

Title:

Preconception care for people with health conditions; what approaches work, for whom, and in what circumstances? A realist review.

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ABSTRACT

Preconception care for people with chronic health conditions is recommended in view of the implications for them and their offspring during pregnancy and beyond. This realist review of published and grey literature explored factors that explain why people seek or receive appropriate preconception counseling, and why they engage in recommended health behavior change prior to pregnancy. 52 studies contributed to phase 1 synthesis and 38 studies provided explanations in phase 2. 10 program theories were developed, explored, and refined, through iterative discussion and coding. Causal explanations of the ways in which components of preconception care contribute to effective access to care and pre-pregnancy behavior change were identified. Beneficial components included continuity of carer (promoting trust), a partnership approach (empowering people who feel valued), promoting an integrated approach across primary and secondary care, offering psychological counseling (recognizing the link between physical and psychosocial aspects of living with health conditions), considering sexual and reproductive health as part of routine care and normalizing conversations about preconception care (to reduce the barrier created by social and cultural norms). These key aspects have been highlighted for consideration when planning, implementing, and improving preconception care services for people with health conditions.

KEY WORDS: chronic health conditions; health behavior change; preconception counseling; realist review; reproductive health

1. INTRODUCTION

Factors influencing parental health and well-being before conception (preconception), including nutrition, lifestyle habits and medication, can influence both gametogenesis and embryogenesis, and the subsequent long-term health and development of offspring (Fleming et al., 2018; Lane et al., 2014; Steegers-Theunissen et al., 2013). Increased incidence of morbidity and mortality demonstrates that health and well-being during the preconception period is particularly important for people with pre-existing chronic physical and mental health conditions (Jack et al., 2008; Knight et al., 2021; Persson et al., 2009).

Several different approaches to preconception care, for people with chronic health conditions, have been identified in the literature including face-to-face clinics targeting people planning a pregnancy (Murphy et al., 2010), opportunistic counseling in primary or secondary care (Shannon et al., 2014) and digital or web-based resources such as smart phone applications (apps) (Nwolise et al., 2017). Evidence is already available to demonstrate that appropriate preconception behavior change, as a result of access to appropriate information or counseling, can be effective in terms of pregnancy outcomes (King et al., 2012; Murphy et al., 2010). Awareness of barriers and challenges to implementing effective preconception care is also apparent (Barker et al., 2018; Murphy et al., 2010; Spence et al., 2010).

There is, however, a lack of systematic consideration of what approaches to preconception care work, for whom, and in what circumstances (Public Health England, 2018). A realist approach exploring mechanisms, contexts, and outcomes, aiming to identify the conditions required for efficacy (Pawson, 2006), is therefore both appropriate and needed, in order to improve care.

Thus, the aim of this realist review was to explore, in relation to people with chronic health conditions, what form of preconception care works for whom, how these approaches work, and in what circumstances. In keeping with realist methodology, the objective of this two-phase review was to develop theory relating to the main factors or mechanisms that are thought (both scientifically and experientially) to explain why people with chronic health conditions a) seek or receive appropriate condition-specific preconception care or advice, and b) engage in recommended behavior change prior to pregnancy (we are defining “engagement in recommended behavior change” as engagement in at least one condition-specific behavior change, which is known to reduce morbidity or mortality, prior to conception).

2. METHODOLOGY AND METHODS

Realist review methods, described as a form of systematic literature review, (Pawson et al., 2005; Pawson, 2006; RAMESES II Project, 2017a) were employed since the nature of preconception care is complex and the aim was to explore what works for whom and how. The overall aim of a realist review is explanation building, and this was achieved through an iterative theory-driven interpretive approach by consulting any relevant material, thereby using evidence from a range of sources (Pawson, 2006). The review was conducted in two main phases: firstly, systematic searches led to the development of initial program theories (from evidence related to preconception care) and secondly, targeted searches were conducted to test and refine these theories (using evidence from other interventions that share the same program theories). As in other realist reviews, stakeholder involvement was embedded throughout this process (Husk et al., 2020), with an international expert

advisory group including health professionals, researchers, and people with lived experience, involved in each phase. Consultation was either face-to-face or by email.

2.1 Phase 1: Initial program theory development

2.1.1 Electronic searches

A search was developed iteratively, with support from an information specialist (AW), based on a known set of key articles. These key includable articles had been identified in an earlier targeted literature search; if they were not returned from a draft search strategy, it was amended until the team was satisfied with the search. The result was a “broad brush” approach that captured all types of preconception care. Terms included: pre-conception, peri-conception, pre-gestational, pre-pregnancy, inter-conception and family planning. Terms relating to specific health conditions were only included in conjunction with “family planning” to exclude many items, related to contraception, that we were not interested in (see supplementary material table 1 for full search strategy).

Databases searched were MEDLINE, Embase, PsycINFO, Cochrane Library, British Nursing Database and CINAHL. Included studies were those written in English and based in the Organization for Economic Co-operation and Development (OECD) countries (OECD, 2021), to select those with similar approaches to healthcare and economic status. There were not an unmanageable number of studies and so no date limit was applied; the search was undertaken in August 2018. Search results were de-duplicated and saved using EndNote X8.

2.1.2 Searching other sources

Supplementary searches were undertaken to ensure relevant evidence was included (Cooper et al., 2018). Strategies used included emailing authors of identified studies,

conducting Google and Google Scholar searches, backwards and forward citation chasing, searching relevant United Kingdom websites, and the British Library EThOS online service.

2.1.3 Screening to identify relevant evidence

Relevant evidence, from a variety of sources, contributed to theory building.

Evidence was included when there were descriptions of who was receiving preconception care, under what circumstances, and what resources were on offer to them.

Initial screening of titles and, where available, abstracts identified in the database searches, was conducted by two team members independently (HH and KM) using the web-based citation management application Rayyan (Ouzzani et al., 2016).

Where these met the inclusion criteria, the full text was obtained and screened again by the same two team members. Any disagreements were resolved through discussion, and a third researcher was available but not needed.

2.1.4 Population inclusion criteria

The population inclusion criteria were: people who had any type of self or clinician-identified chronic physical or mental health condition, and who were seeking or receiving preconception care. This may have been part of routine primary or secondary care related to their condition, or specifically because they were considering planning a pregnancy. Some people in this group may have previously experienced pregnancy, and some may have experienced pregnancy loss, complicated pregnancy, or neonatal loss. Evidence was not restricted by health condition.

2.1.5 Intervention inclusion and exclusion criteria

We included evidence concerning a range of preconception care packages aimed specifically at people with pre-existing health conditions. Some of these targeted anyone of reproductive age, and others were designed for people who were planning a pregnancy. Despite evidence of a bias in the literature towards preconception care for females (as opposed to males), our search included care offered to both sexes including care offered to same-sex couples. The following interventions were excluded: any aimed specifically at people experiencing fertility problems, seeking advice regarding contraception or delaying pregnancy, and any aimed at people seeking termination of pregnancy or pre-pregnancy genetic screening.

2.1.6 Types of study

As with other realist reviews (Wong et al., 2013), evidence that provided descriptions of preconception care using a broad variety of methods was included; both qualitative and quantitative studies as well as non-empirical studies. Since numerous studies were identified, these were prioritized based on relevance and rigor (Pearson et al., 2015) (see below).

2.1.7 Outcomes

The outcomes relating to preconception care for health care professionals included awareness and involvement in preconception care, and for people with health conditions included experiences of preconception advice and appropriate health behavior change(s) dependent on the individual person's health condition(s) or circumstances.

2.1.8 Quality assessment

In line with requirements for realist review, the quality of the data was based on relevance (to the program theory) and rigor (credibility and trustworthiness of the methods used) (Wong et al., 2013) using a hybrid tool classifying sources as

“conceptually-rich, thick or thin” (Pearson et al., 2013). Papers identified as conceptually rich or thick were included in the review (see supplementary material table 2). This enabled focus on stronger sources without exclusion of weaker ones.

2.1.9 Data extraction

The realist approach of “engaging with” the data was used initially, rather than formal data extraction; this involved note-taking, annotation, conceptualization and revision based on regular team discussion and review of the data (Pearson et al., 2015), both on paper and using NVivo 12 Pro (2018) for coding. Specifically, a variety of types of preconception care and factors contributing to identified intended and un-intended outcomes were examined.

2.1.10 Synthesis

Iterative examination of the data by the team (HH, KH and KM) resulted in the identification of three broad areas that were found to contribute significantly to both access and behavior change in relation to preconception care. Further data examination led to the identification of prominent recurring patterns (demi-regularities), which were explained using “if X – then Y” structured statements (Husk et al., 2020). These statements were iteratively refined through discussion and data review by the team (HH, KH, KM and JS) together with input from the expert advisory group, resulting in 35 statements (initial programme theories – see supplementary material table 3) of how preconception care works. Refinement of statements involved tools described in realist literature, in which synthesis is described as gaining a greater understanding of how and why an intervention works (Pawson, 2006; Wong et al., 2013).

Documentation of the reasoning processes used and applied during synthesis promoted transparency throughout the process (Wong et al., 2013).

Due to time and financial constraints, we were unable to further robustly refine all 35 statements and so undertook a web-based nominal group technique prioritisation (Murphy et al., 1998; Husk et al., 2020) with our expert advisory group, using a democratic process whereby all participants have an equal private vote and were asked to rank statements in order of preference for further investigation. This resulted in 10 statements (initial program theories) for further testing in phase 2.

2.2 Phase 2: Program theory testing and refinement

2.2.1 The search for empirical evidence to assess plausibility of initial program theories

The second phase started with 10 targeted searches, using search terms specific to each of the 10 initial program theories identified for further investigation. Use of one large abstract and citation database, Scopus (Elsevier, 2021), enabled identification of relevant empirical evidence related to each program theory to assess plausibility; this was not intended to be an exhaustive systematic search. This effective approach was therefore described as purposive due to the use of specific program theory-related terms and is often referred to as a “spear-fishing” approach (Pawson, 2006) (detail of search strategy available in supplementary material table 4). During this phase we included both qualitative and quantitative studies; we included any primary studies, from any discipline (not necessarily related to preconception care), that provided apposite evidence on the theories that had been identified in phase one and required testing and refining in phase two (see supplementary material table 5). Evidence was again tested for relevance and rigor prior to data extraction and synthesis (Pawson, 2006), with conceptually-rich papers contributing to the review.

2.2.2 Data extraction and synthesis

Data extraction in phase two of this review involved annotation of papers, collation of evidence aligned to the initial program theories identified in phase one, using NVivo 12 Pro (2018), and reportage, using extracts of evidence to identify the basis of inferences used for synthesis (Pawson, 2006). The data were used to clarify and explore contexts, mechanisms and outcomes in order to test and refine program theory indicating causal explanations regarding effective preconception care, and thus contributed to the final synthesis stage. Synthesis followed the strategy outlined in phase one above, with the purpose of refining initial program theory developed during phase one in the light of evidence and analysis of findings from a broader range of empirical studies identified in phase two. This refinement of theory is referred to as abstraction, where the theory still provides an explanation of what is happening in a particular scenario but can also be applied more generically in other similar situations, such as among people with different health conditions; at this stage, the theory is often referred to as “middle-range theory” (RAMESES II Project, 2017).

3. RESULTS

3.1 Search results

The first phase of database searching resulted in 3,161 citations, 37 of which contributed to the synthesis, alongside 15 additional citations from grey literature searches (Figure 1). Most evidence from the first phase searches related to people with Diabetes (type 1 and type 2). Other health conditions identified included obesity, epilepsy, HIV, hypertension, mental health conditions, chronic kidney disease, cystic fibrosis and inflammatory bowel disease (see supplementary material table 2). The

second phase of searching resulted in 4,228 hits, with 38 contributing to context, mechanism, outcome (CMO) configurations (see table 1 and supplementary material table 4).

3.2 Analysis and synthesis (i)

The 35 statements (initial programme theories from iterative data analysis and synthesis following phase 1 searches, listed in full in supplementary material table 3) were divided into three areas that were found to contribute significantly to if and how access to preconception care and behavior change prior to pregnancy takes place, for people with chronic health conditions. These three areas: the intervention (including provider characteristics, content and delivery mechanisms), people's beliefs (including perceptions of self, perceptions of support and perceptions of pregnancy planning) and the process (including guidelines and pathways, and accessibility of services), are interconnected, as illustrated in Figure 2.

3.3 Analysis and synthesis (ii) – deeper explanations of the 10 prioritised theories

The ten prioritised statements were further refined through iterative analysis and synthesis considering evidence from phase 2 searches. These are presented below.

3.4 Programme theories related to the intervention (provider characteristics):

3.4.1 If there is continuity of carer, and this experience is positive, THEN people are more likely to trust their healthcare professional, feel empowered and thus supported to make appropriate choices / behavior change.

Nine studies (Devindo, 2014; Earle et al., 2017; Echenique et al., 2017; King et al., 2012; McCorry et al., 2012; Mortagy et al., 2010; Murphy et al., 2010; O'Higgins et al., 2014; Spence et al., 2010) were included in the review that referred to continuity of carer, or relational continuity; most reported its value. This was both in terms of

accessing preconception care, and for on-going support for behavior change prior to pregnancy. What people gained from such continuity was described in several ways:

“Reassurance and support... trusted expert guidance... feeling cared for... moral and medical support... professional support and understanding...” (O'Higgins et al., 2014 p370)

Relational continuity can, therefore, improve care through trust in a healthcare professional that is established over time and based on knowledge and understanding of an individual. To develop a truly empowering partnership approach to care, however, mutual respect between both parties is required:

“Participants who had developed a strong relationship with their professional carers and had expertise in their diabetes mellitus talked about care as a partnership...” (Earle et al., 2017 p86)

Studies highlighted that continuity of carer alone is not sufficient, but continuity must be accompanied by mutual respect (Devito, 2014; Earle et al., 2017; McCorry et al., 2012; Murphy et al., 2010; O'Higgins et al., 2014; Spence et al., 2010). The extract below illustrates how people feel when the health care professional does not appear to care about them:

“... they don't even let you speak, they just tell you to shut it there that's it ...” (Earle et al., 2017 p87)

Healthcare professionals need to listen and acknowledge that people may be willing to sacrifice their own health in order to fulfil their dreams and goals. The extract below highlight the negative impact of perceived disapproval from health care professionals:

“... she would not seek advice as her health professional was opposed to the prospect of further pregnancies ... I don't want to hear you telling me I can't have more children, so I just went ahead.” (Spence et al., 2010 p1388)

Continuity of carer over time may result in additional barriers, as one study highlighted, if health care providers are viewed as parental figures. A paternalistic approach suggests lack of mutual respect and is therefore unlikely to result in feelings of support and empowerment.

“Most men in this study had been attending the clinic their entire lives and could have viewed providers as parental figures, such that discussions of sexual practice were uncomfortable or could have been considered inappropriate or to stimulate disapproval.” (Echenique et al., 2017 p41)

In our broader targeted (secondary) searches (for search terms see supplementary material table 4), we sought to establish the plausibility of this pathway in other fields. Studies show that continuity of carer can improve outcomes for individuals in both general practice and maternity care (Barker et al., 2017; McLachlan et al., 2012; Parker et al., 2011; Sandall et al., 2016; Van Walraven et al., 2010), supporting our middle-range theory. These studies showed improved patient safety and reduced

mortality rates, with relational continuity, as well as improvement in both patient and health care provider satisfaction.

3.4.2 IF healthcare professionals have a positive, non-judgemental, respectful attitude towards people seeking support, and adopt a partnership approach THEN people will develop trust in their healthcare professional and are likely to feel empowered and supported to make informed and appropriate choices / behavior change.

As identified above, a positive and respectful attitude towards people seeking support, by healthcare professionals, has been recognised as paramount. The nature of interactions with healthcare professionals was mentioned in 18 studies included in the review (Devido, 2014; Earle et al., 2017; Echenique et al., 2017; Health Education England, 2017; Public Health England, 2018; Forde et al., 2016; Frieder, 2010; Kazmerski et al., 2017; Klein et al., 2017; Lawther et al., 2018; McCorry et al., 2012; Mittal et al., 2014; Murphy et al., 2010; Nguyen et al., 2015; O'Higgins et al., 2014; Poels et al., 2016; Spence et al., 2010; Winterbottom, 2012), highlighting that health care professionals' behavior can affect how people perceive they are viewed, and this in turn impacts their behavior and therefore the outcome of the intervention. One study highlighted how this can affect clinic attendance:

“...women are more likely to want to attend clinics when they feel they are treated with respect and dignity.” (Earle et al., 2017 p87)

Some studies highlighted the issue of control, resulting from an authoritative and paternalistic communication style (Earle et al., 2017). Studies also reported how lack of respect can make people feel they are not valued:

“ ... felt 'judged' by the health professional and perceived them to be controlling and directive ... one woman preferred to shy away from advice as she felt doctors attributed all her problems to obesity and made her feel like she was unimportant. (Spence et al., 2010 p1388).

Whilst healthcare professionals may have an opinion on what is right, they must recognise that people have autonomy and agency. Healthcare professionals must adopt a partnership rather than a paternalistic approach:

“The advice one woman gave to [healthcare] providers was, “Don't let [your patient] just assume they're never going to have a baby. Whether you think it's good for their health or not, it's [your patient's] decision.” (Kazmerski et al., 2017 p821)

Our targeted, broader, secondary searches showed that this middle-range theory is well supported in other fields. Meaningful relationships between individuals and their health care providers, characterised by trust, is a recurrent theme of effective health care in a range of settings (Brener et al., 2013; Holt et al., 2018; Lor et al., 2016; Stenner et al., 2011; Sword et al., 2012). Many studies refer to this as personalized care (Deeny et al., 2018; National Health Service, 2019) or a person-centred approach (McCormack & McCance, 2006).

3.5 Programme theories related to the intervention (delivery mechanism):

3.5.1 IF information and/or support is internet-based or available via a mobile phone THEN people can use this to increase knowledge about their condition / progress, which is both empowering and supports a partnership approach between the patient and their healthcare professional.

Four studies included in the review referred to online or internet-based preconception information (Agricola et al., 2014; Barker et al., 2016; Lopez et al., 2015; O'Higgins et al., 2014). In some cases, it was noted that this could be used to complement a face-to-face discussion with a health care professional:

“... talking to a doctor is the preferred way to receive information ... However, searching for information online was seen as a complementary way to receive information and also helped in preparing for their conversation with a doctor.” (Lopez et al., 2015 p567)

The use of online platforms to improve pre-pregnancy health through sustained behavior change has shown promise, with one study identifying how a digital intervention tool can tailor content to meet individual needs (Barker et al., 2016). People who used the tool received regular reminders, tips and feedback, which may have contributed to motivation and empowerment to maintain healthy behaviors. Adding human interaction, however, may result in increased engagement and effectiveness of the online intervention:

“... adding an element of human interaction in the form of telephone coaching increased engagement with, and effectiveness of, the [digital] intervention when

compared with a condition where there was no telephone coaching.” (Barker et al., 2016 p332)

In our targeted, broader, secondary searches, it was evident that online platforms are useful sources of information for some individuals with underlying health conditions, which can empower them through increased knowledge and understanding (Kelly et al., 2018; Madrigal & Escoffery, 2019; Reen et al., 2019). Digital interventions (such as mobile applications) increased self-efficacy and self-management of glycaemic control for some individuals with diabetes; an example of beneficial preconception health behavior change (Jeffrey et al., 2019). Recommendation and involvement of the healthcare professional in use of the mobile application showed increased effectiveness through a partnership approach to care (Kelly et al., 2018).

This middle-range theory is therefore supported, with evidence suggesting that a combination of non-judgemental motivational human contact alongside online or digital interventions may support behavior change prior to pregnancy through choice and autonomy, resulting in feelings of empowerment and self-efficacy.

3.6 Programme theories related to the intervention (content):

3.6.1 IF preconception care is considered as part of a broader sexual and reproductive health continuum in relation to peoples’ health conditions THEN relevant aspects of sexual and reproductive health on the continuum of contraception / pregnancy planning are likely to be discussed and people are less likely to experience unplanned pregnancies.

Preconception care is just one element of a broader sexual and reproductive health continuum, which 13 studies identified as a gap in care for individuals with chronic

health conditions (Charron-Prochownik et al., 2001; Echenique et al., 2017; Fischl et al., 2010; Forde et al., 2016; Frieder, 2010; Holmes et al., 2017; Holmes et al., 2012; Kazmerski et al., 2017; Lopez et al., 2015; McCorry et al., 2012; Mittal et al., 2014; Murphy et al., 2010; Simone et al., 2018; Spence et al., 2013). Taking a broader view of sexual and reproductive health (SRH), rather than just focusing on preconception care provides opportunities for a more personalized approach and may reduce unplanned pregnancies.

“Existing misconceptions around fertility and limited receipt of SRH care lead to sexual risk behaviors and unintended pregnancies..... young women want improved SRH educational resources coupled with routine SRH discussions initiated by their [healthcare] providers.” (Kazmerski et al., 2017 p822)

One study highlighted how preconception advice is sometimes given routinely, and this may be perceived as irrelevant to many people:

“Delivering preconception counseling to younger women, in whom the prospect of pregnancy seems a long way off, is undoubtedly a challenge. Women in this study routinely questioned its effectiveness, suggesting it was often ‘skimmed over’ during annual reviews.” (Murphy et al., 2010 p98)

Another study tested the use of a reproductive life plan for people with chronic health conditions. Both contraception and preconception were included in the brief counseling tool, with people demonstrating increased knowledge and feelings of empowerment as a result:

“...patients who benefited the most from the intervention were the women who started with lower baseline knowledge about childbearing and/or their chronic diseases, and that women without previous children may particularly benefit from reproductive life planning.” (Mittal et al., 2014 p30)

This middle-range theory was partially supported by evidence identified in secondary searches, although it is unclear how long any beneficial effects might last as longer-term follow-up of study participants is required. Studies have shown that discussions should include both sexes, sexual and reproductive health services, contraception, personal life plans, and underlying social and community factors (Dean et al., 2014; Liu et al., 2016; Skogsdal et al., 2019).

3.6.2 IF peer support networks are available THEN people are more likely to feel emotionally / socially supported and will be more inclined to engage with the medical advice and support they are offered

Seven studies included in the review identified peer support as beneficial to individuals with health conditions (Doss, 2017; Grady & Geller, 2016; King et al., 2012; Lopez et al., 2015; O'Higgins et al., 2014; Spence et al., 2010; Spence et al., 2013). Peer support may be face to face or via other routes including social media:

“... women ... repeatedly acknowledged that a ‘real life’ approach was best ... with interview clips from ‘real women’ at the forefront ...” (Spence et al., 2013 p12)

Studies identified the value of people sharing experiences, demonstrating a deeper understanding of common challenges, and creating social norms for people who might otherwise feel that their experiences of pregnancy are “different”:

“... Involvement in pre-pregnancy care enabled women to meet others, promoting an atmosphere of normality and reassurance that they could give birth to a healthy baby ... the comfort of knowing they are not alone in their struggle and frustration.”

(O'Higgins et al., 2014 p370 & 371)

This middle-range theory was supported by our secondary searches, which highlighted the value of peer support for individuals in a range of different circumstances. Common themes include an increased sense of hope, control and ability to effect change, as well as a sense of community belonging (Davidson et al., 2012; Gillard et al., 2015; Naslund et al., 2016). Some studies also showed that volunteer peer educators can influence effective physiological health changes (Gagliardino et al., 2013; Piette et al., 2013); a key element of peer support includes authentic presence, which provides effective support (Schmied et al., 2011).

3.6.3 IF psychological counseling and support is available to address and reduce anxieties regarding pregnancy with a pre-existing health condition THEN people are more likely to be able to access preconception care and engage in appropriate behavior change.

11 included studies acknowledged the impact of psychosocial factors in relation to preconception care (Earle et al., 2017; Forde et al., 2016; Kazmerski et al., 2017; Komiti et al., 2014; Lawther et al., 2018; McCorry et al., 2012; Murphy et al., 2010;

O'Higgins et al., 2014; Pashley & O'Donoghue, 2009; Steel et al., 2015; Winterbottom, 2012):

“Women reported experiencing emotional stress whilst attempting to comply with preconception requirements unique to their diagnosed health condition ...” (Steel et al., 2015 p7)

Some psychological issues may be severe, due to previous experience or poor pregnancy outcome:

“... their experiences of guilt, loss and personal failure are extremely distressing and may not always be recognised during routine clinical care.” (Murphy et al., 2010 p99)

One study suggested that psychological support may be an important element of successful preconception care:

“Changing health behavior, without recognising and addressing the psychosocial and economic environment in which the behavior takes place, is unlikely to be successful.” (Pashley & O'Donoghue, 2009 p155)

Secondary searches supported this middle-range theory. Studies ranged from services implementing a very brief psychological intervention, to those that included a psychologist as part of the team; all demonstrated that implementing a psychological intervention resulted in some improvement in outcomes that were dependent on behavior change (Armitage et al., 2014; Harvey, 2015; Uchendu & Blake, 2017).

Lunn et al. (2017) described a working model for service provision, using an adapted biopsychosocial model to illustrate the inextricable link between physical and psychosocial impacts of living with a chronic health condition.

3.7 Programme theories related to the people's beliefs (perceptions of pregnancy planning):

3.7.1 IF people perceive preconception care as normal THEN they are more likely to seek / be receptive to preconception care and adopt appropriate behavior to optimise pregnancy.

Five studies included in the review (Earle et al., 2017; Forde et al., 2016; Kazmerski et al., 2017; Murphy et al., 2010; Poels et al., 2016) report a desire for normality among people with health conditions, which may be a barrier to accessing preconception care since this does not align with social and cultural norms:

“... women just wanted their lives to be as normal as possible ... the desire for a normal experience of ... pregnancy planning is cited ... as the reason why they would not attend for preconception care.” (Earle et al., 2017 p56)

This links with the benefit of peer support providing a social group in which people with similar health conditions feel normal as a result of shared experiences:

“Involvement in pre-pregnancy care enabled women to meet others, promoting an atmosphere of normality and reassurance ... ‘been involved ... before and felt it was good’.” (O'Higgins et al., 2014 p370)

Secondary searches showed that a 'normal' preconception experience does not involve drastic lifestyle changes or information seeking. Consequently, when these are required it can lead to increased anxiety (Atkinson et al., 2016). A desire to be normal is well documented across patient groups, and perceptions of normality can be socially constructed (Genuis & Bronstein, 2017). New perceptions of reality can be established through shared experiences, which can help individuals with health challenges to accept the need for therapeutic interventions (Genuis & Bronstein, 2017).

3.7.2 IF people believe that their health condition reduces fertility and/or their use of contraception is suboptimal THEN they have a greater chance of unplanned pregnancies.

Three studies included in the review highlighted the impact of inaccurate health beliefs (Earle et al., 2017; Forde et al., 2016; Murphy et al., 2010); specifically, that inaccurate beliefs about contraception resulted in a higher risk of unplanned pregnancy due to suboptimal practises:

“There were beliefs that contraception was not necessary because the woman’s diabetes meant they would not get pregnant or that contraception, particularly oral contraception, was inappropriate for people with diabetes.” (Forde et al., 2016 p697)

In our secondary searches, studies supported this middle-range theory, showing that inaccurate health beliefs can have negative consequences. Two studies highlighted how inaccurate beliefs about the aetiology of a condition or efficacy of treatment can

result in delays seeking medical advice and subsequent increased risk of morbidity and mortality (Jones et al., 2014; Spencer et al., 2012).

3.8 Programme theories related to the process (guidelines and pathways):

3.8.1 IF individualised brief interventions about sexual and reproductive health issues, using the Health Belief Model, are a routine part of on-going care THEN people are more likely to seek / access preconception care, having considered key elements of the Health Belief Model and received ‘cues to action’.

Six studies included in the review demonstrate that including preconception advice in routine medical appointments can act as ‘cues to action’, resulting in intentions to plan future pregnancies (Charron-Prochownik et al., 2001; Charron-Prochownik et al., 2013; Fischl et al., 2010; Mittal et al., 2014; Spence et al., 2010; Spence et al., 2013). What these studies have in common, however, is a broader focus on sexual and reproductive health, including contraception, as well as recognition of wider psychosocial and cognitive factors:

“Women felt empowered to make decisions about their reproductive future in the context of their chronic disease with the use of [a reproductive life plan] and a discussion with the clinician.” (Mittal et al., 2014 p30)

When preconception advice was not focused on individual’s needs, or repeated advice was offered at times that were not relevant, however, this was found to be counterproductive:

“Constant advice that is repetitive and untimely can actually be a disincentive to women in accessing preconception care.” (Earle et al., 2017 p60)

Highlighting a high level of unplanned pregnancies and a limited discussion of contraception in routine medical consultations, McCorry et al. (2012) emphasised the importance of individualized care and advice:

“Healthcare for women with pre-existing diabetes should ... provide individualised care and advice, which is tailored to women’s needs and relevant to their specific life circumstances, including women who are not actively considering pregnancy.”
(McCorry et al., 2012 p401)

Our secondary searches identified studies reporting that a public health policy in the United Kingdom, ‘making every contact count’, aimed at encouraging health care professionals to support and facilitate behavior change through opportunistic interventions, is closely aligned to and supports this middle-range theory (Nelson et al., 2013). In order to effectively deliver this type of brief intervention, however, health care professionals need to identify appropriate recipients and have effective training for efficient delivery of interventions (Keyworth et al., 2018). Studies in secondary searches also found that application of the health belief model has been shown to be effective in relation to contraceptive counseling for people with diabetes from a range of cultural and ethnic backgrounds (Johnson et al., 2021).

3.8.2 IF provision is integrated across specialities and across both primary and secondary care THEN a shared vision for preconception care will result in consistently better provision of preconception care.

12 papers included in the review commented on the organisation of services in some way (Brennand et al., 2016; Public Health England, 2018; Forde et al., 2016; Frieder, 2010; King et al., 2012; Knight et al., 2017; Lawther et al., 2018; Mortagy et al., 2010; Murphy et al., 2010; Nguyen et al., 2015; Simone et al., 2018; Wiles et al., 2015), many recommending an integrated approach, promoting a person-centred rather than a condition-centred approach:

“You’re not dealing with diabetes you’re dealing with a woman with diabetes.”

(Mortagy et al., 2010 p819)

The importance of giving a consistent message was highlighted:

“I don’t think it matters who gives the message as long as we’re all giving the same consistent message, GP’s practice nurses, health visitors, community midwives, everyone.” (Forde et al., 2016 p699)

Studies also recognised system-wide barriers, which may differ between countries in view of the variation in healthcare systems:

“... lack of clarity in responsibilities was further attributed to the targets set up by the commissioning bodies regarded as ‘mixed messages’ and ‘conflicting incentives’ by some GPs.” (Mortagy et al., 2010 p817)

Despite these barriers, there was a clear need for a shared vision and guidelines between primary and secondary care, which would facilitate a consistent message to those seeking support:

“The secondary care team pointed out the need for a joint ... strategy as well as a clear shared vision with primary care, with GPs being active members of the team providing the preconception care service.” (Mortagy et al., 2010 p818)

Studies identified in our secondary searches strongly support this middle-range theory, identifying that an integrated approach to care for people with chronic health conditions was beneficial in terms of improved well-being and a reduced need for healthcare services (Kruis et al., 2013; Nuño et al., 2012). Whilst barriers to delivering integrated care exist, ways to overcome these have been identified and can be used to plan and implement integrated care (Ling et al., 2012).

3.9 Summary of results

Figure 2 illustrates the subdivisions and connections between the three areas that were found to contribute significantly to both access and behavior change in relation to preconception care. The intervention refers to the interaction during which preconception care, advice or support was offered. Provider characteristics that were identified as empowering and most likely to support behavior change were those demonstrating a positive, non-judgemental, respectful attitude towards people, which promoted a partnership approach (Brener et al., 2013; Lor et al., 2016), and this was more likely when a trusting relationship was built through continuity of carer (Barker

et al., 2017; Sandall et al., 2016). Online delivery of information or support was found to complement face-to-face discussions with a health care professional and promote a partnership approach to care (Kelly et al., 2018).

Three aspects of content were explored; the social and emotional benefits of peer support, the availability of psychological counseling and approaching preconception care as part of a broader sexual and reproductive health continuum. All three were identified as beneficial (Lunn et al., 2017; Mittal et al., 2014; O'Higgins et al., 2014), although longer term follow-up studies are needed in relation to the broader sexual and reproductive health continuum for people with health conditions.

A desire to be normal was one of two theories relating to beliefs that people with health conditions shared (Forde et al., 2016; Kazmerski et al., 2017; Lopez et al., 2015). With normality being socially constructed (Genuis & Bronstein, 2017), this highlights a mechanism by which peer support effects access and behavior change in relation to preconception care. Accurate knowledge was also identified as important for this group, in view of the increased risk of unplanned pregnancy due to inaccurate health beliefs (Forde et al., 2016).

Two theories relating to the process of preconception care were explored, including preconception advice in routine medical appointments. This was found to be beneficial, but only if it was individualized, relevant and met appropriate sexual and reproductive health needs (McCorry et al., 2012; Mittal et al., 2014). This finding links to the need for an integrated approach across specialities and services (Kruis et al., 2013). Whilst barriers to achieving a clear shared vision may exist (Ling et al., 2012), it is imperative to develop shared guidelines to achieve consistency and clarity (Mortagy et al., 2010), which in turn will facilitate access and behavior change in relation to preconception care.

4. DISCUSSION

Preconception care for people with health conditions is recognised as an important area for improvement to optimise the health of future generations (Public Health England, 2018). This realist review has produced theories relating to three areas of preconception care: the intervention (including provider characteristics, the content, and delivery mechanisms), people's beliefs (including perceptions of self, perceptions of support and perceptions of preconception care), and the process (including guidelines and pathways, and accessibility of services). These three areas were found to contribute significantly to both access and behavior change in relation to preconception care, for people with chronic health conditions.

The review has identified that preconception care is offered at different times and settings, but there were insufficient data to conclude what may be particularly beneficial and appropriate for different groups of people in different contexts; this is something that we aim to draw out in our realist evaluation, which includes a range of health conditions. Whilst undertaking this review we noted that diabetes has been the subject of most published research in this area and that a wider range of conditions need to be included in future research to reflect the diversity of experience within the population.

The findings of this realist review include themes that resonate with wider areas and experiences of health care, and are well supported by empirical evidence, such as personalization (Lor et al., 2016), continuity of carer (Sandall et al., 2016) and integration of services (Kruis et al., 2013). People are more likely to seek and accept support from a healthcare professional that they trust, and trust can be built through respectful relationships and consistent advice (Earle et al., 2017). People's behavior

is also affected by their values and beliefs (Jones et al., 2014), so their perceptions of self, support and preconception care are equally significant and contribute to accessing advice and support prior to pregnancy. Peer support can be beneficial for some people (O'Higgins et al., 2014), but this review has shown that preconception care and support can be, at times, both unavailable and unwelcomed (Earle et al., 2017; Kazmerski et al., 2017). It is, therefore, not a single intervention, but part of a wider sexual and reproductive health continuum, and one that is likely to be an individualized pathway involving a complex series of interacting elements, all of which need consideration.

A major strength of this review is the inclusion of a wide range of chronic physical and mental health conditions, and the breadth of literature we have included. The involvement of people with lived experience, clinicians and researchers with international representation in our expert advisory group has also been a strength; this group played a key role in developing, testing and prioritizing program theories for more targeted searching, further synthesis and refinement. As with other realist reviews (Wong et al., 2013), these more targeted searches (phase 2) enabled us to provide better explanations of program theories based on empirical evidence.

The review had some limitations; we acknowledge that the availability of evidence we were seeking regarding preconception care was mainly from studies focusing on diabetes, with some other health conditions contributing to this (see Figure 1).

Availability of evidence was also limited by the lack of detailed descriptions in the literature of what was offered, in terms of preconception care.

5. RECOMMENDATIONS

This realist review has identified important approaches and principles that should be incorporated into the delivery of preconception care for people with chronic health conditions. Healthcare professionals must demonstrate a positive, non-judgemental, respectful attitude towards people with health conditions, and adopt a partnership approach to care. Continuity of carer should be promoted to build trusting relationships between people and their healthcare professionals. Healthcare professionals should also encourage a partnership approach through access to accurate online information, which people can use to complement face-to-face discussions. Accurate information must be available to people at all stages along the sexual and reproductive health continuum. This must be relevant and personalized to promote healthy lives and planned pregnancies. Information should also be provided at routine medical appointments as part of a holistic approach to care. Peer support networks should be promoted so that people with chronic health conditions can access social and emotional support from others with similar experiences, providing opportunities for preconception care to be normalised within these networks. Psychological counseling should be offered to everyone seeking preconception care, recognising the additional anxiety people with chronic conditions may face when planning a pregnancy. Shared preconception care guidelines providing consistency and clarity must be developed and an integrated approach across specialities and services is required to facilitate access to preconception care and support people to make appropriate behavior changes where necessary. Finally, further longer-term follow-up studies are needed to evaluate preconception care, as part of the broader sexual and reproductive health continuum, for people with chronic health conditions, and we are undertaking a realist evaluation to conclude what may be particularly beneficial and appropriate for different groups of people in different contexts.

6. CONCLUSIONS

We have explored some of the complexities involved in providing preconception care for people with health conditions and identified specific areas that contribute significantly to effective outcomes. The recommendations regarding the intervention, people's beliefs, and the process in relation to preconception care must be implemented in order to achieve effective access to preconception care and support for behavior change prior to pregnancy. These recommendations alone are unlikely to be a solution for all, and our ongoing research will include exploration of whether particular approaches to preconception care might be appropriate for people in different contexts. They will, however, enable more effective preconception care, which has been shown to reduce morbidity and mortality for people with health conditions by improving health before, during and after pregnancy, and therefore require urgent consideration.

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Declaration of Interests

Competing interests: none.

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The Intervention:	Hits (SCOPUS)	Included
Provider characteristics and delivery mechanism	Continuity of Carer	5
	Respectful attitude	5
	Web-based	4
The Intervention:	Hits (SCOPUS)	Included
Content	Preconception care part of broader Sexual and Reproductive Health	4
	Peer support	6
	Psychological counseling	4
People's beliefs	Hits (SCOPUS)	Included
Perceptions of pregnancy planning	Perceived as normal	2
	Health beliefs	2
The Process	Hits (SCOPUS)	Included
Guidelines and pathways	Part of routine care	3
	Integrated services	3
38 papers contributing to CMO configurations		

Table 1: Second phase targeted searches

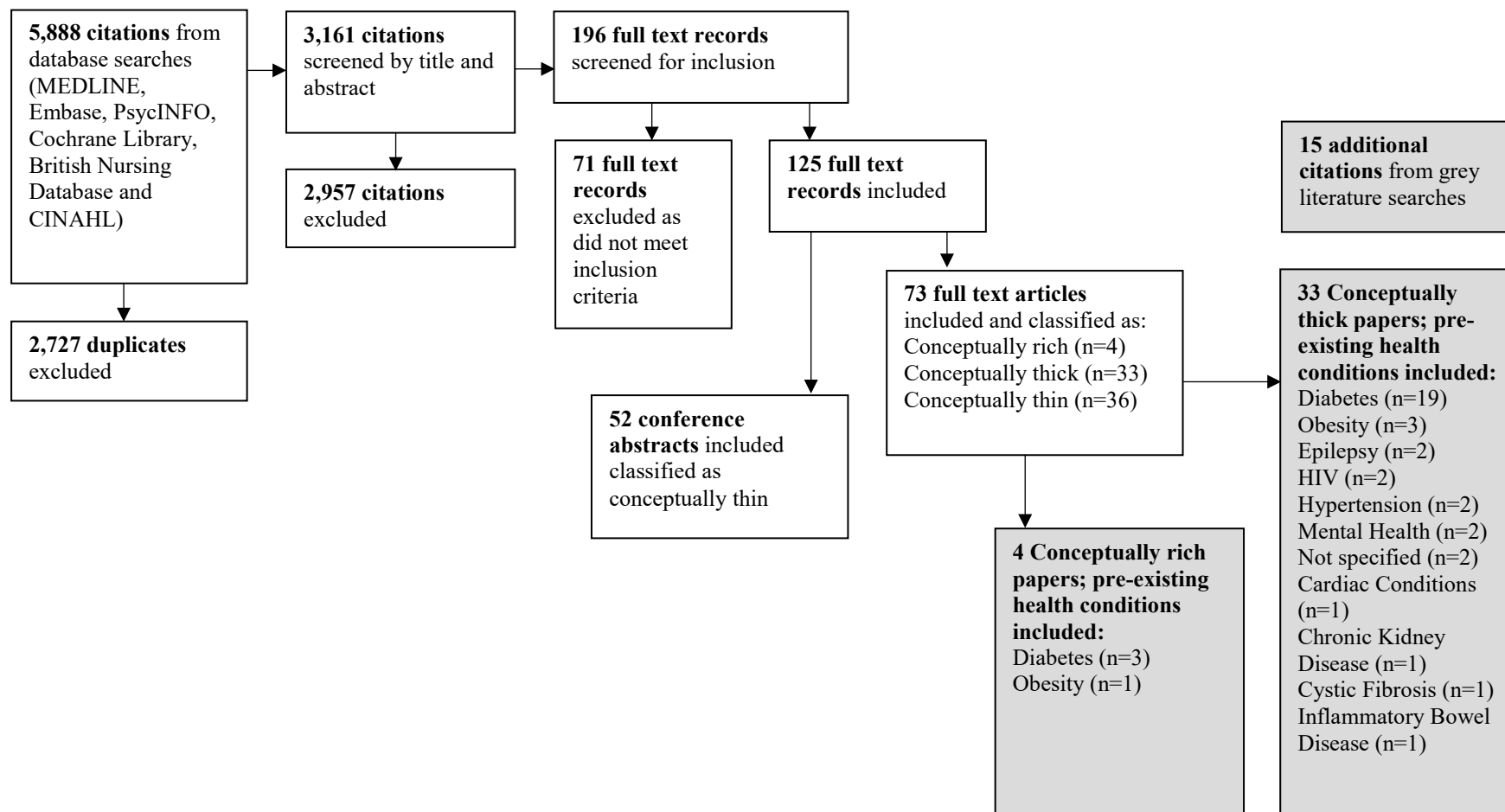


Figure 1: First phase searches: progress and flow of sources through the review

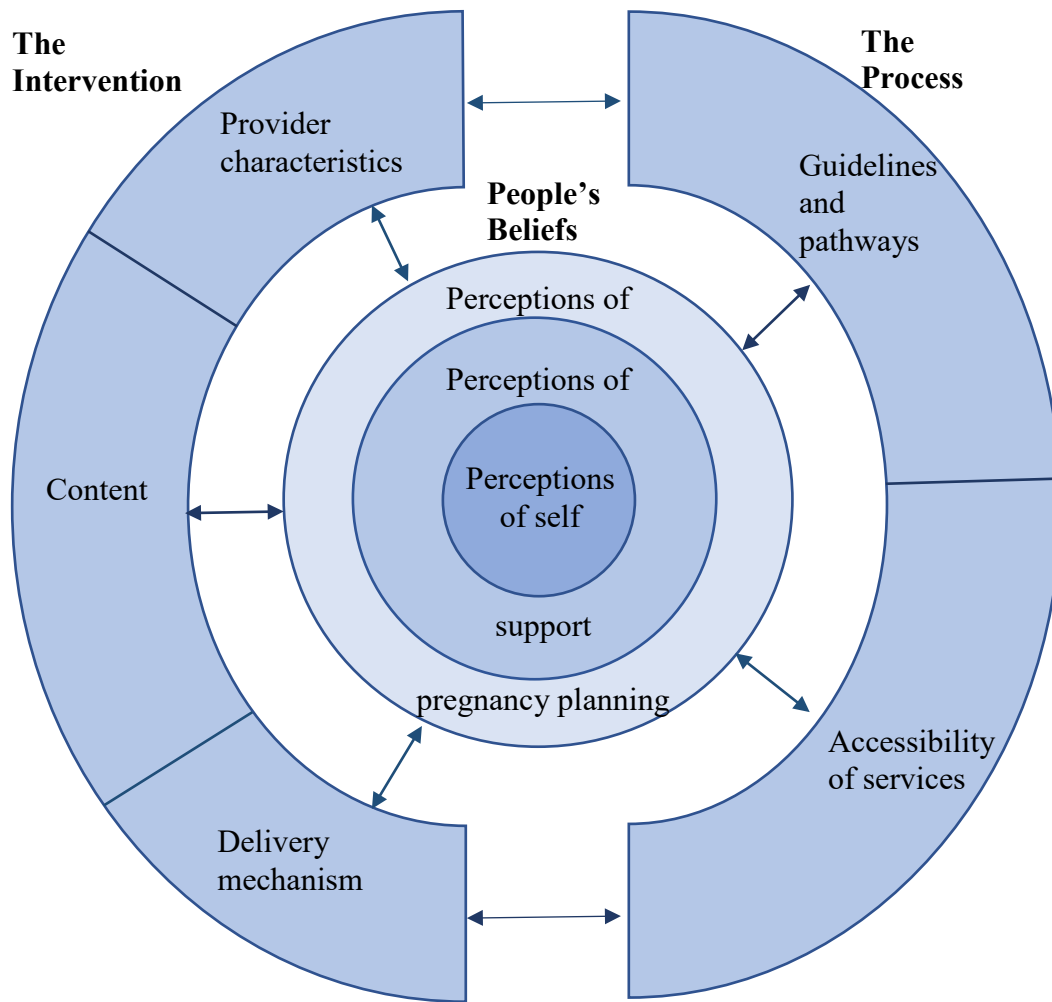


Figure 2: Subdivisions and connections between the three areas that were found to contribute significantly to both access and behavior change in relation to preconception care.