What do we know about the psychosocial issues associated with cancer during pregnancy? A scoping review and gap analysis

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INTRODUCTION

Childbearing age is considered by many, including the Office for National Statistics, to be between 15 and 45 years.1 Although the risk of developing cancer is typically low at this life stage, there is a sustained global trend, particularly in developed nations and where the average number of births per woman is less than two, for women to delay conception.2 In these countries, the average childbearing age has increased to around 32 years.3 It is believed that this rise is coexistent with an observed increase in the numbers of pregnant women with cancer.4–6 An estimate of the incidence of cancer during pregnancy is difficult to determine. Globally, health registries do not combine oncological and obstetric data. Further, some pregnancies result in miscarriage or termination because of the disease and/or its treatment: these pregnancies may not be recorded. However, conservative estimates suggest incidence rates of 17 per 100 000 live births or 25–27 per 100 000 pregnancies.6 Increased prenatal testing to detect fetal chromosomal

ABSTRACT

Objectives There is a global increase in the number of women diagnosed with cancer during their pregnancy and a nascent evidence base to guide their supportive care. The purposes of this study were to (1) map research on the psychosocial issues affecting women and their partners on diagnosis and treatment for cancer during pregnancy; (2) determine available supportive care or educational interventions; and (3) identify knowledge gaps for future research and development.

Design Scoping review.

Search strategy Six databases were searched (Scopus, CINAHL, PsycINFO, Medline, Intermed, Maternal and Infant Health) to retrieve primary research (January 1995 to November 2021) investigating women and/or their partner’s decision-making and their psychosocial outcomes during and after pregnancy.

Data extraction and synthesis Sociodemographic, gestational and disease characteristics of participants and psychosocial issues identified were extracted. Leventhal’s self-regulatory model of illness provided a framework for mapping study findings enabling evidence synthesis and gap analysis.

Results Twelve studies were included, conducted in eight countries in six continents. Most women (70% of 217) were diagnosed with breast cancer during pregnancy. Reporting of sociodemographic, psychiatric, obstetric and oncological characteristics that are important in assessing psychosocial outcomes was inconsistent. None of the studies had a longitudinal design and no supportive care or educational interventions were identified. The gap analysis highlighted the lack of evidence about pathways to diagnosis, impact of late effects and how internal/social resources may affect outcomes.

Conclusions Research has focused on women with gestational breast cancer. Little is known about those diagnosed with other cancers. We encourage future study designs to capture data on sociodemographic, obstetric, oncological and psychiatric characteristics and adopt a longitudinal approach to explore the longer term psychosocial impact on women and their families. Future research should include outcomes that are meaningful for women (and their partners) and draw on international collaboration to accelerate progress in this field.

STRENGTHS AND LIMITATIONS OF THIS STUDY

⇒ First scoping review to use a theoretical framework to map, appraise and evaluate research on the psychosocial issues affecting women and/or their partners diagnosed with cancer during pregnancy.

⇒ As this was a scoping review some studies could have been missed; however, we conducted a comprehensive search of six medical, psychological, nursing and midwifery databases for literature published over a 25-year period in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses extension for Scoping Reviews guidelines.
abnormality has led to asymptomatic women being diagnosed with cancer during pregnancy.7

Research in the field of cancer in pregnancy is gaining momentum in recognition of its incidence and of the complexity it creates for mothers and their partners and clinicians: managing the pregnancy; maintaining the safety of the unborn child; delivering effective treatment; and treatment-related decision-making. There is a recognised focus on single diagnostic groups in previous research, for example, breast cancer: the most common form of cancer during pregnancy.8 An additional focus has been to appraise the evidence in order, for example, to evidence safe and effective treatment and to develop recommendations for practice.9–11 What is missing is an appraisal and synthesis of the psychosocial impact of this experience, both during or shortly after pregnancy. Pregnancy and cancer are two major life-changing events; when they occur simultaneously, psychosocial stressors can be anticipated, but how women’s needs are met remains largely unknown.

**Rationale**

Since the 1990s, there has been a growing body of research seeking to understand the psychosocial issues—the psychological and social aspects—associated with a cancer diagnosis in pregnancy, and the treatment decisions made by parents and clinicians. A recent systematic review12 found common themes in this literature: concerns about infants’ health and their future development; a sense of lost opportunity; and feelings of ‘not fitting-in’. The latter theme related to women feeling isolated from peer support groups and related this to the low prevalence of cancer during pregnancy. Potentially meaning that clinicians, while sympathetic, were unable to meet women’s needs because they were insufficiently experienced at treating and caring for these women.13 Further, women’s decision-making could be affected by their personal circumstances (eg, whether they already had children) and poor communication within multidisciplinary teams could impact their care experiences.12 The evidence base is growing, and the previous review failed to include maternity databases in their search, potentially omitting relevant studies from the search period. Timely systematic appraisal and synthesis are much needed to determine state of the art; also to identify priorities for future research, and to inform the development of interventions aimed at safeguarding the psychosocial well-being of women and their families. Therefore, we undertook a scoping review, rather than a systematic review, because our aim was to map the extent and nature of the emerging evidence and key concepts, and to inform future research directions and priorities,14 15 rather than establish the strength of evidence in an established field.

**Objectives**

This scoping review aimed to: (1) explore the extent, range and nature of research concerning the psychosocial issues affecting women (and their partners) on diagnosis and treatment for cancer during pregnancy; (2) determine what (if any) supportive care or educational interventions are available; and (3) conduct a gap analysis to identify areas for future research and practice development.

These specific objectives are different from a previous systematic review which had the more general aim of identifying the psychological aspects of gestational cancer.12

**METHODS**

**Protocol and registration**

The review methods align with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) extension for Scoping Reviews: checklist and explanation (see online supplemental file 1).16 17 The final protocol was registered prospectively with the Open Science Framework.18

**Patient and public involvement**

Patients and the public were involved in discussing the preliminary findings of this scoping review at an online international priority setting workshop hosted by the Institute of Advanced Studies at the University of Surrey (July 2021). This event was attended by 60 delegates from nine different countries, including women and partners with lived experience, oncology and maternity healthcare professionals (HCP) and support staff, academic researchers and educators and representatives from several charities. The workshop gained consensus on the priorities for future research into the psychosocial well-being and support for women diagnosed with cancer and their families. The discussions and recommendations were captured live by an artist (Katie Chappell, www.katiechappell.com) and used for dissemination across social media outlets (see online supplemental file 2).

**Information sources and searching**

Electronic searches were undertaken in Scopus, Nursing and Medical Cross Search (EBSCO, including CINAHL, PsycINFO and Medline) and Intermed, and Maternity and Infant Care (from January 1995 to November 2021). The start date of 1995 was chosen as this coincides with important policy changes in UK cancer services.19 Our search strategy was piloted and refined by the authors and used the following terms: (‘cancer’ OR ‘neoplasm’ OR ‘tumour’ OR ‘tumor’ OR ‘malignancy’ OR ‘carcinoma’) AND (‘pregnancy’ or ‘maternity’) AND (‘psychology’ OR ‘anxiety’ OR ‘distress’ OR ‘mental health’ OR ‘experience’ OR ‘perceptions’ OR ‘attitudes’ OR ‘views’ OR ‘feelings’ OR ‘perspective’). Database searches were supplemented by extensive cross-referencing of selected studies as well as searches of reference lists of previously published articles. The final search results were exported into EndNote V.X9 with duplicates removed.
Eligibility criteria
This scoping review included primary research published in peer-reviewed journals that explored, measured or focused on the social, psychological or emotional impact of a cancer diagnosis during pregnancy on a woman and her partner (published from 1995 onwards and available in English language). This included research investigating women and/or their partner’s decision-making and their psychosocial outcomes during and after pregnancy as well as HCPs involved in providing support.

Psychosocial issues could include distress, anxiety, depression, poor mental health, quality of life, attachment, relationships with partner/infant/other children and social support/networks in women and/or their partners. Papers were included regardless of pregnancy outcome (miscarriage, termination, stillbirth or live birth). Interventions could be focused on supporting women, their partners or learning for clinicians. Quantitative, qualitative and mixed-methods original research studies were eligible. Studies were excluded if they did not report a specific psychosocial component, involved women diagnosed with cancer before their pregnancy, in the postnatal period or with gestational trophoblastic disease, and/or were grey literature.

Selection of sources of evidence
Two of four reviewers (either CP, JH, KP or SV) screened the titles, abstracts or full texts of publications identified by the searches as potentially relevant publications. All reviewers were psychologists or clinical academics. Any disagreements about selection of studies were resolved by consensus and discussion with a third reviewer.

Data charting and synthesis
Two of four reviewers independently charted the data (any two of CTP, SV, JH or KP) using a data extraction form piloted on a subsample of studies and then refined to include agreed final data items (see online supplemental file 3). For each extracted data point, it was noted where data were not reported/unclear and recorded where data were present. In addition, to identify gaps in current evidence, the themes/focus of the studies were mapped by two researchers (JH and validated by KP) onto concepts from Leventhal’s self-regulatory model of illness (SRMI).21 This is a pragmatic framework for describing how symptoms and emotional experiences during a health threat or diagnosis influence how an individual perceives, interprets, responds and adjusts to such threats.25 It has been widely applied within the context of cancer and other illnesses that affect women of child-bearing age.23–25 This conceptual model of illness cognitions and behaviours provided a framework to synthesise the mix of qualitative and quantitative studies and map the current scope of evidence on psychosocial coping and appraisal in women diagnosed with cancer during pregnancy as well as other important contextual factors affecting psychosocial well-being. Areas mapped included illness representations, coping behaviours and responses, physical and psychosocial outcomes, external resources, healthcare systems and internal/social resources. As this was a scoping review we did not assess the risk of bias of included studies.

RESULTS
Identification of studies
Overall, 2705 records were identified (2539 through database searching and seven from cross referencing) (figure 1). After removing duplicates, 2546 unique records were screened with 2520 excluded as out of scope, based on title and abstract. In total, 25 full-text articles were obtained and assessed further for eligibility. Of these, 13 were excluded due to methodological/design ineligibility (8) or publication type (5) (figure 1). Twelve studies were included in the final scoping review (table 1).

Characteristics and results of sources of evidence
The total sample size of women diagnosed with cancer during pregnancy across all 12 studies was 212, with individual samples ranging in size from 3 to 74 women (table 2) with a median sample size of 8. One study also included 61 partners of women,11 and another included 19 relatives/close contacts (table 1).26 One study focused on clinicians’ (n=12) experiences of caring for eight women and their families.27 Nine studies were qualitative, all using semistructured interviews,26–34 two were quantitative cross-sectional surveys11 35 and one was a mixed-methods cross-sectional survey that included both closed and open-ended items.36 Three qualitative studies specifically reported being informed by a broad theoretical approach such as interpretative feminism,29 interpretative phenomenological analysis,33 and symbolic interactionism.26

Nearly all (11/12) studies were conducted in more economically developed countries. Three studies were conducted in Australia,28 30 34 two were conducted in the USA,25 36 Italy,31 33 and Japan.27 32 The remaining studies were undertaken in the UK,29 Belgium and the Netherlands,11 and Brazil.26 Studies were published over a 25-year period, with six in the past 2 years (table 1). Studies focused on the subjective experiences and perceptions of women,26 28–34 their main concerns during treatment and coping strategies,11 36 the perceived consequences of their cancer diagnosis, care and decision-making on their own psychosocial well-being and that of their family,36 or the demographic and clinical correlates related to higher distress.35 Only one study focused on clinicians’ experiences of shared decision-making in the context of cancer and pregnancy.27 Most accounts were retrospective (range 4 months to 17 years) with few participants reporting on their current experience (table 2). There was inconsistency in data collection and reporting across studies with many important contextual sociodemographic (table 2), obstetric and oncology details (table 3) not reported. Only one study22 considered the temporal order of pregnancy and cancer, with three of the eight participants...
being diagnosed with cancer before they were aware that they were pregnant.

Eight studies\(^1\)\(^\text{11,26-30,33-36}\) included some reporting of trimester or gestational stage at cancer diagnosis, only two studies reported stage of cancer when diagnosed\(^1\)\(^\text{11,35}\) (table 3) and seven reported relationship status (tables 1 and 2).\(^\text{11,26,30,31,33,34,36}\) Only two studies reported mode of delivery (62.0% vaginal, 36.6% caesarean section and 1.4% forceps).\(^\text{22,35}\) Only four studies reported details about all participants’ ethnicity/cultural background\(^\text{26,27,31,32}\) and none explicitly reported sexual orientation or whether the sample included women who were single.

Women’s age at cancer diagnosis ranged from 22 to 43 with the mean/median around mid-30s. Two studies reported women’s age at the time of data collection rather than diagnosis,\(^2\)\(^\text{32,33}\) and this differentiation was unclear in three studies (table 2).\(^\text{30,31,36}\) Six of the 12 studies only recruited women with gestational breast cancer (GBC) (38/212 women)\(^\text{28-31,33,36}\); the remaining studies recruited women with a range of cancer diagnoses,\(^\text{11,26,27,32,34,35}\) but almost two-thirds of these women (113/175) were diagnosed with GBC (table 3). Where treatments during pregnancy were reported these typically included some form of surgery (71/173 women) and/or chemotherapy (134/173 women) with a minority (26/173) receiving radiotherapy (either alone or with other treatments). Where reported, more than a third of the women in the total sample were primiparous

Figure 1 Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flow chart of literature search.
<table>
<thead>
<tr>
<th>Author</th>
<th>Year</th>
<th>Country</th>
<th>Aim</th>
<th>Design</th>
<th>Analysis approach</th>
<th>Overall sample size</th>
</tr>
</thead>
<tbody>
<tr>
<td>Bandyk and Gilmore</td>
<td>1995</td>
<td>USA</td>
<td>To identify the concerns of women who were treated with CT for GBC.</td>
<td>Mixed methods; cross-sectional pilot survey</td>
<td>Descriptive analysis</td>
<td>6</td>
</tr>
<tr>
<td>Henry et al</td>
<td>2012</td>
<td>USA</td>
<td>To explore variables associated with long-term psychological distress in women following cancer during pregnancy. Risk for distress was higher if they did not receive fertility assistance, had been advised to terminate the pregnancy, delivery was preterm, delivery was by C-section, produced insufficient milk to breast feed, experienced recurrence or undergone surgery after pregnancy.</td>
<td>Quantitative; cross-sectional survey using BSI-18 and IES</td>
<td>Bivariate associations</td>
<td>74</td>
</tr>
<tr>
<td>Ives et al</td>
<td>2012</td>
<td>Australia</td>
<td>To describe the psychosocial experiences of pregnancy in women diagnosed with GBC. Women make difficult decisions that impact their own, their family and their unborn fetus/baby. They describe high levels of anxiety, distress and conflict between concerns for their own and their baby’s health and well-being.</td>
<td>Qualitative; semistructured interviews</td>
<td>Thematic analysis</td>
<td>15 (4 women diagnosed during pregnancy, 11 post partum)</td>
</tr>
<tr>
<td>Rees and Young</td>
<td>2016</td>
<td>UK</td>
<td>To explore the experiences of women diagnosed with GBC. Cancer during pregnancy disrupted assumptions and expectations about pregnancy, new motherhood and future life course.</td>
<td>Qualitative; semistructured interviews</td>
<td>Grounded theory</td>
<td>3</td>
</tr>
<tr>
<td>Vandenbroucke et al</td>
<td>2017</td>
<td>Belgium and Netherlands</td>
<td>To identify women and partners at risk of high levels of distress based on their coping profile when diagnosed with cancer during pregnancy. Internalising coping strategies in women and partners was associated with highest levels of distress and these people may benefit from additional psychosocial support.</td>
<td>Quantitative; cross-sectional survey (using CPQ and CERQ)</td>
<td>Bivariate associations, ANOVA, cluster analysis</td>
<td>122 (61 women; 61 partners)</td>
</tr>
<tr>
<td>Hammarberg et al</td>
<td>2018</td>
<td>Australia</td>
<td>To explore the healthcare experiences of women diagnosed with GBC. Interdisciplinary and patient communication were important components of care experience. Comprehensive care was seen to encompass the ‘spirit’, ‘mind’ and ‘body’.</td>
<td>Qualitative; semistructured interviews</td>
<td>Thematic analysis</td>
<td>17</td>
</tr>
<tr>
<td>Faccio et al</td>
<td>2020</td>
<td>Italy</td>
<td>To compare maternal representations in pregnant women with experience of GBC, breast cancer history and those with no cancer history. Main themes were fears and worries for themselves and fetus/baby, meaning of motherhood, mother-fetus relationship and partner support.</td>
<td>Qualitative; case-control semistructured interviews</td>
<td>Thematic analysis</td>
<td>38 (4 with GBC, 15 with cancer history, 19 with no cancer history)</td>
</tr>
<tr>
<td>Kozu et al</td>
<td>2020</td>
<td>Japan</td>
<td>To explore the experiences of pregnant women with cancer in decision-making and their views on the role of nurses in providing care. Decision-making experiences had three themes: interaction between the women and the fetus, family members and staff; dilemma and uncertainty; and redefinition of own decisions.</td>
<td>Qualitative; semistructured interviews</td>
<td>Content analysis</td>
<td>8</td>
</tr>
<tr>
<td>Facchin et al</td>
<td>2021</td>
<td>Italy</td>
<td>To understand the lived experience of women being diagnosed with GBC. Three main themes emerged: overwhelming emotions; a sense of difference; and sources of strength. Stress is caused by the paradoxical coexistence of cancer and pregnancy.</td>
<td>Qualitative; semistructured interviews</td>
<td>Interpretative phenomenological analysis</td>
<td>5</td>
</tr>
<tr>
<td>Gomes et al</td>
<td>2021</td>
<td>Brazil</td>
<td>To explore how the diagnosis of cancer during pregnancy occurred and impact on maternity experience. Diagnosis was based on symptoms that were confused with pregnancy. Two main themes: being surprised by the diagnosis, and suffering from the repercussions of cancer on pregnancy and health.</td>
<td>Qualitative; interviews</td>
<td>Grounded theory</td>
<td>31 (12 women, 19 family members)</td>
</tr>
<tr>
<td>Hori and Suzuki</td>
<td>2021</td>
<td>Japan</td>
<td>To explore the process of shared decision-making for healthcare professionals supporting pregnant patients with cancer and their families. Five themes are integral to providing support; preparing for shared decision-making; healthcare professionals working in a team; confirming intentions to the patient and her family; improving the system to provide support; and providing support that helps informed decisions.</td>
<td>Qualitative; semistructured interviews and focus group</td>
<td>Narrative approach</td>
<td>14 (all healthcare professionals)</td>
</tr>
<tr>
<td>Stafford et al</td>
<td>2021</td>
<td>Australia</td>
<td>To explore women’s quality of healthcare experience in public or private care systems when diagnosed with cancer during pregnancy. Five themes included: control over healthcare; trust in clinicians, hospitals and systems; coordination of care; an uncommon diagnosis; and holistic, future-orientated care.</td>
<td>Qualitative; semistructured interviews</td>
<td>Thematic analysis</td>
<td>23</td>
</tr>
</tbody>
</table>

ANOVA, analysis of variance; BSI-18, Brief Symptom Inventory-18; CERO, Cognitive Emotion Regulation Questionnaire; CPQ, Cancer and Pregnancy Questionnaire; C-section, caesarean section; GBC, gestational breast cancer; IES, Impact of Events Scale.
<table>
<thead>
<tr>
<th>Table 2</th>
<th>Sociodemographic data reported for women diagnosed with cancer during pregnancy in reviewed studies</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Author</strong></td>
<td><strong>Sample of women with cancer diagnosed during pregnancy</strong></td>
</tr>
<tr>
<td>Bandyk and Gilmore&lt;sup&gt;36&lt;/sup&gt;</td>
<td>6</td>
</tr>
<tr>
<td>Henry et al&lt;sup&gt;25&lt;/sup&gt;</td>
<td>74</td>
</tr>
<tr>
<td>Ives et al&lt;sup&gt;28&lt;/sup&gt;</td>
<td>4</td>
</tr>
<tr>
<td>Rees and Young&lt;sup&gt;29&lt;/sup&gt;</td>
<td>3</td>
</tr>
<tr>
<td>Vandenbroucke et al&lt;sup&gt;11&lt;/sup&gt;</td>
<td>61</td>
</tr>
<tr>
<td>Hammarberg et al&lt;sup&gt;30&lt;/sup&gt;</td>
<td>17&lt;sup&gt;+&lt;/sup&gt;</td>
</tr>
<tr>
<td>Faccio et al&lt;sup&gt;31&lt;/sup&gt;</td>
<td>4</td>
</tr>
<tr>
<td>Kozu et al&lt;sup&gt;32&lt;/sup&gt;</td>
<td>8†</td>
</tr>
<tr>
<td>Facchin et al&lt;sup&gt;33&lt;/sup&gt;</td>
<td>5</td>
</tr>
<tr>
<td>Gomes et al&lt;sup&gt;36&lt;/sup&gt;</td>
<td>12‡</td>
</tr>
<tr>
<td>Stafford et al&lt;sup&gt;34&lt;/sup&gt;</td>
<td>23</td>
</tr>
</tbody>
</table>

*Two of these women were diagnosed with cancer post partum and data cannot be differentiated.
†Three women were diagnosed with cancer before they knew they were pregnant.
‡Three of these women were diagnosed with cancer post partum and data cannot be differentiated.
<table>
<thead>
<tr>
<th>Author</th>
<th>Sample size</th>
<th>Trimester at cancer diagnosis or gestation (in weeks)</th>
<th>Primiparous/multiparous</th>
<th>Birth outcomes (including gestational age M [SD] and mode of delivery)</th>
<th>Breast cancer (n)</th>
<th>Other cancers (n)</th>
<th>Cancer stage</th>
<th>Treatment†</th>
</tr>
</thead>
<tbody>
<tr>
<td>Bandyk and Gilmore</td>
<td>6</td>
<td>1st trimester (4) 2nd trimester (2)</td>
<td>Not reported</td>
<td>All ‘healthy’</td>
<td>6</td>
<td>Not applicable‡</td>
<td>Not reported</td>
<td>Surgery+chemotherapy (6)</td>
</tr>
<tr>
<td>Henry et al</td>
<td>74</td>
<td>14.7 [8.4]</td>
<td>Primiparous (25)</td>
<td>Gestational age at delivery (36.4 weeks [3.1], Advised to terminate (19), Mode of delivery: Vaginal birth (44), C-section (26), Forceps (1))</td>
<td>48</td>
<td>Blood cancers (8) Ovarian (4) Melanoma (4) Other (not specified) (10)</td>
<td>Stage I (18) Stage II (37) Stage III (10) Stage IV (6)</td>
<td>Surgery (53) Chemotherapy (59) Radiotherapy (1)</td>
</tr>
<tr>
<td>Ives et al</td>
<td>4</td>
<td>1st trimester (1) 2nd trimester (1) 3rd trimester (2)</td>
<td>Primiparous (1) Multiparous (3)</td>
<td>Live birth (3) Miscarriage (1)</td>
<td>4</td>
<td>Not applicable‡</td>
<td>Not reported</td>
<td>Surgery+chemotherapy (4) Radiotherapy (2) Hormone therapy (3)</td>
</tr>
<tr>
<td>Rees and Young</td>
<td>3</td>
<td>2nd trimester (1) 3rd trimester (2)</td>
<td>Primiparous (1) Multiparous (3)</td>
<td>Live birth (3)</td>
<td>3</td>
<td>Not applicable‡</td>
<td>Not reported</td>
<td>Surgery (3) Chemotherapy (2) Radiotherapy (2) Hormone therapy (2)</td>
</tr>
<tr>
<td>Vandenbroucke et al</td>
<td>61</td>
<td>16 (range 1–36)</td>
<td>Primiparous (27) Multiparous (34)</td>
<td>Not reported</td>
<td>38</td>
<td>Blood cancers (13) Cervical (3) Tongue (1) Ewing (1) gastrointestinal stromal tumor (1)</td>
<td>Stage I (13) Stage II (17) Stage III (11) Recurrence (3)</td>
<td>Surgery+chemo/radiotherapy or both (33) Radiotherapy (2) Hormone therapy (1) None (2)</td>
</tr>
<tr>
<td>Hammarberg et al</td>
<td>17</td>
<td>1st trimester (5) 2nd trimester (4) 3rd trimester (6)</td>
<td>Primiparous (6) Multiparous (11)</td>
<td>Miscarriage (1) Premature birth (9) Full-term birth (7)</td>
<td>17</td>
<td>Not applicable‡</td>
<td>Not reported</td>
<td>Not reported</td>
</tr>
<tr>
<td>Faccio et al</td>
<td>4</td>
<td>Not reported</td>
<td>Primiparous (1) Multiparous (3)</td>
<td>Not reported</td>
<td>3</td>
<td>Not applicable‡</td>
<td>Not reported</td>
<td>Not reported</td>
</tr>
<tr>
<td>Kozu et al</td>
<td>8†</td>
<td>Not reported</td>
<td>Primiparous (4) Multiparous (4)</td>
<td>Termination (2) Premature birth (2) Full-term birth (4)</td>
<td>2</td>
<td>Blood cancers (2) Cervical (3) Digestive (1)</td>
<td>Not reported</td>
<td>Surgery+chemotherapy during pregnancy (6) Surgery, chemotherapy, radiotherapy after pregnancy (8)</td>
</tr>
<tr>
<td>Facchin et al</td>
<td>5</td>
<td>Range 4–26 weeks</td>
<td>Primiparous (2) Multiparous (3)</td>
<td>Pregnant at time of data collection (5)</td>
<td>5</td>
<td>Not applicable‡</td>
<td>Not reported</td>
<td>Chemotherapy</td>
</tr>
<tr>
<td>Gomes et al</td>
<td>12§</td>
<td>Not reported</td>
<td>Not reported</td>
<td>Premature birth (5) Full-term birth (4) Unclear for post-partum diagnosis (3)</td>
<td>10</td>
<td>Blood cancer (1) Cervical (1)</td>
<td>Not reported</td>
<td>Surgery, chemotherapy, radiotherapy</td>
</tr>
<tr>
<td>Stafford et al</td>
<td>23</td>
<td>17.52 [10.09]</td>
<td>Not reported</td>
<td>Live birth (17) Pregnant at time of data collection (6)</td>
<td>15</td>
<td>Blood cancer (5) Bowel (1) Cervical (1) Lung (1)</td>
<td>Not reported</td>
<td>Not reported</td>
</tr>
</tbody>
</table>

*Women diagnosed with cancer during pregnancy.
†Treatment may be during or after pregnancy.
‡Recruitment restricted to women with breast cancer only.
§Three of these women were diagnosed with cancer post partum and data cannot be differentiated.
(67/175 women) and most pregnancies resulted in live births (90/141 women).

**Synthesis of results and gap analysis**

Mapping onto Leventhal’s SRMI we were able to synthesise study results and identify gaps in knowledge surrounding the psychosocial impact of a diagnosis of cancer during pregnancy on women and their partners (figure 2).

The extent, range and nature of research on the psychosocial issues associated with gestational cancer

Only two qualitative studies described women’s experiences leading up to the time of diagnosis; these indicated that pregnancy (and recent motherhood) may impact symptom appraisal and lead to possible delays in help seeking.26 33 In Facchin et al’s33 study, one participant described how she had felt that there was something strange in her breast, people reassured her it was due to pregnancy-related changes, but she was not convinced and wanted to get it checked. She described being unaccompanied at the time of diagnosis and how a lack of information immediately after diagnosis was a source of confusion. Similarly, in Gomes et al’s26 study, one participant described how family members had provided reassurance that breast changes were likely to be related to hormone changes, which led to delays in help seeking.

All studies focused on concerns relating to health-related outcomes for the women, including the impact of cancer treatments on future fertility; recurrence of cancer, health/survival; and continuation with pregnancy and/or the future infant outcomes (where the pregnancy continued).11 26–36 Several studies also reported concerns and distress about ability to breast feed specifically related to cancer and its treatments.26 31 34–36 All studies described adverse psychosocial outcomes for women including psychological distress, anxiety, sense of conflict/uncertainty, loss, guilt and disrupted expectations.11 26–36 The one study that included partners did not find any statistically significant difference between women and their partners’ distress; however, women were more inclined to continue with the pregnancy than their partners.11 Similarly, women in a qualitative study32 described how their partners and/or wider family tried to influence their decision to prioritise their own life rather than their fetus or future fertility which the women felt was not constructive. Four studies included themes that encompassed positive long-term psychosocial outcomes in the face of adversity, including post-traumatic growth29 and a sense of pride, relief and comfort in their ability to cope.29 31 32 However as these studies were retrospective, and none were longitudinal, it is not possible to determine how this may have changed over time or been affected by prognosis.

Overall studies focused extensively on the cognitive and emotional representations of receiving a cancer diagnosis during pregnancy and how this impacted decision-making processes and care plans. An important theme was the perceived threat from cancer and the concept of prioritisation of life, including consideration of the risks to self and fetus and how this may be impacted by partner/family support and whether they were multiparous. Several studies described how the co-occurrence of pregnancy and cancer created cognitive dissonance relating to thoughts and rumination about both mortality and new life.26 29 31–33 The importance of cultural context in decision-making was only highlighted in one study. Conducted in Japan, it noted that family views on fertility and adoption impacted the decision-making process.32 Within this context, communication, continuity of care and emotional/informational support from HCPs were found to be important alongside the support provided.
by intimate partners or family members. The value of peer support, that is, support from those with similar experience, was mentioned in two qualitative studies; however, there were also barriers for pregnant women attending cancer support groups including a sense of ‘feeling out of place’.

One study conducted in Brazil described the importance of spiritual, emotional and practical support provided by religion and faith in helping some women to cope with their diagnosis. This same study also described the importance of social media (WhatsApp) in providing opportunities for peer support for some women and how this might reduce their sense of isolation; however, one woman spoke about negative experiences of finding conflicting information online and learning to trust their HCPs as their primary source of information.

Two studies explored different coping styles. One qualitative study found avoidance was used as a coping strategy by one woman but the impact this had on psychosocial well-being was not explored. A quantitative study used cluster analysis to explore and understand different styles of coping using the Cognitive Emotional and Regulation Questionnaire, which characterises responses to stressful events. Somewhat surprisingly this study suggested that both positive coping (with an emphasis on acceptance, positive reappraisal and putting into perspective) and blaming coping (characterised by self and other blame only) strategies were associated with lower distress for women and their partners compared with internalising coping (rumination, catastrophising, planning combined with self and other blame). The authors concluded that couples using internalising strategies may need greater psychosocial support from HCPs.

However, overall, little is known about the potential long-term impact of different coping styles, or the role of self-regulation and self-efficacy and how these may affect appraisal of goals and coping in this context.

Identification of supportive care or educational interventions

Our second aim was to explore the supportive care interventions available in the peer-reviewed literature. We found no studies reporting interventions or programmes tailored for women or their partners diagnosed with cancer during pregnancy. Similarly, there were no interventions/educational programmes tailored to upskill HCPs, support the workforce in multidisciplinary working or inform organisational service provision. Only one descriptive qualitative study conducted in Japan described the experience of HCPs in providing care to women diagnosed with cancer in and around pregnancy with a focus on the issue of shared decision-making. Their results emphasised the following: the importance of supporting women and their families to make informed decisions, aligned with the women’s intentions; and the need for effective teamwork and coordination of services, with clear roles and responsibilities within and between the different oncology and maternity teams involved.

Identification of gaps in knowledge to guide future research and development

Some clear gaps in knowledge were evident in the literature. No studies assessed whether psychiatric symptoms were at a level requiring clinical intervention and important concepts such as fear of recurrence/progression were completely absent from this literature. Furthermore, none of the studies assessed the issue of late effects in detail and whether long-term psychosocial morbidity and supportive care needs were impacted by the experience of pregnancy during cancer. Similarly, no studies evaluated the impact on work-related issues, their financial supportive care needs (eg, during treatment or maternity leave) or the longer term economic impact of a cancer diagnosis during pregnancy.

None of the studies explored how disease characteristics may impact cognitive and emotional representations, coping and, therefore, psychosocial response. The methodological focus on retrospective accounts means the samples were likely to be biased towards including women diagnosed at an earlier cancer stage, with better cancer outcomes, and as a result little is known about end-of-life decision-making in this group.

Despite the diverse population from the many countries included in these studies, it appears that the samples overwhelmingly represent the experience of white women in stable heterosexual relationships. There was little or no evidence about the impact of the cancer diagnosis on women or their partners in the context of complex healthcare needs and important social characteristics such as language barriers, ethnicity, being single or the role of religiosity/spirituality. There was also no evidence in this specific population about the demographic risk factors that might put women and their partners at risk of poorer psychosocial outcomes.

DISCUSSION

Despite the increasing incidence of cancer during pregnancy, the psychosocial issues associated with a cancer diagnosis at this time remain an under-researched topic. Our scoping review includes only 12 studies. Our knowledge is therefore informed by data from little more than 200 women worldwide, suggesting these women experience significant distress and have considerable supportive care needs, persisting beyond the end of pregnancy. How these needs are being met, if at all, either in the short or long term, remains largely unknown. It remains unclear how their experience of distress may be related to pre-existing vulnerabilities to poor mental health, which have been shown to be important predictors of psychological well-being for women living with and beyond cancer generally. Without this, it is difficult to predict/plan/deliver resources needed to support women in a proactive manner.

We found few studies focused on the experience of pathways to receiving a cancer diagnosis. There was a suggestion that women do not experience diagnostic and
treatment delay intervals after first contact is made with HCPs. There was some evidence, however, of patient delay in making initial contact with HCPs for pregnant or lactating women and this was evidenced by the accounts of women in two studies. In part, this may account for why more pregnant women with breast cancer are diagnosed with advanced disease than non-pregnant women (65%–90% vs 45%–65%). It is important that future research explores how women become aware of and interpret their cancer symptoms in the context of their pregnancy, whether or not they seek help promptly, where they first present with cancer symptoms (primary care, maternity or other settings) and what support they receive from their midwifery and cancer teams after the cancer diagnosis.

Only one small qualitative study was based in the UK (table 1), with the remainder in Europe, North and South America and Oceania. This gap across nations is important because different healthcare systems use different models of funding and care in primary, maternity and cancer services: this will likely impact women’s experiences of these healthcare systems. Reporting of important contextual information was inconsistent across studies, for example, whether the women had had previous pregnancies. This needs to be addressed in future research to enable comparison across studies and determine relevance of research across countries and settings. Notably, whether women had had previous pregnancies was poorly reported. The participants lacked diversity as most diagnosed with breast cancer were white and married. The experiences of non-white, single, non-heterosexual women or socially disadvantaged women are under-represented in existing studies, as are the experiences of wider family members. This could result in the needs of more diverse populations being overlooked and risks exacerbating existing inequalities in experience and outcomes of care.

None of the studies reported on the involvement of service users within their projects and this may be a critical omission in ensuring that research is relevant and valuable to women and their families. Few studies had any theoretical basis or made reference to conceptual models or frameworks, which was surprising, as they are integral to healthcare practice and research. Theory should form the basis for any future complex interventions to improve support for people affected by cancer during pregnancy at the individual, professional, multidisciplinary team, organisational and service levels. Using the Leventhal’s SRMI framework helped shed light on the emergent psychosocial issues associated with a diagnosis of cancer during pregnancy as well as the gaps in evidence. An advantage of using the Leventhal’s SRMI for the gap analysis was that it helped to identify possible targets for intervention. Our scoping review shows two key areas for future research and practice development: peer support and shared decision-making.

Peer support was rarely mentioned, but those that did highlighted challenges associated with accessing such support for an uncommon situation. Where it was accessed, there was emerging evidence that it might lessen the sense of isolation, recognising that digital information and peer forums may prove an important focus for future supportive care interventions in people affected by a cancer diagnosis during pregnancy.

The decision-making process was mentioned in several studies and the important role HCP support and communication play in this. There was, however, a lack of studies exploring the psychosocial experiences of the decision-making process around cancer treatment, who was involved, cross-specialty working between obstetric and oncology teams and possible decisional regret which could affect longer term psychosocial outcomes. Further research is needed to understand what factors might make decision-making optimal for these women, considering their family’s individual circumstances. To do this, HCPs may need access to evidence-based online resources and decision support ‘tools’ to enable holistic care to meet these women’s needs, at the point of diagnosis and beyond.

There are many avenues for future research to inform clinical practice and education for HCPs. First, it is important that future studies take a longitudinal design and capture women’s full sociodemographic, obstetric, oncological, mental health and quality of life data to drive advances in the development of supportive interventions that extend beyond the prenatal period. Second, in realising a research agenda for this relatively rare condition, international collaboration will be essential through specialist networks such as the International Network on Cancer, Infertility and Pregnancy, and the European PosMat initiative (developing maternal HCPs to promote a POSitive MATernal experience for women with cancer). Essential to these future endeavours is service user involvement, this must be at the heart of establishing research priorities and to identify outcomes that are meaningful to women and their families. This can be achieved through collaboration with third sector organisations that have developed specialist peer support and expertise in this field, such as Mummy’s Star (UK and Ireland), Hope for Two (USA) and Stichting Sterk (the Netherlands). Enhanced models of care for women affected by cancer during pregnancy have been proposed, for example, a recent scoping review of ethical issues for cancer during pregnancy suggested the need for an adapted clinical decision-making model—which takes into account the complex ethical and non-medical factors—to reduce psychosocial distress. It is likely that third sector organisations will need to play an important role in the development of such models of care to ensure they meet the psychosocial needs of women and their families.

Strength and limitations

The strength of our findings is enhanced by adherence to PRISMA scoping review principles including independent data sources, systematic identification, and retrieval, cross-referencing and a broad approach.
to literature searching informed by recommendations. By including maternity databases we synthesised a wide range of evidence and use of a theoretical framework provided a useful lens through which to appraise and evaluate the data and delineate the gaps; both of which are strengths compared with previous systematic or narrative reviews. However, several limitations should be noted. As with all scoping reviews, we cannot be certain that we did not exclude or miss some important studies; however, we tried to minimise this by assessing full-text articles for further studies. In line with scoping review methodology, we limited our search to six key databases but acknowledging the exclusion of others could be a potential limitation. A key strength of the review was inclusion of quantitative or qualitative studies to ensure comprehensive assessment of the literature and the focus on women diagnosed during pregnancy specifically. Although this may have reduced the number of available data sources (i.e., those diagnosed after birth), we thought this was necessary to get an accurate picture of the scope of research focused on cancer during a pregnancy. No studies from the Middle East or African countries were found.

CONCLUSIONS

The incidence of cancer during pregnancy is increasing. A diagnosis of cancer during pregnancy can have devastating consequences for women and their families. The complexity of decision-making, that is ethically and emotionally charged, is apparent and requires the close involvement of maternity and cancer teams to guide women towards informed treatment decisions. It is a challenging situation for all, and yet we still know so little about the psychological impact of cancer on pregnant women.

In conclusion, while this scoping review highlights gaps in our understanding and an absence of supportive care interventions, we have identified a nascent research interest in this field. The well-established international specialist network and sector-specific support organisations provide a fertile landscape that is ripe for advancing psychosocial research to bring benefits to women and their families diagnosed with cancer during pregnancy.

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