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QUALITATIVE META SYNTHESIS

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Fathers' experiences of living with a child with a progressive life-limiting condition without curative treatment options: A qualitative systematic review

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Abstract

Aim: To systematically review and synthesize findings across qualitative primary studies about fathers' experiences of living with a child with a progressive life-limiting condition without curative treatment options (C3 conditions).

Design: Systematic review and metasynthesis.

Methods: Sandelowski and Barosso's qualitative research methodology guided this review and metasynthesis. A modification of Ricoeur's interpretation theory, described by Lindseth and Norberg, guided the synthesis of qualitative data. The quality of the studies was evaluated using the Joanna Briggs Institute Checklist for Qualitative Research.

Data Sources: A systematic literature search was conducted on 6 May 2022 and updated on 19 July 2023 on MEDLINE, CINAHL Plus with Full Text, APA PsycInfo and Scopus. Inclusion criteria were English-written qualitative studies from the year 2000, from which we could extract data on fathers' experiences of living with a child from 0 to 18 years with a progressive life-limiting condition without curative treatment options.

Results: Seven reports from Western countries contributed to the review. Through structural analysis, we developed the following themes: 'Being shattered in the perception of fatherhood', 'Establishing a new normal' and 'Striving to be acknowledged as a part of the caring team'.

Conclusion: Fathers had to establish a new normal, and they experienced anticipatory mourning, role conflicts and feeling sidelined in healthcare settings when living with a child with a C3 condition. An important issue for further research on paediatric palliative care (PPC) should be to include fathers in the research sample and report separately on fathers' or mothers' experiences instead of parents' experiences.

Impact: The findings will be of interest to healthcare personnel and multidisciplinary teams working within PPC, as they give insight into fathers' experiences and suggest interventions to increase healthcare personnel's involvement with fathers, such as telemedicine.

Reporting Method: Following EQUATOR guidelines, the study was reported according to the enhancing transparency in reporting the synthesis of qualitative research (ENTREQ) framework.

Patient or Public Contribution: No patient or Public Contribution.

KEYWORDS
child, family-centred care, fathers, hospice and palliative nursing, life-limiting conditions, male, paediatric palliative care, palliative care, parents, qualitative research

1 | INTRODUCTION

Globally, over 20 million children are estimated to have a life-limiting condition with a need for paediatric palliative care (PPC), and the number is increasing with the advent of broader eligibility criteria, improved treatment and medical interventions (Benini et al., 2022; Fraser et al., 2021). Caring for these children puts a considerable physical and emotional burden on their parents, leading to the risk of a low quality of life and elevated stress, anxiety and depression (Collins et al., 2020; Lykke et al., 2019).

PPC may extend from months to several years; thus, healthcare professionals (HCP) will care for the child and parents over long periods. Knowledge about parents' experiences is valuable for providing optimal PPC. Existing research in PPC has predominantly focused on mothers or parents as a whole, overlooking the significance of fathers' unique experiences (Macdonald et al., 2010; Nicholas et al., 2020). In recent years, researchers have become increasingly interested in fathers' experiences. However, existing studies on fathers' experiences have mainly focused on children with cancer (Fisher et al., 2021). Research on fathers' experiences of having a child with a progressive life-limiting condition without hope for a cure is limited and requires systematization. To gain new insights and reveal these fathers' experiences, we aimed to systematically review and synthesize existing research on fathers of children with progressive life-limiting conditions without curative treatment options.

2 | BACKGROUND

Children with progressive life-limiting conditions without curative treatment options require palliative care. In general, diseases in children who require palliation are commonly divided into four main categories (Table 1), and the population addressed in this review belongs to the third category (further referred to as C3 conditions).

C3 conditions differ from the other categories of life-limiting conditions because the conditions are progressive with no hope of cure or survival, and the treatment is exclusively palliative (Together for Short Lives, 2018). These children often have a progressive gene-based, metabolic or neurological illness leading to delayed cognitive development, lack of verbal language and physical ailments, such as pain, feeding difficulties, reflux, epileptic seizures, constipation, breathing difficulties and sleep problems (Pawliuk et al., 2020; Siden, 2018). C3 conditions are rare, and there being few comparable cases leads to unpredictability regarding life expectancy and functional outcomes. As a result, parents may experience a lack of information (Siden, 2018), and the parents become experts on their child's care (Price et al., 2022).

PPC is a multidisciplinary and holistic approach to care, including physical, psychological, social and spiritual elements for children with life-threatening and life-limiting conditions and their families (World Health Organization, 2018). A core concept of PPC is to provide the best possible quality of life for each child and their family, including individualized care in which the child and family are at the centre of decision-making (Benini et al., 2022; World Health Organization, 2018). PPC should start at the time of diagnosis or recognition and run alongside other treatments aiming to prolong life, and it is essential to clarify that PPC is not synonymous with terminal care (Benini et al., 2022). Parents of children with life-limiting conditions bear a heavy responsibility for the child's daily care (Brandt et al., 2022), leading to physiological and psychological exhaustion and the risk of reduced physical health, increased pain and sleep disturbance (Pawliuk et al., 2020;

TABLE 1 Categories of life-limiting and life-threatening diseases in children (Together For Short Lives, 2018, p. 11).

Category 1	Life-threatening conditions for which curative treatment may be feasible but can fail. Example: Cancer
Category 2	Conditions where premature death is inevitable, where there may be long periods of intensive treatment aimed at prolonging life and allowing participation in normal activities. Examples: Cystic fibrosis, Duchenne muscular dystrophy
Category 3	Progressive conditions without curative treatment options, where treatment is exclusively palliative and may commonly extend over many years. Examples: Severe metabolic, genetic and neurological conditions
Category 4	Irreversible but non-progressive conditions causing severe disability, leading to susceptibility to health complications and the likelihood of premature death. Examples: Severe cerebral palsy

Siden, 2018). In addition, parents report increased financial costs (Brandt et al., 2022), strain on relationships (Krantz et al., 2022; Postavaru et al., 2020) and various emotions, such as uncertainty, chaos, sadness, loneliness and grief (Bally et al., 2018). It has also been found that parents express grieving throughout the trajectory of their child's disease, especially at the time of diagnosis and when the child progressively loses functions (Fisher et al., 2022; Price et al., 2022). The grief is commonly referred to as 'anticipatory grief reactions.' This term describes persons going through grief reactions prematurely and detaching themselves under the threat of death (Lindemann, 1994). The concept of anticipatory grief can also represent a multidimensional process, which includes the conflicting demands of holding onto, letting go of, and being drawn closer to the dying patient, including grief over losses associated with the progression of life-limiting illness in the past, present and future (Rando, 1988, 2000).

Family-centred care (FCC) is a widely used philosophy of care in paediatric nursing that recognizes the family as important to a child's life and sees the child in the context of a unique family (Mikkelsen & Frederiksen, 2011). The attributes of FCC are a relationship between parents and HCP that is characterized by mutuality and common goals, shared responsibility, parental autonomy, negotiation and family support (Mikkelsen & Frederiksen, 2011). Close collaboration with parents is necessary to provide PPC. However, FCC is criticized for being an abstract concept, and it has its limitations. Parents express frustration when the HCP positions themselves as an expert (Price et al., 2022), and unclear roles between the HCP and the parents can lead the parents into a passive role (Mikkelsen & Frederiksen, 2011).

The father's role in parenting has gradually evolved from seeing fathers as the family patriarch and breadwinner to fathers playing a more active role in their child's life encompassing multiple roles, including being a carer (Cabrera et al., 2000, 2018; Chin et al., 2011). Despite the development in fathers' role, Mikkelsen and Frederiksen (2011) reveal in their analysis of the concept of FCC that FCC studies mainly include mothers as research participants, therefore representing a narrow understanding of the term 'family'. Even though contemporary fathers are more involved in caring for children than in previous decades (Cabrera et al., 2018; Lamb, 2010; Schoppe-Sullivan & Fagan, 2020), fathers of children with developmental disabilities have reported feeling like a peripheral parent and being overlooked by researchers and practitioners (Macdonald & Hastings, 2010). Likewise, fathers are under-represented in parental research (Cabrera et al., 2018) and PPC research (Macdonald et al., 2010; Nicholas et al., 2020), leading to less knowledge of fathers' experiences of and needs in caring for children with life-limiting conditions. The research on fathers' experiences with PPC reveals that fathers feel helpless (Postavaru et al., 2020) and must come to terms with uncertainty being a part of their lives (Fisher et al., 2021). Furthermore, fathers often tend to grieve in isolation rather than in public and take on the role of family protectors (Fisher et al., 2021; Postavaru et al., 2020). Fathers also describe being the forgotten parent in healthcare settings, leading them to feeling like they are on the periphery of their child's care (Fisher et al., 2021; Postavaru et al., 2020).

Previous reviews have explored parents' experiences of having a child with a malignant disease (Tan et al., 2020) or heterogeneous categories of life-limiting conditions (Bally et al., 2018; Postavaru, 2019) but not fathers in particular. No existing reviews of fathers' unique experiences of having a child with a life-limiting condition provide detailed data on C3 conditions (Fisher et al., 2021; Postavaru et al., 2020). Thus, this review could provide new insights and a systematization of fathers' experiences of living with a child with a progressive condition without curative treatment options.

3 | THE REVIEW

3.1 | Aim

This metasynthesis aims to systematically review the current state of knowledge and synthesize findings across qualitative primary studies about fathers' experiences of living with a child with a C3 condition.

3.2 | Design

We designed a qualitative systematic review based on a previous registered protocol in PROSPERO, with registration number: CRD42021265964.

The qualitative systematic review was guided by Sandelowski and Barosso's (2007) four-step metasynthesis methodology consisting of the following: (1) a comprehensive systematic literature search and retrieval of relevant research reports, (2) a quality appraisal of the included studies, (3) a classification of the findings and (4) creation of metasummaries and synthetization of the findings. In the final step, we were guided by a modification of Ricoeur's interpretation theory, as described by Lindseth and Norberg (2004, 2021). This allowed us to explore fathers' lived experiences from a phenomenological–hermeneutical stance and to incorporate our pre-understanding and theory to obtain a comprehensive understanding of these fathers' experiences.

In the reporting of the study, we were guided by enhancing transparency in reporting the synthesis of qualitative research (ENTREQ) framework (Tong et al., 2012).

4 | SEARCH METHODS

4.1 | Search strategy

We developed a search strategy in collaboration with a librarian who is experienced in systematic literature searching. A stepwise search strategy aimed to find both published and unpublished studies (Aromataris & Munn, 2020). In the first step, we used a pearl-growing

strategy to detect search terms from relevant articles and how they were indexed in the databases. We initially searched for 'paediatric palliative care' in titles, abstracts and index words to build a logic grid of keywords. In the second step, we developed a database-specific search based on *p*opulation, phenomena of *i*nterest and *c*ontext (PICo) (Aromataris & Munn, 2020) (Table 2).

Searching the literature, we used index terms and free-text words detected in the first step and translated them into a search strategy targeting each database. Boolean operators combined index terms and free-text words within the same block with 'OR' and between the blocks with 'AND'. The database-specific search and a complete overview of index terms and free-text words are presented in Supplementary File A.

Data sources for the systematic comprehensive searches were Medline, CINAHL Plus with Full Text (Ebsco Host), APA PsycInfo (Ovid) and Scopus. We searched for grey literature and dissertations in the Healthcare Administration Database, Nursing & Allied Health Database and Public Health Database (Proquest). In addition, we searched Google Scholar and screened for the first 300 findings. The searches were conducted on 3 May 2022. In the third step (June 2022), we performed backward citation searches from reference lists and forward citation searches from included articles to retrieve additional studies in Scopus, CINAHL, Medline (Ebsco host), APA PsycInfo, Ovid Nursing Database (Ovid), Isi Web of Science and Google Scholar. The systematic literature searches were updated 19 July 2023 including both backward and forward citation searches (21 July 2023). However, no new reports eligible for inclusion were identified through these updates. The inclusion and exclusion criteria were based on PICo.

4.1.1 | Population

We included reports if they presented fathers' experiences by quotations or researchers' interpretations. We defined a father as a person who holds a caring role and described themselves as a father, for example biological fathers, stepfathers, adoptive fathers, co-fathers and foster carers (Oxford English Dictionary, 2016). Reports including family members other than fathers were included if it was possible to extract data on fathers' experiences.

4.1.2 | Phenomenon of interest

We included reports that qualitatively explored fathers' experiences of living with a child with a C3 condition. Reports that explored fathers' experiences of death and bereavement were included if they presented experiences from when the child was still alive. We included reports about fathers' experiences from (but not limited to) home, respite care, hospital, hospice services and other care facilities. We aimed to synthesize fathers' experiences of living an everyday life in home settings in addition to being hospitalized and therefore excluded studies with newborn who died before hospital discharge.

TABLE 2 PICo—Form.

Population (P)	Phenomena of interest (I)	Context (Co)
Fathers	Experiences	Children with progressive conditions without curative treatment options

4.1.3 | Context

We included reports that examined fathers' experiences living with children aged from 0 to 18 who were diagnosed with a C3 condition, a disease trajectory clearly described as a C3 condition, or if the corresponding authors confirmed the child's C3 condition by e-mail. Reports with a mixed sample of children's diagnoses were included if we could extract data from fathers of children with C3 conditions.

Additionally, we only included English-written primary studies with qualitative designs published from 2000 to 2022. The publication date was chosen to balance the need for a sufficient number of retrievals and to include relatively new studies.

4.2 | Search outcomes

A total of 7556 identified records were imported to Endnote X9. After duplicate removal, 5075 unique records were screened by title and abstract against the eligibility criteria by two reviewers independently using the Rayyan Screening Tool (Ouzzani et al., 2016). Furthermore, 58 reports were read in full text and assessed blindly for eligibility by two reviewers. MS screened all the records, and MSL, NR and LF screened one-third each. Disagreement was resolved by discussion between all four authors. When in doubt if a diagnosis fell under category 3, we consulted an experienced senior consultant or contacted the corresponding authors of the reports. One author replied (Steele, 1999) and confirmed that the children had a C3 condition. Four reports were included after reading the full text. Forward and backward citation searches identified three eligible reports, and seven reports representing seven studies were included in the review.

One report meeting the eligible criteria (Steele, 2002) presented selected findings from a PhD thesis from 1999 (Steele, 1999). After conducting forward and backward citation searches from Steele (2002) and searching for the authors' publications in APA PsycInfo, Medline, CINAHL, Scopus and Research Gate, we identified five eligible reports presenting duplicate findings from the same PhD thesis (Steele, 2000, 2002, 2005a, 2005b; Steele & Davis, 2006). Accordingly, we included the PhD thesis instead of the reports, even though it was published in 1999, because it generated thicker descriptions of fathers' experiences. Of the seven included reports, six were journal articles, and one was a PhD thesis (Steele, 1999). The

PRISMA flow diagram in Figure 1 summarizes the selection process. Reasons for the exclusion of full text with references are available in Supplementary File B.

4.3 | Quality appraisal

Two authors (MS for every report and MSL, NR and LF for one-third each) conducted the critical appraisal independently using the Joanna Briggs Institute Checklist for Qualitative Research (The Joanna Briggs Institute, 2017) (Table 3). Disagreement was resolved by joint discussion with all four authors until consensus was reached. According to Sandelowski and Barosso (2007), studies should not be excluded based on methodological weaknesses. Even if a study has poor methodological quality, its findings are not necessarily without empirical support. Instead, methodological quality should be treated as one of the many characteristics of a study.

4.4 | Data abstraction and synthesis

We extracted and metasummarized the included study characteristics in Table 4. Sandelowski and Barosso (2007) describe the metasummary as 'a quantitatively orientated aggregation of qualitative findings that are themselves topical thematic summaries or surveys of data' (Sandelowski & Barosso, 2007, p. 151). The first author extracted the data and validated the outcome with the other authors.

In the metasynthesis, we analysed the qualitative data in a phenomenological–hermeneutical approach guided by a modification of Paul Ricoeur's interpretation theory, as described by Lindseth and Norberg (2004, 2021). The analysis consisted of 1: Naive reading, 2: Structural analysis and 3: A critical or comprehensive understanding in which the results were reflected on in relation to the research question and relevant literature in the discussion section.

In the naive reading, we read the text as a whole with a phenomenological attitude to formulate a naive understanding of fathers' experiences. After the naive reading, we imported the included reports to NViVO (QSR International Pty Ltd, 2018). In the structural analysis, we extracted meaning units relevant to the research question from the results/findings section of the included reports. Meaning units were text from direct quotations from fathers of children with C3 conditions and the authors' descriptions and interpretations, also referred to as first- and second-order constructs, respectively (Ludvigsen et al., 2016; Sandelowski & Barosso, 2007). Similar meaning units were then condensed and assembled into subthemes and further developed into themes. All four authors contributed to the structural analysis. An example of

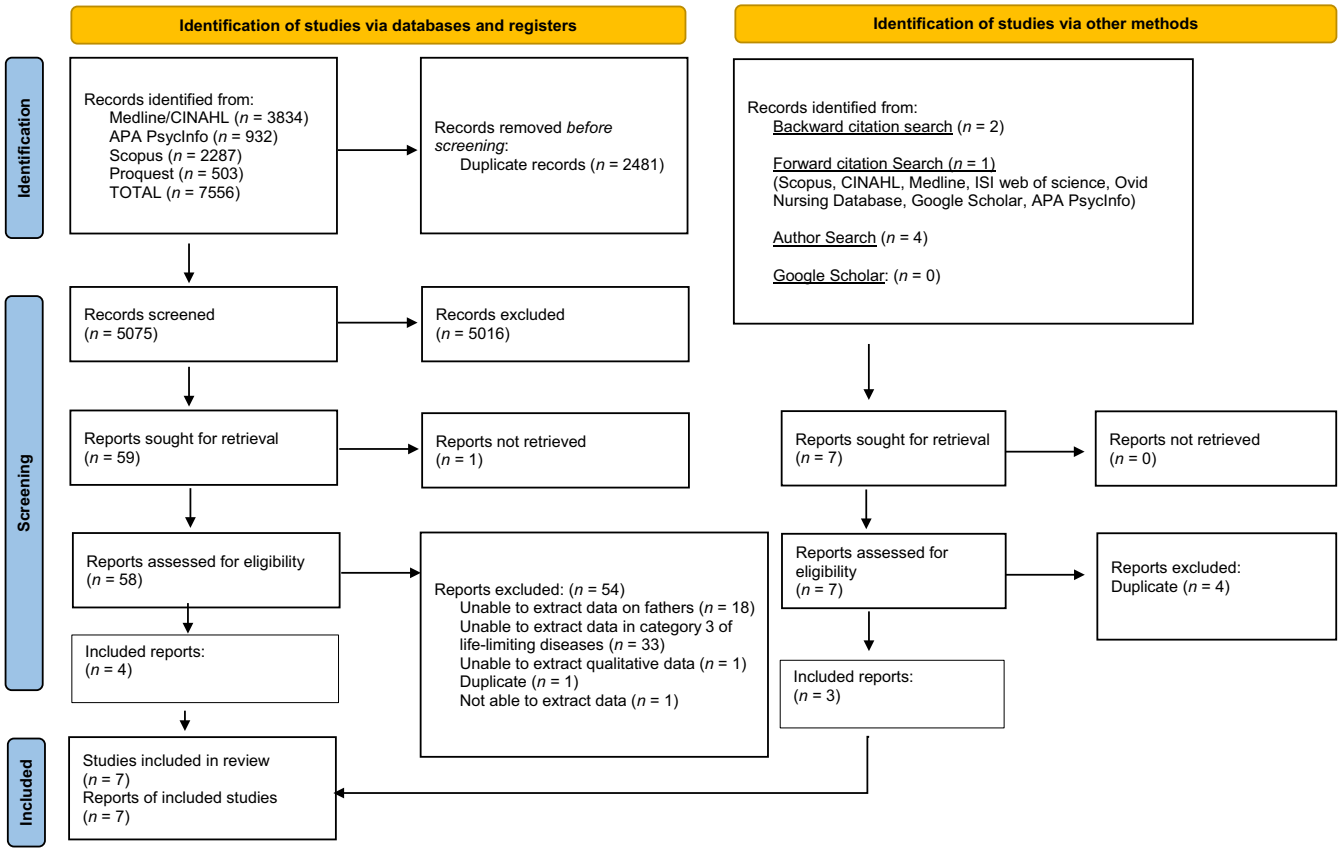

FIGURE 1 PRISMA flow diagram of included studies (Page et al., 2021).

TABLE 3 Critical appraisal of included studies (The Joanna Briggs Institute, 2017).

Author and year	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10
Bose et al. (2019)	Y	Y	Y	U	U	N	Y	Y	Y	Y
Davies et al. (2004)	Y	Y	Y	Y	Y	N	Y	Y	Y	Y
Engler et al. (2020)	U	Y	Y	Y	Y	N	N	Y	Y	N
Rallison and Raffin-Bouchal (2013)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Steele (1999)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Ware and Raval (2007)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Wood and Milo (2001)	Y	Y	Y	Y	Y	N	N	Y	N	Y
Questions										
1. Is there congruity between the stated philosophical perspective and the research methodology?										
2. Is there congruity between the research methodology and the research question or objectives?										
3. Is there congruity between the research methodology and the methods used to collect data?										
4. Is there congruity between the research methodology and the representation and analysis of data?										
5. Is there congruity between the research methodology and the interpretation of results?										
6. Is there a statement locating the researcher culturally or theoretically?										
7. Is the influence of the researcher on the research, and vice versa, addressed?										
8. Are participants, and their voices, adequately represented?										
9. Is the research ethical according to current criteria or, for recent studies, and is there evidence of ethical approval by an appropriate body?										
10. Do the conclusions drawn in the research report flow from the analysis, or interpretation, of the data?										

Abbreviations: JBI-QARI, Joanna Briggs Institute—Qualitative Assessment and Review Instrument; N, no; NA; not applicable; U, unclear; Y, yes.

the process of developing from meaning units to themes is presented in Table 5.

The structural analysis was a 'back-and-forth' process, shifting between the meaning units, subthemes and themes. As described by Lindseth and Norberg (2021), hermeneutical circular movement is repeated several times. Hence, we revised our naive understanding after gaining a deeper understanding of fathers' experiences in the structural analysis until the naive understanding was validated by the structural analysis.

To illustrate the magnitude of the findings across reports, we calculated the effect sizes. The manifest frequency size indicates the number of reports contributing to each theme in the analysis. The intensity effect size calculates the concentration of findings within each report (Table 6) (Onwuegbuzie, 2003; Sandelowski & Barosso, 2007).

5 | RESULTS

5.1 | Metasummaries

The reports included in this review were conducted in Western industrial countries, representing 52 fathers. Family structures were commonly provided in the reports, and most fathers were married or in a relationship with the child's mother. The common data collection method was individual or focus group interviews, and in two studies,

TABLE 4 Characteristics of included studies.

Authors (year)	Country	Objective	*N* fathers	Other participating in the study	Childs diagnoses	*N* children/age (Years)	Methodology/Method	Data analysis
Bose et al. (2019)	USA	To characterize the ZSD caregiver emotional experience in order to develop a comprehensive picture of the specific influences and interactions within a caregiver's daily life	12	25 mothers	32 Zellweger spectrum disorders, 5 D-bifunctional protein deficiency	N = 35/0–18 years, *N* = 3/over 18 years	Qualitative/ semistructured focus group interviews	Content analysis (Elo & Kyngas, 2008)
Davies et al. (2004)	USA	To enhance understanding of fathers' experiences with a child who is seriously ill and dies.	8	0	Cancer: 5 spinal muscular atrophy: 2 Thay-Sachs: 1	*N* = 8/0–14 years	Grounded theory/In-depth unstructured interviews	Constant comparative analysis (Strauss & Corbin, 1998)
Engler et al. (2020)	Germany	Whether hospital care is comparable to paediatric specialized outpatient palliative care (SOPPC) for families, and how parents felt about their children's palliative care situation before entering SOPPC	4	9 mothers	Inborn metabolic disorder, brain malformation, perinatal disorder of the respiratory and cardiovascular system, epilepsy, storage disease, genetic defect, tumour, brainstem injury, undiagnosed.	*N* = 9/0–17 years	Qualitative methodology. Part of mixed–method study/unstructured narrative interviews	Grounded theory approach (Corbin & Strauss, 2008)
Rallison and Raffin-Bouchal (2013)	Canada	To uncover the experience of families living with a child with progressive neurodegenerative Illness	4[a]	8 mothers, 2 siblings, 1 ill child, 2 caregivers considered as family	Progressive neurodegenerative Illness	*N* = 6/NS	Hermeneutic Phenomenology (Gadamer)/Interviews and observations	Interpretative analysis
Steele (1999)	Canada	To enhance understanding of the experiences of families with a child who has a life-threatening neurodegenerative Illness	8[a]	29 (8 families – parents + siblings, grandmother)	Progressive neurodegenerative Illness	*N* = 10/3–13 years	Grounded theory/In-depth interviews and observations	Grounded theory approach (Straus & Corbin, 1990)
Ware and Raval (2007)	United Kingdom	To uncover the experience of families living with a child with progressive neurodegenerative Illness	8	0	Progressive conditions without curative treatment options	*N* = 8/NS	Phenomenology/In-depth semistructured interviews	Interpretative phenomenological analysis (Smith et al., 1999)
Wood and Milo (2001)	USA	To enhance the research on men's bereavement experience and to serve as a companion piece to the former study of maternal responses to the loss of a child with a developmental disability	8	0	Microcephaly, epilepsy and hypomelanosis of Ito, developmental delays and seizures, severe disabilities, cerebral palsy, anoxia at birth	*N* = 8/0–12 years	Qualitative and quantitative methodology/ Individual and focus group semistructured interviews. Grief Experience Inventory (GEI)	Constant comparative analysis (Glaser & Strauss, 1967)

Abbreviation: NS, not say.

[a]Confirmed by e-mail from authors.

TABLE 5 Examples of structural analysis.

Meaning units	Condensed meaning unit	Subtheme	Theme
Rob describes the gradual learning as something 'that just sends waves of fear through you'. While believing he had a healthy baby and enjoying the early period of raising a child, he was also identifying problems at a more gradual pace	Finding out in a gradual pace sent waves of fear.	Getting a diagnosis was a life-changing event	Being shattered in perception of fatherhood
The participants could remember exactly how they received the news, usually in a medical consultation accompanied by their partner. Getting the results of tests/a diagnosis was devastating for all these fathers. Suddenly, their world altered irrevocably. They felt completely overwhelmed. Everything had changed for them and their future life: 'A feeling of devastation, yes, you know that somehow the world changed from what it was an hour and a half before'	Getting a diagnosis was devastating. Their world altered irrevocably		
Yeah, you don't do any sports that could injure yourself. Because, like friends just phoned up, 'Do you want to go skiing with us for the day, come with us?' And I go, 'No'. " I don't ski because I don't want to injure myself. If I injure myself I can't help. So you just don't do things that could injure yourself…	Don't do activities that could injure yourself	Feeling home bound	Establishing a new normal
Thinking about the future when the child had died also caused feelings of guilt: I used to feel guilty whenever I would think about what it would be like afterwards [when the child has died]. I would almost feel giddy because I kept thinking of all this freedom I'd have. Then I'd feel really guilty about it	Feeling guilty of thinking of freedom after the child's death		
Communication between services led to duplication and a lack of clarity and responsibility: 'There is just a deluge of different people, all coming from different angles and it is a minefield really'	Communication between services led to lack of clarity and responsibility	Experiencing lack of coordination of care	Striving to be acknowledged as a part of the caring team
'We have an incredible children's' hospital. We have only a couple of doctors, two, three? Their coordination of care is just awful'	The coordination of care was awful		

TABLE 6 Intrastudy intensity effect sizes and interstudy frequency effect size of themes related to fathers' experiences of living with a child with a C3-condition.

Theme Authors	Being shattered in perceptions of fatherhood	Establishing a new normal	Striving to be acknowledged as a part of the caring team	Intrastudy intensity effect size Individual studies contributing to themes
Bose et al. (2019)	X	X	X	100% (3 out of 3)
Davies et al. (2004)	X	X	X	100% (3 out of 3)
Engler et al. (2020)	X	X	X	100% (3 out of 3)
Rallison and Raffin-Bouchal (2013)	X			33% (1 out of 3)
Steele (1999)	X	X	X	100% (3 out of 3)
Ware and Raval (2007)	X	X	X	100% (3 out of 3)
Wood and Milo (2001)	X	X	X	100% (3 out of 3)
Interstudy frequency effect sizes				
Representation of themes in individual studies	100% (7 out of 7)		86% (6 out of 7)	86% (6 out of 7)

Note. X refers to studies represented in the themes.

it was combined with observations (Rallison & Raffin-Bouchal, 2013; Steele, 1999). One study (Ware & Raval, 2007) focused entirely on fathers and their experience of living with a child with a C3 condition. Furthermore, two studies focused solely on fathers' experiences, but they included fathers with children with various categories of life-limiting conditions (Davies et al., 2004; Wood & Milo, 2001).

Most studies included other family members in addition to fathers, such as mothers, siblings and grandparents. In the included studies, the children with a C3 condition were mainly represented by progressive neurodegenerative and metabolic conditions.

The manifest intensity effect size was 100%—except for one theme, where it was 33%—illustrating that all reports, except for Rallison and Raffin-Bouchal (2013), were represented in each theme. Moreover, the manifest frequency effect size ranged from 86% to 100%, meaning that most reports contributed to the themes in the analysis, and no reports were over- or under-represented in the themes (Sandelowski & Barosso, 2007).

5.2 | Metasynthesis

We synthesized the qualitative findings of fathers' experiences of living with a child with a C3 condition across seven reports. The naive reading revealed that getting a diagnosis was a shock, leading to an unpredictable life in which fathers had to acknowledge their new way of living. The fathers felt that they had to be strong, and in their encounters with HCP, they felt that they were treated differently from the mothers.

Through structural analysis, we developed the following themes describing fathers' experiences of having a child with a C3 condition: 'Being shattered in the perception of fatherhood', 'Establishing a new normal' and 'Striving to be acknowledged as a part of the caring team'.

5.2.1 | Being shattered in the perception of fatherhood

Learning that their child had a progressive life-limiting condition was a life-changing event for fathers causing a double loss. Not only did they know that their child would die, but having a child with a C3 condition also shattered fathers' perception of fatherhood and their future with a healthy child.

> [...] all those new things that are new and fresh and exciting to a kid, things they share with their fathers... that I will never share with my son. That was harder than burying him in some respect... But that loss occurred almost the first day after we got the finding... I lost that particular future with my son at that point [...] (Wood & Milo, 2001, p. 644).

Several fathers had sensed that something was wrong with their child at the time of diagnosis; thus, the time of diagnosis was not marked as a dramatic moment (Ware & Raval, 2007; Wood & Milo, 2001). Other fathers experienced the diagnosis as a shock because they had not noticed a problem with their child until someone else had pointed it out to them (Ware & Raval, 2007). Despite this, the time of the diagnosis was a life-changing moment when they knew that their child was going to die, and the hope of survival was irrelevant.

Even though fathers sensed something was wrong with their children, they were not prepared for what was to follow (Ware & Raval, 2007). The fathers' lives had been tainted permanently, and they sought answers to questions such as 'why did this happen to us' (Ware & Raval, 2007; Wood & Milo, 2001). Fathers with solid religious beliefs expressed how their faith was experienced as solace and support throughout the child's illness trajectory (Ware & Raval, 2007; Wood & Milo, 2001).

Learning about their child's illness could evoke an immense range of emotions, such as fear and devastation (Bose et al., 2019; Davies et al., 2004; Engler et al., 2020; Rallison & Raffin-Bouchal, 2013; Steele, 1999; Ware & Raval, 2007; Wood & Milo, 2001). In addition, fathers experienced grief, sadness and distress. For some, the sadness never lessened, and one father even welcomed the idea of his own death. Feeling like everything was out of their control could add to their feeling of being powerless and overwhelmed (Bose et al., 2019; Davies et al., 2004; Steele, 1999; Ware & Raval, 2007; Wood & Milo, 2001). As one father expressed:

> A part of us is dying with our son. A part of our emotions is going with him. We don't know what is going to happen. We don't even know what state it is going to leave us in at the end. It will be almost like a sponge that has been wrung out. (Steele, 1999, p. 111).

One father left his family when the child was expected to die because it was too difficult to cope with. Instead of emotionally distancing themselves, other fathers experienced a unique and heightened love for the child. Having a child with a C3 condition could help fathers with personal growth and change what they value in life. Even though fathers felt that their child was given a death sentence, they hoped to prolong their child's life and alleviate their suffering. The focus shifted from survival to quality of life, allowing the child to live the course of their life and focusing on their child's abilities (Davies et al., 2004; Steele, 1999; Ware & Raval, 2007).

5.2.2 | Establishing a new normal

Establishing some sort of normal was an ongoing process that changed throughout the child's lifetime (Bose et al., 2019; Ware & Raval, 2007). Their new everyday lives altered all aspects of life, such as work and finances, relationships with friends and family, and their ability to take care of themselves. Fathers used different strategies to establish a new everyday life. Some fathers coped by taking one day at a time and facing difficulties as they appeared, preferring not to look into the future. Others planned for the future by guessing the trajectory of their child's disease to make the transition into the next phase as smooth as possible (Steele, 1999; Ware & Raval, 2007).

Because of their uncertain and unpredictable lifeworlds, fathers could feel isolated, alienated and abandoned. The feeling of

alienation was attributed to the rareness of the child's illness, inadequate follow-up after diagnosis, poor communication, being an at-home parent or the fact that others could not see a problem because their child outwardly appeared to be normal (Steele, 1999; Ware & Raval, 2007; Wood & Milo, 2001). Because of their stressful situation, fathers described how they could no longer keep up with their previous pace at work and in their personal lives. They described feelings of going into slow motion (Steele, 1999). One father said:

> If it takes a normal person one day to do something, it will take us two or three days (Steele, 1999, p. 130).

While some fathers gave up work because they could not concentrate, other fathers explained how supportive and flexible work was crucial to continuing to work. Examples could be flexible hours, taking a nap during the day or going home early if necessary (Davies et al., 2004; Steele, 1999). Having a child with a C3 condition challenged the family economy, their income was reduced, and many costs were not covered. Consequently, some families struggled financially. In addition, families became homebound and unable to move because of the risk of losing benefits. As the primary breadwinner, fathers often handled the finances, while the mother cared for the child (Steele, 1999; Ware & Raval, 2007). 'We had this automatic division of labour and responsibility. [Wife] was going to see to his physical needs and his care and I was just going to make sure I brought home the bacon' (Ware & Raval, 2007, p. 556). Being at work could reduce the father's contact with the child, and some quit their jobs to spend more time with their families, even though this could make their financial situation more challenging (Steele, 1999).

To take care of themselves, fathers found it important to exercise, take up hobbies, attend courses about chronically ill children or go to counselling (Bose et al., 2019; Steele, 1999; Ware & Raval, 2007). Even though fathers emphasized the importance of taking care of themselves and their emotional well-being, they found it difficult to take the time to do so (Engler et al., 2020; Ware & Raval, 2007). Fathers described giving up previous pursuits, such as hobbies or sports. If, for example, they were injured in sports, they could not care for the child and would leave their spouse exhausted. The feeling of being homebound and unable to do activities in the spur of the moment could lead to guilt when thinking about the freedom they would have after the child's death (Steele, 1999):

> I used to feel guilty whenever I would think about what it would be like afterwards [when the child has died]. I would almost feel giddy because I kept thinking of all this freedom I'd have. Then I'd feel real guilty about it. (Steele, 1999, p. 112).

The child's illness affects fathers' relationships with partners, friends and the conflicting needs of their other children (Engler et al., 2020; Steele, 1999; Ware & Raval, 2007). However, their relationships could also be more robust due to their child's illness, and fathers found it essential to have a close relationship with their partner. Still, in some cases, the child's condition required that one parent constantly be with the child, making it impossible for parents to spend time alone, focusing on their marriage (Steele, 1999; Ware & Raval, 2007).

Fathers emphasized the importance of gaining support from friends and family, and they appreciated people talking directly to them rather than making assumptions about their situation (Steele, 1999; Ware & Raval, 2007). Still, they had to draw lines with other people because they were stressed and had too little energy to deal with other people's problems. Instead, fathers found support and communicated better with families in the same situation and valued talking to somebody outside of the family—for instance, support groups or a parent in a similar situation (Steele, 1999; Ware & Raval, 2007):

> Something like this, most people do not experience in their lifetime. You might see it from a distance, but unless you actually go through it, you never know what it is really like (Steele, 1999, p. 115).

In addition, helping others in the same situation was a motivation for participating in research, so other fathers did not have to suffer in the same way as they did (Steele, 1999; Ware & Raval, 2007).

In the process of establishing a new normal, fathers described differences in how the mothers and fathers responded emotionally. In addition, they experienced that society discouraged men from acknowledging their emotions and that women were more actively encouraged to share their thoughts. It was particularly difficult to show emotions together with other men. Fathers experienced grief more privately, and their grief was not always obvious to others. (Steele, 1999; Ware & Raval, 2007):

> How I coped with it was a typical male, the female broke down in tears and someone had to be strong to support the other, but inside I was screwed up about it but someone had to hold it together for everyone else (Ware & Raval, 2007, p. 557).

5.2.3 | Striving to be acknowledged as a part of the caring team

Fathers had varying experiences of care and collaboration with HCP. While several fathers described having an excellent team of professionals taking care of their child, others described a lack of coordination, poor communication and lack of trust in HCP. One father described the hospital as '*a permanent state of emergency, all the time*' (Engler et al., 2020, p. 7). Fathers were told that they were part of a team but had the opposite experience:

> They suck you in at the beginning that you're going to be an important part of this team. And you're fuck all. You're nothing. They don't give a shit what you think or, what you want. Anybody that's an important part of the team, I can't see an important person sleeping on the floor. But that's where you sleep as a parent. You sleep on the floor. Does the doctor sleep on the floor? Does the patient? No. That kind of thing just pissed me off (Steele, 1999, p. 174).

To improve health services, fathers suggested holistic care and a single worker to whom they could talk in order to enhance continuity of care (Bose et al., 2019; Steele, 1999; Ware & Raval, 2007).

Positive experiences were related to continuous care and follow-ups by the same person over the years, good communication and healthcare professionals caring for the child in a personal way (Bose et al., 2019; Davies et al., 2004; Wood & Milo, 2001):

> We, around that time, got a home care nurse. [S]he was just a godsend, really. She was a fabulous, fabulous nurse. She really kind of adopted Rachel as her second child (Davies et al., 2004, p. 127).

Fathers felt that they were treated differently from mothers by HCP. In general, fathers experienced that the child's mother had been given more opportunities to discuss the diagnosis and its implications than they had been given, and they described sparse information provision and insufficient follow-up after the diagnosis (Bose et al., 2019; Ware & Raval, 2007). Because the fathers felt it was more difficult to acknowledge that they were struggling emotionally, they experienced their emotional reactions being more frequently ignored or overlooked by HCP. They suggested that the focus should shift from mothers to both parents and that the system could find more effective ways of engaging fathers, such as including fathers in the consultation or treatment process and adjusting the appointments to when fathers could take time off work (Ware & Raval, 2007).

Fathers became advocates for their children and discovered information and resources that they were entitled to through organizations related to the child's diagnosis. The fathers wanted action before talking and described cases where they had to explode in rage to get people to listen. Seeking information gave fathers a feeling of taking charge, and they tried to educate themselves with as much information as possible. Having a child with a C3 condition could make them fall between two stools, and they had to arrange meetings with appropriate personnel to receive, for example, respite care (Davies et al., 2004; Steele, 1999; Ware & Raval, 2007; Wood & Milo, 2001). One father expressed his frustration with the difference in treatment related to diagnosis:

> And if you get cancer or have cardiac issues, it's like the jackpot. You know, they're cutting edge. And it's hard for me not to be so angry about that (Bose et al., 2019, p. 4).

6 | DISCUSSION

In this systematic review of seven qualitative reports from the Western world, we analysed fathers' experiences of living with a child with a C3 condition. The findings developed in the metasynthesis implied that living with a child with a C3 condition has a profound and life-altering impact on fathers. It shattered their perception of fatherhood, as their anticipation of being a father was not met. Moreover, they had to cope with the grief of not being able to do what other fathers do with their children and the fact that their child would not live into adulthood. Receiving a diagnosis evoked enormous emotional responses, and fathers entered a world of uncertainty and needed to establish a new normal. The fathers described grieving in private and how their child's illness affected their work–life balance. Furthermore, fathers strived to be acknowledged as a part of the caring team and experienced being treated differently from the mothers by the professionals.

Being shattered in their perception of fatherhood revealed aspects of anticipatory mourning when fathers grieved over losses associated with the progression of their child's life-limiting condition (Rando, 1988, 2000). The anticipatory mourning reactions seem to be consistent with findings in previous studies exploring parents' experiences within PPC (Fisher et al., 2021; Hurley et al., 2021; Krantz et al., 2022; Postavaru et al., 2020; Price et al., 2022). In our review, we found that most fathers felt a heightened feeling of love towards their child, which brings to light Rando's (1988) anticipatory mourning theory of being drawn closer to the dying patient. However, our findings also describe anticipatory grief reactions consistent with Lindemann's (1994) anticipatory grief theory of detaching oneself under the threat of death when one father described that he left his family. The various anticipatory grief reactions reveal that fathers not only grieve their dying child but also handle recurrent losses throughout their child's trajectory. We presume that the findings of being shattered in their perception of fatherhood reveal a mismatch in how fathers expected their role as fathers to be. In their metasynthesis of transitioning into fatherhood, Chin et al. (2011) found that fathers with healthy children saw their role towards the child as more physical and playful than that of their partners. In addition to anticipatory grief over their child's progression of condition, loss of functions and future death (Rando, 2000), we suggest it may be that fathers' anticipatory mourning reactions are concurrent with grief and loss over their expected role as fathers towards the child when they were no longer able to do the same thing with their child as fathers with healthy children could.

Being the father of a child with a C3 condition affected every aspect of the father's everyday life. The sick child had to come first, and fathers had to establish a new normal. In line with our findings, several studies, regardless of diagnosis or gender, describe parents as establishing a new normal and refer to it as normality reconstruction (Von Der Lippe et al., 2022), finding normal (Bally et al., 2018) and striving for normality (Price et al., 2022). Having a child with a C3 condition is time-consuming (Lazzarin et al., 2018),

and fathers in our review felt isolated, homebound, and as though their personal relationships had changed. The father's ability to prioritize their marriage, spend time with friends or be spontaneous was lost. Previous studies have established that marriage can be negatively affected when parents have time-consuming care commitments (Lazzarin et al., 2018; Sevin et al., 2022). However, our findings, aligned with Von Der Lippe et al. (2022), revealed that some fathers gained a more robust relationship with their partners. A possible explanation is the strengthened feeling of togetherness, knowing that someone is facing the same stress (Von Der Lippe et al., 2022). In addition to changes in fathers' personal relationships, we found that their perceived social role as breadwinners was challenged. Even though today's society is more equal and fathers play an active role in their children's lives (Cabrera et al., 2018; Cabrera et al., 2000; Lamb, 2010), research confirms our findings that fathers still see themselves as providers (Chin et al., 2011). Gender differences in income, where men often earn more than women (Schoppe-Sullivan & Fagan, 2020), might also contribute to the explanation of traditional gender roles where fathers are viewed as breadwinners.

When fathers could not take time off work and were unable to participate in hospital appointments, they strived to be acknowledged as part of the caring team. International PPC standards recommend that parents should be assisted in maintaining their social roles, such as in work (Benini et al., 2022). However, the conflicting demands between work life and providing care for their child can result in gatekeeping and regulating fathers' possibilities of participating in their child's care (Mikkelsen & Frederiksen, 2011). Our findings might indicate that fathers of children with C3 conditions might not receive the social support recommended by international standards (Benini et al., 2022) when they are struggling to combine their role as carers and breadwinners. Fathers in our review also experienced how women were more encouraged to share their thoughts and feelings. FCC emphasizes the importance of family in a child's life (Mikkelsen & Frederiksen, 2011), and international standards of PPC emphasize providing family members with the opportunity to talk about personal feelings and receive professional support (Benini et al., 2022). Even though fathers' emotional reactions have been well documented in previous research on PPC (Fisher et al., 2021; Postavaru et al., 2020), our findings reveal that fathers might be forgotten parents in healthcare settings. When fathers strive to be acknowledged as part of the caring team, their opportunity to be an important source of information and to be a mutual partner in caring for their child is not being looked after. The feeling of not being acknowledged does not seem to be a unique experience for fathers of children with C3 conditions but is a shared experience by fathers in general (Chin et al., 2011; Høgmo et al., 2021; Leahy-Warren et al., 2022). This might indicate a structural problem in fathers' role within paediatric health care. It is also well established in PPC research that fathers feel like they are treated differently by HCP and like secondary parents compared to mothers (Fisher et al., 2021; Postavaru et al., 2020).

Fathers in our study were empowered by information. However, they could not receive first-hand information from HCP if they were at work or felt they were overlooked by HCP. In contrast to the principles of mutuality, shared responsibility and support of family in FCC (Mikkelsen & Frederiksen, 2011), being overlooked or unable to be present leads fathers to receive second-hand information and mothers to receive information alone and unsupported (Hurley et al., 2021). To compensate for the lack of information and support, the fathers in our review sought information from other sources, such as organizations related to their child's diagnosis. Life-limiting conditions are often rare, and parents become experts on their child's needs (Price et al., 2022). In addition to support from organizations, as described in our study, the Internet and social media could be valuable for peer support and sources of information, with the advantage of easy access, quick responses and not having to take the time to show up in person (Baumbusch et al., 2019; Nicholas et al., 2016). In their conceptualization of the term FCC, Mikkelsen and Fredriksen (2011) found most of the included studies to be based on the perspective on mothers. In light of our findings, we consider that there might be a shortcoming in the care model of FCC for fathers and suggest further interventions and studies to include fathers in care models such as FCC. After the pandemic, the use of digital technology and telemedicine in health care has increased (Omboni et al., 2022). According to international PPC standards (Benini et al., 2022), digital resources—for example, telemedicine—should be integrated into current care models. As a recommendation to practice, we suggest telemedicine as one possible intervention to facilitate fathers' inclusion in healthcare teams if they are unable to be physically present.

6.1 | Limitations and methodological considerations

Despite the thorough literature search, a limitation is that there might be eligible studies on C3 conditions that we did not identify through the systematic literature search, due to a lack of standard terminology in PPC. Several studies written in languages other than English were excluded when screening titles and abstracts; thus, we might have missed studies presented in other languages. Another methodological consideration is the extraction of data on fathers' experiences of having a child with a C3 condition. We excluded several reports because fathers' experiences were not specified in the results and were instead referred to as 'parents' experiences'. Of the reports that explicitly presented fathers' experiences, we were challenged in separating fathers' experiences of having a child with a C3 condition from other life-limiting conditions. Hence, we did not extract data unless we could identify specific data from fathers of children with C3 conditions. It is important to notice that most of the fathers in our review for most represents married or cohabitant fathers' experiences. As a result, experiences of single dads remain unexplored.

Although no reports were excluded due to methodological quality, the critical appraisal of the included reports must be taken

into consideration when reading the review. The overall quality of the included reports was considered to be good, but statements locating the researchers' background and the influence of the researcher on the research and vice versa were especially lacking in several of the included reports. Therefore, we do not have information on how the authors of the included reports might have affected the interpretation of the qualitative data. Because we interpret out of our pre-understanding (Lindseth & Norberg, 2004, 2021), we emphasized presenting our theoretical and practical background as nurses with clinical experience working within paediatrics.

7 | CONCLUSION

This study synthesizes existing knowledge and offers insight into the experiences of fathers of children with C3 conditions. The results indicate that fathers not only cope with their child's inevitable death, but also grieve over the loss of their previously healthy child. Fathers had to establish a new normal, which in turn affected their work and private relationships. Furthermore, our findings indicate that fathers go through anticipatory mourning, encounter role conflicts and often feel sidelined in healthcare settings. The results will be of interest to HCP and multidisciplinary teams working within PPC, and there is a need for interventions to increase healthcare personnel's involvement with fathers. An important issue for further research in the field of PPC and C3 conditions should be to include fathers in the research sample and report separately on fathers' or mothers' experiences. Considering shifts in family dynamics, it is also relevant to further explore single dads, fathers identifying as LGBTIQ+ and rainbow families' within the context of PPC. In addition, research on ethnic minorities or fathers' experiences from non-Western countries has received little attention and should be further explored.

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CONFLICT OF INTEREST STATEMENT

The authors have no conflicts of interest to declare.

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DATA AVAILABILITY STATEMENT

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SUPPORTING INFORMATION

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

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