An exploration of the parental experience of diagnosis of foetal abnormalities during routine antenatal ultrasound screening.

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Chapter One: Systematic Review

The impact of foetal abnormality identification during antenatal ultrasound screening on maternal mental health in the perinatal period: a systematic review.

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Abstract

Background

Prenatal ultrasound screening is a routine aspect of antenatal care for women in the UK, typically at 12 weeks of gestation to confirm pregnancy and at 20 weeks to screen for foetal abnormalities. The diagnosis of foetal abnormality is likely to be an emotional and stressful time for a woman and her family; however, the impact on maternal mental health in the short-term has not been systematically evaluated. This systematic review aimed to assess the mental health consequences of diagnosis of foetal abnormalities by routine ultrasound screening for women in the perinatal period.

Methods

A systematic review of six electronic databases (Embase, Medline, PsycInfo, CINAHL, Scopus, and Web of Science) was conducted to identify relevant studies published before April 2023. A narrative synthesis of findings was produced. Included studies were those using standardised measures to assess maternal mental health following prenatal diagnosis of foetal abnormality.

Results

A total of 3221 studies were screened and 13 met inclusion criteria (eight prospective observational and five cross-sectional studies). A narrative synthesis of findings was presented according to three main mental health outcome domains identified: anxiety, depression, and traumatic stress. Women with recent diagnoses of foetal abnormality on average had anxiety reaching clinical threshold for concern, significantly higher than normal controls. Depression symptoms were, on average, higher than those with normal ultrasound findings. Approximately one third of those surveyed demonstrated traumatic stress above threshold for clinical concern. Risk of bias was assessed using the Joanna Briggs Institute Critical Appraisal Tool for Quasi-Experimental Studies.

Conclusions

Prenatal diagnosis of foetal abnormality appears to be associated with poor mental health, and increased risk of anxiety, depression, and traumatic stress symptoms for women during pregnancy. Studies in this area are limited by the lack of assessment of mental health prior to diagnosis. It is recommended that clinicians providing ultrasound screening and prenatal diagnoses should consider the psychological needs of women during this process. Sources of

support should be considered to minimise the potential impact of these risks on obstetric and child development outcomes.

Background

Routine ultrasound screening is an important aspect of prenatal care for women and their unborn infants, and in the United Kingdom typically involves a scan in the first trimester to provide an estimated date of delivery and again at around 20 weeks of gestation to screen for foetal abnormalities (NHS Inform, 2021). Women typically view ultrasound scanning as integral to their pregnancy experience (Moncrieff et al., 2021), and the uptake of offers of ultrasonography in pregnancy is high internationally (Lalor and Begley, 2006), between 97 and 99 percent in UK samples (Redshaw and Heikkila, 2010). Screening tests offer potential benefits, such as early identification of abnormalities leading to earlier treatment, but also potential harm. This includes psychological distress associated with testing or test findings, including false positives (Marteau, 1990).

Parents typically view ultrasound scans as a time for them to "meet" their baby, and they value being able to share pictures of the unborn baby with family and friends (Moncrieff et al., 2021). It has been suggested that prenatal ultrasound scanning may positively impact the attachment between a mother and her unborn baby, although evidence for this is mixed (Baillie and Hewison, 1999). This gives the ultrasound scan a unique social significance when compared with other forms of prenatal screening, such as amniocentesis or maternal serum screening. This can be problematic, as the mother's expectation of the scan may contrast with the expectations of healthcare providers, who view the identification of foetal abnormalities as the primary purpose of routine ultrasound screening. As a result, parents may be underprepared for the possibility of being told what they may consider bad news about their baby (Luz et al., 2017).

The routine nature of ultrasound screening may lead to informed consent for this procedure being less robust than that of amniocentesis or maternal serum screening (Mitchell, 2004). Ultrasonography also has distinct features to other means of screening, including being in the room with the healthcare provider examining the findings in real time, and being able to visualise any identified abnormality. In their study of 74 women undergoing invasive (amniocentesis) and non-invasive (ultrasonography) diagnostic procedures in Croatia, Nakić Radoš and colleagues (2013) demonstrated that levels of anxiety regarding both procedures were comparable prior to the procedure and above general population norms.

In the year 2019, serious congenital abnormalities were identified in 1512 pregnancies in Scotland (3.06% of live and still births; Public Health Scotland, 2021). For women who do receive a diagnosis of foetal abnormality, the course of expected pregnancy is changed. The care team around the woman changes as she accesses more specialist support, and she may lose the relationships with existing routine supports she has developed (Áskelsdóttir et al., 2008).

Receiving abnormal foetal screening results has been demonstrated to meet the American Psychological Association (APA) definition of traumatic, in that it is "sudden and unexpected", it is disruptive to parents' beliefs about their sense of control, their "basic assumptions about the world and others" and is "usually experienced with intensity, terror and helplessness" (Aite et al., 2011, p1). The acute distress experienced by parents has been shown to have ongoing influence on the remainder of the pregnancy and future pregnancies (Hodgson and McClaren, 2018) and parenting approaches in the longer term (Giuliani et al., 2014). In response to the diagnosis, a considerable proportion of parents report high levels of distress and develop anxiety and depression requiring intervention (Skari et al., 2006). Despite this distress, most parents report feeling glad to have found out about the diagnosis prenatally, and better able to prepare for their child's birth (Hedrick, 2005).

Maternal mental health during pregnancy is associated with health outcomes for the mother and child (Alder et al., 2007). Increased anxiety in pregnancy has been shown to be negatively related to maternal nausea (Fischbein et al., 2019), foetal activity (DiPietro et al., 2003) and obstetric outcomes (Mancuso et al., 2004). Anxiety during pregnancy is a strong predictor of postnatal depression, even when controlling for prenatal depression (Fairbrother et al., 2017). Following prenatal diagnosis, parents may be less confident or consider themselves less capable than parents with normal ultrasound findings (Giuliani et al., 2014) which can have consequences for the effectiveness of parenting practices and longer-term child outcomes (Skreden et al., 2010).

Aims

This study aims to systematically review the literature assessing measurable mental health outcomes of identification of foetal abnormalities by ultrasound screening for women in the perinatal period. The primary research question will focus on mental health outcomes in the period immediately after diagnosis. Where possible, the secondary research question

regarding mental health outcomes for the remainder of pregnancy and/or early postnatal period will be discussed.

Research questions

1. What are the mental health outcomes for women immediately after diagnosis when foetal abnormality is identified through ultrasound screening?

2. How do mental health outcomes develop over the perinatal period (e.g., through repeated measures in the remaining pregnancy/early postnatal period; and/or in response to surgical counselling)?

Methods

Search Strategy

Following initial scoping searches, a search strategy was developed and discussed with a university librarian. Search terms using PICO (population, intervention, control, outcome) criteria can be found in Table 1. An example of the complete search strategy is presented in Appendix 1.1. The search strategy was adapted to make syntax and subject headings relevant for each database. Six electronic databases were searched in October 2022 and April 2023 and were as follows:

- MEDLINE (EBSCOhost)
- EMBASE (Ovid)
- CINAHL (EBSCOhost)
- PsycINFO (EBSCOhost)
- Scopus
- Web of Science - Core Collection

Table 1: PICO Search Terms

PICO Criteria	Example Terms Used
Population: Women in the perinatal period	Antepartum period; expectant (mothers/parents); perinatal period; postnatal period; prenatal care; pregnancy; antenatal; maternity
Intervention: Ultrasound scan identifies foetal abnormality	Congenital (anomaly/abnormality/malformation); foetal (anomaly/abnormality/malformation); fetal (anomaly/abnormality/malformation); structural (anomaly/abnormality/malformation); antenatal diagnosis; prenatal diagnosis; abnormal (ultrasound/sonography); unexpected findings
Control	None
Outcome: mental health outcomes	Affective disorders; anxiety; anxiety disorders; depression; mental health; posttraumatic stress disorder; adjustment disorder; distress; mood; panic; psychiatric; psychological; psychosocial
Quantitative only	Quantitative; questionnaire; measure; inventory; cross-sectional; screening; prospective; observational

No limits were added to searches in terms of publication date, population, or language. Reference lists of included papers and relevant systematic reviews were also examined for additional relevant articles. Results of the search were exported to EndNote reference management software.

Inclusion/Exclusion Criteria

Eligibility criteria for each aspect of PICO criteria are provided in Table 2.

Table 2: PICO Eligibility Criteria

PICO Criteria	Inclusion Criteria	Exclusion Criteria
Population	• Mothers in the perinatal period (pregnancy to one year postnatal)	• Fathers/ non-pregnant partners, or parents without differentiated results for mothers • Beyond one year postnatal
Intervention	• Routine ultrasound scan identifies foetal abnormality before birth • Intention to continue pregnancy (i.e., paper does not focus on termination of pregnancy due to foetal abnormality (TOPFA))	• Other means of screening e.g., amniocentesis, maternal serum screening, NIPT • Ultrasound following bleeding or pain (non-routine) • Ultrasound identifies soft markers, false positive, or no abnormality • Focus of research is TOPFA • Abnormality diagnosed postnatally
Control	Not applicable	Not applicable
Outcome	• Standardised quantitative diagnostic and/or symptomatic measures of mental health conditions • First outcome measure completed during pregnancy • Empirical study, peer reviewed publication • Paper written in English language	• Qualitative outcomes only • Non-symptomatic outcomes e.g., coping styles, emotion list • First outcome measure completed postnatally • Conference abstracts, academic dissertations, systematic reviews • Paper in language other than English

Duplicate articles were removed prior to screening for eligibility criteria (see Figure 1). The first phase of screening involved reviewing titles and abstracts of identified articles (n=3221) and was carried out by the primary author. Relevant articles and those of uncertain relevance

were put forward for phase two (full text review n=51). The primary author then read in full the remaining papers and identified those meeting eligibility criteria for data extraction (n=13) . A co-rater was involved in screening a sample (10/51) of full text papers against eligibility criteria and agreement was reached. The primary author also discussed two further papers with the co-rater due to ambiguity.

Data Extraction and Quality Appraisal

A data extraction tool was developed and can be found in Appendix 1.2. The quality of included studies was appraised using the Joanna Briggs Institute (JBI) Critical Appraisal Checklist for Quasi-Experimental Studies (JBI, 2017; Appendix 1.3) due to the non-randomised nature of articles. The JBI checklist provides nine questions of appraisal to critique the methodological quality of a study to consider whether the risk of bias has been addressed throughout design, conduct, and write up stages (JBI, 2017). All included papers were appraised using the JBI checklist by the primary author, and four (30.77%) were also appraised by the co-rater to assess reliability of quality ratings.

Ratings given are provided in Table 3 (see Appendix 1.4 for full appraisal details). Of the four papers scored by a co-rater, there was agreement on seven items, eight items, nine items, and nine items, respectively. Discrepancies were discussed and agreement reached. Papers were not excluded based on quality appraisal, although strengths and limitations will be discussed in the results section.

Narrative Synthesis

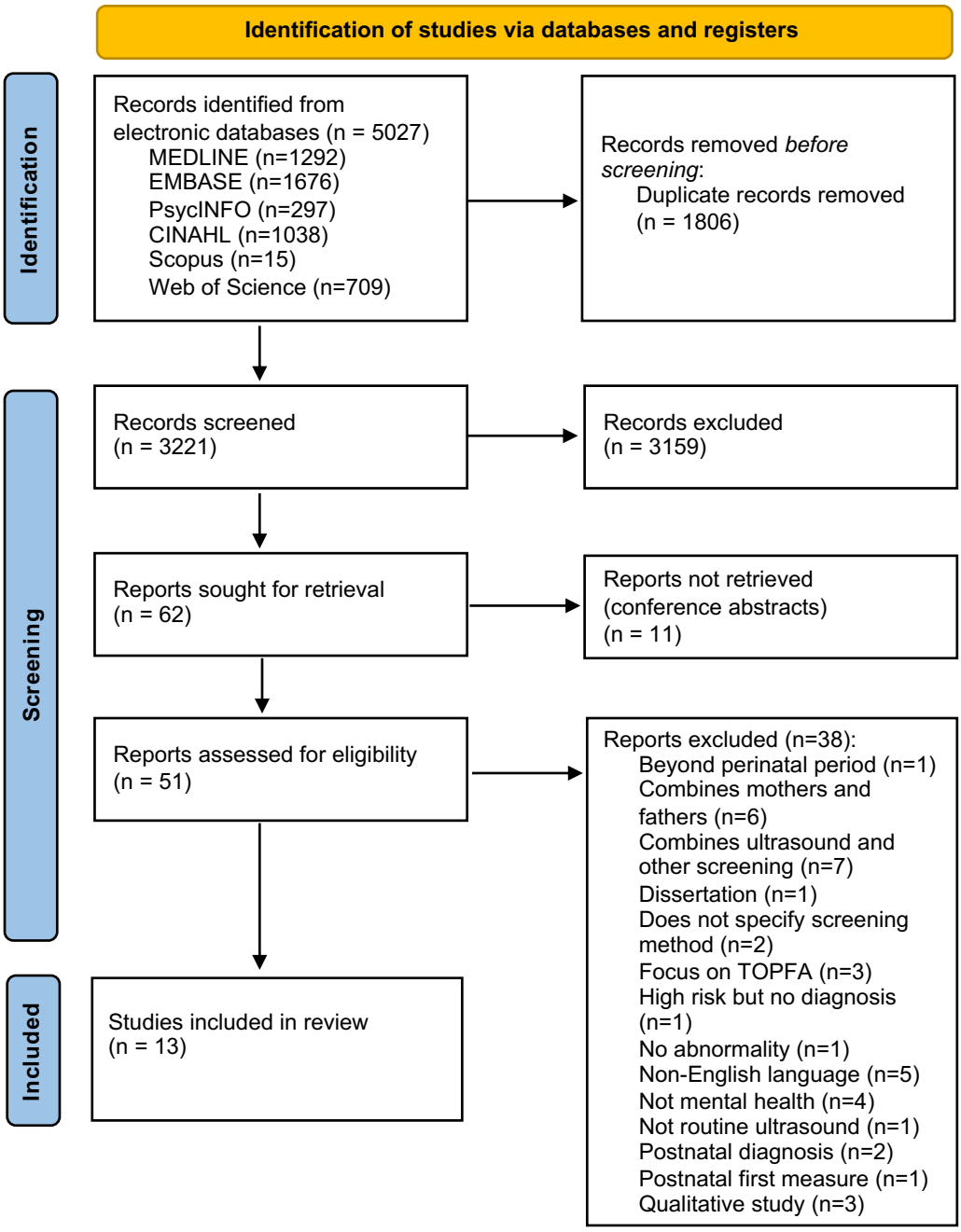
This systematic review provides a narrative synthesis of findings from identified studies due to heterogeneity of studies. Reporting of results is structured according to the aspect of mental health assessed in identified studies (i.e., anxiety, depression, traumatic stress) and grouped according to measures used. The written narrative provides a summary of findings in each area. Summary tables are also provided including sample sizes and mean scores on measures. Effect sizes were calculated and added to summary tables by the systematic review author where possible.

Results

Electronic database searches were carried out in October 2022 and identified 5027 records. After the screening process (see Figure 1), thirteen studies were found to be eligible for inclusion. A repeat search in April 2023 did not identify any additional relevant papers.

No further studies were identified through reference lists of identified papers or systematic reviews.

Figure 1: PRISMA Study Identification Flowchart



Study Characteristics

Details of the thirteen included studies are provided in Table 3. Ten of the studies used measures for anxiety: the General Health Questionnaire (GHQ-28, Goldberg and Hillier, 1979), the state subscale of the State Trait Anxiety Inventory (STAI-S, Spielberger et al., 1971), and the Hospital Anxiety and Depression Scale (HADS, Zigmond and Snaith, 1983). Nine studies used measures of depression, including those for postnatal depression: the GHQ-28, the HADS, the Beck Depression Inventory (BDI-II, Beck et al., 1996), the Edinburgh Postnatal Depression Scale (EPDS, Cox et al., 1987), and the Postpartum Depression Screening Scale (PDSS, Beck and Gable, 2000). Six studies used the Impact of Events Scale – Revised (IES-R, Weiss, 2007) to measure symptoms of traumatic stress. Studies were published by groups from Norway (n=4), the USA (n=4), Italy (n=1), Sweden (n=1), the UK (n=1), Malaysia (n=1), and Thailand (n=1). Study designs were observational, with eight prospective and five cross-sectional studies.

Quality of Studies

The JBI quality appraisal tool has nine items to assess the methodological quality of studies and consider the risk of bias. Scores ranged from three to nine with an average score of six (Table 3). The most common area of strength for the studies was the use of reliable outcome measures. The most common areas of weakness were regarding poorly powered studies based on sample size, a lack of multiple measurements of outcome measures (with studies typically measuring one time point only following diagnosis), lack of pre-diagnosis measure of mental health, and lack of follow up measures. Studies were not excluded based on quality appraisal, and studies were given equal weighting when considering results and drawing conclusions from this systematic review.

Table 3: Study Characteristics

#	Author, Year, Country	Sample Characteristics	Study Design	Mental Health Outcome & Measures	Key Findings	JBI Quality Appraisal Score*
1	Aite et al. (2009) Italy	Pregnant women with prenatal foetal diagnosis of congenital cystic adenomatoid malformation (CCAM) (n=21) or congenital diaphragmatic hernia (CDH) (n=23).	Prospective observational study. Within-group and between-group comparisons. Anxiety measured before and after surgical counselling for CCAM and CDH groups.	Anxiety (STAI-S)	Mean STAI-S scores in both groups were above clinical threshold for anxiety following diagnosis. Following counselling, CCAM group mean anxiety remained above clinical threshold whilst CDH group mean fell below clinical threshold.	7/9
2	Asplin et al. (2015) Sweden	Pregnant women with detected foetal malformation (n=56).	Prospective observational study. Repeated assessment mid-pregnancy, 2 months postpartum, and 1 year postpartum.	Anxiety (STAI-S), depression (EPDS)	In mid-pregnancy, 37% of women assessed were in clinical range for depressive symptoms; 26.5% at two months and 22% at one year. Mean anxiety decreased between mid-pregnancy and two months postpartum and increased again at one year postpartum.	5/8
3	Cole et al. (2016) USA	Pregnant women with a confirmed foetal abnormality (n=1032) and fathers (n=788).	Retrospective medical chart review - cross-sectional.	Depression (PDSS), traumatic stress (IES-R)	19.3% of women fell in the high-risk category (IES over 33) for post-traumatic stress symptoms, and 23% scored above clinical threshold for a major depressive	3/8

			Compared mothers and fathers at one time point.		disorder, 2-3 weeks after prenatal diagnosis was made.	
4	Helbig et al. (2011) Norway	Pregnant women with a newly detected foetal malformation (n=86) and with normal ultrasound findings (n=98).	Cross-sectional. Compared mothers with and without newly detected foetal malformation at one time point.	Anxiety, depression (GHQ 28), traumatic stress (IES-R)	Group with foetal malformation scored significantly higher on all domains of IES and GHQ than those without. Group with diagnosed malformation mean GHQ score was above clinical threshold, whilst control group was below.	6/8
5	Kaasen et al. (2010) Norway	Pregnant women with a foetal structural abnormality detected by ultrasound (n=180) and normal ultrasound findings (n=111).	Cross-sectional. Compared mothers with newly detected foetal malformation and normal ultrasound findings at one time point. Within-group analysis of those with diagnosis separated by severity of abnormality.	Anxiety (GHQ-28) depression (GHQ-28; EPDS), traumatic stress (IES-R)	Women with a newly detected foetal abnormality scored significantly higher on all measures of psychosocial distress than the comparison group. Least severe abnormalities were associated with lowest levels of IES intrusive distress.	6/8
6	Kaasen et al. (2017) Norway	Pregnant women with a structural foetal abnormality (n=48) and a normal ultrasound (n=105).	Prospective observational study. Between-group comparisons. Compared pregnant women with a structural foetal abnormality and those with normal ultrasound findings at 4	Anxiety (GHQ-28) depression (GHQ-28; EPDS), traumatic stress (IES-R)	Psychological distress was highest soon after diagnosis in the group with newly detected structural foetal abnormality. This group scored higher than controls on all measures at all time points, except on two subscales of IES and GHQ	7/9

			time points during pregnancy.		depression at time 4 (36 weeks' gestation).	
7	Kemp et al. (1998) UK	Pregnant women with diagnosed surgical abnormalities (n=26) and normal ultrasound (n=30).	Prospective observational study. Anxiety measured before and after surgical counselling in group of pregnant women with diagnosed abnormalities. Compared with normal controls at one time point in pregnancy.	Anxiety (STAI-S)	State anxiety in pregnant women with recently diagnosed surgical abnormalities was on average above a clinical threshold. Following surgical counselling, this reduced and on average was below clinical threshold. State anxiety was significantly higher in the diagnostic group prior to counselling, but there was not a significant difference following counselling.	5/8
8	Kim et al. (2021) USA	Pregnant women with foetal congenital abnormalities (n=23) and their non-pregnant partners (n=14).	Prospective observational study. Depression measured pre and postnatally. Within group analysis by severity of abnormality.	Depression (EPDS)	21.7% of pregnant women scored above clinical threshold on EPDS prenatally following diagnosis, and 15% postnatally. EPDS scores were correlated with severity of abnormality.	3/9
9	Oftedal et al. (2022) Norway	Pregnant women with foetal abnormality detected by ultrasound (n=81) and their male partners (n=69). Control group of pregnant women with normal ultrasound findings (n=110)	Prospective observational study. Within-group and between-group comparisons. Compared study and control mothers and fathers at four time points (T1 within 72 hours of	Depression (EPDS), traumatic stress (IES-R)	At T1, those with recently detected foetal abnormality had average EPDS score above clinical cut off. This fell below clinical threshold for T2 onwards, although remained statistically significantly higher than mothers in control group. Average IES intrusion subscale was above	7/9

		and their male partners (n=98).	diagnosis, T2 2-3 weeks later, T3 30 weeks' gestation, T4 36 weeks' gestation) in pregnancy and 6 weeks after birth (T5).		threshold for clinical significance for the group with foetal abnormality diagnosis at T1 but fell below threshold for remaining time points. All subscales of IES were significantly higher in group with diagnoses than control group, except for IES arousal subscale at time 4.	
10	Roslan et al. (2021) Malaysia	Pregnant women with foetal structural abnormality diagnosed by ultrasound (n=65) and normal ultrasound findings (n=76) during the covid 19 pandemic.	Prospective observational study. Within-group and between-group comparisons. Both groups assessed at 4 time points (T1 prior to scan, T2 2-4 weeks later, T3 1-2 weeks prior to delivery, T4 1-2 weeks post-delivery).	Anxiety, depression (HADS)	Pregnant women with foetal structural abnormality diagnosed during the covid pandemic had average depression scores in the "high" range at all time points measured. This did not differ significantly from a control group with normal ultrasound findings at T1-3 and was significantly lower than controls at T4. Those with diagnosed structural abnormalities had anxiety in the "moderate" range at all time points. This was significantly higher than normal controls at T2-4.	9/9
11	Rychik et al. (2013) USA	Pregnant women with prenatal diagnosis of foetal congenital heart disease (n=59).	Cross-sectional.	Anxiety (STAI-S), depression (BDI-II),	Clinically important traumatic distress found in 39% of respondents, depression in 22% and state anxiety in 31%.	3/8

				traumatic stress (IES-R)		
12	Titapant et al. (2015) Thailand	Pregnant women with non-lethal congenital abnormalities diagnosed by ultrasound (n=55).	Prospective observational study. Anxiety assessed at each follow up appointment after diagnosis during pregnancy (5 times).	Anxiety (STAI-S)	State anxiety immediately after diagnosis was significantly higher than in next follow up appointment. Mean state anxiety was in a clinical range at all times of assessment, except for time 4.	6/8
13	Wilpers et al. (2017) USA	Pregnant women with foetal abnormalities (n=19) and those with normal ultrasound findings (n=25).	Cross-sectional. Compared anxiety following diagnosis during pregnancy with normal control group at one time point.	Anxiety (STAI-S)	Mean anxiety score for those with diagnosed foetal abnormalities was above clinical threshold. This was significantly higher than normal control group, which had average anxiety below clinical threshold.	6/8

Note:

*A maximum score of 9 is possible on the JBI quality appraisal tool. In papers when a "not applicable" answer was given, the total possible score is reduced to reflect this.

Anxiety

Anxiety was assessed in 10 of the 13 identified studies. This was measured using the STAI-S (six studies), GHQ-28 (three studies), and HADS (one study). Identified studies varied in the timing of delivering assessment measures, with some studies delivering measures once and others using repeated measures for the remainder of pregnancy (20 to 40 weeks) and postnatally (up to twelve months). Some studies included comparisons with other groups, such as women with normal ultrasound findings. The primary research question regarding anxiety following diagnosis will be explored first, followed by subsequent analysis regarding timing of assessment and intervention, when possible, from identified studies.

STAI-S

All six studies utilising the STAI-S (see Table 4) showed that the mean score for women following a diagnosis of foetal abnormality was above 40, indicating mean state anxiety above a clinical threshold, soon after diagnosis.

Table 4: STAI-S Scores

Paper	**Women with a diagnosis of foetal abnormality (n=)**	**Mean STAI-S following diagnosis (SD)**
Aite et al., 2009	44	44.41 (5.39)
Asplin et al., 2015	54	43.41 (11.42)
Rychik et al., 2013	59	44.14 (14.69)
Titapant et al., 2015	55	52.4 (9.08)
Wilpers et al., 2017	19	43.6 (17)
Kemp et al., 1998	30	Median = 49.5 [Interquartile range 27 to 73]

With a clinical cut-off score of 40, Wilpers and colleagues (2017) found that 53% of those with prenatal diagnoses were in the clinical range for anxiety, compared to 12% of normal controls. Rychik and colleagues (2013) also used a severe clinical cut-off of over 65 and found that 31% of pregnant women with antenatally diagnosed foetal congenital heart disease were above this range, suggesting severe anxiety for almost one third of women in their sample.

Two studies which compared women with a diagnosis of foetal abnormality with women with normal ultrasound findings demonstrated that the group with diagnoses scored significantly higher than normal controls (Kemp et al., 1998; Wilpers et al., 2017). Calculation of effect size was possible from data presented by Wilpers and colleagues, and was found to be of medium strength (see Table 5). Quality appraisal for this paper identified that the study sample did not meet power which should be considered alongside this effect size. Insufficient information was presented by Kemp and colleagues (1998) to calculate effect size of this comparison.

Table 5: Comparison of STAI-S Scores for Clinical and Control Groups

Paper	**Women with a diagnosis. M (SD)**	**Normal controls. M (SD)**	**Statistical significance *p***	**Cohen's *d***	**Effect size *r***
Kemp et al., 1998	49.5 (NR)	NR (NR)	.0004	NP	NP
Wilpers et al., 2017	43.6 (17.0)	29.1 (8.5)	.002	1.079	0.475

NR: Not reported

NP: Not possible to calculate

Two further studies demonstrated no significant difference in anxiety when comparing conditions of major or minor severity following diagnosis (Aite et al., 2009; Titapant et al., 2015; see Table 6). Aite and colleagues (2009) had a sample size which did not reach power.

Table 6: Comparison of STAI-S Score in Conditions of Major and Minor Severity

Paper	**Condition of major severity. M (SD)**	**Condition of minor severity. M (SD)**	**Statistical significance *p***	**Cohen's *d***	**Effect size *r***
Aite et al., 2009	44.05 (4.96)	44.80 (5.92)	>0.05	-0.137	-0.069
Titapant et al., 2015	53.50 (10.04)	51.77 (8.57)	0.50	0.185	0.092

GHQ-28

Three studies used the GHQ-28 to assess psychological wellbeing, including anxiety. Mean anxiety subscale scores for those with recent diagnosis of foetal abnormality can be found in Table 7. All three studies compared women with a prenatal diagnosis of foetal abnormality with control groups with normal ultrasound findings. Each study concluded that anxiety scores during pregnancy were significantly higher in the foetal abnormality diagnoses group than those with normal ultrasound results. Calculations of effect sizes demonstrate these associations to be of medium strength. Quality appraisal identified that all three studies had comparison groups which were significantly different (for example, demographic factors for clinical group and control group) and as such the strength of this effect may be explained by other factors.

Table 7: GHQ-28 Anxiety Subscale Scores and Comparisons

Paper	**Women with a diagnosis of foetal abnormality (n=)**	**Clinical group mean (SD)**	**Normal controls (n=)**	**Control group mean (SD)**	**Statistical significance of difference**	**Cohen's *d***	**Effect size *r***
Helbig et al., 2011	86	8.4 (4.3)	98	5.5 (3.3)	<0.001	0.757	0.354
Kaasen et al., 2010	180	8.9 (4.4)	111	5.5 (3.4)	<0.001	0.865	0.397
Kaasen et al., 2017	48	8.5 (4.4)	105	5.5 (3.4)	<0.001	0.763	0.356

HADS

Roslan and colleagues (2021) used the HADS to assess anxiety in 141 women with recently diagnosed foetal abnormalities and demonstrated that average scores fell in the moderate range. This study was the only paper to measure anxiety prior to diagnosis of foetal abnormality identified in this systematic review. When comparing women with a diagnosis of foetal abnormality to those with normal ultrasound findings, authors demonstrated that groups did not significantly vary in their anxiety levels prior to diagnosis (7.92 versus 7.84,

p=.808) but following diagnosis the women with a diagnosis of foetal abnormality had significantly higher levels of anxiety than those with normal ultrasound findings with a small effect size (8.69 versus 7.95, p=0.036, d= 0.358, r= 0.176). No specific concerns were identified at the quality appraisal stage that would indicate cause for concern when interpreting this result.

Secondary Research Questions

Of the thirteen papers identified which assessed maternal anxiety following prenatal diagnosis, five assessed anxiety using repeated measures following surgical counselling over the remaining course of pregnancy. Aite and colleagues (2009) compared measures of anxiety before and after counselling for two conditions with different severity and found that, following surgical counselling for CDH, average maternal anxiety reduced to below the clinical cut-off, whilst anxiety for those receiving surgical counselling for CCAM remained above the clinical cut-off. They hypothesised that this was due to the uncertain nature of the course of CCAM and less clearly defined next steps for surgery than in CDH. Kaasen and colleagues (2017) measured anxiety at four timepoints throughout pregnancy while women with a diagnosis of foetal abnormality were offered regular surgical counselling appointments. Anxiety was found to reduce over this period but remained significantly higher than anxiety in normal controls. In contrast, Kemp and colleagues (1998) demonstrated that anxiety after counselling reached comparable levels to normal controls. Roslan and colleagues (2021) demonstrated that anxiety increased over the course of pregnancy despite surgical counselling, during the covid-19 pandemic. Titapant and colleagues (2015) demonstrated falling levels of anxiety through pregnancy with ongoing surgical counselling at each measured timepoint, other than their final measurement in the final weeks before birth, which they suggested may be a typical increase in anxiety in preparation for delivery.

Two studies included assessment of anxiety postnatally in addition to antenatal measurements of anxiety. Asplin and colleagues (2015) found that state anxiety decreased at measurement two months postpartum but was observed to increase when reassessed at 12 months postpartum. Roslan and colleagues (2021) demonstrated that those with prenatal diagnosis remained significantly more anxious than normal controls one to two weeks postnatally, during the covid-19 pandemic.

Summary

Across the three measures used to assess anxiety, women with recent diagnosis of foetal abnormality typically had on average levels of anxiety reaching a clinical threshold. When this was compared to those with normal ultrasound findings, women with diagnosis had significantly higher anxiety levels. It appears that diagnosis is anxiety-provoking regardless of the severity of diagnosis. Surgical counselling may influence anxiety in the remainder of pregnancy following foetal abnormality diagnoses, although evidence for this is mixed and appears to be influenced by other factors, such as the covid pandemic.

Depression

Depression was an outcome of interest in nine of the 13 identified studies. This was measured using the EPDS (five studies), GHQ-28 (three studies), PDSS (one study), BDI-II (one study), and HADS (one study). Some studies included comparisons with normal controls whilst others estimated prevalence of depression within groups of women with a diagnosis of foetal abnormality. The primary research question regarding depression following diagnosis in the antenatal period will be examined first, followed by subsequent assessment of secondary research questions available in identified studies.

EPDS

Three papers used the EPDS to measure antenatal depression scores of women with a diagnosis of foetal abnormality and compared this with scores of a control group with normal ultrasound findings (Kaasen et al., 2010; Kaasen et al., 2017; Oftedal et al., 2022). Mean EPDS scores are presented in Table 8 and were in the mild range (>10) for all studies. All three papers demonstrated significantly higher EPDS scores in the group of women with a diagnosis of foetal abnormality than normal control groups. Effect size calculations find these associations to be of a large effect. As noted previously, both papers by Kaasen and colleagues (2010; 2017) compared groups which differed significantly on factors such as demographics, and this was also the case for the paper by Oftedal and colleagues (2022).

Table 8: EPDS Scores and Comparisons

Paper	**Women with a diagnosis of foetal abnormality (n=)**	**Clinical group mean (SD)**	**Normal controls (n=)**	**Control group mean (SD)**	**Statistical significance of difference**	**Cohen's *d***	**Effect size *r***
Kaasen et al., 2010	180	12.3 (5.9)	111	3.1 (3.1)	<0.001	1.952	0.698
Kaasen et al., 2017	48	10.5 (6.0)	105	3.1 (3.1)	<0.001	1.550	0.612
Oftedal et al., 2022	81	11.26 (6.18)	110	3.18 (3.15)	<0.001	1.647	0.636

Oftedal and colleagues (2022) separated women with a diagnosis of foetal abnormality into high and low prognostic ambiguity conditions and found no significant difference in EPDS score between these subgroups. Kaasen and colleagues (2017) demonstrated that 65% of their study group were in the mild depression range (EPDS>10), and 44% were in the moderate range (EPDS >13).

Two further studies used the EPDS to provide an estimated prevalence of depressive symptoms in groups of women with diagnosed foetal abnormalities during pregnancy. Asplin and colleagues (2015) administered the EPDS to women informed of a foetal malformation during pregnancy and found that 20 of 54 (37%) women in the study had depressive symptoms above the moderate clinical cut-off of 13. Kim and colleagues (2021) assessed depression following antenatal diagnosis of foetal abnormality using the EPDS and found that 21.7% of their sample had a positive screening for depression, which they classed as a score over 10 (mild range). They found a moderate correlation between EPDS score and diagnostic severity of foetal abnormality of r=-0.49 (p=.02).

GHQ-28

Two studies (Kaasen et al., 2010; 2017) used the GHQ in addition to the EPDS, whilst Helbig and colleagues (2011) used the GHQ alone to measure symptoms of depression for those with

recent diagnosis of foetal abnormality (see Table 9 for sample sizes and mean subscale scores).

Table 9: GHQ-28 Depression Subscale Scores and Comparisons

Paper	**Women with a diagnosis of foetal abnormality (n=)**	**Clinical group mean (SD)**	**Normal controls (n=)**	**Control group mean (SD)**	**Statistical significance of difference**	**Cohen's *d***	**Effect size *r***
Helbig et al., 2011	86	1.7 (2.8)	98	0.3 (0.9)	<0.001	0.673	0.319
Kaasen et al., 2010	180	2.0 (3.1)	111	0.4 (1.3)	<0.001	0.673	0.319
Kaasen et al., 2017	48	1.8 (3.0)	105	0.3 (0.9)	<0.001	0.677	0.320

All three studies compared women with a diagnosis of foetal abnormality to those with normal ultrasound findings. Each study concluded that depression score during pregnancy was significantly higher in women with a diagnosis of foetal abnormality than those with normal ultrasound findings. Calculation of effect size demonstrates that this association is of medium strength. This effect may be influenced by demographic differences between the clinical and control groups.

BDI-II, PDSS and HADS

Rychik and colleagues (2013) used the BDI-II and found that 22% of 59 women with prenatal diagnosis of foetal congenital heart disease (CHD) met criteria for clinically important depressive symptoms (BDI>16). Cole and colleagues (2016) used the PDSS to assess prevalence of depressive symptoms in pregnancy following diagnosis of foetal abnormality and found that 23% of 1032 women screened positive for a major depressive disorder (PDSS>80) and a further 27.3% had signs of depressive symptoms (60<PDSS<79) following diagnosis of foetal abnormality.

Roslan and colleagues (2021) used the HADS to assess depressive symptoms in 65 women with recently diagnosed foetal abnormalities and demonstrated that average scores fell in the

high range (HADS>11) and were comparable to normal controls (p=0.760, d=-0.052, r= 0.026). These authors also measured symptoms prior to diagnosis, and it is important to note that the average depression score was in the high range prior to diagnosis for the clinical and non-clinical control groups.

Secondary Research Questions

Three studies which assessed depression prenatally also repeated measures in the early postnatal period. Asplin and colleagues (2015) followed up women with a diagnosis of foetal abnormality at two months and 12 months postpartum. At two months postpartum, 26.5% of those assessed met the moderate depression threshold on the EPDS, falling to 22% at 12 months postpartum. Kim and colleagues (2021) found no significant difference when the EPDS was repeated two weeks after birth between antenatal and postnatal mean scores. Roslan and colleagues (2021) found no significant difference between women with a diagnosis of foetal abnormality and those with normal ultrasound findings one to two weeks prior to delivery but did identify significantly higher depression scores in women with diagnoses of foetal abnormality one to two weeks postnatally.

Summary

From the above findings, it appears that women with recent diagnoses of foetal abnormality have significantly higher levels of depressive symptoms in pregnancy than those with normal ultrasound findings.

Traumatic Stress

Six studies used the IES-R to measure traumatic stress; four to assess mean traumatic stress scores following diagnosis and two to estimate prevalence of traumatic symptoms in those with prenatal diagnoses. When assessing mean traumatic stress scores, all four relevant papers (see Table 10) demonstrated mean intrusion subscales in the clinically significant range (IES intrusion >20).

Table 10: IES-R Subscale Scores and Comparisons

Paper	IES Subscale	**Women with a diagnosis of foetal abnormality (n=)**	**Clinical group mean (SD)**	**Normal controls (n=)**	**Control group mean (SD)**	**Statistical significance *p***	**Cohen's *d***	**Effect size *r***
Helbig et al., 2011	Intrusion	86	21.8 (8.9)	98	9.3 (6.3)	<0.001	1.621	0.630
	Avoidance		10.4 (6.3)		2.1 (3.8)	<0.001	1.595	0.624
	Arousal		14.5 (8.9)		4.0 (4.7)	<0.001	1.475	0.594
Kaasen et al., 2010	Intrusion	180	22.1 (8.6)	111	9.2 (6.3)	<0.001	1.711	0.650
	Avoidance		11.1 (7.3)		2.4 (4.0)	<0.001	1.478	0.594
	Arousal		14.6 (8.8)		4.0 (4.6)	<0.001	1.510	0.602
Kaasen et al., 2017	Intrusion	48	21.1 (10.7)	105	9.8 (6.6)	<0.001	0.769	0.359
	Avoidance		9.4 (7.2)		2.5 (4.1)	<0.001	1.178	0.507
	Arousal		11.6 (7.8)		3.8 (4.3)	<0.001	1.238	0.526
Oftedal et al., 2022	Intrusion	81	22.93 (10.29)	110	9.49 (6.60)	<0.001	1.555	0.614
	Avoidance		10.34 (8.36)		2.45 (4.05)	<0.001	1.201	0.515
	Arousal		12.09 (9.95)		3.68 (4.25)	<0.001	1.099	0.482

The above four papers also compared scores of those with diagnosed foetal abnormality to scores of a control group with normal ultrasound findings. Following diagnosis, all four studies found that those with confirmed foetal abnormality diagnoses had significantly higher IES-R scores on all domains than normal controls. Calculations of effect sizes show this to be a medium to large effect across all subscales. This effect may have been influenced by differences between group demographics.

Rychik and colleagues (2013) used the IES-R to determine the rate of traumatic stress in a population of 59 mothers whose infant had been diagnosed antenatally with CHD and found that 39% of those assessed had "clinically important" traumatic stress (IES-R≥24). Cole and colleagues (2016) also used the IES-R to determine rate of traumatic stress in response to confirmed foetal abnormality in pregnancy and showed that 32.9% of 1032 women had "clinically important" traumatic stress (IES-R≥24). 19.3% were found to have "high risk PTSD symptoms" (IES≥33).

Secondary Research Questions

Two studies administered the IES-R at multiple points throughout the remaining pregnancy, following diagnosis of foetal abnormality. Kaasen and colleagues (2017) administered the IES-R a few days after detection of the abnormality, 3 weeks later, at 30 weeks' gestation and 36 weeks' gestation. IES-R scores fell over time with counselling and close follow-up. When compared with normal controls, the clinical group scored significantly higher at all time points, other than at 36 weeks' gestation when only IES avoidance was significantly higher in the clinical group. Oftedal and colleagues (2022) also compared clinical and control groups at the same time points as Kaasen and colleagues (2017), plus an additional measurement taken at six weeks postpartum. It was demonstrated that the clinical group scored significantly higher on all subscales of the IES at all timepoints than normal controls, except for arousal at 36 weeks' gestation ($p=0.050$).

Summary

Diagnosis of foetal abnormality during pregnancy appears to be associated with traumatic stress of clinical concern for approximately one third of those surveyed and is significantly higher than for those with normal ultrasound findings. Symptoms of intrusion, arousal and avoidance are higher for those with foetal abnormality diagnosis than normal controls. This appears to last throughout pregnancy, although does appear to reduce with prenatal counselling.

Discussion

This review systematically searched for and evaluated the available literature on mental health outcomes on symptomatic and/or diagnostic measures for mothers with diagnosed foetal abnormalities by ultrasound during pregnancy. The identified research focused on anxiety, depression, and traumatic stress, using a range of outcome measures.

In measures of anxiety, women with recent diagnoses of foetal abnormality had anxiety levels above clinical threshold. This is significantly higher on average than for women with normal ultrasound findings with medium effect. Prenatal diagnosis appears to be anxiety-provoking regardless of the severity of the diagnosis. Surgical counselling may be associated with a reduction in anxiety over the remainder of the pregnancy. Depression levels are also higher on average for those with prenatal diagnosis than those with normal ultrasound findings, with medium to large effect. Evidence is mixed when examining the severity of diagnosis and its impact on depression scores. Traumatic stress reached a threshold of clinical concern for approximately one third of those surveyed in relevant studies, and this appeared to improve with surgical counselling in the prenatal period. Women with diagnoses of foetal abnormality scored higher on measures of traumatic stress in all domains, with medium to large effect.

Whilst comparisons across all domains demonstrated medium to large effect sizes when calculated, it is important to consider this information alongside quality appraisal information for the studies. Studies with sample sizes not reaching power are at increased risk of detecting false positives or false negatives (that is, finding an association that does not exist, or missing an association that does exist) due to increased influence of potential outliers in the data. Studies which were found to have significant differences between groups compared (clinical and non-clinical groups) may be influenced by these differences, such as in demographics. As such, additional factors beyond the presence or absence of foetal abnormality may be represented in the presented association. The findings of this study should therefore be considered alongside quality appraisal findings.

When considering what might explain the relationship between diagnosis of foetal abnormality and poor maternal mental health, it has been suggested that a contributing factor to increased risk of poor mental health for mothers with prenatal diagnosis of foetal abnormality is the increased uncertainty for the remainder of the pregnancy. Parents may need to wait for further medical testing postnatally to answer some questions that have begun prenatally (Áskelsdóttir et al., 2008). The uncertainty caused by prenatal diagnosis may also

explain differences observed in mental health outcomes according to the severity of the abnormality identified, or prenatal counselling offered by medical staff, as some conditions have more predictable outcomes and defined treatment plans than others (e.g., see Aite et al., 2009).

Brisch and colleagues (2003) highlighted that studies in this area examining the psychological impact of detection of foetal abnormality typically have a range of limitations, including limited information on the mental health of pregnant women, studies which are typically retrospective, small sample sizes and limited use of healthy controls. In this systematic review, although seven studies referred to participants undergoing surgical counselling, there was limited information provided to understand what this included. This may vary according to clinical need; however, this made it challenging to comment on the potential impact this counselling had on mental health outcomes during pregnancy.

Only one study provided a pre-diagnosis assessment of maternal mental health (Roslan et al., 2021). As such, it is difficult to assess the true impact of prenatal diagnosis on maternal mental health. Use of a control group with normal ultrasound findings is helpful, although as identified in quality appraisal, most studies used control groups which were different from clinical groups on factors such as demographic variables. As a result, findings in this area may be influenced by individual factors of those compared, rather than present a true reflection of the impact of diagnosis alone.

Limitations

To examine the current literature on mental health outcomes in the perinatal period of prenatal diagnosis, diagnostic and/or symptomatic measures were selected as a key criterion of included studies. This allowed the consideration of diagnosable mental health conditions, although it is recognised that tools of this kind alone are not enough to provide a clinical diagnosis. It is possible that relevant papers exist which may not have used such tools, for example using clinician impression and diagnostic criteria, and this review may have been enhanced by such studies' inclusion. In addition, focusing on the immediate term following diagnosis may not give a true reflection of mental health conditions as formal diagnostic criteria typically require symptoms to have endured for a period of weeks or months prior to diagnosis. This does however provide an effort to quantify the psychological distress experienced by women in clinical terms. Variation in reporting in papers (i.e., some

providing percentages of samples over clinical cut-off, some providing mean scores only) made meaningful comparisons challenging in this study.

Whilst this study included only quantitative measures of mental health outcomes, the addition of qualitative studies of mental health outcomes as reported by patients or healthcare professionals may have enhanced this study and allowed further consideration of the experience for those involved during the diagnostic process. This may also have improved understanding of the factors leading to increased anxiety, depression, and traumatic stress for those receiving a diagnosis of foetal abnormality.

Recognising that the diagnosis of foetal abnormality which results in the decision to terminate a pregnancy has unique emotional demands and psychological outcomes (e.g., see Brondino et al., 2013), the decision was made to exclude studies with a focus on diagnoses resulting in decision to terminate pregnancy. It is important to note that this review is relevant to non-lethal diagnoses only, however it may be of interest to consider similar studies focused on diagnoses leading to termination.

In conducting this systematic review, a co-rater was included for quality appraisal of four papers. Including the co-rater for appraisal of all papers would have further improved reliability of results. On the advice of a subject librarian, a comprehensive search strategy was developed for several databases; however, it is possible that relevant literature exists in other databases. Papers written in languages other than English were excluded due to lack of resource for translation in this student report, however there may be relevant literature that has been excluded written in other languages.

Implications for Future Research, Policy, and Practice

Whilst the JBI quality appraisal checklist was used to consider the risk of bias in identified papers, conclusions of this study were not weighted on the quality of papers, with the author attempting to provide a summary of all identified results. As such, poorer quality papers may influence presented results. It may be beneficial in similar reviews in the future to weight conclusions according to effect sizes and quality appraisal. Whilst statistical significance and effect sizes were examined in this study, this may not capture the clinical significance of comparisons either between groups or within groups over time. Statistically significant changes on a measure may or may not capture perceivable “real world” changes for study participants. Future research could use measures of clinical significance to enhance reviews in this area.

Given that foetal abnormalities occur in approximately three percent of pregnancies, with a significant proportion detected before birth by routine ultrasound, the potential influence of prenatal diagnoses on maternal mental health is relevant to over 1500 women in Scotland per year (Public Health Scotland, 2021) and is important from a public health perspective. These findings highlight the importance of keeping the mental health of mothers in mind as they undergo prenatal ultrasound screening for foetal abnormalities. Mothers may be underprepared for abnormal findings (Luz et al., 2017) and may be at risk of poor mental health in the perinatal period which may place their infant at risk of poor outcomes (Alder et al., 2007). A diagnosis of foetal abnormality is likely to be a shock to parents and some distress may be a likely outcome; however, clinicians should be aware of the potential negative association with maternal mental health in the short term.

Current routine practice in Scotland is that women should receive written information in advance of prenatal screening informing them of their right to decide whether to engage in screening (see Public Health Scotland, 2021b). However, the only risk associated with testing discussed in this document is related to increased risk of miscarriage following amniocentesis, with no discussion of risks including psychological harms associated with ultrasonography. To provide informed consent to participate in antenatal screening, women should be made aware of the potential distress that foetal abnormality diagnosis could cause.

Although this study focused on the perinatal period of pregnancy up to one-year, further research on the longer-term impact of prenatal diagnosis on maternal mental health and child outcomes would be beneficial in considering the consequences of diagnosis. Further research on the specific aspects included in prenatal surgical counselling would also help to establish the impact of these interventions for maternal mental health during this period and to make recommendations for the most effective counselling for families in the future. Typically, counselling is used as a generic term to describe input from a range of professionals including surgeons and medics. The present study suggests that diagnoses may be associated with poor mental health for some women at least in the short term of diagnosis, and it may be beneficial to consider the role that mental health professionals could offer to families at this time. Across Scotland, the recent expansion of Maternity and Neonatal Psychological Intervention (MNPI) services (Scottish Government, 2023) means clinical psychologists with specialist knowledge in this area should be increasingly available to women who have received antenatal diagnoses for individual psychological support where required. More broadly, revising public health messaging including written information provided antenatally

regarding antenatal screening with potential mental health association in mind may be beneficial. Whilst this study focused on the mental health of mothers only, it is acknowledged that prenatal diagnosis effects fathers/co-parents (e.g., Skreden et al., 2010) and further research could also examine the interaction between parents' reactions to diagnosis.

The mental health of mothers during pregnancy has implications for their health in pregnancy, and the health of the unborn child. Prenatal maternal mental health has been linked to several obstetric outcomes, including miscarriage, low birth weight and prematurity (see Fairbrother et al., 2017). Anxiety in the prenatal period has been shown to influence how a mother communicates with her child postnatally, resulting in less skilful interaction (Field et al., 2005). Children with a prenatal diagnosis of foetal abnormality may already be at risk of poorer health and developmental outcomes (Skreden et al., 2010) and as such it is important to consider the impact that maternal mental health may have on these already vulnerable children in both the short- and longer-term.

Conclusions

This systematic review has examined the mental health of women receiving diagnoses of foetal abnormalities in pregnancy. For a considerable number of these women, their mental health is likely to be negatively associated with prenatal diagnoses in the perinatal period. When compared to women with normal ultrasound findings, these women are significantly more likely to report a range of mental health difficulties in the short term. Methodological limitations of studies of this kind include a lack of assessment of mental health prior to diagnosis, unclear reporting on the care given to women and families following diagnosis by services, and poorly powered studies based on sample size. When working with women given prenatal diagnoses of foetal abnormalities, clinicians should be aware that this diagnosis may be associated with poor maternal mental health for the remainder of pregnancy and consider ways to support women during this time.

References

(References in bold indicate papers included in systematic review).

Aite, L., Zaccara, A., Mirante, N., Nahom, A., Trucchi, A., Capolupo, I., & Bagolan, P. (2011). Antenatal diagnosis of congenital anomaly: A really traumatic experience. *Journal of Perinatology*, *31*(12), 760–763. https://doi.org/10.1038/jp.2011.22

Aite, L., Zaccara, A., Trucchi, A., Brizzi, C., Nahom, A., Iacobelli, B., Capolupo, I., & Bagolan, P. (2009). When uncertainty generates more anxiety than severity: The prenatal experience with cystic adenomatoid malformation of the lung. *Journal of Perinatal Medicine*, *37*(5), 539–542. https://doi.org/10.1515/JPM.2009.098

Alder, J., Fink, N., Bitzer, J., Hösli, I., & Holzgreve, W. (2007). Depression and anxiety during pregnancy: a risk factor for obstetric, fetal and neonatal outcome? A critical review of the literature. *The journal of maternal-fetal & neonatal*, *20*(3), 189–209. https://doi.org/10.1080/14767050701209560

Áskelsdóttir, B., Conroy, S., & Rempel, G. (2008). From diagnosis to birth parents' experience when expecting a child with congenital anomaly. *Advances in Neonatal Care*, *8*(6), 348–354. https://doi.org/10.1097/01.ANC.0000342768.94734.23

Asplin, N., Wessel, H., Marions, L., & Georgsson Öhman, S. (2015). Maternal emotional wellbeing over time and attachment to the fetus when a malformation is detected. *Sexual and Reproductive Healthcare*, *6*(3), 191–195. https://doi.org/10.1016/j.srhc.2015.04.003

Baillie C. & Hewison J. (1999). Should ultrasound scanning in pregnancy be routine? *J Reprod Infant Psychol,* 17:149–57. https://doi.org/10.1080/02646839908409094

Beck, C. T. & Gable, R. K. (2000). Postpartum Depression Screening Scale: development and psychometric testing. *Nursing Research, 49*(5), 272–282. https://doi.org/10.1097/00006199-200009000-00006

Beck, A. T., Steer, R. A., & Brown, G. (1996*). Beck Depression Inventory: Manual.* (2nd ed). Psychological Corp. San Antonio.

Brisch, K. H., Munz, D., Bemmerer-Mayer, K., Terinde, R., Kreienberg, R., & Kächele, H. (2003). Coping styles of pregnant women after prenatal ultrasound screening for fetal malformation. *Journal of Psychosomatic Research*, *55*(2), 91–97. https://doi.org/10.1016/S0022-3999(02)00572-X

Brondino, N., Colombini, G., Morandotti, N., Podavini, F., De Vidovich, G., Formica, M., Arossa, A., De Silvestri, A., Montanari, L., & Caverzasi, E. (2013). Psychological correlates of decision-making during prenatal diagnosis: A prospective study. *Journal of Psychosomatic Obstetrics and Gynecology*, *34*(2), 68–74. https://doi.org/10.3109/0167482X.2013.797404

Cole, J. C. M., Moldenhauer, J. S., Berger, K., Cary, M. S., Smith, H., Martino, V., Rendon, N., & Howell, L. J. (2016). Identifying expectant parents at risk for psychological distress in response to a confirmed fetal abnormality. *Archives of Women's Mental Health*, *19*(3), 443–453. https://doi.org/10.1007/s00737-015-0580-6

Cox, J. L., Holden, J. M., & Sagovsky, R. (1987). Detection of postnatal depression: development of the 10-item Edinburgh Postnatal Depression Scale. *The British Journal of Psychiatry*, *150*(6), 782-786.

DiPietro, J. A., Costigan, K. A., & Gurewitsch, E. D. (2003). Fetal response to induced maternal stress. *Early Human Development*, *74*(2), 125–138. https://doi.org/10.1016/j.earlhumdev.2003.07.001

Fairbrother, N., Young, A. H., Zhang, A., Janssen, P., & Antony, M. M. (2017). The prevalence and incidence of perinatal anxiety disorders among women experiencing a medically complicated pregnancy. *Archives of Women's Mental Health*, *20*(2), 311–319. https://doi.org/10.1007/s00737-016-0704-7

Field, T., Hernandez-Reif, M., Vera, Y., Gil, K., Diego, M., Bendell, D., & Yando, R. (2005). Anxiety and anger effects on depressed mother-infant spontaneous and imitative interactions. *Infant Behavior & Development, 28*(1), 1–9. https://doi.org/10.1016/j.infbeh.2004.06.003

Fischbein, R. L., Nicholas, L., Kingsbury, D. M., Falletta, L. M., Baughman, K. R., & VanGeest, J. (2019). State anxiety in pregnancies affected by obstetric complications:

A systematic review. *Journal of Affective Disorders*, *257*, 214–240. https://doi.org/10.1016/j.jad.2019.07.007

Giuliani, R., Tripani, A., Pellizzoni, S., Clarici, A., Lonciari, I., D'Ottavio, G., & Schleef, J. (2014). Pregnancy and postpartum following a prenatal diagnosis of fetal thoracoabdominal malformation: The parental perspective. *Journal of Pediatric Surgery*, *49*(2), 353–358. https://doi.org/10.1016/j.jpedsurg.2013.07.025

Goldberg, D. P., & Hillier, V. F. (1979). A scaled version of the General Health Questionnaire. *Psychological medicine*, *9*(1), 139-145. https://doi.org/10.1017/s0033291700021644

Hedrick, J. (2005). The lived experience of pregnancy while carrying a child with a known, nonlethal congenital abnormality. *Journal of Obstetric, Gynecologic, and Neonatal Nursing*, *34*(6), 732–740. https://doi.org/10.1177/0884217505282020

Helbig, A., Kaasen, A., Malt, U. F., & Haugen, G. (2011). Psychological distress after recent detection of fetal malformation: Short-term effect on second-trimester uteroplacental and fetoplacental circulation. *BJOG: An International Journal of Obstetrics and Gynaecology*, *118*(13), 1653–1657. https://doi.org/10.1111/j.1471-0528.2011.03155.x

Hodgson, J., & McClaren, B. J. (2018). Parental experiences after prenatal diagnosis of fetal abnormality. *Seminars in Fetal and Neonatal Medicine*, *23*(2), 150–154. https://doi.org/10.1016/j.siny.2017.11.009

Joanna Briggs Institute. (2019). *Checklist for quasi-experimental studies*. https://joannabriggs.org/ebp/critical_appraisal_tools

Kaasen, A., Helbig, A., Malt, U. F., Næs, T., Skari, H., & Haugen, G. (2010). Acute maternal social dysfunction, health perception and psychological distress after ultrasonographic detection of a fetal structural anomaly: Fetal medicine. *BJOG: An International Journal of Obstetrics and Gynaecology*, *117*(9), 1127–1138. https://doi.org/10.1111/j.1471-0528.2010.02622.x

Kaasen, A., Helbig, A., Malt, U. F., Næs, T., Skari, H., & Haugen, G. (2017). Maternal psychological responses during pregnancy after ultrasonographic detection of

structural fetal anomalies: A prospective longitudinal observational study. *PLoS ONE*, *12*(3), 1–16. https://doi.org/10.1371/journal.pone.0174412

Kemp, J., Davenport, M., & Pernet, A. (1998). Antenatally diagnosed surgical anomalies: The psychological effect of parental antenatal counseling. *Journal of Pediatric Surgery*, *33*(9), 1376–1379. https://doi.org/10.1016/S0022-3468(98)90011-2

Kim, A. J. H., Servino, L., Bircher, S., Feist, C., Rdesinski, R. E., Dukhovny, S., Shaffer, B. L., & Saxton, S. N. (2021). Depression and socioeconomic stressors in expectant parents with fetal congenital anomalies. *Journal of Maternal-Fetal and Neonatal Medicine*, *0*(0), 1–7. https://doi.org/10.1080/14767058.2021.1992379

Lalor, J., & Begley, C. (2006). Fetal anomaly screening: What do women want to know? *Journal of Advanced Nursing*, *55*(1), 11–19. https://doi.org/10.1111/j.1365-2648.2006.03884.x

Luz, R., George, A., Spitz, E., & Vieux, R. (2017). Breaking bad news in prenatal medicine: a literature review. *Journal of Reproductive and Infant Psychology*, *35*(1), 14–31. https://doi.org/10.1080/02646838.2016.1253052

Mancuso, R. A., Schetter, C. D., Rini, C. M., Roesch, S. C., & Hobel, C. J. (2004). Maternal prenatal anxiety and corticotropin-releasing hormone associated with timing of delivery. *Psychosomatic Medicine*, *66*(5), 762–769. https://doi.org/10.1097/01.psy.0000138284.70670.d5

Marteau, T. M. (1990). Screening in practice: Reducing the psychological costs. *British Medical Journal*, *301*(6742), 26. https://doi.org/10.1136/bmj.301.6742.26

Mitchell, L. M. (2004). Women's experiences of unexpected ultrasound findings. *Journal of Midwifery and Women's Health*, *49*(3), 228–234. https://doi.org/10.1016/j.jmwh.2003.11.004

Moncrieff, G., Finlayson, K., Cordey, S., McCrimmon, R., Harris, C., Barreix, M., Tunçalp, Ö., & Downe, S. (2021). First and second trimester ultrasound in pregnancy: A systematic review and metasynthesis of the views and experiences of pregnant

women, partners, and health workers. *PLoS ONE*, *16*(12): e0261096 https://doi.org/10.1371/journal.pone.0261096

Nakić Radoš, S., Košec, V., & Gall, V. (2013). The psychological effects of prenatal diagnostic procedures: Maternal anxiety before and after invasive and noninvasive procedures. *Prenatal Diagnosis*, *33*(12), 1194–1200. https://doi.org/10.1002/pd.4223

NHS Inform (2021). *Ultrasound scans during pregnancy*. Retrieved 26 November 2022, from https://www.nhsinform.scot/healthy-living/screening/pregnancy/ultrasound-scans-during-pregnancy.

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 Obstetricia et Gynecologica Scandinavica*, *101*(11), 1291–1299. https://doi.org/10.1111/aogs.14453

Public Health Scotland (2021). *Congenital anomalies in SCOTLAND 2000 to 2019*. Publications - Public Health Scotland. https://publichealthscotland.scot/publications/congenital-anomalies-in-scotland/congenital-anomalies-in-scotland-2000-to-2019/

Public Health Scotland (2021b) *You're pregnant! scans and tests*, Public Health Scotland. https://www.healthscotland.com/documents/30532.aspx

Redshaw, M., & Heikkila, K. (2010). *Delivered With Care. A National Survey of Women's Experience of Maternity Care 2010.* Technical Report. National Perinatal Epidemiology Unit, University of Oxford, Oxford, United Kingdom. https://researchonline.lshtm.ac.uk/id/eprint/2548656

Roslan, N. R., Fauzi, M. F. M., Teng, L. W., & Azurah, A. G. N. (2021). Maternal mental health following ultrasonographic detection of fetal structural anomaly in the midst of the covid-19 pandemic. *International Journal of Environmental Research and Public Health*, *18*(24). https://doi.org/10.3390/ijerph182412900

Rychik, J., Donaghue, D. D., Levy, S., Fajardo, C., Combs, J., Zhang, X., Szwast, A., & Diamond, G. S. (2013). Maternal psychological stress after prenatal diagnosis of

congenital heart disease. *Journal of Pediatrics*, *162*(2), 302-307.e1. https://doi.org/10.1016/j.jpeds.2012.07.023

Scottish Government (2023) *Perinatal and Infant Mental Health Services: Update*, *Scottish Government*. The Scottish Government. https://www.gov.scot/publications/perinatal-infant-mental-health-services-update/documents/

Skari, H., Malt, U. F., Bjornland, K., Egeland, T., Haugen, G., Skreden, M., Björk, M. D., Ostensen, A. B., & Emblem, R. (2006). Prenatal diagnosis of congenital malformations and parental psychological distress - A prospective longitudinal cohort study. *Prenatal Diagnosis*, *26*(11), 1001–1009. https://doi.org/10.1002/pd.1542

Skreden, M., Skari, H., Malt, U. F., Haugen, G., Pripp, A. H., Faugli, A., & Emblem, R. (2010). Long-term parental psychological distress among parents of children with a malformation - A prospective longitudinal study. *American Journal of Medical Genetics, Part A*, *152*(9), 2193–2202. https://doi.org/10.1002/ajmg.a.33605

Spielberger, C. D., Gonzalez-Reigosa, F., Martinez-Urrutia, A., Natalicio, L. F., & Natalicio, D. S. (1971). The state-trait anxiety inventory. *Revista Interamericana de Psicologia/Interamerican Journal of Psychology*, *5*(3 & 4). https://doi.org/10.30849/rip/ijp.v5i3 & 4.620

Titapant, V., & Chuenwattana, P. (2015). Psychological effects of fetal diagnoses of non-lethal congenital anomalies on the experience of pregnant women during the remainder of their pregnancy. *Journal of Obstetrics and Gynaecology Research*, *41*(1), 77–83. https://doi.org/10.1111/jog.12504

Weiss, D. S. (2007). The impact of event scale: revised. *Cross-cultural assessment of psychological trauma and PTSD*, 219-238. https://doi.org/10.1007/978-0-387-70990-1_10

Wilpers, A. B., Kennedy, H. P., Wall, D., Funk, M., & Bahtiyar, M. O. (2017). Maternal Anxiety Related to Prenatal Diagnoses of Fetal Anomalies That Require Surgery. *JOGNN - Journal of Obstetric, Gynecologic, and Neonatal Nursing*, *46*(3), 456–464. https://doi.org/10.1016/j.jogn.2017.02.001

Zigmond, A. S., & Snaith, R. P. (1983). The hospital anxiety and depression scale. *Acta psychiatrica scandinavica*, *67*(6), 361-370. https://doi.org/10.1111/j.1600-0447.1983.tb09716.x

Chapter Two: Major Research Project

Parental experience of uncertainty and decision making during the diagnosis and management of congenital pulmonary airway malformation (CPAM).

Prepared in accordance with the author requirements for BMC Pediatrics

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Plain Language Summary

Title

Parental experience of uncertainty and decision making during the diagnosis and management of congenital pulmonary airway malformation.

Background

In the UK, pregnant women are offered ultrasound scans at around 20 weeks of pregnancy to check that their baby is healthy. Usually this is a happy time. However, this scan can sometimes identify problems. This allows doctors to begin to plan how to look after the baby but can be a shock for parents.

One problem that can be seen at the 20-week scan is CPAM. This stands for congenital pulmonary airway malformation, an area of abnormal lung tissue. When the baby is born, the CPAM might not cause any problems, and doctors cannot always tell if the baby will have problems later. Doctors can feel unsure about whether surgery is the best treatment plan or if they should "watch and wait". Sometimes parents are asked what they think should happen to help make the decision, especially if the baby is born without any problems.

Aims

This research project examined the experience of parents whose infant was diagnosed with CPAM which was not causing any obvious problems when the baby was born. This focused on parents' decisions about whether they wanted their baby to have surgery, to find out what it was like for parents to find out about CPAM during pregnancy, and what it was like to make decisions about whether the baby should have surgery.

Methods

This research interviewed four parents (three mothers and one father) of children who were born with a CPAM and who had attended appointments at the CPAM clinic. The study asked parents to "look back" on their experiences during pregnancy and once their baby was born. Participants were recruited through staff at the CPAM clinic, who sent them information about the study and a letter inviting them to take part. The parents were interviewed by a researcher and their interview was recorded and later transcribed. Interpretative Phenomenological Analysis (IPA) was used to analyse the interviews. This is a way of asking people to make sense of what has happened to them. The researcher then looks for themes across all the interviews to find themes across the experience of all participants.

Main Findings and Conclusions

Parents told the researcher about their experience of diagnosis of CPAM for their baby, and the researcher examined their accounts to generate four themes. These were “diagnosis changed the expected course of pregnancy”, “searching for information about the condition”, “making decisions about surgery” and “life after decision making”.

This study adds to understanding about what it is like for parents to be told about a CPAM at their 20-week scan, how this impacts the rest of their pregnancy, how they find out more about the condition, and how it impacts them today. The results lead to suggestions for ways healthcare staff can support families receiving diagnosis of CPAM in the future.

Abstract

Background

Congenital pulmonary airway malformation (CPAM) is a term used to describe a range of foetal abnormalities of the lung typically identified during routine antenatal ultrasound screening at 20 weeks' gestation. Babies are typically born without symptoms and as such there is uncertainty about the best course of treatment: to operate, or to watch and wait. This uncertainty can have psychological consequences for the parents receiving such a diagnosis. This study aims to explore the experience of parents who have been involved in decision making about their infant's care following diagnosis of CPAM in the context of uncertainty about treatment.

Methods

This study used a qualitative approach in which four participants participated in a semi-structured interview about their experiences. Interviews were recorded and transcribed verbatim. Transcripts were then analysed using Interpretative Phenomenological Analysis (IPA) to generate group experiential themes.

Results

Four group experiential themes were developed across the four interviews conducted: diagnosis changed the expected course of pregnancy; searching for information about the condition; making decisions about surgery; and life after decision making. Eleven subthemes were identified and are discussed.

Conclusions

The findings of this study provide an insight into the experiences of parents who received a diagnosis of CPAM during routine antenatal screening. Participants highlighted the changes to the course of pregnancy, the importance of finding out more information about the condition, the process of making decisions about surgery, and the impact this continued to have on their lives in the present day. Recommendations are made for clinicians when providing similar diagnoses, particularly with regards to families' informational needs. Possible future research directions are also discussed.

Background

Antenatal Sonography

Since January 2010, pregnant women in Scotland have routinely been offered an ultrasound scan mid-pregnancy (typically 18 to 21 weeks' gestation) to screen for major congenital abnormalities (Public Health Scotland, 2021). The use of ultrasound screening routinely, rather than in response to specific identified risks, means that women typically view scans as non-threatening, which can lead to shock if major abnormalities are identified (Lalor et al., 2007). Whilst most pregnant women will be unaffected by such a diagnosis, for approximately three percent a diagnosis of major abnormality will be given; this figure appears consistent across countries where mid-pregnancy routine ultrasound screening occurs, including Scotland (Public Health Scotland, 2021) and the United States of America (Tucker and Christian, 2022).

Expectant parents given diagnoses of major congenital abnormalities frequently experience post-traumatic stress symptoms (Cole et al., 2016), with mood and anxiety scores in the period immediately after diagnosis comparable to patients with major depression (Leithner et al., 2004). Diagnoses of "simple-to-fix" abnormalities with good outcomes have been shown to be as distressing for parents as potentially lethal conditions during pregnancy (Aite et al., 2011).

Congenital Pulmonary Airway Malformation (CPAM)

The British Lung Foundation describes CPAM as "abnormal areas of tissue (lesions) on the lung [that] happen when the airway and surrounding lung tissue does not develop properly" (BLF, 2022). They report that CPAM is the most common congenital lung abnormality and most babies "can be delivered normally and will not have any obvious symptoms after they are born". The term CPAM is used to refer to a range of lung malformations, including congenital cystic adenomatous malformation (CCAM), bronchopulmonary sequestration, and bronchogenic cysts. Improvement in ultrasound technology has led the rates of CPAM diagnosis to increase over time (Morini et al., 2018) to an estimated one in 10,000 births (Mehta and Sharma, 2022).

As babies are often born without symptoms, there is debate in clinical practice about how best to manage CPAM. Surveys conducted in the UK, Europe, and USA (Peters et al., 2013; Morini et al., 2018; and Berman et al., 2018 respectively) demonstrated a lack of consensus as to whether surgical intervention is indicated for asymptomatic infants. As a result,

variation exists in approaches between settings dependent on clinician judgment, with some favouring "surgical excision for nearly all detected lesions, whereas others are more cautious and recommend intervention only after the development of symptoms or complications" (Stanton et al., 2009, p. 1028).

This ambiguity may have psychological consequences for families. In earlier quantitative research, Aite and colleagues (2009) carried out a study with families in Italy who received prenatal diagnosis of congenital diaphragmatic hernia (CDH) or CCAM. They demonstrated that receiving a diagnosis of CDH, which has a higher mortality rate than CCAM but also a clearly defined treatment protocol, was experienced as less traumatic than diagnosis of CCAM, immediately following counselling from healthcare professionals. They suggested that "parents seem to relate better to defined management plans rather than the more pragmatic, yet entirely honest approach: 'we will see what it is like when the baby is born'" (p.1).

Decision Making

This ambiguity in approach to CPAM treatment gives space for discussion between clinicians and parents regarding the best course of action for the infant; including to operate when the baby is born or to "watch and wait". Decision making of this kind is influenced by multiple factors and involves complex interaction between those making the decisions. Decision-making models specific to medical settings between patients and healthcare professionals have been developed, although less so in neonatal or paediatric settings. Decision making is complicated when patients cannot voice their own preferences, for example neonates, and must be represented by surrogate decision-makers, usually their parents (Krick et al., 2020), who must make decisions at a highly emotive time (Sullivan and Cummings, 2020).

Historically, medical decision making has been based on either Best Interest Standard (BIS) or Shared Decision Making (SDM) approaches (Sullivan and Cummings, 2020). The BIS asserts that the course of action followed should be that with the highest overall benefit for the individual and which places the individual patient's best interests above all other factors, although it has been criticised for excluding the views and values of the wider network around the patient. In contrast, SDM considers the views of multiple stakeholders including healthcare professionals, the patient where possible, and their family. The goal with SDM is to make a decision that is medically beneficial whilst also considering what matters to the patient and their family.

Krick and colleagues (2020) presented a decision-making model specific to uncertainty in neonatal medicine. They proposed that "the presence of greater uncertainty ought to permit parents greater latitude to incorporate family values into their decision making even if these decisions are contradictory to the recommendations of the medical team" (p.1). They suggested the term "zone of parental discretion" (ZPD) to refer to the context of increased uncertainty and limited evidence of any greater risk or benefit from one decision or another. When there is little uncertainty, they noted that the ZPD becomes smaller and allows medical providers more confidence in mandating a treatment.

Research Aims

This study aims to explore the experience of parents who have previously received an antenatal diagnosis of CPAM and subsequently been involved in decision making about their infant's care. The study targets those within the ZPD regarding surgery for their infant; where there was uncertainty about the best course of treatment from healthcare professionals, due to limited evidence of harm or benefit of surgery.

Methods

Design

A qualitative design was utilised to explore the experience of parents who had received a prenatal diagnosis of CPAM for their child who was subsequently asymptomatic at birth, leading to uncertainty about the course of treatment. An Interpretative Phenomenological Analysis (IPA) approach was chosen. This methodology focuses on the experience of a small number of individuals and how they make sense of that experience (Smith, Flowers, and Larkin, 2009).

Theoretical Framework

IPA is underpinned by theoretical principles of phenomenology, hermeneutics, and an idiographic approach. It focuses on exploratory research, rather than explanatory, and is phenomenological in that it examines the lived experience of participants in its own terms, rather than pre-existing categories, or hypotheses. To present the lived experience of another, IPA recognises that the individual must first make interpretations of their experience which is in turn interpreted by the researcher. This second order interpretation is referred to as "double hermeneutics". Rather than making generalisations at a larger population level, the

idiographic aspect of IPA means that researchers prioritise the rich and detailed understanding of individuals in response to a specific situation. This leads to in-depth interviews, typically with small, homogenous samples, with Smith and colleagues (2009) suggesting that a sample size of three to six participants is reasonable for a student project using IPA and one-off interviews. IPA has been identified as an appropriate approach for topics which participants consider significant in their life course, often with emotional significance, and is often used in health contexts (Smith and Nizza, 2022).

Participants

This study took place within the NHS Greater Glasgow and Clyde (GGC) CPAM clinic, based at the Royal Hospital for Children. Participants were four unrelated parents (three mothers and one father) whose child had been diagnosed with CPAM antenatally, and who had subsequently been involved in decisions about the treatment plan for their child with clinical staff. Targeted parents were those with children aged over two years old, as the focus of the study was looking back on the experience of diagnosis and subsequent decision making, rather than those still going through the process.

Thirty-five potential participants who met eligibility criteria for the study (see Table 1) were identified by clinical staff and were sent recruitment materials through the mail by administrative staff. This included a recruitment letter, the participant information sheet and consent form for the study (see Appendices 2.1-2.3). Those sent recruitment materials were parents whose children continued to engage with the NHS GGC CPAM clinic for regular reviews, or if reviews had come to an end, had consented to be contacted for follow up for research purposes through the clinic. Those who wished to participate contacted the principal investigator (PI) to opt-in to the study, and an interview was arranged in person or via Microsoft Teams, depending on participant preference. Participants were asked to sign and return their consent form through the mail, at their in-person interview, or email a copy. For video interviews, consent was verified when the consent form was not signed in front of the researcher.

Table 1: Eligibility Criteria

Criteria	**Inclusion**	**Exclusion**
Child's diagnosis.	CPAM diagnosed during routine antenatal ultrasound screening.	Other diagnoses.
Child's health at birth.	Child born without symptoms/in good health. Uncertainty about whether surgery or watch and wait best approach.	Child symptomatic at birth/ acutely unwell. Child's presentation such that management was clear i.e., surgery definitely required.
Number of babies at scanning stage.	Singleton.	Multiple.
Child's age.	Over two years old.	Under two years old.
Child's surgical/health status.	Decision-making process about surgery for child is not currently ongoing. Parents of those who did and did not have surgery are both eligible to participate. Child is currently in good health and stable.	There are ongoing and current discussions about whether child should have surgery. Child is currently unwell or medically unstable.
Parents' language.	Parent can understand and communicate in English to an adequate level that does not require an interpreter to be present.	Parent requires an interpreter to engage with clinic visits.

Interviews

A semi-structured interview schedule was developed by the PI following discussion with clinical staff providing field supervision (two respiratory neonatal doctors and one consultant clinical psychologist) and initial reading of the literature regarding CPAM diagnosis. The schedule was further developed in supervision with the research supervisor. The research interview was structured in a way that asked participants to talk through their experience following a timeline beginning in pregnancy and ending in present day. Key topics and questions are presented in Table 2 (see Appendix 2.4 for full details and prompts).

Table 2: Interview Questions

A.	Your child now
	1. Please can you start by telling me a bit about your child?
B.	Pregnancy – before diagnosis
	2. How was your (/your partner's) pregnancy with {child} prior to the diagnosis? 3. What do you remember about going for scans?
C.	Pregnancy – diagnosis
	4. Can you tell me about when you first found out there was an unusual finding on the scan?
D.	Pregnancy – after diagnosis
	5. So you've had your scan (/partner's scan) and you've been told about the finding. What happened next?
E.	Birth
	6. How was the birth?
F.	Responding to the CPAM
	7. When did you first have to make decisions about baby's CPAM? What do you remember about the decision-making process?
G.	Follow up with CPAM clinic
	8. What involvement have you had with the clinic since then?
H.	Bringing it to a close
	9. Looking back on the experience, is there anything you would tell families starting out with a CPAM diagnosis? 10. Is there anything else you want to add? Do you have any other questions for me?

Interviews took place at the Royal Hospital for Children, or online using Microsoft Teams. Interviews lasted between 39 and 52 minutes, and each participant was interviewed once. Interviews were recorded with participant consent, using a Dictaphone if conducted in-person or Microsoft Teams software if online. Interview recordings were stored securely on a OneDrive account within the NHS. Participants received a £10 voucher for their participation and a debrief document once the interview ended (see Appendix 2.5).

Interviews were transcribed verbatim by the PI. Transcripts were pseudonymised prior to analysis and identifiable information stored separately. As this study focused on a rare condition, participants were offered the opportunity to review their transcript prior to analysis to ensure that they were satisfied with the level of pseudonymisation, to allow editing or removal as required.

Analysis

Analysis was completed by the PI and guided by Smith and Nizza (2022). The first stage of analysis involved taking each transcript in turn and developing exploratory notes and experiential statements (for an example see Appendix 2.6). This involved working through the transcript line by line and capturing thoughts and ideas in the exploratory notes. These notes were then used to formulate experiential statements, beginning to summarise key aspects of the transcript. Experiential statements were then used to develop connections between statements to generate themes for each participant's experience (see Appendix 2.7). This process was repeated for every transcript and produced tables of experiential themes for each participant, with corresponding quotations to hold the participant voice in mind at each stage of analysis. Cross-case analysis was then carried out, looking for similarities and differences between participants and considering themes across multiple accounts. Group experiential themes were then generated and are discussed in the results section.

Reflexivity

In conducting qualitative research, good practice guidelines (CORE-Q, Tong et al., 2007) highlight that researchers are unable to completely avoid personal bias due to their engagement with those participating in research and suggest that researchers should clarify their position in relation to participants and the research question. The PI in this study is a female trainee clinical psychologist. The PI had no previous relationship with any study participants. The PI has no children and has not accessed maternity or neonatal health services as a patient. The PI was working as a trainee clinical psychologist within maternity and neonatal services at the time of the study, including with women undergoing routine ultrasound screening. This was discussed during research supervision to attempt to reduce likelihood of this influencing the PI's interpretation of the data. The PI kept a reflective diary to enhance transparency of the process, including "bracketing off" of assumptions and preconceptions, and discussed this in research supervision to improve rigour of the process.

Ethics

This study was reviewed by the West of Scotland Research Ethics Committee 3 and ethical approval was granted by the committee (IRAS 309326) and by the NHS Greater Glasgow and Clyde Research and Innovation Department (GN22MH163). Correspondence is provided in Appendix 2.8.

Results

Four parents participated in the study. Relevant characteristics of parents are provided below in Table 3. Participants' names and children's names have been pseudonymised. All children discussed were female.

Table 3: Participant Characteristics

Parent	**Child**	**Child age (at time of interview)**	**Child had surgery**	**Child was first baby**
Amy	Erin	6	Yes	No
Ellen	Alice	8	No	Yes
Craig	Isla	6	Yes	Yes
Gemma	Olivia	8	Yes	No

Four group experiential themes were developed and are outlined in Table 4. The themes and related subthemes are discussed, with relevant quotations provided.

Table 4: Group Experiential Themes and Subthemes

Group Experiential Themes	**Subthemes**
Diagnosis changed the expected course of pregnancy.	No longer a "normal" scan.
	A move from generic services to specialist services.
	The emotional impact of diagnosis during pregnancy.
Searching for information about the condition.	Accessing information was challenging.
	Searching online.
	Meeting with specialist clinicians.
Making decisions about surgery.	Attitudes towards watching and waiting.
	Expectations about the role of a doctor.
	Approach to decision making.
Life after decision making.	Uncertainty about health does not end once the decision is made.
	Choosing to continue engagement with the clinic.

DIAGNOSIS CHANGED THE EXPECTED COURSE OF PREGNANCY

No longer a "normal" scan

All four participants described finding out about a problem with their baby's lungs during routine 20-week screening. Participants with older children (Amy and Gemma) were alerted to the presence of a potential problem by the actions of the sonographer, noticing differences from previous scans.

> *"So we went into the scan, she started scanning and then she asked me to empty my bladder and I was like "right there's something the matter, she's looking for something, I've seen enough scans, been to enough scans to know she's looking for something"."* (Amy)

The participants described being told the sonographer had identified something on the scan and that they would seek advice from a consultant obstetrician in the clinic. This took place on the same day for all participants although involved a period of waiting. This appeared to communicate to participants that something beyond the norm was identified and required additional expertise.

> "*…we got put into a room after the scan and we had to wait for the consultant."* (Craig)

At this stage, three participants described questioning the viability of the pregnancy: two raised by staff and one by the participant. Craig reported that they were told the pregnancy was not viable at the initial meeting with an obstetrician, whilst Gemma recalled being told there was the option to terminate her pregnancy.

> *"As [the obstetrician] explained it, Isla had these lesions on her lungs and the impression we were given was "yeah, it's not a viable pregnancy, you're not going to go anywhere", which was quite hard to take."* (Craig)

> *"… [the obstetrician] had said that we've got an option of {pause} basically getting rid of the baby."* (Gemma)

For Amy, questions about the viability of her pregnancy came to mind without staff prompting and she described leaving the clinic without having been able to discuss this worry. Amy was not provided with any pictures after her scan, which added to her distress.

> *"…we left there not knowing if she was going to die or if the pregnancy was going to continue… […] Cause I left there, and I thought "what if she… what if we were to lose her", we wouldn't even have a picture of her last being alive as such." (Amy)*

A move from generic to specialist services

Although consultant obstetricians were called upon to offer more expertise than sonographers, they did not have specialist knowledge of CPAM. Participants were subsequently offered appointments with specialists through the CPAM clinic who took the

lead on remaining antenatal care. For all participants, this brought about a loss of relationships developed over antenatal care to date.

> *"And they sent me then to specialist people who took over my pregnancy until Olivia was born."* (Gemma)

Amy and Gemma continued to access support in their local health board, although through different teams. For Ellen and Craig, accessing this specialist care was not possible in their local health boards, and as such they were referred to clinicians in Glasgow. This required travel to access specialist care and communicated further deviation from expected course of care.

> *"He said it was a lesion and he was going to send us over to Glasgow with specialists." (*Craig)

Participants also described the experience when generic services were no longer able to meet their needs but before they were linked in with specialist services. This left participants unsure of who to speak to about their concerns. The loneliness of this time was captured by Amy:

> *"Like the midwifes couldn't help you, they didn't know about that, like doctors couldn't help you cause they also never knew but your people that are meant to be your team there to help you, they weren't there... […] So, I found that probably the hardest part of that whole thing, just being alone.*" (Amy)

The emotional impact of diagnosis during pregnancy

The emotional impact of diagnosis was described differently by participants but was significant for all. Amy felt the diagnosis had "ruined" her pregnancy, and her reaction was like experiencing a loss.

> *"I never seen any family, I just stayed away from them. I got all the baby stuff out my house, I was like "take it away, cause I don't know what's going to happen"."* (Amy)

Craig described mixed emotions for his wife during pregnancy. Following previous discussions about viability of the pregnancy, ongoing anxiety led to frequent checking of the unborn baby's movements.

> *"I know certainly that my wife, she kept sort of poking and prodding her stomach to see if Isla was still alive, feeling the bump, feeling for kicks, so she had a really tough time... It was, it was a mixed bag of emotions the whole pregnancy."* (Craig)

Gemma reflected that pregnancy with a diagnosis was fundamentally different to her previous pregnancies. This comparison added to distress associated with the diagnosis itself.

> *"It was absolutely, it was heart wrenching to hear all the things that we were getting told. Do you know what I mean? It was the worst pregnancy in my life."* (Gemma)

However, Ellen described being able to continue to enjoy her pregnancy despite the diagnosis and felt it important to continue to do so. When considering advice for other families receiving a diagnosis, she recommended:

> *"Yeah, try and not let it, not like let it… detract from the magical nature of being pregnant. Because that would be awful if that was maybe somebody's only child and they were worrying about this the whole time instead of enjoying the fact that your body is doing an amazing thing, I would say."* (Ellen)

SEARCHING FOR INFORMATION ABOUT THE CONDITION

Accessing information was challenging

Participants described leaving their scan with little to no information about the diagnosis. Although they were told about the arrangements to see a specialist, they were left without information in the interim which led to feelings of confusion.

> *"So, we got nothing, we left there empty handed with a piece of paper with [the name of] this condition, with no information cause we never knew what this was, no leaflet which I think is really important to have."* (Amy)

> *"…like normally, if you go to the hospital and you get told your child's got this or that, you get all this information home with you. But we weren't given that. We literally were not given any information at all."* (Gemma)

When participants tried to do their own research, this could be complicated by the range of terms used to describe the condition, both by clinicians and online. Participants experienced a range of terminology in their appointments with clinicians which made it difficult to find relevant information.

> *"It was all words at the time, you try to read into it, but it was just so broad I think, the whole CPAM umbrella term was massive."* (Ellen)

Two participants (Gemma and Amy) described being explicitly told not to research the condition by clinicians before their specialist appointments. This warning away from information sources was not accompanied by reputable information to meet the needs of participants.

> *"…she went "have you ever heard of a thing called CPAM?" and I said "no". She said, "do not google it", that was the worst thing she ever said to me, "don't google it" because that was the first thing I done as soon as I left the hospital..."* (Gemma)

Searching online

All four participants turned to the internet for information, although two had been warned against it. At the time (between 2014 and 2016), information online was reported to be very limited. This was a barrier to accessing information about the condition.

> *"It has to be said, looking back at the stuff I read that's on the internet is limited in scope and probably a waste of space for the most part, so as I say I'm grateful I've got through the other side to see that it isn't as bad as it is online."* (Craig)

> *"…it was like American websites so there was no kind of UK websites which you're looking for, you're looking for that wee bit of, even like Great Ormond Street, I*

couldn't find anything there. I was going to the main places like NHS, Great Ormond Street, nothing. I couldn't find anything." (Amy)

The information that participants could find online was often found to be negative and could add to their concerns. When presented with a range of information, participants acknowledged that they could find themselves being drawn in by more negative information.

"I would be researching that. And you know google, you always find the worst-case scenarios..." (Amy)

"…cause google was telling me that 9 times out of 10 the baby doesn't survive…" (Gemma)

When limited information was available from formal sources, families looked to stories of others who had been through the process of diagnosis in the past. For Amy and Gemma, this was accessed through a national Facebook group for parents of babies with CPAM. Individual stories increased parents' understanding of the condition.

"I did find support through stories, saying "this is my wee CPAM baby, and this is how well they're doing" and like showing pictures of their scars, and some of them showed pictures of their babies when they were still in hospital which I... that helped me a wee bit to prepare." (Amy)

Meeting with specialist clinicians

Participants described their experience once they met with specialist clinicians from the CPAM clinic, ranging from a few days later (Ellen and Gemma) to two weeks after the scan (Craig and Amy). Accessing the clinic provided participants with information from clinicians with more expertise than those they had met during routine antenatal care and their 20-week scans. For Craig, the provision of information helped him to feel more grounded:

"And we did, we met with the three of them [2 surgeons and neonatologist] a number of times, as I say I can't fault them for giving us the information they did, and they

were good at trying to give me the right information and keep my hopes up, but not too over the top, at the same time as grounding me…" (Craig)

Having previously had questions about viability of pregnancy in some cases, and difficulties in accessing information, meeting clinicians with expertise in this area provided more accurate and balanced information for participants. This was reassuring to participants after days to weeks of uncertainty and anxiety.

"…and then that's kinda like where the scans all started happening, and then we met with [consultant neonatologist] who then put my mind at ease a lot better. He explained it a lot better…" (Amy)

Regular contact with specialist clinicians also helped Ellen to manage her worries during pregnancy, and she was reassured by regular scans and appointments:

"So, you just kinda went with it. And I think because you were having scans every week it was fine, because someone was looking at baby, and baby was alright." (Ellen)

MAKING DECISIONS ABOUT SURGERY

Attitudes towards watching and waiting

During the antenatal period, specialist clinicians introduced the parents to uncertainty regarding CPAM treatment. Participants were made aware of the plan to "watch and wait" antenatally, before deciding whether surgery is indicated to address the condition once the baby was born. This was explained by Ellen:

"I think obviously they give you the worst advice over if she becomes unwell and if she has chest infection after chest infection which, touch wood, she's never had one, then they'd have to review things and see how she is. So it's just again a kind of sit with the waiting and see what happens." (Ellen)

Participants varied in their perspectives regarding this period of watching and waiting into the postnatal period. For Amy, the thought of allowing uncertainty to continue into her daughter's childhood was intolerable.

> *"I just didn't want to be going into childhood and having that worry, I thought if we can get it away the now why no? Why have this worry for years and years to come? And then have to get surgery anyway?"* (Amy)

Ellen described being told about the possible outcomes and found that knowing what the plan would be in both situations reassuring. She appeared to have been able to tolerate the uncertainty and was accepting of this plan.

> *"Aye basically they gave us the two scenarios that maybe we'd be fine, and it would never cause them any problems, or they might need an operation. And I think those two, having been told that, obviously you hold onto the fact that hopefully nothing will ever happen, but if it does then this is what they do."* (Ellen)

For Gemma, being provided with varying possible outcomes appeared to have led to confusion. She described her interpretation of this information as clinicians changing their minds.

> *"I was getting quite annoyed with them because one minute we were getting told she needed this operation, then we were getting told we'll just leave it, then we were getting told we'll see about it. It was just constantly changing so I was getting dead annoyed…"* (Gemma)

Craig recalled that possible outcomes were explained to him, based on how his daughter would present at birth. He described struggling to remember his thoughts on this at the time and attributed this to the demands of travelling out of area to the appointments.

> *"But yeah, they did talk to us [about the different options]. I fear I probably, having driven two hours up and facing the two-hour drive down, an hour-long scan, some of the stuff I probably wasn't on the planet for listening to."* (Craig)

Expectations about the role of a doctor

Participants reflected on the experience of having a doctor introduce uncertainty and invite parental opinions when deciding on the treatment plan for their child. This was described by Amy who highlighted that there were challenges associated with being offered input in the process.

> *"So they just kinda took us through the part that it wasn't growing, they were on the fence, on whether they would operate, and it was down to us as parents...[…]It would have been easier had they said, "we are doing it" [surgery] or "no we're not doing it".*" (Amy)

Participants had expectations about the role of a doctor in decision making. Being included in discussions and asked for opinions as parents appeared to challenge these expectations. Gemma described this as:

> *"[Doctor] kept saying to me as well, "look what do you think should happen?" and I'm like "well you're the doctor, you tell me, I'm not medically trained for this, I don't know", [doctors should be] saying "listen this is the problem, this is what we're going to do..." ."* (Gemma)

Despite there being room for parental viewpoints, Ellen and Craig described relying on doctors in guiding decisions. This was the case in deciding to have surgery (Craig), and to not have surgery (Ellen), and there was reassurance offered in following the advice of doctors.

> *"…they tested her, they said "ok we're definitely going to need to do some operation here"."* (Craig)

> *"…people are keeping an eye on things, then put faith in them and trust in them to help you make the right decision and give you the right information."* (Ellen)

Approach to decision making

Parents described how they approached deciding whether their child would have surgery. Ellen was the only participant whose child did not have surgery. She described making the

decision based on her daughter being healthy at birth. She acknowledged openness to continue to monitor her daughter's health.

> *"… it's just if she has any chest infections or becomes unwell and breathing is an issue, then we'll have to review again."* (Ellen)

Amy recognised that there was more than one option available. She described inferring meaning from the ambiguity presented by clinicians. Amy took clinicians' openness to mean that surgery was a preferred option which influenced her decision.

> *"Cause if they thought... they wouldn't operate on a baby if they didn't need to, they wouldn't cut her open for no reason, so the fact they were on the fence, I was thinking "oh they think we should go for it then" as well."* (Amy)

Although her daughter was healthy at birth, Gemma described worries about deterioration in her daughter's condition in the future. This led to her wanting to pursue surgery for her daughter.

> *"They [CPAM clinic staff] kept saying "but she looks well just now". She might look well to you just now but tomorrow night that can change, do you know what I mean?"* (Gemma)

Similarly, Craig's daughter was healthy at birth. However, the perceived threat that she might be more prone to chest infections in the future led to the decision to pursue surgery.

> *"…the prognosis we were told at the time was that she might have more chest infections if she didn't have the operation."* (Craig)

LIFE AFTER DECISION MAKING

Uncertainty about health does not end once the decision has been made

Although decisions were made about surgery for all four participants' children in early childhood, they described levels of ongoing uncertainty about their children's health up to the present day. Participants described continued views of their children's vulnerability to lung conditions in childhood and during the recent coronavirus pandemic.

"We were so scared with covid." (Amy)

"I had never experienced or heard croup in my life before. And I took her into [the GP] like, she was feeding fine, and she was fine, but you had this diagnosis in the back of your head, and you think "gosh I maybe better go and see somebody about this." (Ellen)

Gemma shared that she had thought that the purpose of surgery would be to remove future risk of chest infections. When the surgeon explained to her that this was not a guarantee, she was surprised.

"[Surgeon] says to me "after the operation… after the operation this might not work, she might still get chest infections" and I'm like "so what did you do that for then?"" (Gemma)

Gemma also described worries that, although her daughter had surgery to remove the CPAM, it could grow back as she got older.

"It always has scared me, the worry that it'll come back, cause that's what they told us. That the lung can grow and when the lung grows back the CPAM can grow back." (Gemma)

Craig discussed his concerns regarding the prognosis for his daughter in the next phase of her life. He highlighted that conversations at the time focused on more immediate outcomes.

"Certainly going forward I don't know… the prognosis we were told at the time was that she might have more chest infections if she didn't have the operation, but what's the prognosis of, now she's had the operation, what are we thinking of she'll get in the future years, what do we need to watch out for, what are the signs we need?" (Craig)

Choosing to continue engagement with the clinic

Participants' children were healthy in the present day with limited need for ongoing review with the CPAM clinic. Despite this, they continued to engage with clinic follow-up for

research purposes. Participating in research was meaningful for participants in contributing to knowledge about CPAM.

> *"... they were really keen to keep in contact with us cause she would be their patient as such, until she was like an adult, basically she's research, she's a number, they need to know how she's doing and how she's getting on, obviously that's really important to parents in the future that's going to get diagnosed... […], so we are like really keen to stay in touch with them."* (Amy)

> *"I think we just keep going with it cause its, that's what we try and tell her, its research for people who aren't as able as her, there's people out there that are struggling with this and if we can gather some research and help other people out, then we should do that."* (Ellen)

For participants, ongoing engagement with follow up was a decision they continued to make. Craig highlighted that engaging in research is an ongoing decision which must be weighed up. As his daughter got older, he wondered whether continuing to go through lung function tests for research purposes was something that should continue.

> *"I don't know if we'll get that [lung function test for research], I don't know if I want to be pushing Isla for stuff that I don't have to."* (Craig)

Ellen also highlighted that her daughter was old enough to have her own views on clinic involvement. For Ellen's daughter, having a lung condition was something she perceived made her different to others and Ellen wondered how to approach this going forward. This suggested that there are potential downsides to engagement, along with the perceived value of engaging in research, for parents.

> *"Like she knows she's got something with her lung, which she's raging because [laughs] she doesn't really want to go and see anybody so... and she doesn't like being different. So, we don't really chat about it much to her, which I don't know if that's right or wrong, I don't know*." (Ellen)

Discussion

The current study explored the experience of parents who had been involved in decision making about their infant's care following the diagnosis of CPAM. Four themes were developed through the analysis and are discussed below in the context of existing literature. Study limitations and implications for clinical practice and future research are also discussed.

Key Findings

Participants in this study were asked to discuss their experiences of CPAM diagnosis and subsequent decision making, beginning with their experience of pregnancy and diagnosis, and working through the narrative chronologically. Subsequent generated themes were also structured around the chronological account of experiences described by participants. Participants described a fundamental change to the course of pregnancy following prenatal diagnosis, beginning during the ultrasound scan. This change to expected scanning procedures set the tone for the uncertainty of pregnancy with a diagnosis of CPAM for their unborn baby. The move towards specialist services indicated that this condition was beyond the clinical expertise of those providing routine antenatal care. In their systematic review of delivering diagnoses in prenatal medicine, Luz and colleagues (2017) highlighted that the quality of communication from sonographer to parents can influence the long-term consequences for parents of the diagnosis. The participants in this study highlighted that they experienced communication in both the spoken and unspoken.

The diagnosis of CPAM also introduced the inaccurate suggestion that the pregnancy may not be viable, or termination may be indicated. This is in keeping with Aite and colleagues' (2011b) study in which 50% of those with newly diagnosed CCAM were advised to terminate their pregnancy by obstetricians. This was despite termination not being indicated for the condition due to the likelihood of good outcomes. The authors attributed this recommendation to uncertainty in clinicians in generic services regarding CCAM and highlighted the distress this is likely to cause families unnecessarily. Similarly, participants in this study described increased distress in response to the diagnosis of CPAM due to the incorrect suggestion that termination may be indicated, or that their baby may not survive.

Parents interviewed described a range of emotional responses to the diagnosis. Aite and colleagues (2011) also described the experience of loss by parents following diagnosis of foetal abnormality, including loss of joy in pregnancy, loss of the dreamed about child, and loss of current understanding of the world. This was mirrored in findings in the present study,

in which participants described their diagnoses in terms of loss (Amy), loss of enjoyment of pregnancy (Gemma and Craig), and commitment to not lose this enjoyment (Ellen).

Participants described their attempts to find information and understand more about their unborn baby's diagnosis through clinicians, online resources, and the stories of others who had been through the diagnosis. Learning about a diagnosis of foetal abnormality is typically a shock to women. Whilst learning about this diagnosis, they are then presented with complex and sometimes incomplete information (Tucker and Christian, 2022). Whilst women may think they have received sufficient information at the time, they may later feel confused and in need of further information (Asplin et al., 2012). A novel finding from this study specific to CPAM diagnosis was the confusion that was added to the information-seeking phase by the various conditions that fall under the umbrella term of CPAM. This made it difficult for participants to access information relevant to their baby's specific condition.

Another interesting finding was that two participants reported the significance of being told not to research the condition online ("do not google it"), although they proceeded to do so. The reasons behind this from clinicians' perspectives are unknown. Providing information whilst dissuading parents from looking into it appeared to have an unsettling effect and ultimately did not prevent parents from looking to the internet for information. Asplin and colleagues (2012) have suggested that a long wait between initial diagnosis and subsequent appointment with specialists makes it more likely that women will look to the internet for more information. Ensuring a quick allocation to a specialist clinician may therefore be more effective in reducing online searching than discouraging parents from doing so. With a wait of a few days to weeks to speak to specialists, participants in this study had looked online for information, with one (Amy) doing so whilst still in the hospital after receiving the initial diagnosis. As such, providing helpful information in a timely fashion is recommended.

When considering what information is likely to be most helpful, participants in this study discussed the support offered by specialist clinicians through the CPAM clinic. This appeared to provide reassurance for families and expertise that was not available online or through generic services. Appointments with specialist staff have been shown to reduce the anxiety experienced by parents following diagnosis of a foetal abnormality (Aite et al., 2009), and parents have been shown to value surgical expertise in allaying fears that come about due to lack of information or misinformation provided during ultrasound scans (Statham et al., 2000).

Parents responded differently in their tolerance of uncertainty in the prenatal period and in their approach to "watching and waiting" as the baby got older. Some participants found it difficult to tolerate this uncertainty, whilst others reported feeling more accepting. Uncertainty appeared to have relevance to how participants coped with the diagnosis. Those more accepting of the uncertainty described their understanding of possible outcomes (for example Ellen's description of "the two scenarios") whilst those less accepting felt less clear of the plan (for example Amy's "and then have to get surgery anyway").

Expectations regarding the role of doctor and patient, or parent, were also discussed. For some, the idea that parents could have a say in decisions was challenging, and others continued to take the lead from clinicians or make assumptions about underlying preferences based on ambiguity. In their systematic review, Wilpers and colleagues (2021) demonstrated that parents feel helpless during their experience of prenatal care following diagnosis and may feel unable to "do their job" of being a parent. As such, they look to healthcare professionals to guide their decision making and to validate their choices. This appeared to be the case in this study. Even when clinicians were non-directive in the case of Amy's baby, she reported taking meaning from this and being guided by her perception of what doctors thought.

Parents varied in what factors motivated them to make decisions about surgery for their infant. Whilst all babies were healthy at birth, parents responded differently to this. For example, Ellen found this reassuring and was accepting of not pursuing surgery. For Craig, Amy, and Gemma, this was not reassuring, and the threat of future illness was a more pressing factor which ultimately led to the decision to have surgery. Despite the decision having been made about surgery for the participants' children, they all described ongoing uncertainty about their child's health when faced with typical childhood illnesses and with the recent coronavirus pandemic.

Parents in this study showed awareness of the lack of available long-term data on infants born with CPAM and felt it important to contribute to this evidence base for other families. Parents have been shown to value and find meaning in participating in research after experiencing prenatal diagnosis (Wilpers et al., 2021). Whilst parents placed value on this involvement, two participants (Ellen and Craig) discussed the recent need to consider their daughters' views on ongoing involvement with the clinic.

Limitations

Findings of this study are based on the account of four parents who received antenatal diagnosis of CPAM between 2014 and 2016. These interviews were analysed using IPA to generate rich accounts of the experience. The focus of IPA is on telling the stories of participants, rather than generating broader theories with generalisability to the wider population. As such, whilst these findings can offer some insight into the experience of these participants, caution should be exercised before drawing conclusions about wider populations. However, this study may serve as a preliminary investigation and inform future research with parents going through similar experiences.

Although the population of targeted participants covered a wide age range, respondents had children between six and eight years old. This added some strength to the study, as IPA recommends having a homogenous sample where possible. However, time had passed between the experience, including subsequent decision making, and the research interview. This may have had an impact on participants' recall of discussions with staff and their views on decision making at the time. Accuracy of recall of the event may have been influenced by the length of time between the experience and the interview, with parents looking back on an experience which happened years in the past. The semi-structured interview encouraged families to tell their story of CPAM diagnosis and subsequent decision making. This is a story that families are likely to have retold and reappraised over time, either with staff in the CPAM clinic, with family and friends, or to themselves. This retelling of the story may have influenced the narrative they developed about the experience. Asking participants to present their account chronologically may also have influenced the themes identified and it is acknowledged that participants may not have spontaneously presented their experiences in this way.

Implications for Future Research

Future research in which families are targeted prospectively whilst going through the diagnosis and decision-making process may enhance understanding of the experience. Participants in this study may have become more assured of their decision over time and, as such, may have reported reduced uncertainty compared to those going through the process contemporaneously. Participants described the factors which led to their decision making (i.e., the reassurance offered by a healthy child at birth versus anxiety about the potential for illness in the future). As far as the researcher is aware, there has not been previous research

into why parents of babies with CPAM make the decisions they do regarding surgery. Further research in this area may be beneficial for informing further service provision.

Parents in this study were selected due to some uncertainty at the time of diagnosis about the best course of treatment for their unborn infant and their decision-making taking place within the "zone of parental discretion" described by Krick and colleagues (2020), with scope for parents to incorporate their family values and viewpoints in the process. Despite this, parents in this study did not describe incorporating their perspectives or values into the decision-making process. When they were invited to provide their perspectives, parents often described discomfort and preferred a position in which clinicians were the key decision makers regarding treatment for CPAM. This clinician-led approach is more in keeping with the "best interest standard" of decision making. This is of note as clinicians within the CPAM clinic value involving parents in decisions about their infant's treatment and hope to increase this shared approach to decision making. Further research examining how parents respond to shared decision making in the zone of parental discretion, concerns they may have about having a say in decision making, barriers to involving parents, and considering ways to increase parental involvement, may be beneficial in shaping service-level approaches to these issues.

Implications for Clinical Practice

As noted previously, participants in this study first accessed services between 2014 and 2016. Although recommendations are made for clinical practice based on their accounts, it is acknowledged that services may have already made developments since this time and online resources have improved.

Interactions with staff were key aspects of the accounts of parents, both with staff in generic services when attending for routine ultrasound, and with specialist services through onward referral following the initial diagnosis of a lung problem. Healthcare staff should be mindful that they communicate in the spoken and unspoken with families, and where possible be explicit and transparent in their thought process. At each stage in the process, clinicians should be mindful of the finding that questions about viability of pregnancy are raised at the point of diagnosis. Those providing diagnosis and consultation through generic services may benefit from further training on the good outcomes for those diagnosed with CPAM to reduce likelihood of recommending termination or questioning viability of pregnancy. Specialists in

the CPAM clinic should be aware of the possibility that these discussions have taken place in generic services, and act quickly to provide accurate information to reduce parental distress.

Specialists introducing shared decision making should consider the potential discomfort this could cause families and be mindful that parents may look for meaning in their ambiguity. At all timepoints, staff should be aware of the likelihood that parents will look to the internet for further information. Rather than discourage them from doing so, which may shut down their confidence in discussing what they have read, clinicians should create a space in which parents can discuss what they have read, correcting misinformation if required.

Involving specialist staff as early as possible may help to reduce the uncertainty and distress experienced by parents. Parents spoke of the perceived benefits of having a named contact throughout the remaining pregnancy and postnatally. Within NHS GGC, cardiac liaison nurses offer a similar role for families whose infant is diagnosed with cardiac conditions prenatally, and a similar role may be of benefit in this case for infants with lung conditions. In Scotland, the recent expansion of Maternity and Neonatal Psychological Interventions (MNPI) services nationally (Scottish Government, 2023) has led to increased access to clinical psychology staff within maternity and neonatal settings. Whilst all families receiving diagnosis of CPAM may not require individual therapeutic work with a psychologist, these staff can also support clinicians working with families to consider how to communicate the diagnosis in a psychologically informed way, to reduce potential distress. They may also offer consultation to staff working with families to consider when distress reaches a threshold requiring psychological intervention and to make recommendations for helpful interventions. This MNPI provision was not widely available at the time to the parents who participated in this study, however, may facilitate additional support during a time of uncertainty to families receiving similar diagnoses in the present day.

Conclusions

This study explored in-depth accounts of the experience of parents who had received a diagnosis of CPAM antenatally for their unborn baby, and their involvement in decision making about their infant's care. The findings provide an insight into experiences during pregnancy and into childhood, with participants highlighting the change from expected pregnancy, their subsequent search for information to better understand the condition, the process of making decisions about surgery, and the impact this continued to have on their

lives in the present day. Findings demonstrate areas of challenge for families, and where clinicians may be able to support families during this uncertain time. It is hoped that the findings of this study can inform future research in this evolving area, and support service development as the needs of this population are considered.

References

Aite, L., Zaccara, A., Mirante, N., Nahom, A., Trucchi, A., Capolupo, I., & Bagolan, P. (2011). Antenatal diagnosis of congenital anomaly: A really traumatic experience. *Journal of Perinatology*, *31*(12), 760–763. https://doi.org/10.1038/jp.2011.22

Aite, L., Zaccara, A., Trucchi, A., Brizzi, C., Nahom, A., Iacobelli, B., Capolupo, I., & Bagolan, P. (2009). When uncertainty generates more anxiety than severity: The prenatal experience with cystic adenomatoid malformation of the lung. *Journal of Perinatal Medicine*, *37*(5), 539–542. https://doi.org/10.1515/JPM.2009.098

Aite, L., Zaccara, A., Trucchi, A., Nahom, A., Capolupo, I., Mobili, L., & Bagolan, P. (2011b). Is counselling for CCAM that difficult? Learning from parental experience. *Journal of prenatal medicine*, *5*(3), 65–68.

Asplin, N., Wessel, H., Marions, L., & Georgsson Öhman, S. (2012). Pregnant women's experiences, needs, and preferences regarding information about malformations detected by ultrasound scan. *Sexual and Reproductive Healthcare*, *3*(2), 73–78. https://doi.org/10.1016/j.srhc.2011.12.002

Berman, L., Jackson, J., Miller, K., Kowalski, R., Kolm, P., & Luks, F. I. (2018). Expert surgical consensus for prenatal counseling using the Delphi method. *Journal of Pediatric Surgery*, *53*(8), 1592–1599. https://doi.org/10.1016/j.jpedsurg.2017.11.056

British Lung Foundation. (2022). *Congenital pulmonary airway malformation (CPAM)*. Asthma + Lung UK. https://www.blf.org.uk/support-for-you/congenital-pulmonary-airway-malformation-cpam

Cole, J. C. M., Moldenhauer, J. S., Berger, K., Cary, M. S., Smith, H., Martino, V., Rendon, N., & Howell, L. J. (2016). Identifying expectant parents at risk for psychological distress in response to a confirmed fetal abnormality. *Archives of Women's Mental Health*, *19*(3), 443–453. https://doi.org/10.1007/s00737-015-0580-6

Krick, J. A., Hogue, J. S., Reese, T. R., & Studer, M. A. (2020). Uncertainty: An Uncomfortable companion to decision-making for infants. *Pediatrics*, *146*(August 2020), S14–S17. https://doi.org/10.1542/peds.2020-0818E

Lalor, J. G., Devane, D., & Begley, C. M. (2007). Unexpected diagnosis of fetal abnormality: Women's encounters with caregivers. *Birth*, *34*(1), 80–88. https://doi.org/10.1111/j.1523-536X.2006.00148.x

Leithner, K., Maar, A., Fischer-Kern, M., Hilger, E., Löffler-Stastka, H., & Ponocny-Seliger, E. (2004). Affective state of women following a prenatal diagnosis: Predictors of a negative psychological outcome. *Ultrasound in Obstetrics and Gynecology*, *23*(3), 240–246. https://doi.org/10.1002/uog.978

Luz, R., George, A., Spitz, E., & Vieux, R. (2017). Breaking bad news in prenatal medicine: a literature review. *Journal of Reproductive and Infant Psychology*, *35*(1), 14–31. https://doi.org/10.1080/02646838.2016.1253052

Mehta, P. A., & Sharma, G. (2022). *Congenital Pulmonary Airway Malformation*. National Center for Biotechnology Information. https://www.ncbi.nlm.nih.gov/books/NBK551664/

Morini, F., Zani, A., Conforti, A., Van Heurn, E., Eaton, S., Puri, P., Rintala, R., Lukac, M., Kuebler, J. F., Friedmacher, F., Wijnen, R., Tovar, J. A., Pierro, A., & Bagolan, P. (2018). Current Management of Congenital Pulmonary Airway Malformations: A European Pediatric Surgeons' Association Survey. *European Journal of Pediatric Surgery*, *28*(1), 1–5. https://doi.org/10.1055/s-0037-1604020

Peters, R. T., Burge, D. M., & Marven, S. S. (2013). Congenital lung malformations: An ongoing controversy. *Annals of the Royal College of Surgeons of England*, *95*(2), 144–147. https://doi.org/10.1308/003588412X13373405387735

Public Health Scotland. (2021). *Congenital Anomalies in Scotland (2000-2019)*. Public Health Scotland. https://publichealthscotland.scot/media/9504/2021-10-05-congenitalanomaliesinscotland2019-main-report.pdf

Scottish Government (2023) *Perinatal and Infant Mental Health Services: Update*, *Scottish Government*. The Scottish Government. https://www.gov.scot/publications/perinatal-infant-mental-health-services-update/documents/.

Smith, J. A., Flowers, P., & Larkin, M. (2009). *Interpretative Phenomenological Analysis: Theory, Method and Research* (2nd ed.). SAGE Publications.

Smith, J. A. & Nizza, I. E. (2022) *Essentials of Interpretative Phenomenological Analysis.* American Psychological Association.

Stanton, M., Njere, I., Ade-Ajayi, N., Patel, S., & Davenport, M. (2009). Systematic review and meta-analysis of the postnatal management of congenital cystic lung lesions. *Journal of Pediatric Surgery*, *44*(5), 1027–1033. https://doi.org/10.1016/j.jpedsurg.2008.10.118

Statham, H., Solomou, W., & Chitty, L. (2000). Prenatal diagnosis of fetal abnormality: Psychological effects on women in low-risk pregnancies. *Bailliere's Best Practice and Research in Clinical Obstetrics and Gynaecology*, *14*(4), 731–747. https://doi.org/10.1053/beog.2000.0108

Sullivan, A., & Cummings, C. (2020). Shared decision making in the NICU. *NeoReviews*, *21*(4), e217–e225. https://doi.org/10.1542/neo.21-4-e217

Tong, A., Sainsbury, P., & Craig, J. (2007). Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *International journal for quality in health care*, *19*(6), 349-357.

Tucker, J. A., & Christian, B. J. (2022). Maternal Distress in Disclosing a Nonlethal Fetal Anomaly Diagnosis To Family and Friends. *The American Journal of Maternal/Child Nursing*, *47*(4), 220–226. https://doi.org/10.1097/NMC.0000000000000829

Wilpers, A., Bahtiyar, M. O., Stitelman, D., Batten, J., Calix, R. X., Chase, V., Yung, N., Maassel, N., & Novick, G. (2021). The parental journey of fetal care: a systematic review and meta synthesis. *American Journal of Obstetrics and Gynecology MFM*, *3*(3), 100320. https://doi.org/10.1016/j.ajogmf.2021.100320

Appendices

Systematic Review

Appendix 1.1: Example Search Strategy – PsycInfo

Appendix 1.2: Data Extraction Template

Appendix 1.3: Joanna Briggs Institute Checklist for Quasi-Experimental Studies

Appendix 1.4: Quality appraisal tool items (JBI Quasi-Experimental Checklist)

Major Research Project

Appendix 2.1: Recruitment Letter

Appendix 2.2: Participant Information Sheet

Appendix 2.3: Consent Form

Appendix 2.4: Interview Schedule

Appendix 2.5: Debrief Document

Appendix 2.6: Exploratory Notes and Experiential Statements (Extract)

Appendix 2.7: Participant Experiential Themes (Extract)

Appendix 2.8: REC Correspondence

Appendix 2.9: Final Approved MRP Proposal

Appendix 1.1: Example Search Strategy – PsycInfo

Search terms were adapted according to the database searched. An example from one database is provided below.

#	Query	Results
S1	DE “Affective Disorders”	15,491
S2	DE “Anxiety”	92,681
S3	DE “Anxiety Disorders” OR DE “Generalized Anxiety Disorder” OR DE “Obsessive Compulsive Disorder” OR DE “Panic Disorder” PR DE “Phobias” OR DE “Separation Anxiety Disorder” OR DE “Trichotillomania”	62,799
S4	DE "Major Depression" OR DE "Postpartum Depression" OR DE "Reactive Depression" OR DE "Depression (Emotion)" OR DE "Internalizing Symptoms"	173,761
S5	DE “Mental Health”	90,512
S6	DE "Stress and Trauma Related Disorders" OR DE "Acute Stress Disorder" OR DE "Adjustment Disorders" OR DE "Attachment Disorders" OR DE "Posttraumatic Stress Disorder" OR DE "Prolonged Grief Disorder"	41,937
S7	TI (adjustment or anxiet* or anxious or coping or depress* or distress* or grief or "mental health" or mood or panic* or psychiatr* or psychological* or psychology or psychosocial or traum* or OCD or "obsessive compulsive") OR AB (adjustment or anxiet* or anxious or coping or depress* or distress* or grief or "mental health" or mood or panic* or psychiatr* or psychological* or psychology or psychosocial or traum* or OCD or "obsessive compulsive")	1,506,193
S8	S1 OR S2 OR S3 OR S4 OR S5 OR S6 OR S7	1,535,301
S9	DE "Congenital Disorders"	3,419
S10	DE "Prenatal Diagnosis"	1,225
S11	TI ("congenital anomal*" or (foetal N2 anomal*) or (fetal N2 anomal*) or (structural N2 anomal*)) OR AB ("congenital anomal*" or (foetal N2 anomal*) or (fetal N2 anomal*) or (structural N2 anomal*))	892
S12	TI ("congenital malformation" or "fetal malformation" or "foetal malformation") OR AB ("congenital malformation" or "fetal malformation" or "foetal malformation")	180
S13	TI ("congenital abnormal*" or (foetal N2 abnormal*) or (fetal N2 abnormal*)) OR AB ("congenital abnormal*" or (foetal N2 abnormal*) or (fetal N2 abnormal*))	455
S14	TI ((antenatal* N2 diagnos*) or (prenatal* N2 diagnos*) or (abnormal* N2 ultraso*) or (abnormal* N2 sonogra*) or "unexpected finding*")) OR AB ((antenatal* N2 diagnos*) or (prenatal* N2 diagnos*) or (abnormal* N2 ultraso*) or (abnormal* N2 sonogra*) or "unexpected finding*"))	1,917
S15	S9 OR S10 OR S11 OR S12 OR S13 OR S14	7,070
S16	DE "Antepartum Period"	394
S17	DE "Expectant Mothers"	927
S18	DE "Expectant Parents"	216
S19	DE "Perinatal Period"	3,743
S20	DE “Postnatal Period”	6,009
S21	DE “Prenatal Care”	3,846
S22	DE "Pregnancy"	50,067
S23	TI (antenatal* or antepart* or (before N2 birth) or matern* or perinatal* or peri-natal* or peripartum or peri- partum or postnatal* or post-natal* or postpartum or post-partum or pregnan* or prenatal* or pre-natal* or prepartum or pre-partum) OR AB (antenatal* or antepart* or (before N2 birth) or matern* or perinatal* or peri-natal* or peripartum or peri-partum or	136,195

	postnatal* or post-natal* or postpartum or post- partum or pregnan* or prenatal* or pre-natal* or prepartum or pre-partum)	
S24	S16 OR S17 OR S18 OR S19 OR S20 OR S21 OR S22 OR S23	145,780
S25	DE "Quantitative Methods"	3,847
S26	((DE "General Health Questionnaire" OR DE "Screening Tests") OR (DE "Psychological Screening Inventory")) OR (DE "State Trait Anxiety Inventory")	9,209
S27	TI (quantitative* or questionnaire* or measure or inventory or longitudinal or "cross- sectional" or "cross sectional" or "screening tool*" or "prospective observational" or "impact of events scale" or IES or "edinburgh postnatal depression scale" or EPDS or "general health questionnaire" or "ghq" or "state trait anxiety inventory" or STAI) OR AB (quantitative* or questionnaire* or measure or inventory or longitudinal or "cross- sectional" or "cross sectional" or "screening tool*" or "prospective observational" or "impact of events scale" or IES or "edinburgh postnatal depression scale" or EPDS or "general health questionnaire" or "ghq" or "state trait anxiety inventory" or STAI)	1,087,347
S28	S25 OR S26 OR S27	1,091,331
S29	S8 AND S15 AND S24 AND S28	297

Appendix 1.2: Data Extraction Template

General Information			
Researcher performing data extraction			
Date of data extraction			
Features of the study			
Record number			
Author			
Article title			
Journal		Year	
Country of origin			
Source of funding			

Study Characteristics	
Aim/objectives of the study	
Study design	
Study inclusion and exclusion criteria	
Recruitment procedures used (e.g., details of randomisation, blinding)	

Participant Characteristics	
Age	
Ethnicity	
Socio-economic status	
Number of participants at outset	
Scanning Details	
Type of scan	
Stage of pregnancy scan administered	
Setting where scan administered	
Anomaly Details	

Anomaly/anomalies identified	
Number of participants in each group	
Prognosis of diagnosis	
Details of any controls or comparisons	
Control group characteristics	
Number of participants in each group	

Outcome Data/Results	
Mental health outcome(s) assessed	
For each mental health outcome	
Definition used in study	
Measurement tool or method used	
Length of follow up, number and/or times of follow-up measurements	
For all intervention group(s) and control group(s)	
Number of participants enrolled	
Number of participants included in analysis	
Number of withdrawals, exclusions, lost to follow-up	
Summary outcome data	*e.g., Dichotomous: number of events, number of participants* *Continuous: mean and standard deviation*
Type of analysis used in study	*e.g., intention to treat, per protocol*
Results of study analysis	*e.g., Dichotomous: odds ratio, risk ratio and confidence intervals, p values* *Continuous: mean difference, confidence intervals*
Additional outcomes	
Costs	
Resource use	

Adverse events	

Study Conclusions	
Conclusions of study by author	
Limitations of study by author	
Limitation of study by reviewer	
Quality assessment - reviewer	

Notes

Appendix 1.3: Joanna Briggs Institute Checklist for Quasi-Experimental Studies

JBI Critical Appraisal Checklist for Quasi-Experimental Studies (non-randomized experimental studies)

Reviewer_____Date_____

Author _____Year_____Record Number_____

		Yes	No	Unclear	Not applicable
1.	Is it clear in the study what is the 'cause' and what is the 'effect' (i.e. there is no confusion about which variable comes first)?	☐	☐	☐	☐
2.	Were the participants included in any comparisons similar?	☐	☐	☐	☐
3.	Were the participants included in any comparisons receiving similar treatment/care, other than the exposure or intervention of interest?	☐	☐	☐	☐
4.	Was there a control group?	☐	☐	☐	☐
5.	Were there multiple measurements of the outcome both pre and post the intervention/exposure?	☐	☐	☐	☐
6.	Was follow up complete and if not, were differences between groups in terms of their follow up adequately described and analyzed?	☐	☐	☐	☐
7.	Were the outcomes of participants included in any comparisons measured in the same way?	☐	☐	☐	☐
8.	Were outcomes measured in a reliable way?	☐	☐	☐	☐
9.	Was appropriate statistical analysis used?	☐	☐	☐	☐

Overall appraisal: Include ☐ Exclude ☐ Seek further info ☐

Comments (Including reason for exclusion)

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_____

_____

Appendix 1.4: Quality Appraisal Tool Items (JBI Quasi-Experimental Checklist)

#		**Q1**	**Q2**	**Q3**	**Q4**	**Q5**	**Q6**	**Q7**	**Q8**	**Q9**	**TOTAL**
1*	Aite et al., 2009	✓	✓	✓	✓	X	✓	✓	✓	✗	7/9
2	Asplin et al., 2015	✓	✓	NA	✗	✗	✗	✓	✓	✓	5/8
3	Cole et al., 2016	✓	?	?	✗	✗	NA	✗	✓	✓	3/8
4	Helbig et al., 2011	✓	?	✓	✓	✗	NA	✓	✓	✓	6/8
5	Kaasen et al., 2010	✓	✗	✓	✓	✗	NA	✓	✓	✓	6/8
6	Kaasen et al., 2017	✓	✗	✓	✓	✗	✓	✓	✓	✓	7/9
7	Kemp et al., 1998	✓	✗	✓	✓	✗	NA	✓	✓	✗	5/8
8	Kim et al., 2021	✓	?	?	✗	✗	?	✓	✓	✗	3/9
9	Oftedal et al., 2022	✓	✗	✓	✓	✗	✓	✓	✓	✓	7/9
10*	Roslan et al., 2021	✓	✓	✓	✓	✓	✓	✓	✓	✓	9/9
11*	Rychik et al., 2013	✓	?	?	✗	✗	NA	?	✓	✓	3/8
12	Titapant et al., 2015	✓	✓	NA	✗	✗	✓	✓	✓	✓	6/8
13*	Wilpers et al., 2017	✓	✓	✓	✓	✗	NA	✓	✓	✗	6/8

✓ - yes
✗ - no
? - unclear
NA – not applicable

* Indicates assessed by co-rater

Appendix 2.1: Recruitment Letter

Available for download at: https://osf.io/35cmn

Appendix 2.2: Participant Information Sheet

Available for download at: https://osf.io/mwtrx

Appendix 2.3: Consent Form

Available for download at: https://osf.io/zsvkh

Appendix 2.4: Interview Schedule

Available for download at: https://osf.io/sekuz

Appendix 2.5: Debrief Document

Available for download at: https://osf.io/arwyh

Appendix 2.6: Exploratory Notes and Experiential Statements (Extract)

Experiential Statement		**Original Transcript** I: Interviewer P: Participant	**Exploratory Notes** descriptive linguistic *conceptual*
Don't google it. Given a label on a piece of paper and sent home alone.	178 179 180 181 182 183 184 185 186 187 188 189 190 191 192 193 194	He did mention CPAM, said that was a newer name they were coming up with to cover the wider range of things, but then it got wrote on a piece of paper. He says "I'll write it for you, write it on a piece of paper for you and you can google it if you want. I would advise you not to but I know parents like to so I'll write it" so that's what he gave and we left. So I've no pictures, I've got no scan pictures cause, I don't know if Claire thought we were going to get them off the doctor, or the doctor thought Claire… so we got nothing, we left there empty handed with a piece of paper with this condition, with no information cause we never knew what this was, no leaflet which I think is really important to have. We actually done fundraising for the rare diseases [department]. We asked for something like… I think it was over £5000 for, at least print a leaflet and give it to these parents because, there's nothing, there was no website, there was no information, we left there not knowing if she was going to die or if the pregnancy was going to continue, we just didn't know what the future was going to hold,	Described it as CPAM – *was this confusing for family? Change in terms over the course of a day.* Doctor wrote CPAM on paper for family to take away and google, but also advised not to google. *Inconsistency, confusing contradiction? Not providing any alternative information to what is found on google.* "I've got no scan pictures" – *further deviation from "normal" pregnancy/ procedures.* Family later raised money to provide information on the condition. *Meaning making from their experiences?* Suggestion that leaflet would be helpful to take information away.

Questions about viability of pregnancy. Hardest part was feeling alone without contact from professionals.	195 196 197 198 199 200 201 202 203 204	and then it was 6 days before we got contacted again, so we had nothing from nobody. No contact, no checking in, no like counsellor option, nobody whatsoever, no phone numbers and we just left the hospital and that was us for 6 days. And then the appointment again wasn't for a further week, so it was actually 2 weeks before we had seen anybody in person to speak to this. So, I found that probably the hardest part of that whole thing, just being alone. **I:** Yeah, feeling alone in that. **P:** Uh huh. Definitely.	"Not knowing if she was going to die or if the pregnancy was going to continue". Left appointment with uncertainty and questions. 6 days before professionals made contact again. 2 weeks before another appointment. *Isolated? Not given any information or support, just name to google.* "The hardest part… being alone".

Appendix 2.7: Participant Experiential Themes (Extract)

Theme 3: Looking to specialists for information and support		
Doctors in generic services didn't understand the rare condition.	2.48	*I found myself at A and E quite a few times, but they never knew what it was.*
Struggled to find people who can help or who understood; waited for appointment with specialist team.	9.264	*…at that point we hadn't been appointed to like Dr [redacted], we weren't put to him yet, so we were just left alone for two weeks and I think its quite important to maybe be like contacted the following day or later that day or whatever or within a day or two, contacted quite quickly so that you can get like that bit of support that you're looking for.*
Generic services need more information through training	30.890	*These professionals knowing a wee bit more about it. I dunno if there's a training course or… I don't know, I don't know how they work that but maybe just having these professionals having more information to give to the parents.*
CPAM is less well known than other conditions	23.681	*If a child gets diagnosed with, say Down Syndrome or other like medical conditions, there is a lot [of information available].*
Lack of continuity of care from clinicians led to parents having more expertise in some areas. – *Responsibility?*	10.295	*I know you can't always make it the same person, but I had to tell one of the person that was doing the scan how to measure it, which I don't think was very professional either.*

Appendix 2.9: Final Approved MRP Proposal

Available for download at: https://osf.io/ejrqs