Decision-making and future pregnancies after a positive fetal anomaly screen: A scoping review

1 Abstract

Aims and Objective: To examine and consolidate literature on the experiences and decision-making of parents following a screen positive result for a potential fetal anomaly and/or diagnosis of an actual anomaly in a previous pregnancy.

Background: Prenatal screening consists of any diagnostic modality that is aimed at acquiring information about a foetus or an embryo; however, the entire process is highly stressful for parents, especially if there was a previous screen positive result, but no abnormality was detected in the final result.

Methods: Eight electronic databases (PubMed, Embase, CINAHL, PsycINFO, Scopus, Web of Science, ProQuest Theses and Dissertations and ClinicalTrials.gov) were searched from each database's inception until February 2022. This scoping review was guided by Arksey and O'Malley's framework and was reported in accordance with the PRISMA-ScR checklist. Braun and Clarke's thematic analysis framework was utilized.

Results: Thirty-one studies were eligible for inclusion. Two main themes (reliving the fear while maintaining hope, and bridging the past and future pregnancies) and six sub-themes were identified.

Conclusions: A fetal anomaly diagnosis in pregnancy had a mixed impact on the attitudes of parents toward a future pregnancy. Some parents were fearful of reliving a traumatic experience, while others were determined to have a healthy child and grow their family. Parents generally expressed a greater preference for non-invasive over invasive prenatal testing due to the procedural risks involved.

Relevance to clinical practice: There is a need for healthcare professionals to provide psychosocial and emotional support to parents so that they can achieve resolution for their previous pregnancy. Healthcare professionals' ability to provide informational support also enables these parents to make informed decision and understand their reproductive outcomes. Additionally, healthcare administration and policymakers should reconsider current neonatal or pregnancy loss bereavement guidelines to improve the inclusivity of fathers.

Patient or Public contribution: No patient or public contribution.

Keywords: Congenital abnormalities; forecasting; pregnancy; prenatal diagnosis; parents; review

2 Introduction

Prenatal screening consists of any diagnostic modality that is aimed at acquiring information about a foetus or an embryo, specifically for identifying prenatal genetic disorders and their characteristics (Wieacker & Steinhard, 2010). Screening for birth defects was initiated in the 1950s with the use of an ultrasound, and it has continued to remain the primary, routine, and established screening method to date (Carlson & Vora, 2017). The World Health Organization recommends pregnant women to have their first prenatal ultrasound within the first 12 weeks of gestation (World Health Organization, 2016). Ultrasound is a basic screening test on foetus, and women who receive a screen positive result for potential abnormalities are generally offered more definitive diagnostic tests which can be invasive (usually more precise), or non-invasive (usually less precise) (Allyse et al., 2015). The process of waiting to receive a diagnostic test and undergoing the procedure, particularly if it is invasive, and waiting for the results to be released can be highly stressful, even if the final result does not indicate the presence of an abnormality. If an abnormality is present, the stress is accentuated by the need for the woman, partner and family to decide on the next step (Lotto et al., 2017).

Prenatal ultrasound is usually a positive experience as parents can receive visual confirmation of their pregnancy and many expectant parents consider this as a defining step in their journey toward becoming a parent (Carlsson & Mattsson, 2018). However, in approximately three percent of all cases, parents would receive a screening test result which suggests a fetal anomaly, leaving them with the difficult decision to: (1) continue the pregnancy without further testing and experience ongoing uncertainty until or even after birth, or (2) undergo invasive diagnostic tests that may provide either more assurance of fetal well-being, or certainty of abnormality but risking the loss of a healthy foetus (Carlson & Vora, 2017). If a fetal anomaly is found, parents face a greater difficulty to decide whether to terminate the pregnancy, or continue on with the pregnancy but bring an "affected" child into the world (Carlson & Vora, 2017). Parents are often ill-prepared to receive adverse outcomes and experience a wide range of emotions ranging from anger, disbelief, grief, isolation, and adapting to adjustment (Lotto et al., 2017). In a recent review, a few women who had healthy babies, but initially had screen positive result on their ultrasound which turned out to be negative later on, continued to believe that their babies were not healthy. Hence, they were reluctant to be pregnant again, even though they had originally planned to have more children (Moncrieff et al., 2021).

To date, research on experiences of parents following a screen positive test result for potential or actual fetal anomaly predominantly focused on the experiences and decision-making of parents during the eventful pregnancy. There is a lack of systematic reviews which explore the experiences and decision-making of parents during the current and subsequent pregnancies. We managed to retrieve two qualitative systematic reviews that were similar to our proposed review, but both only briefly touched on our phenomenon of interest (Blakeley et al., 2019; Moncrieff et al., 2021). Blakeley et al. (2019) synthesized factors which influenced parents who considered terminating their pregnancy, or those who continued with the

pregnancy following the identification of lethal, life-limiting, or severely debilitating fetal abnormalities. On the other hand, Moncrieff et al. (2021) examined the short- and longer-term beliefs, concerns, experiences and views of women, partners and health workers on routine ultrasound in the first and second trimesters of pregnancy. Only five studies that focused on future pregnancy outcomes out of a possible 104 included articles were found in both reviews; one was from Blakeley et al. (2019) and the other four were from Moncrieff et al. (2021). This shows that future pregnancy experiences and decision-making of parents following a screen positive result for a potential fetal anomaly in a previous pregnancy, whether or not it was later confirmed by other diagnostic procedures, is an emerging topic; literature on this topic has different focuses and study designs. Therefore, we undertook a scoping review approach over a traditional systematic review (Munn et al., 2018).

3 Methods

The methodological approach of this scoping review was guided by Arksey and O'Malley (2005)'s five-stage framework consisting of: (1) identifying the research question, (2) identifying relevant studies, (3) study selection, (4) charting the data, and (5) collating, summarizing and reporting the results. This review was reported according to the guidelines of the Preferred Reporting Items of Systematic Reviews and Meta-Analyses Extension for scoping reviews (PRISMA-ScR) checklist (see Supplementary File 1) (Tricco et al., 2018).

3.1 Stage 1: Identifying the research question

This stage encompassed the identification of the research question and the purpose of the scoping review. The research questions were as follows:

1. How does a positive fetal anomaly screening result in a previous pregnancy impact the experiences and decision-making of parents (e.g., whether or not to pursue a future pregnancy) before a future pregnancy?

2. How does a positive fetal anomaly screening result in a previous pregnancy impact the experiences and decision-making of parents (e.g., about fetal anomaly screening during a future pregnancy) during a future pregnancy?

3.2 Stage 2: Identifying relevant studies

Eight electronic databases (PubMed, Embase, CINAHL, PsycINFO, Scopus, Web of Science, ProQuest Theses and Dissertations, and ClinicalTrials.gov) were searched from each database's point of inception until February 2022. An academic librarian was consulted to guide the search process, and an initial search was conducted in PubMed using the following concepts: ("forecasting" OR "pregnancy") AND ("prenatal diagnosis" OR "ultrasonography") AND ("congenital abnormalities" OR "life change events"). Keywords and index terms were combined using Boolean operators and truncation symbols to optimize the results. To ensure greater comprehensibility, a search on gray literature resources (OpenGrey and MedNar) was carried out alongside a thorough hand-search of the bibliographies of relevant studies. The detailed search strategies for the databases are presented in Supplementary File 2.

3.3 Stage 3: Study selection

All types of quantitative and qualitative study designs were eligible if they focused on the experiences and decision-making of parents following a screen positive result for a potential or actual diagnosis of a fetal anomaly in a previous pregnancy for future pregnancies. Studies where parents received any abnormal prenatal screen positive result or definite diagnosis during scheduled prenatal screenings for fetal anomaly, or as a chance or an unexpected finding in any previous pregnancies were included even if the foetus or child was found to be free of an anomaly upon detailed investigation (i.e., false-positive results). Studies exploring all fetal anomalies regardless of lethality, life-limitability, severity of debilitation, and length of impact were included, and participants were included regardless of their choice to continue or

terminate a previous affected pregnancy. To gather more inclusive and holistic evidence, all publication types (e.g., conference abstracts, editorials and opinions) were included except for books, book reviews and studies without full texts. Although the reviewers' native language is English, no language restrictions were set and Google Translate was utilized to translate studies outside of the reviewers' native language. Studies where parents: (1) received an expected diagnosis, or who were known carriers of genetic conditions, (2) received an abnormal diagnosis postnatally, and/or (3) received a non-fetal-related diagnosis during pregnancy such as maternal or placental conditions, were excluded. The Endnote X9 program was used to organize search results and remove duplicate records (The EndNote Team, 2013). Next, the titles and abstracts of all the studies were screened according to the inclusion and exclusion criteria. Thereafter, the full texts of selected studies were assessed for eligibility. Two independent reviewers conducted the screening process using Rayyan (Ouzzani et al., 2016), and any disagreements were discussed until consensus was met. An inter-rater reliability of 95% was maintained for the title or abstract screening, and full-text screening.

3.4 Stage 4: Charting the data

Based on the research question, the reviewers independently extracted the following: study author(s), year, country, study design, aim, population characteristics (number of parents, parents' age, and gender), type of diagnostic test, fetal diagnosis, pregnancy outcome, methodology (data collection, measurement points), outcome measures (if applicable), and findings related to the experiences, or decision-making of parents for future pregnancies (qualitative themes or subthemes, quantitative data, narrative summary). Any disagreements were discussed until consensus was met.

3.5 Stage 5: Collating, summarizing and reporting the results

A narrative synthesis approach guided by Popay et al. (2006) was utilized to summarize the data from the included quantitative and qualitative studies. First, textual summaries of each included study was systematically produced (including similar information for all studies where possible and in the similar order) for the reviewers to gain familiarity with the included studies, and to identify similarities and differences in findings across studies. Thereafter, the reviewers organized the included studies into groups and clusters according to the research questions to enhance the process of description and analysis, and identified patterns across and within these groups and clusters. These initial groups were refined as the synthesis developed. Subsequently, data from the included studies (e.g., study design details, outcome measures, results, or themes) were presented in a tabular form to visually represent both quantitative and qualitative data. Tabulation aided the reviewers in the preliminary synthesis of data across studies and provided important foundations for future elements of the synthesis process. Next, Thomas and Harden (2008)'s thematic synthesis approach was utilized to translate and interpret the data from the included studies. This process consisted of inductive coding of the data, development of descriptive themes and generalization of analytical themes. Inductive codes were generated using manual color-coding method and then compared, grouped and organized into subcategories to form descriptive themes(Nowell et al., 2017). These descriptive themes underwent a process of constant targeted comparison with textual data of the included studies to uncover and collate new and systematic understanding of parental experiences and decision-making for future pregnancies, and eventually the development of analytical themes (Sandelowski & Barroso, 2007). To enhance the reflexivity of the scoping review, a collaborative approach between the reviewers from screening to analysis was adopted to examine and reduce the influence of each reviewer's own beliefs, judgments and practices on the research process (Dodgson, 2019). The inter-rater reliability between the reviewers was approximately 90% for the data extraction and thematic analysis process, and any

disagreements were resolved through discussions. As the objective of scoping reviews was to provide an overview of existing evidence, a quality appraisal of the included studies was not conducted (Arksey & O'Malley, 2005; Levac et al., 2010). However, during data extraction, the reviewers ensured that all included studies stated ethical approval and/or implemented appropriate methodologies to obtain knowledge from their participants.

4 Results

4.1 Database search

A total of 39,333 studies were identified from the database search and manual search. Upon the removal of 17,003 duplicate studies, the titles and abstracts of 22,330 studies were screened based on the eligibility criteria, and 22,014 studies were excluded. The remaining 316 studies had their full texts screened for eligibility and 31 studies were included in the review. The detailed PRISMA flow diagram along with reasons for exclusion is presented in Figure 1.

Figure 1: PRISMA flow diagram (Insert Figure 1 above)

4.2 Characteristics of the included studies

All included studies were published between 1984 and 2021. The 31 studies consisted of three publication types: peer-review (n=27), theses or dissertations (n=3), and conference abstract (n=1). Three study types were identified: qualitative (n=21), quantitative (n=9), and mixed-method (n=1). Twenty-eight single-country studies were conducted across 14 different countries: United States (USA; n=7), Brazil (n=3), Sweden (n=3), United Kingdom (UK; n=3), Canada (n=2), the Netherlands (n=2), and one each in Belgium, Germany, Iceland, Iran, Scotland, South Africa, Switzerland, and Vietnam. Three studies conducted multi-country research: Lafarge et al. (2019) (France and UK), Samango-Sprouse et al. (2020) (Australia, Canada, Israel, UK and USA) and Hammond et al. (2021) (Netherlands and UK). In total, 28

English language studies and three foreign language studies were retrieved, i.e., German (Götzmann et al. (2002) and Wollenschein et al. (2007)) and Portuguese (Benute et al. (2006)).

4.3 Characteristics of the population

A total of 1338 responses were analyzed across 31 studies, of which 1192 were mothers, 121 were fathers or partners aged from 13 to 55 years old (25 participants' gender was unidentifiable). Approximately 1251 pregnancies were recorded from the responses. Twenty-one studies consisting of approximately 820 pregnancies reported on the type of prenatal diagnostic test utilized by participants in their previous pregnancy. The use of only non-invasive prenatal testing to confirm a prenatal diagnosis (i.e., ultrasound only or ultrasound with a combination of maternal serum screening, e.g., free β-hCG level and PAPP-A level, nuchal translucency, and/or fetal echography) was utilized in 504 pregnancies. Conversely, ultrasound with invasive prenatal testing (i.e., amniocentesis, chorionic villus sampling, exome sequencing, and/or fluorescence *in situ* hybridization) was utilized to confirm a prenatal diagnosis in 316 pregnancies. Amniocentesis and chorionic villus sampling were the most common choices, accounting for 241 and 62 invasive prenatal tests utilized, respectively. Twenty-six studies consisting of approximately 967 pregnancies reported the decision of parents after receiving their prenatal diagnosis. Termination of pregnancy was chosen by 750 parents,while 217 parents chose to continue the pregnancy. Approximately 65 unique fetal conditions were reported; the most common diagnoses reported in the studies were trisomy 21 (n=14) and anencephaly (n=8). Detailed characteristics of the included studies and the specific fetal diagnoses identified are presented in Supplementary Files 3 and 4, respectively.

The thematic analysis generated two main themes: (1) re-living the fear while maintaining hope, and (2) bridging the past and , future pregnancies. Six subthemes were also generated. Further details on the themes and subthemes are provided in Supplementary File 5.

4.4 Theme 1: Re-living the Fear while Maintaining Hope

This theme explored the experiences and decision-making of parents before a subsequent pregnancy. They were categorized into three subthemes: (1) too traumatized to move on; (2) seeking closure with future pregnancies; and (3) connecting the dots.

4.4.1 Too traumatized to move on

Nineteen studies (Baillie et al., 2000; Brandenburg et al., 1992; Bryar, 1997; Carolan & Hodnett, 2009; Dallaire et al., 1995; Evers-Kiebooms et al., 1988; Fernandes et al., 2020; Gammeltoft et al., 2008; Hammond et al., 2021; Irani et al., 2019; Jones et al., 1984; Kelly, 2009; Leuthner et al., 2003; Menary, 1987; Ndjapa-Ndamkou et al., 2013; Pelly, 2003; Rillstone, 1999; White-Van Mourik, 1989; Wollenschein et al., 2007) highlighted the reasons why parents were reluctant to become pregnant again following a diagnosis of a fetal anomaly in a previous pregnancy. For these parents, the fear of the anomaly recurring and reliving the trauma was sufficient to prevent them from becoming pregnant again (Baillie et al., 2000; Brandenburg et al., 1992; Bryar, 1997; Carlsson & Mattsson, 2018; Fernandes et al., 2020; Ferreira da Costa et al., 2005; Gammeltoft et al., 2008; Hammond et al., 2021; Irani et al., 2019; Kelly, 2009; Leuthner et al., 2003; Menary, 1987; Rillstone, 1999; White-Van Mourik, 1989). This fear also persisted in parents whose children received a normal diagnosis following detailed investigations (Baillie et al., 2000). Additionally, parents felt that choosing not to become pregnant again would allow them to ultimately avoid the difficult decisions associated with another screen positive result, i.e. whether to do a test at all, terminate or continue the pregnancy, or “choose” to bring a child with anomalies into the world (Kelly, 2009; Rillstone, 1999). Parents who previously believed a diagnosis of fetal anomaly only happen to others were left feeling vulnerable to a sense of the arbitrariness of a diagnosis, particularly when they had done all they could to ensure a healthy pregnancy (Baillie et al., 2000; Bryar, 1997; Carolan

& Hodnett, 2009; Gammeltoft et al., 2008; Kelly, 2009; Leuthner et al., 2003; Menary, 1987; Rillstone, 1999; White-Van Mourik, 1989). A previous positive diagnosis of fetal anomaly caused women to lose confidence in their reproductive capacity to produce a healthy baby (Carolan & Hodnett, 2009; Fernandes et al., 2020; Hammond et al., 2021; Pelly, 2003), and sowed doubts about their worth as wives and mothers. (Gammeltoft et al., 2008; Menary, 1987). In contrast, men in two studies reported fewer fears and vulnerabilities, and a shorter duration of negative emotions compared to women (Leuthner et al., 2003; White-Van Mourik, 1989). Parents' willingness to only consider a future pregnancy if there is zero probability of fetal anomalies coupled with the limitations of current medicine to predict and control risks inherent to reproduction with absolute certainty, was another reason they reported for not embarking on a subsequent pregnancy (Kelly, 2009; Rillstone, 1999).

Parents were more likely to decide against a future pregnancy if: (1) women were of advanced maternal age due to concerns of reduced fertility and increased complications following a possible future termination, or during birth and in the postpartum period; (2) they received a more severe and potentially recurring fetal diagnosis (e.g., chromosomal anomalies) previously, or (3) they had previously borne children with disability, or experienced fetal death, stillbirth or neonatal death (Brandenburg et al., 1992; Menary, 1987; White-Van Mourik, 1989). Rates of subsequent pregnancies in these groups were lower than that of parents with only healthy children, or no previous children (Brandenburg et al., 1992; Menary, 1987; White-Van Mourik, 1989). Notably, parents in two studies were able to find closure with their previous pregnancy through therapy, focusing on their education and careers, and adoption (Menary, 1987).

4.4.2 Seeking closure with future pregnancies

Eighteen studies (Baillie et al., 2000; Brandenburg et al., 1992; Bryar, 1997; Carolan & Hodnett, 2009; Dallaire et al., 1995; Evers-Kiebooms et al., 1988; Fernandes et al., 2020; Gammeltoft et al., 2008; Irani et al., 2019; Kelly, 2009; Lafarge et al., 2013; Lafarge et al., 2019; Menary, 1987; Ndjapa-Ndamkou et al., 2013; Pelly, 2003; Rillstone, 1999; White-Van Mourik, 1989; Wollenschein et al., 2007) highlighted the reasons why parents had a future pregnancy, or tried for one following a screen positive result for a potential or actual fetal anomaly in a previous pregnancy. Parents in seven studies highlighted that giving birth to a healthy child was essential to achieve a closure for their previous pregnancy since they could: (1) heal emotional wounds, (2) restore a sense of equilibrium in the family unit, and (3) remove doubts about their reproductive capabilities (Dallaire et al., 1995; Gammeltoft et al., 2008; Lafarge et al., 2013; Menary, 1987; Rillstone, 1999; White-Van Mourik, 1989). Additionally, a new pregnancy helped them focus on the future instead of the past and allowed them to rebuild their lives (Dallaire et al., 1995; Menary, 1987). These parents overcame the situation and accepted their experience with the previous pregnancy, and were optimistic that their subsequent child would be a different child, a child unto itself, and not a replacement or a means to forget the previous child (Gammeltoft et al., 2008; Lafarge et al., 2013; Lafarge et al., 2019; Rillstone, 1999). However, a new pregnancy also created ambivalence for parents who dealt with feelings of excitement and hope at a chance at regaining normality, while still feeling anxious about their increased risk of adverse obstetrical and perinatal consequences (Bryar, 1997; Lafarge et al., 2019; Pelly, 2003). Parents' determination to conceive again transcended these fears (Carolan & Hodnett, 2009; Ferreira da Costa et al., 2005; Gammeltoft et al., 2008; Menary, 1987; Pelly, 2003; Rillstone, 1999), and this desire was more significant for parents who did not have children and those who had experienced spontaneous fetal loss or terminated their previous pregnancy following a diagnosis of fetal anomaly (Brandenburg et al., 1992; Fernandes et al., 2020; Irani et al., 2019).

For other parents, the source of their determination came from their faith and their confidence in surviving another "ordeal" from previous hardships and losses in their life, which demonstrated their post-traumatic growth (Menary, 1987). Other parents felt they were capable of parenting another affected child after already caring for one and highlighted that increasing social acceptance of individuals with disabilities reaffirmed their decision (Kelly, 2009). While parents in three studies waited before trying for a future pregnancy to recuperate physically and emotionally, strengthen their marriage, or seek a medical opinion (Gammeltoft et al., 2008; Ndjapa-Ndamkou et al., 2013; Wollenschein et al., 2007), others did so as soon as their postpartum fertility returned. At times this was earlier than the medical staff advised (Menary, 1987). A small group of parents in the review reportedly tried for a future pregnancy without first resolving their grief and issues from the previous pregnancy, or did not have a plan on how they would cope if they receive a screen positive result for a fetal anomaly again (Ferreira da Costa et al., 2005; Menary, 1987).

4.4.3 Connecting the dots

Nine studies (Gammeltoft et al., 2008; Irani et al., 2019; Kelly, 2009; Menary, 1987; Pelly, 2003; Rillstone, 1999; Samango-Sprouse et al., 2020; Smith et al., 2021; White-Van Mourik, 1989) highlighted how parents navigated the period before or immediately after a screen positive result for a potential or actual fetal anomaly. Parents who wanted a future pregnancy reportedly had unprotected sex and copulated more frequently to reinforce closeness and to conceive, while parents who were against a future pregnancy abstained from intercourse (Kelly, 2009; Menary, 1987; White-Van Mourik, 1989). Parents who abstained from copulation did not trust contraceptives to avert an unplanned pregnancy, and their depression and sadness contributed to their lack of interest in a future pregnancy (Menary, 1987; White-Van Mourik, 1989). Parents in three studies underwent invasive permanent birth control (e.g.,

tubal ligation, vasectomy) to alleviate their worries (Kelly, 2009; Menary, 1987; White-Van Mourik, 1989).

Attitudes toward genetic counselling were largely positive, and parents in this review reported having sought a genetic counsellor, or intended to do so for a future pregnancy. Parents in seven studies viewed genetic counselling essential to increase awareness of their unique risks and the likelihood of recurrence for anomalies, enhance their odds of having a healthy child, ensure the well-being and safety of their future child, organize treatments and link parents with resources, and advocate for and provide objective, non-judgmental and compassionate support to improve parents' quality of life (Gammeltoft et al., 2008; Irani et al., 2019; Menary, 1987; Pelly, 2003; Rillstone, 1999; Samango-Sprouse et al., 2020; Smith et al., 2021). Some parents reluctantly attended genetic counselling as it was compulsory for prenatal testing (Smith et al., 2021). Parents did not utilize this service when they had negative past experiences with genetic counsellors, or when they already possessed the information they desired (Smith et al., 2021). Only a few parents in the review mentioned seeking professional psychological counselling for previous traumatic pregnancy experiences (Gammeltoft et al., 2008; Menary, 1987; Rillstone, 1999).

4.5 Theme 2: Bridging the Past and Future Pregnancies

This theme detailed parents' attitudes and behaviors in making decisions during a future pregnancy. They were categorized into three subthemes: (1) taking charge of the situation, (2) attitudes toward invasive prenatal testing, and (3) managing expectations and staying guarded during future pregnancy.

4.5.1 Taking charge of the situation

Fifteen studies (Bakkeren et al., 2020; Brandenburg et al., 1992; Carlsson & Mattsson, 2018; Evers-Kiebooms et al., 1988; Georgsson Öhman et al., 2006; Götzmann et al., 2002;

Irani et al., 2019; Kelly, 2009; Kristjansdottir & Gottfredsdottir, 2014; Larsson et al., 2010; Leuthner et al., 2003; Pelly, 2003; Rillstone, 1999; Samango-Sprouse et al., 2020; White-Van Mourik, 1989) highlighted parents' attitudes toward non-invasive prenatal testing for future pregnancies. Experiencing fetal anomaly detection changed parents' views and awareness about non-invasive prenatal testing and its medical purpose (Carlsson & Mattsson, 2018; Götzmann et al., 2002; Irani et al., 2019; Samango-Sprouse et al., 2020). They reported that non-invasive prenatal testing allowed fetal issues to potentially be detected early and allowed them to be well-equipped for the future, such as healing their psycho-emotional wounds and grief to ensure a smooth postpartum period, preparing for how their child will look and making arrangements for a good delivery, or death post-birth if the child had a poor outcome (Georgsson Öhman et al., 2006; Götzmann et al., 2002; Larsson et al., 2010; Leuthner et al., 2003; Rillstone, 1999; Samango-Sprouse et al., 2020). Additionally, an early diagnosis gave parents more time to research, and discuss different prognoses and views about the treatment to have a better understanding of their child's diagnosis (Georgsson Öhman et al., 2006; Götzmann et al., 2002; Larsson et al., 2010; Rillstone, 1999; Samango-Sprouse et al., 2020). Consequently, parents who took up this option were not only more proactive and more medically capable to care for their children, but were also advocates for their child's needs. Parents engaged in assembling and coordinating a team of medical specialists (on their own and without referrals) from different disciplines to ensure the best care plans, sought out community doctors and therapists and supplemental resources such as enrichment activities and support groups, and applied early for treatments, therapies and subsidies which often had long waiting lists (Larsson et al., 2010; Rillstone, 1999; Samango-Sprouse et al., 2020).

Parents in eleven studies had utilized or intended to use non-invasive prenatal testing (especially ultrasound examinations) for a future pregnancy (Bakkeren et al., 2020; Evers-Kiebooms et al., 1988; Georgsson Öhman et al., 2006; Götzmann et al., 2002; Irani et al., 2019;

Kelly, 2009; Kristjansdottir & Gottfredsdottir, 2014; Larsson et al., 2010; Leuthner et al., 2003; Samango-Sprouse et al., 2020; White-Van Mourik, 1989). The skills of the healthcare professionals who performed the procedure, the comprehensiveness and comprehensibility of the information received, and confidence with the efficiency and diagnostic reliability of non-invasive prenatal testing were the common external reasons why parents chose non-invasive prenatal testing in a future pregnancy (Götzmann et al., 2002; Samango-Sprouse et al., 2020).

Parents in two studies felt that the window of optimal intervention for their future child was narrow, hence they felt responsible to strategize their child's care. This meant that they were not willing to wait until after birth to decide on care plans and treatment options (Leuthner et al., 2003; Samango-Sprouse et al., 2020). To diminish the fear and emotional pain associated with waiting for a diagnosis confirmation, parents in one study sought to "accelerate the diagnosis" by pushing for non-invasive prenatal testing to be done at the earliest possible date, choosing alternative tests with shorter waitlists or quicker results, and traveled across states or cities if the technology was not available in their area (Rillstone, 1999). Parents in the aforementioned study even opted for invasive prenatal testing (which can be done as early as in the first trimester) rather than waiting and seeing, despite the increased risks; this will be discussed in the subsequent section (Rillstone, 1999).

Parents in three studies reported either believing that non-invasive prenatal testing should be a mandatory and routine part of prenatal care (Götzmann et al., 2002; Samango-Sprouse et al., 2020), or had undergone multiple non-invasive prenatal testing and opted in for optional incidental findings for comprehensiveness (Bakkeren et al., 2020). Parents in three studies (Kelly, 2009; Kristjansdottir & Gottfredsdottir, 2014; Samango-Sprouse et al., 2020) were against utilizing non-invasive prenatal testing for future pregnancies as they did not wish to be confronted with the choice of continuing or terminating an affected pregnancy. They felt such procedures caused psychological suffering and prevented a "happy" pregnancy, or they

were not going to terminate the foetus regardless of the diagnosis, or they had doubts over the reliability and consistency of non-invasive prenatal testing.

4.5.2 Attitudes toward invasive prenatal testing

Twelve studies (Brandenburg et al., 1992; Carlsson & Mattsson, 2018; Evers-Kiebooms et al., 1988; Georgsson Öhman et al., 2006; Jones et al., 1984; Kelly, 2009; Kristjansdottir & Gottfredsdottir, 2014; Larsson et al., 2010; Pelly, 2003; Rillstone, 1999; Samango-Sprouse et al., 2020; White-Van Mourik, 1989) highlighted parents' attitudes toward invasive prenatal testing for future pregnancies following a screen positive result for a fetal anomaly in a previous pregnancy. Parent in six studies did not, or had no intention to utilize invasive prenatal testing for a future pregnancy (Georgsson Öhman et al., 2006; Kelly, 2009; Kristjansdottir & Gottfredsdottir, 2014; Rillstone, 1999; Samango-Sprouse et al., 2020; White-Van Mourik, 1989). Parents, especially women of younger maternal age, were concerned with procedural complications such as injury to the foetus or mother, infection, preterm labour, miscarriage and stillbirth (Evers-Kiebooms et al., 1988; Kelly, 2009; Rillstone, 1999; Samango-Sprouse et al., 2020). Choosing whether or not to have invasive testing appeared to either be an emotional decision or based on incorrect beliefs on the distinct differences between non-invasive prenatal testing and invasive prenatal testing. Parents in two studies refused invasive prenatal testing. They chose to deal with the anomalies (if any) and stressors after the child was born as they believed that despite very low risks, their odds would somehow not be in their favor. Others believed that invasive prenatal testing was unnecessary as their experience with their previous affected pregnancy or child adequately prepared them to manage any impairments (Kelly, 2009; Rillstone, 1999). Parents in four studies reported that they used or intended to use invasive prenatal testing in a future pregnancy as they believed that invasive prenatal testing would enhance the safety and well-being of the foetus, and had higher

sensitivity and specificity compared to non-invasive prenatal testing (Evers-Kiebooms et al., 1988; Jones et al., 1984; Larsson et al., 2010; Samango-Sprouse et al., 2020).

4.5.3 Managing expectations and staying guarded during a future pregnancy

Fifteen studies (Baillie et al., 2000; Benute et al., 2006; Bryar, 1997; Carolan & Hodnett, 2009; Gammeltoft et al., 2008; Kelly, 2009; Lafarge et al., 2013; Lafarge et al., 2019; Leuthner et al., 2003; Menary, 1987; Rillstone, 1999; Samango-Sprouse et al., 2020; Smith et al., 2021; White-Van Mourik, 1989; Wollenschein et al., 2007) highlighted how parents navigated the prenatal period in a subsequent pregnancy following a screen positive result for a fetal anomaly in a previous pregnancy. If faced with a similar situation as their previous pregnancy, parents reported wanting to change how they would deal with the question of terminating or continuing the pregnancy. Parents in three studies who were younger women not of advanced maternal age were more accepting of their decision to terminate their previous pregnancy and were in favor of doing so again if an abnormality is diagnosed in a future pregnancy (Benute et al., 2006; Kelly, 2009; White-Van Mourik, 1989). Conversely, women of advanced maternal age, who generally were pro-terminating their previous pregnancy , had lower expectations and gravitated toward continuing the pregnancy regardless of the diagnosis (Kelly, 2009; Menary, 1987; Rillstone, 1999). These parents greatly reconsidered their hopes and desires for a “perfect baby” who would outlive them since the ability to journey through life as a “normal” family became their priority (Kelly, 2009).

A large number of parents included in the review approached their subsequent pregnancy, or intended to approach their future pregnancy with caution and not take things for granted. To manage this uncertainty, they expected and prepared for the worse (Baillie et al., 2000; Bryar, 1997; Carolan & Hodnett, 2009; Gammeltoft et al., 2008; Kelly, 2009; Leuthner et al., 2003; Menary, 1987; Rillstone, 1999; White-Van Mourik, 1989). This attitude was

associated with an increase in fear, insecurity, worry and stress (Rillstone, 1999; Wollenschein et al., 2007). To manage this, parents in three studies deliberately delayed acknowledging and investing in the pregnancy and avoided developing emotional attachment with their foetus (Lafarge et al., 2013; Lafarge et al., 2019; Wollenschein et al., 2007). They did so either out of self-protection and self-preservation against a bad fetal outcome, or because they were still grieving the loss of the previous child and could not bond with the present child (Rillstone, 1999). Parents who did not resolve this "denial" promptly found themselves investing late into the pregnancy and this left them with less time to prepare for the birth and more anxiety closer to the delivery (Rillstone, 1999).

Compared to previous pregnancies, more parents withheld or said they would withhold news of a new pregnancy until they were completely certain that their child had a normal diagnosis (Lafarge et al., 2019). Parents were afraid that receiving love and support from others would prematurely raise their hopes and worsen their pain if unsuccessful, and having to break bad news to others and witnessing their pain, would add to their pain (Rillstone, 1999). On the other hand, parents in two studies (Rillstone, 1999; Wollenschein et al., 2007) experienced a better pregnancy experience in their subsequent pregnancy. By acknowledging they could lose their child at any moment, and not wishing to have any regrets, these parents actively shared about their pregnancy and made a bond with their child to ensure that their child felt important and loved.

Parents needed an "enhanced level of hand-holding" during the subsequent pregnancy. Compared to an average pregnant woman or birthing person, they made more visits to medical institutions and even requested for extra checkups, saw various healthcare professionals, depended more heavily on healthcare professionals for informational and emotional support, and had a more therapeutic physician-patient relationship (Lafarge et al., 2013; Lafarge et al., 2019; Rillstone, 1999). Additionally, parents in two studies reported utilizing local and online

pregnancy support groups to share their experiences, seek connection, reassurance and support from "credible" parents, and manage plans while coping with the pain of a difficult pregnancy (Rillstone, 1999; Smith et al., 2021).

5 Discussion

This review consolidated evidence on the experiences and decision-making of parents for future pregnancies following a screen positive result for potential or diagnosis of an actual fetal anomaly in a previous pregnancy. In subsequent pregnancy, many parents reported higher levels of anxiety due to the increased risk of an adverse outcome. This phenomenon was also identified by Campbell-Jackson et al. (2014) who found that women who suffered a perinatal loss experienced higher levels of post-traumatic stress, anxiety and depression compared to women who have not had any experience of loss. Poorer prenatal psychological health was also associated with an increased risk of negative perinatal outcomes in a future pregnancy such as preterm birth and low birth weight (Grote et al., 2010). Furthermore, compared to uneventful pregnancies which are generally associated with positive emotions and expectations, pregnancies following a perinatal loss (stillbirth or neonatal death) are emotional-laden and increases anxiety level due to parental fears of a recurring loss (Mills et al., 2014). Additionally, studies found that women who underwent a previous induced abortion also had higher anxiety and depression scores in their subsequent pregnancy (particularly in the first trimester), and these scores were statistically similar to women who had experienced spontaneous abortion (Broen et al., 2005; Huang et al., 2012). Therefore, healthcare professionals should routinely consider women's history of prenatal or perinatal loss and history of positive screening test results during their risk assessment for prenatal and postpartum anxiety and depression so that high-risk women and partners can be identified and supported.

Parents in this review reported that giving birth to a healthy child would help to heal emotional wounds, rebalance their family, and alleviate the anxieties surrounding their reproductive ability. This finding was corroborated by Barr (2006) and Gold et al. (2010) who found that a future pregnancy would provide the wished-for-baby to facilitate the integration of the prior loss, while a healthy pregnancy would restore maternal self-esteem often ravaged by perinatal loss. However, Blackmore et al. (2011) showed that the depression and anxiety symptoms related to a previous perinatal loss can persist well beyond the subsequent pregnancy despite the birth of a healthy child. Furthermore, Dekel et al. (2017) reported that despite a successful birth, post-traumatic stress response brought about by the childbirth experience may still manifest in mothers and they may also suffer from childbirth-related postpartum traumatic stress disorder. Therefore, healthcare professionals must remain vigilant to recognize early symptoms of these phenomena (especially in new mothers) and provide ongoing long-term support even after a subsequent healthy pregnancy to promote smooth adaptation. Although many parents were prepared to move on and proceed with a subsequent pregnancy, they were adamant that doing so was not a means to forget or replace their previous child. These findings were similarly reported in Campbell-Jackson et al. (2014) and Brooten et al. (2015) who found that although the parents' emphasis was on the next child, it was still important for them to maintain a "relationship" and keep the memory of their previous child alive for themselves, their families and communities. Healthcare administrations should ensure that healthcare professionals receive adequate and necessary training so that they can be more wary and sensitive to such needs and allow open conversations where parents can discuss previous losses.

While there were parents in our review who chose to wait to process the physiological and emotional stress of a previous pregnancy, other parents (specifically those who did not have children and were of advanced maternal age) were determined to conceive again as soon

as possible. Although this group of parents were often the more fearful and had more unresolved previous pregnancy issues, they were more likely to go against medical advice regarding recovery. This could be because they might feel that they had limited time as their biological clock was ticking. However, the World Health Organization (2007) recommends that women should have a minimum birth interval of 6 months after an abortion or fetal loss, or a minimum of 24 months after a live birth for optimal maternal and perinatal outcomes. Conversely, other studies have shown that pregnancy spacing of 18-23 months was associated with lower incidence of low birth weight, preterm birth and small for gestational age, among other adverse perinatal outcomes (Conde-Agudelo et al., 2006; Grisaru-Granovsky et al., 2009). This waiting period poses a particular problem for parents in high-income countries who may be toward the end of their childbearing years, given the growing trend of delayed childbearing due to economic and social reasons (Nargund, 2009). There is a positive relationship between increasing maternal age and adverse maternal (i.e., gestational diabetes, hypertension and preeclampsia) and child outcomes (cesarean birth, fetal growth restriction, miscarriage, placental abruption, preterm birth) (Lean et al., 2017). Aref-Adib et al. (2008) reported a 30% increase in miscarriages for mothers who were 40 years of age, which then rose to 50% after 45 years old. Additionally, Alio et al. (2012) reported a 24% increase in miscarriages for paternal ages ranging 40-45 years old, which further rose to 50% after 45 years old. As reflected in our review, parents of advanced childbearing age (≥35 years) were aware that every delay in attempting conception would severely lower the chance of a healthy baby. Therefore, parents (especially first time parents) were more likely to plan a subsequent pregnancy shortly after the first one. Healthcare professionals should provide parents, especially bereaved parents, counselling on how to optimize their health before preparing for a future pregnancy, and discuss the medical risks and benefits of delaying versus trying to ensure that parents can make an informed decision.

Parents who chose not to have a future pregnancy wanted to avoid reliving their traumatic pregnancy and felt that they were more vulnerable to the recurrence of fetal anomalies. This was the case even for parents whose subsequent diagnosis ruled out abnormality after an initial screen positive result. Healthcare providers need to understand this phenomenon since the usual assumption of a woman who had a healthy baby is to assume that there are no residual concerns following an earlier screen positive result. This area is underreported and requires more primary research.

Our findings showed that men reported a lower level of fear and vulnerability, and experienced a shorter duration of negative emotions compared to women. These findings were also reported by Kersting and Wagner (2012) who found that men grieved less intensely and for a shorter period compared to women after prenatal loss. However, this does not conclusively reflect a causal relationship between gender and how they cope with perinatal loss. We hypothesize that men are generally less likely to outwardly express emotions due to societal expectations of how men should behave (Obst et al., 2020). This is further supported by Due et al. (2017) who showed that men had a higher propensity than women to engage in maladaptive compensatory mechanisms (e.g., substance abuse), had higher scores on avoidance scales and expressed greater difficulty approaching and accessing support services. Additionally, Williams et al. (2020) found that men whose wife experienced a miscarriage felt less entitled than women to describe their feelings due to the fear of being shamed and rejected since it was not them who physically lost the baby. Our hypothesis was also reinforced when Miller et al. (2019) reported that although men experience grief of a similar intensity to women over a perinatal loss, they understand that their primary role is to support their partner and are less likely to report their feelings to, or in the presence of their partners. Given that current perinatal support services and information are largely targeted at women, men are often unintentionally neglected by healthcare professionals. Therefore, more research into the

experiences of men in perinatal research, specifically regarding their coping mechanism and how they express themselves in situations like this, are warranted.

According to Cacciatore et al. (2013), positive experiences with healthcare professionals increase psychosocial care and support group participation, and reduce grief and depressive symptoms in men. As there is a tendency to focus on women's expression of grief and view men in a primarily supportive role to women (McCreight, 2004), healthcare professionals must first recognize and break negative preconceived notions related to paternal grief. Healthcare administrations should ensure that healthcare professionals receive adequate and necessary training so that they can better validate the experiences of men, and display empathy and sensitivity toward their needs, similar to how they would with women. Doing so could build more therapeutic alliance between healthcare professionals and men, and improve male receptiveness to subsequent psychosocial interventions. Additionally, healthcare administrations should also reform current neonatal or pregnancy loss bereavement guidelines which primarily focus on the experiences and needs of women.

We realize that parents' emotional state plays a significant role in deciding whether or not they choose to have invasive testing, i.e., some parents are willing to absorb the risks associated with invasive testing, while others avoid receiving a diagnosis. Additionally, we also highlighted that there were parents who associated non-invasive prenatal testing with confirmation of a diagnosis. This indicates that those parents either did not comprehend the information or were not adequately educated about non-invasive prenatal testing. Non-invasive prenatal testing is a screening test that can only determine the risk profile of women toward a fetal anomaly, and not refute or confirm a diagnosis (Allyse et al., 2015). Misinformation is further exacerbated as parents tend to have a tunnel vision on the safety aspects of different prenatal tests. This is confirmed by Hill et al. (2012) who found that a prenatal test with zero or negligible risk of procedure-related complications, or loss was the most important factor

that women considered before deciding to undergo prenatal testing. Compared to parents of older childbearing age, younger parents were more likely to decline invasive prenatal testing due to concerns with complications. Both Mujezinovic and Alfirevic (2007) and Hill et al. (2012) reported similar findings observed in our review. They also found that women of advanced maternal age generally valued tests with the highest accuracy and produced the quickest results. However, these tests often presented with greater risks. Conversely, younger women (<35 years) generally valued tests with the lowest possible false-positive rate and risk. However, these tests usually had longer waiting times and lower accuracy. Mujezinovic and Alfirevic (2007) and Hill et al. (2012) hypothesized that this difference between age groups could be due to personal experiences and experiential knowledge. Therefore, as safety weighs heavily on parents' minds, healthcare professionals must clearly and carefully counsel parents on the differences between the available prenatal tests and their implications to ensure that parents can make informed decision and achieve reproductive autonomy, and tailor a care pathway according to their needs and level of risk.

Parents, especially women, in our review tended to compartmentalize their pregnancy to avoid the emotional aspects for as long as possible until they received a greater certainty of success, or had a live baby in their arms. More parents withheld news of their subsequent pregnancy and only informed a smaller circle of contacts. This conscious and at times subconscious defensive mechanism has been investigated in previous studies and is known as 'bracing for the worst' (Bailey et al., 2019; Ockhuijsen et al., 2013), 'emotional cushioning' (Côté-Arsenault & Donato, 2011), or 'holding back emotions' (Cĵté-Arsenault & Dombeck, 2001). Generally, this is a normal and adaptive process as parents are still able to maintain good quality relationships and function normally in society while being protected from the potential pain of another loss (Côté-Arsenault & Donato, 2011). However, maternity care professionals must still recognize the existence and prevalence of this behaviour, and keep a close eye as

parents (especially men) may not reach out for support and underreport their true anxiety levels. Maternity care professionals should adopt an authentic listening approach and provide ample space and time for parents to make a decision on their support needs.

5.1 Limitations

A limitation of this review is that although efforts were made to include non-English language articles in this review using Google Translate, relevant studies may have been left out due to limitations in translating. Furthermore, the majority of the articles included in this review were from high-income settings (n=24), where prenatal testing and termination of pregnancy for fetal abnormality are generally (though not always) socially acceptable. Thus, our findings may not be generalizable to other contexts with a different ethical and legal landscape. More geographically and culturally diverse research on the experiences of parents from Africa, Asia or Middle East is warranted in future. This is to account for socio-cultural and political differences, and inequalities in access to good quality maternity care, screening and counselling services in some of these settings. Although the Arksey and O'Malley (2005) framework highlights that a review of the findings by stakeholders and consumers with an interest in the topic would provide additional references and insights beyond those in literature, we did not undertake this step due to time and resource constraints. Additionally, we did not register an *a priori* protocol as scoping reviews are presently ineligible for registration on PROSPERO. However, we acknowledge that doing so aids in enhancing the comprehensibility, reproducibility and transparency of a scoping review, and we encourage future researchers to undertake this step in alternative registries such as Figshare (https://figshare.com/) or Open Science Framework (https://osf.io/) (Aromataris & Munn, 2020).

5.2 Relevance to future research

The Patterns, Advances, Gaps, Evidence for practice and Research recommendations (PAGER) framework guided this section (see Table 1) (Bradbury-Jones et al., 2021).

Table 1: PAGER framework for practice & research implications (insert Figure 1 above)

Men accounted for less than 10% of the responses in the included studies, and only one study (Carlsson & Mattsson, 2018) focused exclusively on the experiences and decision-making of men. Future studies should focus on improving the representation of fathers in perinatal health research, specifically understanding how fathers cope with and express grief, and identifying strategies to increase male involvement. As all of the included studies were conducted before the coronavirus disease 2019 (COVID-19) pandemic, future studies should account for the effects of the COVID-19 pandemic restrictions and their concomitant impact on healthcare delivery when examining the phenomenon of interest. Additionally, as the objective of scoping reviews is to provide a holistic overview of existing evidence, we did not set any date restrictions, and thus some dated studies were included. Within the past few years, social and political views regarding termination of pregnancy access have shifted dramatically across continents, particularly in the United States. Additionally, the introduction of new technologies in the last decade has also drastically changed the current practice of prenatal screening and testing for fetal anomalies. Therefore, as parents' experiences and views may be influenced by societal discourses and what is available to them, the consensus of some dated studies may not be relevant in today's society. Therefore, there is an urgent need for newer primary qualitative and quantitative studies, ideally with a longitudinal design, so that a comprehensive understanding of the phenomenon can be achieved and parents can be provided with timely support.

6 Conclusion

This review consolidated the experiences and decision-making of parents for future pregnancies following a screen positive result for potential fetal anomaly or diagnosis of an actual problem in a previous pregnancy. Our findings demonstrated that both scenarios had a mixed impact on the attitudes of parents toward having a future pregnancy. While some parents became more fearful of living through a traumatic experience again (in some cases even if their baby was healthy), other parents were more determined to give themselves a chance to have a healthy child and a normal family. Many parents expressed a greater preference for non-invasive prenatal testing over invasive prenatal testing due to the procedural risks involved. Our findings highlight the need to focus on the roles that healthcare professionals play in terms of providing psychosocial and emotional support to parents so that they can achieve resolution for their previous pregnancy, and also as a source of informational support to ensure that parents make informed decisions and understand the reproductive outcomes. Additionally, our findings also indicate the need to reform current neonatal or pregnancy loss (bereavement) guidelines to create greater inclusivity by including bereaved men.

7 Relevance to Clinical Practice

Healthcare professionals must ensure that parents are well supported from the moment they receive a screen positive result for potential fetal anomaly or a diagnosis of actual fetal abnormality until well after, regardless of whether they decide on a future pregnancy. During this process, healthcare professionals must be sensitive to the needs of parents and allow them to openly discuss their experiences to aid in the grieving process. For parents who wish to embark on a subsequent pregnancy, healthcare professionals should provide continuity of care, if possible. Even if this is not possible, those caring for parents in this situation should carefully counsel them on inter-pregnancy interval timings, the various types of prenatal tests available and their implications, and the distinct differences between non-invasive prenatal testing and invasive prenatal testing to ensure that they make informed decisions and obtain optimal

reproductive outcome. Healthcare professionals should also refrain from stereotyping men, and neglecting their experiences and needs. This would enable the development of a therapeutic relationship and improve health-seeking behaviors in men. Lastly, healthcare administrations and policymakers could work toward reforming current neonatal or pregnancy loss bereavement guidelines, which currently are primarily focused on the experiences and needs of women, to ensure greater inclusivity for bereaved men.

Data availability statement: The data that supports the findings of this study are available in the supplementary files of this article

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