Chronic Paradoxes

A Systematic Review of Qualitative Family Perspectives on Living with Congenital Heart Defects

Svensson, Marie Kofod; Wahlberg, Ayo; Gislasson, Gunnar Hilmar

Published in:
Qualitative Health Research

DOI:
10.1177/1049732319869909

Publication date:
2020

Document version
Peer reviewed version

Citation for published version (APA):
Svensson, M. K., Wahlberg, A., & Gislasson, G. H. (2020). Chronic Paradoxes: A Systematic Review of Qualitative Family Perspectives on Living with Congenital Heart Defects. *Qualitative Health Research*, *30*(1), 119-132. https://doi.org/10.1177/1049732319869909

Chronic Paradoxes: A Systematic Review of Qualitative Family Perspectives on Living with Congenital Heart Defects

Marie Kofod Svensson, Ayo Wahlberg, and Gunnar H. Gislason

Published in:
Qualitative Health Research

DOI:
10.1177/1049732319869909

Publication date:
2020

Document version:
Peer reviewed version (Post-print)

Citation for published version (APA):
Svensson, M. K., Wahlberg, A., & Gislason, G. H. (2020). Chronic Paradoxes: A Systematic Review of Qualitative Family Perspectives on Living with Congenital Heart Defects. *Qualitative Health Research*, *30*(1), 119–132. https://doi.org/10.1177/1049732319869909

Chronic Paradoxes: A Systematic Review of Qualitative Family Perspectives on Living with Congenital Heart Defects

Marie Kofod Svensson[1,2], Ayo Wahlberg[2], and Gunnar H. Gislason[1,3]

[1] The Danish Heart Foundation, Copenhagen, Denmark
[2] Department of Anthropology, University of Copenhagen, Copenhagen, Denmark
[3] Department of Cardiology, Copenhagen University Hospital Herlev and Gentofte, Denmark

Corresponding Author
Marie Kofod Svensson, The Danish Heart Foundation, Vognmagergade 7, 3rd floor, 1120 Copenhagen K, Denmark. Email: marieks@hjerteforeningen.dk
ORCID iD: Marie Kofod Svensson https://orcid.org/0000-0003-4271-9569

Acknowledgments
We would like to thank Signe Lindgård Andersen for her comments on earlier drafts of this article, Vibeke Steffen for help in designing the initial ideas for the review, and Matthew Phelps for proofreading. Ayo Wahlberg acknowledges the European Research Council Starting Grant "The Vitality of Disease—Quality of Life in the Making" (Grant No. 639275).

Funding
This work is part of a PhD study funded by the Innovation Fund, Denmark and the Children's Heart Foundation, Denmark.

Author Biographies
Marie Kofod Svensson is industrial PhD fellow at the Danish Heart Foundation and the Department of Anthropology, University of Copenhagen conducting ethnographic research on family perspectives on living with CHDs in Denmark. Her research has focused on child and youth health, disability, chronic illness, visuality and visual representation.

Ayo Wahlberg is professor MSO at the Department of Anthropology, University of Copenhagen. Working broadly within the field of social studies of (bio) medicine, his research has focused on the social study of traditional herbal medicine, reproductive technologies, and health metrics. He is associate editor of BioSocieties and author of Good Quality - the Routinization of Sperm Banking in China (University of California Press, 2018).

Gunnar H. Gislason is professor of cardiology at the University of Copenhagen, research director at the Danish Heart Foundation, and at Copenhagen University Hospital Herlev and Gentofte. His research has focused on large-scale register-based studies related to epidemiology and treatment of cardiovascular disease, implementation of evidence-based treatments and cardiovascular complications of cardiovascular and non-cardiovascular treatments.

Abstract

There have been substantial advances in the diagnostics and treatment of congenital heart defects (CHDs) in recent decades, and this has improved survival significantly. Consequently, there is a growing interest in how CHDs affect the daily lives of children and youth. We examine life with CHDs as a particular kind of living from the perspectives of both children and youth with CHDs and their families through a systematic review of existing qualitative research. Based on a meta-ethnographic analysis of 20 articles (identified through PubMed, EMBASE, EBSCOhost, PSYCHinfo, Scopus, and Web of Science from January 7 to 12, 2016), we argue that living with CHDs is characterized by chronic paradoxes arising out of the transitions, normalities, and futures that families have to navigate.

Keywords

Congenital heart defects; chronic illness; chronic paradoxes; families; children's health; meta-ethnography; qualitative research; systematic review

Congenital heart defects (CHDs)[1] – structural defects in the heart present at birth (National Heart, Lung, and Blood Institute, 2017) – are generally described as the most common major birth defect (van der Linde et al., 2011). Diagnostics, surgery, and medical treatments of CHDs have seen considerable advances through the last 3 decades (Dodge-Khatami, 2016; Larsen, Olsen, Emmertsen, & Hjortdal, 2017; van der Bom et al., 2011), affecting survival rates so significantly that CHDs are now often perceived as chronic conditions (e.g., Connor, Kline, Mott, Harris, & Jenkins, 2010; Kosta et al., 2015; Moola, Fusco, & Kirsh, 2011). Such advances notwithstanding, an increasing focus on chronicity (given its often inherent definition against acute disease) can be problematic (Manderson & Smith-Morris, 2010). Looking at CHDs as chronic conditions can elide uneven access to and varying quality of treatments around the world (e.g., Musa et al., 2017), blur experiences of suffering that persist despite or perhaps because of improvements in survival rates, and impede simultaneous examination of acute and stable periods. Consequently, in this review, we do not simply approach CHDs as conditions that were previously acute and now chronic. Instead, we emphasize CHDs as one type of condition among many, where biomedical advances have affected survival so profoundly that a new research field has emerged focused on everyday living with and not only surviving CHDs (Dodge-Khatami, 2016; Moola, Faulkner, Kirsh, & Kilburn, 2008).

Two previous systematic reviews included qualitative perspectives, although both were based mainly on quantitative psychosocial ratings, as they examined parental coping strategies and the psychological impact of CHDs. From them, we learned that many parents of children with CHDs are anxious, stressed, and depressed (Wei, Roscigno, Hanson, & Swanson, 2015), and that fewer psychosocial resources and support impact negatively while family cohesiveness and adaptive parenting strategies enable successful adjustment (Jackson, Frydenberg, Liang, Higgins, & Murphy, 2015). The authors of a newer review focused on children and young adults' qualitative illness experiences with CHDs (ages 6–21 extracted from studies focused on ages 6–67) and found them to be facing prognostic uncertainty, traumatic medical treatments, limitations of physical stamina, and unsure future goals, all of which are better coped with by those who have good social support, continuous and empathetic healthcare, and personal determination (Chong et al., 2018). We add to these reviews by exclusively exploring qualitative perspectives, combining child/youth and adult perspectives for a wider family focus, and going beyond the aggregation of themes to provide a new interpretive framework through the approach of meta-ethnography. Analytically, we focus on what can be thought of as the "certain kinds of living" that CHDs bring in their wake, "characterised by therapy, regular medication, visits to the hospital, and/ or reliance on medical devices which in turn can impede upon or shape daily living" (Wahlberg, 2018, p. 734). This entails aspects of coping and quality of life, but more in the sense of "how" than "how well." Thus, the aim of this article is to

synthesize existing qualitative research on CHDs to identify the kinds of lives that families lead, characterized by particular rhythms, disruptions, and limitations, as well as opportunities and possibilities for improvements. We do so by focusing on how children and youth with CHDs and their families relate to and negotiate everyday life with CHDs, and on how they orient them- selves toward the future.

Method

Review Approach

Despite a recent surge in systematic qualitative reviews and a proliferation of approaches, there is no consensus on one particular approach. The method has been heavily debated, particularly what is seen as the more rigid, linear, and quantitatively inspired aspects of the genre (Hawker, Payne, Kerr, Hardey, & Powell, 2002; Popay & Mallinson, 2010; Thorne, 2017). Consequently, many advocate for, and we follow, an approach that is interpretive, iterative, and generative of new theories or metaphors rather than aggregating, summarizing, and developing thematic analysis (Popay & Mallinson, 2010; Thorne, 2017). One of the most common approaches, and the one that has inspired our approach, is that of metaethnography (France et al., 2014; Popay & Mallinson, 2010), which focuses on interpretation, translation, and choice of form depending on how the included studies relate to each other (Noblit in Thorne, Jensen, Kearney, Noblit, & Sandelowski, 2004).

The authors of the included articles examine different aspects of living with CHDs—from children and youths' attitudes to physical activity to grandparents' involvement with siblings. Consequently, we have chosen to undertake "a-lines-of-argument synthesis" (Noblit & Hare, 1988, p. 62). We aim to say something about the whole – living with CHDs from a family perspective – based on different parts, that is, articles focused on different themes, informant groups, time periods, and settings. Together, they provide insights into the everyday challenges, frustrations, and possibilities that families living with CHDs face.

Search Method

We did Boolean searches in PubMed, EMBASE, EBSCOhost, PSYCHinfo, Scopus, and Web of Science January 7 to 12, 2016 using the following combinations: Congenital AND (heart disease OR heart defect OR cardiac disease OR cardiac defect OR heart malformation) AND (family OR child OR youth OR infant OR adolescent OR sibling OR parent OR grandparent OR mother OR father) AND (qualitative study OR qualitative method OR interview OR participant observation) NOT (quantitative study OR quantitative method OR questionnaire). To counter the barriers to qualitative

research in biblio- graphic databases (Dixon-Woods et al., 2006), we also manually searched a range of journals and the references of included articles.

Selection Process

Articles were included if they examined aspects of living with CHDs from the perspective of parents, grandparents, siblings to children or youth with CHDs between 0 and 18 years old, or from children and youth with CHDs in the same age group. Studies had to be empirically based, peer-reviewed articles from "Western countries" (Danmarks Statistik, n.d.), in English, Norwegian, Swedish, or Danish. We focused on the last 30 years due to advances in diagnostics and treatment in this period (Bjorbækmo & Engelsrud, 2008; Bouma & Mulder, 2017; Dodge-Khatami, 2016). We included articles with the same empirical data if analytical focus varied, and excluded ones that evaluated interventions, or included health professionals, or other diagnoses. See Figure 1 for an overview of the selection process.

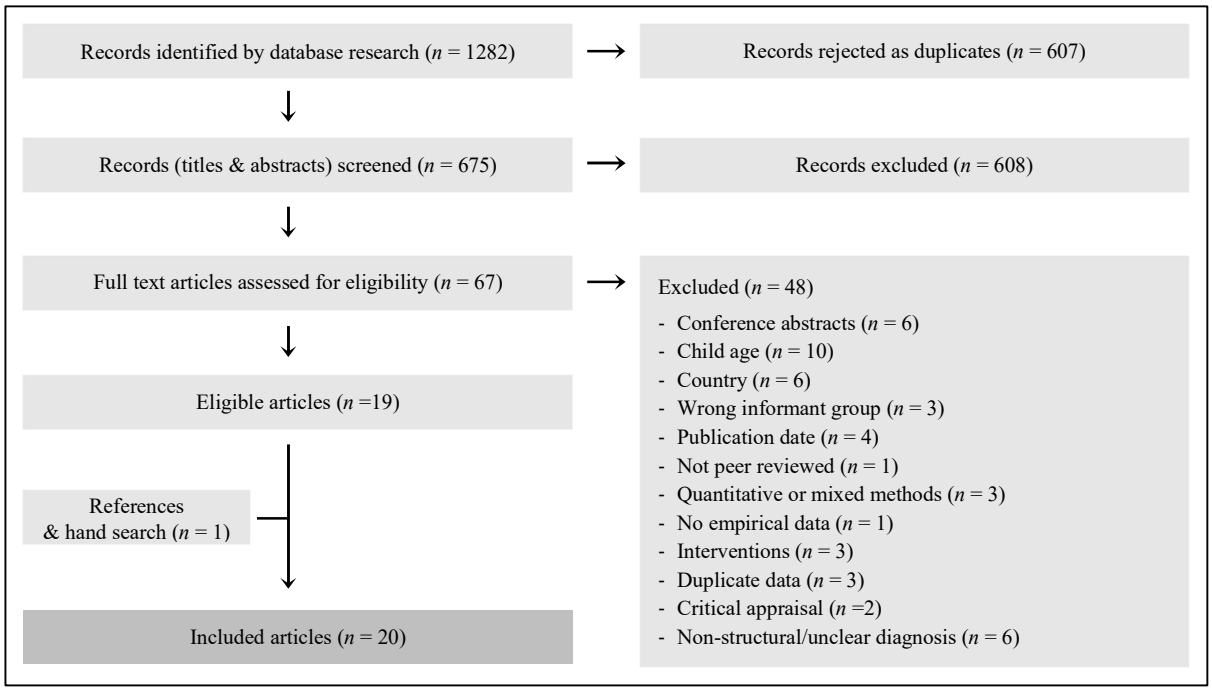

Figure 1. Flow diagram of selection process.

Classification and terminology is a general problem when it comes to CHDs (Edwards, 2001; Garne et al., 2012). We found both structural and nonstructural conditions (e.g., arrhythmia, cardiomyopathy, and genetic disorders) interchangeably labeled with terms such as congenital heart disease and congenital heart defects as well as a lack of explicit descriptions of included diagnoses.2

When diagnoses were missing, we included or excluded articles based on descriptions of illness trajectories (particularly surgeries as treatment), references, and other descriptions of the children's conditions. The process of excluding non- structural defects was collective. However, Gislason, a professor of cardiology, made the final decision.

Critical Appraisals Skills Programme (2014) was used for critical appraisal in combination with the appraisal tool developed by Hawker et al. (2002). Following Carroll, Booth, and Lloyd-Jones (2012), we excluded articles based on critical appraisal, as they showed that inadequately reported studies tend to lack details and contribute little to the analysis. We excluded articles if they scored more than two poor ratings in Categories 1 to 7 (abstract/title, introduction/aims, method/data, sampling, data analysis, ethics/bias, findings/results), or if they had any poor rating in Category 7 (findings/results). There was a general lack of information on sampling, ethics, and bias, so we did not exclude based on these ratings alone. Twenty articles were included (see Table 1), and they mainly focus on parent perspectives (15 articles), whereas children and youth with CHDs (seven articles) and grandparents (two articles) are given less attention, and no articles include siblings. Female informants, particularly mothers, are overrepresented. Sixteen articles look at multiple types of CHDs. However, there is little discussion of diagnosis and severity differences. The articles are primarily constructed through one-off interview studies and published in nursing journals.

Table 1. Overview of Included Articles (Sorted According to Year of Publication).

Author	Aim	Sample and Country	Child/Youth Characteristics	Method	Findings
Kendall, Sloper, Lewin, and Parsons (2003a)	Examines parental views on the need for, and shape of, services for rehabilitation.	17 parents (♀♂ unclear) ⌂ United Kingdom	5–18 y/o (♀♂ unclear). Different CHDs (broad range of complexity).	One-off semistructured interviews at home.	Parents welcome more help and support from health professionals. Particular areas of concern are information about CHDs, communication with health professionals, establishing safe levels of activity, and managing CHDs in school.
Kendall, Sloper, Lewin, and Parsons (2003b)	Explores the views of youth on what would help them better deal with their CHDs.	16 children/youth ⌂ United Kingdom	8–17 y/o (7♀ 9♂). Different CHDs (broad range of complexity, however, the majority had good physical functioning).	One-off semistructured interviews at home.	While most of the young people actively manage their condition, they think more support and understanding from others would help—especially regarding issues of activity and communication.
Fernandes (2005)	xplores what it is like for parents of a newborn infant to be faced with the diagnosis of CHD.	10 parents (5♀ 5♂) ⌂ United States	Age <3 y/o (♀♂ unclear). Diagnosis focus/distribution unclear.	Phenomenologically informed study. One-off duo interviews at home and one telephone interview. Interview structure unclear.	Parents felt disbelief, shock, and like time stopped. Together with hospitalization and surgery, the diagnosis was experienced as an emotional roller coaster. Health professionals are crucial in providing information, translation, and involvement in care. Memories of this period weigh differently on parents.
Rempel and Harrison (2007)	Describes experiences of parents whose child with HLHS underwent Norwood surgery.	16 parents (9♀ 7♂) ⌂ Canada	2 months–5 y/o (6♀ 3♂). All had HLHS and had been through one or several surgeries starting with the Norwood surgical procedure.	Multiple, individual, unstructured, interactive interviews with parents in person (setting unclear) or by telephone. Based on constructivist grounded theory.	Extraordinary parenting in a context of uncertainty was evident as parents simultaneously safeguarded their child's precarious survival as well as their own survival.
Moola, Faulkner, Kirsh, and Kilburn (2008)	Explores perceptions toward physical activity and sport in the lives of youth with CHDs.	13 children/youth + parents (*n* and ♀♂ unclear) ⌂ Canada	10–18 y/o (7♀ 6♂). Different CHDs (all complex).	One-off, semistructured, individual (12 with parents present) interviews at the hospital based on social self-efficacy theory.	Sport was not valued despite the belief that it is essential for good health. Low self-efficacy and fatigue were influenced by covert fears and exclusion and decreased the value of physical activity. Nontraditional activities, support from others, and perceptions of mastery enabled participation.
Bjorbækmo and Engelsrud (2008)	Examines how children with serious CHDs experience movement and physical activities.	11 children/youth + parents (*n* and ♀♂ unclear) ⌂ Norway	7–12 y/o (♀♂ unclear). Different CHDs (all had undergone multiple and complex surgical corrections within the first year of life).	One-off interviews mainly at home and inspired by the new empathic approach followed by observations during a day at school. Interview structure unclear.	They moved to be social, and despite physical limitations for which they adjusted, they considered themselves to have the same capabilities as peers. They wanted others to take their limitations into consideration but simultaneously disliked disclosure.

(continued)

Table 1. (continued)

Author	Aim	Sample and Country	Child/Youth Characteristics	Method	Findings
Connor, Kline, Mott, Harris, and Jenkins (2010)	Describes the cost burden of CHDs and the associated social impact experienced by families.	32 parents (♀♂ unclear) ⌂ United States	Age unclear, either 0–5 or 0–15 y/o (♀♂ unclear). Currently admitted for surgery. Different CHDs (various degrees of complexity).	Exploratory semistructured interviews at the hospital + follow-up call. In all, 12 of 20 were with both parents, the rest with the parent responsible for household finances.	Cost was described as financial, emotional, and family burden. Disease complexity and parent's socioeconomic status was linked to higher levels of stress, whereas prenatal diagnosis enabled early considerations of the financial situation.
Moola, Fusco, and Kirsh (2011)	Explores barriers to physical activity for youth with CHDs.	17 children/youth ⌂ Canada	11–17 y/o (10♀ 7♂). Different CHDs (severe and complex with complicated and intensive medical histories.	One-off, individual, semistructured interviews at the hospital informed by the social model of disability (five with parents present).	Misrepresentations and misunderstandings of CHDs, disclosure dilemmas, inappropriate social and structural conditions in physical education, and the youths struggle to understand themselves as normal were described as barriers.
Ravindran and Rempel (2011)	Looks at grandparents' involvement with siblings of preschool children with HLHS.	15 grandparents (10♀ 5♂) ⌂ Canada	Precise age unclear but preschool children (♀♂ unclear). All had HLHS. Siblings 1½–6 y/o, were healthy, active, and experiencing normal development (♀♂ unclear).	One-off individual interviews either in person (setting unclear) or by telephone. Interview structure unclear.	Grandparents stepped into a parent role with siblings by attending to their daily care, play, and relational needs, while parents were occupied with the hospitalized child. They also tried to sustain parent-child and child-sibling relationships.
Lee and Rempel (2011)	Describes the role of normalization for parents of children with HLHS.	16 parents (9♀ 7♂) ⌂ Canada	2–60 m/o (6♀ 3♂). All had HLHS and had undergone a series of high-risk surgeries, incl. Norwood procedure.	Multiple, individual, semistructured interviews with mothers and fathers at home or by telephone.	Parents engaged in a balancing act between worrying about their children's vulnerability and marveling at their children's survival, using normalization as a coping strategy.
Rempel, Rogers, Ravindran, and Magill-Evans (2012)	Conceptualizes needs of parents of young children with HLHS.	25 parents (15♀ 10♂) + 28 grandparents (17♀ 11♂) ⌂ Canada	½–4½ y/o (♀♂ unclear). All had HLHS and had undergone the Sano surgical approach.	One-off individual interviews. Interview structure and setting unclear.	Found five facets of parenting: Hands-off parenting, expert parenting, uncertain parenting, and supported parenting.
Harvey, Kovalesky, Woods, and Loan (2013)	Examines the experiences of mothers of infants undergoing complex heart surgery.	8+7 parents (♀) ⌂ United States	0–5 y/o (♀♂ unclear). Different CHDs (moderate to severe diagnosed postnatally, and needed heart surgery before their first birthday).	Written journals about days before surgery, the day of surgery, and days after surgery.	Mothers described the surgical period as one of intense fluctuating emotion, where they had to navigate the medical world, deal with the unknown, face the possibility of their child's death, search for meaning, and "mother through it all."
Re, Dean, and Menahem (2013)	Explores experiences of mothers of infants with CHDs who required surgery in early infancy.	26 parents (♀) ⌂ Australia	2 m/o (♀♂ unclear). Different CHDs (all complex and subjected to recent cardiac surgery).	One-off semistructured interviews. Setting unclear.	Almost all participants described acute stress symptoms relating to the diagnosis and the infant's surgery. Most mothers reported that the interview helped them to think through what had happened to them and their infant.
Shearer, Rempel, Norris, and Magill- Evans (2013)	Looks at how adolescents with CHDs describe everyday life and relate to quality of life.	10 children/youth ⌂ Canada	13–17 y/o (6♀ 4♂). Different CHDs (had at least two heart surgeries).	Individual semistructured interviews, but most youth participated in several interviews. In all, 10 interviews took place in homes, seven by telephone, and five at the hospital.	Most viewed themselves as normal, situating their CHDs into the foreground or background as it suited their needs. They spoke of quality-of-life issues in a concrete manner, focusing on physical limitations and their need to fit in.

Table 1. (continued)

Author	Aim	Sample and Country	Child/Youth Characteristics	Method	Findings
Sikora and Janusz (2014)	Describes the caregiving behavior and experience of mothers after their child's surgery.	4 parents (♀) ⌂ Poland	5 weeks–1¼ y/o (♀♂ unclear). Different CHDs (had gone through at least one surgery).	One-off, individual, narrative interviews. Interview structure and setting unclear.	Mothers described how surgery could lead to loss of control and the feeling of maternal competence. They also described coping mechanisms to ward off fear, the difficulties of support, ambivalent relationships with medical staff, and being torn between hope and fear.
Bruce, Lilja, and Sundin (2014)	Illuminates the meanings of support as disclosed by mothers of children with CHDs.	10 parents (♀) ⌂ Sweden	3–12 y/o (♀♂ unclear). Different CHDs (the children visited the pediatric cardiac outpatient clinic regularly, at least once per year).	One-off, individual, narrative interviews (1 with spouse) based on a phenomenological- hermeneutic approach. Interview structure unclear. In all, three interviews at the university, one at the mother's office, and five at libraries.	Mothers wanted to be supported in being near their children, while also being able to take a break. They wanted personal, holistic, and informative care from healthcare staff, and good support was experienced as highly contextual.
Moola, Faulkner, White, and Kirsh (2015)	Explores the meaning of camp for children with CHDs.	15 children/youth ⌂ Canada	9–16 y/o (9♀ 6♂). Different CHDs.	One-off semistructured interviews at the hospital after camp based on an interpretive phenomenological approach.	Camp allowed youth to engage in free, unrestricted movement and forge friendships with peers with CHD. Social media allowed youth to maintain camp friendships over time.
Burström, Öjmyr-Joelsson, Bratt, Lundell, and Nisell (2016)	Describes the needs of adolescents with CHDs and their parents in regard to transition to adult care.	13 children/youth + 12 parents (7♀ 5♂) ⌂ Sweden	Age unclear, but the inclusion criteria were 16–18 y/o (7♀ 6♂). Different CHDs (moderate to complex).	One-off, individual, semistructured interviews at the hospital (two interviews at parent's office).	Transfer to adult care was considered a natural step but caused concerns about continuity of care, relationships with medical staff, and transferal of knowledge and responsibility. Some parents experienced transition as a relief, others as sorrow. Youth were still very confident with parents in charge.
Obas, Leal, Zegray, and Rennick (2016)	Explores parents' experiences of transition from PICU to the surgical ward.	9 parents (♀♂ unclear) ⌂ Canada	2 m/o–14 y/o (♀♂ unclear). Different CHDs (first time transferring from the PICU to the surgical ward).	One-off, individual, semistructured interviews in the hospital following the transfer from PICU to the surgical ward.	Parents described mixed feelings of happiness and uncertainty, and how nurses played an important role in preparing for transfer. Once transferred, parents had to come to terms with new expectations and a challenging new environment.
Bruce, Lindh, and Sundin (2016)	Illuminates the meanings of support by fathers of children with CHDs.	5 parents (♂) ⌂ Sweden	3–12 y/o (1♀ 4♂). Different CHDs (had been visiting the pediatric cardiac reception at least once per year).	One-off, individual, narrative interviews (one with spouse) based on a phenomenological- hermeneutic approach. In all, four interviews at the university, one at home.	Support was closely connected to being in a sharing relationship with others—both within the family and with healthcare professionals. Healthcare professionals are important for involving fathers in care.

Analytical Approach

As is often the case with meta-ethnography (Noblit & Hare, 1988), our reading, coding, and analyzing was a repeated and dynamic process, starting well before the final selection of articles. Translations (Noblit & Hare, 1988; Thorne et al., 2004) were done through "initial coding" (Saldaña, 2009, pp. 81–85) where empirical quotes and analytical points were coded if they seemed intuitively relevant, interesting, or telling, followed by "focused coding" (Saldaña, 2009, pp. 155–159) through continuous readings of articles, codes, and juxtapositions of these, resulting in overall themes with subthemes.

Due to the heterogeneity of the included articles, a new interpretive framework was needed that aimed "to discover a 'whole' among a set of parts" (Noblit & Hare, 1988, p. 63) and would reach beyond the original interpretations. As we developed themes, reread, and compared the articles, we found that they collectively described different aspects of continuous paradoxes. Therefore, we developed a new interpretive framework called the chronic paradoxes of CHD living defined as experiences and strategies that address the often life- long and contradictory nature of living with CHDs. We argue that these chronic paradoxes are a foundational and constant aspect of living with CHDs. Although some authors in the included articles used "paradox," "ambivalence," or "contradiction" when describing findings, many of the paradoxes we cover emerged in our iterative readings and comparisons, and so the framework transformed isolated descriptions of paradox to a condition of life with CHDs. Our framework was also inspired by, and extends, concepts found in research on children's illness, in general, such as "the embrace of paradox" (Larson, 1998) and "the paradox of hope" (Mattingly, 2010), as well as research on parents' experiences of CHDs such as "walking a tight- rope" (Brown, 2003). Where these authors focus on how a paradoxical space between grief and joy as well as fear and hope is the outset for parents' emotional well-being and care work, our framework extends this with family perspectives beyond parents and by including more paradoxes than those related to grief and joy, hope and fear.

Results

In the following analysis, we trace the multiple paradoxes faced by families during times of transition, as they seek out normalities, and when imagining futures (see Figure 2). We emphasize how these paradoxes are perceived differently in the family and at different times.

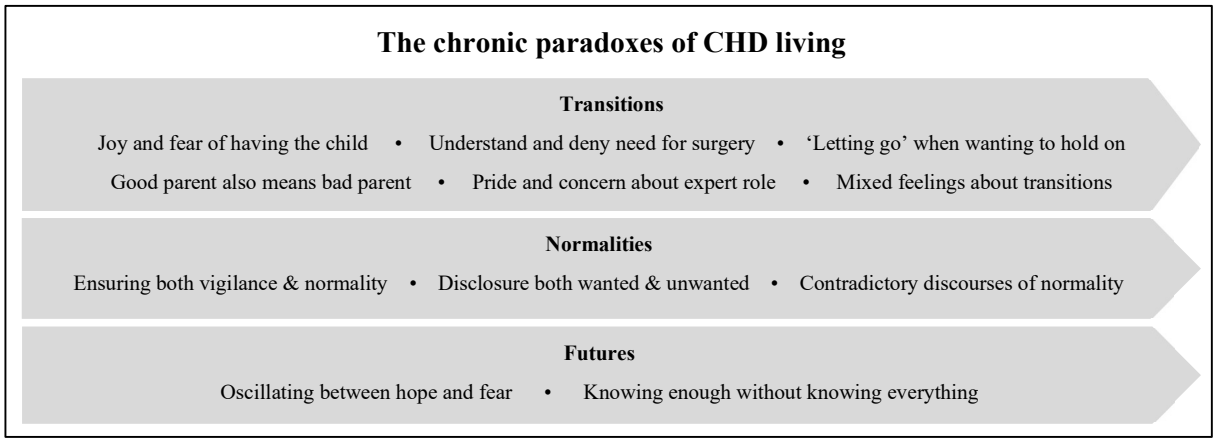

Figure 2. The chronic paradoxes of CHD living within the themes of transitions, normalities, and futures

Transitions

Individually, the included articles focus on a limited age group or period. Together, they provide comprehensive insights into the illness trajectories of children and youth with CHDs and their families. As such, we learn how living with CHDs is characterized by a series of transitions that generate a number of paradoxical experiences and actions.

The first paradox emerges at the time of diagnosis, when the joy of expecting or becoming a parent collides and coexists with the sorrow and shock of learning that one's child has a CHD (Fernandes, 2005; Harvey, Kovalesky, Woods, & Loan, 2013; Sikora & Janusz, 2014). A mother describes it as "thrilling, then devastat- ing, all in one little short period" (Fernandes, 2005, p. 325). A transition from having a surgically untreated to a treated child often follows, which many parents describe as the most difficult period of living with CHDs (Harvey et al., 2013; Re, Dean, & Menahem, 2013; Sikora & Janusz, 2014), a time filled with "intense fluctuating emotion" (Harvey et al., 2013, p. 401). Parents, for example, both understand and deny or are insecure about the need for and possible outcomes of surgery (Re et al., 2013; Rempel & Harrison, 2007):

> This is to save him, this is to save him. If I run away with him, it's not going to help him at all, you know . . . I just thought I was gonna lose him . . . it was all . . . Yeah. I thought they're gonna take him and kill him [original emphasis], you know, [laughs]. (Mother of an infant with CHD; Re et al., 2013, p. 280)

A diagnosis of CHD with subsequent hospitalizations and surgical treatment plunges families into an unfamiliar medical world (Harvey et al., 2013) and demands major family reorganizations. One parent

(often the mother) usually stays at the hospital, while the other parent is at home taking care of siblings, often with the help of grand- parents (Ravindran & Rempel, 2011). In general, much family attention is focused on the child with CHD. This leads to the paradox of being a good parent (to the child with CHD) while simultaneously being a bad or less present parent (to siblings); a situation that parents and grand-parents describe as the cause of many worries and as something that affects siblings significantly (Bruce, Lilja, & Sundin, 2014; Bruce, Lindh, & Sundin, 2016; Connor et al., 2010; Harvey et al., 2013; Ravindran & Rempel, 2011). A mother of a child with a Ventricular Septal Defect (VSD) describes this as, "I have to split myself in two" (Obas, Leal, Zegray, & Rennick, 2016, p. 6).

Parents also have to engage in what Rempel, Rogers, Ravindran, and Magill-Evans (2012) call "expert parenting" (pp. 4–5). They acquire vocabulary and knowledge of CHDs, observe, coordinate, and make decisions concerning their child's care, and achieve new skills for taking care of a symptomatic or newly surgically treated child (Bruce et al., 2014; Fernandes, 2005; Lee & Rempel, 2011; Obas et al., 2016; Rempel & Harrison, 2007; Sikora & Janusz, 2014). However, in the hospital during and just after surgery, expert parenting mainly relates to giving consent and grasping the diagnosis and treatments. For a short period, expert parenting is coupled with "hands-off parenting" (Rempel et al., 2012, pp. 3–4) or "letting go" (Harvey et al., 2013, p. 402), as they have to hand over all care of their child to nurses and doctors. This causes a paradox characterized by "a painful split between the desire for intimacy and providing care for the infant on their own and the necessity of subordination to the requirements of treatment," as Sikora and Janusz (2014, p. 441) put it, a point that is echoed in other studies (Bruce et al., 2014, 2015; Fernandes, 2005; Harvey et al., 2013; Rempel et al., 2012).

Parents' expert role and responsibility increases in times of transition within or from the hospital, as care and monitoring of the child with CHD are transferred from machines and medical staff to parents (Rempel et al., 2012). Paradoxically, although becoming more responsible for their child's care is what parents have yearned for, and transfer is a positive sign, parents have "mixed feelings" (Obas et al., 2016, pp. 4–5). They are concerned about the less vigilant care their child will receive and their increased role in caretaking (Obas et al., 2016; Rempel & Harrison, 2007). In general, involvement in care is a balance, as parents want to be involved but do not want to be managers of their child's medical care (Bruce et al., 2014). However, once at home, parents experience generalist health professionals to have limited knowledge about CHDs (Kendall, Sloper, Lewin, & Parsons, 2003a; Rempel et al., 2012), and, despite little recognition, parents consider themselves to be more knowledgeable, which they paradoxically express both "concern and pride" about (Rempel et al., 2012, p. 5).

Parents' role as experts, or the ones "in charge" as some youth in Burström, Öjmyr-Joelsson, Bratt, Lundell, and Nisell (2016) frame it (p. 401), can change as the children transition to adolescence and adulthood. Much like transition within or from the hospital, transitions to adult care are experienced with mixed feelings. It is both considered a natural step by youth with CHDs and somewhat of a relief by parents (Burström et al., 2016), while also causing sorrow for some parents and concerns for both groups about the transition of the expert role, transferal of knowledge, and loss of continuity in relationships with medical staff (Burström et al., 2016).

Even though living with CHD means living through many transitions and accompanying paradoxes, there are important differences in perspectives. While there is a before and after CHD for relatives, this is rarely the case for children and youth with CHDs:

> Ever since I remember, I've had it so it's not like I can tell you about a time when I didn't have it. So it's not like it really matters, or it's just a part of me now 'cause I grew up with it. (Female with CHD, 15 years old; Shearer, Rempel, Norris, & Magill-Evans, 2013, p. 32)

Normalities

In Moola et al.'s (2011) study, a 14-year-old girl with CHD poignantly says, "You want to live a normal life, but you can't live a normal life because you have this. And like I said, when you're not in the hospital, your disease doesn't exist" (p. 68). Many of the authors describe what might be thought of as paradoxes of normality as children and youth with CHDs and their families strive for, work to uphold, or describe themselves in terms of normality (Bjorbækmo & Engelsrud, 2008; Bruce et al., 2016; Burström et al., 2016; Harvey et al., 2013; Kendall et al., 2003a; Kendall, Sloper, Lewin, & Parsons, 2003b; Lee & Rempel, 2011; Moola et al., 2008; Moola, Faulkner, White, & Kirsh, 2015; Moola et al., 2011; Ravindran & Rempel, 2011; Rempel & Harrison, 2007; Shearer et al., 2013). Efforts to live a normal life seem crucial to the well-being of families while also being a stress factor (Kendall et al., 2003a), and going from the acute, hectic, and traumatic periods of hospitalization to a more stable everyday life at home, and sometimes back again, is not easy.

Parents struggle to be both vigilant and cautious expert parents, while not being excessively limiting so that their child can lead a "normal life" (Lee & Rempel, 2011, p. 185). Feeling responsible for both vigilance and normality is a paradox and a continuous "balancing act" (Lee & Rempel, 2011, p. 185). It is less so with children that have been "fixed" (Moola et al., 2008, p. 59), and more so with a child that "hasn't been fixed" (Connor et al., 2010,

p. 323), requiring multiple surgeries, such as children with Hypoplastic Left Heart Syndrome (HLHS; Rempel et al., 2012).

Children and youth recognize that it is a difficult balancing act for parents, but emphasize the importance of allowing them to control their limitations (Kendall et al., 2003b; Moola et al., 2015; Shearer et al., 2013). When this balance is not found, some defy parents (Shearer et al., 2013), whereas others come to embody their fears and become "super nervous because my family has always been so cautious of what I do," as explained by a girl in Moola et al.'s (2015) study (p. 281).

However, parents are not alone in restricting, and normality is especially threatened and at stake when it comes to physical activities (Moola et al., 2011; Shearer et al., 2013). Children and youth not only struggle with limited physical stamina due to their CHDs (Bjorbækmo & Engelsrud, 2008; Burström et al., 2016; Moola et al., 2008; Moola et al., 2011), but are also sometimes restricted by teachers and peers (Burström et al., 2016; Kendall et al., 2003a; Moola et al., 2011), even if medically imposed restrictions are relatively rare (e.g., Shearer et al., 2013). Limited physical abilities and unnecessary restrictions can make children and youth with CHDs feel different, left out, and sometimes even bullied (Kendall et al., 2003b; Moola et al., 2008; Moola et al., 2015; Moola et al., 2011). An 11-year-old girl with HLHS says,

> I'm always getting teased by the other kids. Which isn't very nice, and it really bothers me a lot. I try my best but then other people always swipe the ball from me. And I always end up having to chase them. And then I get really tired, so I have to sit down, so someone else has to take my place on the team that I'm on. (Moola et al., 2008, p. 60)

To avoid negative social reactions, children and youth with CHDs and their families navigate a paradox of (non) disclosure. Bjorbækmo and Engelsrud (2008) argue that the children and youth in their study "both want and do not want the others to know and to take into account the fact that they have 'this (heart) condition'" (p. 786), and Moola et al. (2011) argue that normalcy depends on resolving disclosure dilemmas. So, balancing the fear of being treated differently or overly cautiously if disclosing their CHDs, with the need for empathy and adjustment from others gained by disclosure, is crucial.

Disclosure is such an issue because children and youth with CHDs often "look okay" (Kendall et al., 2003b, p. 16) or "normal" (Kendall et al., 2003a, p. 23)—meaning CHDs are not necessarily readily visible or identifiable. This places a burden of disclosure on the shoulders of the children and youth with CHDs who wish people around them would "remember all the time, instead

of me having to tell them," as a boy with aortic stenosis (AS) aged 9 describes it (Kendall et al., 2003b, p. 16). Parents also feel responsible as "gatekeepers of information" (Kendall et al., 2003a, p. 23), and although less described, they also seem to struggle with the timing and amount of information necessary, as it is difficult "putting it across to people so they are not frightened, but yet they know that if there is a problem they can deal with it," as one parent put it (Kendall et al., 2003a, p. 23). Underlying disclosure paradoxes seem to be particular understandings and misconceptions of CHDs, associating them with risks of collapsing (Kendall et al., 2003b), and understandings of the heart as a particularly important organ so that CHDs must cause severe debilitation (Moola et al., 2011).

Another paradox concerns the arbitrary nature of normality, as children and youth with CHDs "simultaneously constructed themselves as both normal and not, the same and different, at one and the same time" (Moola et al., 2011, p. 68). Buttressed by the congenital nature of CHDs (Burström et al., 2016; Shearer et al., 2013), their relative invisibility (Kendall et al., 2003a; 2003b), and their skills in adjusting to physical limitations (Bjorbækmo & Engelsrud, 2008), as well as mentally foregrounding or backgrounding their CHDs as needed (Shearer et al., 2013), children and youth, supported by their parents, strive to and sometimes succeed in feeling normal in comparison with healthy peers (Bjorbækmo & Engelsrud, 2008; Moola et al., 2011; Shearer et al., 2013). However, another kind of normality is experienced together with peers with CHDs, because "they are just like you," as a girl in Moola et al. (2015) says (p. 282), and parents feel much the same about being with other parents of children or youth with CHDs (Bruce et al., 2016; Bruce et al., 2014). Still, even in CHD contexts, difference can exist as it "puts it more into perspective about how you might not necessarily have it the worst," as one youth says (Moola et al., 2015, p. 283), echoed elsewhere by parents (Bruce et al., 2014). So, "discourses of normality were contradictory" (Moola et al., 2011, p. 68), and a matter of normalities rather than one specific normality.

Futures

Thoughts about the future are present in many of the articles, especially in parent perspectives focused on the time of diagnosis or surgery, or in cases where children have the complex diagnosis of HLHS. Also, these thoughts are often of a paradoxical nature, as "you have to deal, in your head . . . with the good and the bad," in the words of a mother (Rempel et al., 2012, p. 5). On one hand, parents and grandparents express a range of fears concerning death, deterioration, or comorbidities (Bruce et al., 2014; Connor et al., 2010; Fernandes, 2005; Harvey et al., 2013; Lee & Rempel, 2011; Rempel & Harrison, 2007; Rempel et al., 2012), limits to and possibilities for children's future employment and

physical abilities (Kendall et al., 2003a; Rempel & Harrison, 2007), impact on relationship between parents (Connor et al., 2010), and future emotional trauma for siblings (Ravindran & Rempel, 2011). On the other hand, hope and positive outcomes are created by downplaying or pushing away concerns (Fernandes, 2005; Rempel & Harrison, 2007), normalizing the conditions and referring to the children and youth as "normal" as well as "extraordinary" (Lee & Rempel, 2011, pp. 184, 185; Rempel & Harrison, 2007, pp. 830– 831), and putting faith in God or doctors (Harvey et al., 2013; Lee & Rempel, 2011; Rempel & Harrison, 2007), or in medical advances, as this quote illustrates:

> This Fontan hopefully keeps her going till her 20s and then hopefully we've got some other option, maybe even something other than a heart transplant. Like a ventricular replacement or something like that, since we expect it'll be the ventricle that's going to be the first problem. I kind of base my hopes on that. (Father of a child with HLHS; Rempel & Harrison, 2007, p. 831)

These strategies of curbing fear and worry do not mean that parents are in denial about their child's prognosis, but rather that the two opposing forces seem to exist simultaneously (Rempel et al., 2012). It is described as "oscillating" between or "a mixture" of hope and fear (Lee & Rempel, 2011, p. 185; Rempel et al., 2012, p. 5; Sikora & Janusz, 2014, p. 441) or "two extremes of vulnerability and optimistic appraisal" (Lee & Rempel, 2011, p. 185). In the center of the two extremes is the ideal of "a normal life," and Lee and Rempel (2011) suggest that this "emotional oscillation" is the motivation for parents to become active and creative agents of their child's care and their own well-being (p. 185).

Although there is sparse focus on child and youth perspectives on the future in the included articles, there are a few indications of a paradox similar to parents' oscillation between hope and fear. However, it is mainly related to information about CHDs:

> That's enough, I don't need to know . . . "your heart is pumping so and so . . .," but it should be pumping so . . ., If I knew everything that was wrong with my heart . . . that like, I could die . . ., I'd just get depressed and want to crawl into a hole. (An 18-year-old female with CHD; Burström et al., 2016, p. 401)

Most information on CHDs passes through parents (Burström et al., 2016; Kendall et al., 2003a, 2003b) who struggle with timing and how much they should disclose to their children (Kendall et al.,

2003a). Furthermore, as many children and youth with CHDs do not remember their surgeries or have a complete understanding of their condition (Burström et al., 2016; Kendall et al., 2003b), they, therefore, have little information to imagine their futures with and through. However, they do express some concerns about future physical abilities (Moola et al., 2008), impact on or possibility for pregnancy and issues of heredity (Burström et al., 2016), side effects of medication (Shearer et al., 2013), possible deteriorations (Burström et al., 2016), and in a few instances direct fears of death (Burström et al., 2016). Although attitudes differ on how much information is ideal (Kendall et al., 2003b, p. 16), there seems to be a paradox and a difficult balance of knowing enough or knowing what it is all about without knowing everything (Burström et al., 2016).

Discussion

Based on 20 articles from seven countries that included perspectives from children and youth with CHDs as well as parents and grandparents, we have argued that immense medical progress notwithstanding, living with CHDs is characterized by chronic paradoxes that emerge out of multiple transitions, negotiations of normality, and orientations toward the future.

Further to our analytical inspirations and the literature reviewed here, experiences of paradox (or related concepts of ambivalence and dilemma) have been found in studies on other kinds of children's illness (e.g., Jones, Parker-Raley, & Barczyk, 2011; Rallison & Raffin-Bouchal, 2013), on congenital anomalies in general (Rempel, 2005), as well as in studies on acquired cardiovascular disease in adults such as heart failure (e.g., Hellesø, Eines, & Fagermoen, 2012; Overgaard, Grufstedt Kjeldgaard, & Egerod, 2012). Particularly on CHDs, authors of a previous study found fathers of newly diagnosed infants with CHDs to experience "conflicting responses," as they are both happy and sad, want and fear attachment, need control while losing it, and struggle to support others by hiding their own emotions (Clark & Miles, 1999).3 Furthermore, studies of teenage years as well as young adulthood (age range = 13–39) with CHDs point to experiences of being in between sick and healthy, normal and different, vigilant and unworried, hopeful and anxious, dependent or independent, and open and closed about the diagnosis, which indeed indicate a chronicity of paradoxes beyond childhood (e.g., Berghammer, Dellborg, & Ekman, 20064; Sparacino et al., 1997; Tong et al., 1998).5

The novelty is, therefore, not the finding of paradox itself, but the finding that experiences of paradoxes in life with CHDs are not isolated cases pertaining only to a particular age or place in the illness trajectory. Rather, what we are highlighting is the chronic nature of these para- doxes, and, consequently, how these chronic paradoxes profoundly shape the everyday lives of children and youth

living with CHDs and their families, as they together navigate and weave their way through schooling, medical care, and social life.

The question then remains why paradox is so inherent in experiences of chronic illness in general and experiences of CHDs, in particular. Gitte Wind (2009), who has researched chronic illness narratives, argues that people with chronic conditions inhabit a land in-between the sick and the well. While this is a meaningful frame for under- standing the paradoxical nature of living with chronic conditions in general, there are several factors unique to CHDs that contribute to the creation of paradoxes. First, children and youth with CHDs have experienced continuous improvements in survival rates so that most of them can now expect to reach adulthood (Larsen et al., 2017; Lüscher, 2017). However, they still have lower survival rates than background populations and live with lifelong medical checkups, possible neurodevelopmental disorders, reduced exercise capacity and need for new surgical or catheter-based interventions, as well as risks of complications such as arrhythmia, hypertension, valvular heart disease, and heart failure (Bouma & Mulder, 2017; Larsen et al., 2017; Lüscher, 2017). Also, the visual subtlety of CHDs, indicated in our review and echoed elsewhere (e.g., Berghammer et al., 2006; Chiang et al., 2015; Messias, Gilliss, Sparacino, Tong, & Foote, 1995), coexists with a stigma and drama around heart conditions found both in the included articles as well as studies on heart conditions in children in general (e.g., Desai, Sutton, Staley, & Hannon, 2014; McMurray et al., 2001), perhaps due to understandings of the heart as a particularly vital organ and its symbolic connection to personhood and life itself (Jensen, 2009, 2011). Furthermore, prognostic in-betweenness, invisibility, stigma, and drama are wrapped into extreme opposites in the illness trajectory, as seen in the section “Transitions.” Finally, as described in this review, the illness trajectory is not equally understood or remembered in the family, and previous research has also found children and youth to lack knowledge and understanding of their CHDs (Janssens et al., 2016; Veldtman et al., 2000), whereas parents overall have a good understanding (Chessa et al., 2005; Löbel, Geyer, Grosser, & Wessel, 2012).

We have suggested that the paradoxes of CHD living are chronic, although some paradoxes relate to issues that are transitory while others are long term. Even though not all paradoxes are lifelong or long term, there is chronicity to the experience of paradox in living with CHDs as well as to the conditions from which they stem. Therefore, it seems pertinent that health professionals recognize and address the chronic paradoxes of CHDs in their interactions with families living with CHDs and that they are attentive to and encourage discussion of experiences of transition, normalizing strategies, and thoughts about the future. Simply telling children and youth with CHDs and their families that they are normal and should live a nor- mal life is to ignore half of the paradoxical

realities they have to navigate. Furthermore, attention should be paid to the continuous transferal of knowledge from parent, nurse, and doctor to child and youth, as well as different knowledge perspectives and wishes. It is also important to engage parents in a continuous conversation about their role as expert parents, as well as encourage and facilitate positively valued involvement in care. Finally, health professionals should discuss, and partner with families around disclosure issues, and, together with patient associations, provide tailored information on CHDs for schools, afterschool clubs, and the parents of children's friends.

Conclusion and Suggestions for Future Research

By introducing an analytical frame that points to the chronic paradoxes of CHD living, we have highlighted the complexities of living with CHDs as chronic conditions and demonstrated that balancing and negotiating many different paradoxes generates both possibilities and struggles. As the effects of biomedical advances within the field of CHDs continue to shape the lives of new generations, we urge continued scholarly attention to examining if and how paradoxes remain quintessential to the experience of living with CHDs. Moreover, future research should engage with the limitations we found, such as the lack of perspectives from siblings and young children with CHDs (under the age of 7), prolonged engagement or observations, discussion of severity differences, and the underrepresentation of male perspectives.

Notes

[1] Congenital heart defects are often abbreviated as CHDs (see, for example, Bruce, Lindh, & Sundin, 2016; Jenkins, 2017); however, this should not be confused with coronary heart disease, which often goes under the same abbreviation.

[2] Some of the previous reviews mentioned in the introduction, for example, included articles with both structural and nonstructural conditions under the term congenital heart disease.

[3] Although predominantly focused on structural CHDs, this study also includes at least one father of an infant with a nonstructural heart condition (supraventricular tachycardia), and it was, therefore, excluded from this review.

[4] Although predominantly focused on structural CHDs, this study also includes one adult with a nonstructural heart condition (dilated and restrictive cardiomyopathy).

[5] The review by Dellafiore et al. (2017; building on six articles whereof only four were specifically focused on CHD) also found contradictions to be a theme in the experiences of parents of adolescents with CHD.

References

Berghammer, M., Dellborg, M., & Ekman, I. (2006). Young adults experiences of living with congenital heart dis- ease. International Journal of Cardiology, 110, 340–347. doi:10.1016/j.ijcard.2005.08.006

Bjorbækmo, W., & Engelsrud, G. (2008). "I am almost like a fish": An investigation of how children with congenital heart disease experience and perform movement in daily life. Child: Care, Health and Development, 34, 781–788. doi:10.1111/j.13652214.2008.00851.x

Bouma, B. J., & Mulder, B. J. M. (2017). Changing landscape of congenital heart disease. Circulation Research, 120, 908–922. doi:10.1161/CIRCRESAHA.116.309302

Brown, P. (2003). Walking a tightrope: Living with your child's congenital heart disease (Doctoral dissertation). Retrieved from ProQuest dissertations and theses database. (3090854)

Bruce, E., Lilja, C., & Sundin, K. (2014). Mothers' lived experiences of support when living with young children with con- genital heart defects. Journal for Specialists in Pediatric Nursing, 19, 54–67. doi:10.1111/jspn.12049

Bruce, E., Lindh, V., & Sundin, K. (2016). Support for fathers of children with heart defects. Clinical Nursing Research, 15, 254–272. doi:10.1177/1054773815586351

Burström, Å., Öjmyr-Joelsson, M., Bratt, E.-L., Lundell, B., & Nisell, M. (2016). Adolescents with congenital heart dis- ease and their parents: Needs before transfer to adult care. The Journal of Cardiovascular Nursing, 31, 399–404. doi:10.1097/JCN.0000000000000288

Carroll, C., Booth, A., & Lloyd-Jones, M. (2012). Should we exclude inadequately reported studies from qualitative systematic reviews? An evaluation of sensitivity analyses in two case study reviews. Qualitative Health Research, 22, 1425–1434. doi:10.1177 /1049732312452937

Chessa, M., De Rosa, G., Pardeo, M., Negura, D. G., Butera, G., Giamberti, A., . . . Carminati, M. (2005). What do par- ents know about the malformations afflicting the hearts of their children? Cardiology in the Young, 15, 125–129. doi:10.1017/S1047951105000284

Chiang, Y.-T., Chen, C.-W., Su, W.-J., Wang, J.-K., Lu, C.-W., Li, Y.-F., & Moons, P. (2015). Between invisible defects and visible impact: The life experiences of adolescents and young adults with congenital heart disease. Journal of Advanced Nursing, 71, 599–608. doi:10.1111/jan.12546

Chong, L. S. H., Fitzgerald, D. A., Craig, J. C., Manera, K. E., Hanson, C. S., Celermajer, D., . . . Tong, A. (2018). Children's experiences of congenital heart disease: A sys- tematic review of qualitative studies. European Journal of Pediatrics, 177, 319–336. doi:10.1007/s00431-017-3081-y

Clark, S. M., & Miles, M. S. (1999). Conflicting responses: The experiences of fathers of infants diagnosed with severe congenital heart disease. Journal of the Society of Pediatric Nurses, 4, 7–14. doi:10.1177/1359105316628757

Connor, J. A., Kline, N. E., Mott, S., Harris, S. K., & Jenkins, K. J. (2010). The meaning of cost for families of children with congenital heart disease. Journal of Pediatric Health Care, 24, 318–325. doi:10.1016/j.pedhc.2009.09.002

Critical Appraisals Skills Programme. (2014). CASP checklists. Retrieved from http://www.casp-uk.net/#!checklists/cb36

Danmarks Statistik. (n.d.). Begreber [Concepts]. Retrieved from http://www.dst.dk/da/Statistik/dokumentation/hvad- betyder.aspx

Dellafiore, F., Domanico, R., Flocco, S. F., Pittella, F., Conte, G., Magon, A., . . . Caruso, R. (2017). The life experience of parents of congenital heart disease adolescents: A meta-synthesis. Archives of Nursing Practice and Care, 3, 031–037. doi:10.17352/anpc.000022

Desai, P. P., Sutton, L. J., Staley, M. D., & Hannon, D. W. (2014). A qualitative study exploring the psychosocial value of weekend camping experiences for children and adoles- cents with complex heart defects. Child: Care, Health and Development, 40, 553–561. doi:10.1111/cch.12056

Dixon-Woods, M., Bonas, S., Booth, A., Jones, D. R., Miller, T., Sutton, A. J., . . . Young, B. (2006). How can systematic reviews incorporate qualitative research? A critical perspective. Qualitative Research, 6, 27–44. doi:10.1177/1468794106058867

Dodge-Khatami, A. (2016). Advances and research in con- genital heart disease. Translational Pediatrics, 5, 109–111. doi:10.21037/tp.2016.05.01

Edwards, W. D. (2001). Classification and terminology of cardiovascular anomalies. In H. D. Allen, H. P. Gutgesell, E. B. Clark, & D. J. Driscoll (Eds.), Moss and Adam's heart disease in infants, children and adolescents including the fetus and young adult (6th. ed., pp. 118–142). Philadelphia: Lippincott Williams & Wilkins.

Fernandes, J. (2005). The experience of a broken heart. Critical Care Nursing Clinics of North America, 17, 319–327. doi:10.1016/j.ccell.2005.08.001

France, E. F., Ring, N., Thomas, R., Noyes, J., Maxwell, M., & Jepson, R. (2014). A methodological systematic review of what's wrong with meta-ethnography report- ing. BMC Medical Research Methodology, 14, Article 119. doi:10.1186/1471-2288-14-119

Garne, E., Olsen, M. S., Johnsen, S. P., Hjortdal, V., Andersen, H. Ø., Nissen, H., . . . Videbæk, J. (2012). How do we define congenital heart defects for scientific studies? Congenital Heart Disease, 7, 46–49. doi:10.1111/j.1747- 0803.2011.00581.x

Harvey, K. A., Kovalesky, A., Woods, R. K., & Loan, L. A. (2013). Experiences of mothers of infants with congenital heart disease before, during, and after complex cardiac surgery. Heart & Lung: The Journal of Acute and Critical Care, 42, 399–406. doi:10.1016/j.hrtlng.2013.08.009

Hawker, S., Payne, S., Kerr, C., Hardey, M., & Powell, J. (2002). Appraising the evidence: Reviewing disparate data systematically. Qualitative Health Research, 12, 1284–1299. doi:10.1177/1049732302238251

Hellesø, R., Eines, J., & Fagermoen, M. S. (2012). The significance of informal caregivers in information management from the perspective of heart failure patients: Significance of informal caregivers. Journal of Clinical Nursing, 21, 495–503. doi:10.1111/j.1365-2702.2011.03927.x

Jackson, A. C., Frydenberg, E., Liang, P.-T., Higgins, R. O., & Murphy, B. M. (2015). Familial impact and coping with child heart disease: A systematic review. Pediatric Cardiology, 36, 695–712. doi:10.1007/s00246-015-1121-9

Janssens, A., Goossens, E., Luyckx, K., Budts, W., Gewillig, M., Moons, P., & i-DETACH investigators. (2016). Exploring the relationship between disease-related knowledge and health risk behaviours in young people with con- genital heart disease. European Journal of Cardiovascular Nursing, 15, 231–240. doi:10.1177/1474515114565214

Jenkins, K. J. (2017). Congenital Heart Defects: Amazing advances and ongoing challenges. Journal of the American College of Cardiology, 69, 2733–2734. doi:10.1016/j. jacc.2017.04.008

Jensen, A. M. B. (2009). Mistede liv og nye chancer: Kropsdelenes komplekse sociale betydninger i organdonationsfeltet [Lost lives and new chances: The complex social meaning of body parts in the field of organ donation]. Tidsskrift for Forskning i Sygdom Og Samfund, 11, 31–50. Retrieved from https://tidsskrift.dk/sygdomogsam- fund/article/view/2699/2324

Jensen, A. M. B. (2011). Orchestrating an exceptional death: Donor family experiences and organ donation in Denmark (Doctoral dissertation, University of Copenhagen, Denmark). Retrieved from https://www.researchgate.net/ publication/291957907_Orchestrating_an_Exceptional_Death_Donor_Family_Experiences_and_Organ_Donation_in_Denmark_Phd_Series_no_69_Department_ of_Anthropology_University_of_Copenhagen_2011

Jones, B. L., Parker-Raley, J., & Barczyk, A. (2011). Adolescent cancer survivors: Identity paradox and the need to belong. Qualitative Health Research, 21, 1033–1040. doi:10.1177/1049732311404029

Kendall, L., Sloper, P., Lewin, R. J., & Parsons, J. M. (2003a). The views of parents concerning the planning of services for rehabilitation of families of children with congenital cardiac disease. Cardiology in the Young, 13, 20–27. doi:10.1017/S1047951103000052

Kendall, L., Sloper, P., Lewin, R. J., & Parsons, J. M. (2003b). The views of young people with congenital cardiac disease on designing the services for their treatment. Cardiology in the Young, 13, 11–19. doi:10.1017/S1047951103000040

Kosta, L., Harms, L., Franich-Ray, C., Anderson, V., Northam, E., Cochrane, A., . . . Jordan, B. (2015). Parental experiences of their infant's hospitalization for cardiac surgery. Child: Care, Health & Development, 41, 1057–1065. doi:10.1111/cch.12230

Larsen, S. H., Olsen, M., Emmertsen, K., & Hjortdal, V. E. (2017). Interventional treatment of patients with congenital heart disease. Journal of the American College of Cardiology, 69, 2725–2732. doi:10.1016/j.jacc.2017.03.587

Larson, E. (1998). Reframing the meaning of disability to families: The embrace of paradox. Social Science & Medicine, 47, 865–875. doi:10.1016/S0277-9536(98)00113-0

Lee, A., & Rempel, G. R. (2011). Parenting children with hypoplastic left heart syndrome: Finding a balance. Journal for Specialists in Pediatric Nursing, 16, 179–189. doi:10.1111/ j.1744-6155.2011.00289.x

Löbel, A., Geyer, S., Grosser, U., & Wessel, A. (2012). Knowledge of congenital heart disease of mothers: Presentation of a standardized questionnaire and first results. Congenital Heart Disease, 7, 31–40. doi:10.1111/ j.17470803.2011.00591.x

Lüscher, T. F. (2017). Congenital heart disease: Some progress, but still the challenge of a lifetime! European Heart Journal, 38, 2021–2023. doi:10.1093/eurheartj/ehx372

Manderson, L., & Smith-Morris, C. (2010). Chronic conditions, fluid states: Chronicity and the anthropology of illness. New Brunswick, NJ: Rutgers University Press.

Mattingly, C. (2010). The Paradox of hope: Journeys through a clinical borderland (1st ed.). Berkeley: University of California Press.

McMurray, R., Kendall, L., Parsons, J. M., Quirk, J., Veldtman, G. R., Lewin, R. J. P., & Sloper, P. (2001). A life less ordinary: Growing up and coping with congenital heart disease. Coronary Health Care, 5, 51–57. doi:10.1054/ chec.2001.0112

Messias, D. K. H., Gilliss, C. L., Sparacino, P. S. A., Tong, E. M., & Foote, D. (1995). Stories of transition: Parents recall the diagnosis of congenital heart defect. Family Systems Medicine, 13, 367–377. doi:10.1037/h0089280

Moola, F. J., Faulkner, G. E., Kirsh, J. A., & Kilburn, J. (2008). Physical activity and sport participation in youth with congenital heart disease: Perceptions of children and parents. Adapted Physical Activity Quarterly, 25, 49–70. doi:10.1123/apaq.25.1.49

Moola, F. J., Faulkner, G. E., White, L., & Kirsh, J. (2015). Kids with special hearts: The experience of children with congenital heart disease at Camp Willowood. Qualitative Research in Sport, Exercise and Health, 7, 271–293. doi:1 0.1080/2159676X.2014.926968

Moola, F. J., Fusco, C., & Kirsh, J. A. (2011). "What I wish you knew": Social barriers toward physical activity in youth with congenital heart disease (CHD). Adapted Physical Activity Quarterly, 28, 56–77. doi:10.1123/apaq.28.1.56

Musa, N. L., Hjortdal, V., Zheleva, B., Murni, I. K., Sano, S., Schwartz, S., & Staveski, S. L. (2017). The global burden of paediatric heart disease. Cardiology in the Young, 27, S3–S8. doi:10.1017/S1047951117002530

National Heart, Lung and Blood Institute. (2017). What are congenital heart defects? Retrieved from https://www. nhlbi.nih.gov/health/health-topics/topics/chd#

Noblit, G. W., & Hare, D. (1988). Meta-ethnography. Thousand Oaks, CA: Sage.

Obas, K. A., Leal, J. M., Zegray, M., & Rennick, J. E. (2016). Parental perceptions of transition from intensive care fol- lowing a child's cardiac surgery. Nursing in Critical Care, 21, e1–e9. doi:10.1111/nicc.12202

Overgaard, D., Grufstedt Kjeldgaard, H., & Egerod, I. (2012). Life in transition: A qualitative study of the illness experience and vocational adjustment of patients with left ventricular assist device. The Journal of Cardiovascular Nursing, 27, 394–402. doi:10.1097/JCN.0b013e318227f119

Popay, J., & Mallinson, S. (2010). Qualitative research review and synthesis. In I. Bourgeault, R. Dingwall, & R. De Vries (Eds.), The SAGE handbook of qualitative methods in health research (pp. 289–306). London: Sage. doi:10.4135/9781446268247.n16

Rallison, L. B., & Raffin-Bouchal, S. (2013). Living in the in- between: Families caring for a child with a progressive neurodegenerative illness. Qualitative Health Research, 23, 194–206. doi:10.1177/1049732312467232

Ravindran, V. P., & Rempel, G. R. (2011). Grandparents and siblings of children with congenital heart disease. Journal of Advanced Nursing, 67, 169–175. doi:10.1111/j.1365-2648.2010.05482.x

Re, J., Dean, S., & Menahem, S. (2013). Infant cardiac surgery: Mothers tell their story: A therapeutic experience. World Journal for Pediatric & Congenital Heart Surgery, 4, 278– 285. doi:10.1177/2150135113481480

Rempel, G. R. (2005). Parenting a child with HLHS whose Treatment includes the Norwood surgical approach (Doctoral dissertation). Retrieved from ProQuest dissertations and theses database. (NR08720)

Rempel, G. R., & Harrison, M. J. (2007). Safeguarding precarious survival: Parenting children who have life-threatening heart disease. Qualitative Health Research, 17, 824–837. doi:10.1177/1049732307303164

Rempel, G. R., Rogers, L. G., Ravindran, V., & Magill-Evans, J. (2012). Facets of parenting a child with hypoplastic left heart syndrome. Nursing Research and Practice, 2012, Article 714178. doi:10.1155/2012/714178

Saldaña, J. (2009). The coding manual for qualitative researchers. Los Angeles: Sage.

Shearer, K., Rempel, G. R., Norris, C. M., & Magill-Evans, J. (2013). "It's no big deal": Adolescents with congenital heart disease. Journal of Pediatric Nursing, 28, 28–36. doi:10.1016/j.pedn.2012.03.031

Sikora, K., & Janusz, B. (2014). Maternal bond with cardiosurgically treated infant. Qualitative analysis of mothers' narratives. Developmental Period Medicine, 18, 439–446. Retrieved from http://www.medwiekurozwoj.pl/articles/ 2014-4-4.pdf

Sparacino, P. S. A., Tong, E. M., Messias, D. K. H., Foote, D., Chesla, C. A., & Gilliss, C. L. (1997). The dilemmas of par- ents of adolescents and young adults with congenital heart disease. Heart and Lung: Journal of Acute and Critical Care, 26, 187–195. doi:10.1016/S0147-9563(97)90055-8

Thorne, S. (2017). Metasynthetic madness: What kind of monster have we created? Qualitative Health Research, 27, 3–12. doi:10.1177/1049732316679370

Thorne, S., Jensen, L., Kearney, M. H., Noblit, G., & Sandelowski, M. (2004). Qualitative metasynthesis: Reflections on methodological orientation and ideologi- cal agenda. Qualitative Health Research, 14, 1342–1365. doi:10.1177/1049732304269888

Tong, E. M., Sparacino, P. S. A., Messias, D. K. H., Foote, D., Chesla, C. A., & Gilliss, C. L. (1998). Growing up with congenital heart disease: The dilemmas of adolescents and young adults. Cardiology in the Young, 8, 303–309. doi:10.1016/S01479563(97)90055-8

van der Bom, T., Zomer, A. C., Zwinderman, A. H., Meijboom, F. J., Bouma, B. J., & Mulder, B. J. M. (2011). The changing epidemiology of congenital heart disease. Nature Reviews Cardiology, 8, 50–60. doi:10.1038/nrcardio.2010.166

van der Linde, D., Konings, E. E. M., Slager, M. A., Witsenburg, M., Helbing, W. A., Takkenberg, J. J. M., & Roos-Hesselink, J. W. (2011). Birth prevalence of congenital heart disease worldwide. Journal of the American College of Cardiology, 58, 2241–2247. doi:10.1016/j. jacc.2011.08.025

Veldtman, G. R., Matley, S. L., Kendall, L., Quirk, J., Gibbs, J. L., Parsons, J. M., & Hewison, J. (2000). Illness understanding in children and adolescents with heart disease. Heart, 84, 395–397. doi:10.1136/heart.84.4.395

Wahlberg, A. (2018). The vitality of disease. In M. Meloni, J. Cromby, D. Fitzgerald, & S. Lloyd (Eds.), The Palgrave handbook of biology and society (pp. 727–748). London: Palgrave Macmillan.

Wei, H., Roscigno, C. I., Hanson, C. C., & Swanson, K. M. (2015). Families of children with congenital heart dis- ease: A literature review. Heart & Lung: The Journal of Acute and Critical Care, 44, 494–511. doi:10.1016/j. hrtlng.2015.08.005

Wind, G. (2009). Stiltiende fortællinger: Livet med kronisk sygdom i et antropologisk perspektiv [Tacit narratives: Life with chronic illness in an anthropological perspective] (Doctoral dissertation, University of Copenhagen, Denmark).