Managing broken expectations after a diagnosis of fetal anomaly

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\textbf{ABSTRACT}

The prenatal diagnosis of a fetal anomaly is unexpected, and for many parents it is devastating. It is considered a traumatic perinatal event that presents a crisis for parents. Expectant parents dealing with a lethal anomaly are particularly at increased risk for perinatal depression, anxiety, and traumatic stress. A growing number of qualitative researchers have examined the experience of fetal anomaly. Despite the accumulating knowledge, theory has not been developed. The purpose of this study is to analyze and synthesize evidence drawn from grounded theory research in order to develop a theory that describes and explains the process parents use to manage the diagnosis and predict their needs. A systematic search was conducted in 5 databases. Fourteen grounded theory research articles were chosen and were analyzed with grounded theory methods. Parents’ expectations about pregnancy and future parenting were broken with the diagnosis of a fetal anomaly. Parents use a six-stage process of repositioning to work through the problem and direct their course on the best new pathway for their family. The factors that enhance parents’ repositioning can facilitate the development of interventions to improve the care for these families.

1. Background

Fetal anomalies affect one in every 33 babies born in the United States each year, and are the leading cause of infant mortality, accounting for 20% of all infant deaths (Center for Disease Control, 2022). The prenatal diagnosis of a fetal anomaly is unexpected, and for many parents it is devastating. It is considered a traumatic perinatal event that presents a crisis for parents (Aite et al., 2011). After diagnosis, parents begin a complex, emotionally challenging process (Hodgson & McClaren, 2018). Expectant parents dealing with a lethal anomaly are particularly at increased risk for perinatal depression, anxiety, and traumatic stress (Dempsey, Chavis, Willis, Zuk, & Cole, 2021; Oftedal et al., 2022). To provide care for these families, healthcare providers need to understand the experience and the process that parents use to cope with this crisis.

A growing number of qualitative researchers have examined the experience of fetal anomaly in a variety of contexts. Methods used to summarize the findings have included integrative reviews (Berry & Colorafi, 2019; Kravotovil & Julian, 2017) and systematic reviews of qualitative research (Lou, et al., 2017). Interpretive synthesis and meta ethnography have been used to synthesize multiple qualitative studies on the parent journey (Wilpers et al., 2021), birth experiences during termination (Jones, Baird, & Fenwick, 2017), and decision making for termination in lethal anomaly (Blakeley, Smith, Johnstone, & Wittkowski, 2019; Lafarge, Mitchell, & Fox, 2014). Despite the accumulating knowledge about the experience, theory has not been developed. Although synthesis methods are ideal for the development of theory (Lier & Smith, 2017), investigators have been slow to utilize this method to develop practice theories. This may be due to a lack of awareness of how these theories could be applied in the clinical setting (Finfgeld-Connett, 2016).

A theory, grounded in data, would explain the expectant family process and predict their needs. Theory in this childbearing context is necessary to guide practice, develop and test interventions, and improve patient care. The following question guided the inquiry: what is the process used by expectant parents to deal with a diagnosis of fetal anomaly? The purpose of this paper is to describe the development of a clinically relevant theory that describes and explains the process used by expectant families.

2. Method

The goal of the grounded theory method is the discovery of the basic
social processes that are used by persons in response to specific problems (Glaser & Strauss, 1967). Substantive, or situation-specific theory is developed from data and explains the problems of individuals and the process (Basic Social Process) and the strategies they use to resolve or cope with the problem. The core category, which explains variation in the theory, is identified during the analysis (Glaser, 2007). A growing body of grounded theories around fetal anomaly has been conducted.

Once several substantive theory reports on the same area are evaluated together, common patterns can be identified. Sociologists combine multiple substantive theories together to produce a higher level grounded formal theory that describes an experience that applies to many contexts (Glaser & Strauss, 1967). Although grounded theory has been used increasingly as a qualitative research method in health care research, very few authors have developed a higher-level theory from the synthesis of multiple grounded theories. An adaptation of formal grounded theory methods (Kearney, 1998a) was used to develop theories (AlOmeir, Patel, & Donyai, 2020; Kearney, 1998b, 2001) to guide understanding of specific health and illness experiences and develop clinically useful models and interventions. While narrative reviews and meta synthesis create a summary or synthesis across a variety of samples, this method goes beyond merely assembling and describing past work and develops a theoretical explanation for differences in individual outcomes (Kearney, 1998a). We used this clinically focused approach of grounded theory synthesis to develop a higher-level theory to guide practice.

2.1. Literature search strategy

To identify relevant literature, we consulted with a medical librarian to conduct a comprehensive search of multiple computerized library databases, including PubMed, Scopus, CINAHL, Ovid PsycInfo, and Google Scholar. Search terms included MeSH terms such as fetal anomaly, congenital anomalies, congenital birth defects, fetal deformity, maternal behavior, expectant parents, parental responses, obstetric care, experiences, coping, and qualitative study or research. Limits were set to include only peer reviewed research articles published in English from 2000 to 2020. Quantitative or mixed methods studies, scale validation studies, literature reviews and systematic reviews and gray literature were excluded. Rayyan, a web-based tool was used to conduct and coordinate the collaborative screening and selection process. We identified additional resources through a manual search and review of reference lists from the initial search and systematic reviews. The search and article selection processes, modeled after the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) guidelines (Page et al., 2021), are outlined in Fig. 1.

2.2. Selection and eligibility criteria

Criteria for inclusion of studies were the use of a grounded theory or a theory-generating approach method, with findings that went beyond description to generate a theory, and discover at least a core category or a basic process (Kearney, 1998a). All relevant studies meeting these criteria were considered for inclusion. Publications describing other qualitative methods were considered if the findings included theoretical components that could be analyzed with constant comparative techniques. The judgement for inclusion was based on the title and subsequently the abstract. If it was unclear, then the entire paper was read and assessed against the inclusion and exclusion criteria. Two authors reviewed all articles independently and any conflicts were discussed to reach consensus on whether the criterion was present. Reviewer opinion differed on six reports. These were reviewed and discussed to reach consensus on whether the criterion was present. Thirty-three reports were retrieved, and 28 were evaluated. Nineteen studies were excluded due to methods (qualitative or descriptive), infant care focus, or lack of...
adequate description of an analytic approach. The final sample included nine studies. Five studies were included from additional searching. The 14 primary research articles retained in the sample used grounded theory methods or techniques and demonstrated building of concepts or theories from direct quotations.

2.3. Critical appraisal

The appraisal of the individual studies was evaluated using criteria specific to the grounded theory method. The Guideline for Reporting and Evaluating Grounded Theory Research Studies (Bertheisen, Grimshaw, & Hanson, 2018) was used to determine the methodological quality of the individual studies. Of the articles, 7 identified a basic social process (BSP) and four identified a core category. Two authors independently reviewed the selected articles and then discussed whether the criteria was present. All articles met the criteria and all authors used constant comparison to analyze their data and generate theory.

2.4. Data analysis and theory development

Data analysis and theory development was conducted using grounded theory research methods (Glaser & Strauss, 1967). The first author created a data matrix tool to extract descriptive data about the studies. Details included the year, author and discipline, context, purpose or research question, methodology, description of the sample, categories, the basic process, and core category, if identified, from each article. A second author independently checked and added to the extraction process.

The researchers' theoretical findings and data were used to develop a broader, more useful theory. From the research reports, direct quotes of participants and author narratives were imported into a word processing program. The substantive reports were interrogated as if they were interview transcripts in which the participant has offered a summary of her or his experience of a phenomenon (Kearney, 1998a). Data was coded by both authors, using an inductive method. During open descriptive coding, constant comparison was used to identify similarities and differences among codes. Codes were developed and placed into categories. Categories were constantly compared and merged. Selective coding was performed until a core category emerged. Theoretical sampling of data was then used to develop the theory. Higher level theoretical coding further refined the theory by identifying process, stages, strategies, context, varying conditions, and the core category and the basic social process. The emerging theory was compared to other theories of responding to an unexpected crisis in several different contexts and transitions in perinatal settings (e.g., preterm labor, fetal loss, and admission to the Neonatal Intensive Care Unit). Memos were written during both the authors' analysis and meetings to discuss and compare our analysis throughout. Data analysis continued until theoretical saturation was reached. After the theory was developed it was evaluated for quality using method-specific criteria (Glaser, 1978). Two nurses working in the clinical specialty area and one who had experienced a fetal anomaly with her first pregnancy reviewed the theory. Positive feedback and confirmation of clarity and usefulness and improvement suggestions were provided to further refine the theory.

3. Results

3.1. Characteristics of included studies

The 14 studies reviewed included a total of 240 total participants reported as 169 women and 71 men or partners with an age range of 21–44 years who obtained care at multidisciplinary obstetrical care centers providing fetal diagnosis and treatment for expectant parents across eight countries. The diagnosis of a fetal anomaly in most women occurred during the 8–20-week period by prenatal genetic screening or screening ultrasound. In one study, 20 women whose babies had died within 15 months, continued the pregnancy with the expectation of a still birth, or learned of the anomaly at the birth, or up to after 30 days of the birth (n = 4) (Welch, 2018). Women who experienced medical termination of pregnancy (n = 89) were identified in 5 studies (Im et al., 2018; Lafarge, Mitchell, & Fox, 2013; Lalor, Begley, & Galavan, 2009; Maijala, Paavilainen, Astedt-Kurki, & Paavilainen, 2003; Rillstone, et al., 2001). The remainder of the women (n = 80) delivered their baby for further care as needed. The categories of fetal anomaly included both structural and functional anomalies with varying severity. Severity of fetal diagnosis ranged from conditions like choroid plexus or complex genetic conditions including Trisomy 21, Edwards and Noonan syndromes to immediate life-threatening conditions such as severe cardiac abnormalities and anencephaly. The publication date range of the studies spanned from 2001 to 2019, across eight countries, including, the United States of America, United Kingdom, Ireland, Sweden, Finland, Australia, Korea, and China. Disciplines represented in the studies included nurse researchers, nurse midwives, psychologists, social workers, geneticists, and genetic counselors. An overview of included studies can be found in Table 1.

3.2. Repositioning: managing broken expectations after a diagnosis of fetal anomaly

Broken Expectations of their Path to Parenthood was the major problem faced by parents. Expectant parents eagerly anticipated seeing their healthy baby on the monitor during their routine ultrasound at mid pregnancy. Others completed prenatal genetic screening tests. Soon thereafter they received the unexpected news of a fetal anomaly. Their innocence of creating joyful plans became a crisis, and their expectations of a joyous birth had been lost. They struggled with feelings of shock, unreality, disbelief, grief, and denial. They felt devastated and expressed worries about their baby and their postnatal future. Many had questions, and most desired an immediate need for information to understand their situation. Previously many parents had envisioned a particular life path for their anticipated healthy baby. A few parents had experienced a prior loss due to fetal anomaly. Once they received the diagnosis the expected path toward parenthood changed direction. This required parents to make sense of and reassess their current path position and view of the future. The main concern for parents was managing their broken expectations.

Parents used the basic process of repositioning to manage their broken expectations. The core category, repositioning, initially involves determining current location or position on the path. Following that, repositioning involves changing direction by navigating to the best position to travel the path. Some paths were straightforward, others were more complicated. Repositioning brought participants to a resolution of the problem of the broken expectations. This process occurred in six stages and was influenced by both environmental and personal factors or conditions. The unique combination of these factors determined the best pathway for each family.

3.2.1. Stage 1 living in innocence

At mid-pregnancy, an ultrasound is commonly performed to evaluate fetal anatomy. Women anticipated the upcoming ultrasound. They were expecting to see a healthy fetus and to find out if it was a girl or a boy. “We were full of expectation and thought it would be very exciting to see our baby” (Larsson, Svalenius, Lundquist, & Dykes, 2010, p 4). Though the parents were aware that the examination could indicate or confirm an abnormality, they expected their baby would be healthy.

I had heard about it [ultrasound] from other people so it was something I was looking forward to. I probably think it was to check for dates and just to check everything was okay. This time she [community midwife] gave me a leaflet, I didn't even read it (Lalor et al., 2009, p. 466).
3.2.2. Stage 2 receiving the diagnosis
The stage of living in innocence ends abruptly with getting the unwelcome news of an abnormal ultrasound or genetic screening finding from their provider. Some participants were present when the results were shared, and others received the information by telephone. Getting the unwelcome news changes the feeling of innocent anticipation when the parent learns that there is a fetal anomaly.

Receiving the diagnosis redirects the care for the baby and mother most often to a specialty clinic. The clinic consists of a multidisciplinary provider team composed of maternal-fetal medicine providers, nurses, and pediatric subspecialists. The participants entered this phase with differing amounts of knowledge about the diagnosis of anomaly. The parents received the news of the diagnosis of a life-limiting congenital anomaly. The new and awful reality is that the baby will die at birth or during the delivery. A father speaking for his wife and himself noted “it was a shock, and we’re not going to be the one … it was a shock, and we were obviously totally rattled” (Hickerton et al., 2012, p. 376). The process of transition from initial shock to restructuring required learning new vocabulary and communication skills, consuming their time and energy. Simultaneously, they struggled to cope while experiencing multiple and powerful emotions. Disbelief was often accompanied by the shock at receiving the news and the parents began to understand that their pregnancy position is altered from normal expectations to a new reality. A father speaking for his wife and himself noted that, “You think you’re not going to be the one … it was a shock, and we were obviously totally rattled” (Hickerton et al., 2012, p. 376). The emotions were described as anguish, which was intense, severe, and extreme. A challenge for some parents was the “roller coaster” effect, a constant shifting in the timing, type, and intensity of emotions. Some parents received the news of the diagnosis of a life-limiting congenital anomaly. The new and awful reality is that the baby will die at birth or within a brief time after birth.

This stage marks the beginning of the process of understanding the news and applying early management strategies of denial, hoping, detachment, and distancing. The participants’ actions at this stage are more passive than in subsequent stages. Strong fluctuating emotions continue. A sense of control and meaning has not yet been established. They are beginning the process of determining their new position on their path to parenthood. Parents developed multiple strategies deal with the crisis. The types and number of these varied with the severity of the
anomaly. Though the increased speed of information was viewed as helpful it did not necessarily bring the family to fully believe in the condition.

Moving from stark unreality towards the current reality of the condition facing the baby was accomplished through the healthcare team. During this stage most healthcare providers offered key information, education, support, and often further testing. The level of involvement and support by the healthcare provider team increased parents’ coping by reducing uncertainty and increasing understanding. Obtaining understandable information and more time to discuss, discern and absorb it was a need expressed by many.

3.2.3. Stage 3 determining truth

This stage is punctuated by turning towards active repositioning, defined as an evolving process of understanding, seeking, confirming, and believing the information gathered from trusted or available resources, then finally adapting and accepting the information that has been presented. Some parents could not accept the situation. One mother recalled, “I find it difficult to give up my expectations. I mean, maybe something is wrong, maybe they (healthcare providers) gave me the results of another pregnant woman” (Qin et al., 2019, p. 88). Parents held onto hope in their minds, even if their hope was unrealistic, and made extensive efforts to determine the correctness of the diagnosis. They also asked for professional advice from providers at different hospitals in different cities to confirm the diagnosis. The need for information depended on the level of uncertainty about the diagnosis. After confirming the diagnosis, one mother recalled, “Although I knew that little hope existed, I kept a glimmer of hope until it was broken by all the rechecking” (Qin et al., 2019, p. 88). Parents described feeling a loss of control when receiving the diagnosis. Eventually parents came to an acceptance of the diagnosis. Acceptance was influenced by the visibility of the anomaly and information validating the diagnosis. Acceptance was a critical turning point to move along in the process. From the acknowledged truth about the condition of the baby, parents developed an understanding of their current position. The result of this phase of the process was increased control. Parents were better able to mobilize their own resources to a position of action and move forward when their need for information was met. This stage ends when the truth of reality is fully understood with the best available information at the time. Acceptance and understanding of their position on the pathway are a consequence of this stage. Management strategies included information-seeking, reconstructing reality, altering expectations and reassessing the future, and gathering support. Influences on this stage include severity of the anomaly, family, culture, social norms, and healthcare providers.

3.2.4. Stage 4 choosing direction

Choosing direction began with the movement from being passive recipients to becoming increasingly actively involved in their own care. Although they had received information about the anomaly, many continued to need information and support to make decisions about the direction of the next steps on the pathway. Some parents did not consider or need to decide. They confirmed their prior thought process to continue the pregnancy when the anomaly was not as serious. For a small group of parents, a decision was not considered or needed if their belief was that termination is wrong. After determining their truth and current position, they moved directly into repositioning and navigating their new pathway.

Others continued to the stage of choosing direction. The two major decisions were electing to perform genetic testing and continuing or terminating the pregnancy in cases of severe or lethal anomaly. Two main styles of decision making were identified: analytical and emotional (Lotto et al., 2018). This has been described as “head or heart” (Lotto, Smith, & Armstrong, 2018, p. 281). The head-led approach was a more rational style of weighing options. Parents using an organized analytical style used the strategies of gathering pros and cons or weighing options. Others approached the issues in a haphazard way. In the heart-led approach, or emotion-led style feelings or “gut reactions” were used in place of or in addition to practicalities. These parents were already grieving for their lost dream of a ‘perfect baby’ and were not able to weigh their options. Others did not decide as a passive reaction to an uncertain situation, or feeling unable to act, and the pregnancy continued by default (Lotto et al., 2018).

Following an abnormal ultrasound report and discussion of further testing, many parents found genetic testing to be a difficult decision because diagnostic testing procedures such as amniocentesis or chorionic villi sampling carry a risk of miscarriage. Parents needed to balance the risk to their baby with the need for information to inform their decision. The decision to perform genetic testing also could involve an additional concern: A moral dilemma. One father commented,

We were then faced with not only the dilemma… of raising a child with a… terminal disability but also… the moral decisions of, ‘Okay, we’ll do the testing, and then what do we do after that? Do we terminate or do we continue on?’

So there’s another sort of moral decision [that] never would normally have entered our minds. (Hickerton, et al., 2012, p. 377)

If the diagnosis was a severe or a lethal anomaly, parents moved into a decision about the next direction of the pathway. Regaining some form of control over the situation began with determining the truth and culminated with the decision to either continue or terminate the pregnancy. The degree and seriousness of the anomaly, and parents’ attitudes and cultural or religious beliefs about pregnancy termination and disability influenced the need for further information and their choices during decision making. Cultural norms and religious beliefs created a dilemma for some women, as they agonized over whether they should maintain the pregnancy. Termination may conflict with their cultural sense of duty to produce a healthy child (Im et al., 2018), or norms about acceptability or availability of termination. A serious or lethal anomaly created a position of emotional and moral difficulty in decision making. The process of choosing whether to continue the pregnancy was described as a dilemma and an emotionally painful time for parents. Most felt they had only negative choices. One father reflected, “In my mind, there was no choice,” while another father described it as “a choice between terrible and horrible and whichever way you go you're going to have to suffer major consequences and there's no real… win-win solution” (Rillstone & Hutchinson, 2001, p. 293). The decision to end or not to end their child’s life was recalled as agonizing. One mother explained, “because you have to be involved and be an active participant in the whole decision-making process. It's much, much, much easier to have things happen to you… as opposed to deciding that things will happen.” Another mother reported, “I think the thing that intensifies it to some degree is [the] incredible guilt associated at the same time.” (Rillstone & Hutchinson, 2001, p. 293).

When speaking about their choice to continue the pregnancy, participants primarily spoke of personal reasons such as not agreeing with termination of pregnancy or their perception of quality of life for the child (Hickerton et al., 2012). Religious or ethical conviction was also a clear reason for continuing the pregnancy. Although several parents considered themselves to be religious, this was not stated as a primary reason for continuing the pregnancy. In the situation of expected eventual death of the baby in infancy, decisions may also go beyond elective termination or continuing the pregnancy to delivery. After delivery, decisions may include resuscitation efforts at delivery and palliative care.

Parents who chose termination of pregnancy (TOP) felt that this was best for the baby in terms of potential suffering or the belief that coping with a disabled child was a situation that parents could not envision themselves in. One couple regretted the decision they made to terminate. All wished that they had never been placed in the position of having to
make the decision. Most parents felt that they had “made the right decision at that point... for that baby” (Billstone & Hutchinson, 2001, p. 293) using the information they had at the time. Due to the stigma surrounding abortion, most parents tended not to share their experience openly, causing them additional pain. When a decision had been made to terminate the pregnancy, couples faced another decision, the method. The choices included dilation and evacuation (D&E) or induction of labor and delivery. Considerations included gestation time and personal choice. Most women decided to have labor induced. Regardless of the termination method, many parents wanted to say goodbye to their baby (Welch, 2018).

Most parents valued being actively involved and appreciated receiving respect for their decisions. Health care providers played a significant role in supporting parents during the decision-making phase through providing information and guidance. While some parents needed support and guidance from the provider, others preferred to make the decision independently. Management strategies included venting, staying positive, religion, and seeking support.

3.2.5. Stage 5: navigating

During this stage, parents acted on their decisions and repositioned their path. In most cases this new pathway was the best choice for each family based on the condition of the baby, their beliefs, resources, and personal situation. As their pregnancies progressed, the baby was evaluated with ongoing monitoring and testing. Some parents expressed significant worries about the delivery and worsening of the baby’s status related to mortality, surgery outcomes, and neonatal intensive care. Steps taken to prepare for parenting were influenced by fetal and future child health and the degree of attachment, and the meaning of the diagnosis. Some women found it difficult to become attached to the baby or delayed attachment. This was often due to uncertainty about the baby’s condition and fear that developing a bond would intensify the emotional pain felt if the baby subsequently died (Lalor et al., 2009). They felt disconnected and viewed themselves as uncertain future parents. Others viewed themselves as parents and formed a loving bond (McKechnie, et al., 2015). They expressed closeness and affection.

Parents with a baby with a severe or lethal anomaly who chose termination described the procedure as the most difficult experience. Support from partners, family, and providers was essential. Many used distancing and distraction during the procedure. They discussed the loss of the normal post birth experience and closeness to the infant (Lafarge et al., 2013). Women receiving a diagnosis of a lethal anomaly frequently hoped that their baby would soon die, as it marked an end to waiting for the inevitable. For those diagnosed with an anomaly likely to survive up to birth or for a brief time afterwards, many hoped for a live birth. Women described leaving the hospital without their baby as heart-breaking, and the process of saying goodbye as surreal. Regardless of the method, the delivery experience was found to be emotionally painful. Management strategies included preparing, taking control, seeking support, social isolation (continuing pregnancy); distancing, receiving support, acknowledging the baby, creating memories, remembering the baby, letting go (termination and death).

3.2.6. Stage 6 going on

Going on requires regaining a sense of normalcy. Attributing meaning to the losses along the way and facing a new reality requires another layer of management strategies. These include a range of new tasks, including developing altered parenting, and finding meaning to the birth or death. For those who delivered a baby diagnosed with an anomaly who is not expected to die, the parents must go on with more complex parenting and medical needs than they had originally expected. Normalcy strategies may begin prior to birth, including shielding the baby by delaying sharing of the diagnosed anomaly with others.

For others, the stage of going on begins with termination or the death of the baby (Welch, 2018). Coping with the grief of the loss of the perfect baby or the actual or anticipated death of the baby creates sadness for the parents and the family. The concerns of this stage include trying to live with the decision, and grieving the loss of their baby, and consideration of future pregnancy. Going on does not mean getting over the loss of the baby. Rather it involves doing what needs to be done to live life. The parent goes back to work, marriage, and being a parent if applicable. However, life has forever changed. The duration of this phase differed. Most parents recalled that the months immediately following the death of their child were the most intense periods of anguish. They reported that it was impossible to return to the time before the diagnosis of a fetal anomaly. Baby clothes, pregnant women, or babies in the vicinity, and even things about babies mentioned by others, would trigger their traumatic memories and grief. One mother commented that “My heart would tremble if I recalled this thing TOP (termination of pregnancy)” (Qin et al., 2019, p. 88) while another recalled, “(It) is always in my mind, sometimes coming out in my dreams, so sad” (Qin et al., 2019, p. 88).

The intensity of anguish diminished over time. Almost all the parents described the anguish as “never going away.” “It isn’t going to go away, and I don’t think anybody else understands what we’ve been through” (Billstone & Hutchinson, 2001, p. 293). This anguish had a rippling effect on many aspects of their lives. Most parents did not want to forget about it. Many reported feelings of isolation and misunderstanding in their grief. Looking to the future was also linked to achieving closure. All women reported the process of healing as being uneven, lengthy, and like a roller-coaster. It could last years for some parents. For some participants, the termination also instigated personal growth, with some women reporting feeling stronger and more confident as a result. Readiness for another pregnancy was individual. Most women could see a future and were willing to consider another pregnancy. A small number were adamant that they would never put themselves in this position again. Management strategies included making meaning, denial, returning to work, thinking about the future.

As parents moved forward to a subsequent pregnancy, the previously experienced anguish returned to the forefront; parents were all too aware that the past could recur. The fear of experiencing additional anguish during a subsequent pregnancy caused these parents to develop protective strategies based on their prior experience. Management strategies included developing emotional armor, limiting disclosure, and lowering expectations.

3.2.7. Healthcare experiences and expectations throughout the process

Throughout all stages of repositioning, interactions with healthcare providers strongly influenced parents’ experiences both positively and negatively. Parents valued open and honest communication. Their priority was receiving accurate and understandable information about the condition of the baby and thorough and concrete explanations of the probable course of future events, with positive and negative aspects of management. They wanted support with decision-making if needed (Majala et al., 2003), to feel comfortable in the setting, adequate time for discussions, and having consistent providers throughout their care. They viewed the ideal healthcare provider as compassionate, non-judgmental, and empathetic. They recalled that providers demonstrated active caring by listening to and addressing fears, considering the parents’ perspective of the situation, and encouraging parents to make contact whenever necessary. Unfortunately, many parents reported a lack of information and poor communication. Some felt that their concerns and fears were not taken seriously or that they had been abandoned. Following termination of pregnancy, very few received follow-up care or services.

3.2.8. Influences on the process

There is considerable variation in how parents responded and managed the crisis (see Table 2, Variation in Influences). Both personal and environmental factors affected the complexity of the pathway and time to completion. Several factors facilitated repositioning. Personal factors were acceptance of anomaly, alteration of expectations, desire to be actively involved in their care, ability to understand the diagnosis and
Table 2
Variation in influences

<table>
<thead>
<tr>
<th>Influence</th>
<th>Supporting Quotes</th>
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<tbody>
<tr>
<td><strong>Situational</strong></td>
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<tr>
<td>Degree of Seriousness</td>
<td>“The doctors think our son won't make it. I used to think of a baby as far away from death as you can possibly get. Now death's at my side every day.” Non-Serious “At no point did I think about terminating the pregnancy, the baby was either going to make it or it wasn't. We weren't talking about dealing with a child that would be ill for the rest of our lives.”</td>
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<tr>
<td>Need for Decisions</td>
<td>“We had decided that even if he should have functional malformations, we should keep him.” Non-Need “I could see how that would have been the 'best' thing for some people … But that just wasn't an option for us.”</td>
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<tr>
<td>Support from Family</td>
<td>“... we rang my parents … and they just said, 'Look, she'll keep us all alive and she's come to a great family [crying] ... they could see the big picture …’” Non-Support “I got zero support from my family and they made it very clear that they didn't really want Emma to be part of their lives and focused a lot more on [our other two children], with presents.”</td>
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<tr>
<td><strong>Personal</strong></td>
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<tr>
<td>Acceptance of Diagnosis</td>
<td>Accept “I just accept the truth now.” Not Accept “I doubted the diagnosis based on the B ultrasound. It must be wrong … I still thought that it was a misdiagnosis after I got the results of the amniocentesis.”</td>
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<td>Need for Information</td>
<td>“I'm currently in a period when I read everything I come across … It's difficult, but I need to go through this. I need to understand what is happening!! I have such a hard time processing it all …” Not Need “I don't want to read anything on the internet; I don't want to go on any websites, I don't want to look at any pictures.”</td>
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<td>Managing</td>
<td>“I wanted them to know that we are okay with this [fetal diagnosis and having a child with medical needs] and life will go on as normal.” Not Managing “I am avoiding it completely. I have closed all doors on it.”</td>
</tr>
<tr>
<td>Involvement with Problem</td>
<td>Involved “She [radiographer] was concentrating very heavily on one particular area and she said I have to go and get someone else. But my straight away reaction was 'right, tell me what is going on'. Not Involved “When coming out [from the ultrasound] … The coming days I was in low spirits, it [the abnormality] circulated in my head the whole time and I could not do anything.”</td>
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<tr>
<td>Feelings About Abortion</td>
<td>Acceptable “I've always been brought up believing termination is wrong, but with all the things wrong with her, I just couldn't put her through [being born]. I mean the suffering would have been terrible.” Not Acceptable “… abortion was never something that I … really agreed with. I mean each case is different obviously.”</td>
</tr>
<tr>
<td>Attitudes About Disability</td>
<td>Acceptance “She's still a baby, and we're still happy to have her.” Non-Acceptance “My biggest fear is that the baby will survive and I'll be left with a seriously handicapped baby.” “I could not think about what it would be like if I had a congenitally abnormal child.”</td>
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<tr>
<td>Attachment</td>
<td>Attached “Even though she does have the problems that she does … I love her so much. …” Not Attached “Some women found it difficult to become attached or delayed attachment. This was often due to uncertainty about the baby's condition and fear that developing a bond would intensify the emotional pain felt if the baby subsequently died.” (Lalor et al., 2009, p.468)</td>
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<tr>
<td>Preparation</td>
<td>Preparing “We both instantly just viewed ourselves as parents, and what we needed to do to prepare, and taking the steps that we needed to take to be good parents.” Not Preparing “Distancing themselves from the pregnancy could involve stopping preparations for the infant: Decorating the nursery room, and buying baby clothes could all come to a halt.” (McKechnie et al., 2015, p. 119).</td>
</tr>
<tr>
<td>Readiness for Future Pregnancy</td>
<td>Desire “I am always wondering what went wrong during the previous pregnancy, and what I need to do better for the next pregnancy. I yearn for a healthy baby.” No Desire “A small number were adamant that they would never put themselves in this position again.” (Lalor et al., 2009, p. 469).</td>
</tr>
<tr>
<td>Culture</td>
<td>“When we got married, I promised I'd bear him a pretty and healthy baby …” “An assumption of the fetus's being is tied to the practice of encouraging early prenatal interactions with the fetus, referred to as Taekyo, a traditional concept thought to contribute to fetal development.” (Im et al., 2018, p. 5)</td>
</tr>
</tbody>
</table>
information, and develop and follow through on plans. Environmental factors were adequate information, feeling connected to providers, and support from providers, families and other groups and services. Many factors could either enhance or delay repositioning (see Fig. 2).

4. Summary and discussion

The data from the studies enabled an in-depth analysis of parents’ experiences across the course of mid pregnancy through postpartum. The theory developed from this synthesis identified the various paths that parents may need to travel, and how each path differs in complexity. This theory moves beyond a description of the experience, to identification of the stages, strategies, and the factors that enhanced their efforts in responding to the crisis. The core category, repositioning, explains the variation in the family response to the crisis and the strategies used for management.

4.1. Theory evaluation

When evaluated for quality (Glaser, 1978) this theory fits the area of fetal anomaly, is general enough to apply in multiple contexts, and has been evaluated by both a participant and clinicians working in the clinical area as understandable.

4.2. Comparisons

Broken or shattered expectations were a key finding in all of the studies. This concept was also reported in the findings of other qualitative perinatal studies of parents’ experiences of unexpected perinatal events such as fetal loss (Lusiuik, Comeau, & Newburn-Cook, 2013) and admission to the Neonatal Intensive Care Unit (Lowenstein, Barroso, & Phillips, 2019).

Many similar concepts of the theory were confirmed when it was compared to other qualitative synthesis studies of fetal anomaly and perinatal loss. As found by others, (Berry, Tarko, & Oneal, 2021; Lafarge et al., 2014), loss was a major concept. We identified similar coping strategies for dealing with the initial diagnosis and loss of a baby (Blackley et al., 2019; Fernandez-Basanta, Coronado, & Movilla-Fernandez, 2020). Findings about variation in the amount and sources of information parents wanted (Jones et al., 2017), the parental role in decision making (Rosenthal & Nolan, 2013), and decision styles (Paton, Armstrong, Smith, & Lotto, 2020) were confirmed. We identified similar needs for their care expressed by parents (Lafarge et al., 2013; Wilpers et al., 2021).

The discovery of a fetal anomaly transforms the pregnancy experience into an unexpected crisis. The theory has similar concepts of the Transactional Model of Stress and Coping Theory (Lazarus & Folkman, 1984), and the Crisis Decision Theory (Sweeney, 2008). There are also concepts similar to the Theory of Transitions (Meleis, Sawyer, Im, Messias, & Schumacher, 2000), which focuses on the transition process involved in parenthood and adapting to a change in health.

4.3. Limitations

The findings of this study are limited to a homogenous sample of mostly female participants from European, American, and Asian populations. The participants had successfully repositioned themselves through the diagnosis of an anomaly experience, which may not reflect those people who did not cope successfully through the crisis. The original researchers’ published findings were based on verbatim interviews, but our analysis was of a secondary nature and limited by the published results which were analyzed for this synthesis and theory. A potential for variation in the healthcare specialty experience exists due to the breadth of countries/states included in the study. There were no in-depth descriptions of insurance, access, or other care coverage issues. This theory is not generalizable to other populations. The theory can be built on by using other sources of data.

4.4. Implications for practice and research

Dealing with a diagnosis of fetal anomaly is a process that unfolds over time. The duration of time in each stage of the path is individual. Healthcare providers must anticipate and listen to the mother and the family and establish where they are in this path. The key influences identified by this theory will guide assessment and the development of evidence-based interventions to improve the care of these families. To assist parents to determine their best path and guide them as they navigate, interventions need to be individualized and tailored to parents’ stage and influences on the repositioning process.

Peer groups in the studies we reviewed were a positive source of support for women. Group prenatal care for women with fetal anomaly...
provided enhanced support, continuity, and learning opportunities (Schwartz et al., 2020). Further studies using this model could be needed.

The findings of the studies that were synthesized here completed their research at the stage just after delivery, and did not identify how families coped over time. Further research in this area would assist in anticipatory guidance and follow up of these families, particularly those with lethal anomaly and termination. We found limited research about the experiences and needs of fathers and partners participated in some of the studies.

5. Conclusion

Parents manage the crisis of fetal anomaly through a multi-stage process. The process of repositioning facilitates their travel on the new pathway. Both internal and external factors affect their decisions and strategies they use to manage the crisis. Several factors enhance a positive completion of their journey. These factors can be utilized by health care providers to guide and support parents as they navigate this new pathway.

CRediT authorship contribution statement

RT: Initiated the concept for the review, determined review design, developed eligibility criteria and conducted the search.

RT & CH; determined design; assessed the research studies for eligibility; evaluated the studies for quality, extracted the data, and conducted the analysis. Both obtained feedback on content from outside sources. Both reviewed the findings with the ENRQ Checklist. The final synthesized findings were written and agreed on. The final manuscript was written and approved by both RT and CH.

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Ethical approval

No human participants were involved in the preparation of this manuscript.

Data availability statement

The data used for this review is not available for upload. The reason is that the data we analyzed for the review came from published research articles that are available in the journals that we have cited. This data does not belong to us.

Submission declaration & verification

This manuscript has not been published previously, and is not under consideration elsewhere.

The Author Guideline has been reviewed.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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