

From: Through the Unexpected
To: [Birth Trauma](#)
Subject: Re: Birth Trauma - Post-hearing responses - 8 April 2024
Date: Monday, 29 April 2024 10:24:52 PM
Attachments:

Dear Julianna,

Thank you for the reminder email. Sorry for my delay in replying, COVID-19 stole a week from this household.

I do not have corrections to the transcripts. I have attached two journal articles:

- Psaila 2023: This is the study that was requested on notice about the PEARLS clinic.
- Shakes 2023: This is my PhD research, which identified the disassociation experienced by participants and described the larger threat and challenge of adaptation within the experience of prenatal diagnosis. I mentioned the state of disassociation and my research in my discussion; therefore, I wish to share this with the committee in case this is of interest.

With thanks,



Pieta Shakes (she/her) | Executive Director
Through the Unexpected

www.throughtheunexpected.org.au



Through the
Unexpected

Exploring continuity of care for women with prenatal diagnosis of congenital anomaly: A mixed method study

Kim M. Psaila RN, RM, PhD, Nurse Academic¹  | Virginia Schmied RN, RM, PhD, Nurse Academic¹  | Susan Heath RN, RM, Clinical Nurse Consultant²

¹School of Nursing and Midwifery, University of Western, Sydney, New South Wales, Australia

²PEARLS Team, Westmead Hospital, Sydney, New South Wales, Australia

Correspondence

Kim M. Psaila, Faculty of Nursing, Midwifery and Paramedicine (NSW/ACT) Division Health Sciences, Australian Catholic University, P.O. Box 175, Richmond, NSW 2753, Australia.
Email: Kim.Psaila@acu.edu.au

Funding information

Western Sydney University

Abstract

Aims: To map the pathway and service provision for pregnant women whose newborns require admission into the surgical neonatal intensive care unit at or soon after birth, and to examine the nature of continuity of care (COC) provided and the facilitators and barriers to woman- and family-centred care from the perspective of women/parents and health professionals.

Background: Limited research exists on current service and care pathways for families whose babies are diagnosed with congenital abnormality requiring surgery.

Design: A mixed method sequential design adhering to EQUATOR guidelines for Good Reporting of a Mixed Methods Study.

Methods: Data collection methods included: (1) a workshop with health professionals ($n=15$), (2) retrospective maternal record review ($n=20$), prospective maternal record review (17), (3) interviews with pregnant women given a prenatal diagnosis of congenital anomaly ($n=17$) and (4) interviews with key health professionals ($n=7$).

Results/Findings: Participants perceived care delivered by state-based services as problematic prior to admission into the high-risk midwifery COC model. Once admitted to the high-risk maternity team women described care 'like a breath of fresh air' with a 'contrast in support', where they felt supported in their decisions.

Conclusion: This study highlights provision of COC, in particular relational continuity between health providers and women as essential to achieve optimal outcomes.

Relevance to Clinical Practice: Provision of individualised COC offers an opportunity for perinatal services to reduce the negative consequences of pregnancy-related stress associated with diagnosis of foetal anomaly.

Patient or Public Contribution: No patient or public was involved in the design, analysis, preparation or writing of this review.

KEYWORDS

congenital anomaly, continuity of care, high-risk pregnancy, MAPS, transition of care

1 | INTRODUCTION

Congenital anomalies, defined as structural or functional anomalies that develop prenatally, may be identified before or at birth or later in

life. Some congenital anomalies treated with surgery, such as cardiac and neural tube defects, may have lifelong impacts. An estimated 6% of babies worldwide are born with a congenital anomaly, resulting in hundreds of thousands of associated deaths. In Australia in 2018, of

the 31,838 newborns admitted to special care nurseries or neonatal intensive care units (NICUs), 2400 families received a prenatal diagnosis of one or more major congenital anomalies (AIHW, 2020). This paper describes the experience of the care journey from admission to a high-risk maternity unit for the birth of their baby from the perspective of women who received an antenatal diagnosis of congenital anomaly.

2 | BACKGROUND

Parents receiving an antenatal diagnosis of a congenital anomaly are understandably distressed and apprehensive regarding the birth, which potentially may impact parental mental health, parent–infant attachment and infant development (Dagklis et al., 2016; Meaney et al., 2016). This significant distress is exacerbated by the practical and emotional difficulties parents encounter attending consultations with a diverse range of health professionals during antenatal and early postnatal periods (Munch et al., 2019).

Maternity, neonatal and child and family health (CFH) professionals, nationally and internationally, have emphasised the importance of continuity of care (COC) for all women and their families during pregnancy, birth and postnatal periods for over a decade (Fox et al., 2022; Psaila et al., 2013). Increasingly, care coordinator or liaison roles are being implemented to guide and advise women and their families on how to navigate their health journey across multiple services (Baldwin et al., 2019; Olley et al., 2017). In some services in Australia and England, midwives work as a core part of the multidisciplinary team offering women experiencing complications in pregnancy, COC across pregnancy, labour, birth and the postnatal period (Fernandez Turienzo et al., 2020), in line with the International Confederation of Midwives (ICM) perspective on continuity of midwifery care (ICM, 2021). ICM (2021) recognises the importance of COC with a known midwife for all women, encouraging midwives to work collaboratively with other health providers caring for child-bearing women and newborns by promoting respect, trust and open communication among all levels of healthcare providers.

Having a 'known' midwife is optimal for the woman and baby, demonstrating improved clinical outcomes for women with a low risk for poor outcomes (Sandall et al., 2016) and an increase in maternal satisfaction for women experiencing a medically high-risk pregnancy (de Wolff et al., 2021; Fox et al., 2022). A high-risk pregnancy refers to women who either have a pre-existing health condition or develop a condition during pregnancy, potentially causing complications for her or her baby. Therefore, women whose babies have been diagnosed with a congenital anomaly requiring surgery at or soon after birth are referred to major referral centres, such as the PEARLS Service, Westmead Hospital for maternal–fetal medicine (MFM) in Sydney, Australia.

This service supports women who receive a prenatal diagnosis of congenital anomaly within a woman-centred model of care with a known midwife, in collaboration with a multidisciplinary team. The service provides psychosocial support throughout the prenatal, intrapartum and postnatal period, coordinates services and guides women

What does this paper contribute to the wider global community?

- The PEARLS model of care has enabled midwives to support high-risk pregnant women at the multitude of critical antenatal time points they experience.
- The provision of informational, managerial and relational continuity provided in this model of care is essential to enable women and their families to cope and contribute to optimal outcomes.
- This study highlights the advantages of allocating COC midwives to women with high-risk pregnancies, particularly psychosocial care.

through the multidisciplinary networks, aiming to bring some normality to the transition to parenthood despite the woman experiencing a high-risk pregnancy. In addition, a multidisciplinary team specific to the baby's needs, including the cardiac, surgical, neurology, renal and gastrointestinal specialties from the co-located Children's Hospital, consult with families before the baby's birth. After birth, babies requiring surgery are transferred across hospital campuses to Children's Hospital Westmead for assessment and treatment.

The PEARLS care model offers continuity across the antenatal, intrapartum and early postnatal period up to maternal discharge, for over 300 women and their babies annually. PEARLS is a collaborative service between Westmead Hospital and Children's Hospital Westmead, providing NICU medical and nursing support.

The term 'continuity of care' refers to the service's care processes that are designed to achieve continuity, whereas, care provided to a woman by one or a small team of known midwives is termed 'continuity of carer' (McLachlan et al., 2012; NHS, 2016). Continuity of carer, or relationship continuity, refers to a therapeutic relationship between the service user and one or more health professionals over time. However, very little research has explored models of care that offer COC including a relationship with a known care provider for women with a high-risk pregnancy requiring care from multiple professionals, such as women who have received a diagnosis of congenital abnormality.

McCarthy describes the approach to documenting the patient journey as 'mapping a consecutive series of touch points between the patient and the service where the patient experience is actively shaped' and then illustrated visually (McCarthy et al., 2016). This helps to identify all the stages in the pathway process as well as routine investigations and consultations. While not all patient journey mapping captures service user perspectives, we considered in this study women's perspectives of their pregnancy, birth and early postnatal journey were essential to capture. From a person-centred view, COC is defined as 'the patient's experience of coordinated and smooth progression of care' over an appropriate period (O'Brien et al., 2021; Reid, Glazener, et al., 2002; Reid, Haggerty, & McKendry, 2002).

This multidisciplinary model of care provides an opportunity to map women's journeys from prenatal diagnosis of a congenital anomaly to their discharge from maternity services, to determine what is working well for women and families and improvements needed. A study exploring the practice advantages of providing midwifery COC for women with high-risk pregnancies, and the relationships midwives develop with the interdisciplinary team may increase our understanding of the care needs of these families. Our increased insight may reduce the potential for conflicting advice to families and facilitate the continuation of care provided by the primary midwife even when complications arise.

3 | AIM

The aim of this study was to (1) map the care pathway and service provision (patient journey) for pregnant women whose newborns require admission into the surgical NICU at or soon after birth from the perspective of women/parents and health professionals, (2) examine the nature of COC provided and the facilitators and barriers to woman and family-centred care.

4 | METHODS

4.1 | Research design

This was a mixed methods study design (Cresswell & Plano Clark, 2011), and was undertaken according to the criteria set out in Good Reporting of a Mixed Methods Study (GRAMMS) (O'Cathian et al., 2008, pp. 92–96, see Appendix S1). As quantitative and qualitative data were required to capture the complexities of the care journey and to map the processes and experiences from the perspective of mothers and health professionals a mixed method approach was selected (Guetterman et al., 2015). Priority was placed on qualitative aspects of design to capture the experiences and complexities of the care journey from the perspective of women and health professionals. The qualitative data were supplemented with record audit data that was required to map care processes. The audit data were quantified using descriptive statistics (Guetterman et al., 2015).

In a mixed methods design study, integration can occur at one or more points throughout the research process (Cresswell & Plano Clark, 2011, pp. 66–67; Fetters et al., 2013). Cresswell and Plano Clark (2011) describe integration at the method level as occurring through 'building', referring to the data collected in one phase informing the next phase of data collection. The quantitative data collected in the Phase 1 maternal record review, and the journey map were used to guide interviews of women who had experienced the journey to hear their stories and to describe care continuity and discontinuity. Finally, integration occurred at the interpretation level through our narrative interpretation of the combined quantitative and qualitative findings (Fetters et al., 2013). The information generated contributed to an understanding of COC for these women and families.

4.2 | Study setting, participants and recruitment

The study site was the PEARLS clinic located within the (MFM) service of a major referral Hospital for a range of specialty health areas. A mix of convenience and purposive sampling techniques was used. Convenience sampling was used to recruit women currently in PEARLS service and to recruit stakeholder health professionals. Purposive sampling was used to recruit key health professionals to interview. Our purpose was to interview those health professionals specifically identified by women as supporting them in some way throughout their journey. Study participants included 17 pregnant women who had received a prenatal diagnosis of congenital anomaly requiring the admission of their baby to a surgical NICU soon after birth. Twenty-two staff participated in the study. This number included the 15 key stakeholder health professionals recruited to attend the stakeholder workshop and 7 key health professionals recruited for interview. Health professionals who had a role in establishing the service were recruited for the stakeholder workshop and included COC midwives employed within PEARLS, staff employed within MFM and clinical nurse consultants and neonatal nurses from both Westmead NICU and Children's Hospital NICU teams. Seven key health professionals were identified in interviews with women as being extremely supportive and were therefore recruited for interview. They included the Children's Hospital cardiac team's clinical nurse consultant, obstetric medical team professionals, PEARLS midwife and Children's Hospital NICU nursing clinical coordinators. Each of these individuals had extensive experience in their specific areas.

Women were provided with information about the study by PEARLS midwives. If women expressed an interest to participate, they were invited to contact the researchers for further information and were provided with consent forms and a hard copy of study information, and signed the consent form if they agreed to participate following assurance their participation was voluntary and they could withdraw from the research at any time.

Similar information was provided to potential health professional participants and key stakeholder health professionals from the MFM and NICU using communication channels used within the service, specifically the regular monthly team meeting. If agreeing to participate, the external researcher (KP) provided potential participants with study information before obtaining consent. Ethics approval for this study was obtained from SCHN Human Ethics Research Committee (HREC) and the Western Sydney Local Health District Ethics committee. The HREC reference is available upon request.

4.3 | Data collection

Data collection methods are described in Table 1. Data were collected in two phases

1. Phase 1: Data collected in this phase were used to construct the women/family (patient) journey map. Data collection methods included a workshop with 15 stakeholder health professionals and 7 key health professionals and an audit of 20 randomly

TABLE 1 Description of data collection methods in chronological order.

Phase 1 construction of journey map

Stakeholder workshop: Two researchers (RR and JJ) facilitated a workshop with 15 stakeholder health professionals employed within PEARLS, maternal–foetal medicine (MFM), Westmead NICU and Children's Hospital NICU teams to develop a draft patient journey map for women who had received a prenatal diagnosis of congenital anomaly. Each phase of the women's journey was considered, routine consultations, procedures, processes and perceived facilitators/barriers to COC identified

Retrospective case file review: A retrospective case file review of 20 randomly chosen case records of women admitted to the PEARLS clinic over the previous year was completed. Maternity computer-based and hard copy documentation, from antenatal diagnosis of anomaly through to discharge from midwifery care were reviewed. Our objective was to trial the data collection tool for the prospective case file review and to use data collected to inform the patient journey map

Phase 2 perspectives of participants to inform, validate and give voice to the journey

Prospective case file review: Prior to the interviews with pregnant women who had received a prenatal diagnosis of congenital anomaly requiring the admission of their baby to a surgical NICU soon after birth, and with the study's 17 participants permission, we conducted a prospective case file review using the data collection tool trialled in the case file review undertaken in Phase 1. Records reviewed included participants maternity computer-based and hard copy documentation of their maternity care progress, from antenatal diagnosis of anomaly through to discharge from midwifery care. Data collected were expected to inform data collection in the participant women interviews and inform the developing patient journey map

Interviews with pregnant women: Seventeen pregnant women who had received a prenatal diagnosis of congenital anomaly requiring the admission of their baby to a surgical NICU soon after birth were recruited to the study. A research team member (RR) interviewed each woman face-to-face, exploring their perceptions their health services experience throughout their pregnancy, from antenatal diagnosis to the interview point. Interviews were scheduled at a time convenient for women, and were held within a private interview room in maternity services. The semi structured interviews lasted between 40 and 90 min, were digitally recorded and later transcribed. Data collected within these interviews were expected to inform the developing patient journey map and identify key health professionals to be subsequently interviewed

Interviews with key health professionals: Interviews with key health professionals (representatives from CHW cardiac team, obstetric team professionals, PEARLS midwife and CHW NICU CNC) were digitally recorded and then transcribed. Interviews were semi-structured, using open-ended questions held at a time and place convenient to them. Ascertain their perceptions of COC, around transition of care between teams and services. These health professionals were deemed able to provide additional information to inform the women's care pathway under their maternity health service role. Data collected were expected to inform the developing patient journey map

selected retrospective case records of women who had traversed the service in the previous 2 months.

- Phase 2: Data collection in Phase 2 aimed to explore the perspectives of participants to inform, validate and give voice to the woman/family (patient) journey map. Data were collected via face-to-face interviews and audits of the records of 17 women currently enrolled with the service. Only partners who accompanied women to the interview, were consented before the commencement of the interview. Three partners joined with mothers interviewed. Interviews were also undertaken with seven health professionals.

All interview data were transcribed verbatim and de-identified using codes. Audit data collected included demographic data, external referral notes, health history, investigations, procedures undertaken throughout pregnancy, clinic attendance, booked consultations and consultant notes and birth records. Audit data were also de-identified using codes. Data collection methods are detailed in [Table 1](#).

4.4 | Data analysis

As the focus of the study was on the presence/absence of continuity across the care continuum and how continuity was facilitated, our analysis was informed by concepts of continuity as advocated

by Reid, Haggerty and McKendry (2002). Commentators have defined the dimensions of continuity as 'managerial continuity', 'relational continuity' and 'informational continuity' (O'Brien et al., 2021; Reid, Glazener, et al., 2002; Reid, Haggerty, & McKendry, 2002). Managerial continuity refers to the coordination and provision of timely and complementary services within a service and is most vulnerable during care transitions. Relational continuity refers to an ongoing therapeutic relationship between a woman, her family and health providers that support present care by linking it to recorded knowledge of the family's past and future circumstances (O'Brien et al., 2021; Reid, Glazener, et al., 2002; Reid, Haggerty, & McKendry, 2002). Informational continuity refers to sharing information (current and historical) between healthcare providers to ensure clear communication, consistency and appropriateness of care for the individual's/family's condition (Reid, Glazener, et al., 2002; Reid, Haggerty, & McKendry, 2002).

The qualitative data collected from 17 pregnant women and 22 health professionals were analysed using Braun and Clarke's model of reflexive thematic analysis (Braun & Clark, 2013). Emphasis is placed on the constant revisiting of earlier stages in the analysis process to confirm and establish the adequacy of the analysis and continued refinement of themes (Braun & Clark, 2013, 2021). Complete immersion in the data was achieved through repeated readings of all transcripts. Initially, all ideas and patterns were allocated to as many codes as they fit. Coding was undertaken line by line, with data extracted by coding inductively (Braun &

TABLE 2 Maternal demographics.

Maternal age (years)	N=17	Marital status	N=17	Country of birth	N=17	Parity	N=17	Co-existing problems	N
<30	5	Married	14	Australia	12	G1P0	4	Congenital anomaly	2
31-35	6	Partner	2	America	2	G1P1	1	Asthma	1
36-41	6	Single	1	Indian	3	G2P1	4	Depression and/or anxiety	8
>41	0			Asian	1	G3P1	3	Hypertension	2
						G3P2	2	Thyroid disease	2
						G4P5	1	Anaemia	2
						<G5P1	2	Obesity/diabetes	2

Clark, 2013, 2021). Initially, 80 codes were identified and subsequently developed into concepts by grouping existing codes into 18 concept groups. The coding map was then examined to identify patterns and linkages across the concept groups. The process produced seven themes across the datasets. Coding and preliminary analysis were initially undertaken by RR and themes were confirmed by other researchers. NVivo (released in March 2020) was used to analyse all qualitative data.

Quantitative data collected from audits of the records of 17 women who were enrolled with the service were analysed using basic descriptive statistics.

4.5 | Ethical considerations

It is widely acknowledged that the pregnant woman is vulnerable and potentially disadvantaged regarding research, exacerbated in cases of high-risk pregnancy where women rely heavily on those caring for her and her unborn child. Therefore, in addition to the ethical principle of autonomy, non-maleficence, beneficence and justice, several additional issues required consideration by researchers undertaking this study.

1. The well-being and care of the woman who is pregnant and of her foetus always takes precedence over research considerations (NHMRC, 2007), addressed through the voluntary consent process. Study information, provided to women before consenting to study participation, included a description of the nature of the research. Women were also informed that they may experience some emotional discomfort while sharing their information during interviews. Confidentiality and anonymity were promised in the study information and achieved by ensuring data was initially collected appropriately. Interview data collection occurred in a designed space separate to clinical areas and was undertaken by experienced researchers. Data were de-identified at the collection and kept in files on a secure computer.
2. The possibility of providing access to counselling for the woman should be part of this discussion (NHMRC, 2007). The study information provide to women confirmed they would have access to free counselling services should this be required. Access to service was arranged before data collection.

3. Researchers should ask the woman, whether, in her decisions about the research, she wishes to involve others for whom the research may have implications (NHMRC, 2007). Although not offered specifically in the study information, this point was addressed by the inclusion in the interview process of partners who attended the interview with women when requested.

5 | RESULTS/FINDINGS

5.1 | Characteristics of participants

Table 2 describes the 17 women participants who were recruited in Phase 2. Participating women were between 19 and 38 weeks gestation when they participated and their mean age was 34 years, range 22–41 years. Most participants had been pregnant previously, four were pregnant for the first time. Twelve participants were born in Australia, and five born overseas (India, South America and Sri Lanka). As described in Table 2 participants had a variety of co-existing medical problems including maternal congenital anomaly, asthma, depression and anxiety, hypertension, thyroid disease, anaemia, obesity and diabetes.

Table 3 provides information related to antenatal diagnosis and referral details. Figure 1 provides a flow chart of identified aspects of each stage of the journey including data contributing to identification of themes within each stage. All participant data were de-identified at the point of data collection.

5.2 | Patient journey map

A draft map of the typical patient journey was developed during the workshop with 17 stakeholder health professionals. Each stage of the patient journey was later described and confirmed through Phase 2 maternal document review data and interview data from women and health professionals. The stages of the final confirmed patient journey map are presented below in chronological order. The identified aspects of each stage of the journey are included in Figure 1. The themes identified in each stage of the journey were developed from the analysis of interview data and reflected the women's experience of the journey and the factors that either

TABLE 3 Antenatal diagnosis and referral details.

Gestation first ultrasound	N= 17	Suspected diagnosis	N= 17	Diagnosis confirmation	N= 17	Referred by	N= 17	PEARLS admission	N= 17
Not recorded	1	Not recorded	3	Cardiologist	8	Cardiologist	6	Not recorded	1
<16weeks	6	Sonographer	4	Surgeon	2	Surgeon	4	<20weeks	3
16–25 weeks	8	Antenatal clinic	2	Obstetrician	1	Antenatal clinic	5	21–30weeks	11
26–35 weeks	2	General practitioner	5	MFM	6	External MFM	2	31–40weeks	3
		Obstetrician	3						

facilitated or hindered continuity at each stage of the journey. In each of the chronological stages of the patient journey facilitators of continuity are identified as examples of continuity while barriers to continuity are identified as examples of discontinuity at each stage. Data contributing to the themes identified at each stage of the journey along with examples of facilitators of continuity (continuity) and barriers of continuity (discontinuity) are included in Figure 2.

5.3 | The initial diagnosis: Informational and managerial discontinuity

Women's experiences of this phase of the journey were captured in two themes associated with receiving the diagnosis—'Going it alone' and 'There's something wrong but we don't know what the problem is'. Participants received a preliminary diagnosis at different times during their pregnancy dependent upon when signs of a problem developed. Commonly the preliminary diagnosis was established during routine antenatal ultrasound examinations, before admission to PEARLS.

Table 3 shows six participants in the prospective group received preliminary diagnosis after their first routine ultrasound before 16 weeks gestation, eight between 16 and 25 weeks gestation, and two between 26 and 35 weeks gestation before PEARLS care. In each case the news was unexpected, initially explained to participants by either a sonographer (4), antenatal clinic (2), general practitioner (5) or obstetrician (3), then referred to a consultant specialist for confirmation and discussion of prognosis, with delays for families varying from days to over a week. Interviews with women/parents confirmed this as very distressing (*informational discontinuity*).

5.3.1 | Going it alone

Women sometimes received the diagnosis alone, without a support person being present, because the woman had attended what she thought maybe a routine antenatal scan alone. Other women were given the diagnosis alone when called back by their family doctor. In neither scenario, were participants advised ahead of the appointment to attend with a support person, even when there was prior knowledge of the suspected diagnosis (*managerial discontinuity*).

'Going it alone' also refers to participants reporting feeling their partners could not support them, or feeling they must be strong for their partners. In several instances, after being given the diagnosis alone, participants were offered the opportunity to call a support person but believing themselves emotionally unable to make that phone call, opted not to contact their partner, they were ill equipped emotionally or cognitively to take in all the information being given to them at that time (*informational discontinuity*). Despite almost 50% of participants having a history of mental health issues, mothers revealed that their distress went unacknowledged. Despite her partner being with her earlier, this participant left her partner outside and was given the diagnosis alone.

I knew it was the heart because I've had a lot of echos done before. I was just thinking, why are they looking at the baby's heart so much? What's wrong? Then they told us to go out into the foyer until we were called back in. He's just sitting on Facebook so I just said to him, 'Just wait there. Don't worry about it'. I went in alone when called.

(Woman ANT)

While this participant was offered the opportunity to call someone, emotionally by that point it was too late, she was unable to call for help.

The sonographer's words were, 'Alarm bells are ringing. Now you wait here, I'll get the doctor'. She just left me there for fifteen minutes. When she came back, she said, 'Oh, you better call your husband'. I thought, 'Oh God'. There's no way I could call him at that moment! I had to look at the texture of the curtains to distract myself, so I could get through the next bit to focus on what they were saying.

(Woman AN H)

Parents viewed the experience of being given the preliminary diagnosis as a particularly significant event. When participants perceived the communication of the diagnosis as poor, it was usually due to the context in which the preliminary diagnosis was delivered. There is something wrong but we don't know what the problem is. This sub-theme refers to participants receiving a preliminary

DATA CONTRIBUTING TO FINDINGS	ASPECTS OF THE JOURNEY	STAGE ONE Confirmation of pregnancy and early care - referral for first ultrasound	STAGE TWO Diagnosis of congenital anomaly suspected - referral to consultant specialist, obstetrician or hospital	STAGE THREE referral to Maternal Fetal Medicine unit - for study participants this was Westmead Hospital and the PEARLS team	STAGE FOUR - Birth of baby	STAGE FIVE Mother is transferred to postnatal ward and transfer of baby to NICU.
Maternal interview supported by details from case record review.	Family participants interacting with health system at each stage	Mother with/without Father	Mother with/without Father	Mother with/without Father	Mother with/without Father	Mother with/without Father
Maternal interview supported by details from case record review.	Individual who coordinated care at each stage	Mother initially, Later GP or midwife	Mother with/without Father	Continuity of Care midwife	Birthing unit or Operating Theatre staff	Postnatal ward staff
Maternal interview supported by details from case record review.	Location of service provision at each stage	GP / Sonographers Office	Specialist Consultant offices, antenatal clinic at local hospital, GP office	High risk maternity clinic	Birthing unit or Operating Theatre	Postnatal ward
Maternal and staff interviews supported by details from case record review.	Specialist Consultations, Health Providers and other staff involved in care at each stage	GP, Sonographer, receptionists	Obstetrician, Specialist Consultants (Surgeon, Cardiologist, Neurologist etc), GP, Midwife	Bi-weekly visits to PEARLS clinic to see COC midwife. COC midwife may coordinate for family at same visit to see MFM team, Obstetrician, Cardiac CNC, Surgeon, Neonatologist dependant on needs.	Birthing unit maternity staff or Operating Theatre nursing staff, Obstetrician, Neonatologist, midwife	Postnatal ward midwives, Obstetrician or Obstetric registrar, Surgeon, Neonatologist, clerical staff
Case record review details confirmed in Maternal and staff interviews.	Investigations completed and Information created at each stage	Blood Screen, Ultrasound/ Patient notes, referrals,U/S record	Blood Screen, Ultrasound/ Patient notes, referrals,U/S record	COC midwife coordinates investigations and records - Blood Screen, Ultrasound, urinalysis, Care plans for birth and NICU transfer, Patient notes, referrals, pathology and imaging investigations, clinic card (record of visits - investigation/ results, examination record, weight and urinalysis results, midwifery record)	Dependant on mode of delivery; Vital signs, CTG, Blood gas, Blood screen/ Fluid balance, Drug sheet, Observation chart, Birthing suite or Operating suite notes, Ventilation chart, OT procedure sheet	Vital signs, Blood screen/ Fluid balance, Drug sheet, Observation chart, patient record notes, discharge paperwork, referral for postnatal check

FIGURE 1 Flow chart of identified aspects of each stage of the journey. Data which contributed to the identification of aspects of each stage are included. COC, continuity of care; NICU, neonatal intensive care unit.

diagnosis that was either incorrect or unable to be confirmed at that time, resulting in significant distress and uneasiness because they could not begin to plan or seek help and support without enough information.

At the twenty weeks scan, they said, 'There's something wrong with his heart... we don't know what it is'. So I was very calm, trying to take in all the information. They said, 'It could mean that the baby dies, or the baby has a disability. We have no idea, we can't tell you what the problem is. You won't find out for a while, we'll send you to Dr. C but you won't get in for several weeks'.

(Woman AN I)

While participants were distressed at being told that there was a problem, in most instances, it was the extent of the problem that was unknown which called the most distress. For others, it seemed the health professional did not have adequate knowledge of the problem (informational discontinuity). Several participants were given an incorrect initial diagnosis, inadvertently resulting in them in future questioning the advice provided and appearing to have lost trust in health services (relational discontinuity).

The family doctor phoned me the next day and said you need to come in. He wouldn't tell me anything over the phone. We went in and he said your baby doesn't have a brain, you need to have a curette because the baby is not going to survive past 10 weeks. He referred us to our local hospital. When they read the report they said, no, that's not the problem, this is the problem. We don't specialise in this so we will refer you to Westmead Hospital.

(Woman Ant E)

5.4 | Care prior to admission to the PEARLS team: Risk of discontinuity

This time point on the care continuum refers to the interval between participants receiving initial diagnosis, and attending an appointment with a specialist who could confirm the diagnosis and provide concrete information regarding the prognosis. Table 3 reports the range of specialist consultants families were referred to for confirmation of diagnosis; cardiologist (8), surgeon (2), obstetrician (1) or MFM specialists (6). A single theme captured at this point of the journey is 'Everyone should be in the know'.

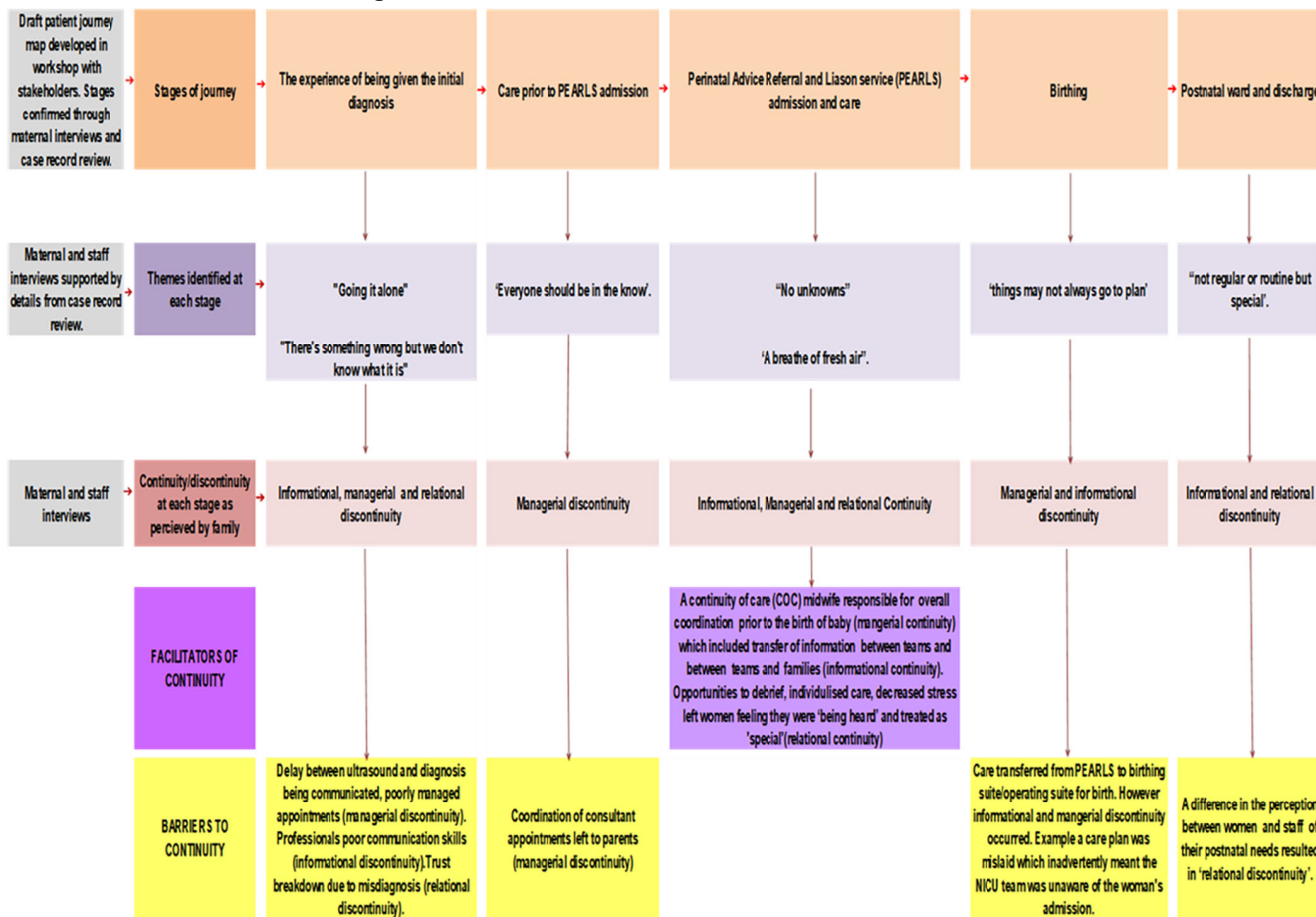


FIGURE 2 Flow chart of data contributing to the themes identified at each stage of the journey along with examples of continuity/discontinuity.

5.4.1 | Everyone should be in the know

This sub-theme refers to parents' perceptions of the difficulty they experienced accessing their care, specifically how poorly information is sometimes communicated between teams (informational discontinuity). As with the initial diagnosis, often there was a delay between ultrasound confirmation of an anomaly and discussion with specialist consultants (informational discontinuity). Although parents were provided with referrals they were usually expected to organise and coordinate their appointments. The informational discontinuity often experienced at this point of the care journey is captured in the following quote.

At our first appointment, 'Dr. X had said I'll send this report to you, and I'll send this to your doctor at the last Hospital' But ...he sent it somewhere else, or it didn't come through in time, I don't know! Anyway, when I went to theHospital, they didn't have the report. I waited two hours in the waiting room to hear the obstetrician say, 'There's nothing we can really talk about because I don't have the report that was supposed to be sent here'.

(Woman AN H)

The following woman described how she received the suspected diagnosis and waited for confirmation. Unfortunately, her partner was also due to travel for business which would keep him away for several weeks. The couple found the organisation of appointments extremely stressful (managerial discontinuity).

I stayed home from work so I could be on the phone, I was waiting by the phone all day but no call! My husband did know at that point that there was something wrong, but he didn't know what the problem was. He's an anxious person,... anyway, the doctor I finally got on to the next day said, 'I've got you in for next Monday. Can your husband push back his trip?'

(Woman ANT I)

Table 3 demonstrates that most participants were referred to PEARLS between 21 and 30weeks gestation (11). Referral often involved four to nine pre-admission visits to a variety of health professionals, local clinics and then metropolitan hospitals, which together with the delay between appointments increased parental stress significantly as described below.

When they read the report they said, this is the problem. We don't specialise in this so we will refer you to [local fetal medicine unit]. We had to wait another week to get an appointment down at their unit and had lots of scans. Also, we had the CVS testing, and genetic testing was done just to see what we were dealing with. Then from there, they referred us to Westmead for specialist treatment... so, a lot of waiting around for test results and stuff. They told us what it was but they couldn't really tell us how it's going to be fixed. They told us that there was a high chance of other abnormalities, so we had to deal with that for weeks. It was pretty stressful.

(Woman Ant 10)

care plans for birth, transfer and NICU admission, encompassing the predicted condition of the neonate at birth.

Once a month we'll have a surgical meeting where we go down to the CHW and the surgeons get together and talk about the antenatal and postnatal cases. So, it's still that communication and it's also where our obstetricians turn around and ask for their advice; what do you think? We saw this... what do you think of these pictures looking at the scan? So it's a real education session and exchange of information and we keep them updated on the pregnancy as well.

(PEARLS Midwife Interview 1)

5.5 | Admission to the PEARLS team: Relational and managerial continuity

Table 3 reports referrals to the PEARLS and MFM teams at Westmead Hospital were made by cardiologists, surgeons, staff working in the Westmead antenatal clinic and external MFM clinics. This point in the journey describes care provided to participants after admission to the PEARLS team, with an allocated PEARLS midwife.

The midwife was responsible for antenatal care in collaboration with an allocated MFM consultant and the overall coordination of investigations, introductions and appointments with other specialist care teams for the participant or her newborn (managerial continuity) and for information transfer both between the teams and between the teams and the family (informational continuity). In describing their role, midwives emphasised how important coordination of care was for women (managerial continuity). This is illustrated in the following quote.

So that's how we build our appointments. Just for continuity, trying to do it all in one day: So when I do appointments, Dr N might be scanning her [woman] this week on a Tuesday, so instead of coming to the Wednesday clinic I see her on Tuesday. They can't come twice in one week, they'd go insane. That's the same as the tours, if parents miss the tour but they are coming back to the cardiologist let's do it all at one time. Continuity is important.

(Stakeholder health professional workshop participants)

We noted midwives negotiate on behalf of participants with the health professionals in other teams to coordinate several appointments to occur in 1 day, aiming to decrease visit numbers participants made to the clinic and avoid long wait times (managerial continuity). In addition, the PEARLS midwives attend weekly cross-campus multidisciplinary team meetings (managerial continuity), documenting clinical

To facilitate a smooth transition across teams and appropriate NICU bed management, a copy of the register of newborns, regularly updated, is held in all relevant departments and has been refined further to an electronic version accessible across both campuses (informational continuity). Notification of impending birth occurs via group text paging to the multidisciplinary teams (NICUs, neonatology, management, midwifery) at four time points; when each woman is in early labour, just before birth, after birth and on transfer of the neonate to the surgical NICU (informational continuity). Two themes emerged capturing participants' experiences of the journey: 'No unknowns' and 'A breath of fresh air'.

5.5.1 | No unknowns

The theme 'No unknowns' refers to participants always knowing what to expect, with PEARLS midwife always able to access information on their behalf, an example of 'informational continuity'.

Woman: everything's been thought out and considered and planned. Even though it's been quick we've had doctor O who talked us through all the details. We knew exactly what we were dealing with- even though it's a little bit unknown.

Partner: I think it's just getting all the information we need, getting all the details, knowing what's happening next. No unknowns, which is nice.

Participants reported being appreciative of the efforts taken by midwives in care coordination (managerial continuity) predominantly because they were often overwhelmed by the situation they had found themselves in:

Well, the fact that [PEARLS midwife] has been organising the meetings and it's not like... 'Well, you go and organise the meeting'. Which, for us, ...I have no idea what I'm supposed to do. The fact that she has

organised everything and she has just been saying, 'It's this day and this time okay? This is where you have to go and talk'. That has been helpful for us, it has been easy.

(Couple Ant C)

5.5.2 | A breath of fresh air

Several participants provided comparisons of their care before their PEARLS admission, describing coordination (managerial continuity) as 'a breath of fresh air' making the experience less stressful and more personal as parents viewed themselves as 'special'. In the following excerpt we begin to see evidence of relational continuity.

The care here has been exceptionally good, compared to our previous hospital. It's just they're approachable, we can ask BB [PEARLS midwife] any questions, any number of times and she'll explain it and John (her partner) will probably ask the same questions again and she'll explain again. It's not just professional, 'here's the list now go away'. It's not that, she goes beyond that. Yeah, especially with a baby that's got a condition and we're worried. Coming from previous hospital to this hospital, I think, I'm more hopeful than I was.

(Woman Ant P)

PEARLS midwives worked as a liaison or coordinators to smooth a path through the system on behalf of participants, an important aspect of the role in any discipline (managerial continuity). This entails the midwife being approachable and flexible both in interactions with participants but also with professional peers. This flexibility facilitated 'relational continuity'.

Around coordination of services, I think has been exceptional. It's very rare to find someone who goes out of their way for someone... going that one extra mile. DD [PEARLS midwife] was always very available if I had any questions she made it clear that I was able to contact her any time with the mobile number in and out of hours. There was always a phone number to contact somebody else if there were any problems or any questions and, I was followed up regularly.

(Couple Ant I)

Participants perceived that they received preferential treatment, leading them to feel 'special' and 'very supported'. The PEARLS midwives provided participants with opportunities to talk and debrief, which left them feeling that they were 'being heard' (relational continuity).

They assist you with whatever problems that you have, they don't push you out and say sit down and wait. They do assist you if you have problems, when I came and I said to them I cannot feel my baby I am worried. They said why didn't you call? They don't make you wait.

(Couple Ant D)

The following participant had attended the regular hospital antenatal clinic until the diagnosis of congenital anomaly. She speaks about the contrast in care once under PEARLS care.

Woman: I also find DD [PEARLS midwife], wants to listen to me and answer my questions. Now I get to talk and she asks how I'm doing, there's a lot – of that emotional side. We connected. When I saw the doctors I was just another patient

I'm voicing my feelings and my concerns whereas before it was just, okay tick, tick we'll move your file along.

(Woman Ant F)

The PEARLS midwives' role in facilitating continuity by providing care coordination was recognised and appreciated by other health professionals (managerial continuity). Importantly, they perceived women cared for by PEARLS have better knowledge and understanding of their case and less anxiety.

I don't think we could have that continuity of the journey if I couldn't just ring up the PEARLS girls and say, okay - your patient's here now. They arrange everything for them. Which is good, they have it in their mind for each of their patients, it's like another check as well for me to make sure, okay, that's right you can have that scan done and we'll organise that.

(Medical Officer Interview 1)

Not showing up for appointments and not knowing how to make appointments, particularly if there's a language barrier, is such a common problem. In this group that's avoided as all appointments are made for them. There's much less anxiety as they seem far more connected to the system.

(Medical Officer Interview 2)

5.6 | Birth care: managerial and informational discontinuity.

This time point refers to the family's experiences around birth. A single theme was identified at this point 'things may not always go to plan'. The PEARLS midwife coordinates care of women up to

the birth. Once women go into labour, care is shared between the PEARLS team and birth unit health professionals, who follow the woman's birth plan. If the birth is planned as an elective caesarean section the PEARLS midwives provide all care. In an induction of labour, cervical ripening balloons are inserted by PEARLS midwives and intrapartum care is shared with birth unit midwives. A copy of the plan is also held by the woman. This preparation aims to ensure the birth goes as smoothly as possible, inclusive of the woman's requests.

5.6.1 | Things may not always go to plan

The excerpt below demonstrates that despite all efforts' *things may not always go to plan* and demonstrates 'managerial discontinuity', stimulating a needed change in the model of care to now encompass continuity of midwifery care at caesarean section.

I don't think the delivery team knew what was wrong with the baby...because I was to have a planned C-section. They prepped me, the obstetrics registrar introduced himself. They were about to wheel me into the theatre when my husband said something about our baby's heart condition. The mood changed and all of a sudden they stopped, the NICU team came down. The midwife asked me for my birth plan... they didn't seem to have it so we had to get my yellow card. Suddenly the consultant obstetrician came in to deliver our baby.

(Couple Ant I)

Immediately after the birth, depending on the neonate's level of acuity, participants may or may not be able to hold their newborn, other than while the cord is being clamped, before the neonate's transfer to the NICU. Depending on the transfer plan, the neonate was transferred to either the Westmead Hospital NICU or to the Children's Hospital Westmead usually accompanied by the father as a support person.

5.7 | Postnatal ward: Relational discontinuity

Following the birth, women's care was transferred to postnatal ward staff. The woman's partner or support person accompanied the team transferring the newborn to NICU care, and then returned to inform their partner of their newborn's condition. Once the woman was feeling well enough, her partner or a health professional help her travel to see the neonate.

PEARLS midwives provide midwifery at home service to women following discharge from the postnatal ward, either in their home if residing locally, but more often in the CHW accommodation provided for women travelling large distances from home, placing them in an ideal position to monitor women's postnatal

journeys. Midwifery at home care is provided by PEARLS midwives for up to 2 weeks, hence PEARLS is an expanded MAPS model of care.

Women were typically discharged home after several days while their babies remained in NICU. This short period between birthing their baby and discharge was problematic for the majority of mothers interviewed. Most couples had envisaged postnatal ward accommodation until their baby's condition was stable; however, in reality many were discharged while their babies were critically ill. The theme identified at this stage was 'not regular or routine, but special', as parents perceived their situation as different from others, requiring them to have special care. In contrast, however, postnatal ward health professionals saw their duty as providing routine postnatal care resulting in 'relational discontinuity'.

5.7.1 | Not regular or routine, but special

As their newborns are critically ill, women spend many hours over the first few days in the NICU. Although postnatal ward health professionals were aware of the neonate's admission to NICU, the parents' perceived their actions to be unsympathetic regarding their situation and did not seem to understand the absence of women.

I was expecting the four days promised so I planned to be there ...the day after delivery, I was told they needed the bed for sick mums. Under regular circumstances I probably would have been 'yep, of course I'm heading home' and it wouldn't have affected me as much. But at that point in time, it was just really insensitive. I was quite devastated, felt really alone and unsupported. I understand the logistics, but I also think my situation is special.

(Couple Ant J)

The participant in the next excerpt, while not asked to leave, was equally upset when paperwork prompting her to consider discharge was left on her bed. She has reflected on what she thought should happen.

They're not dealing with a regular mum who's just given birth, they're dealing with a mum who's separated from her baby, unsure of what's about to unfold so they need to be able to support her an extra night. I think it was, for health professionals, a case of this is just what we do so we're just going to keep on doing it. Rather than stepping back and going okay, so maybe something needs to be put in place - this is a regular thing, mums that are going to be separated from their babies.

(Couple Ant B)

6 | DISCUSSION

The study's aim was to map the pathway and service provision for pregnant women whose newborns require admission into the surgical NICU at, or soon after birth, examine the nature of COC provided, and facilitators and barriers to the woman and family-centred care (Figure 1). In addition, we hoped to identify opportunities and strategies for service redesign to improve continuity for women.

We found minimal evidence of coordination (managerial continuity) before COC by PEARLS, with services primarily focused on requirements of their services rather than the coordination across services. Haggerty et al. (2013) purport that managerial and informational continuity, which ensure patients know what to expect, are extremely important to the development of relational continuity. Knowing what to expect is seen as positive, providing security to patients (Haggerty et al., 2013). When patients experience discontinuity they experience uncertainty, insecurity or mistrust (Haggerty et al., 2013). This was evident in our study, particularly concerning women's experience of receiving the diagnosis of congenital anomaly. Managerial discontinuity was reflected in the lengthy time delays experienced by women between suspected and confirmed diagnoses, miscommunication of information within and between health services and professionals and absence of support when negotiating appointments with consultant physicians. This has previously been confirmed in the literature where women recounted their frustration at having to become quite assertive to coordinate their care among many different specialists (Haggerty et al., 2013).

In Australia, ultrasound technology has dramatically improved the detection rate of major congenital anomalies over the last decade (Molander et al., 2010; Tan et al., 2012). Most pregnant women view the ultrasound as non-threatening and do not consider the possibility of a foetal anomaly diagnosis (Thomas et al., 2017). Literature confirms women are often poorly informed about the purpose of ultrasound (Edvardsson et al., 2015). Certainly, this was the case in our study. Women in our study reported being given distressing information without adequate emotional preparation beforehand or support afterward. Literature supports that parents have vivid memories of the circumstances surrounding the diagnosis (Carlsson et al., 2017).

Parents confronted with the diagnosis of congenital anomaly in the antenatal period must make critical decisions based on information provided at diagnosis, often within very short time frames (Lotto et al., 2018). Decision-making hinges on being given accurate information in a way that can be understood clearly and the parents' interpretation, understanding, and retention of the information provided (Lotto et al., 2018). In a systematic review of prenatal counselling for the congenital anomaly, parents expressed a preference for counselling on all aspects of the anomaly as soon as possible and desired written visual and web-based information resources, and support group contacts to reduce parental anxiety (Marokakis et al., 2016). As has been reported in the literature (Karim et al., 2016), some women perceived health professionals were

unsure of the diagnosis or provided an incorrect diagnosis. While the extent of some defects may not be able to be clearly defined until later in the pregnancy (Karim et al., 2016), women presumed these inconsistencies were due to inadequate skills or knowledge of the professional or service. Irrespective of the cause, when a health professional appears unsure of a diagnosis or provides inaccurate diagnosis trust in the health professional deteriorates (relational discontinuity) (Carlsson et al., 2015).

While parents were satisfied overall with their care in the birth unit, they became more dissatisfied with any negative aspects of the birth if they encountered an unsupportive postnatal ward environment. Women were quite distressed at being seen as 'just another family' in the postnatal ward. This may be due to their perception of receiving exceptional 'individualised' care from their PEARLS midwife. The PEARLS midwives had gone to great lengths to ensure the women were well informed and cared for, whereas on the postnatal ward as the women were for much of the day in NICU, their needs were 'invisible' to health professionals. Women contrasted the discontinuity they experienced prior admission to PEARLS and in postnatal ward with the continuity they experienced with their PEARLS midwife. Women seemed most distressed by the postnatal care. While women's expectations for continuity may have been beyond what was possible within the maternity service model of care, it alternatively may reflect a staffing skill mix that varied considerably to the experienced midwives in the PEARLS team.

While the relationship developed with their PEARLS midwife was important to families, the level of expertise and coordination brought to the PEARLS midwives' liaison role was equally important. Women with high-risk pregnancies admitted into midwifery COC models had positive experiences and outcomes, which were consistent with findings about low-risk women (Fox et al., 2022). Offering continuity of carer enabled midwives to the optimal work environment to facilitate development of relationships with the women, their midwifery peers and medical colleagues.

6.1 | Study strengths and limitations

The diversity of data sources is a strength of the study offering the opportunity for integration of the study findings. The study is however limited because only 17 women participated in the interviews. We also did not interview health professionals other than those who participated in the workshop and interviews. We were not able to interview any health professionals who cared for women in other parts of their journey before diagnosis or at diagnosis and in the postnatal period.

6.2 | Suggestions for future research

Our findings suggest that research should investigate models of care that facilitate COC for high-risk women during the immediate

antenatal and postnatal period and determine the maternal and infant outcomes across models. Further research in this area should also seek the perspective of health professionals providing care to women diagnosed with congenital anomalies along each stage of their journey. Comparative studies of parents in programs like PEARLS that promote continuity and parents who are not part of such programs may illuminate differences in experience and perspective.

7 | CONCLUSION

The PEARLS model of care has enabled midwives to support high-risk pregnant women at the multitude of critical antenatal care touch points they experience. The movement from initial shock to acceptance for expectant parents faced with a prenatal diagnosis of congenital anomaly varies between families. In successfully mapping the pathway and service provision for pregnant women whose newborns require admission into the surgical NICU at or soon after birth, we were able to examine the nature of COC provided and the facilitators and barriers to the woman and family-centred care.

The provision of informational, managerial, and relational continuity provided in this model of care is essential to enable women and their families to cope and contribute to optimal outcomes. The results from this study highlight the advantages of allocating COC midwives to women with high-risk pregnancies, particularly psychosocial care, enabling women to benefit from care coordination and midwifery COC, within a collaborative practice with MFM consultants. Comorbidities in pregnancy, whether foetal or maternal, should always include midwifery care delivered through a continuity of carer model.

Enhancing care to improve women's experience of prenatal ultrasound, by skilled staff communicating with empathy the preliminary diagnosis, and the way forward, in line with national standards for communication in obstetric ultrasound (ASUM, 2022), is required from local through to tertiary level services. Steps need to be taken to improve women's experience of prenatal screening. At the time of diagnosis women must be adequately supported by skilled staff who can deliver bad news in a compassionate way and are able to clearly outline the way forward.

Parents' perception of the information communicated to them about their unborn infant's diagnosis affects parents' ability to cope with the diagnosis (Kratovil & Julion, 2017). In this study parents specifically stated the time delay between receiving a preliminary diagnosis, having it confirmed, and being referred on to tertiary maternity services was problematic. If a definitive diagnosis cannot be provided due to a developing structural anomaly, this should be explained to families with the potential worst scenario being explained. Empowering families by providing evidence-based resources can provide preliminary support, but effective multidisciplinary strategies to reduce waiting times to definitive diagnosis is needed to mitigate further trauma to parents.

Families in this study saw the role of the COC midwife as one of navigation. Essential characteristics of a liaison for high-risk pregnancy include the ability to individualise care, demonstrate empathy

and communicate effectively and to work well in an interprofessional team (Naughton et al., 2021) The liaison should be equipped to provide emotional support, arrange appointments with consultant healthcare professionals, interpret investigation results, align multiple appointments for single days, arrange medical interpreters, arrange travel support and schedule multidisciplinary case meetings, and should commence at preliminary diagnosis rather than wait until confirmation at the tertiary centre. Remodelling of care will therefore require a model which supports midwives coordinating the woman's care from preliminary diagnosis, confirmation of diagnosis and continuity of midwifery care through antenatal, birth, postpartum care and navigate discharge and ongoing care.

8 | RELEVANCE TO CLINICAL PRACTICE

Given the difficulties experienced by families in this study pre and post-admission to PEARLS, families need to be allocated a clinician immediately after the initial suspected diagnosis who then walk the journey with them through to discharge. In Australia, and other countries with vast geographical distances, service development is focusing on providing families with earlier contact and continuity with the tertiary centre teams through telehealth and other shared electronic imaging capabilities.

Models of care should be designed to provide a sensitive and flexible approach to care, with collaborative multidisciplinary care close to the woman's home, when possible. This work requires midwives who are sensitive to the needs of these families and able to communicate effectively in situations where families are likely to be extremely anxious. Fox et al. (2022) found that by demonstrating mutual respect for the expertise of others and clarifying each care provider's role and goals, positive experiences were facilitated. Enhanced communication is vital given the heightened complexity of maternity care with high-risk pregnancies. Staff may require additional education to improve communication, provide trauma informed care with shared decision making, and increase awareness of the emotional toll families carry with a baby in NICU.

AUTHOR CONTRIBUTIONS

Conception and design, or acquisition of data or analysis and interpretation of data; manuscript drafting and revising: KP, SH and VS. All authors have given final approval of the version to be published. Each author has participated sufficiently in the work to take public responsibility for appropriate portions of the content and agreed to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

FUNDING INFORMATION

Funding for the project was provided through an internal Western Sydney University research grant. There were no restrictions placed on authors regarding data collection, analysis or publication by the funding organisation.

CONFLICT OF INTEREST STATEMENT

No conflict of interest has been declared by the author(s).

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

ETHICS STATEMENT

Gained through The SCHN HREC and WSLHD Ethics committees (HREC Reference: HREC/16/SCHN/77).

ORCID

Kim M. Psaila  <https://orcid.org/0000-0002-7757-8615>

Virginia Schmied  <https://orcid.org/0000-0002-4066-1781>

REFERENCES

- AIHW. (2020). *Australia's mothers and babies 2018: In brief* (Perinatal statistics series no. 36. Cat. no. PER 108. Issue).
- Australasian Society for Ultrasound in Medicine (ASUM). (2022). Parent-centred communication in obstetric ultrasound. <https://www.asum.com.au/files/public/SoP/curver/Obs-Gynae/Parent-centred-communication-in-obstetric-ultrasound.pdf>
- Baldwin, A., Harvey, C., Willis, E., Ferguson, B., & Capper, T. (2019). Transitioning across professional boundaries in midwifery models of care: A literature review. *Women and Birth: Journal of the Australian College of Midwives*, 32(3), 195–203.
- Braun, V., & Clark, V. (2013). *Successful qualitative research: A practical guide for beginners*. Sage.
- Braun, V., & Clark, V. (2021). One size fits all? What counts as quality practice in (reflexive) thematic analysis? *Qualitative Research in Psychology*, 18(3), 328–352. <https://doi.org/10.1080/14780887.2020.1769238>
- Carlsson, T., Bergman, G., Marttala, M. U., Wadensten, B., & Mattsson, E. (2015). Information following a diagnosis of congenital heart defect: Experiences among parents to prenatally diagnosed children. *PLoS ONE*, 10, e0117995. <https://doi.org/10.1371/journal.pone.0117995>
- Carlsson, T., Starke, V., & Mattson, E. (2017). The emotional process from diagnosis to birth following a prenatal diagnosis of fetal anomaly: A qualitative study of messages in online discussion boards. *Midwifery*, 48, 53–59. <https://doi.org/10.1016/j.midw.2017.02.010>
- Cresswell, J., & Plano Clark, V. (2011). Choosing a mixed methods design. In J. Cresswell & V. Plano Clark (Eds.), *Designing and conducting mixed methods research* (2nd ed., p. 11). Sage.
- Dagklis, T., Papazisis, G., Tsakiridis, I., Chouliara, F., Mamopoulos, A., & Rousso, D. (2016). Prevalence of antenatal depression and associated factors among pregnant women hospitalized in high-risk pregnancy unit in Greece. *Social Psychiatry and Psychiatric Epidemiology*, 51, 1025–1031. <https://doi.org/10.1007/s00127-016-1230-7>
- de Wolff, M. G., Midtgaard, J., Johansen, M., Rom, A. L., Rosthoj, S., Tabor, A., & Hegaard, H. K. (2021). Effects of a midwife-coordinated maternity care intervention (ChroPreg) vs. standard care in pregnant women with chronic medical conditions: Results from a randomized controlled trial. *International Journal of Environmental Research and Public Health*, 18, 7875.
- Edvardsson, D., Mogren, I., Lalos, A., Persson, M., & Small, R. (2015). A routine tool with far-reaching influence: Australian midwives' views on the use of ultrasound during pregnancy. *BMC Pregnancy & Childbirth*, 15, 195. <https://doi.org/10.1186/s12884-015-0632-y>
- Fernandez Turienzo, C., Bick, D., Briley, A. L., Bollard, M., Coxon, K., Cross, P., Silverio, S. A., Singh, C., Seed, P. T., Tribe, R. M., Shennan, A. H., Sandall, J., & Vogel, J. (2020). Midwifery continuity of care versus standard maternity care for women at increased risk of preterm birth: A hybrid implementation-effectiveness, randomised controlled pilot trial in the UK. *PLoS Medicine*, 17(10), e1003350.
- Fetters, M. D., Curry, L., & Creswell, J. (2013). Achieving integration in mixed methods designs—Principles and practices. *Health Services Research*, 48(6), 2134–2156. <https://doi.org/10.1111/1475-6773.12117>
- Fox, D., Scarf, V., Turkmani, S., Rossiter, C., Coddington, R., Sheehy, A., Catling, C., Cummins, A., & Baird, K. (2022). Midwifery continuity of care for women with complex pregnancies in Australia: An integrative review. *Women and Birth: Journal of the Australian College of Midwives*. <https://doi.org/10.1016/j.wombi.2022.07.001>
- Guetterman, T. C., Fetter, M. D., & Cresswell, J. W. (2015). Integrating quantitative and qualitative results in health science mixed methods research through joint displays. *Annals of Family Medicine*, 13(6), 554–561. <https://doi.org/10.1370/afm.1865>
- Haggerty, J., Roberge, D., Freeman, G., & Beaulieu, C. (2013). Experienced continuity of care when patients see multiple clinicians: A qualitative metasummary. *Annals of Family Medicine*, 11(3), 262–271. <https://doi.org/10.1370/afm.1499>
- ICM. (2021) PS2021_ENMidwife-led Continuity of Care (MLCC) [Position Statement]. <https://internationalmidwives.org/our-work/policy-and-practice/icm-position-statements/>
- Karim, J., Roberts, N., Salomon, L., & Papageorghios, A. T. (2016). Systematic review of first trimester ultrasound screening in detecting fetal structural anomalies and factors affecting screening performance. *Ultrasound Obstetric Gynecology*, 50(4), 429–441. <https://doi.org/10.1002/uog.17246>
- Kratovil, A., & Julion, W. (2017). Health-care provider communication with expectant parents during a prenatal diagnosis: An integrative review. *Journal of Perinatology*, 37, 2–12. <https://doi.org/10.1038/jp.2016.123>
- Lotto, R., Smith, L. K., & Armstrong, D. (2018). Diagnosis of a severe congenital anomaly: A qualitative analysis of parental decision making and the implications for healthcare encounters. *Health Expectations*, 21(3), 678–684. <https://doi.org/10.1111/hex.12664>
- Marokakis, S., Kasparian, N. A., & Kennedy, S. E. (2016). Prenatal counselling for congenital anomalies: A systematic review. *Prenatal Diagnosis*, 36(7), 662–671. <https://doi.org/10.1002/pd.4836>
- McCarthy, S., O'Raghallaigh, P., Woodworth, S., Lin Lim, Y., Kenny, L. C., & Adam, F. (2016). An integrated patient journey mapping tool for embedding quality in healthcare service reform. *Journal of Decision Systems*, 25, 354–368. <https://doi.org/10.1080/12460125.2016.1187394>
- McLachlan, H., Forster, D., Davey, M., Farrell, T., Gold, L., Biro, M., Albers, L., Flood, M., Oats, J., & Waldenström, U. (2012). Effects of continuity of care by a primary midwife (caseload midwifery) on caesarean section rates in women of low obstetric risk: The COSMOS randomised controlled trial. *British Journal of Obstetrics and Gynaecology*, 119(12), 1483–1492. <https://doi.org/10.1111/j.1471-0528.2012.03446.x>
- Meaney, S., Lutomski, J. E., O'Connor, L., O'Donoghue, K., & Greene, R. A. (2016). Women's experience of maternal morbidity: A qualitative analysis. *BMC Pregnancy and Childbirth*, 16(184), 1–6. <https://doi.org/10.1186/s12884-016-0974-0>
- Molander, E., Alehagen, S., & Bertero, C. M. (2010). Routine ultrasound examination during pregnancy: A world of possibilities. *Midwifery*, 26(1), 18–26.
- Munch, S., McCoyd, J. L., Curran, L., & Harmon, C. (2019). Medically high-risk pregnancy: Women's perceptions of their relationships with health care providers. *Social Work in Health Care*, 59(1), 20–45.

- National Health and Medical Research Council. (2007). Women who are pregnant and the human foetus. In National Health and Medical Research Council, Australian Research Council, Australian Vice-Chancellors' Committee (Eds.), *The National statement on ethical conduct in human research* (pp. 51–53). [cited 2022 Nov 10]. Available from: http://www.nhmrc.gov.au/_files_nhmrc/publications/attachments/e72.pdf
- Naughton, S. L., Harvey, C., & Baldwin, A. (2021). Providing woman-centred care in complex pregnancy situations. *Midwifery*, *102*, 103060.
- NHS. (2016). *National Maternity Review (Better births: Improving outcomes of maternity services in England)*. The National Maternity Review. england.maternityreview@nhs.net, www.england.nhs.uk/ourwork/futurenhs/mat-review
- O'Brien, D., Butler, M. M., & Casey, M. (2021). The importance of nurturing trusting relationships to embed shared decision-making during pregnancy and childbirth. *Midwifery*, *98*, 102987–102987. <https://doi.org/10.1016/j.midw.2021.102987>
- O'Cathian, A., Murphy, E., & Nicholl, J. (2008). The quality of mixed method studies in health services research. *The Journal of Health Services Research & Policy*, *13*(2), 92–98. <https://doi.org/10.1258/jhsrp.2007.007074>
- Olley, H., Psaila, K., Fowler, C., Kruske, S., & Homer, C. (2017). Being the bridge and the beacon: A qualitative study of the characteristics and functions of the liaison role in child and family health services in Australia. *Journal of Clinical Nursing*, *26*(1–2), 91–102.
- Psaila, K., Schmied, V., Fowler, C., & Kruske, S. (2013). Discontinuities between maternity and child and family health services: Health professional's perceptions. *BMC Health Services Research*, *14*(4), 1–12. <https://doi.org/10.1186/1472-6963-14-4>
- Reid, M., Glazener, C., Murray, J., & Taylor, G. S. (2002). A two-centred pragmatic randomised controlled trial of two interventions of postnatal support. *British Journal of Obstetrics and Gynaecology*, *109*, 1164–1170.
- Reid, R. J., Haggerty, J., & McKendry, R. (2002). *Defusing the confusion: Concepts and measures of continuity of healthcare*. Canadian Health Services Research Foundation.
- Sandall, J., Soltani, H., Gates, S., Shennan, A., & Devane, D. (2016). Midwife-led continuity models versus other models of care for childbearing women. *Cochrane Library*, *4*, CD004667.
- Tan, W. S., Guaran, R., & Challis, D. (2012). Advances in maternal fetal medicine practice. *Journal of Paediatrics and Child Health*, *48*, 955–962. <https://doi.org/10.1111/j.1440-1754.2012.02596.x>
- Thomas, G. M., Roberts, J., & Griffiths, F. E. (2017). Ultrasound as a technology of reassurance? How pregnant women and health care professionals articulate ultrasound reassurance and its limitations. *Sociology of Health and Illness*, *39*(6), 893–907. <https://doi.org/10.1111/1467-9566.12554>

SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

How to cite this article: Psaila, K. M., Schmied, V., & Heath, S. (2023). Exploring continuity of care for women with prenatal diagnosis of congenital anomaly: A mixed method study. *Journal of Clinical Nursing*, *00*, 1–15. <https://doi.org/10.1111/jocn.16777>



Threat and adaptation: The maternal lived experience of continuing pregnancy after receiving a prenatal diagnosis of agenesis of the corpus callosum

Pieta Shakes^{a,*}, Andrew Cashin^a, John Hurley^b

^a Faculty of Health, Southern Cross University, PO Box 157, Lismore, NSW, 2480, Australia

^b Faculty of Health, Southern Cross University, Hogbin Drive, Coffs Harbour, NSW, 2450, Australia

ARTICLE INFO

Handling Editor: Medical Sociology Office

Keywords:

Agenesis of corpus callosum
Prenatal diagnosis
Ultrasonography
Prenatal
Hermeneutic phenomenology
Corpus callosum disorder
Pregnancy
Lived experience
Prenatal care

ABSTRACT

One stated objective of prenatal screening and diagnosis is the preparation for delivering a baby with medical needs or disability, however, psychosocial outcomes of parents who received a prenatal diagnosis suggest that this objective is not yet realised. Preparation may be complicated by diagnostic and prognostic uncertainty. A prenatal diagnosis that includes significant uncertainty due to the heterogeneous presentations, classifications, causes and outcomes is agenesis of the corpus callosum. As a neuroanatomical anomaly identified in the second or third trimesters, the diagnosis is likely to cause distress for expectant mothers, yet there is limited guidance for holistic support. To begin to address the paucity of research, this hermeneutic phenomenological study sought to explore, and provide a telling of the maternal experience of continuing pregnancy after a prenatal diagnosis of agenesis of the corpus callosum. Through interviews and a series of online, asynchronous and facilitated focus groups, lived experiences during pregnancy from the time of diagnosis to birth were explored with 26 mothers who participated in this international study. Themes were constructed through reflexive thematic analysis to describe the experience of the lived phenomenon. The first theme, *Under Threat*, included subthemes of *The Threat to the Life of the Baby* and *Threatened Image of the Expected Family*. The second theme, *Day to Day Toward Adaptation*, included subthemes of *Holding it Together and Falling Apart*, and *More Than Information, Searching for Meaning, Hope and Control*. To realise the commonly stated objective of prenatal diagnosis, to support maternal preparation, healthcare professionals require awareness of the profound, yet individual experience of prenatal diagnosis to adequately respond and support mothers through their continued pregnancies. Healthcare services should be designed to flexibly respond in a woman- and family-centred manner to reduce the threat and support maternal adaptation after a prenatal diagnosis.

1. Introduction

Two commonly stated objectives of prenatal diagnosis are to inform reproductive decisions and support preparation for birthing a baby with a known anomaly (Michie, 2020). If psychosocial outcomes are a marker of preparation, parents who continued pregnancy after receiving a prenatal diagnosis should show improved psychosocial outcomes compared to parents who received a postnatal diagnosis, reflective of this preparation. However, comparison studies have identified similar levels of state anxiety (Fontoura et al., 2018; Werner et al., 2019) and medical traumatic stress (Bevilacqua et al., 2021) in the postnatal period. Parents who received a prenatal diagnosis of congenital heart

disease also reported lower levels of life satisfaction and sense of coherence than parents who received a postnatal diagnosis (Bratt et al., 2019).

This potential discordance with one of the two objectives of antenatal screening is significant as a prenatal diagnosis can trigger antenatal distress. Between 35% and 66% of mothers who received a prenatal diagnosis experienced clinically relevant acute stress or met full diagnostic criteria for post-traumatic stress disorder (PTSD) (Aguar and Abrahão, 2022; Fonseca et al., 2011; Rychik et al., 2013). Antenatal mental health conditions influence postnatal outcomes (Eastwood et al., 2017; Grigoriadis et al., 2019) and may harbour further consequences for a developing fetus with a known anomaly (Kasparian, 2019).

* Corresponding author.

E-mail addresses: p.shakes.10@student.scu.edu.au (P. Shakes), andrew.cashin@scu.edu.au (A. Cashin), John.hurley@scu.edu.au (J. Hurley).

<https://doi.org/10.1016/j.socscimed.2023.116391>

Received 17 July 2023; Received in revised form 11 October 2023; Accepted 31 October 2023

Available online 8 November 2023

0277-9536/© 2023 The Authors. Published by Elsevier Ltd. This is an open access article under the CC BY license (<http://creativecommons.org/licenses/by/4.0/>).

Maternal stress after prenatal diagnosis was flagged as a potential iatrogenic factor that influenced the earlier spontaneous birth and poorer postoperative outcomes of prenatally-diagnosed infants with transposition of the great arteries (Namachivayam et al., 2023).

Psychosocial outcomes and the pregnancy experience after a prenatal diagnosis warrant investigation. Research that explored experiences of mothers described shock at the unexpected news of a fetal anomaly, followed by a period of crisis, before adjustment and goal remodelling (Carlsson et al., 2017; Côté-Arsenault and Denney-Koelsch, 2016). Prognostic ambiguity, however, stalled progression toward adjustment (Côté-Arsenault and Denney-Koelsch, 2016) and delayed parental trajectories of connection to the fetus (McKechnie et al., 2015a). Prognostic ambiguity and diagnostic changes were associated with increased distress after a prenatal diagnosis (Oftedal et al., 2022) and uncertainty or contradictory information, increased parental feelings of being overwhelmed and emotionally exhausted (Marokakis et al., 2017). High uncertainty within a fetal anomaly diagnosis may hinder adaptation and maternal and fetal outcomes. A specific prenatal diagnosis associated with high uncertainty is agenesis of the corpus callosum (ACC).

ACC is a neuroanatomical anomaly characterised by the absence or atypical development of the corpus callosum, the largest interhemispheric pathway in the brain, which integrates cognitive and sensory information between the left and right hemispheres (Raybaud, 2010). Individuals born with an anomaly of the corpus callosum demonstrate highly variable neurodevelopmental outcomes, including typical development, developmental delays, mild to profound intellectual disability, or even early death (Shakes et al., 2020). Outcomes are attributed to additional neuroanatomical or extracranial anomalies and variable causes, including over 200 different syndromes (Smith et al., 2023). An atypical corpus callosum can present as the primary anomaly alongside secondary neuroanatomical changes, often termed *isolated ACC* (Shakes et al., 2020). These secondary features, described by Raybaud (2010), include an absent cavum septum pellucidum, poorly defined cingulate gyrus, lateral ventricles in a colpocephalic shape, a raised third ventricle, a rounded hippocampus and differences to the other interhemispheric commissures. Secondary anomalies have been interpreted as being included and excluded from the category of isolated ACC by different researchers, which increases prognostic ambiguity, as isolated and non-isolated ACC are associated with different outcomes (Shakes et al., 2020).

The high heterogeneity in presentation, interpretation, cause and outcomes of ACC increases the complexity of this diagnosis, which is commonly diagnosed after 18 weeks gestation and as late as 40 weeks (Smith et al., 2023). While there are recommendations for diagnosis and prognostication, there is a deficit of guidance to support parents through the high uncertainty of a prenatal diagnosis of ACC. Support for mothers who continue their pregnancies should be grounded by an exploration of the maternal experience of continuing pregnancy after a prenatal diagnosis.

2. Methods

2.1. Aim

This study aimed to provide a telling of the phenomenon of the maternal lived experience of pregnancy after receiving a prenatal diagnosis of ACC. This telling offers healthcare professionals insight into the complex experience, which may provide a foundation for empathy, understanding and systemic changes to improve the experience and outcomes of mothers. Patient experience should not be viewed as simply the interactions with healthcare professionals, but as multidimensional and multifaceted complex human experiences (Oben, 2020). Therefore, this study sought to explore the holistic experience of continuing pregnancy after a prenatal diagnosis of ACC.

2.2. Design

Hermeneutic phenomenology was the methodology for this study, informed by readings of Heidegger (1927/1962) and Gadamer (1975/2013) and selected due to the focus on uncovering and making manifest a phenomenon of experience (Moran, 2000). Three distinct features distinguish hermeneutic phenomenology from other qualitative methodologies: the interpretive focus on experiences-as-lived, involvement of the researcher and the dynamic and thoughtful reflecting and writing in analysis (Bynum and Varpio, 2018). Within Heidegger's adoption of hermeneutic phenomenology as methodology, he returned to primordial, pre-reflective accounts of his phenomenon of interest, and moved past a descriptive report of findings (Carr, 1999). Through a circular process of interpretation grounded by understanding and with a central tenant of temporality, Heidegger showed meaning as bound within historicity and facticity (Heidegger, 1927/1962). Gadamer progressed Heidegger's phenomenological methodology through his investigation that demonstrated the universality of hermeneutics for all understanding, in which fore-understanding and openness to new possibilities leads to new understandings through a "fusion of horizons" (Gadamer, 1975/2013, 305). Hermeneutic phenomenology framed how experience could be accessed and interpreted to disclose an experienced phenomenon which is temporally situated and open for reinterpretation. A benefit of this approach is that the telling is based on concrete, *as-lived*, accounts of experience rather than participants' reflections and perceptions, which continue to shift and change with time. The framing is demonstrated in the study design schematic (Fig. 1).

2.3. Ethical approval

This research study was approved by the Southern Cross University Human Research Ethics Committee, approval number: ECN-19-200.

2.4. Participant selection

Purposive sampling allowed recruitment of mothers with lived experience of the shared phenomenon of continuing pregnancy after a prenatal diagnosis of ACC. Inclusion criteria required confirmation that mothers (1) were the biological mother of a child prenatally diagnosed with ACC and who was between 6 months and 4 years at study enrolment; (2) spoke English as their first language; (3) lived in Australia, New Zealand, the United States of America, the United Kingdom or Canada; (4) had access to participate through online videoconferencing or focus groups; (5) were over 18 years and (5) provided consent. Child age boundaries reduced potential burden for pregnant or new mothers and to support recall. International sampling with health services and providers who were likely to be informed by similar English-based medical literature was selected to recruit an appropriately sized sample. While ACC may be one of the more common neuroanatomical anomalies identified during pregnancy, it remains a rare condition (Smith et al., 2023). As a study that included dialogic intersection (Vandermause and Fleming, 2011), conversational co-elicitation and exploration of experience between the researcher and participants, shared fluency in English was required to reduce additional translation-based complexities. Administrators of formal and informal ACC support organisations (Supplementary File 1) were asked to share the study flyer which directed interested mothers to email the lead researcher. There was no payment or reimbursement for participation.

2.5. Data collection and analysis

While there are variety of methods for data collection and analysis in hermeneutic phenomenology, the methodology guides the interpretative process of data collection and analysis, which commence together (Vandermause and Fleming, 2011). Two stages in this study were delineated by the method of interaction with participants; Stage 1

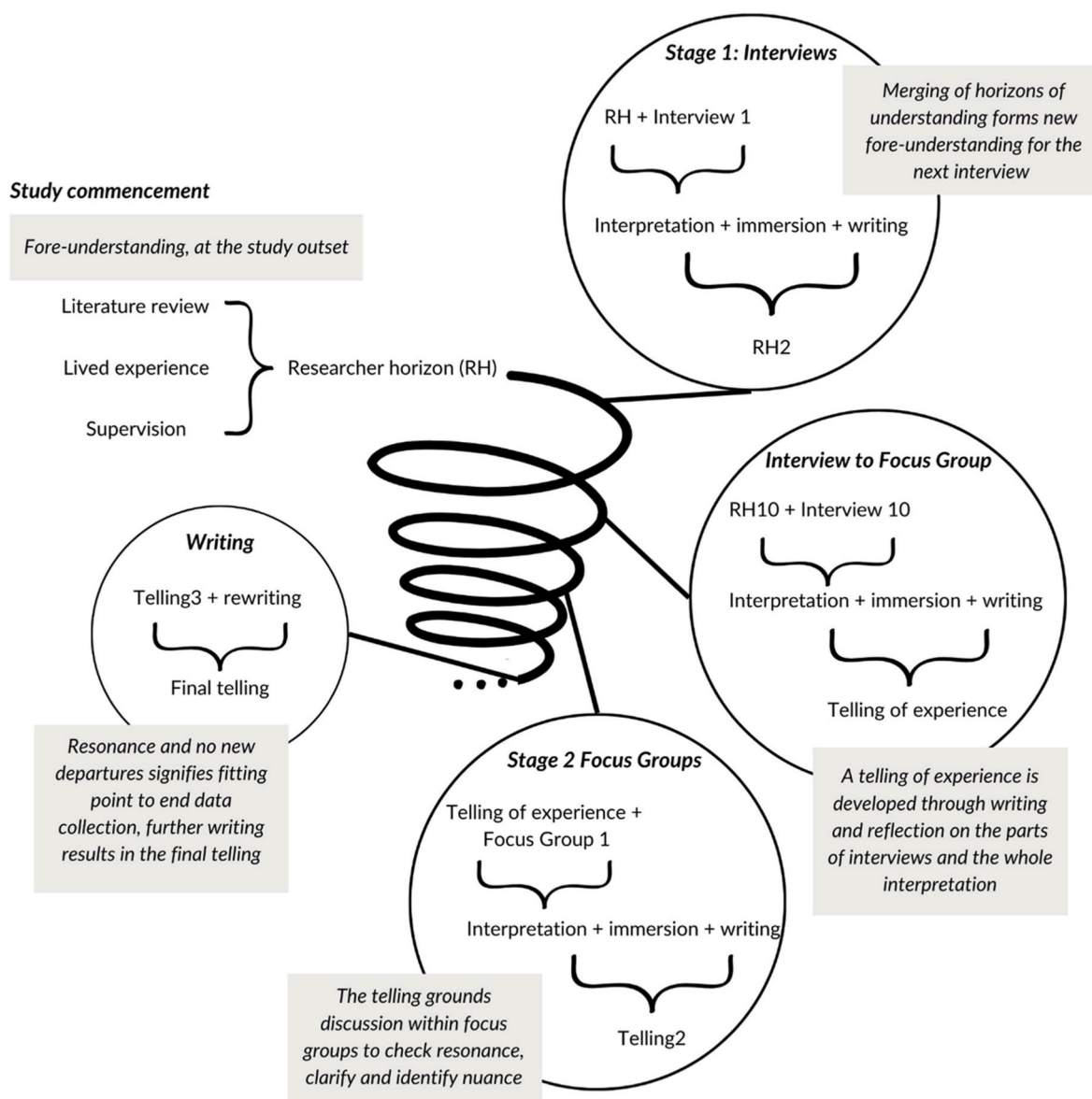


Fig. 1. Research design schematic.

involved one-on-one semi-structured interviews via videoconferencing and Stage 2 involved online, asynchronous and facilitated focus groups. Participants self-selected their involvement and were eligible to participate in both stages.

Interviews were semi-structured to guide the recall of experiences from the point of diagnosis through to birth and yield moments as-lived rather than participants' reflections. Transcription of recorded interviews was completed through Otter.ai (n.d.) and read in full alongside listening to the recording to begin immersion in the data and correct transcription errors. Transcripts were uploaded to NVivo (QSR International Pty Ltd, 2018) for further reading, inductive coding, chunking and re-splitting codes, mind mapping, writing and rewriting to disclose patterns and generate initial themes (Braun and Clark, 2021). Coding and condensing of like-codes followed each interview, and then shaped the horizon of understanding taken to the following interview, allowing the researcher to explore complexity. Interviews continued until diminished returns were noted, this point should not be confused with saturation which is incongruent with the epistemological assumptions of hermeneutic phenomenology and reflexive thematic analysis, as meaning is temporal, generated through interpretation and open for reinterpretation (Braun and Clark, 2021).

Online focus groups with up to eight mothers were engaged to confirm resonance from Stage 1 interpretations to increase study trustworthiness. Focus groups also allowed discussion to elucidate additional experiences for identifying nuance. Mothers participated in the 5-day, asynchronous discussion under usernames, Participant 1–8, allocated by the FocusGroupIt (n.d.) platform. Data were extracted via PDF and uploaded into NVivo (QSR International Pty Ltd, 2018) for coding and analysis. Confirmation of resonance and diminished returns signified a suitable end of Stage 2. Final analysis and interpretation occurred during the re-writing of the findings, as theme development was an ongoing process and reflected the approach described by Trainor and Bundon (2020). Reporting of this study was influenced by the Consolidated Criteria for Reporting Qualitative Research (Tong et al., 2007), although context-dependant information was prioritised in this report, and the checklist is comprehensively addressed in Supplementary File 2. Participant quotes do not include an identifier to mitigate the heightened potential of triangulation and reidentification of participants within research with families affected by rare conditions (Joseph, 2023).

Positionality of the researcher is also significant for framing the analytic process and theme development (Braun and Clark, 2021). This research was completed by a doctoral candidate who continued

pregnancy after a prenatal diagnosis of ACC eight years before the study’s conceptualisation. Her role within formal and informal peer support organisations surfaced additional considerations for recruitment, data collection and reflexive analysis, described in Supplementary File 2.

3. Findings

In total, 26 mothers who continued pregnancy after receiving a prenatal diagnosis of ACC participated in this study (Table 1).

Stage 1 comprised interviews with 10 mothers between December 2019 and April 2020. Interview recording commenced after introductions and consent, and averaged 71 min. Stage 2 comprised three online, asynchronous and facilitated focus groups between June 2020 and March 2022 (Table 2). Seven mothers who completed interviews chose to participate in the focus groups.

Table 1
Participant demographic information.

Demographic	N	%
Total participants	26	100
Country		
Australia	4	15.4
Canada	1	3.8
United Kingdom	3	11.5
United States of America	18	69.2
Mother’s age (years)		
18–22 years	1	3.8
23 - 27	2	7.7
28 - 32	5	19.2
33 - 37	7	26.9
38+	4	15.4
Not stated	7	26.9
Mother’s highest completed level of education		
High school	1	3.8
Vocational award	4	15.4
Bachelor degree	9	34.6
Postgraduate degree	1	3.8
Master degree	6	23.1
Not stated	5	19.2
Mother’s occupation		
Health professional	6	23.1
Educator	4	15.4
Stay at home mother	4	15.4
Business owner	3	11.5
Student	2	7.7
Other professional	2	7.7
Office worker	1	3.8
Not stated	4	15.4
Child’s prenatal diagnosis^a		
ACC	2	7.7
Complete ACC	4	15.4
Isolated complete ACC	5	19.2
DCC and ventriculomegaly ^b	10	38.5
ACC plus ^c	3	11.5
Uncertain specific diagnosis	1	3.8
Not stated	1	3.8
Child’s age at enrolment		
6 months to <1 year	3	11.5
1 - <2 years	10	38.5
2 - <3 years	7	26.9
3 - <4 years	6	23.1
Gestation of prenatal diagnosis		
18–22 weeks	12	46.1
23–27 weeks	6	23.1
28–32 weeks	4	15.4
>33 weeks	2	7.7
Not stated	2	7.7

ACC: Agenesis of the corpus callosum.

^a Prenatal diagnosis as reported and grouped.

^b Inclusive of ventriculomegaly, colpocephaly, enlarged ventricles and hydrocephalus expressed as an additional diagnosis to ACC prenatally.

^c Plus inclusive of a genetic variant or additional anomalies.

Table 2

Focus group participant numbers, words retrieved and interactivity within the group.

Focus Group	1	2	3
Mothers sent link (n)	8	8	10
Participants joined (n)	7	8	9
Participants withdrew after joining (n)	0	0	1
Words exported (n)	15,363	31,079	30,531
Posts to group (n)	135	156	134
Posts per participant, average (range)	12 (3–16)	15 (9–18)	14 (3–20)

Aus: Australia, UK: United Kingdom, US: United States of America, Can: Canada.

Two distinct themes of the experience of continuing pregnancy after a prenatal diagnosis of ACC were identified (Fig. 2). The first theme, *Under Threat*, encapsulated the immediate and ongoing threat presented with the prenatal diagnosis of ACC. The two subthemes, *The Threat to the Baby’s Life* and *Threatened Image of the Expected Family* encompassed the two dimensions of being under threat. The second theme, *Day by Day Toward Adaptation*, was constructed through mothers’ continual assimilation and accommodation as they moved through pregnancy. Actions undertaken by mothers disclosed two subthemes, *Holding it Together and Falling Apart* and *More than Information, Searching for Meaning, Hope and Control*. These two major themes and their two subthemes revealed the profound experience of continuing pregnancy after a prenatal diagnosis of ACC and were illuminated within the first stage of the study, although the specific labelling and categorisation was arrived at through the ongoing analysis and identification of nuance. The final focus group confirmed resonance with the interpretation and did not yield further points of departure.

3.1. Under threat

The expected pregnancy includes a series of normalised milestones and ends with the birth of a healthy baby. The prenatal diagnosis of ACC exposed possibilities that threatened the norm and as such, profoundly altered the experience of pregnancy. The complexities of ACC meant that possibilities remained, suspended in uncertainty, without closure. Accounts of lived responses, actions and emotions illuminated how the baby’s life and the image of the expected family were under threat.

3.1.1. The threat to the Baby’s life

The threat to the baby’s life was experienced in both a concrete and abstract manner. The indication of something unexpected through professionals’ demeanour or words during an ultrasound or follow up appointment surfaced the immediate threat to the baby’s life.

I can’t remember how long afterwards but the more experienced sonographer turned to us and said, ‘I’m really sorry but your baby is missing part of its brain. We’ll give you some time, we need to take you up to the ward’. They then both left the room. I asked my husband to repeat what she had said, he did. I remember discussing what that meant, was the pregnancy over? Was I being sent to the ward to terminate the pregnancy?

The threat to the baby’s life was often immediate, “So, I’m like, well, is the baby going to die?”, or triggered by the option of termination. This threat was evidenced by a commonly shared “shutting down”, a dissociative response disclosed through physiological responses, “I went completely numb”. The response included changes to sensory, cognitive and motor functions. “I couldn’t stand up; I was in complete shock. I couldn’t even get the words out of my mouth.” Dissociation was further identified by the impaired ability to take in information, “I feel like I blacked out at everything she said from that point on”. Mothers recounted inability to recall parts of the conversation, as exemplified by the mother who asked her own mother important information about the diagnosis after attending the appointment together. “When I was told, my mum was in the room with me, and I just shut off completely. I

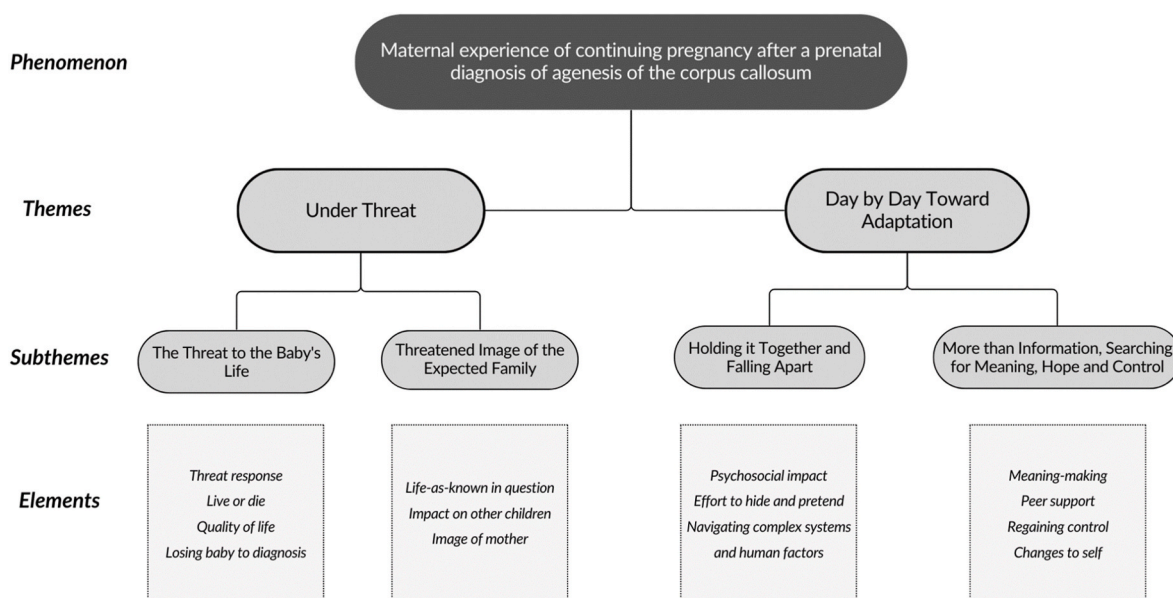


Fig. 2. Thematic map of the maternal experience of continuing pregnancy after a prenatal diagnosis of agenesis of the corpus callosum.

totally forgot what they even said, and my mum had to tell me later. It was like I blacked out.”

While many mothers experienced disassociation, a few mothers specifically denied a dissociative response to the news. “I don’t feel like I shut down that first day, because I wanted to soak up every bit of information to be able to google it later, but I did feel overwhelmed with it all.” Despite not shutting down, threat remained, highlighted through being overwhelmed and responses that included fear and worries. “I felt so vulnerable and scared and worried.” The baby’s quality of life was also under threat. “My biggest fear was, like, will they know love? Will they be able to know that I love them, will they be able to smile and stuff?” The threat to the baby’s life remained for many mothers, illuminated through their avoidance of buying baby clothes, preparing the nursery, or having a baby shower. “I chose to postpone the baby shower my home-school co-op friends wanted to throw until after [the baby] was here so that we would know more about him and know if he was going to make it.”

For a while, I didn’t want to name my baby, because the name I picked out I really loved, and I didn’t want to ‘waste’ the name on a baby that wouldn’t live. This is a thought I have only expressed once and never directly to anyone I am close with, not even my partner. I definitely have a lot of guilt associated with that, too.

The threat to the baby’s life was also experienced in an abstract manner, where the diagnosis and the uncertainty overshadowed the life and personhood of the baby. “I felt in some ways it was like a ticking time bomb I was carrying as we just were never sure what the future would look like once he arrived.” Actions of mothers sought to reclaim babyhood from diagnosis further disclosed the threat to baby’s life and flattened identity of the baby. “We did find out the gender of this baby as we felt she had become this terrible unknown that we were fearful of. We needed to humanise the whole experience.” However, the possibility of disability was consuming for some, overshadowing the baby’s individuality, who instead became a child with special needs. “I thought about that a lot during my pregnancy, it’s what made me the most depressed, thinking I was going to have a child with special needs”.

Appointments often perpetuated the threat to the baby’s life. “So many conversations with doctors were so cold and clinical. We needed to remember that we were talking about a tiny baby”. A “doom and gloom” focus or language that obscured the baby further suggested that professionals also reflected this threat. “Some specialists were horrific

and referred to our son as ‘the pregnancy’, urging us to terminate, this was at 35 weeks now.” The prenatal diagnosis triggered protocols that differed from the norm, as shared by a mother who received an unexpected phone call to ask her intent to continue or terminate her pregnancy; when her own mother took the phone in response to her distress and inability to talk, the caller stated, “she has a special needs baby, its protocol, we call and do that.” However, professionals also reduced this threat to the baby’s life and personhood, by supporting mothers to reclaim the baby from uncertainty.

She saw me being upset and started to ask me about my son, what his name would be, what kind of family he was coming into. She never mentioned his diagnosis but just talked about his future as if nothing on the paper mattered. It really helped me in the moment remember at the end of the day I was bring home a baby, not a diagnosis on a screen. She told from what she could see on the screen he was going to bring us joy, and laughter, and love just like how our other children did. She said she knew he would be loved and that’s all a baby really needs. I needed to hear those words and I’m very thankful that she was the last doctor I saw before I gave birth.

3.1.2. Threatened image of the expected family

The exposed possibilities meant that the expected family was under threat. “I was terrified that we had just had our last day together as a ‘normal’ [sic] family and after perhaps our lives would never look like this again.” With the suspended uncertainty, the threat to the family wasn’t easily contained. “That day we called our parents and took turns crying and wrapping our heads around a new parenting path we hadn’t planned for”. Mothers described fears of needing to “sacrifice everything”, that lives, as-known, were over and that the family may no longer be able to do things that are enjoyable or meaningful to them.

Mothers felt responsible for potentially hurting their family by choosing to have another baby that could have high medical or support needs. “Did my greed for another child cause this? Was I being punished for not being happy with what I had?” Feeling responsible for the diagnosis appeared intertwined with fears of not being “a good enough” mother. The image of the good mother in the expected family was threatened by the self-blame and guilt of causing the anomaly, “I broke my baby”. For one mother, the weight of this fear of causing her baby to suffer led to suicidal ideation. Within this fear of not being “enough” of a mother were concerns about coping with their baby’s potential needs or

their inability to continue to mother other children. “Would my other child’s needs be put on the backburner?” The threat to the image of mother within the expected family extended beyond responsibilities and tasks, the prenatal diagnosis of ACC thrust a mother’s love into question.

I was so afraid. I was like, what if the baby is like one of the babies who look weird and then I don’t love it because it doesn’t look cute. What kind of mum doesn’t love their baby because it’s not cute? Like, that’s horrible.

The mothers described the way their thoughts challenged some of their long-standing moral principles tied to reproduction and expected families. “I am also very pro-choice but for myself I always knew I could never have an abortion which made me feel all the more guilty when I considered it while pregnant with my daughter”. The image of the selfless or expected mother was under threat by both thoughts of continuing and ending pregnancies, evident through focus group discussions where mothers shared similar experiences despite divergent long-standing beliefs about termination. “I’m actually very pro-life and I felt the exact same way as you said! I felt so guilty having thoughts like ‘would it, my, life be easier without this baby?’ I felt terrible!” For some, the weight of decision-making about termination prompted mothers to consider how a miscarriage could be protective of the family, “that it would be okay if my baby didn’t pull through; both to save him from a life of possible endless suffering and pain, and to save our family from possibly having to see that”. However, this thought also simultaneously threatened the image of the mother.

It felt unnatural to all the sudden have these conflicting feelings; I love you more than anything, but I hate you right now. You’re so wanted and loved but it’s okay if you miscarry. I mean what parent thinks that? And how do you get over the guilt of those feelings after your child is born?

The expected family’s image was also threatened by the potential of becoming a different type of family than expected. “Were we about to become special needs parents?” These different types of families included the possible shift to a “one-income family” so a parent could remain home as carer, which also thrust into question the affordability of living in their house or current residential area.

3.2. Day by day toward adaptation

The mothers’ experiences of the days from diagnosis to birth varied greatly due to differences in healthcare systems, workplaces and roles and their own positionality in adapting to their changed pregnancy. As a time-limited and embodied experience, pregnancy demanded coping and self-regulatory processes as mothers moved through the days responding to the threat of the prenatal diagnosis of ACC. Mothers cycled through holding it together and falling apart, while they searched for more than information and yearned for meaning, hope and control.

3.2.1. Holding it together and falling apart

Accommodating the unexpected diagnosis was emotionally laborious, although the duration of accommodation processes varied. Mothers engaged a range coping processes to “hold it together” to be present in appointments and tests, workplaces, and even at home. Proactive coping thresholds were often exceeded resulting in periods of falling apart, until the mother was able to re-establish some sense of equilibrium. The cyclical nature was highlighted through changes in thoughts and mood. “I would swing from trying to be so hopeful in 1 s and then falling apart other days”. Expressions of breaking down often occurred outside clinics, in bathrooms or the car, or at home, when they could no longer restrain themselves. “In the lift, a lady who had dementia or something akin to that said, in such a sweet, childlike voice, ‘oh dear, you have such sad eyes whatever is wrong?’ I just broke”.

Anxiety and dread within appointments were the resurfacing of the threat. “I felt the same feelings of worry and anxiety and when we were

waiting to be called for the ultrasound in our private waiting room I started to cry. The feelings just resurfaced every time.” For many, this ongoing threat reminder demanded an accommodative coping response so they could physically be present and engage at the appointment. “I mostly internalised everything and kept the anxiety hidden at appointments”. Difficult thoughts, emotions and somatic symptoms were not limited to appointments, though, disclosed through accounts of waking with nightmares, changes in appetite, loss of interest in usual activities, migraines, “severe panic attacks” and “ruminating” worries in addition to mood changes that were evident to others.

I have two very close friends who I think at the time we’re like walking on eggshells around me and trying really hard not to, like [upset me], because I think they could tell that there was something up, but I wasn’t in a place to talk about it yet.

Mothers engaged strategies to manage the emotional labour and somatic symptoms of stress, such as setting an alarm as a reminder to eat, journalling, walking, meditation or seeking mental health support. “I had to work actively each day to keep myself calm and not over-stimulated.” When asked, these strategies were often an attempt to hold it together specifically “for the health of the baby”. Accommodative coping also surfaced through the avoidance of triggers. For some mothers, this meant stopping talking to their baby to “shut down the connection”, avoiding the nursery, “the nursery was a no-go area” and isolating themselves, “I was closed off from everybody”. This avoidance sometimes meant mothers avoided both informal and formal supports. “My closest friends didn’t know about it.”

I was also already seeing a therapist when I found out about her anomaly but stopped going soon after because the therapist herself was pregnant and just about as far along as I was. I couldn’t bear to go anymore!

When avoidance was impossible, mothers attempted to hide their circumstances and hold it together, recalling they would “pretend to be happy and pretend to smile”. “When I went to work a couple days after the ultrasound, everyone was asking, ‘are you so excited?’ I had a really hard time putting on a happy face”. Pretending and hiding was a strategy to avoid exhausting questions, the “look of shock and horror” from others and insensitive comments. Hiding was also an attempt to protect the baby from the threat, as explored earlier, where the diagnosis may overshadow the baby. “I felt like our baby would never be looked at as a normal baby to whoever knows her diagnosis.” Hiding and pretending surfaced as experiences within workplaces, the public and appointments with professionals. The few mothers who were pregnant during COVID19, specifically disclosed the protective nature of staying home due to the reduced visibility and need to “concoct a fake smile if a stranger approached me about my growing bump”. Hiding was complex, as it sometimes meant hiding frustrations and challenges but also hiding hope and attempts to adapt.

I was ‘warned’ by more than one provider not to take too much to heart what I saw on Facebook, and that really just infuriated me because they were 100% accepting of some study from 30 years ago listing the disabilities of kids with ACC as one of their diagnoses. They had zero idea of what a child with isolated [ACC], as ours is, could be or do. Towards the end, I stopped bringing it up or discussing the baby’s future with those providers that I knew had the doom scenario in mind.

There were also mothers who sought and received support of friends and family. “My mother-in-law was brilliant, so positive, challenging us to embrace this.” Although mothers were sometimes forced to hold it together to manage questions or responses from family and friends, “but it added so much extra stress for me.” While worries and questions from others were challenging to manage, friends who had this “idealistic mindset that a miracle would happen” or who commended the mothers on their strength surfaced difficult emotions for the mothers. “This

[notions of strength] really made me mad and made me feel belittled”.

Mothers who encountered systems-based challenges had additional demands upon them to hold it together while they navigated complexities, such as the need to ring around services to find shorter wait times for tests, travel long distances, wait in clinic rooms “surrounded by all these women with their perfect pregnancies” and actively managing “the nightmare that was trying to get the MRI records transferred!” While mothers had typically accommodated the loss of their initial hopes for a certain type of birth, significant distress was caused by late and unexpected decisions to admit their new babies to the neonatal intensive care unit (NICU) for monitoring when there was no clear clinical need. Mothers who had expected a NICU admission due to a complex presentation of ACC felt prepared, although described feeling detached from their baby, “I felt like my baby boy hadn’t belonged to me either”. Yet, the abrupt and unplanned separation resulted in additional emotional labour while mothers tried to hold it together to challenge the decision to separate them from their new baby. The advocacy of a private midwife allowed one mother to remain with her baby, while another was threatened with child protective services when she was seeking understanding of the clinical need and indicators when her baby would return to her from NICU, thus resulting in significant additional distress to hold.

3.2.2. *More than information, Searching for Meaning, hope and control*

Mothers described their search for information as “obsessive”. Information was sought to reduce the vastness of possibilities presented within the incomplete or inconsistent information offered by healthcare professionals. “There was so much conflicting information at my doctors’ visits. No one was able to get me a definite answer on anything”. Initial communication about ACC was often with healthcare professionals who expressed limited experience with ACC and commonly focused on the “worst case scenario”, “like there was no sense that it could actually be ok”. Mothers who later met with specialists with expertise in ACC recounted receiving more balanced or favourable prognostic information. “All of a sudden, it was a completely different scenario ... she said, ‘I see this, I deal with this [I am] comfortable telling you that he has a very good chance of being fine’”.

The search was for more than just information, it was about making sense of what the possibilities meant for the baby and family, to reach a near enough understanding to contain the threat and ground preparation. “I needed to gather as much information as I could, as a means to processing what was happening and begin to set myself up for what the eventual outcome would be.” This meaning-making was typically the task of the mother or couple with limited professional support, “there was no counselling”, with exception of two mothers who engaged in therapeutic counselling. Meaning-making was illuminated through accounts of accommodating possibilities within the expected family life, at times reigniting hope and reducing the impact of the threat.

After the diagnosis, my husband and I talked about all the worst-case scenarios. We talked about our son possibly never learning to walk or to talk, could we handle that? What would life then look like for us? We are avid travellers and we talked about how we would possibly no longer be able to travel the world with our child like we dreamt of. Of course, we felt sad, zooming out on what our life could look like. My husband said something that touched me, he said, ‘our son will have his own joys and we will encourage them, maybe his thing is birds. So, then we would make it a goal to travel places based on seeking out birds for our son to marvel at. We would still keep true to ourselves, all the while catering to him as well’.

In searching of meaning, mothers found their way to ACC organisations or online support groups. Many mothers spoke of the benefits of these groups, specifically the access to real-life stories about what it means to live with ACC. “It was hard for me to accept what my child’s life would look like until I found the Facebook groups”. The groups showed the diversity of lived experiences and outcomes associated with

ACC and offered the chance to “talk to other parents and hear their stories and experiences, both scary and shockingly normal”. There was a difference in the way mothers discussed the “larger groups” and the “babies’ groups”, with the former described as “the most challenging” due to the confrontation of stories that included outcomes they had not yet accommodated. Although “seeing people ‘living that life’ in the larger groups made things seem more doable.” While there was visibility of the spectrum of outcomes, access to real-life stories reduced some of the uncertainty by showing “that if things were going poorly as doctors predicted that our kids are still lovable and perfect just the way they are”. Lived experience further grounded the possibilities in a way that statistics alone couldn’t. “These were real people with real children with real experiences and challenges. Doctors could not offer anything tangible or concrete for us”.

The birth year groups provided access to “woman and men whose babies were in the similar stages of development”, which not only included “information to know what questions to ask, what tests to inquire about and how to set up early intervention” but access to others with a shared experience, where mothers could be open and honest about what was happening. “I would talk to just people in the group about it because I didn’t want to [talk to] my mum and like, I didn’t want to talk to anyone else.”

The search for information, meaning and hope was also an effort to regain control when so much seemed out of control. The uncontrolled possibilities meant mothers set to unpack their prior assumptions of parenting to accommodate the uncertain outcomes and focus on what they could control as mothers to support their child. “It really forced me to re-evaluate what it meant to provide healthier support to our child”.

I felt like I wasn’t in control of the ACC or the outcome of how my daughters ACC would affect her, so reading for hours and scouring support groups for happy stories and anecdotes was within my control and it consumed me.

4. Discussion

This study disclosed that the maternal experience continuing pregnancy after a prenatal diagnosis of ACC is an experience of being under threat and moving day by day toward adaptation. The diagnosis exposed possibilities that threatened the baby’s life and image of the expected family. Adapting to the threat while moving through pregnancy after the prenatal diagnosis demanded cognitive and emotional coping responses, both proactive and defensive to allow mothers hold it together while they searched for information, meaning, hope and control.

Threat surfaced within some, but limited, prior studies that have explored experiences and psychosocial outcomes after a prenatal diagnosis. Threats to the baby’s health were connected with a loss of the self-value as a mother within an analysis of interviews with 16 mothers who received a prenatal diagnosis of cleft lip and/or palate in Taiwan (Hsieh et al., 2013). Maternal appraisals of threat or loss, rather than challenge, were identified within a cross-sectional study that used several psychosocial measurement scales with 40 mothers who continued their pregnancies after a prenatal diagnosis in the UK (Horsch et al., 2013). The authors also found that primary threat appraisal was positively correlated with anxiety and depression. While threat surfaced in these studies and suggested a connection to psychosocial outcomes, this dimension of experience may be potentially concealed in other literature by a focus on the shock of an unexpected diagnosis. The experience of prenatal diagnosis as a threat may be specific to certain diagnoses such as ACC or those with high uncertainty. Although noting the high incidence of PTSD among mothers who have received a prenatal diagnosis (Aguar and Abrahão, 2022; Fonseca et al., 2011; Rychik et al., 2013), further research exploring threat in prenatal diagnosis and interventions to reduce threat appears warranted.

The acknowledgement of threat within the experience of receiving a prenatal diagnosis and the defence cascade in response to threat may

offer healthcare professionals a useful framework for understanding and the vast range of emotional responses from parents and the support they require. The automated and hard-wired defence to threat includes the physiological, sensorial and cognitive states, arousal, fight-or-flight, freeze and immobility within which a range of emotions and behaviours can be activated, including denial, anger, panic and dissociation (Kozłowska et al., 2015). These states may have implications for clinical care, for example, the physiological responses in the defence cascade may mean mothers struggle to hear and process news at the time of receiving a prenatal diagnosis. Many mothers in this study described lived experiences that suggested a dissociative response leading to confusion, inability to process news and ask questions or recall things discussed. Disassociation and lowering of consciousness is a normal neurobiological function when under threat (Paulsen and Lanius, 2014). Consideration of dissociation may be critical for informing support at the interpersonal and systems level. Acknowledgement of the potential for threat and peritraumatic reactions within prenatal diagnosis could underpin healthcare service design that supports clinicians to adapt communication and provide timely continuity within follow-up to accommodate disassociation. Trauma-informed prenatal care could be inclusive of the assessment of psychosocial needs, psychoeducation, streamlined referral to psychosocial supports and active follow-up as standard, rather than waiting for overt signs of distress considering the different physiological reactions of the threat response and the surfaced experience of holding it together for appointments.

Prenatal diagnosis is a unique practice in healthcare, in the sense that a diagnosis is received prior to birth and before a baby becomes known by their individual identity and personhood. A living person is understood not by their biological elements but by the interaction and qualities of the biological system of a human (Greve and Strobl, 2004). Prenatal diagnosis places significant focus on a diagnosis and biological elements before a baby's individual identity can manifest beyond imagined futures, which may perpetuate the threat of the loss of the baby to diagnosis. A diagnosis focus, often coupled with a focus on deficiency, may hinder the hopeful possibilities that are typically afforded to a baby without a diagnosis or disability (Johnson, 2023). Hope, after a prenatal diagnosis, was previously identified as a common and potentially important component of the maternal experience, which led to recommendations that healthcare professionals ensure they provide factual information about the diagnosis while also leaving room for positive emotions such as hope and optimism, as opposed to presenting a deterministic view which can undermine coping (Fonseca et al., 2011). This study disclosed that hope need not be just about typical development but can present through the accommodation of possibilities within the expected family life and through the reclamation of baby from diagnosis. High uncertainty within prenatal diagnosis may interrupt hope and attachment, influencing the environment, opportunities, and expectations a baby is born into. An interruption to bonding was not universal in this study but was common and remained a significant challenge for mothers to process years after their experience. These considerations highlight the critical need for further research and investment in supporting maternal adaptation to reduce potential harm, particularly in the climate of expanding prenatal testing.

This study disclosed mothers' many proactive and defensive actions as they moved through their pregnancy toward adaptation and that there was no single way to experience pregnancy after the prenatal diagnosis. Findings that some mothers enjoyed further ultrasounds and celebrated typical pregnancy milestones, while these experiences heightened distress for other mothers could be further considered in relation to the dual-process framework by Brandtstädter and Rothmund (2002). Assimilation-accommodation responses to divergences between desired and factual circumstances may ground individual adaptive processes. Assimilation comprises the maintenance of expectations and actions to incorporate circumstances into projections of the desired life course, reflected in mothers' accounts of self-care to protect their baby's development and within aspirations that incorporated space

for disability. Assimilation responses may also include denying the threat and challenging possibilities after a prenatal diagnosis, while these cannot be accommodated. Accommodation signifies processes that are not internally originated, which are cognitively and emotionally demanding due to the changing of internal structures, the modification of expectations and the creation of new schemas (Hanfstingl et al., 2022). Accommodation is a defence against an anxiety-arousing threat (Hayes et al., 2015). Accommodative responses were evidenced through the reorganisation of hopes, reevaluation of threatened future images, and defensive coping reactions. As the assimilation-accommodation framework incorporates individual and social influences within coping and adaptation, viewing the experience of prenatal diagnosis with consideration of the framework may ground research into supporting maternal adaptation after a prenatal diagnosis.

This study identified that access to supportive teams and streamlined services appeared to reduce the maternal experience of being overwhelmed. Patient journey mapping of an Australian high-risk midwifery model for women offered insight into the opportunity to improve the experience of prenatal diagnosis through holistic continuity of care, yet elicited challenges of discontinuity for parents before and after the specialist service (Psaila et al., 2023). A broad perspective of the complex human experience through prenatal diagnosis and into parenting, opposed to a narrow focus on prenatal diagnosis as an antenatal medical event could guide health policy that seeks to reduce systems gaps and improve psychosocial outcomes. Where continuity of care can't be achieved, a relational intervention such as the 15-Minute Interview may optimise communication to address the individual needs of the mother and family (McKechnie et al., 2015b).

5. Limitations

A phenomenological study does not intend to produce generalisable or representative findings and present an objective truth of a phenomenon. The findings of this study are specific to the participants' experiences and must be read with this limitation in mind. While eligibility criteria were broader, mothers who volunteered to participate were those who had engaged with ACC support groups before or after birth, spoke English as a first language, were in heteronormative parenting relationships during their prenatal diagnosis, had commonly completed education post-high-school and a high proportion were health professionals. Data collection and analysis was undertaken during COVID-19.

6. Conclusion

This hermeneutic phenomenological study provided a telling of the maternal experience of continuing pregnancy after receiving a prenatal diagnosis of ACC. The news of the suspected neuroanatomical anomaly was experienced as a threat to the life of the baby and the image of the expected family. Mothers engaged in active and defensive cognitive and emotional processes to hold it together while they searched for meaning, hope and control to accommodate their changed pregnancies and expectations. To realise the commonly stated objective of prenatal diagnosis, to support maternal preparation, healthcare professionals require awareness of the profound, yet individual experience of prenatal diagnosis to adequately respond and support mothers through their continued pregnancies. Healthcare services should be designed to flexibly respond in a woman- and family-centred manner to reduce the threat and support maternal adaptation after a prenatal diagnosis.

Data statement

To protect the anonymity of the mothers and in line with the ethics approval, data is not available for sharing, due to detailed experiences which may allow triangulation and reidentification.

Funding statement

This research was supported by an Australian Government Research Training Program (RTP) Scholarship.

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.

Data availability

The data that has been used is confidential.

Acknowledgements

We would like to thank the ACC support groups for the assistance with recruitment and all the parents who expressed interest, with an extra special thank you to the mothers who generously shared their experiences.

Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.socscimed.2023.116391>.

References

- Aguiar, M.A.V., Abrahão, A.R., 2022. Post-traumatic stress disorder and congenital anomalies in prenatal care. *Rev. Bras. Saúde Materno Infant.* 22, 517–525.
- Bevilacqua, F., Morini, F., Ragni, B., Braguglia, A., Gentile, S., Zaccara, A., et al., 2021. Pediatric medical traumatic stress (PMTS) in parents of newborns with a congenital anomaly requiring surgery at birth. *J. Pediatr. Surg.* 56, 471–475.
- Brandstädter, J., Rothermund, K., 2002. The life-course dynamics of goal pursuit and goal adjustment: a two-process framework. *Dev. Rev.* 22, 117–150.
- Bratt, E.-L., Järholm, S., Ekman-Joelsson, B.-M., Johannsmeyer, A., Carlsson, S.-Å., Mattsson, L.-Å., et al., 2019. Parental reactions, distress, and sense of coherence after prenatal versus postnatal diagnosis of complex congenital heart disease. *Cardiol. Young* 29, 1328–1334.
- Braun, V., Clark, V., 2021. *Thematic Analysis*. SAGE Publications Ptd, London: UK.
- Bynum, W., Varpio, L., 2018. When I say ... hermeneutic phenomenology. *Med. Educ.* 52, 252–253.
- Carlsson, T., Starke, V., Mattsson, E., 2017. The emotional process from diagnosis to birth following a prenatal diagnosis of fetal anomaly: a qualitative study of messages in online discussion boards. *Midwifery* 48, 53–59.
- Carr, D., 1999. *The Paradox of Subjectivity: the Self in the Transcendental Tradition*. Oxford University Press, New York.
- Côté-Arsenault, D., Denney-Koelsch, E., 2016. "Have no regrets:" Parents' experiences and developmental tasks in pregnancy with a lethal fetal diagnosis. *Soc. Sci. Med.* 154, 100–109.
- Eastwood, J., Ogbo, F.A., Hendry, A., Noble, J., Page, A., Early Years Research, G., 2017. The impact of antenatal depression on perinatal outcomes in Australian women. *PLoS One* 12, e0169907.
- FocusGroupIt. (n.d.). *Focus groups made easy*. (p. Online focus group platform).
- Fonseca, A., Nazaré, B., Canavaro, M.C., 2011. Patterns of parental emotional reactions after a pre- or postnatal diagnosis of a congenital anomaly. *J. Reprod. Infant Psychol.* 29, 320–333.
- Fontoura, F.C., Cardoso, M., Rodrigues, S.E., Almeida, P.C., Carvalho, L.B., 2018. Anxiety of mothers of newborns with congenital malformations in the pre- and postnatal periods. *Rev. Lat. Am. Enfermagem.* 26, e3080.
- Gadamer, H.-G., 1975/2013. *Truth and Method*. Bloomsbury Academic, London, United Kingdom.
- Greve, W., Strobl, R., 2004. Social and individual coping with threats: outlines of an interdisciplinary approach. *Rev. Gen. Psychol.* 8, 194–207.
- Grigoriadis, S., Graves, L., Peer, M., Mamisashvili, L., Tomlinson, G., Vigod, S.N., et al., 2019. A systematic review and meta-analysis of the effects of antenatal anxiety on postpartum outcomes. *Arch. Womens Ment. Health* 22, 543–556.
- Hanfstingl, B., Arzenšek, A., Apschner, J., Göllly, K.I., 2022. Assimilation and accommodation. *Eur. Psychol.* 27, 320–337.
- Hayes, J., Schimmel, J., Williams, T.J., Howard, A.L., Webber, D., Faucher, E.H., 2015. Worldview accommodation: selectively modifying committed beliefs provides defense against worldview threat. *Self Ident.* 14, 521–548.
- Heidegger, M., 1927/1962. *Being and Time*. Blackwell Publishers, Oxford, United Kingdom.
- Horsch, A., Brooks, C., Fletcher, H., 2013. Maternal coping, appraisals and adjustment following diagnosis of fetal anomaly. *Prenat. Diagn.* 33, 1137–1145.
- Hsieh, Y.-T., Yu Chao, Y.-M., Shiao, J.S.-C., 2013. A qualitative study of psychosocial factors affecting expecting mothers who choose to continue a cleft lip and/or palate pregnancy to term. *J. Nurs. Res.* 21, 1–9.
- Johnson, B., 2023. Babies with disabilities and their entitlement to imagined hopeful futures. In: Beckett, A.E., Callus, A.-M. (Ed.), *The Routledge International Handbook of Children's Rights and Disability*. Routledge, pp. 263–264.
- Joseph, P., 2023. 'It's all public anyway': a collaborative navigation of anonymity and informed consent in a study with identifiable parent carers. *Ethics Soc. Welfare* 17, 191–205.
- Kasparian, N.A., 2019. Heart care before birth: a psychobiological perspective on fetal cardiac diagnosis. *Prog. Pediatr. Cardiol.* 54, 101142.
- Kozłowska, K., Walker, P., McLean, L., Carrive, P., 2015. Fear and the defense cascade: clinical implications and management. *Harv. Rev. Psychiatr.* 23, 263–287.
- Marokakis, S., Kasparian, N.A., Kennedy, S.E., 2017. Parents' perceptions of counselling following prenatal diagnosis of congenital anomalies of the kidney and urinary tract: a qualitative study. *BJU Int.* 119, 474–481.
- McKechnie, A.C., Pridham, K., Tluczek, A., 2015a. Preparing heart and mind for becoming a parent following a diagnosis of fetal anomaly. *Qual. Health Res.* 25, 1182–1198.
- McKechnie, A.C., Pridham, K., Tluczek, A., 2015b. Walking the "emotional tightrope" from pregnancy to parenthood. *J. Fam. Nurs.* 22, 74–107.
- Michie, M., 2020. Is preparation a good reason for prenatal genetic testing? Ethical and critical questions. *Birth Defects Res.* 112, 332–338.
- Moran, D., 2000. *Introduction to Phenomenology*. Routledge, Oxon: UK.
- Namachivayam, S.P., Butt, W., Brizard, C., Millar, J., Thompson, J., Walker, S.P., et al., 2023. Potential benefits of prenatal diagnosis of TGA in Australia may be outweighed by the adverse effects of earlier delivery: likely causation and potential solutions. *Arch. Dis. Child.*
- Oben, P., 2020. Understanding the patient experience: a conceptual framework. *J. Patient Exp.* 7, 906–910.
- Oftedal, A., Bekkhus, M., Haugen, G.N., Czajkowski, N.O., Kaasen, A., 2022. The impact of diagnosed fetal anomaly, diagnostic severity and prognostic ambiguity on parental depression and traumatic stress: a prospective longitudinal cohort study. *Acta Obstet. Gynecol. Scand.* 101, 1291–1299.
- Otter.ai (n.d.). *Otter.ai*. (p. Automated transcription).
- Paulsen, S.L., Lanius, U.F., 2014. Introduction: the ubiquity of dissociation. In: Lanius, U. F., Paulsen, S.L., Corrigan, F.M. (Eds.), *Neurobiology and Treatment of Traumatic Dissociation: towards an Embodied Self*. Springer Publishing Company.
- Psaila, K.M., Schmied, V., Heath, S., 2023. Exploring continuity of care for women with prenatal diagnosis of congenital anomaly: a mixed method study. *J. Clin. Nurs. QSR International Pty Ltd*, 2018. NVivo, Version 12.
- Raybaud, C., 2010. The corpus callosum, the other great forebrain commissures, and the septum pellucidum: anatomy, development, and malformation. *Neuroradiology* 52, 447–477.
- Rychik, J., Donaghue, D.D., Levy, S., Fajardo, C., Combs, J., Zhang, X., et al., 2013. Maternal psychological stress after prenatal diagnosis of congenital heart disease. *J. Pediatr.* 162, 302–307 e301.
- Shakes, P., Cashin, A., Hurlley, J., 2020. Scoping review of the prenatal diagnosis of agenesis of the corpus callosum. *J. Obstet. Gynecol. Neonatal Nurs.* 49, 423–436.
- Smith, C.J., Smith, Z.G., Rasool, H., Cullen, K., Ghosh, M., Woolley, T.E., et al., 2023. Unravelling the clinical Co-morbidity and risk factors associated with agenesis of the corpus callosum. *J. Clin. Med.* 12.
- Tong, A., Sainsbury, P., Craig, J., 2007. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *Int. J. Qual. Health Care* 19, 349–357.
- Trainor, L.R., Bundon, A., 2020. Developing the craft: reflexive accounts of doing reflexive thematic analysis. *Qualitat. Res. Sport Exer. Health* 13, 705–726.
- Vandermause, R.K., Fleming, S.E., 2011. Philosophical hermeneutic interviewing. *Int. J. Qual. Methods* 10, 367–377.
- Werner, O., El Louali, F., Fouilloux, V., Amedro, P., Ovaert, C., 2019. Parental anxiety before invasive cardiac procedure in children with congenital heart disease: contributing factors and consequences. *Congenit. Heart Dis.* 14, 778–784.