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# Understanding Parental Challenges in the Neonatal Intensive Care Unit: Psychological Support and Decision Making

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# Chapter 1: Systematic Review

## Reducing parental distress in the neonatal intensive care unit: a systematic review of psychological interventions

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# Abstract

Background: An infant's admission to the Neonatal Intensive Care Unit (NICU) is a distressing experience for parents. The NICU environment, their infant's physical health and the alteration to the parental role can leave parents with feelings of fear, powerlessness and guilt, causing symptoms of depression, anxiety and trauma. Parental distress in a child's early years can have significant developmental consequences. There are various interventions recommended for reducing parental distress in the NICU. Several studies investigating different psychological interventions have been published in recent years, however there is no standardised approach for this population. This review aimed to systematically search and synthesise studies investigating the effectiveness of psychological interventions in reducing parental distress in the NICU, addressing which approaches are most effective in alleviating anxiety, PTSD, stress and depression.

Methods: A systematic search of four electronic databases (MEDLINE, EMBASE, PsychINFO and CINAHL) was carried out to identify studies published before November 2024. A narrative synthesis of findings was produced. Included studies were randomised control trials investigating psychological interventions aimed at reducing various measures of parental distress. Risk of bias was assessed using the RoB2.

Results: A total of 3388 records were screened and 15 (n=1087) were included for review. A narrative synthesis of findings was produced according to the therapeutic approach to intervention: cognitive behavioural; attachment focused; non-directive/alternative. Cognitive behavioural approaches were the most effective at reducing symptoms of depression, anxiety and post-traumatic stress. Attachment focused interventions did not consistently show reductions in distress and there was a small effect for non-directive/alternative approaches.

Conclusions: This review provides a more nuanced understanding of which approaches are most effective at reducing parental distress, expanding the findings of previous reviews in this area. Future research should prioritise the inclusion of fathers and parents of full-term infants. Structured, brief, group-based and individual cognitive behavioural interventions are a promising and cost-effective way for reducing parental distress and should be utilised in clinical settings.

# Introduction

Around 100,000 infants born in the UK each year are admitted to the neonatal intensive care unit (NICU); equating to one in seven infants born (NDAU, 2016). This is due to factors including prematurity, congenital abnormalities, respiratory difficulties and infection with admission length ranging significantly, according to severity and gestation (Battersby et al., 2017). Admission to the NICU is often unexpected, leaving parents to navigate considerable uncertainty during an already challenging transition to parenthood following birth.

Parents of infants admitted to the NICU commonly experience heightened psychological distress (Al Maghaireh et al., 2016; Malouf et al., 2020).  The clinical setting of the NICU, characterised by medical equipment, healthcare staff and continuous noise, exacerbates the sense of physical and psychological separation parents feel from their infant (Treherne et al., 2017). Parents frequently express concern about their infant's physical appearance, including their colour, size, and fragility (Sweet & Mannix, 2012; Govindaswamy et al., 2019). Furthermore, the NICU environment often prevents parents from immediately participating in their child's care (Ioni et al., 2016), leaving many feeling unable to fulfil their parental role (Montirosso et al., 2012). These challenges leave parents processing difficult feelings such as powerlessness, fear, loss and guilt (Watson, 2011). Review studies consistently identify these feelings as some of the primary sources of parental distress (Govindaswamy et al., 2019).

Although this response may be considered a normative reaction under the circumstances, emotional difficulties can persist beyond hospitalisation. There is a significantly higher rate of post-partum depression among parents of infants admitted to the NICU, with almost 40% meeting the criteria (Lefkowitz et al., 2010) compared with 10-15% in the general population (Putnam et al, 2015). Additionally, Malouf and colleagues' systematic review analysed results from across the globe (N=6036) and identified a rate of anxiety and post-traumatic stress of approximately 40% for NICU parents one month postpartum. Although this rate declined to around 25% over the first year following birth, post-traumatic stress remained notably high beyond one year, persisting at 27% (Malouf et al., 2022). This is considerably higher than rates of anxiety among mothers in the perinatal period (20%) (Fawcett et al., 2019) and women in the general population (5-9%) (Remes et al., 2016). Postnatal mental health difficulties are linked to various emotional, behavioural and cognitive outcomes in children (Rodgers et a., 2020). These findings show the complex interplay of psychosocial factors in shaping parental mental health outcomes in the NICU, which can have consequences for infant development (Sinclair & Murray, 1998; Beck, 1995; Grunberg et al., 2019). While many factors can contribute to parental distress, a UK study involving parents of children admitted to paediatric intensive care found that psychological factors may have a stronger impact than demographic variables (such as ethnicity) or illness-related aspects (like length of hospital stay or severity of the illness; Colville & Pierce, 2012). This highlights the need for tailored support that considers both individual and contextual vulnerabilities.

Previous reviews on parental interventions have primarily focused on parents of premature infants and parenting outcomes (Girabent-Farres et al., 2021) or exclusively on mothers, without assessing study quality (Chertok et al., 2014). In contrast, Sabnis et al. (2019) reviewed 58 studies (23 in meta-analysis) that included both parents of NICU infants beyond prematurity. They categorised interventions into medical care changes (n=11), complementary/alternative medicine (n=12), family-centred instruction (n=30), and psychotherapy (n=5), overall finding modest long-term reductions in parental distress. Family-centred interventions, particularly in-hospital parent education, emerged as a clinical priority, aligning with neonatal care models that emphasise active parental involvement in care and decision-making (Lee, 2023; Roué et al., 2017). However, Sabnis et al. (2019) found medical care changes had no effect, and alternative medicine and psychotherapy were under-researched. Given the need for evidence-based mental health support, further investigation into the effectiveness of psychological interventions in the NICU is warranted (Hynan, 2015; Lee, 2023).

Psychological interventions in the NICU lack standardisation, with significant variability in content, delivery, and approach. Some use structured methods like trauma-focused cognitive behavioural therapy (CBT) (Shaw et al., 2023), while others adopt transdiagnostic approaches, such as emotional regulation programmes (Sin Choi et al., 2021). Others target the parent-child relationship by measuring interactive behaviours alongside mental health outcomes (Yu et al., 2023). Interventions also differ in delivery (group vs. individual) and duration (3–11 sessions). While previous studies have addressed specific concerns like trauma (Shaw et al., 2013) or anxiety (Yilmaz & Alemdar, 2022), NICU parents frequently experience overlapping psychological distress, including anxiety, stress, depression and PTSD (Malouf et al., 2022; Shetty et al., 2023), all of which share symptoms and underlying mechanisms (Zhu et al., 2022; Kalin, 2020). Sabnis et al. (2019) reviewed five psychological interventions, primarily cognitive-behavioural and unstructured psychotherapy, with four showing positive effects on parental distress. However, several relevant studies have emerged since then, highlighting the need for an updated review of evidence.

In the UK, maternity and perinatal mental health services within the NHS have expanded significantly over the past decade (Cantwell, 2023). Despite this expansion, gaps remain, particularly for women experiencing mental health challenges in a maternity context, for instance, birth trauma or adjustment to neonatal illness and complications (Cantwell, 2023). NHS England proposed the development of maternity psychological services as part of its ten-year Long-Term Plan (NHS England, 2019). Similarly, Scotland has been expanding Maternity and Neonatal Psychological Intervention (MNPI) services as part of a £50 million investment in perinatal mental health services (Scottish Government, 2019), currently under review by Public Health Scotland (2024). Given the diversity of psychological interventions in the NICU, a systematic review synthesising evidence is essential for informing clinical practice, enhancing parental well-being, and guiding future research. With Scotland expanding MNPI services, a comprehensive review could support the development of evidence-based strategies tailored to this population.

This review aims to evaluate the effectiveness of psychological interventions in reducing parental distress in the NICU. It specifically focuses on interventions that target parents' cognitions, emotions, and behavioural responses, as these psychological domains are key to understanding and mitigating distress. The review will aim to address the question: Which psychological interventions are most effective in alleviating symptoms of anxiety, PTSD, stress, and depression in parents of infants in the NICU? This will focus on quantitative studies, prioritising randomised controlled trials (RCTs) for their reliability in evaluating intervention effectiveness. Feasibility of implementing these interventions in clinical settings will be discussed.

## Methods

### Protocol and Registration

This review was conducted and reported in accordance with Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) 2020 statement and checklist (Page et al., 2021) (Appendix B.). Details of review registration can be found in Appendix A.

### Search strategy

After several scoping searches, a search strategy was designed and discussed with an experienced subject librarian. Search strategy using PICO (population, intervention, control, outcome) criteria can be found in Table 1. Four electronic databases, PsycINFO, Medline, EMBASE, and CINAHL were searched in November- December 2024 using the OVID and EBSCO platforms. The search strategy was adapted to make syntax and subject headings relevant for each database. An example of a completed search is presented in Appendix C.

**Table 1**

*PICO Search Terms*

| PICO Criteria | Search terms |
|---|---|
| Population: 1. Parents | parents/ or fathers/ or mothers/ or single parent/ Caregivers/ ("Parent*" or "Mother*" or "Father*" or "Caregiver*") |
| 2. NICU | intensive care, neonatal/<br>intensive care units, neonatal/<br>("NICU" or "Neonatal Intensive Care Neonatal Care" or "Neonatal Unit" or "Premature infant*" or "Preterm infant*" or "neo-nat*") |
| Intervention: | (therap* or intervention* or treatment*) |
| Control: | N/A |
| Outcome: Distress | anxiety/ or depression/ or psychological distress/ "trauma and stressor related disorders"/ or stress disorders, traumatic/("Parenting stress" or "Parental distress" or "Parental anxiety" or "Parental depression" or "parental coping")<br>("stress" or "distress" or "Emotion*" or "Anxi*" or "Depression" or "Coping" or "Mental health" or "Psychological*") |

Searches were limited to studies published in the English language. Reference lists of included papers and previous systematic reviews were also searched for additional articles.

## Inclusion/Exclusion Criteria

Studies with quantitative data pertaining to the effectiveness of psychological interventions aimed at reducing distress among parents of infants admitted to the NICU were included in this review. Psychological interventions were defined as non-pharmacological approaches that aim to modify emotional states, cognitions or behavioural responses, underpinned by a therapeutic modality (Hodges et al, 2011). Studies could employ any type of control condition for instance, treatment as usual or active control group. RCTs that were feasibility or pilot studies were eligible for inclusion if they provided sufficient pre- and post-test data and included a comparison with a control group.  Qualitative research and grey literature were not within the scope of the current review. Full eligibility criteria are provided in Table 2.

**Table 2**

*PICO Eligibility Criteria*

| | Inclusion | Exclusion |
|---|---|---|
| Population | Parents of infants admitted to NICU | Start of intervention after infant discharged from NICU |
| Intervention | Structured psychological intervention which addresses parents thoughts/emotions/ coping behaviours and aims to reduce psychological distress | Exclusively educational interventions (e.g. about caring for an preterm infant/admitted to NICU)<br>Exclusively Environmental/physical interventions e.g. physical touch, music therapy |
| Control | Participants randomised to Control Group/Treatment As Usual/Waitlist Control/Active Control | Non- RCT studies |
| Outcome | Studies report an outcome of psychological distress (stress, depression, anxiety, PTSD) using a validated outcome measure | Studies which do not report an outcome of psychological distress (stress, depression, anxiety, PTSD) using a validated outcome measure |
| Study design | Published RCTs | Grey literature; Qualitative research; Quasi-experimental design; Pilot and |

| | |
|---|---|
| | feasibility studies without sufficient pre and post-intervention data and control group |

### Study selection

Search results from each database were imported to Rayyan, an online systematic review tool. Duplicates were removed and references collated. Studies were then screened by the primary researcher using the inclusion/exclusion criteria, at first on the basis of their title and abstract and subsequently full text. The PRISMA flowchart was used to record the search and screening process. In the second stage, full texts of potentially eligible papers were retrieved and reviewed for eligibility. An independent reviewer who was a Trainee Clinical Psychologist screened 50% of full texts (n= 26). There was 97% inter-rater agreement. The only disagreement concerned the inclusion of Bagheri et al. (2024), which was ultimately excluded by consensus, as it did not include an adequate measure of psychological distress.

### Data extraction

The following information was extracted for each study: author, year; country; study design; outcomes measures; sample; features of psychological intervention; features of control condition; and reported findings related to distress in parents following admission of their infant to neonatal intensive care.

### Risk of Bias

The Cochrane Risk of Bias tool was used to assess the risk of bias. Two reviewers independently assessed risk of bias and disagreement was discussed until consensus was reached. Results from this assessment are summarised in Table 4.

### Synthesis

This systematic review provides a narrative synthesis of findings. Due to heterogeneity among studies a meta-analysis was deemed inappropriate. Included studies varied in the psychological intervention administered, outcome measures (anxiety, depression, traumatic stress) and several studies did not report sufficient data (i.e. post-treatment mean scores). Instead, a narrative synthesis was employed to provide detailed evaluation of intervention effectiveness, feasibility and quality of evidence.

### Certainty of Evidence

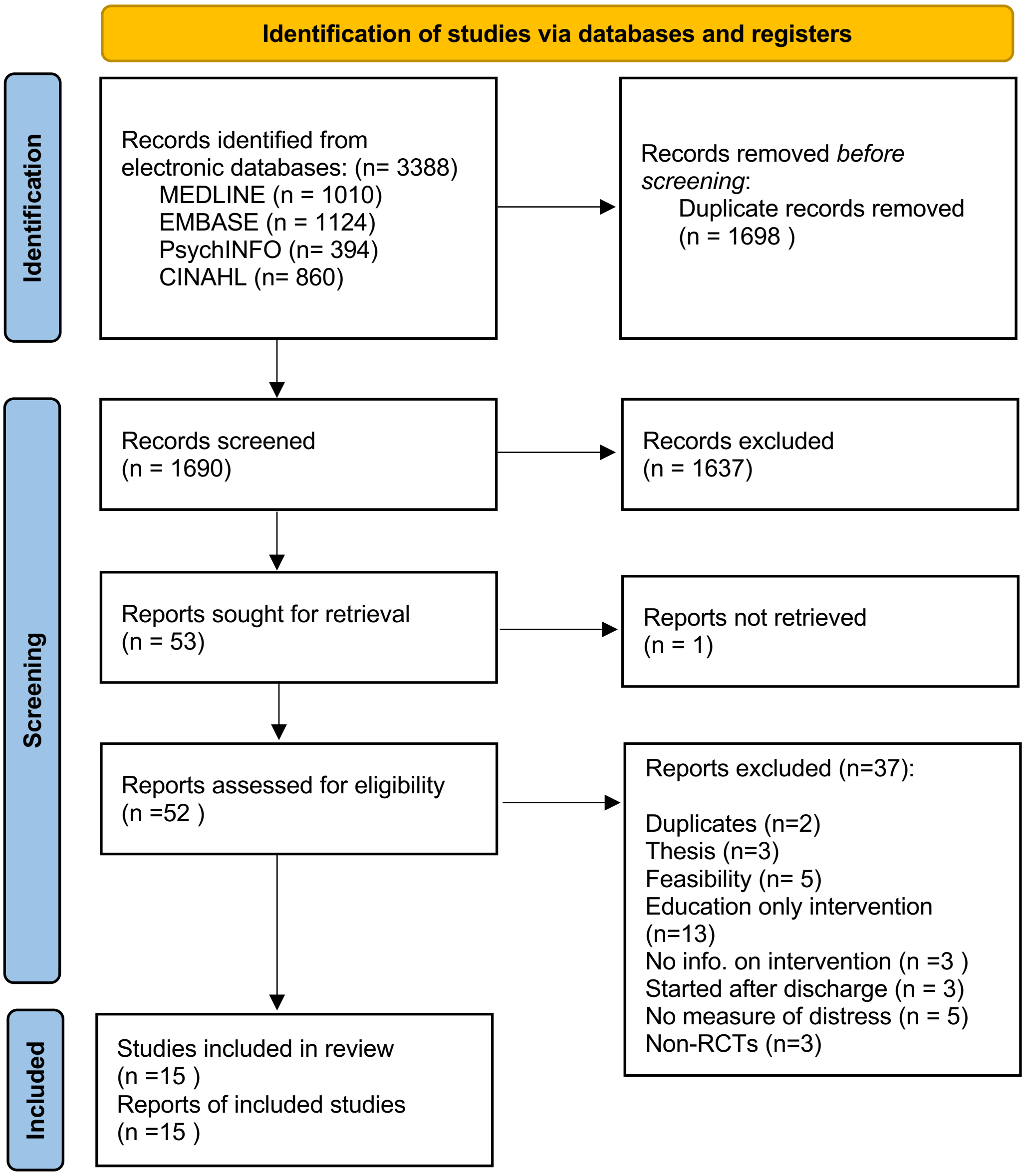
The certainty of evidence presented was assessed using an adapted form of GRADE (Grading of Recommendation, Assessment, Development and Evaluation), for narrative synthesis (Murad et al., 2017).

## Results

Electronic searches of MEDLINE, EMBASE, PsychINFO and CINAHL were carried out in November 2024 and identified 3380 records. 1698 duplicates were removed. After the screening process (see Figure 1), 15 studies were eligible for inclusion. No further studies were found through reference lists of identified papers or systematic reviews.

**Figure 1**

*PRISMA Study Identification Flowchart*



## Study Characteristics

Details of the 15 included studies are provided in Table 3. Studies were published between 2005 and 2024 and comprised 15 randomised control trials (n=1087). The majority of studies recruited mothers exclusively, two included both parents and one study consisted of fathers only. Sample sizes varied from 26 to 122. Most studies recruited parents of preterm or low birth weight infants. One study recruited parents of infants with further complexities. The studies were conducted globally, with four in Europe, four in North America and seven across Asia. The studies measured various outcomes of psychological distress including, parenting stress, anxiety, depression and post-traumatic stress.

## Outcomes

### *Stress*

Eleven studies included a measure of parental stress, seven of which utilised the Parental Stressor Scale: NICU (PSS-NICU) (Miles et al., 1993). Three studies used the Parental Stressor Scale (PSS) (Berry & Jones, 1995) and one used the Parental Stress Index (PSI) (Adibin, 1995).

### *Anxiety and Depression*

Three studies examined symptoms of anxiety and depression using one measure, with two studies using the General Health Questionnaire (GHQ-28) (Goldberg & Williams, 1988) and one using the Hospital Anxiety and Depression Scale (HADS) (Stern, 2014).

### *Anxiety*

Anxiety was assessed in seven studies. Three utilised the Beck Anxiety Inventory (BAI) (Beck et al., 1988) and four used the State-Trait Anxiety Inventory (STAI) (Spielberger, 1983).

### *Depression*

Eight studies included measures of depression, with four using the Beck Depression Inventory-II (BDI-II) (Beck et al 1996) and four used the Edinburgh Postnatal Depression Scale (EPDS) (Cox et al., 1987).

### *Post-Traumatic Stress*

Six studies assessed post-traumatic stress, employing four different scales, making it the outcome with the greatest variability in measurement tools. The most common was the Davison Trauma Scale (Davison et al., 1997) which was used in three studies, followed by the Perinatal Posttraumatic Stress Disorder Questionnaire (PPQ) (Callahan & Borja, 2008) used in two studies. The PTSD Symptom Scale (Ghorbani et al., 2015) was used in one study.

**Table 3**

*Study Characteristics*

| Author | Year | Country | Sample | Study design | Distress Outcome |
|---|---|---|---|---|---|
| Ouyang et al. | 2024 | China | 39 mothers of preterm infants (<32 weeks) | RCT | PSS;<br>HADS |
| Karimi et al | 2024 | Iran | 80 mothers of preterm infants (28-34 weeks) | RCT | PSS |
| Shaw et al. | 2023 | USA | 26 mothers of pre-term infants (25-34 weeks) | RCT | Davison Trauma Scale; BDI-II; BAI |
| Yu et al. | 2023 | China | 60 mothers of preterm infants | RCT | PSI<br>EPDS |
| Yimaz & Alemdar. | 2022 | Turkey | 85 mothers of preterm infants (28-38 weeks) | RCT | PSS-NICU;<br>STAI |
| Seiiedi-Biarag et al. | 2021 | Iran | 66 mothers of pre-term infants (28-33 weeks) | RCT | GHQ-28 |
| Samani et al. | 2020 | Iran | 50 mothers of preterm infants (32-34 weeks) | RCT | GHQ-28 |
| Koockaki et al. | 2018 | Iran | 90 mothers preterm infants (<37 weeks) | RCT | PTSD Symptom Scale |
| Fotiou et al. | 2015 | Greece | 59 parents of preterm infants (<37 weeks) | RCT | PSS; STAI; |
| Hoffenkamp et al. | 2015 | The Netherlands | 75 parents of pre-term infants (32-37 weeks) | RCT | PSS- NICU;<br>EPDS;<br>STAI |
| Borghini et al | 2015 | Switzerland | 60 mothers of preterm infants (<33 weeks) | RCT | PPQ |
| Shaw et al. (follow up to Shaw et al. 2013) | 2014 | USA | 105 mothers of preterm infants (25-34 weeks) | RCT | Davison Trauma Scale;<br>PSS- NICU;<br>BDI-II; BAI |
| Shaw et al. | 2013 | USA | 105 mothers of preterm infants (25-34 weeks) | RCT | Davison Trauma Scale;<br>PSS- NICU;<br>BDI-II; BAI |
| Ravn et al. | 2011 | Norway | 65 fathers of preterm infants (30-36 weeks) | RCT | PSI |
| Zelkowitz et al. | 2011 | Canada | 122 mothers of preterm infants (weight <1500g) | RCT | STAI; PSS-NICU; PPQ;<br>EPDS |

### Interventions

Different approaches to psychological interventions were utilised, varying in the therapeutic mechanism underpinning them. Eight studies used cognitive behavioural approaches. Six used attachment informed approaches by targeting the parent-infant relationship. Four were non-directive and did not follow a clearly defined underlying therapeutic mechanism. Further details of each intervention are reported in Table 4. All interventions were delivered at the same hospital site as the NICU, with one study providing a follow-up session at home (Borghini et al., 2014) and varied from three to fourteen sessions. Five studies administered group interventions and ten studies administered individual interventions.

### Risk of bias

Studies (n=15) varied significantly in quality, however the vast majority (n=11) showed a high level of bias. Details of scores for individual studies for each domain are reported in Table 4. Most RCTs implemented the randomisation processes effectively, resulting in a low risk of bias in this domain. However, several had issues with allocation concealment, lack of details about randomisation methods, or there were significant differences in baseline levels of distress between control and intervention, leading to some concerns in ensuring equal groups (Shaw et al., 2013, Zelkowitz et al., 2011). Due to the nature of psychological intervention studies, it was often not feasible to blind participants or therapists to the intervention. Some studies did attempt this by providing the same information about the study to both the intervention and control. A few studies also blinded therapists, however due to their professional knowledge it is possible some bias was introduced. Studies varied in their adherence to interventions, with some studies specifying strict protocols (e.g. Shaw et al, 2013; 2014; 2023). The reliance on subjective, self-reported measures created a high risk of bias in most studies where efforts were not made to blind participants to the condition. Attrition rates raised challenges in several studies (e.g., Shaw et al., 2023; Koockaki et al., 2018; Ravn et al. 2011), resulting in a high risk of bias. However, missing data was generally well reported across studies. Six studies reported pre-specified outcomes or published protocols, resulting in low risk of bias in this domain (e.g., Hoffenkamp et al., 2015; Zelkowitz et al., 2011;). However, the remaining nine did not explicitly state if their analyses were pre-specified, raising some concerns.

**Table 4**

*Cochrane risk of bias assessment 2.0 (RoB 2) for RCTs*

| Author, Year | Domain | | | | | | |
|---|---|---|---|---|---|---|---|
| | 1. Randomisation | 2a. Assignment to intervention | 2b. Adhering to intervention | 3. Missing data | 4. Outcome measurement | 5. Data reporting | Overall **r**isk of bias |
| Ouyang et al. 2024 | Low | Some | Some | Low | High | Low | High |
| Karimi et al, 2024 Iran | Low | High | Some | Low | High | Some | High |
| Shaw et al. 2023 USA | Some | Some | Low | High | High | Some | High |
| Yu et al. 2023 | Low | Some | Low | Low | Some | Some | Some |
| Yimaz & Alemdar. 2022 | Low | Some | Some | Some | High | Some | High |
| Seiiedi-Biarag et al. 2021 | Low | Some | High | Some | High | Some | High |
| Samani et al. 2020 Iran | Some | Some | Some | Low | High | Some | High |
| Koockaki et al., 2018 | Some | High | Some | Some | Some | Low | High |
| Fotiou et al. 2015 | Low | Some | Some | Low | Some | Low | Some |
| Hoffenkamp et al. 2015 | Low | Some | Some | Low | Some | Low | Some |
| Borghini et al. 2015 | Some | Some | Some | Some | High | Some | High |
| Shaw et al. 2014 | Low | Some | Low | Low | High | Low | High |
| Shaw et al. 2013 | Some | Some | Low | Low | High | Some | High |
| Ravn et al. 2011 | Low | Some | High | High | High | Some | High |
| Zelkowitz et al. 2011 | Low | Some | Some | Low | Some | Low | Some |

## Certainty of Evidence

The GRADE approach was used to assess the certainty of evidence across studies. This is based on guidance provided by Murad et al., (2017) when results have been synthesised narratively. Based on reviewing the risk of bias and variation in results, across the studies, the certainty of evidence was deemed to be low. See Appendix D for full details of appraisal.

## Synthesis of Results

Results are synthesised narratively according to the therapeutic modality underpinning each intervention. Nine studies did not report effect sizes and five did not report means or standard deviations either at baseline or post-treatment. Additionally, there was significant heterogeneity in the direction and size of effect of results reported. Details and results of studies are highlighted in Table 5.

### ***Cognitive- Behavioural Approaches***

Four studies using a cognitive-behavioural approach focused on PTSD management (Shaw et al., 2023, 2014, 2013; Koockaki et al., 2018). All interventions significantly reduced PTSD symptoms, as measured by the DTS and PTSD Symptom Scale, compared to an educational control group. Shaw et al. (2013) found six sessions of TF-CBT reduced trauma (Cohen's $d$=0.41, $p$<.001) and depression ($d$=0.59, $p$<.001), with further reductions at six months (trauma: $d$= 0.74, $p$<.001; depression: $d$= -0.64, $p$=.002) (Shaw et al., 2014). Anxiety initially declined in both groups, likely due to control group support, but showed a significant long-term reduction in the intervention group ($d$=0.63, $p$=.001) (Shaw et al., 2014). Extending the intervention with three additional sessions did not enhance outcomes, suggesting six sessions are sufficient for maintaining long-term benefits (Shaw et al., 2014). A group-based adaptation (Shaw et al., 2023) reduced trauma, depression, and anxiety symptoms but was less effective than individual therapy (mean score differences: -5.9 to -12.4; trauma: $d$=0.48, $p$=.016; depression: $d$=0.76, $p$=.003; anxiety: $d$=0.50, $p$=.042). Koockaki et al. (2018) also found a group-based TF-CBT intervention superior to routine care ($p$<.001), with sustained effects three weeks post-intervention ($p$<.001).

The three remaining studies used general cognitive-behavioural strategies to address negative thinking and manage emotional distress through relaxation and breathing techniques. Fotiou (2015) conducted a five-session group intervention targeting stress symptoms, finding a significant reduction in trait anxiety ($p$ = .02) but no significant effects on parental stress ($p$ =

.69) or state anxiety (*p* = .18). Similarly, Yilmaz & Alemdar (2022) provided seven 90-minute group sessions and reported a significant reduction in anxiety (*p* < .05) but no effect on parental stress (*p* > .05). Both studies included an educational control group, which may have contributed to the findings. Samani et al. (2020) implemented an individual CBT intervention, finding significant reductions in physical health symptoms (*p* = .005), anxiety (*p*= .001), and overall mental health (*p* = .001). However, reductions in depression (*p* = .07) and social function symptoms (*p* =.64) were not significant, and the control condition was insufficiently described, limiting interpretation. Notably, Samani et al. (2020) and Fotiou et al. (2015) did not conduct a priori power calculations, raising concerns about statistical power in detecting effects on parental stress. Despite methodological limitations, CBT appears effective in reducing psychological distress in NICU parents.

### ***Relational Approaches***

Five studies used an attachment-based approach (Bowlby, 1967) to address parents' perceptions of their child and help them recognise and manage distress in themselves and their infant. This approach showed more varied effects on distress than cognitive-behavioural interventions. Yu et al. (2023) found that four individual sessions with a clinical psychologist significantly reduced stress (*d* = -0.76, *p* < .001) and depression (*d* = -0.76, *p* < .05) at six months, with a large effect size. In contrast, four studies found no significant reductions in parental distress. Rayn et al. (2011), using a similar approach to Yu et al. (2023) but with fathers, reported increased parental stress in both intervention and control groups, with high dropout rates (average attendance: five of 11 sessions). Borghini et al. (2015) implemented a six-session Video Interaction Guidance (VIG) intervention, finding PTSD symptom reductions within the intervention group over 12 months (*p* = .045), though not significantly different from the control (*p* = .07). Hoffenkamp et al. (2015) found no significant reductions in parental stress or depression (*p* > .05) after a three-session VIG intervention. Zelkowitz et al. (2011) offered six individual sessions to enhance maternal sensitivity and distress recognition, finding reduced trauma and anxiety symptoms in both groups but no significant differences (*p* > .05). Their active control group, which included educational materials, may have contributed to these results.

Among the higher-quality studies, such as those by Yu et al. (2030), Hoffenkamp et al. (2015), and Zelkowitz et al. (2011), only Yu et al. reported significant improvements in psychological distress. This suggests some promise for relational interventions, particularly when delivered with methodological rigour. However, the lack of consistent effects across other relatively robust studies indicates that the effectiveness of these approaches may depend on factors such as intervention intensity, or population characteristics. While quality enhances confidence in the findings, it does not fully account for the variability in outcomes. Overall, attachment-informed interventions showed less consistency in reducing psychological distress compared to cognitive-behavioural approaches.

### ***Alternative/Non-directive Approaches***

Three studies used a nondirective approach to address parental distress within a structured therapeutic context. Ouyang et al. (2024) combined mindfulness training with Kangaroo Care, finding significant reductions in anxiety and depression (*mean difference* = -2.87, $p = .001$) but no effect on parenting stress. Two studies used group counselling with NICU education, emotional expression, and relaxation techniques. Karimi et al. (2024) reported a substantial reduction in parenting stress ($d = 3.90$, $p > .001$), and Seiiedi-Biarag et al. (2021) found significant distress reduction (*mean difference* = -9.8, $p < .001$), as measured by the GHQ-28. However, both studies had a high risk of bias due to the unstructured nature of counselling, which relies on participant generated content, leading to variability in intervention quality, creating challenges for consistent evaluation. Additionally, the lack of details on routine care in control groups complicates interpretation. The educational component in Karimi et al.'s (2024) intervention may have contributed to stress reduction, as observed in other study control groups.

**Table 5**

*Study Results*

*Cognitive- behavioural approaches*

| Author, Year | Intervention Details | Control | Mean (SD) baseline | Mean (SD) post-treatment | Mean (SD) follow-up | Mean difference (95% CI) | Effect size Post-treatment | Effect size follow-up | Significance |
|---|---|---|---|---|---|---|---|---|---|
| **Shaw et al. 2023** | I: 6x sessions manualised group TF-CBT n=26<br>I: 6x sessions manualised individual TF-CBT n=62 | N/A | Group<br>DTS: 48.19(27.02)<br>BDI-II: 18.23(8.31)<br>BAI: 21.85(12.13)<br>Individual<br>DTS: 49.40(25.49)<br>BDI-II: 20.60(9.48)<br>BAI: 21.97(11.83) | NR | NR | Baseline- 6 months<br>DTS -12.4(2.4, 22.4)<br>BDI-II -6.9 (2.4,11.5)<br>BAI -5.9(0.2, 11.6) | 3 months<br>DTS: *d*=0.35<br>BDI-II: *d*=0.83*<br>BAI: *d*=0.66 | 6 months<br>DTS: *d*=0.48*<br>BDI-II: *d*=0.76**<br>BAI: *d*=0.50** | Longitudinal mixed effects modelling showed significant difference between groups across measures, in favour of individual input.<br>Baseline-post<br>DTS *p*=.083<br>BDI-II *p*= .001<br>BAI- *p*=.042 |
| **Yimaz & Alemdar. 2022** | Education about NICU and 7x90 min group sessions discussing feelings and developing cognitive and behavioural stress management strategies<br>n=45 | TAU: Education about NICU<br>n=40 | PSS- NICU<br>I: 2.03(0.38)<br>C:2.06 (0.28)<br>STAI-2<br>I: 47.72 (5.64)<br>C: 49.80 (5.34) | PSS-NICU:<br>I:1.58 (0.39)<br>C: 1.72 (0.38)<br>STAI-2<br>I: 46.80* (4.76)<br>C: 50.15 (5.35) | N/A | NR | NR | NR | Between groups testing showing significant reduction for anxiety only. STAI-2: *p* < .05 No significant reduction observed for overall PSS- NICU score. |

| **Author, Year** | **Intervention Details** | **Control** | **Mean (SD) baseline** | **Mean (SD) post-treatment** | **Mean (SD) follow-up** | **Mean difference (95% CI)** | **Effect size Post-treatment** | **Effect size follow-up** | **Significance** |
|---|---|---|---|---|---|---|---|---|---|
| **Samani et al. 2020** | 6x 45 minute individual sessions of CBT<br>n=25 | TAU (no details reported)<br>n=25 | N/R<br>I: 29.24<br>C: 21.76 | GHQ-28<br>I: 17.36*<br>C: 33.64 | N/A | NR | NR | N/A | Between group testing showed significant reduction for I group. (*p* = .001) |
| **Koockaki et al., 2018** | 8x 60 mins sessions group CBT for PTSD+ routine care counselling (education and care skills for NICU)<br>n=42 | Active control: 8x sessions routine care counselling with educational package<br>n=39 | PTSD SS<br>I: 8.09(5.20)<br>C:6.82(4.28) | I: 4.55 (2.30)**<br>C: 6.56 (4.29) | 3 weeks<br>I: 5.00** (2.24)<br>C: 14.21 (4.86) | NR | NR | NR | Generalised estimating equation showing between group difference post-intervention. *p*< .001 and at 3-week follow up (*p* <.001) |
| **Fotiou et al. 2015** | 5x 90 min group sessions. Educational sessions + cognitive and behavioural strategies to manage stress<br>n=31 | Active control: 5x 90 min educational sessions on NICU and premature infants<br>n=28 | PSS<br>I: 28.3(8.0)<br>C: 27.3(8.1)<br>STAI-1<br>I: 46.3 (11.4)<br>C: 45.7 (13.6)<br>STAI-2<br>I: 41.7 (11.3)<br>C: 38.1 (14.2) | PSS<br>I: 25.2(7.5)<br>C: 24.4(9.4)<br>STAI-1<br>I: 41.6 (10.5)<br>C: 41.5 (11.8)<br>STAI-2<br>I: 37.9 (12.5)*<br>C: 42.6 (11.8) | N/A | NR | NR | NR | Between group testing showed a significant reduction for trait anxiety only (STAI-2) (*p*= .02). No sig. differences between groups in total scores for stress *p*=0.69 or state anxiety, *p*= 0.51. |
| **Shaw et al. 2014 USA** | I: 6 x sessions Manualised TF-CBT, infant redefinition, and parenting | (Reported below)<br>n=43 | DTS;<br>I:49.40 (25.49)<br>C: 42.35 (27.05) | NR | NR | Baseline-6 months<br>DTS: -15.96 [-23.13, -8.86] | 1 month<br>DTS<br>*d*=0.33* | 6 months<br>DTS<br>*d*=0.74** | At six months, mixed effects modelling shows differences between intervention and control: |

| **Author, Year** | **Intervention Details** | **Control** | **Mean (SD) baseline** | **Mean (SD) post-treatment** | **Mean (SD) follow-up** | **Mean difference (95% CI)** | **Effect size Post-treatment** | **Effect size follow-up** | **Significance** |
|---|---|---|---|---|---|---|---|---|---|
| **(follow up to Shaw et al. 2013)** | guidance.<br>I: 9 x sessions (As above with additional 3 sessions aimed at identifying trauma triggers and education about parenting patterns)<br>n=62 | | BDI-II<br>I: 20.60 (9.48)<br>C: 17.49 (10.68)<br>BAI<br>I: 21.97 (11.83)<br>C: 20.30 (12.49) | | | BDI-II: 5.12 [-8.38, 1.86]<br>BAI: -5.31 [-8.36, 3.25] | BDI-II *d*=-0.55*<br>BAI *d*=-0.19 | BDI-II *d*=-0.64**<br>BAI *d*=-0.63** | trauma (*p*=.001), depression (*p* = .002), and anxiety (*p*= .001). No additional benefit from 3 additional sessions. |
| **Shaw et al. 2013** | I: 6x 45 min individual sessions. Manualised TF-CBT,<br>infant redefinition, and parenting guidance.<br>N=62 | Active control: 1x 45 min educational session on NICU and parenting pre-term infant<br>n=43 | DTS;<br>I:49.40 (25.49)<br>C: 42.35 (27.05)<br>BDI-II<br>I: 20.60 (9.48)<br>C: 17.49 (10.68)<br>BAI<br>I: 21.97 (11.83)<br>C: 20.30 (12.49) | NR | N/A | 1 month<br>DTS<br>-7.378 (13.72, -1.03)<br>BDI-II<br>–4.352 (6.79,1.91)<br>BAI<br>–1.675 (-4.93,1.58) | DTS<br>*d*=0.41*<br>BDI-II<br>*d*= 0.59**<br>BAI<br>*d*= 0.16 | NR | Mixed effects modelling found significant reduction for I group for trauma (*p*<.001) and depression (*p*<.001). There was reduction in anxiety symptoms over time but no difference between groups. |

*Relational approaches*

| Author, Year | Intervention Details | Control | Mean (SD) baseline | Mean (SD) post-treatment | Mean (SD) follow-up | Mean difference (95% CI) | Effect size Post-treatment | Effect size follow-up | Significance |
|---|---|---|---|---|---|---|---|---|---|
| **Yu et al. 2022** | 4x individual sessions of manualised modified MITP with clinical psychologist (understanding, recognising and responding to parent and infant cues).<br>n=30 | Preterm control TAU- education about NICU and 2x non-directive psychology sessions<br>n=30 | PSI<br>I: 89.9 (11.7)<br>C: 87.3 (20.8)<br>EPDS<br>I: 17.9 (4.1)<br>C: 18.8 (5.5) | PSI<br>I: 78.0** (14.3)<br>C: 95.1 (17.8)<br>EPDS<br>I:15.0 (4.0)**<br>C: 19.4 (4.8) | PSI<br>I: 71.2** (12.5)<br>C: 86.1 (22.9)<br>EPDS<br>I:14.1 (3.4)*<br>C:17.4 (4.7) | NR | 1 month<br>PSI<br>*d*= −0.94**<br>EPDS<br>*d*= -0.91** | 6 months<br>PSI<br>*d*= −0.76**<br>EPDS<br>*d*= -0.76* | A multi-level growth curve analysis found reduction for intervention group compared to control for PSI (*p*<.001) and EPDS (*p*=.004) at post-treatment and 6-month follow up. |
| **Hoffenkamp et al. 2015** | 3x sessions Video Interaction Guidance with clinical psychologist<br>n=75 | Preterm control TAU- education about NICU informal support from staff<br>n=75 | PSS-NICU<br>I: 71.95 (2.59) (mothers)<br>I: 61.99 (2.28) (fathers)<br>C: 72.97 (2.59) (mothers)<br>C: 63.82 (2.34) (fathers)<br>EPDS- NR | PSS-NICU- NR<br>EPDS<br>I:6.71 (0.60) (mothers)<br>I: 4.01 (0.40) (fathers)<br>C: 7.34 (0.62) (mothers)<br>C: 3.54 (0.44) (fathers) | NA | PSS-NICU<br>-1.02 ( 8.23, 6.21) (mothers)<br>-1.83 (8.31, 4.64) (fathers)<br>EDPS<br>-0.63 (2.30 to 1.04) (mothers)<br>0.47 (0.72 to 1.66) (fathers) | NR | NA | Between group analyses showed no significant differences on PSS-NICU (*p* >.05) or EPDS (*p* >.05) |

| **Author, Year** | **Intervention Details** | **Control** | **Mean (SD) baseline** | **Mean (SD) post-treatment** | **Mean (SD) follow-up** | **Mean difference (95% CI)** | **Effect size Post-treatment** | **Effect size follow-up** | **Significance** |
|---|---|---|---|---|---|---|---|---|---|
| **Borghini et al. 2014** | 6 x individual sessions (3 following discharge) based on family systems theory with components of and VIG aimed at improving parents' sensitivity and responsiveness<br>n=36 | Preterm control (n=29) and full-term control (n=23) groups<br>TAU:<br>1x educational session regarding infant also offered to PC group. | PPQ:<br>I: 4.62 (3.54)<br>PC: 3.55 (2.88)<br>FC: 1.52 (1.64) | I: 3.31* (3.38)<br>PC: 3.59 (3.31)<br>FC: 1.35 (1.69) | I: 2.69* (2.58)<br>PC: 3.17 (2.66)<br>FC: 1.26 (1.60) | | NR | *R*² = 0.39<br>Results from ANCOVA with PC groups only. FC not included. | Within group testing found significant reduction over time in intervention (*p*=.045). Between group testing was not significant (*p* = .07) |
| **Ravn et al. 2011** | 11x 60 min sessions MITP for fathers aimed to increase sensitivity and responsivity<br>n=32 | Preterm control (n=37) and full term control (n=39). TAU-no other info reported | NR | PSI (6 months)<br>I: 60.8 (12.7)<br>PC: 62.2 (16.1)<br>FC: 52.2 (10.4) | PSI (12 months):<br>I: 195.7 (27.9)<br>PC: 200.5 (30.5)<br>FC: 186.0 (25.8) | | NR | NR | No significant differences between intervention and PC at 6 (*p*=.69) or 12 months (*p*=.65) |
| **Zelkowitz et al. 2011** | "Cues" programme: 6x 45-75 min individual sessions to support mothers recognise and respond to their own distress and enhance their sensitivity to infant's distress<br>n=46 | Preterm active control: "Care" programme: 6x 'contacts' given general information about infant care.<br>N=46 | PSS-NICU (total NR)<br>PPQ<br>I: 5.7 (3.1)<br>C: 5.6 (2.9)<br>STAI<br>I:48.4 (13.8)<br>C:47.9(14.1)<br>EPDS<br>I: 39<br>C: 41 | PSS-NICU (total NR)<br>PPQ<br>I: 2.9 (2.9)**<br>C: 3.2 (2.8)**<br>STAI<br>I:26.6 (6.8)**<br>C:27.8 (8.2)** | N/A | PPQ<br>(0.8 to 1.5)<br>STAI<br>(-2.0 to 4.3) | NR | NR | Within group differences for I and C but no between group differences for STAI (*p*=.46) or PPQ (*p*=.54)<br>EPDS- NR |

*Alternative/Non-directive Approaches*

| Author, Year | Intervention Details | Control | Mean (SD) baseline | Mean (SD) post-treatment | Mean difference (95% CI) | Mean (SD) follow-up | Effect size Post-treatment | Effect size follow-up | Significance |
|---|---|---|---|---|---|---|---|---|---|
| **Ouyang et al. 2024** | 14x 30 min sessions of mindfulness training during Kangaroo Mother Care (KMC): n=47 | Active preterm control: KMC only<br>n=44 | PSS- NICU<br>I: 47.89 (2.965)<br>C: 48.18(2.814)<br>HADS<br>I: 23.57(3.44)<br>C: 24.18 (3.72) | PSS- NICU<br>I: 42.47 (4.62)<br>C: 41.05 (4.24)<br>HADS<br>I: 15.06(1.37)**<br>C: 17.93 (2.27)** | HADS<br>-2.868 (0.643, -2.093)** | N/A | NR | NR | Significant reduction for intervention group for HADS only. (*p*=.001). |
| **Karimi et al, 2024** | 4x 45-60 min individual non-directive counselling sessions centred around 'empowering' mothers in NICU<br>n=40 | Preterm control<br>TAU: no other info reported<br>n=40 | PSS- NICU<br>I:155.42 (36.74)<br>C: 155.45(36.74) | PSS- NICU<br>I: 61(10.98)**<br>C: 154.65 (32.15) | | N/A | *d*= 3.90 | N/A | Significant reduction for intervention group (*p*<.001) |
| **Seiiedi-Biarag et al. 2021** | 6x 45-60 min group sessions non-directive 'supportive' counselling by research assistant<br>n=34 | Preterm control<br>TAU: no other info reported<br>n=32 | GHQ-28<br>I: 19.8 (2.4)<br>C: 18.6 (2.6) | I: 14.0 (4.8)**<br>C: 22.6 (7.0) | -9.8 (-12.5 to -7.1)** | NA | NR | NA | Significant reduction for intervention group (*p*< 0.001) |

Note: NR= Not Reported; I= Intervention; C= Control **p*<.05 ***p*<.001; CI=Confidence Interval

## Discussion

This review aimed to explore the effectiveness of psychological interventions on parental distress in the NICU. Where previous reviews in this area have examined all forms of hospital-based interventions, this review focuses on those aimed at parents cognitions, emotions and behavioural responses. Fifteen studies from North America, Europe, China, Turkey and Iran met the eligibility criteria for review. Eleven studies showed a significant reduction in a measure of distress when compared with the control, six of which reported effect sizes ranging from small to large in magnitude.

### *Summary of results*

Seven studies utilised a cognitive-behavioural approach, all of which found a significant effect for the intervention group. The majority of these studies (n=6) also utilised an active control group consisting educational sessions about the NICU and caring for a NICU infant. The findings from Shaw et al. (2013, 2014, 2023) and Koockaki et al. (2018) on trauma-focused cognitive behavioural therapy (TF-CBT) suggest that while both manualised TF-CBT group interventions and general CBT approaches were effective, a structured, individualised intervention may yield stronger outcomes. However, a group-based intervention may still be a cost-effective. The results from studies employing a generalised CBT approach suggest that cognitive-behavioural strategies, such as addressing thinking styles, maladaptive cognitions, and behavioural coping mechanisms, may be effective in reducing more entrenched forms of anxiety. This is particularly evident in reductions in trait anxiety, but not necessarily state anxiety (Fitiou et al., 2015; Yilmaz & Alexander, 2022). However, parents may still continue to experience situational anxiety and stress related to the ongoing challenges of their infant's NICU admission. This supports evidence from Van Dis et al's (2019) systematic review and meta-analysis, indicating the benefits of CBT for reducing anxiety post-treatment and long-term within the general adult population. Although Shaw et al (2014; 2023) examined long term effects, the remaining studies (Fitiou et al., 2015; Samani et al 2020, Koockaki et al., 2018; Yilmaz & Alexander, 2022) would be strengthened by gathering follow- up data to examine the long-term impact of CBT interventions for parents.

Five studies examined the effect of relationally focused interventions, aimed at improving maternal sensitivity, responsiveness and managing stress reactions. There were conflicting results, with one study finding a significant effect of interventions on symptoms of depression, stress and PTSD (Yu et al., 2022). Four studies found no significant effect on parental stress, depression or anxiety (Borghini et al., 2015; Hoffenkamp et al., 2015; Rayn et al., 2011; Zelkowitz et al., 2011). Rayn et al's study (2011) with fathers had a high drop-out rate, with most fathers attending an average of five out of 11 sessions. Research about engaging fathers in parenting interventions has found that practitioner competence and organisational support are predictive of father engagement (Tully et al., 2018). It is possible this contributed to the results observed.

The results for these interventions may indicate a need to address parents' own mental health concerns prior to providing input aimed at improving parent-infant bonding. Symptoms of depression, anxiety and stress can hinder parents' ability to recognise and respond sensitively to their infant's cues (Radoš, 2021). Research indicates that anxiety is associated with lower sensitivity during mother-infant interactions, which can adversely affect the child's social and emotional development (Smith et al., 2022). Therefore, prioritising parental mental health in the NICU may be important for fostering healthy parent-infant relationships.

Finally, mindfulness and group counselling interventions also appeared to reduce aspects of distress (Ouyan et al., 2024; Karimi et al., 2024; Seiiedi-Biarag et al., 2021). Mindfulness, defined as the practice of observing and acknowledging thoughts, emotions, and bodily sensations, demonstrated effectiveness in alleviating anxiety and depression. This finding aligns with previous research highlighting its role in reducing symptoms of depression and anxiety in adults (Bhattacharya & Hofmann, 2023). The findings for group counselling also support previous research. However, they may have been obscured by the educational content in the intervention groups, which have been found to reduce parental distress (Sabnis et al., 2019).

Overall, although the results are varied, studies which adopted a cognitive behavioural approach appeared to reduce symptoms of parents' distress, most consistently. This suggests that interventions specifically designed to address parents' cognitions, emotions, and behavioural responses may be the most effective in reducing distress both in the NICU and long term. While attachment-focused interventions and counselling approaches also aim to influence parents' thoughts and behaviours, they do so in a less direct and targeted manner. Interventions had a lesser effect on parental stress than on depression, anxiety, and trauma symptoms, likely due to the inherent stress of having a baby in the NICU. The lack of significant reduction in measures of parental stress may also reflect the benefits of educational control interventions in reducing anxiety and aiding parental adjustment. This aligns with Sabnis et al. (2019), who found educational interventions significantly reduced parental stress.

### ***Strengths and limitations***

This review provides a comprehensive and up-to-date synthesis of evidence on the effectiveness of psychological interventions in reducing psychological distress, including symptoms of depression, anxiety, trauma, and stress, among parents of infants admitted to the NICU. By including studies based on psychological interventions, this review builds upon the recommendations of Sabnis et al. (2019), who identified psychotherapeutic interventions as a key area for further investigation. Additionally, this review incorporates eight studies published since 2019, providing an analysis of recent findings. Focussing on a broad range of outcomes enabled synthesis of findings examining various forms of distress, recognising their frequent co-occurrence and interrelated nature. The studies included in the review were conducted in Europe, Asia, the middle East and North America, which supports the generalisability of the review findings. However, it is possible that social context and norms across cultures may impact on treatment effectiveness, which may account for the variation in results.

Only a small number of studies (n=4) included fathers and parents of infants with a congenital abnormality (n=1), which somewhat limits the findings to mothers of preterm infants. Fathers and parents of full-term infants admitted to the NICU are also susceptible to psychological distress (Noergaard, 2019). Therefore, further research and targeted interventions are needed to address their specific mental health needs. All studies also excluded parents with a prior history of mental health difficulties or previous psychological input. This population are more vulnerable to mental health problems worsening and therefore research and recommendations for clinical practice for this population is required. There were significant methodological limitations in the studies included, such as unreported data, lack of effect size calculations and the use of self-reported measures, meaning the certainty of evidence was deemed to be low. The methodological and conceptual heterogeneity among studies meant that a meta-analysis was deemed inappropriate. When more research is available, a future review and meta-analysis could include a narrower inclusion criterion focusing solely on one specific intervention, for instance trauma-focused CBT. Future interventions studies should ensure transparent data reporting and calculation of effect sizes in order to strengthen the generalisability of findings. They should also include a priori power calculations to ensure sample sizes are sufficient to detect statistical significance.

### *Implications for future research*

Future research should prioritise addressing the methodological limitations highlighted by the current review. The use of clinical interviews to assess clinical symptoms in addition to self-reported measures could be considered to reduce risk of bias pertaining to outcome measurement. Furthermore, collecting long-term follow-up data would provide valuable insights into the enduring effects of interventions, allowing researchers to understand their sustained impact on symptoms and how they may influence individuals' well-being over time. Ensuring fidelity in the delivery of interventions would help to minimise variations in the intervention itself, as well as mitigating potential individual differences in therapists' delivery. Research should prioritise incorporating fathers and parents of infants admitted to the NICU for reasons other than low birth weight or prematurity. With 60% of UK NICU admissions being full-term infants (NDAU, 2022), current research excludes a significant portion of parents. As infant health severity in the NICU correlates with higher parenting stress and negative parenting styles (Grunberg et al., 2019), psychological support is crucial to mitigate long-term adverse outcomes for this population.

### *Clinical Implications*

The psychological impact of having an infant admitted to the NICU has gained increasing attention in recent years (Saxton et al., 2020), reflected by the expansion of psychological care within maternity and neonatal settings (NHS England, 2023; NHS Scotland, 2024). While the quality of evidence in this review was limited, brief and structured cognitive-behavioural interventions show promise and offer an important avenue for further development and application in clinical practice. Although individual interventions appear more effective, group-based interventions, such as TF-CBT could be a feasible and cost-effective means to improving parents' wellbeing. Furthermore, non-psychology staff could be trained in the delivery of brief, manualized cognitive-behavioural interventions. This approach aligns with the Scottish Government's initiative to equip neonatal staff with the skills necessary to support and manage parental distress effectively (NHS Scotland, 2024). The studies analysed in this review could serve as a basis for further development of a manualised approach tailored to this population.

## Conclusions

Reducing distress among parents of infants in the NICU is a public health priority due to the potential long-term effects on parental mental health and subsequent child emotional, cognitive, and behavioural development. While this review did not provide sufficient evidence to draw firm conclusions, the interventions that most consistently demonstrated a reduction in parental distress were those employing a cognitive-behavioural approach, with the addition of a NICU-educational component. Non-directive counselling, and mindfulness also showed some positive outcomes; however, further research is needed to validate these approaches. The slight superiority of cognitive-behavioural approaches suggests that interventions targeting parents' thoughts, emotions, and behavioural responses to distress may be particularly effective in reducing long-term symptoms of psychological distress, including depression, anxiety, and PTSD. Future research should prioritise rigorous methodologies and broaden inclusion criteria for intervention studies to strengthen the evidence base.

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# Chapter 2: Major Research Project

## *"The Hope Dies Last"* Exploring parents experiences of decision-making in Congenital Diaphragmatic Hernia: a qualitative analysis

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## Plain Language Summary

Title: *“The Hope Dies Last”* Exploring parents experiences of decision-making in Congenital Diaphragmatic Hernia

Background: CDH (congenital diaphragmatic hernia) is a disease that some babies are born with. This is usually diagnosed at a scan of the mother when she has been pregnant for 20 weeks. This means that when the baby is developing in the womb, there is a hole in its diaphragm. A diaphragm is a thin muscle that separates the stomach from the chest and helps us breathe. When there is a hole in this, the stomach contents (like the intestine for example) move through the hole and into the chest. This means that it presses on other important organs in the chest (like the heart and lungs) and can stop them from growing. This makes it very difficult for babies to breathe properly when they are born. This can cause a lot of worry for parents and they might have to make important and difficult decisions like when to give the baby an operation (and how many operations) or if the baby needs a machine to breathe for them. CDH can affect babies in different ways and there are sometimes different decisions for parents. Parents and doctors sometimes find it difficult to know what the best decision is, and when they should stop giving treatment. This project will examine the experience of decision making for parents by answering: What was it like to make those decisions; who and what information helped you make the decisions at certain times from when you found out your baby had CDH?

Methods: Twenty-one parents were sent a letter about the research by their doctor at the time they were in hospital. Four mothers and one father of babies born with CDH in Scotland from 2021 and 2023 wanted to take part and were interviewed. Parents were interviewed about their experience and the decisions they made at the time, from when they found out about the CDH, to when the baby was born and was in hospital.

Results: Parents discussed four main experiences: processing the diagnosis and looking for information; trusting staff; making decisions; and looking back on their journey. These experiences are compared with previous research.

Conclusions: This study gives important insights into what parents go through and what they need, which can help guide future research and improve support for families with CDH. Research in the future could interview staff to understand more about their experiences.

## Abstract

Background: Congenital diaphragmatic hernia (CDH) is a rare birth defect which causes abdominal contents to move through a hole in the diaphragm and into the chest. This can have a significant impact on the infant's lung development and results in death in approximately 30% of cases. This can cause considerable distress for parents at the point of diagnosis (usually prenatally), throughout pregnancy and at birth. The treatment of children born with CDH involves careful multi-disciplinary planning and can often involve aggressive treatment such as surgery and heart and lung bypass from birth. Disease progression can vary greatly resulting in a variety of treatments at different time points. Parents of infants with CDH have reported feeling insecure and powerless due to uncertainty and lack of transparent communication with medical professionals. This study aims to explore parents’ decision-making process during the care of their child with CDH, focusing on what information influenced their decisions around their child’s care.

Methods: Qualitative, semi-structured interviews were conducted with five parents whose children have received treatment for CDH in a hospital in Scotland between 2021 and 2023. Interpretative Phenomenological Analysis (IPA) was used to analyse interview content.

Results: Four main themes emerged and are presented chronologically, in line with parents’ journey of care: understanding the diagnosis and the search for answers; developing trust in the team; navigating decisions; reflecting on the journey. Parents emphasised the importance of specialist information in building trust with staff, which shaped their decision-making and emotional coping. Hope played a key role throughout their journey, influencing their perspectives and choices. The relationship between themes and their subthemes are discussed with reference to existing literature.

Conclusion: This study provides valuable insights into the experiences and needs of parents within this population, offering important implications for future research and clinical practice.

# Introduction

Congenital diaphragmatic hernia (CDH) is a rare birth defect that affects approximately 200-300 new-borns in the UK annually (Wang et al, 2019). CDH occurs when the diaphragm fails to form properly. This causes the protrusion of abdominal contents (e.g. stomach, intestine, liver) through the diaphragm and into the chest cavity. This means there is often significant deficits in lung development, heart defects and gastrointestinal abnormalities (Van den Hout et al, 2010). CDH often requires aggressive treatment methods from birth. Patients born with CDH require specialist multi-disciplinary care, involving advanced cardiorespiratory management and surgery in the neonatal period (birth-28 days) and beyond. Infant survival for those with a diagnosis of CDH ranges between 60-80% worldwide however this can involve multiple surgeries and an average hospital stay of at least 5 weeks following birth (Politis et al, 2021; Putnam et al, 2015).

CDH is diagnosed at the routine 20-week pregnancy scan in 70% of cases. The number of parents who make the decision to terminate the pregnancy at this stage varies across the world. Up to 40% of pregnancies in some countries (e.g. Australia and The Netherlands) are terminated (Lee et al, 2018; Horn-Oudshoorn et al, 2023), compared with 16% of pregnancies in the UK (Politis et al, 2020) perhaps reflecting a complex interplay of medical, legal, cultural, and ethical factors. Decisions to terminate the pregnancy in The Netherlands appear to be related to disease severity and are seemingly independent of maternal factors such as age, marital status and socioeconomic status (Horn- Oudshoorn et al, 2023). This disparity may be influenced by differing national attitudes toward termination and societal values regarding disability and neonatal outcomes. These variations highlight the importance of context when interpreting international data on CDH outcomes.

It is important to consider the emotional and psychological impact this can have on parents at the point CHD is diagnosed. Carlsson and colleagues (2017) found that following the diagnosis of a foetal abnormality parents report experiencing grief, shock, disbelief and anger. A systematic review reported that several factors influence parents' decisions around the pregnancy at this point including hope, mortality, and the implications for their own and unborn child's future (Blakeley et al, 2019). While prenatal diagnosis can provide an early opportunity for treatment planning and psychological adjustment, it has been associated with higher levels of psychological distress compared to postnatal diagnosis (Skari et al., 2006).

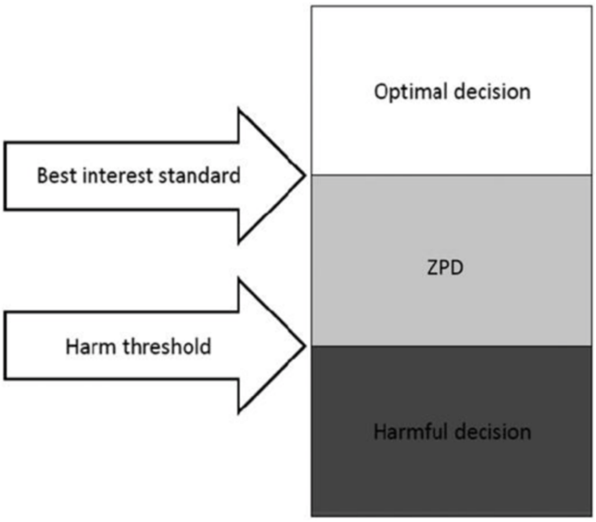
However, research has shown that depression scores among parents of children with CDH tend to decrease following prenatal counselling (Aite, 2009). This highlights the value of counselling and education during the prenatal period in preparing parents for potential challenges and helping them manage uncertainty. This also raises questions about what parents experience in relation to CDH. It may evoke similar feelings to those identified by Carlsson et al. (2017), yet there is a notable lack of research specifically exploring the impact of CDH and how parents navigate these challenges.

Despite advances in care over recent years, treatment protocols within the UK vary across different centres (Long et al, 2018). Decisions of when and what treatment to offer is based on several different variables, including disease stage and progression and additional health complexities. This therefore requires complex decision making for clinician's and parents from point of diagnosis, during birth planning, and post- birth treatments. It is necessary to consider the mechanisms that underlie decision making within a health context. Very traditional models of clinical decision making adopt a paternalistic approach, whereby the clinician is deemed to know what is in the best interests of the patient (Komrad, 1983), despite the patient being capable of making decisions for themselves. This dynamic has faced significant criticism over the years (Komrad, 1983). In an attempt to move away from this, alternative models placed increased emphasis on patient autonomy, where the clinician presents information and can aid the patient's interpretation, but the ultimate decision lies with the patient (Charles et al, 1997). Although patient autonomy is central to medical ethics, a more collaborative approach consisting of shared decision making has been adopted in recent decades (Elwyn et al, 2010). This can be defined as an approach where clinicians and patients share information and patients are supported to consider all options and make informed choices in line with their preferences.

In severe cases of CDH, care can often involve balancing decisions around aggressive clinical management and palliation, which can be highly distressing for parents to navigate. Treatment typically involves immediate intubation, ventilation (life support), sedation and corrective surgery once they are stabilised. In severe cases, ECMO, (Extracorporeal Membrane Oxygenation), heart and lung bypass is sometimes used. Some consider the use of ECMO controversial due to the risk of severe side effects and complications (Kays, 2017). It is recognised it can be distressing for parents to see. There is significant uncertainty for parents when required to make decisions about their infant's care. This encompasses uncertainty around the diagnosis, prognosis as well as parents' role within the care team (Han et al, 2011). This is particularly prevalent in a neonatal setting, where outcomes are highly variable. As infants are incapable of making decisions around their care, parents are assumed to make decisions based on their family values and what is 'right' for their child (Krick et al, 2020). Disagreements between medical professionals and parents regarding care decisions can be a source of distress. In certain circumstances, clinicians may overrule parental decisions if they pose a risk of harm to the child, guided by the harm principle (Diekema, 2004). However, there remains a significant gap between what is considered optimal care and what constitutes harm, leaving potential for complexity. This gap is often referred to as the 'zone of parental discretion' (ZPD) (Gilliam, 2016), illustrated in Figure 1.

**Figure 1**

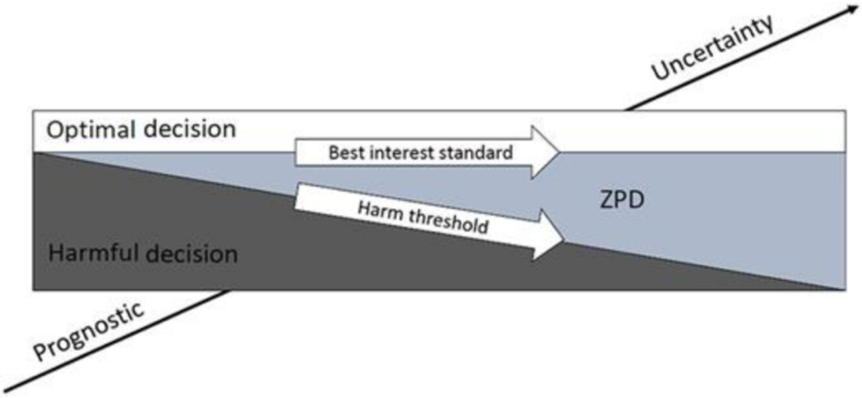
The ZPD



Within this model, parents have the flexibility to make complex decisions based on their own values, regardless of medical opinions. Krick and colleagues propose a development of this model, which incorporates prognostic uncertainty (Krick et al, 2020). This is illustrated in *Figure 2.*

**Figure 2**

Uncertainty and the ZPD: an integrated model



They propose that due to the varying levels of uncertainty within neonatal care, the ZPD will vary in size according to the level of uncertainty. For instance, if there is increased uncertainty about the outcome, the ZPD will be greater, and parents will have more flexibility to make decisions that align with their values. This model incorporates parents' decisions, while providing guidance on where there are limits to this, if there is uncertainty about prognosis. This is particularly relevant for infants diagnosed with CDH, where there is a degree of uncertainty around prognosis at various points from diagnosis to birth (Politis et al, 2021).

A significant amount of research in CDH has focused on treatment methods and survival as the primary outcome. Very few studies to date have focused solely on parents' perspectives and identifying their priorities for decision making and care. One study conducted in the Netherlands examined parents'(n=29) experiences with a child diagnosed with CDH by analysing information on 17 online blogs and three focus groups. They used discourse analysis to examine information that had been written by parents on blogs and the focus groups were discussion forums where parents could provide a written response to questions posed in the online 'room'. They found that insecurities (the main source of distress) were exacerbated by: parents and clinicians having limited specialised knowledge of specific long-term consequences; logistical problems such as waiting times and hospital transfers and non-transparent communication with health professionals. This resulted in feelings of frustration and powerlessness (Steeghs et al, 2019). This highlights the role that uncertainty has in parents' levels of distress, and the need to attempt to mitigate the impact of this where possible. While this study offers insights into parents' experiences, the inclusion criteria for participants were unclear. Additionally, there was a lack of context for the quotes provided, and no medical or demographic information about the parents or their child was included. Further research would deepen understanding of parents experiences. It is important to understand how parents manage their own uncertainty and what information they seek out and value, throughout their child’s care.

### ***Research Aims***

This study will seek to explore parents’ experience of the care they received and the communication they valued from diagnosis to post-birth treatment. It aims to identify key factors influencing parental decision-making about their child's care, providing valuable insights for NICU clinicians treating infants with CDH.

## Methods

### ***Design***

A qualitative design was utilised to explore parents experiences of making decisions during the care of their infant with CDH. Interviews were analysed using Interpretative Phenomenological Analysis (IPA). This method of analysis was selected due to its inductive nature and emphasis on idiographic enquiry and how individuals make sense of their experiences (Smith, 2004). This allowed the researchers to explore personal experiences at a greater depth, within a specific, small population.

### ***Theoretical framework***

IPA is rooted in principles of phenomenology, hermeneutics and idiographic analysis. It takes an exploratory rather than explanatory approach, examining participants' lived experiences on their own terms instead of through predetermined frameworks or hypotheses. The methodology acknowledges that interpretation occurs over two levels; participants interpret their experiences, which researchers then interpret in turn, creating a "double hermeneutics." Rather than pursuing broad population-level generalisations, IPA's idiographic nature emphasises obtaining deeper, nuanced understanding of how individuals understand and experience a specific situation. This approach typically involves conducting semi-structured interviews where the interview schedule is used flexibly, according to what the participant discusses. Smith and colleagues suggest that a sample of between four and ten participants is a suitable number for a project of this nature (Smith, et al, 2009). This small number allows for greater in-depth analysis of any similarities and differences within experiences that may be generated. IPA is noted to be a particularly useful methodology for understanding experiences that may be emotionally complex and is often used in healthcare settings (Smith & Osborn, 2015).

### *Participants*

The study was conducted within a children's hospital in Scotland. Parents were five unrelated parents whose children had received a diagnosis and treatment of CDH between 2021 and 2023. Targeted parents were those who had received a diagnosis at least one year ago, as the purpose of the study was to understand parents' experiences, retrospectively. This also ensured that this process did not interfere with any ongoing clinical decisions. There were no additional exclusion criteria (e.g. parental history of mental health difficulties or bereaved parents). Initial scoping revealed parents with a history of mental health difficulties or who's child did not survive are often excluded from research. Consultation with the parents through the charity CDH UK revealed that bereaved parents expressed a strong desire to be given the opportunity to participate in research of this nature.

Twenty-one parents met eligibility criteria for the study. These participants were identified by clinical staff who hold records for every child diagnosed with CDH. This includes infants who did not survive treatment. Due to the potentially sensitive nature of this a letter (Appendix I) was sent from their neonatal consultant during their period of care. The invitation letter also enclosed a participant information sheet and contact details for the Principal Investigator (PI) (Appendix J). Parents were then asked to contact the PI by email if they were interested in participating. Seven parents contacted the PI expressing interest in participating. Two parents did not provide contact details and did not respond to further correspondence. Five parents were interviewed about their experiences. There were four mothers and one father. Two participants had a child who did not survive treatment. Details of participants and their infant's diagnosis and treatment outcomes are reported in Table 1. All participants have been given a pseudonym. Four participants were White and one participant was Black. These have not been linked to participant details to protect anonymity.

Following the fifth interview, a high level of data richness and recurrence of key themes was observed, suggesting that further data collection was unlikely to yield substantially new insights relevant to the research question. Therefore, the sample was considered sufficient to meet the aims of the study while maintaining the detailed case-by-case analysis central to IPA.

**Table 1**

Participant Characteristics

| **Name** | **Age** | **Gender** | **CDH severity** | **Time infant in NICU** | **Outcome** |
|---|---|---|---|---|---|
| Jane | 32 | F | Left sided (size D) | [redacted] | [redacted] |
| Jon | 32 | M | Left sided (size n/k) | [redacted] | [redacted] |
| Ashley | 27 | F | Left sided (size D) | [redacted] | [redacted] |
| Amy | 35 | F | Left sided (size n/k) | [redacted] | [redacted] |
| Claire | 36 | F | Left sided (size C/D) | [redacted] | [redacted] |

***Interviews***

A semi-structured interview schedule (Appendix L) was developed by the PI in consultation with field supervisors (one consultant neonatologist and specialist in CDH and one consultant clinical psychologist from the maternity and neonatal psychology team) and the research supervisor. The interview schedule followed a chronological structure, from the time of diagnosis (typically at the 20-week scan) to after they were born and the treatment their infant received. Questions also related to how parents experienced making decisions and what factors helped inform this process.

Interviews were arranged to be conducted either online via Microsoft Teams, or in person at the Royal Hospital for Children, according to participant preference. Participants were asked to sign and return their consent form 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. Interviews conducted online were recorded using MS Teams software and stored on the secure NHS Drive. Interviews conducted in person were recorded on a Dictaphone and stored securely. Interviews were transcribed verbatim by the PI. Transcripts were pseudonymised prior to analysis and identifiable information stored separately.

### *Analysis*

Analysis was conducted by the PI, guided by Smith et al (2021), with supervision from the research supervisor. A detailed analysis plan can be located in Appendix M. Transcripts were read multiple times and the PI freely examined any semantic content and language, making exploratory notes. The PI then constructed experiential statements, by attempting to consolidate the key features of the exploratory notes. These were then organised according to 'personal experiential themes' and named according to the experience described. Within these themes, sub-themes were derived based on the original experiential statements. Extracts of the analysis are provided in Appendices N-O. This process was repeated for each individual interview. Themes were then examined across each case, giving attention to similarities and differences, to identify experiences at a group level. To ensure transparency and validity of the analysis, emergent themes were reflected upon in supervision.

### *Reflexivity*

Due to the subjective nature of qualitative research, guidelines recognise the possibility of researcher bias due to the researcher's interactions with participants (Olmos et al, 2022). It is therefore important to explicitly acknowledge their relationship to the research objective and participants to maintain transparency. The PI in this study is a female trainee clinical psychologist. The PI has no children and has not accessed maternity or neonatal health services as a patient. The PI has never worked in maternity or neonatal services but did spend time shadowing members of the team, to understand the service and what is involved in the diagnosis and treatment of CDH. The PI maintained reflective notes throughout the process to document any possible assumptions and biases, discussing these in supervision to enhance methodological rigour and transparency.

### *Ethics*

This study was reviewed by the East of Scotland Research Ethics Committee 3 and ethical approval was granted by the committee (IRAS 339918) and by the NHS Greater Glasgow and Clyde Research and Innovation Department (UGN24MH237). Correspondence is provided in Appendices G-H. The Ethics Committee raised concerns about parental distress during or after the interview if they were discussing potentially highly emotive memories. The PI and interviewer was a Trainee Clinical Psychologist with extensive training and clinical experience in managing high levels of distress and providing a compassionate and supportive response to individuals who are distressed. At the end of each interview the PI had some informal discussion with the participant about how they found the interview experience and directed them to third sector resources. A participant de-brief sheet was also sent that directed participants to the third sector and their GP. The PI also utilised supervision to reflect on the emotional experience of the interview. The Ethics Committee were reassured by the views of parents from CDH UK, who helped inform the inclusion criteria and interview questions.

## Results

Participants in this study were asked to discuss their experiences of CDH diagnosis and treatment, beginning with their pregnancy and diagnosis and following their ‘journey’ through the treatment their child received and the outcome of this. Four themes were identified related to parents’ experiences of care, the communication they valued and what factors were involved in making decisions about their infant’s care. These were explored alongside related subthemes (Table 2). Each theme is supported by verbatim quotes from the interview transcripts. The themes are structured chronologically to align with the way parents were asked to share their journey, reflecting the timeline of their experiences, from receiving the diagnosis, treatment, and looking back. Through these experiences, factors that influence parents’ decisions were explored. This approach helps capture how their perceptions and challenges evolved over different time points. A visual diagram of themes and subthemes is presented in Figure 3.

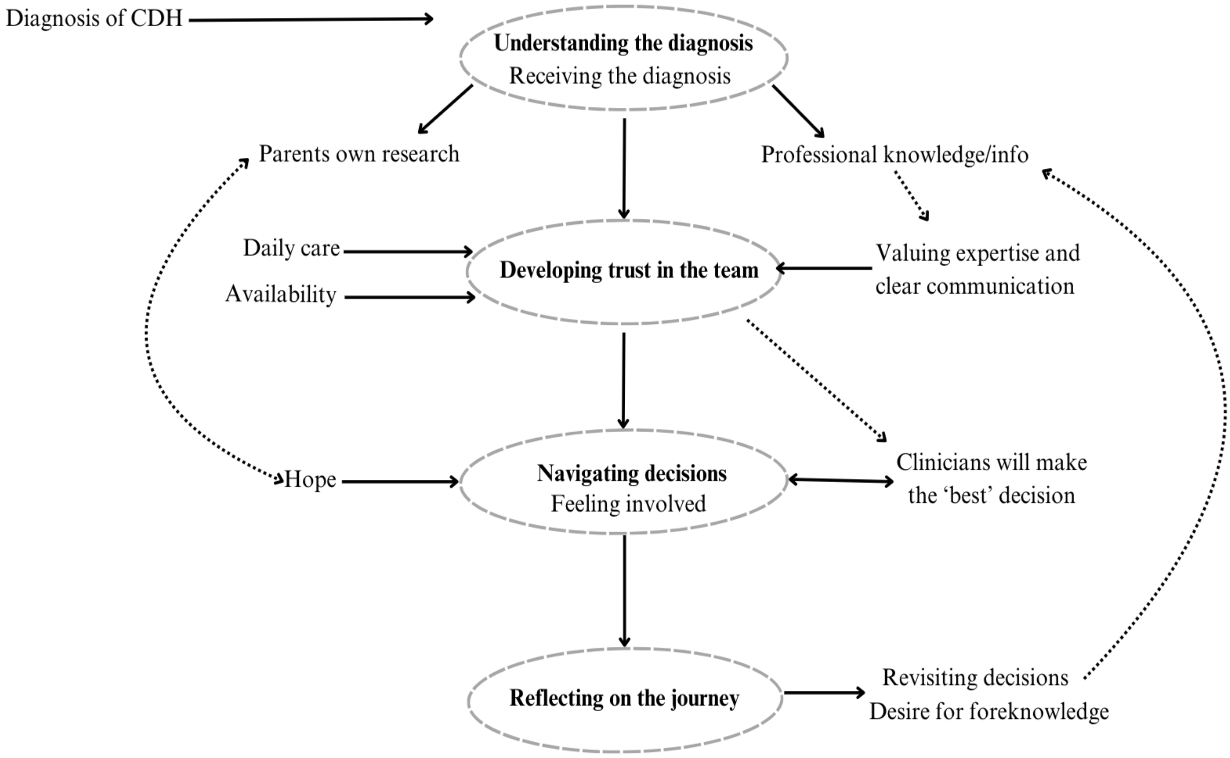
**Table 2**

*Group experiential themes and subthemes*

| Group experiential themes | Subthemes |
|---|---|
| Understanding the diagnosis and the search for answers | Receiving the diagnosis |
| | Seeking professionals' knowledge |
| | Searching for knowledge as parents |
| Developing trust in the team | Feeling confident in baby's daily care |
| | Reassured by specialist knowledge and clear communication |
| | Availability of staff provided comfort |
| Navigating decisions | Feeling involved in decisions |
| | Trust in clinicians to make the best decisions |
| | Hope |
| Reflecting on the journey | Revisiting decisions |
| | Desire for foreknowledge |

**Figure 3**

*Themes and Subthemes.*



Note: Major themes are circled, and subthemes are in standard font

## Understanding the diagnosis and the search for answers

### ***Receiving the diagnosis***

Most parents received the CDH diagnosis at the 20-week scan. Most parents seemed to remember this moment in detail and the initial shock they experienced was evident. Parents seemed to find it difficult to process information provided at the time, and perhaps focused on the details that carried the most emotional weight. Both Claire and Ashley described their experiences of the diagnosis being communicated in a way that heightened their anxiety.

> *"...my world had been turned upside down because then she's talking about survival chances and severity...I was terrified, I was devastated...I just thought...there's no chance she's going to survive ...* (Claire)
> *"I mean everything, everything they told us every time it was just bad news, after bad news...* (Ashley)

Information Claire and Ashley were given by clinicians around severity and survival chances was clearly distressing. Perhaps this level of transparency was not valued at this point, while parents were still adjusting to the sudden news of the diagnosis. Their descriptions of receiving the news highlight the significant emotional impact of the diagnosis and suggests that the way information was communicated to them may have influenced their ability to process, understand, and come to terms with their situation.

### ***Seeking professionals' knowledge***

None of the parents were familiar with CDH before the diagnosis. They all expressed a need to know more about how the diagnosis might impact their child and what it could mean for their future. There was variation in the knowledge of those delivering the diagnosis, which appeared to influence how parents processed the news. This was partly due to where parents lived, as their local hospital was not a specialist centre, but also reflected generic maternity services.

Claire, who described her pregnancy as "*a very stressful time*" expressed that her local hospital and GP did not seem to have much knowledge about CDH:

> *"…it just felt like CDH was another language, nobody knew what it was, nobody understood"*. (Claire)

Ashley also had a similar experience at her local hospital when her baby was diagnosed:

> *"I got nothing, like no kind of knowledge. So I kind of just left with what they told me…I do feel like maybe a wee leaflet or something would have went a long way..."*. (Ashley)

The use of the phrase 'another language' to describe knowledge about CDH among some care services conveys the sense of unfamiliarity and disconnect that parents perceived in these settings. Some basic knowledge provided by clinicians during the early stages of pregnancy might have helped parents manage their anxiety. This could have alleviated some of the uncertainty they felt about the future implications of the diagnosis.

***Searching for knowledge as parents***

As their pregnancies progressed, parents varied in terms of how much they had sought information about potential treatment and what the diagnosis meant for their child's future. For some, this was an additional source of uncertainty and worry and for others was an important part of their decision about whether to continue with the pregnancy. Some parents always knew they wanted to continue with the pregnancy, and prioritised finding out more about treatment, and what the potential implications of this might be:

> *"I even went down as far as looking to see what kind of scars he's be left with… So I saw for babies having the trachea *[tracheostomy]* put in as well as the scar and being left on oxygen, feeding tubes… To the other end, where it was just literally keyhole."* (Amy)

At this stage, it was still uncertain what treatment he would require. While learning about potential treatment outcomes might have heightened anxiety, it also seemed to provide a sense of relief by offering some clarity.

Although the desire for knowledge was evident, most parents described actively avoiding stories where CDH had resulted in a tragic outcome while conducting their own research. This was summarised well by Jon:

> "*you're not really looking for the worst things on the websites and on the forums. You're looking for the best-case scenarios. When people say the hope dies last***,** *that's true.*" (Jon)

While parents expressed a strong desire for knowledge, maintaining hope seemed even more important to them during the pregnancy. Despite the uncertainty of the future, the information they received from professionals and their own research provided some reassurance, perhaps by offering them a sense of clarity in a situation still fraught with uncertainty. Parents placed great value on information that instilled hope and appeared reluctant to consider potential negative outcomes. This may also reflect parents individual coping styles, in an attempt to maintain their emotional wellbeing throughout the pregnancy.

### Developing Trust in the Team

#### *Feeling confident in baby's daily care*

Daily interactions with staff and experiences on the unit shaped parents' perceptions of the care their child was receiving. This was important for developing a sense of trust with clinicians, which had the potential to ease distress. Ashley describes how important it was to feel her baby was being care for the same way she would:

> *"Just having someone that looked after her the way I would have looked after her, obviously if she was at home...and they all kind of looked after her as if she was their own"* (Ashley)

This seemed to help her build a sense of trust with the nursing staff as well as the wider neonatal team. In contrast, Claire perceived the care her baby received as being of a different quality, which was a significant source of stress for her.

> *"...you are leaving your baby in the care of someone else and you expect them to be looked after, you know the basics and to me they were basic things...that was probably one of the most stressful things because you're completely powerless"* (Claire)

While Claire felt this experience was limited to some staff, it seemed to challenge her role as a parent and left her feeling powerless. Although she did not directly acknowledge that this affected her trust in other clinicians, it highlights the importance of daily nursing care being as crucial as specialised treatment in fostering trust. This trust may be a key factor in parents' confidence in the abilities and care provided by the neonatal team.

### ***Reassured by specialist knowledge and clear communication***

The specialist knowledge clinicians had about CDH was also important for building trust. This was evident at various stages in each of the parents' journeys. For Claire, this shift was notable when her baby's care was transferred to a specialist care centre:

> *"It totally changed when we came to appointments here and we felt safer here because of the knowledge that they had."* (Claire)

Parents also described feeling very reassured by the experience of the surgeons, as well as the detailed medical information they were provided with throughout their stay in the NICU. Parents noted how some clinicians had extensive knowledge and experience in treating CDH, which they valued and felt reassured by.

Clear and transparent communication was important for allowing parents to feel involved in aspects of their child's care. All parents described feeling comfortable approaching nurses, doctors and surgeons, and valued their efforts to try and provide them with some clarity.

> *"if there's ever anything that I'm unsure about or don't agree with, I will just say and they'll explain it a bit further to me so I do understand that obviously, and they always explain why they're doing certain things like changing certain things"* (Ashley)

Most parents also commented on the availability of clinicians to answer their questions, suggesting an ongoing, collaborative approach. The level of transparency between parents and clinicians seemed to ease some uncertainty and fostered a sense of collaboration. It was clear that the knowledge shared, whether through clear communication or through the confidence that comes from understanding the professionals' experience, parents' anxieties were significantly reduced.

### ***Availability of staff provided comfort***

All parents also spoke of how much they valued the availability of staff to be able to answer their questions. Most parents described having very regular contact with the doctors and surgeons and were always reassured by nursing staff that they could phone at any time and as often as they wanted.

> *I could talk to him all day, every day, if I needed to. So it was…it was really, really good that way with me, so he was."* (Amy)

Where one parent did not experience this, partly due to being transferred to a local hospital, they named it as being something that would have helped significantly. This highlights how important accessible and open communication was for ensuring parents felt informed and supported.

### Navigating decisions together

#### *Feeling involved in decisions*

All parents shared that they felt involved in decisions about their care. However, there was an emotional burden of parents' involvement in decision making. Two parents described feeling involved in their infants' care but simultaneously felt limited in their choices.

> *"ehm…it's…it just felt like you were forced. But you weren't- I'm not saying that anyone forced us. It just felt like you had no choice. And again it was just part of that lack of control over anything was just you know…this was happening whether you liked it or not and this was what was best for her at the time"* (Claire)

Here, Claire struggles to articulate her conflicting feelings. While she felt very involved in her child's care, she also experienced a sense of having no control over what was happening. This internal conflict was shared by other parents, who expressed feeling that, ultimately, they had to give choice over to clinicians, to ensure their child received the life-saving treatment they needed. This was particularly evident in discussions about major surgeries and critical moments when urgent care was required. One parent expressed relief that she fortunately did not have any major decisions to make, as her son's treatment was relatively straightforward. Regardless of whether they were directly involved in decision-making, they still reported feelings of helplessness. This perhaps reflected the frustration of feeling 'involved' yet not having the ability to directly influence their child's condition.

### *Trust in clinicians to make the best decisions*

Parents’ confidence in clinicians’ decisions was particularly evident during critical moments when they felt uncertain or powerless. They seemed to have faith that the neonatologists and surgeons, specifically knew ‘best’. This stemmed from the strong foundation of trust that had already been established. It played a crucial role in helping parents navigate highly distressing situations, such as their child undergoing surgery or making decisions about life support and end-of-life care. This was apparent for Jon and Ashley when they were considering ECMO:

> *“Obviously he couldn't tell me what to do, but I just needed to hear from someone that we were maybe doing the right thing.” (*Ashley)
>
> *“They said in a way it would be like a torture, so we just, there's no point pushing them to ask to, to connect her to the ECMO more...It was quite important knowing that, you know, there was, there was just no more options. Obviously I can, you can deal with it. So yeah…”* (Jon)

Jane also described the moment where one of the doctors discussed withdrawing care for her child:

> *“Doctor [*redacted*] told us that there's nothing they could do anymore and he would like to turn off the oxygen now because that's what’s doing everything for them. And even at that it’s not better. That he is not asking us to make that decision, that he would make that decision for us. “*(Jane)

All parents appeared to trust that clinicians would make the most informed and compassionate decisions on behalf of their child. For parents of babies who did not survive, knowing that doctors had exhausted all possible options and were guided by their baby’s best interests helped them come to terms with the situation. The use of emotive language, such as ‘torture,’ seemed to reinforce their certainty that further treatment was unlikely to succeed. Similarly, Jane noted the directness of her doctor, whose explicit and unambiguous language removed any uncertainty about the decision. This clarity may have provided parents with a small sense of comfort, knowing they were not left to face such a difficult decision alone.

### *Hope*

Holding onto hope was a protective factor for most parents, providing them with the strength to persevere. This seemed to manifest in how they viewed their baby. Parents often referred to their child as being a ‘fighter’:

> “*I was kind of hopeful that, you know, it was going to be ok because he was a fighter… because he was, you know. He was getting through everything” (*Jane)

There was a sense that their child had a natural resilience or ‘strength’ which was as important as their clinical prognosis, in influencing their recovery. Additionally, parents appeared to grow more hopeful as their child survived various setbacks. This sense of resilience may have reinforced their belief that their child could overcome further challenges, motivating them to pursue additional treatment. Hope not only seemed to help parents to persevere, but it perhaps guided their decision to pursue further treatment or affirmed their choices during challenging situations.

## Reflecting on the journey

Parents also discussed the decisions they made, the feelings of agency, and how they would have approached things differently with more knowledge or time to reflect. Sometimes this was related to knowing more during pregnancy whereas at other times it was about what specific treatment would be involved.

### *Revisiting Decisions*

Jane and Jon, whose infants did not survive, both reflected on their care in a different way. Jon held the view that it was impossible to see into the future and know what the outcome would have been. There was a sense that he did not find it helpful to look back and think about what he might have wanted to be different. While this could be considered a positive way of coping, it might serve as a protective mechanism for him.

Whereas Jane was very emotional when reflecting back on decisions that were made about her child’s care before they died. Here she describes wishing she had known more about the potential side effects of a specific treatment.

> *“The things I regret most was allowing them to give [*redacted*] because maybe if I had known better, I would have made an informed choice...”* She later continued *“and*

> *that just made me feel really bad because it felt like I could have just let him go more peacefully... I felt like I made a decision to worsen his situation"*

Although this decision was made with the best intentions and with the support of the medical team, Jane seems to regret that he was given this treatment, as she feels she was not fully aware of how it could have affected him. Making these decisions under such complex and uncertain circumstances is profoundly challenging, and Jane's experience highlights the painful feelings parents can be left processing when their baby does not survive.

***Desire for foreknowledge***

Parents had varying expectations about the realities and complexities of their child's treatment. Some parents expressed not being prepared for when surgery would take place and how long it would take. One parent spoke about the shock and the emotional impact of seeing her baby on ECMO, which reflects how parents may not always anticipate the full scope of medical interventions. The majority of parents did not expect there to be so much complexity. One parent reflected on how things might have been different had they known what was going to happen.

> *"Looking back if I had the knowledge I had then I don't know…I actually don't know whether…All I can say now is that if I was told I had a future pregnancy and they had CDH I wouldn't go through with it."* (Claire)

Although Claire does not seem to regret any of her decisions, likely because her child is now doing well, she may not have fully anticipated the level of complexity or stress that she would face. Ashely shared that, while she wished she had done more research, doing so would have increased her anxiety. Parents' expectations around treatment, whether in terms of timing, complexity, or ongoing care, were often met with unexpected realities. Claire's perspective could be seen as pragmatic, whereas other parents adopted more idealistic approaches.  Naturally, parents' reflections may mirror the variation in prognosis of CDH, and, to some extent, their differing approaches to facing challenges.

## Discussion

The current study explored parents' experiences of care of their child with a diagnosis of CDH and what factors contributed to their decision making during their child's treatment. Four themes and eleven subthemes were developed through the analysis. Parents described their journey as a process of understanding the diagnosis and searching for answers, developing trust in the medical team, navigating decisions, and reflecting on their experiences.

Participants described receiving the diagnosis as sudden and unexpected. This is consistent with previous research which found parents experience shock, disbelief and anger following a diagnosis of foetal abnormality (Carlssson et al., 2017). Their lack of knowledge around the condition meant they looked to professionals for support as well as trying to search for information independently. Parents reported variability in professionals' knowledge when delivering the diagnosis. This aligns with Luz et al.'s (2016) review, which highlights how the quality of communication and information provided by professionals significantly influences how parents interpret and process their child's diagnosis. Parents varied in how much information they sought about the diagnosis, with some noting that it played a role in their decision about whether to continue the pregnancy. However, it did not appear to be the decisive factor for most, suggesting they perhaps focused on the potential for a positive outcome. Previous research suggests that disease severity and additional diagnoses were the main factors in decisions to terminate (Horn-Oudshoorn et al., 2023), however these results would suggest it is a more nuanced process.

Trust in medical staff emerged as a key theme while infants were receiving treatment. This seemed to manifest through how parents perceived the quality of the daily care, the specialist knowledge and expertise in CDH, and through a process of transparent and regular communication. This appeared to facilitate parents trusting clinicians to make the 'best' decisions on behalf of their child. This validates and expands the findings of Petit-Steeghs's and colleagues (2019) qualitative study of online forums for parents of CDH infants. They found that the lack of specialised knowledge significantly exacerbated parent's distress. Bry and Wigert (2019), who interviewed parents of preterm babies also found that when this trust was impaired, either through a lack of specialist knowledge or their perceptions of care, parents stress was exacerbated. Communication also appeared to be important for creating trust. As with specialist knowledge, non-transparent communication with health professionals also emerged as contributor to parents' dissatisfaction with care in Petit-Steegh et al's (2019) study. This points to why transparency and the accessibility of clinicians may have helped to create a sense of collaboration between parents and clinicians, contributing to the development of a trusting relationship. Petit-Steegh et al (2019) describe this as affecting parents' sense of control. The current study results suggest that this sense of collaboration, fostered by transparency and availability of clinicians, may mitigate parents' distress through reducing the level of uncertainty, to some degree. These findings deepen findings of previous research, highlighting the potential mechanisms through which trust is developed.

Navigating decisions was an important part of parents' experiences of care in this study. Although there was a sense that parents felt involved, the degree to which control could be exerted over this felt limited, particularly when it came to critical moments. Petit-Steegh et al (2019) suggested that parents wished they were more involved in decisions pertaining to hospital transfers and treatment options. Parents in the current study generally described a sense of powerlessness when it came to making major decisions about their child's care. This may be due to the severity of CDH in the majority of cases in this study, meaning parents sense of powerlessness was more pronounced. Parents appeared to accept their influence on treatment options were somewhat limited, although generally felt involved. Qualitative research exploring parents' experiences of palliative care found a paradox in their involvement in decision-making. While parents acknowledged having only one viable choice, they still perceived the process as conscious and deliberative (Denny et al., 2023). Similarly, in the current study, parents' experiences of being involved in decisions also appeared paradoxical. They felt limited in their choices, particularly in decisions involving life-sustaining treatments or, conversely, withdrawing care. Despite expressing a sense of helplessness, feeling involved in the decision-making process perhaps affirms their role as a parent, by actively contributing to their care. Parents' sense of involvement and their perception of themselves as important agents in their child's recovery seemed closely tied to the trust they developed in the neonatal team. This trust allowed parents to rely on doctors to make the 'best' decisions on behalf of their child. Parents found reassurance in their belief that clinicians prioritised their child's best interests, further reducing the emotional burden of making difficult decisions.

Having hope was also central to parents navigating decisions. Hope is known to be a significant coping mechanism for parents in a neonatal context, for managing the emotional impact of uncertainty (Silveria et al., 2023). The current study suggests hope might influence parents' decisions, in the sense that some parents perceived their child to be 'strong' or resilient, because they had survived previous challenges. This helped them to persevere with treatment, underpinned by the hope for a successful outcome. The intersection of parental involvement and trust highlights a delicate balance: parents desired to feel included in decision-making while simultaneously relying on clinicians' knowledge and expertise to determine what was ultimately 'best' for their child. Hope appeared to play a pivotal role in sustaining parents' perseverance through these challenges. These dynamics highlight the emotional complexity of shared decision-making, with trust in clinicians acting as a stabilising force in the face of an often uncertain outcome.

Finally, parents' reflections on their journey revealed a complex interplay of agency, emotional coping, and evolving perspectives on their experiences. While some accepted the uncertainty of outcomes, others struggled with regret or a sense of missed opportunities for more informed decision-making. The variation in reactions, from not dwelling on the 'what-ifs' versus feelings of regret and self-blame has been observed in recent research investigating the experiences of bereaved parents (Feifer et al., 2023; Denny et al., 2024), with the former being argued as a protective mechanism from experience regret (Denny et al., 2024). It seems that this desire for more knowledge, which was also expressed by parents in the early stages of pregnancy, is also paradoxical in nature, as parents also described actively avoiding or rejecting information that felt overwhelming or discouraging. This aligns with research that an 'appropriate' level of information is required for parents to minimise anxiety and distress (Luz et al., 2015). These findings suggest that providing tailored information in manageable amounts, and at the relevant 'stages' of care/treatment, alongside emotional support, may help parents feel more prepared without overwhelming them.

The findings of this study suggest that there is a complex interplay of factors that inform parental decision making in CDH care. Krick et al's (2020) model of uncertainty and the ZPD suggests that the parents have greater flexibility to make decisions about their child's care, that align with their values, where there is greater prognostic uncertainty and while maintaining thresholds around best interests and harm. The results of this study would suggest that although parents still have a 'choice', their anxiety increases as the level of uncertainty increases, causing them to rely more heavily on clinicians to guide decisions about their child's care. Clinicians may then adopt a more directive approach, which can alleviate parental distress to a degree, by reducing the emotional burden of making decisions. The trust that has previously been established with the medical team, as described earlier, enables parents to process information and come to terms with their situation.

## Strengths and limitations

To the author's knowledge, this is the first study in the UK that has examined parent's experiences of care and decision-making pertaining to their infants CDH, using a qualitative approach. This data provides a rich account of what parents receiving a diagnosis of CDH might experience. The results deepen preliminary research in this area (Petit-Steegh et al., 2019), providing insight into how the impact of uncertainty may be mitigated, and the emotional complexity involved in navigating shared decisions. The sample of parents who participated all received care between 2021 and 2022. This provided a relatively homogenous sample, as recommended by IPA, therefore contextual factors which may have affected participants experiences (such as the covid-19 pandemic) was consistent for all participants. However, there are a several limitations that impact the reliability and generalisability of the findings. The outcomes of treatment were vastly different for the participants, with two children not surviving, and one with considerable complex, ongoing needs. While it was valuable to represent the spectrum of experiences that can arise from a CDH diagnosis, the varied nature of these experiences makes it challenging to draw generalised conclusions.

The semi-structured nature of the interview allowed participants to freely recall their experiences and subsequent decision making. As the researcher conducting the interviews was not part of the medical team, this may have influenced how parents disclosed their experiences. The separation from the clinical context could have created a sense of neutrality, perhaps allowing parents to feel freer to express their thoughts and emotions about the care and treatment they received. While this retrospective account creates space for reflection, and provides opportunities for learning from parents' experiences, it may not be possible to generalise these findings to parents currently experiencing diagnosis/treatment. It also meant that the chronological way in which participants told their 'stories' may have influenced the themes identified, as opposed to participants spontaneously presenting their experiences. Additionally, while this time period was relatively recent, participants recall of events may have been slightly impaired. As previously noted, the principal investigator had no prior experience working in or accessing maternity services. However, their training in clinical psychology provided a strong foundation in critical self-reflection, which was practiced throughout the research process. While the researcher did not share the lived experiences of participants, this distance may have facilitated a more objective and balanced analysis by preventing over-identification with the interview content. Nonetheless, a deeper understanding of the medical aspects of CDH treatment may have enriched the contextual interpretation of participants' narratives.

### Implications for clinical practice and policy

Despite the extensive body of work focused on supporting parents in the NICU, few studies have explored the in-depth experiences of decision-making, particularly in the context of CDH. This study provides valuable insights for clinicians into what parents experience and highlights the potential gap between clinicians' expectations and parents' realities. It is evident that providing an appropriate amount of information at the point of diagnosis is highly valued by parents. While some parents expressed a desire for more detailed knowledge about what treatment might involve, care providers must balance this need with maintaining hope, especially as parents process the diagnosis. Standardised information packs, distributed across all centres involved in diagnosis rather than only specialist centres, could address this need and ensure consistent support for parents. The type of information parents seek may evolve if their child requires further, potentially more complex treatment. Staff should strive to provide clear and transparent information about potential side effects and prognosis while thoughtfully balancing this with maintaining a hopeful and supportive tone for parents. The development of clear policy guidance could ensure consistent and transparent delivery of diagnostic information, across different health boards.

Transparent, regular, and empathetic communication was highly valued by parents and considered essential for building trust and helping them feel involved in their infant's care. Maintaining this standard of communication should remain a central priority in neonatal settings. Equally important is the need for healthcare professionals to acknowledge the significant emotional burden parents carry when faced with complex decisions. Families approach these decisions with diverse values, priorities, and beliefs, particularly regarding interventions such as termination or life-sustaining treatment. While some parents look to clinicians to lead or even make the most difficult decisions, many still wish to feel included and respected as part of the process. Policy frameworks should therefore support clinicians in engaging with families in a non-directive and respectful manner, allowing space for cultural, ethical, and personal perspectives to inform care planning. Future research can contribute to these efforts by identifying specific stages in the care journey where informational and emotional support is most needed and effective.

In addition to structured support for parents, it is also crucial to provide reflective spaces for staff. These can help clinicians process their own emotional responses to ethically challenging situations and maintain compassionate, patient-centred care.

## Recommendations for future research

Future research could focus on the experiences of staff, exploring their perspectives on making complex decisions and examining their priorities. Comparing points of similarity and divergence between staff and parents could provide a more comprehensive understanding of the decision-making process in this context.

## Conclusions

This study explored parents' experiences of care and decision-making following their infant's diagnosis of CDH. Their experiences highlight the importance of accessible specialist information and knowledge, which play a key role in building trusting relationships with clinicians. These relationships, in turn, shape parents' decision-making experiences and help mitigate the emotional impact of the diagnosis, both during the acute stages of treatment and afterward, as they process what has happened. Hope emerged as a recurring theme throughout each stage of their journey, influencing parents' actions—from seeking stories that reinforced their hopes for their child's prognosis to shaping how they viewed their child and approached decisions. This study provides valuable insights into the experiences and needs of parents within this population, offering important implications for future research and clinical practice.

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# Appendices

## Chapter 1: Systematic Review

## Chapter 2: Major Research Project

# Appendix A. Systemic Review Registration

Prospero Registration number: CRD420256283

# Appendix B. PRISMA Reporting Guidelines

## PRISMA 2020 Checklist

| Section and Topic | Item # | Checklist item | Location where item is reported |
|---|---|---|---|
| **TITLE** | | | |
| Title | 1 | Identify the report as a systematic review. | |
| **ABSTRACT** | | | |
| Abstract | 2 | See the PRISMA 2020 for Abstracts checklist. | |
| **INTRODUCTION** | | | |
| Rationale | 3 | Describe the rationale for the review in the context of existing knowledge. | |
| Objectives | 4 | Provide an explicit statement of the objective(s) or question(s) the review addresses. | |
| **METHODS** | | | |
| Eligibility criteria | 5 | Specify the inclusion and exclusion criteria for the review and how studies were grouped for the syntheses. | |
| Information sources | 6 | Specify all databases, registers, websites, organisations, reference lists and other sources searched or consulted to identify studies. Specify the date when each source was last searched or consulted. | |
| Search strategy | 7 | Present the full search strategies for all databases, registers and websites, including any filters and limits used. | |
| Selection process | 8 | Specify the methods used to decide whether a study met the inclusion criteria of the review, including how many reviewers screened each record and each report retrieved, whether they worked independently, and if applicable, details of automation tools used in the process. | |
| Data collection process | 9 | Specify the methods used to collect data from reports, including how many reviewers collected data from each report, whether they worked independently, any processes for obtaining or confirming data from study investigators, and if applicable, details of automation tools used in the process. | |
| Data items | 10a | List and define all outcomes for which data were sought. Specify whether all results that were compatible with each outcome domain in each study were sought (e.g. for all measures, time points, analyses), and if not, the methods used to decide which results to collect. | |
| | 10b | List and define all other variables for which data were sought (e.g. participant and intervention characteristics, funding sources). Describe any assumptions made about any missing or unclear information. | |
| Study risk of bias assessment | 11 | Specify the methods used to assess risk of bias in the included studies, including details of the tool(s) used, how many reviewers assessed each study and whether they worked independently, and if applicable, details of automation tools used in the process. | |
| Effect measures | 12 | Specify for each outcome the effect measure(s) (e.g. risk ratio, mean difference) used in the synthesis or presentation of results. | |
| Synthesis methods | 13a | Describe the processes used to decide which studies were eligible for each synthesis (e.g. tabulating the study intervention characteristics and comparing against the planned groups for each synthesis (item #5)). | |
| | 13b | Describe any methods required to prepare the data for presentation or synthesis, such as handling of missing summary statistics, or data conversions. | |
| | 13c | Describe any methods used to tabulate or visually display results of individual studies and syntheses. | |
| | 13d | Describe any methods used to synthesize results and provide a rationale for the choice(s). If meta-analysis was performed, describe the model(s), method(s) to identify the presence and extent of statistical heterogeneity, and software package(s) used. | |
| | 13e | Describe any methods used to explore possible causes of heterogeneity among study results (e.g. subgroup analysis, meta-regression). | |
| | 13f | Describe any sensitivity analyses conducted to assess robustness of the synthesized results. | |
| Reporting bias assessment | 14 | Describe any methods used to assess risk of bias due to missing results in a synthesis (arising from reporting biases). | |
| Certainty assessment | 15 | Describe any methods used to assess certainty (or confidence) in the body of evidence for an outcome. | |

# PRISMA 2020 Checklist

| Section and Topic | Item # | Checklist item | Location where item is reported |
|---|---|---|---|
| **RESULTS** | | | |
| Study selection | 16a | Describe the results of the search and selection process, from the number of records identified in the search to the number of studies included in the review, ideally using a flow diagram. | |
| | 16b | Cite studies that might appear to meet the inclusion criteria, but which were excluded, and explain why they were excluded. | |
| Study characteristics | 17 | Cite each included study and present its characteristics. | |
| Risk of bias in studies | 18 | Present assessments of risk of bias for each included study. | |
| Results of individual studies | 19 | For all outcomes, present, for each study: (a) summary statistics for each group (where appropriate) and (b) an effect estimate and its precision (e.g. confidence/credible interval), ideally using structured tables or plots. | |
| Results of syntheses | 20a | For each synthesis, briefly summarise the characteristics and risk of bias among contributing studies. | |
| | 20b | Present results of all statistical syntheses conducted. If meta-analysis was done, present for each the summary estimate and its precision (e.g. confidence/credible interval) and measures of statistical heterogeneity. If comparing groups, describe the direction of the effect. | |
| | 20c | Present results of all investigations of possible causes of heterogeneity among study results. | |
| | 20d | Present results of all sensitivity analyses conducted to assess the robustness of the synthesized results. | |
| Reporting biases | 21 | Present assessments of risk of bias due to missing results (arising from reporting biases) for each synthesis assessed. | |
| Certainty of evidence | 22 | Present assessments of certainty (or confidence) in the body of evidence for each outcome assessed. | |
| **DISCUSSION** | | | |
| Discussion | 23a | Provide a general interpretation of the results in the context of other evidence. | |
| | 23b | Discuss any limitations of the evidence included in the review. | |
| | 23c | Discuss any limitations of the review processes used. | |
| | 23d | Discuss implications of the results for practice, policy, and future research. | |
| **OTHER INFORMATION** | | | |
| Registration and protocol | 24a | Provide registration information for the review, including register name and registration number, or state that the review was not registered. | |
| | 24b | Indicate where the review protocol can be accessed, or state that a protocol was not prepared. | |
| | 24c | Describe and explain any amendments to information provided at registration or in the protocol. | |
| Support | 25 | Describe sources of financial or non-financial support for the review, and the role of the funders or sponsors in the review. | |
| Competing interests | 26 | Declare any competing interests of review authors. | |
| Availability of data, code and other materials | 27 | Report which of the following are publicly available and where they can be found: template data collection forms; data extracted from included studies; data used for all analyses; analytic code; any other materials used in the review. | |

*From:* Page MJ, McKenzie JE, Bossuyt PM, Boutron I, Hoffmann TC, Mulrow CD, et al. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. BMJ 2021;372:n71. doi: 10.1136/bmj.n71.

Appendix C. Example Search Strategy

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

EMBASE

| | | |
|---|---|---|
| 1 | parents/ or fathers/ or mothers/ or single parent/ | 204144 |
| 2 | caregiver/ | 130366 |
| 3 | ("Parent*" or "Mother*" or "Father*" or "Caregiver*").ab,ti. | 956213 |
| 4 | anxiety/ or anxiety disorder/ | 394511 |
| 5 | depression/ | 491536 |
| 6 | physiological stress/ | 209785 |
| 7 | distress syndrome/ | 72512 |
| 8 | ("Parenting stress" or "Parental distress" or "Parental anxiety" or "Parental depression" or "parental coping").ab,ti. | 6655 |
| 9 | ("stress" or "distress" or "Emotion*" or "Anxi*" or "Depression" or "Coping" or "Mental health" or "Psychological*").ab,ti. | 2592147 |
| 10 | prematurity/ or newborn intensive care/ or neonatal intensive care unit/ | 158423 |
| 11 | 1 or 2 or 3 | 1004305 |
| 12 | ("NICU" or "Neonatal Intensive Care Neonatal Care" or "Neonatal Unit" or "Premature infant*" or "Preterm infant*" or "neo-nat*").ab,ti. | 80982 |
| 13 | 4 or 5 or 6 or 7 or 8 or 9 | 2797591 |
| 14 | 10 or 12 | 176081 |
| 15 | clinical trial/ | 954645 |
| 16 | randomization/ | 91937 |
| 17 | intervention study/ | 71405 |
| 18 | (therap* or intervention*).ab,ti. | 6406511 |
| 19 | 15 or 16 or 17 or 18 | 7029386 |
| 20 | 11 and 13 and 14 and 19 | 2654 |
| 21 | limit 20 to (english language and "remove medline records") | 1124 |

Appendix D. Certainty of Evidence

## Applying the GRADE approach when evidence for an effect is summarised narratively (a meta-analysis is not available)

| **GRADE domain** | **How to apply the GRADE domain to evidence that has been summarised narratively** |
|---|---|
| Methodological limitations of the studies | Make a judgement on the risk of bias across studies for an individual outcome. A sensitivity analysis is not possible to determine if the effect changes when studies at high risk of bias are excluded. It is possible to consider the size of a study, its risk of bias and the impact it would have on the summarised effect. |
| Indirectness | Make a global judgement on how dissimilar the research evidence is to the clinical question at hand (in terms of population, interventions and outcomes across studies). |
| Imprecision | Consider the optimal information size (or the total number of events for binary outcomes and the number of participants in continuous outcomes) across all studies. A threshold of 400 or less is concerning for imprecision.[15] Results may also be imprecise when the CIs of all the studies or of the largest studies include no effect and clinically meaningful benefits or harms. |
| Inconsistency | Judge inconsistency by evaluating the consistency of the direction and primarily the difference in the magnitude of effects across studies (since statistical measures of heterogeneity are not available). Widely differing estimates of the effects indicate inconsistency. |
| Likelihood of publication bias | Publication bias can be suspected when the body of evidence consists of only small positive studies or when studies are reported in trial registries but not published. Statistical evaluation of publication bias is not possible in this case. Publication bias is more likely if the search of the systematic review is not comprehensive. |

| | |
|---|---|
| Factors that can raise certainty in evidence:<br>• Large effect<br>• Dose–response gradient<br>• Plausible confounders or other biases increase the certainty in the effect | If one of the three domains that can increase certainty in a body of evidence (typically from non-randomised studies) is noted, consider rating up the grade of certainty, particularly if it is noted in the majority of studies. |

| **GRADE Domain** | **Judgement** | **Concerns about certainty domains** |
|---|---|---|
| Methodological limitations of the studies | Of 15 RCTs, all presented with some- high risk of bias. 9 studies did not report effect sizes and 5 studies did not report means or SDs either at baseline or post-treatment. Studies are deemed to have serious methodological limitations. | Serious |
| Indirectness | Participants, interventions and measures all provide direct evidence regarding the research question. There was some variability in type of intervention administered and duration of the intervention. Most had a similar model of delivery. There was significant heterogeneity in outcome measures however the research is deemed as direct. | Not serious |
| Imprecision | The total number of participants included across trials was 1087. Two studies did not include sample size calculations, of which both reported insignificant results. Evidence has borderline imprecision. | Borderline, not serious. |
| Inconsistency | The presence and size of effect varied across studies. Effect size was not reported in 9 studies. Of studies reported they ranged from small-large. 11 studies showed significant effect. | Serious |
| Likelihood of publication bias | Bias not suspected as throughout screening and search completed. Studies reporting no effect were also reported. | Not suspected. |

## Appendix E. CORTEQ Reporting Checklist

| No | Item | Guide questions/description |
|---|---|---|
| **Domain 1: Research team and reflexivity** | | |
| Personal Characteristics | | |
| 1. | Interviewer/facilitator | Which author/s conducted the interview or focus group? |
| 2. | Credentials | What were the researcher's credentials? *E.g. PhD, MD* |
| 3. | Occupation | What was their occupation at the time of the study? |
| 4. | Gender | Was the researcher male or female? |
| 5. | Experience and training | What experience or training did the researcher have? |
| Relationship with participants | | |
| 6. | Relationship established | Was a relationship established prior to study commencement? |
| 7. | Participant knowledge of the interviewer | What did the participants know about the researcher? e*.g. personal goals, reasons for doing the research* |
| 8. | Interviewer characteristics | What characteristics were reported about the interviewer/facilitator? e.g. *Bias, assumptions, reasons and interests in the research topic* |
| **Domain 2: study design** | | |
| Theoretical framework | | |
| 9. | Methodological orientation and Theory | What methodological orientation was stated to underpin the study? *e.g. grounded theory, discourse analysis, ethnography, phenomenology, content analysis* |

| No | Item | Guide questions/description |
|---|---|---|
| Participant selection | | |
| 10. | Sampling | How were participants selected? *e.g. purposive, convenience, consecutive, snowball* |
| 11. | Method of approach | How were participants approached? e*.g. face-to-face, telephone, mail, email* |
| 12. | Sample size | How many participants were in the study? |
| 13. | Non-participation | How many people refused to participate or dropped out? Reasons? |
| Setting | | |
| 14. | Setting of data collection | Where was the data collected? e*.g. home, clinic, workplace* |
| 15. | Presence of non-participants | Was anyone else present besides the participants and researchers? |
| 16. | Description of sample | What are the important characteristics of the sample? *e.g. demographic data, date* |
| Data collection | | |
| 17. | Interview guide | Were questions, prompts, guides provided by the authors? Was it pilot tested? |
| 18. | Repeat interviews | Were repeat interviews carried out? If yes, how many? |
| 19. | Audio/visual recording | Did the research use audio or visual recording to collect the data? |
| 20. | Field notes | Were field notes made during and/or after the interview or focus group? |
| 21. | Duration | What was the duration of the interviews or focus group? |
| 22. | Data saturation | Was data saturation discussed? |
| 23. | Transcripts returned | Were transcripts returned to participants for comment and/or correction? |
| **Domain 3: analysis and findings** | | |

| No | Item | Guide questions/description |
|---|---|---|
| Data analysis | | |
| 24. | Number of data coders | How many data coders coded the data? |
| 25. | Description of the coding tree | Did authors provide a description of the coding tree? |
| 26. | Derivation of themes | Were themes identified in advance or derived from the data? |
| 27. | Software | What software, if applicable, was used to manage the data? |
| 28. | Participant checking | Did participants provide feedback on the findings? |
| Reporting | | |
| 29. | Quotations presented | Were participant quotations presented to illustrate the themes / findings? Was each quotation identified? e*.g. participant number* |
| 30. | Data and findings consistent | Was there consistency between the data presented and the findings? |
| 31. | Clarity of major themes | Were major themes clearly presented in the findings? |
| 32. | Clarity of minor themes | Is there a description of diverse cases or discussion of minor themes? |

Appendix F. Final MRP Approved Proposal

https://osf.io/cv7mg

## Appendix G. REC Approval Letter

Content on pages 83-84 removed due to confidentiality issues

## Appendix H. Health Board Approval Letter

Content on pages 83-84 removed due to confidentiality issues

Appendix I. Recruitment Letter
https://osf.io/qfb2j

Appendix J. Participant Information Sheet

https://osf.io/8d49t

Appendix K. Participant Consent Form

https://osf.io/6wd83

Appendix L. Interview Schedule

https://osf.io/yg2an

## Appendix M. Data Analysis plan

Parents will be interviewed about their experience making decisions about their child's care. This method of analysis has been selected due to its inductive nature and emphasis on idiographic enquiry and how individuals make sense of their experiences (Smith, 2004). This will allow the researchers to explore personal experiences at a greater depth, within a specific, small population.

### *Transcription*

Participants will be given pseudonyms. Verbatim transcriptions of the interviews will be created by the PI. This will include pauses, hesitations and emotional expressions. Names of doctors, medication prescribed, or any precise details of infant's medical treatment will be removed to preserve anonymity. Any exact dates given will also be removed. Any additional content not relating to the interview topics will be redacted from transcripts.

### *Familiarisation and Initial Coding*

Transcripts will be read and re-read to immerse oneself in the data. The PI will then freely examine any semantic content and language (Smith et al, 2021), nothing initial impressions and key points. This will begin to develop an understanding of what matters most to the participant (e.g. values, relationships, events) and the meaning of these (Smith et al, 2021).

### *Developing Emergent Themes*

The PI will then construct experiential statements, by attempting to consolidate the key features of the exploratory notes. These will then be organised according to 'personal experiential themes' and named according to the experience described.

### *Within Case Analysis*

Personal experiential themes will then be examined to explore how themes connect within each participants transcripts. These will be organised into main themes and subthemes. This process will then be repeated for each individual interview.

### *Cross-Case Analysis*

Themes will then be examined across each case, giving attention to points of convergence and divergence across different participants. Themes that appear consistently across the cases will be grouped. Transcripts will continue to be revisited to ensure themes remain grounded in the data. These themes will then be interpreted, represented visually and summarised narratively.

### *Reflexivity*

A reflective journal will be maintained throughout the interview, transcription and analysis process to record any analytical decisions and personal reflections on the research.

Appendix N. Exploratory Notes and Experiential themes (Extract)

| Original transcript | Exploratory notes<br>descriptive linguistic *conceptual* | Experiential statements |
|---|---|---|
| I: And how did you cope with any like worries or uncertainties you had, you know, during the pregnancy?<br>P: So I tried to avoid google because kind of everything that comes up is like low chance of survival. I mean, I did, curiosity got the better of me sometimes, but I didn't spend hours and hours because…I just really didn't want to know about what the bad things would be but then that didn't help when she was born, obviously because it went from bad to worse when she was born. But, Yeah, I think I had an appointment with a clinical psychologist over the phone as well. Just one appointment though. That was just after we got the MRI results. | Avoiding googling negative/info “curiosity got the better of me” suggests internal conflict? *Not wanting to know the ‘odds’- coping through avoidance?* “that didn't help me when she was born” also a realisation that avoidance didn't protect from later distress | Need for information to process diagnosis- Valuing professionals knowledge but also looking for information as parents |
| We couldn't really do anything because it was lock down as well, so couldn't really go see friends. Really. We were kind of just stuck with our own thoughts and then not wanting to google then sometimes googling and seeing negative… like obviously negative things and yeah, so… | No social support “stuck with our own thoughts” – isolation/emotional burden, exacerbated by lockdown | Looking back/Retrospective regret but also recognising why she did this |
| I: And is there anything you would have done differently or anything during pregnancy you would have liked to be different?<br>P: Yeah, I probably would have… Like probably would have researched a bit more so I kind of knew like… I mean the doctors and consultants were brilliant. They could kind of tell us, like your baby's likely to, like, go on to ECMO or like have an extensive stay in hospital. But I didn't really understand what any of that meant. Like, I thought it would just be a surgery and then that would be it. But then there's a whole range of issues that can obviously arise from it. So I'd probably researched a bit more, but I was scared at the time to do it because I didn't really want to know the answers… | Valued the doctors knowledge but mismatch in expectations *unable to comprehend complexity of medical treatment?*<br>Uncertainty and fear | Emotional impact |

## Appendix O. Participant Theme (Example)

| **The power of ‘hope’**<br>Personal experiential themes | |
|---|---|
| Searching for positive stories provided comfort | “…we saw stories of people with CDH who survive and grew up to be humans and we even saw somebody who ran a marathon and things like that. So in a way, when that comes to your mind, you're not really looking for the worst things on the websites and on the forums. You're looking for the best case scenarios. So anyway, when people say the hope dies last, that's true.” |
| Hope played a vital role during critical moments | “And I asked them, do they think if there's still a chance for, for everything to be normal in a few years, no matter how many years, maybe four, maybe 5. Do they think if this baby is gonna be a normal person and it's not be like disabled for the rest of her life or whatever and they basically told me they wouldn't ask her to be put on ECMO machine if they thought there's no chance. Yeah. So obviously we said, OK, we went so far, we're gonna keep going.”<br>“You have to question yourself about are you doing the right decision for the baby, especially now that the baby's out and it's, you know…basically in our minds, as long as they told us there's still a chance, we just decide to keep going.” |
| Helped parents to persevere through challenges | “But we just we sat in and we had conversations and we decided we're gonna keep going and in a way it is just it was not an option for us to give up at this point because it was more like we didn't give up at the beginning after we found out about the CDH. Why would you give up now? She, you know, she's out.” |
| | “Well, the most important thing was the knowledge that there was still a chance that everything would be okay. Eventually, she might heal completely and might not have half a lung, but she might go out to be a healthy person in a way… that that you might not even run marathons, but you'll be sufficient in all that. There was always a chance…It wouldn’t be so like doom and gloom from the beginning to the end.” |

## Appendix P. Data Availability Statement

The data supporting this study’s findings are not publicly available due to ethical and privacy considerations but can be requested from the corresponding author subject to appropriate institutional approvals.