherapy to the flank, abdomen or pelvis had increased odds of premature birth (<37 weeks gestation) (OR 3.69 CI [2.82, 4.81] p=<0.00001) and miscarriage (OR 1.59 CI [1.37, 1.84] p=<0.00001), when compared to CCS that had not had radiotherapy. CCS exposed to radiotherapy had increased odds of stillbirth (OR 1.72 [1.08, 2.74] p=0.02) when compared to non-CCS controls. Data for live birth rates were not analysed due to heterogeneity and control group variance.

Female CCS warrant high-risk antenatal care and ongoing surveillance throughout pregnancy. Further research investigating toxic thresholds of the uterus is recommended. Female CCS require detailed communication of future pregnancy risk before pregnancy occurs.

Ethical approval: This study was approved by Coventry University Ethics, project numbers P46688 and P60599.

Trial registration: The protocol was registered with PROSPERO (CRD42017054533) (https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=54533)

Keywords: Childhood cancer survivor, adult cancer survivor, childhood neoplasms, survivorship, pregnancy, birth, live birth, adverse outcome, radiotherapy, Evidence Based Midwifery

Background

The survival rate for children, adolescents and young adults with cancer is approximately 84 per cent at five years following diagnosis (Cancer Research UK 2021). However, cancer treatments given to a child are known to cause significant long-term, chronic health problems in adulthood (Children's Cancer and Leukaemia Group 2021).

Many female childhood cancer survivors (CCS) now maintain their fertility and can go on to conceive a natural pregnancy (van de Loo et al 2019, van der Kooi et al 2019). The number of CCS in the United Kingdom (UK) is currently estimated to be around 35,000 (Cancer Research UK 2021) and rising. Therefore, it is important to consider the long-term health outcomes of CCS, including reproductive outcomes for future pregnancy and birth. Risk of adverse outcome linked to prior cancer treatments merits further investigation with a need to increase awareness of health care professionals, female CCS, and their families about potential risks (van de Loo et al 2019, van der Kooi et al 2019).

Treatment for childhood cancer often requires a combination of chemotherapy, radiotherapy, surgery, and immunotherapy treatments (Children's Cancer and Leukaemia Group 2021). The use of radiotherapy to the flank, abdomen or pelvic areas is dependent on the site, stage, and type of tumour (Children's Cancer and Leukaemia Group 2021). Tumours that often require radiotherapy to the flank, abdomen or pelvis include Wilms' tumour, neuroblastoma, leukaemia (when total body irradiation is used), Hodgkin lymphoma, rhabdomyosarcoma, and germ cell tumours (Children's Cancer and Leukaemia Group 2021).

Radiotherapy delivered to the uterine area in a child that has yet to reach puberty, has been reported to increase the likelihood of abnormal organ development and growth; resulting in an inability to carry a pregnancy to full term (van der Kooi et al 2021, van de Loo et al 2019, Larsen et al 2004). This increases the likelihood of adverse pregnancy and birth outcomes, such as preterm birth (<37 weeks gestation) (van de Loo et al 2019, van der Kooi et al 2019).

The pre-menarche uterus has been reported to be progressively radio-sensitive, which increases the risk of abnormal development in adolescence (Larsen et al 2004). Furthermore, van de Loo et al (2019) reported that female CCS treated with radiotherapy to the abdomen had lower uterine volumes than general population controls and a higher risk of premature labour. Additional radiotherapy-induced malformations of the uterus have been reported as abnormal placental formation, abnormal conversion of uterine spiral and distal arteries, and abnormal placentation (placenta previa, percreta or accreta) (Lie Fong et al 2010). Female CSS treated with radiotherapy to the abdominal area were also found to be at risk of uterine rupture and cervical insufficiency (Lie Fong et al 2010, Reulen et al 2009).

Research to determine the causal link between level of radiotherapy treatment received and risk of adverse outcomes in pregnancy and birth for female CCS is lacking (Reulen et al 2009). The voice of the female CCS and their families within the design, prioritisation and dissemination of research in this area is also absent. This systematic review highlights the need for embedded Patient and Public Involvement and Engagement (PPIE) in CCS research. Likewise, the ability to use PPIE within a systematic review design has been demonstrated by this review, illustrating how PPIE can be embedded into any type of research. To achieve this, data were collected from female CCS and their parents using an online survey to determine patient/parent research priorities for the area of future treatment-related pregnancy risks. Data obtained from the online surveys were compared to the selected primary and secondary outcomes to ensure compatibility. This approach aimed to ensure that the focus of the review aligned with the patient-reported research priorities and needs.

The aim of the review was to evaluate, appraise and synthesise the existing data for live birth rates of female CCS who received flank, abdominal or pelvic radiotherapy as treatment for childhood cancer. The results aimed to provide health care professionals and female CCS with an additional evidence base when planning a pregnancy or accessing maternity services.

Methods

The PRISMA flow chart and reporting checklist were used to structure the systematic review and meta-analysis (Moher et al 2009). Ethical approval was gained from Coventry University Ethics Committee (P46688 and P60599) and the systematic review was registered with PROSPERO (CRD42017054533). An extensive bibliographic search was conducted, and risk of bias performed using the Newcastle Ottawa Scale (NOS) (Wells et al 2018). Meta-analysis of outcomes was undertaken if more than three reports with the same clinical outcome and population/comparators were found.

Review questions

1. What is the impact of flank, abdominal or pelvic radiotherapy given to female childhood/adolescent/young adult cancer survivors upon subsequent live birth outcomes?
2. Are there any identified perinatal risks directly attributable to radiotherapy to the flank, abdomen, or pelvis as a child/adolescent/young adult?

Study selection criteria

Inclusion criteria:

- Women who had given birth (aged ≥16 years)
- Women who received a diagnosis of cancer as a child or adolescent/young adult (up to age 24 years inclusive) who had flank, abdominal or pelvic radiotherapy as part of their treatment
- Naturally occurring pregnancy without fertility treatment, including in-vitro fertilisation (IVF)
- Pregnancy, not within one year of active cancer treatment

Sources were selected from recognised data registries, from the United States of America (USA), Australia, Canada, and other European Union (EU) member countries. Studies were cohort or case-controlled by design and published in English. Control or comparator groups were deemed eligible for data correlation/comparison if derived from non-cancer affected siblings/general population or non-radiotherapy exposed CCS.

Exclusion criteria:

- Male CCS
- Surrogate pregnancies of CCS
- Females treated for adult cancer >age 25 at diagnosis
- Females treated for cancer during pregnancy or pregnancies <1 year from end of treatment
- Pregnancies achieved using artificial reproductive techniques, such as IVF
- Female CCS treated with radiotherapy to other areas of the body, or where treatment site or type were not able to be extrapolated from data
- Data that could not extrapolated to distinguish number of male/female CCS

These exclusion criteria were applied to ensure coherent and consistent analysis of variables in this particular field of research, that is, health care systems and access to health care comparable to those in the UK.

Search approaches

A search of MEDLINE, PubMed, CINAHL, Google Scholar, Scopus, Trip and ProQuest databases was performed on 30 September 2017 with an aggregate review conducted on 11 June 2020. The databases were selected to ensure a wide representation of studies from the nursing, oncology, obstetric and psychological disciplines. The reference lists of included studies were scanned, forward-cited and back-referenced.

All titles and abstracts were scanned by AP. Those not deemed ineligible were further assessed in full-text format. A selection of 10 per cent of the titles/abstracts was screened by NA. This also applied to papers selected for full-text assessment. Conflicts in inclusion of studies to the review were not found. However, a third independent reviewer was available throughout the process to ensure methodological compliance. PRISMA reporting guidelines were followed and a modified Cochrane data extraction template used. Risk of bias and data extraction was checked by NA prior to meta-analysis.

Outcome selection

An online pseudo-anonymised survey of 12 questions was completed by 26 female CCS (aged ≥16 years) who had given birth to a child, and parents of female CCS survivors yet to conceive a child. Participants were asked to complete the short survey and rank the importance of selected outcomes for the review. *Qualtrics XM* software was used to design the survey and collect data.

Participants for the PPIE survey were recruited via an invitation posted on CCS online support groups and social media platforms. The results of the survey were used to verify the selected primary and secondary outcomes of the review.

Outcomes

Primary outcome:

- Live birth at term (37 weeks of completed pregnancy)

Secondary outcomes:

- Pregnancy outcome (live birth, miscarriage, stillbirth, neonatal death up to 28 days and intrauterine death), premature birth (24 weeks to 36+6 weeks' gestation), fetal growth restriction (below tenth centile of predicted growth projection), low birth weight (<2.5kgs at birth), caesarean section rate (elective or emergency), onset of labour type (spontaneous, induced or augmented), uterine dysfunction (defined as delayed first stage requiring syntocinon augmentation and postpartum haemorrhage) and neonatal congenital abnormality.

Data analysis

A Cochrane data extraction template was modified to enable collection of data including cancer type, treatment and dose, age at treatment, ethnic background, age at pregnancy and other adverse obstetric events. Raw binary data were extracted from the individual studies for outcomes by the first reviewer using a 2x2 contingency table. If raw data could not be found within the paper the authors were contacted to provide this information, with studies excluded for meta-analysis if no response was received or data were unobtainable two weeks after the request.

Data synthesis

Outcome measures from included studies were recorded, tabulated and meta-analysis subsequently considered. Narrative review was used for data from outcomes deemed too heterogeneous for meta-analysis and sub-group analysis was planned for, but not undertaken, due to limitation of available studies and heterogeneity of studies.

Meta-analysis was undertaken using *RevMan 5* software and a random effects model. This represented the belief that true effect size might differ from study to study due to variables in population demographics. An initial consideration of clinical homogeneity was undertaken to decide if an outcome was matched in the data of at least three of the included studies (including control group data). If this was not apparent, then meta-analysis was not undertaken.

Risk of bias assessment

This was assessed at the individual study level using the NOS Scale (Wells et al 2018) by AP and NA. Studies were categorised, with the most robust studies (highly assessed and rigorous research) achieving up to nine stars. High-quality studies were defined as those which had achieved a score of seven or more, based on similar NOS categorisations used in systematic reviews of this kind (Kabak et al 2019). Risk of bias across studies was not assessed due to methodological heterogeneity.

Results

PPIE survey

The PPIE online survey was completed by 24 participants using the *Qualtrics XM* software survey tool. The demographic background of the participants represented 19 parents of children who had had cancer, three female CCS who had given birth and two female CCS who had not had a child. Sixteen participants recorded that they/their child had received radiotherapy to the 'tummy'. The three top-ranking future pregnancy concerns were identified as:

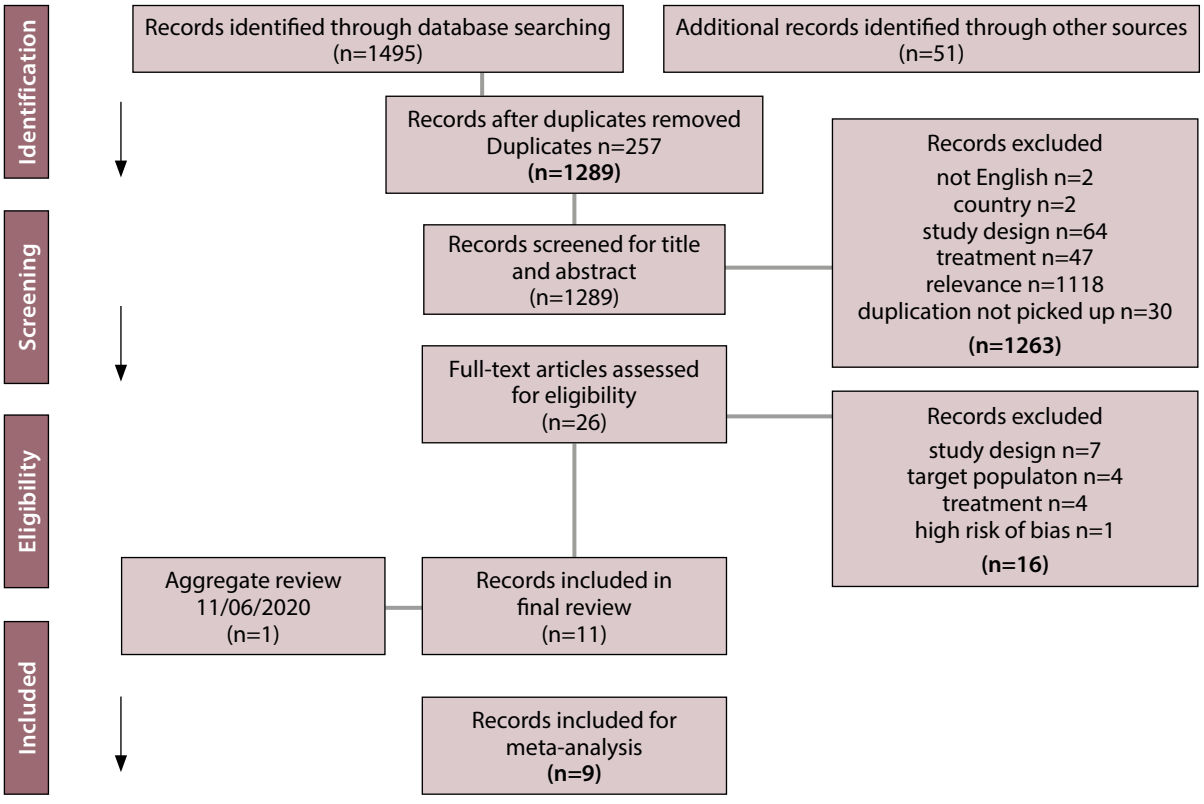
1. Risk of pregnancy complications in the mother
2. Risk of miscarriage and abnormality in the baby
3. Risk of early labour

The outcome of 'live birth' was not directly asked, however the term 'a healthy baby' was included and ranked fourth in the survey.

Review results and meta-analysis

The database search identified 1495 records, 26 were reviewed in the full text and 10 included in the final review (see Figure 1). Following aggregate review on 11 June 2020, a further study was added to the final number (final number n=11 included and n=9 used for meta-analysis). Data from this study were added to the existing summary table and meta-analysis repeated to include the new data. Two studies were excluded for meta-analysis due to unavailability of raw data.

Figure 1. PRISMA flow diagram



Source: Mother et al 2009.

The final included studies consisted of 11 retrospective cohort studies; a summary of the included studies is in Supplementary information. The studies represented recognised data registries including the British Childhood Cancer Survivor Study (BCCSS) (Hawkins et al 2008) and the Childhood Cancer Survivor Study (CCSS) (Robison et al 2002).

Several of the studies used medical records to corroborate patient-reported outcomes, however authors included missing treatment data where possible. The included studies varied in population size from less than 1000 to more than 34,000. All included studies had one or more comparator control groups. Data were provided in three studies for two different categories of control (non-CCS sibling and general population controls) (Green et al 2002, Winther et al 2008, van de Loo et al 2019). Sample sizes ranged from <1000–3000+ and included a variety of convenience, purposeful (sibling matches) and random (data linkage comparisons from data registry) sampling methods.

Of the 11 included studies, five studies provided data for CCS exposed versus CCS non-exposed groups and eight studies provided data for the general population or sibling matches. The outcomes reported by the authors included an extensive list of obstetric pregnancy and birth complications, such as anaemia, gestational diabetes, pre-eclampsia, neonatal and fetal complications, live births, and pregnancy loss data. Detailed social demographics of the population were rarely reported, and control/comparison groups were not always matched rigorously within the CCS radiotherapy-exposed data sets. Sub-group analysis was not possible due to sample heterogeneity.

All 11 studies were classified as 'high' quality based on the NOS risk of bias assessment scale (defined in this review as a score of over seven stars). Meta-analysis was possible for five outcomes. Three outcomes, live birth (childhood cancer survivors who had radiotherapy versus survivors that did not), live birth (childhood cancer survivors who had radiotherapy versus a non-childhood-cancer-affected control group) and low-birth weight (<2.5kgs) (childhood cancer survivors who had radiotherapy versus survivors who did not have radiotherapy), did not meet the criteria for heterogeneity (I^2 result of >50% or χ^2 result with a p value significance of <0.05).

Three of the meta-analysis outcomes, premature birth, stillbirth, and miscarriage met the criteria of significance. Increased odds of premature birth (<37 weeks gestation) (OR 3.69 CI [2.82, 4.81] p=<0.00001) (Table 1) and miscarriage (OR 1.59 CI [1.37, 1.84] p=<0.00001) (Table 2) were found when female CCS exposed to radiotherapy were compared to non-exposed CCS. Female CCS exposed to radiotherapy to the abdominal area were also found to have increased odds of stillbirth (OR 1.72 [1.08, 2.74] p=0.02) (Table 3) when compared to non-CCS controls. This supports the findings of Signorello et al (2010) who reported that radiotherapy to the uterine area significantly increased the risk of stillbirth and neonatal death of female CCS when delivered at doses greater than 10 Gray (Gy).

Table 1. Premature birth (CCS exposed versus non-exposed CCS)

Study or Subgroup	CCS exposed Events	Total	non-exposed CCS Events	Total	Weight	Odds Ratio M-H, Random, 95% CI
Green et al 2010	62	437	19	480	25.2%	4.01 [2.36, 6.83]
Reulen et al 2009	90	419	95	1327	72.6%	3.55 [2.59, 4.85]
van de Loo et al 2019	6	8	9	24	2.2%	5.00 [0.83, 30.28]
Total (95% CI)		864		1831	100.0%	3.69 [2.82, 4.81]
Total events	158		123			

Heterogeneity: Tau² = 0.00; Chi² = 0.27, df = 2 (P = 0.87); I² = 0%
Test for overall effect: Z = 9.59 (P < 0.00001)

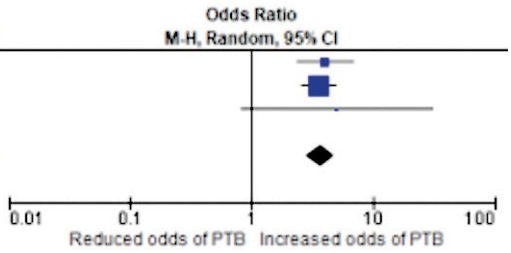


Table 2. Miscarriage (CCS exposed versus non-exposed CCS)

Study or Subgroup	CCS exposed Events	Total	CCS non-exposed Events	Total	Weight	Odds Ratio M-H, Fixed, 95% CI
Green et al 2002	463	1886	239	1441	70.9%	1.64 [1.38, 1.95]
Reulen et al 2009	96	413	209	1213	28.3%	1.45 [1.11, 1.91]
van de Loo et al 2019	4	10	7	26	0.8%	1.81 [0.39, 8.39]
Total (95% CI)		2309		2680	100.0%	1.59 [1.37, 1.84]
Total events	563		455			

Heterogeneity: Chi² = 0.54, df = 2 (P = 0.76); I² = 0%
Test for overall effect: Z = 6.21 (P < 0.00001)

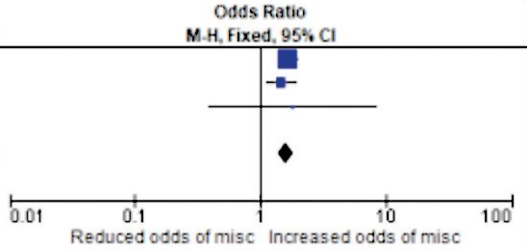
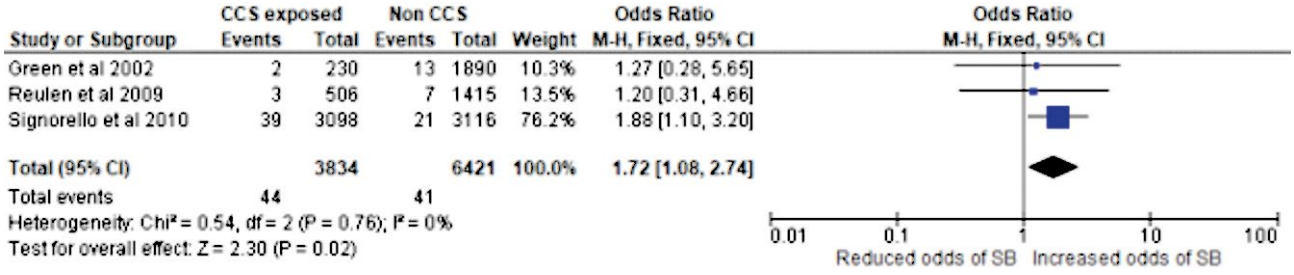


Table 3. Stillbirth (CCS exposed versus non-CCS controls)



A narrative synthesis of the remaining data for additional adverse outcomes (see secondary outcomes) revealed congenital abnormalities were no more likely to occur in pregnancies of female CCS treated with radiotherapy to the flank, abdomen, or pelvis (Mueller et al 2009, Green et al 2010, Haggar et al 2014).

Notably, female CCS with this treatment exposure history were found to have an increased risk of low-birth-weight babies (Green et al 2002, Signorello et al 2006, Reulen et al 2009, Mueller et al 2009, Haggar et al 2014, van de Loo et al 2019), however meta-analysis was not possible due to control group variance. This increased risk was not reflected in the limited data for small-for-gestational-age babies, supporting the theory that radiotherapy damage is linked to abnormal pathophysiology of the uterus. Extensive control group variation between the studies prevented meta-analysis for any additional outcomes, as identified in the secondary outcomes of this review.

Notably, the narrative data within the included studies, were highly suggestive of an increased risk of maternal and fetal adverse outcomes, such as hypertension (OR 3.6) or pre-eclampsia (OR 17.07) (Green et al 2010, Lie Fong et al 2010). Birth complications, such as malposition of the fetus (OR 4.06), haemorrhage (OR 9.16) and manual removal of the placenta (OR 6.71) (Green et al 2010, Lie Fong et al 2010, Reulen et al 2017).

Meta-analysis was not possible as data were not reported within three studies or more, however further research is recommended to evaluate this potential risk. Future prospective longitudinal research with comparable control and CCS controls would be advised to provide more rigorous comparisons and evidence to support a correlation with these life-threatening perinatal complications.

Discussion

This review reported a link between female CCS treated with radiotherapy to the abdomen, flank or pelvis as a child/adolescent/young adult, and increased odds of premature birth, stillbirth, and miscarriage. The results from this review support a growing body of evidence for health care professionals responsible for the obstetric care of female CCS (van der Kooi et al 2021). Despite this evidence base, an increased awareness of future pregnancy and childbirth risk by health care professionals is needed, alongside a multi-disciplinary communication model to facilitate optimal management of CCS in pregnancy and birth.

Implications and recommendations for maternity care

A high-risk pregnancy care plan is recommended for female CCS, in particular if exposed to radiotherapy to the abdomen, flank or pelvis as a child. This recommendation should be communicated to the patient and their family as early as possible in pregnancy to allow for timely referral to a high-risk obstetric team.

The recent publication by van der Kooi et al (2021) supports this recommendation and provides an excellent example guidance document for health care professionals. However, the guidance by van der Kooi et al (2021) gives no specific clinical interventions or surveillance methods. The guidance also has limited generalisability when applied to National Health Service (NHS) maternity care systems and the UK midwifery care model for pregnancy. Wallace et al (2013) produced guidance to assist with the risk-stratification of CCS in Scotland, highlighting pregnancy as a notable risk. However, a more specific and collaborative care pathway is needed to reflect patient need and encourage multi-disciplinary working and early referral for expert advice.

Obstetric and maternity care providers should consider the evidence and implement increased surveillance and/or interventional measures for radiotherapy-exposed female CCS in pregnancy. This may help to reduce the risk of adverse outcomes such as miscarriage, premature birth, and stillbirth. Examples of such interventions include early pregnancy ultrasound scanning, serial cervical length assessment and early induction of labour. However, further evidence is needed to assess the need and impact of such interventions upon adverse outcomes.

Likewise, the increased odds of premature birth, miscarriage and stillbirth in this patient group highlights potential health economic ramifications. Prevention of miscarriage, preterm birth and stillbirth have all been identified by the NHS as key priorities for improvement in the health care of pregnant women (National Institute for Health and Care Excellence (NICE) 2015, NHS England 2016a,

2016b). Health care arising from a premature birth costs the NHS millions per year and extends to costs associated with maternal psychological distress, birth trauma, infection, perineal trauma, and post-natal depression (NICE 2015, NHS England 2016a, 2016b, Tommy's 2018a, 2018b). Therefore, the increased risk of premature labour in female CCS warrants further consideration and economic analysis by maternity services. Furthermore, more qualitative studies in this area would be recommended to explore and evaluate the psychological experiences surrounding future pregnancy and birth of female CCS.

However, achieving a tailored pregnancy care package for CCS is a challenge for maternity services already under pressure with an increasing number of women with highly complex medical histories and multiple co-morbidities requiring an individualised and multi-disciplinary care approach (NHS England 2016a, 2016b). CCS are an increasing population, with multi-variate individual and complex needs (Reulen et al 2009). CCS are also 60 per cent more likely to have a co-morbidity related to their prior cancer treatment which, in turn, is amplified if they have received radiotherapy (Aslett et al 2007). This puts female CCS into a high-risk population.

Clinical guidelines for women in pregnancy with complex medical histories has led to a marked reduction in perinatal morbidity, stillbirth rates and maternal deaths (for example, guidelines for epilepsy, congenital heart conditions) (NHS England 2020). This success in the reduction of adverse outcomes for complex health populations serves as an exemplar and demonstrates the need for an evidence-based guideline for this patient group.

Maternity care professionals must actively support and advocate birth choices of women in pregnancy, even in cases of complex medical and psychological need (NHS England 2016a, 2020). The needs of the woman and her family should be upheld to ensure that women are cared for, and give birth in the right place, at the right time, with the right professional leading their care; a model advocated by the Maternity Transformation Programme (NHS England 2020). Despite this, no referral care pathway or clinical guideline exists for women with a history of CCS in pregnancy. A clinical guideline for maternity care in collaboration with NICE, the Royal College of Midwives (RCM) and the Royal College of Obstetricians and Gynaecologists (RCOG) is needed to support the standardisation of maternity care for this patient group, based upon existing evidence and the results of this review.

Additionally, the communication of risk for future adverse outcome in pregnancy and birth for female CCS exposed to radiotherapy to the abdomen, flank or pelvis, is an important issue to be addressed by further research. Female CCS and their families should be informed and empowered to be active partners in their pregnancy care. They should also be provided with a full clinical picture of evidence-based research to make informed health care choices. The communication of potential risks should take place during or after cancer treatment, in the pre-conception period and/or very early in pregnancy. This recommendation is supported by van der Kooi et al (2021) and van de Loo et al (2019). Further research exploring how female CCS feel about future pregnancy and birth after treatment is also needed to provide health care professionals with context from which to design a patient-centred care pathway.

Research into toxic radiotherapy thresholds of the uterus is lacking, as demonstrated in the review (Reulen et al 2009). This evidence is needed to ensure that female CCS survivors treated with radiotherapy can be risk-stratified. This would also ensure that any obstetric interventions or enhanced surveillance during pregnancy are directed only to those at very high-risk of complications.

Limitations and strengths

This review reported no significant result for the primary outcome 'live birth' (due to insufficient and heterogeneous data). 'Live birth' was found to be under-reported or unclassified as an outcome within the data. This might be explained by a historical tendency in quantitative research to measure or report solely adverse event outcomes within a patient group, however this assumption has not been explored (Smyth et al 2011). Risk of bias assessment utilised a recognised tool suitable for the assessment of cohort studies of population-based cohorts. However, there is a possibility that relevant studies were not included due to the extensive inclusion criteria or terminology of the key words used within the databases. The risk of publication bias is also present due to only including peer-reviewed journal studies.

Up-to-date treatment

Evidence surrounding the pregnancy and birth outcomes of female CCS is limited in quantity and quality and bears little resemblance to modern treatments for childhood/young adult cancers. Included studies in the review acknowledged missing data in their results, pertaining to treatment modality and correct dosage information.

Likewise, data included in this review rely heavily on self-reported patient outcomes. Self-reported outcome data collection facilitates recruitment of adequate sample sizes; however it is often criticised due to the potential for significant recall bias of participants (for example, participants were typically asked to recall information about miscarriages and pregnancies via questionnaire) (Overbeek et al 2012). This could lead to data being reported that are not representative of the population and/or significant loss of data and ungeneralisable results (Overbeek et al 2012).

Ambiguous data surrounding radiotherapy toxicity thresholds for organs such as the uterus, and the associated risk of adverse effects in future pregnancy and birth, suggests a need for more in-depth research, reflective of up-to-date treatments for children with cancer.

Data registries used in this review represent the cohorts from the BCCSS (Hawkins et al 2008), CCSS (Robison et al 2002) and the Dutch DCOG LATER-VEVO study (Skion LATER 2020). In the CCSS a large cohort of 20,276 eligible five-year survivors of childhood and adolescent cancer were recruited (with a diagnosis prior to age 21 years between 1970–1986). The CCSS study addressed important long-term health issues related to treatment for CCS in the USA (Robison et al 2002). The UK registry, the BCCSS, aimed to determine the risks of adverse health and social outcomes among childhood cancer survivors diagnosed between 1940 and 1991, and who had survived five years. The BCCSS cohort of 17,981 forms the basis of many population-based studies of late mortality, including the risks/causes of second malignant neoplasms by using national registration systems (Hawkins et al 2008). The Dutch registry counterpart is more recent, with 1944 CCS recruited with a data collection period of 2008–2014 (Skion LATER 2020).

The cancer registry data sets discussed above, although vast and detailed, do not adequately reflect recent novel treatments, dosages, or risk stratifications. Patient cohorts were relatively young when data were collected, limiting data from CCS of reproductive age and their reproductive outcomes. A more recent or prospective data collection method for CCS and subsequent reproductive outcomes would be beneficial.

Patient and Public Involvement and Engagement

Patient and Public Involvement and Engagement (PPIE) is rare within a systematic review. Traditionally, research studies perform an analysis of secondary data sources to answer a research question, with the aims of the study being completely researcher driven. However, clinical academic researchers are now encouraged to prioritise the needs and views of patients within their research design, allowing for faster translational impact of their results into clinical practice (Gray 2017). This patient-driven, collaborative approach to research is also supported by the James Lind Alliance (James Lind Alliance 2021). The Alliance leads priority-setting partnerships for areas of need and works together with multi-disciplinary and multi-stakeholder groups to prioritise research questions of direct relevance to patients (James Lind Alliance 2021). Using a collaborative PPIE approach within research methodologies, including systematic reviews, helps to improve the applicability, relevance and justification of important research questions and is recommended for future studies of this kind.

This review utilised a collaborative PPIE approach, to ensure that the selected outcomes of the review were of direct relevance to the patient group. However, it is acknowledged that PPIE inclusion could have been further embedded to include activities within the review team, the development of the research question and in the oversight of the meta-analysis process.

Conclusion

Female CCS who have received radiotherapy to the flank, abdomen or pelvis as a child have increased odds of premature birth (<37 weeks gestation) (OR 3.69 CI [2.82, 4.81] p=<0.00001) and miscarriage (OR 1.59 CI [1.37, 1.84] p=<0.00001) when compared to CCS not exposed to radiotherapy. CCS exposed to radiotherapy to this area as a child also have increased odds of stillbirth (OR 1.72 [1.08, 2.74] p=0.02) when compared to non-CCS controls.

Female CCS treated with radiotherapy to the flank, abdomen or pelvis warrant early high-risk antenatal care referral and ongoing surveillance throughout pregnancy. Female CCS and their families require detailed communication of future treatment-related pregnancy risk from health-care professionals when considering pregnancy in adulthood.

Further investigation into the toxic radiotherapy thresholds of the uterus is needed to ensure that female CCS survivors can be risk-stratified for obstetric interventions in future pregnancy care. This in turn will assist to achieve optimal outcomes and shared decision making for female CCS in future pregnancy and birth.

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Conflicts of interest

We declare no competing interests or conflicts of interest. The author declares no permissions required or commercial affiliations. Supporting data related to this review can be obtained by emailing the corresponding author.

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Ethical approval

This study was approved by Coventry University Ethics, project numbers P46688 and P60599.

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Contributions

AP conducted the review and PPIE survey, data collection and analysis, statistical analysis and writing of the review. EB provided support as director of studies and preparation and review of the manuscript. JC and BP reviewed the manuscript and provided supervisory support to the review, including the first draft of the publication. NA contributed as the second reviewer of data.

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Supplementary information

Summary table of included studies

* Low birth weight (LBW) <2500grams, ^ premature birth <37 weeks gestation, ~ small for gestational age (SGA) <10th birth percentile, † all CCS vs siblings (not treatment-specific), ‡ premature rupture of membranes (PROM), § gestational diabetes mellitus (GDM).

Author	Cases n=	Control n=	Comparator n=	Control/ comparison background	Outcomes	Crude data cases	Crude data control	OR	RR	Main findings
Green et al 2002	4029	1903	1680	Siblings	**Sibling control**					• Overall no significant results in outcome by treatment • Higher risk of miscarriage for female CCS treated with ovarian radiotherapy • Female CCS treated with radiotherapy more likely to have LBW infant
				CCS exposed vs CCS non-exposed	Live birth	131/101	1349/554	0.53	0.79	
					Stillbirth	2/230	13/1890	1.26	1.26	
					Miscarriage	46/186	279/1624	1.43	1.35	
					LBW†	172/2376	36/1313	2.64	2.52	
					Abortion	39/193	220/1683	1.54	1.45	
					CCS exposed vs CCS non-exposed					
					Live birth	1472/877	1076/604	0.91	0.97	
					Miscarriage	463/1886	239/1441	1.48	1.38	
					Stillbirth	25/2326	14/1666	1.27	1.27	
					Abortion	460/1889	304/1376	1.1	1.08	
Green et al 2010	312	0	187	CCS exposed vs CCS non-exposed	Live birth	312/187	187/312	2.78	1.66	• Increase in LBW and premature labour increased in female CCS who received radiotherapy • No trend found for risk of congenital abnormalities • Increased risk of hypertension, malposition of fetus for female CCS who received radiotherapy (increased with radiation dose)
					Hypertension	74/425	23/476	3.6	3.21	
					Premature labour	79/420	28/471	3.16	2.82	
					Malposition	31/468	8/491	4.06	3.87	
					Obstructed labour	23/476	12/487	1.96	1.91	
					Abnormality of force	18/481	14/485	1.29	1.28	
					Cord complications	77/422	36/463	2.34	2.13	
					Premature birth	62/437	19/480	3.58	3.26	
					PROM‡	11/488	11/488	1	1	
					Congenital abnormality	28/284	16/171	1.06	1.04	

Reulen et al 2017	2783	25000	0	General population	Hypertension (pre-existing and non)	101/2682	2508/22492	0.33	0.36	• 3-fold increase in hypertension found in female CCS who received radiotherapy • Increased risk of GDM and anaemia found for female CCS treated with radiotherapy • Female CCS more likely to have caesarean section
					Live birth (exposed CCS vs non-exposed CCS)	326/2457	860/1923	0.29	0.37	
					GDM§	16/2767	390/24610	0.36	0.36	
					Anaemia	27/2756	1099/23901	0.21	0.22	
					Growth issues	24/2759	1431/23569	0.14	0.15	
					Post-term pregnancy	18/2765	1349/23651	0.11	0.11	
					Labour complications	127/2656	10448/14552	0.06	0.1	
					PROM	21/2762	1913/23087	0.09	0.09	
					Malpresentation	17/2766	1058/23942	0.13	0.14	
					Caesarean	122/2661	5423/19577	0.16	0.2	
					Haemorrhage	41/2742	2179/22821	0.15	0.16	
Lie Fong et al 2010	40 (6 had RT)	9031	0	General population	Congenital abnormality	0/6	145/8834	4.67	4.4	• Pregnancy outcome not different for either group • Female CCS exposed to abdominal radiotherapy had more preterm babies and haemorrhage • Normal birth weight was found for babies of female CCS after adjustment for age at birth
					Pre-eclampsia	0/6	40/8991	17.07	15.92	
					Haemorrhage	02-Apr	449/8232	9.16	6.44	
					Manual removal	01-May	251/8430	6.71	5.76	
					Caesarean (emergency and elective)	0/6	1296/7735	0.45	0.49	
Signorello et al 2006	1264 (2201 births)	601 (1175 births)	0	Siblings	Live birth†	2309/1220	1209/491	0.76	0.92	• Female CCS more likely to be premature birth • Female CCS treated with abdominal radiotherapy at increased risk of premature birth • Female CCS at increased risk of small for gestational age and LBW babies
					Live birth (CCS exposed vs non-exposed)	1116/1085	617/558	0.93	0.96	
					Premature birth (RT uterus)	252/864	145/1007	2.02	1.79	
					Premature birth (RT ovary)	172/701	145/1007	1.7	1.56	
					Premature birth (cumulative)	424/1565	145/1007	1.88	1.69	

					Low birth weight (RT uterus)	106/1026	48/1094	2.35	2.22	
					Low birth weight (RT ovary)	65/814	48/1094	1.81	1.75	
					Low birth weight (cumulative)	171/945	48/1094	2.11	3.64	
					Small for gestational age (cumulative) (ovary+uterus RT)	159/1758	101/1002	0.89	0.9	
Reulen et al 2009	509	0	1422	CCS exposed vs CCS non-exposed	Live birth	351/158	1048/374	0.79	0.93	• Female CCS treated with radiotherapy at increased risk of preterm birth and LBW and small increased risk of miscarriage • Live birth rate was two-thirds lower than expected for female CCS (particularly when exposed to abdominal radiotherapy)
					Miscarriage	96/413	209/1213	1.34	1.28	
					Stillbirth	3/506	7/1415	1.19	1.19	
					Premature delivery	90/419	95/1327	3	2.64	
					LBW	75/276	77/971	3.42	2.9	
					Termination	59/450	158/1264	1.04	1.04	
Signorello et al 2010	1014	0	596	CCS exposed vs CCS non-exposed	Live birth (all CCS vs exposed CCS)	3077/60	4853/93	0.98	0.99	• Female CCS exposed to abdominal radiotherapy at increased risk of stillbirth and neonatal death
					Stillbirth/ neonatal death	39/3098	21/3116	1.86	1.85	
van de Loo et al 2019	14	33	37	CCS exposed vs CCS non-exposed	**CCS exposed vs CCS non-exposed**					• CCS exposed were at increased risk of reduced uterine volume (<44.3ml) when compared to general population • CCS exposed had an increased risk of premature labour, pregnancy complications and low-birth-weight babies than general population • CCS exposed had an increased risk of low-birth-weight babies than CCS non-exposed • Uterine exposure to radiotherapy increases risk of pregnancy complications and adverse outcomes. Pre-conception counselling and obstetric monitoring recommended
				CCS exposed vs general population	Small uterus	04-Oct	Dec-21	0.7	0.78	
					Pregnancy complication	10-Apr	13/20	3.84	1.81	
					Miscarriage	04-Oct	Jul-26	1.48	1.34	

					Premature delivery^	06-Aug	Sep-24	2	1.57	
					LBW*	05-Sep	Mar-30	5.55	3.92	
					SGA~	Jan-13	Feb-31	1.19	1.17	
					CCS exposed vs general population					
					Small uterus	04-Oct	Jul-30	1.71	1.51	
					Pregnancy complication	10-Apr	Aug-29	9	3.3	
					Miscarriage	04-Oct	Jul-30	1.71	1.51	
					Premature delivery^	06-Aug	Mar-34	8.5	5.28	
					LBW*	05-Sep	Jan-36	20	13.21	
					SGA~	Jan-13	Feb-35	1.34	1.32	
Winther et al 2008	1688	16700	2737 (siblings)	General population	Miscarriage (siblings)	44/413	27989/1718	0.006	0.1	• Female CCS at increased risk of miscarriage • No other differences noted
				Siblings	Live birth (all CCS vs general population)	1022/666	19335/2635	0.2	0.68	
					Stillbirth (all CCS vs general population)	5/1683	94/16606	0.52	0.52	
Haggar et al 2014	1894	4138	0	General population	**UNABLE TO DO META-ANALYSIS DUE TO LACK OF RAW DATA**					• Female CCS at increased risk of miscarriage, GDM, pre-eclampsia, haemorrhage, caesarean, hospitalisation post-partum • Female CCS have no excess risk of premature labour, antepartum haemorrhage, PROM, labour prolongation, retained placenta
Mueller et al 2009	1898	14278	0	General population						• Offspring of female CCS at increased risk of premature birth and LBW • No increase of congenital abnormalities, growth restriction, neonatal complications or perinatal deaths in offspring of female CCS • Female CCS more likely to have preterm birth and LBW • Female CCS at no increased risk of congenital abnormalities, neonatal death

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