Survey Research with Families in the Context of Pediatric Chronic Health Conditions: Key Considerations and Future Directions

Aimee K. Hildenbrand, PhD[1,2,3], Marieke Van Schoors, PhD[4], and Melissa A. Alderfer, PhD[1,3]

[1] Center for Healthcare Delivery Science, Nemours Children's Health System

[2] Division of Behavioral Health, Nemours/ Alfred I. duPont Hospital for Children

[3] Department of Pediatrics, Sidney Kimmel Medical College at Thomas Jefferson University

[4] Department of Experimental Clinical and Health Psychology, Ghent University

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Author Biographical Paragraphs

Aimee K. Hildenbrand, PhD, is an assistant research scientist in the Nemours Center for Healthcare Delivery Science, pediatric psychologist in the Division of Behavioral Health at Nemours/Alfred I. duPont Hospital for Children, and Assistant Professor of Pediatrics at Sidney Kimmel Medical College of Thomas Jefferson University, USA. Her research program focuses on improving healthcare delivery to optimize pain management and psychosocial functioning for youth with chronic health conditions, with an emphasis on sickle cell disease and cancer. Dr. Hildenbrand also provides clinical care to children with sickle cell disease, cancer, and chronic pain and their families. Recent publications include:

Hildenbrand, A. K., & Alderfer, M. A. (2019). Survey and interview assessment approaches in research with couples and families. In B. H. Fiese, M. Celano, K. Deater-Deckard, E. Jouriles, & M. Whisman (Eds.), *APA Handbook of Contemporary Family Psychology: Foundations, methods, and contemporary issues across the lifespan* (pp. 257-279). American Psychological Association. https://doi.org/10.1037/0000099-015

Hildenbrand, A. K., Barakat, L. P., Alderfer, M. A., & Marsac, M. L. (2015). Coping and coping assistance among children with sickle cell disease and their parents. *Journal of Pediatric Hematology/ Oncology*, *37*(1), 25-34. https://dx.doi.org/10.1097%2FMPH.0000000000000092

Marieke Van Schoors, PhD, is a clinical psychologist and post-doctoral researcher at Ghent University in Belgium. She is trained in psycho-oncology and completed a PhD on the impact of childhood cancer on families. She is a board member of the European Pediatric Psychology Network and member of the Belgian Society of Paediatric Haematology and Oncology Young Investors network. Recent publications include:

Van Schoors, M., Caes, L., Goubert, L., Verhofstadt, L. L., & Alderfer, M. A. (2015). Family resilience after pediatric cancer diagnosis: A systematic review. *Journal of Pediatric Psychology, 40*(9), 856-868. https://doi.org/10.1093/jpepsy/jsv055

Van Schoors, M., Caes, L., Knoble, N. B, Goubert, L., Verhofstadt, L. L. & Alderfer, M.A. (2017). Systematic review: Associations between family functioning and child adjustment after pediatric cancer diagnosis: A meta-analysis. *Journal of Pediatric Psychology*, *42*(1), 6-18. https://doi.org/10.1093/jpepsy/jsw070

Van Schoors, M., De Paepe, A. L., Norga, K., Cosyns, V., Morren, H., Vercruysse ,T., Goubert, L., & Verhofstadt, L. L. (2019). Family members dealing with childhood cancer: A study on the role of family functioning and cancer appraisal. *Frontiers in Psychology*, *10*(1405), 1-14. https://doi.org/10.3389/fpsyg.2019.01405

Melissa A. Alderfer, PhD, is the Delaware Valley Center Director for the Nemours Center for Healthcare Delivery Science and a Principal Research Scientist at Nemours. She is also a Professor of Pediatrics at Sidney Kimmel Medical College of Thomas Jefferson University, USA. Dr. Alderfer is a licensed Clinical Psychologist with over 20 years of research experience. Her research focuses on the impact of childhood chronic medical conditions on families and

improving health care to better serve them. She combines qualitative and quantitative methods in her community-engaged work to conceptualize family adjustment and develop and test multi-level interventions and models of care. Recent publications include:

Christofferson, J., Stenman, K., Kazak, A. E., Pierce, J., Kelly, C., Schifano, E., Sciolla, J., Deatrick, J., & Alderfer, M. A. (2020). Family consequences of potentially traumatic pediatric medical events: Implications for trauma-informed care. *Journal of Family Psychology, 34*(2), 237-246. https://doi.org/10.1037/fam0000597

Tomatsu, S. C., Stanley, C., Conroy, R., & Alderfer, M. A. (2021). Method matters: Comparing cancer-related adjustment of siblings to norms and well-matched peers. *Journal of Pediatric Psychology, 46*(3), 324-331. https://doi.org/10.1093/jpepsy/jsaa110

Alderfer, M. A. (2017). Commentary: Family processes and outcomes: Research advances and future directions. *Journal of Pediatric Psychology, 42*(1), 125-129. https://doi.org/10.1093/jpepsy/jsw100

Abstract

Self-report family functioning measures play a critical role in advancing our understanding of how families are impacted by, and adapt to, the demands of childhood health conditions. In this paper, we present key considerations when conceptualizing, assessing, and analyzing dynamic family processes in research, discusses related implications for selecting instruments, and provides an update on the evidence base of self-report family functioning measures. Researchers need to consider theory, definitions of the family, informants, instruments, and procedural and data analytic issues when designing family research. Examples of questionnaires assessing general family functioning, dyadic relationships, and family functioning within the context of pediatric health conditions are provided. Additional evidence of validity, reliability, clinical utility, and cultural sensitivity of these measures is needed within pediatric chronic illness populations. Future research should include multiple family members and utilize varied assessment methods to obtain a comprehensive understanding of family functioning in the context of pediatric health conditions.

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Address all correspondence to: Aimee K. Hildenbrand, PhD, 1600 Rockland Road, RC-1, Suite 160, Wilmington, DE 19803, USA; Email: Aimee.Hildenbrand@nemours.org

Families play a fundamental role in the lives of all youth, including those with chronic health conditions. Pediatric researchers are frequently interested in evaluating how childhood chronic illness impacts the family, and simultaneously, how family processes can impede or facilitate adaptation to and management of pediatric chronic health conditions (Knafl et al., 2015). However, conceptualizing, assessing, and analyzing dynamic family processes presents many challenges (Alderfer et al., 2008; Holmbeck & Devine, 2011). Researchers must evaluate relevant theoretical models, operationalize the family system, and decide which domains of family functioning are important to assess (e.g., communication, cohesion, problem solving) at what levels of measurement (e.g., individual family members, dyads, family as a whole) and in what contexts (e.g., general vs. illness-related family processes). Whether quantitative, qualitative, or mixed methods approaches are selected to assess families, researchers must develop or select corresponding assessment tools, determine which family members should provide data, and formulate data analytic strategies that can accommodate complex, nested data.

Fortunately, for researchers aiming to apply quantitative methods to examine family processes, various surveys, observational tools, and clinician-rated interviews have demonstrated reliability and utility in pediatric samples. Alderfer et al. (2008) systematically reviewed the evidence base of family measures relevant to pediatric chronic health conditions and categorized 19 instruments as “well-established” and 10 as “approaching well-established” in the general population. The majority of these instruments assess perspectives of those inside the family system (i.e., self-report questionnaires). Self-report family assessment tools offer several advantages. Relative to other assessment methods (e.g., clinician-rated interviews, observational coding systems), self-report questionnaires are accessible, inexpensive, and efficient. Many self-report measures are also flexible, as they can be administered in various settings (e.g., research

lab, clinic or hospital, community space), over the phone, or online, which has become particularly useful in the context of the ongoing COVID-19 pandemic. As a result, self-report questionnaires are widely used in family research, which facilitates replication of prior research, systematic and meta-analytic reviews, comparisons across different pediatric chronic illness populations, and refinement of existing or development of new theoretical models of family functioning in the context of pediatric chronic health conditions.

In this paper, we discuss key considerations in designing and conducting research using self-report family functioning measures in the context of pediatric chronic health conditions. We provide a range of examples of such instruments, including updated information on the measures highlighted by Alderfer et al. (2008), and present promising new family assessment tools that have been developed over the past decade. Lastly, we provide recommendations and describe a research agenda to advance family assessment in pediatric chronic illness populations. While the use of validated family assessment tools is not restricted to research, a comprehensive review of the utility of these instruments in clinical practice is beyond the scope of this paper.

Theoretical Issues

General Systems Theory (Engel, 1980; von Bertalanffy, 1968) posits that systems are unified wholes comprised of interdependent components, their organization and relationships, and rules that shape their behavior and interactions (Stanton, 2009). Open systems, such as families, are constantly in flux; they achieve homeostasis through continuous exchange with the larger systems in which they are embedded (e.g., extended family, community). Accurately capturing family systems requires that all components, and the bidirectional relationships between them, are taken into account (Kazak et al., 2009). Open systems also demonstrate equifinality, as they can achieve the same final state from various starting points and through

divergent paths. Over time, systems transform from homogenous wholes to hierarchically organized, specialized subsystems with defined purposes and processes (Hildenbrand & Alderfer, 2019).

Based on principles of General Systems Theory, various models of family functioning have been proposed. Some of these models, including the Beavers Systems Model (Beavers & Hampson, 2003), the Circumplex Model of Marital and Family Systems (Olson & Gorall, 2003), the McMaster Model of Family Functioning (Ryan et al., 2005), and the Process Model of Family Functioning (Skinner et al., 2000), describe aspects of families considered fundamental to how all families function. These include family constructs, such as structure and organization (e.g., roles, rules, leadership, adaptability), relationship patterns (e.g., communication, conflict), and emotional environment (e.g., warmth, cohesion; Alderfer et al., 2008; Bray, 2013; Lebow & Stroud, 2012). For researchers interested in examining family adjustment and functioning in the context of pediatric chronic health conditions, the Process Model of Family Functioning may be especially informative, as it focuses on a family's ability to accomplish basic, developmental, and hazardous/crisis tasks through differentiation, assignment, and performance of specified roles, communication, affective expression, involvement with one another, flexibility and control, and a system of values and norms (Skinner et al., 2000).

Other models describe family processes specifically within the context of a stressor. These family stress and coping models (Hobfoll & Spielberger, 1992), including Hill's ABC-X model (Hill, 1949), which was later expanded on in the Double ABCX model (McCubbin & Patterson, 1983), and the Family Adjustment and Adaptation Response (FAAR) model (Patterson, 1988; Patterson & Garwick, 1994), posit that, when faced with a stressor (e.g., childhood illness), families cope by using individual family member strengths (e.g., self-esteem)

and family-level resources (e.g., flexibility, cohesion), changing perceptions of the stressor and their resources, or removing demands associated with the stressor, in order to adjust to day-to-day challenges and restore balance or equilibrium within the family. The Double ABCX and FAAR models have demonstrated utility in guiding studies of family adaptation and resilience across a wide range of childhood chronic health conditions (Patterson, 2005).

Given increasing recognition of the unique demands of chronic health conditions for families, frameworks such as the Family Systems Illness Model (Rolland, 1984, 1987, 2018) were developed to describe family adaptation to illness and disability. This resilience-based model proposes bidirectional interactions between characteristics of the family unit (e.g., organization, communication), illness attributes (e.g., onset, course, level of uncertainty, outcome), and illness time phases (e.g., initial crisis, chronic, terminal). The Family Systems Illness Model can inform research on how families' strengths and vulnerabilities interact with the psychosocial demands of chronic illness over time to shape family-level adaptation (Rolland & Walsh, 2006).

Family researchers should carefully consider which theoretical model they will apply when studying families of youth with chronic health conditions. Indeed, family functioning frameworks should guide all aspects of research conceptualization and implementation, including the development of research questions, decisions about study design and measure selection, and data analysis (Davey et al., 2014). In turn, research on family functioning among youth with chronic illness has the potential to advance and refine these theoretical frameworks.

Operationalizing the Family

Prior to studying family functioning among youth with chronic health conditions, researchers should stipulate how they will define the family system (Feetham, 2018). This task

can be more challenging than it appears at first glance, as families are increasingly diverse and complex. In the United States, for example, many youth are raised in households with single parents, same sex parents, grandparents, blended families including stepparents, stepchildren, and/or half-siblings, or foster families (Teachman et al., 2013; Widiss, 2016). Some may also consider extended relatives, fictive kin, and/or friends as part of their primary family unit. As such, the common practice of defining the family as individuals connected by blood, marriage, adoption, or living within the same household may be incomplete, or inappropriate, in some contexts. In addition, family composition changes over time and in response to life events. For instance, after the diagnosis of pediatric cancer, grandparents or other extended relatives may take a more prominent role in caregiving for healthy siblings (Van Schoors et al., 2018). In these circumstances, operationalizing the family based on identifying who meets important family functions (e.g., decision making, caregiving) can be particularly useful (Feetham, 2018).

Alternatively, families can be conceptualized as overlapping networks that encompass multiple households (Cherlin, 2010), though this approach may complicate assessment and data analysis. Specifically, researchers who apply this social network strategy will likely receive different lists of family members from individuals within the same household. Researchers might assess characteristics of these family networks (e.g., perceptions of intimacy and communication between individuals) and the extent of overlap between different household members' family networks, variables that can be used to provide additional context and nuance in subsequent analyses of individual- and family-level outcomes of interest. For additional discussion on this social network approach, see Amato (2014).

Selecting Informants and Survey Instruments

After considering relevant theoretical models and defining the family system, researchers must decide who to include in the measurement protocol. Given that perspectives on family functioning can differ across individual family members (Alderfer et al., 2009), it is recommended that researchers target multiple reporters within the family (Alderfer, 2017; Bray, 2013). However, obtaining information from every family member is seldom feasible, particularly for pediatric health researchers who may be recruiting participants in settings such as busy medical clinics. In addition, some argue that assessing dyadic processes (e.g., partner-partner, parent-child, sibling-sibling interactions and relationships) may provide more focused information to inform interventions that may still result in changes for the family as a whole (Bray, 2013). Researchers must consider whose perspectives are most important to gather to understand the dyadic or family construct of interest. For example, Coakley et al. (2002) noted that, when assessing parent-child conflict, low correlations between reporters can be expected. As such, they measured mothers', fathers', and children's perspectives on parent-child conflict and used each of these variables to examine family conflict over time in youth with spina bifida. If children are selected as informants, researchers must also consider the impact of potential cognitive deficits related to pediatric chronic health conditions. In populations where cognitive impairments are likely, researchers should carefully review an instrument to determine whether it is developmentally appropriate (e.g., reading level, complexity of response options, length) and whether modifications in administration are needed (e.g., reading questions aloud, allowing written, spoken, or physical responses such as pointing, using visual stimuli and response cards).

The next step is to design an assessment protocol. To assist researchers in selecting an appropriate tool, we compiled examples of various self-report instruments designed to assess general family functioning (see Table 1), dyadic family relationships (see Table 2), and family

functioning within the context of childhood health conditions (see Table 3). These measures were selected based on those included in the systematic review conducted by Alderfer et al. (2008) and review of relevant recent literature. We also included frequently employed measures identified in the International Family Nursing Association (IFNA) 2017 Family Measures Survey, a project conducted to identify family functioning measures used by nursing researchers across 20 countries. The instruments included in Tables 1 to 3 do not represent a comprehensive list of family assessment tools; rather, we aimed to provide targeted information (e.g., theoretical basis, constructs assessed, format, prior use in pediatric chronic illness populations) on well-established and newly developed (i.e., published between 2008 - 2020) self-report family measures that are relevant to pediatric researchers. We focused specifically on measures of systemic processes rather than instruments that assess individual family member functioning. Below, we turn to additional considerations for researchers in selecting a self-report family assessment tool.

[Insert Table 1 about here.]

[Insert Table 2 about here.]

[Insert Table 3 about here.]

Measures for the General Population vs. Families of Children with Chronic Health Conditions

When selecting a survey, researchers must decide whether to use a measure of family functioning developed for the general population of families or one designed specifically for use with families of children with chronic health conditions. Generic family functioning measures may be advantageous, as they can serve as a common metric thereby enhancing our ability to draw comparisons across different populations (e.g., families of children with chronic illness and

healthy comparisons) and contexts, and advance theoretical frameworks of family functioning that have broad applicability across pediatric chronic health conditions (Leeman et al., 2016). Conversely, it has also been argued that measures developed for use with pediatric chronic illness populations assess more salient aspects of family functioning that may be strongly linked to health outcomes (Long & Marsland, 2011). Specifically, these instruments can help to elucidate the impact of pediatric chronic health conditions on various domains of family life, including relationships, communication, roles, illness management activities, and daily activities (see Table 3). Moreover, some family processes that are described as abnormal or unhealthy, based on measures designed for the general population (e.g., very high cohesion and flexibility), may actually be protective when families are confronted with pediatric chronic illnesses (Alderfer et al., 2008).

Ultimately, the decision to administer a general family instrument, or one developed specifically for pediatric chronic health conditions, depends on the question to be answered. Some aspects of family life may differ in relation to a child's health condition (e.g., family communication, conflict management, problem solving in normative family contexts vs. in pediatric illness management). Family researchers might consider using both types of family measures to compare general and illness-related family processes, each of which can inform models of family functioning and clinical interventions. This approach may be especially relevant for multi-informant research given the limited availability of instruments specifically developed for use in pediatric chronic illness populations that assess family functioning from children's perspectives.

Psychometric Properties

A critical consideration when selecting a family assessment tool is its psychometric properties. As outlined by Holmbeck and Devine (2009), an instrument demonstrates high validity when the construct of interest, purpose of assessment, target population, and context are clearly defined and items were generated and revised using multiple strategies (e.g., expert input, focus groups with target population, review of theory and empirical research). Measures with sound validity are also robustly related to other assessments of the same construct, divergent from measures of unrelated concepts, concurrently related or predictive of criterion measures, and explain a greater proportion of variance in important outcomes relative to similar measures (Holmbeck & Devine, 2009). Empirically supported measures demonstrate high internal consistency, inter-rater reliability and agreement, and temporal stability when constructs are thought to remain constant. Alderfer et al. (2008) outlined specific standards for psychometrically-sound family measures, including "internal consistency (coefficient α) $\geq$ .70, test-retest reliability consistent with the purported stability of the construct, inter-rater reliability (α or ICC) $\geq$ .70 and/or inter-rater agreement (κ) $\geq$ .61, and at least two forms of evidence of concurrent/ predictive or convergent validity" (p. 1048). Readers are referred to Holmbeck and Devine (2009) for a checklist that can inform measure selection and Hamilton and Carr (2016) for additional details on the psychometric properties of many of the instruments included in Table 1. Additional information on the psychometric properties of the measures included in Tables 1 and 2 is included in Supplementary Tables S1 and S2.

In addition, whenever possible, researchers should select measures that have been validated in samples similar to the population under study. This is difficult in the context of pediatric chronic health conditions, as many family measures were developed, tested, and normed on typically developing youth and their families. As mentioned above, some family

interaction patterns may be adaptive and expected within the context of childhood chronic illness but mistakenly described as maladaptive when using general population norms (Alderfer et al., 2008). In addition, some instruments have only undergone empirical validation with predominantly White, English-speaking, well-educated, and middle to high income samples (Sanderson et al., 2009). Many family constructs vary in meaning and significance across cultural groups (e.g., communication, affective expressiveness, roles, responsibilities); as a result, factor structures and cutoff scores likely differ across cultures (Lebow & Stroud, 2012). Moreover, some family constructs may only be salient in certain populations (e.g., familism). Indeed, measures developed with families of majority ethnic/racial backgrounds or traditional structures may contain embedded cultural biases that threaten the validity of data obtained and conclusions drawn when those measures are applied to diverse, nontraditional families (Linville et al., 2014). Examples of instruments validated in culturally diverse samples include the Systemic Clinical Outcome and Routine Evaluation (SCORE; Carr & Stratton, 2017), Relationship Assessment Scale (Hendrick, 1988), the Family Problem-Solving Communication Index (McCubbin et al., 1996), and the PROMIS Pediatric and Parent Proxy Family Relationships scales (Bevans et al., 2017).

While many family functioning measures have been translated into additional languages (see Supplementary Tables S1 and S2), researchers who aim to use translated versions of these instruments should consider the process through which they were adapted and validated. For instance, many translations of the McMaster Family Assessment Device (FAD) are available, but these translated versions demonstrate varying validity, reliability, and factor structures (e.g., Barroilhet et al., 2009; Juliusdottir & Olafsdottir, 2015; Speranza et al., 2012; Tsamparli et al., 2018). Whenever possible, researchers should seek to employ adapted instruments that have

demonstrated linguistic, construct, and measurement equivalence to the original versions. For more information about cross-cultural adaptation of questionnaires, see Byrne (2016) and Epstein et al. (2015).

Sensitivity to Change

Family researchers are often interested in examining change in family processes during and after intervention, in conjunction with changes in health status, or across developmental periods. Unfortunately, few family assessments have demonstrated responsiveness to change (Hamilton & Carr, 2016). One exception is the SCORE (Carr & Stratton, 2017), though its sensitivity to change has not yet been examined for families of youth with chronic health conditions. Measures that are more likely to capture change are those that contain easily understood, non-redundant items and response options, assess a wide range of levels in the latent construct (i.e., not prone to ceiling or floor effects), are culturally sensitive for the target population, and are flexible across diverse contexts (e.g., surveys that can be re-administered as youth and families progress through developmental stages; Fok & Henry, 2015). Families of youth with chronic illness, particularly those marked by a progressive course, unpredictability, and/or demanding treatment regimens, must continually reorganize family roles and responsibilities to adapt to ongoing health-related stressors (Alderfer et al., 2008; Rolland & Walsh, 2006; Van Schoors et al., 2018). These stressors are in addition to the typical challenges that many families face (e.g., births, separation/ divorce, relocation, employment changes). As such, there is an urgent need for instruments that can detect fluctuations in family processes in the context of pediatric chronic health conditions.

Implementing Self-report Family Functioning Measures

Using self-report family functioning measures in the context of pediatric chronic health conditions can present unique challenges. Data are frequently collected in healthcare settings, which have high potential for distractions and interruptions (e.g., during clinic visits). In addition, negative affect related to the hospital environment, upcoming procedures, or concurrent symptoms (e.g., pain, nausea, fatigue) may influence ratings of family functioning. Holmbeck and Devine (2011) proposed that home-based data collection is convenient and acceptable to families of youth with chronic illness and can improve enrollment of multiple family members. Alternatively, researchers might consider administering measures online when budgetary or logistical constraints preclude home visits (e.g., organization serves a very large catchment area).

As all self-reported methods have potential for response bias, researchers must take special precautions when administering these instruments with families (Linville et al., 2014). For example, family members completing measures concurrently may interact in ways that shape their responses (e.g., viewing others' responses, discussing questions). If multiple individuals within a family will complete surveys without research team oversight (e.g., at home, online), researchers should attempt to minimize response bias by providing clear instructions about whether family members should answer questionnaires together or independently. In the event that questionnaires are likely to cause distress or conflict between family members, researchers are ethically obligated to ensure participant safety and provide resources as needed. Pilot testing family assessments is recommended to determine how to prevent or mitigate respondent distress as well as threats to the reliability and validity of data obtained.

After collecting data and prior to conducting primary analyses, researchers should examine how the selected family functioning instrument performed in their sample. At the very least, internal consistency within the sample under investigation should be calculated and

reported (Alderfer et al., 2008). For repeated measurement of constructs thought to be relatively stable, test-retest reliability should also be evaluated. When a survey is administered to multiple family members, researchers should also evaluate the extent of non-independence between scores (e.g., Pearson correlation, intraclass correlation, Kappa) to ensure that these relationships are accounted for in primary analyses (Kenny et al., 2006). In addition, measurement invariance (i.e., stability of associations between survey items and latent factors) should be examined, as relational concepts can differ in salience and meaning across family members (e.g., mothers vs. fathers) and within the same individual over time (Busby & Poulsen, 2014). When the factor structure of a tool varies over time or across family members, traditional statistical methods that assume measurement invariance (e.g., growth curve analysis) may be inappropriate (Busby & Poulsen, 2014). Alderfer et al. (2008) noted that such information is rarely reported. Examining papers published since then suggests that, with the exception of internal consistency, this continues to be a limitation of the evidence base.

Analyzing Data from Self-report Family Functioning Measures

Generally speaking, theoretical models of family functioning have advanced at a faster pace than data analytic methods needed to test these models (Ram et al., 2014). Many traditional statistical approaches (e.g., analysis of variance, multiple regression) assume that data obtained from one individual are unrelated to that of other individuals in the sample (Kenny et al., 2006). When the non-independence of linked or nested observations is not accounted for in the statistical analysis, estimates are likely to be biased (Kenny et al., 2006). As a result, some researchers aggregate the responses of multiple family members to create summary or mean scores (Sayer & Klute, 2005). However, this approach may disguise meaningful differences in perspectives within the family system. Alternatively, researchers sometimes conduct parallel

analyses on subgroups (e.g., mothers vs. fathers) and compare findings, though this strategy likely oversimplifies the complex, interactional processes inherent in dyadic relationships and family groups (Fuligni, 2014; Lebow & Stroud, 2012).

Newer statistical approaches have facilitated more sophisticated analysis of systemic observations. For example, hierarchical linear modeling (HLM), a complex variation of ordinary least squares regression, allows researchers to nest data from individuals within hierarchical levels (e.g., families, clinics, regions) in order to examine variance in outcomes between and within groups and over time (Davey et al., 2014; Kenny et al., 2006). HLM approaches were developed across various fields simultaneously, and thus this method is referred to by many names (e.g., multilevel, mixed level, growth mixture, mixed linear, mixed effects, random effects, and random-coefficient modeling; Woltman et al., 2012).

Similar to HLM, structural equation modeling (SEM) enables researchers to model associations between different individuals, couples, families, and/or higher units (Busby & Poulsen, 2014). SEM tests theoretical relationships between a series of observed and latent independent and dependent variables (Schumacker & Lomax, 2010). Although HLM and SEM yield similar parameter estimates in measurement and factor analytic models, SEM has unique benefits. For example, SEM offers greater flexibility in model specification and constraints, more information that can be used to test and refine theoretical models, and many extensions appropriate for a wide range of systemic research questions (Wendorf, 2002). In addition, some common dyadic analysis models (e.g., Actor-Partner Interdependence Model; Cook & Kenny, 2005) may be more easily conducted using SEM (Kenny et al., 2006). SEM can also examine curvilinear relationships, which is advantageous for situations in which moderate levels of a family construct (e.g., cohesion) are considered optimal. For further information on extensions

and applications of HLM and SEM in dyadic and family research, see McHale et al. (2014), Keiley, Dankowski, et al. (2005), Keiley, Martin, et al. (2005), and Kenny et al. (2006).

Future Directions for Assessing Family Processes in Pediatric Chronic Health Conditions

Self-report family functioning measures play a critical role in advancing our understanding of how families are impacted by and adapt to the demands of childhood chronic health conditions. As highlighted in Tables 1 and 2, many family and dyadic measures developed in the general population have been used with samples of families facing pediatric chronic health conditions. However, rarely have these questionnaires undergone comprehensive psychometric evaluation in pediatric chronic illness populations. This is problematic considering the family-wide changes that can occur in response to unique challenges associated with pediatric chronic health conditions. For example, a study conducted by Marsac and Alderfer (2011) raised concerns regarding the construct validity of some subscales of the Family Adaptability and Cohesion Scale IV (FACES-IV) among families of youth with cancer. Additional investigation of the psychometric properties of general family functioning measures in pediatric chronic illness populations will fill a significant gap in the field and enable researchers to select the most valid and reliable instruments.

Of particular concern for those interested in family research is the clinical utility of questionnaires (IFNA, 2017). Unfortunately, given lack of information regarding the predictive validity and sensitivity to change of general family functioning measures within pediatric chronic illness populations, it is challenging to determine which of these instruments may be best for informing clinical care (see Supplementary Tables S1 and S2 for information on measures that have demonstrated predictive validity and/or sensitivity to treatment effects in other populations). One exception is the Family Impact of Childhood Disability (FICD; Trute et al.,

2007), which has demonstrated predictive validity in mothers of children with a broad array of disabilities including complex health conditions (Benzies et al., 2010). Additional longitudinal work is needed to examine how family functioning instruments perform in predicting important outcomes over time and detecting meaningful changes that occur as individuals and families move through stages of illness, treatment, and development. Relatedly, in order to appropriately allocate psychosocial resources to those most in need, improved norms on general family and dyadic functioning measures are needed for families coping with pediatric chronic health conditions. Continued development and refinement of efficient tools to assess family functioning are also warranted, as many existing measures are lengthy and may not be feasible to integrate into fast-paced healthcare settings.

Across both generic family functioning measures and those developed for pediatric chronic illness populations, additional work to validate these instruments in culturally diverse families is a high priority for the field. Many generic family and dyadic functioning tools were developed using samples of primarily White, English-speaking, two-parent, and middle to high income families (Hamilton & Carr, 2016; Lebow & Stroud, 2012; Sanderson et al., 2009). This is a significant concern given the increasing variation in family structures and the high proportion of ethnically diverse youth with chronic illness (Mitchell et al., 2011). Aspects of family life may vary across cultures, and applying family functioning surveys developed with one cultural group to other populations may increase the risk of biased results and erroneous conclusions. Future work could address this limitation by validating these family instruments in culturally diverse samples, which will likely require engagement of key stakeholder partners, multisite collaboration, and use of coordinated, multipronged sampling, recruitment, data collection, and retention strategies to ensure adequate sample diversity and size. For more comprehensive

reviews of specific strategies to reach underserved populations in research, see Bonevski et al. (2014) and Yancey et al. (2006).

Future research on family functioning in pediatric chronic illness populations should also move beyond single-informant protocols by assessing perspectives of multiple members within the family system. Similar to the trend noted by Alderfer et al. (2008) over a decade ago, the predominance of what we currently know about family functioning in the context of pediatric chronic illness comes from mothers and patients. Assessing other members of the family, including fathers, other caregivers, and siblings, will contribute to a more complete understanding of family functioning. Including multiple family members in assessment protocols also enables researchers to examine the proportion of variance in outcomes explained by differences within and between families, which may have important implications for intervention development. Because survey items, response options, and the broader constructs they assess can vary in their salience and meaning across different respondents within the same family, additional research establishing measure equivalence for existing family assessment tools is critical to facilitate research incorporating multiple family members. In addition, further development of measures designed to assess the perspectives of children in families facing pediatric chronic health conditions is needed, as most family measures developed specifically for these populations rely on parent report.

Finally, it is important to note that self-report methods will always involve some disadvantages (e.g., recall, social desirability bias). To capture the true complexity of family systems, application of diverse methodologies is needed (Davey et al., 2014; Stanton, 2009). While this review focused on self-report family functioning questionnaires, observational tools and interviewer-rated assessments may also provide informative outsider perspectives on family

interaction patterns that may not be readily apparent to those inside the family system. Moreover, qualitative methods such as interviews and focus groups may offer opportunities to obtain deeper insight on bidirectional, complex associations between health and family systems processes. As such, mixed methods research may be particularly valuable to advancing the scientific study of families. In turn, a more thorough understanding of how families function in the context of pediatric chronic health conditions can improve the design of rigorous family-based research, provision of impactful family-centered care, and advocacy efforts for policies to better address families' unmet needs when coping with pediatric chronic health conditions.

Conflicts of Interest

The authors declare that there are no conflicts of interest.

References

Alderfer, M. A. (2017). Commentary: Family processes and outcomes: Research advances and future directions. *Journal of Pediatric Psychology*, *42*(1), 125-129. https://doi.org/10.1093/jpepsy/jsw100

Alderfer, M. A., Fiese, B. H., Gold, J. I., Cutuli, J. J., Holmbeck, G. N., Goldbeck, L., Chambers, C. T., Abad, M., Spetter, D., & Patterson, J. (2008). Evidence-based assessment in pediatric psychology: Family measures. *Journal of Pediatric Psychology*, *33*(9), 1046-1061. https://doi.org/10.1093/jpepsy/jsm083

Alderfer, M. A., Navsaria, N., & Kazak, A. E. (2009). Family functioning and posttraumatic stress disorder in adolescent survivors of childhood cancer. *Journal of Family Psychology*, *23*(5), 717-725. https://doi.org/10.1037/a0015996

Amato, P. R. (2014). Approaches to measuring families. In S. M. McHale, P. R. Amato, & A. Booth (Eds.), *Emerging methods in family research* (pp. 179-193). Springer International Publishing. https://doi.org/10.1007/978-3-319-01562-0_11

Barroilhet, S., Cano-Prous, A., Cervera-Enguix, S., Forjaz, M. J., & Guillén-Grima, F. (2009). A Spanish version of the Family Assessment Device. *Social Psychiatry and Psychiatric Epidemiology*, *44*(12), 1051-1065. https://doi.org/10.1007/s00127-009-0022-8

Beavers, W. R., & Hampson, R. B. (2003). Measuring family competence: The Beavers systems model. In F. Walsh (Ed.), *Normal family processes: Growing diversity and complexity* (3rd ed., pp. 549-580). Guilford Press. https://doi.org/10.4324/9780203428436_chapter_20

Benzies, K. M., Trute, B., Worthington, C., Reddon, J., Keown, L. A., & Moore, M. (2010). Assessing psychological well-being in mothers of children with disability: Evaluation of the Parenting Morale Index and Family Impact of Childhood Disability Scale. *Journal of Pediatric Psychology*, *36*(5), 506-516. https://doi.org/10.1093/jpepsy/jsq081

Bevans, K. B., Riley, A. W., Landgraf, J. M., Carle, A. C., Teneralli, R. E., Fiese, B. H., Meltzer, L. J., Ettinger, A. K., Becker, B. D., & Forrest, C. B. (2017). Children's family experiences: Development of the PROMIS pediatric family relationships measures. *Quality of Life Research*, *26*(11), 3011-3023. https://doi.org/10.1007/s11136-017-1629-y

Bonevski, B., Randell, M., Paul, C., Chapman, K., Twyman, L., Bryant, J., Brozek, I., & Hughes, C. (2014). Reaching the hard-to-reach: A systematic review of strategies for improving health and medical research with socially disadvantaged groups. *BMC Medical Research Methodology*, *14*, Article 42. https://doi.org/10.1186/1471-2288-14-42

Bray, J. H. (2013). Couple and family assessment. In J. H. Bray & M. Stanton (Eds.), *The Wiley-Blackwell handbook of family psychology* (pp. 151-163). Wiley-Blackwell.

Busby, D. M., & Poulsen, F. O. (2014). Measurement issues with couple- and family-level data. In R. B. Miller & L. N. Johnson (Eds.), *Advanced methods in family therapy research: A focus on validity and change* (pp. 44-58). Routledge. https://doi.org/10.4324/9780203084526

Byrne, B. M. (2016). Adaptation of assessment scales in cross-national research: Issues, guidelines, and caveats. *International Perspectives in Psychology: Research, Practice, Consultation*, *5*(1), 51-65. https://doi.org/10.1037/ipp0000042

Carr, A., & Stratton, P. (2017). The SCORE family assessment questionnaire: A decade of progress. *Family Process*, *56*(2), 285-301. https://doi.org/10.1111/famp.12280

Cherlin, A. (2010). Demographic trends in the United States: A review of research in the 2000s. *Journal of Marriage and Family*, *72*(3), 403-419. https://doi.org/10.1111/j.1741-3737.2010.00710.x

Coakley, R. M., Holmbeck, G. N., Friedman, D., Greenley, R. N., & Thill, A. W. (2002). A longitudinal study of pubertal timing, parent-child conflict, and cohesion in families of young adolescents with spina bifida. *Journal of Pediatric Psychology*, *27*(5), 461-473. https://doi.org/10.1093/jpepsy/27.5.461

Cook, W. L., & Kenny, D. A. (2005). The Actor–Partner Interdependence Model: A model of bidirectional effects in developmental studies. *International Journal of Behavioral Development*, *29*(2), 101-109. https://doi.org/10.1080/01650250444000405

Davey, M., Zeytinoglu, S., & Lynch, L. (2014). Integrating theory and research. In R. B. Miller & L. N. Johnson (Eds.), *Advanced methods in family therapy research: A focus on validity and change* (pp. 28-43). Routledge. https://doi.org/10.4324/9780203084526

Engel, G. L. (1980). The clinical application of the biopsychosocial model. *The American Journal of Psychiatry*, *137*(5), 535-544. https://doi.org/10.1176/ajp.137.5.535

Epstein, J., Santo, R. M., & Guillemin, F. (2015). A review of guidelines for cross-cultural adaptation of questionnaires could not bring out a consensus. *Journal of Clinical Epidemiology*, *68*(4), 435-441. https://doi.org/10.1016/j.jclinepi.2014.11.021

Feetham, S. (2018). Revisiting Feetham's criteria for research of families to advance science and inform policy for the health and well-being of families [Guest Editorial]. *Journal of Family Nursing*, *24*(2), 115-127. https://doi.org/10.1177/1074840718767831

Fok, C. C., & Henry, D. (2015). Increasing the sensitivity of measures to change. *Prevention Science*, *16*(7), 978-986. https://doi.org/10.1007/s11121-015-0545-z

Fuligni, A. J. (2014). The benefits and challenges of modeling intra-family variability. In S. M. McHale, P. R. Amato, & A. Booth (Eds.), *Emerging methods in family research* (pp. 57-69). Springer International Publishing. https://doi.org/10.1007/978-3-319-01562-0_4

Hamilton, E., & Carr, A. (2016). Systematic review of self-report family assessment measures. *Family Process*, *55*(1), 16-30. https://doi.org/10.1111/famp.12200

Hendrick, S. S. (1988). A generic measure of relationship satisfaction. *Journal of Marriage and the Family*, *50*(1), 93-98. https://doi.org/10.2307/352430

Hildenbrand, A. K., & Alderfer, M. A. (2019). Survey and interview assessment approaches in research with couples and families. In B. H. Fiese, M. Celano, K. Deater-Deckard, E.

Jouriles, & M. Whisman (Eds.), *APA Handbook of Contemporary Family Psychology*. American Psychological Association. https://doi.org/10.1037/0000099-015

Hill, R. (1949). *Families under stress: Adjustment to the crisis of war, separation, and reunion*. Harper and Row.

Hobfoll, S. E., & Spielberger, C. D. (1992). Family stress: Integrating theory and measurement. *Journal of Family Psychology*, *6*(2), 99-112. https://doi.org/10.1037/0893-3200.6.2.99

Holmbeck, G. N., & Devine, K. A. (2009). An author's checklist for measure development and validation manuscripts [Editorial]. *Journal of Pediatric Psychology*, *34*(7), 691-696. https://doi.org/10.1093/jpepsy/jsp046

Holmbeck, G. N., & Devine, K. A. (2011). Commentary: Family assessment in pediatric psychology. *Journal of Pediatric Psychology*, *36*(5), 642-646. https://doi.org/10.1093/jpepsy/jsr004

International Family Nursing Association (IFNA). (2017). IFNA Position Statement on Advanced Practice Competencies for Family Nursing. https://internationalfamilynursing.org/2017/05/19/advanced-practice-competencies/

Juliusdottir, G. M., & Olafsdottir, H. (2015). An Icelandic version of McMasters Family Assessment Device (FAD). *Research on Social Work Practice*, *25*(7), 815-827. https://doi.org/10.1177/1049731514538673

Kazak, A., Rourke, M. T., & Navsaria, N. (2009). Families and other systems in pediatric psychology. In M. C. Roberts & R. G. Steele (Eds.), *Handbook of pediatric psychology* (4th ed., pp. 656-671). Guilford Press.

Keiley, M. K., Dankoski, M., Dolbin-MacNab, M., & Liu, T. (2005). Covariance structure analysis: From path analysis to structural equation modeling. In D. H. Sprenkle & F. P. Piercy (Eds.), *Research methods in family therapy* (2nd ed., pp. 432-460). Guilford Press.

Keiley, M. K., Martin, N. C., Liu, T., & Dolbin-MacNab, M. (2005). Multilevel growth modeling in the context of family research. In D. H. Sprenkle & F. P. Piercy (Eds.), *Research methods in family therapy* (2nd ed., pp. 405-431). Guilford Press.

Kenny, D. A., Kashy, D. A., & Cook, W. L. (2006). *Dyadic data analysis*. Guilford Press.

Knafl, K., Leeman, J., Havill, N., Crandell, J., & Sandelowski, M. (2015). Delimiting family in syntheses of research on childhood chronic conditions and family life. *Family Process*, *54*(1), 173-184. https://doi.org/10.1111/famp.12101

Lebow, J., & Stroud, C. B. (2012). Assessment of effective couple and family functioning: Prevailing models and instruments. In F. Walsh (Ed.), *Normal family processes: Growing diversity & complexity* (4th ed., pp. 501-528). Guilford Press.

Leeman, J., Crandell, J. L., Lee, A., Bai, J., Sandelowski, M., & Knafl, K. (2016). Family functioning and the well-being of children with chronic conditions: A meta-analysis. *Research in Nursing & Health*, *39*(4), 229-243. https://doi.org/10.1002/nur.21725

Linville, D., Todahl, J. L., & O'Neil, M. E. (2014). Using questionnaires in clinical couple and family research. In R. B. Miller & L. N. Johnson (Eds.), *Advanced methods in family therapy research: A focus on validity and change* (pp. 94-111). Routledge. https://doi.org/10.4324/9780203084526

Long, K. A., & Marsland, A. L. (2011). Family adjustment to childhood cancer: A systematic review. *Clinical Child and Family Psychology Review*, *14*(1), 57-88. https://doi.org/10.1007/s10567-010-0082-z

Marsac, M. L., & Alderfer, M. A. (2011). Psychometric properties of the FACES-IV in a pediatric oncology population. *Journal of Pediatric Psychology*, *36*(5), 528-538. https://doi.org/10.1093/jpepsy/jsq003

McCubbin, H. I., & Patterson, J. M. (1983). The family stress process: The Double ABCX Model of adjustment and adaptation. *Marriage and Family Review*, *6*(1), 7–37. https://doi.org/10.1300/J002v06n01_02

McCubbin, M. A., McCubbin, H. I., & Thompson, A. I. (1996). Family Problem Solving Communication (FPSC). In H. I. McCubbin, A. I. Thompson, & M. A. McCubbin (Eds.), *Family assessment: Resiliency, coping and adaptation - Inventories for research and practice* (pp. 639-686). University of Wisconsin.

McHale, S. M., Amato, P. R., & Booth, A. (Eds.). (2014). *Emerging methods in family research*. Springer International Publishing. https://doi.org/10.1007/978-3-319-01562-0

Mitchell, M. J., Patterson, C. A., & Boyd-Franklin, N. (2011). Commentary: Increasing cultural diversity in pediatric psychology family assessment research. *Journal of Pediatric Psychology*, *36*(5), 634-641. https://doi.org/10.1093/jpepsy/jsr019

Olson, D. H., & Gorall, D. M. (2003). Circumplex model of marital and family systems. In F. Walsh (Ed.), *Normal family processes: Growing diversity & complexity* (3rd ed., pp. 514-548). Guilford Press. https://doi.org/10.4324/9780203428436_chapter_19

Patterson, J. M. (1988). Families experiencing stress: I. The Family Adjustment and Adaptation Response Model: II. Applying the FAAR Model to health-related issues for intervention and research. *Family Systems Medicine*, *6*(2), 202–237. https://doi.org/10.1037/h0089739

Patterson, J. M. (2005). Weaving gold out of straw: Meaning-making in families who have children with chronic illnesses. In W. M. Pinsof & J. L. Lebow (Eds.), *Family psychology: The art of the science* (pp. 521–548). Oxford University Press.

Patterson, J. M., & Garwick, A. W. (1994). Levels of meaning in family stress theory. *Family Process*, *33*, 287–304. https://doi.org/10.1111/j.1545-5300.1994.00287.x

Ram, N., Shiyko, M., Lunkenheimer, E. S., Doerksen, S., & Conroy, D. (2014). Families as coordinated symbiotic systems: Making use of nonlinear dynamic models. In S. M. McHale,

P. R. Amato, & A. Booth (Eds.), *Emerging methods in family research* (pp. 19-37). Springer International Publishing. https://doi.org/10.1007/978-3-319-01562-0_2

Rolland, J. S. (1984). Toward a psychosocial typology of chronic and life-threatening illness. *Family Systems Medicine, 2*, 245–262. https://doi.org/10.1037/h0091663

Rolland, J. S. (1987). Chronic illness and the life cycle: A conceptual framework. *Family Process, 26*, 203–221. https://doi.org/10.1111/j.1545-5300.1987.00203.x

Rolland, J. S. (2018). *Helping couples and families navigate illness and disability: An integrated approach.* Guilford Press.

Rolland, J. S., & Walsh, F. (2006). Facilitating family resilience with childhood illness and disability. *Current Opinions in Pediatrics*, *18*(5), 527-538. https://doi.org/10.1097/01.mop.0000245354.83454.68

Ryan, C. E., Epstein, N. B., Keitner, G. I., Miller, I. W., & Bishop, D. S. (2005). *Evaluation and treating families: The Mcmaster approach*. Routledge.

Sanderson, J., Kosutic, I., Garcia, M., Melendez, T., Donoghue, J., Perumbilly, S., Franzen, C., & Anderson, S. (2009). The measurement of outcome variables in couple and family therapy research. *The American Journal of Family Therapy*, *37*(3), 239-257. https://doi.org/10.1080/01926180802405935

Sayer, A. G., & Klute, M. M. (2005). Analyzing couples and families: Multilevel methods. In V. L. Bengtson, A. C. Acock, K. R. Allen, P. Dilworth-Anderson, & D. M. Klein (Eds.), *Sourcebook of family theory and research* (pp. 289-314). SAGE Publications. http://dx.doi.org/10.4135/9781412990172.n12

Schumacker, R. E., & Lomax, R. G. (2010). *A beginner's guide to structural equation modeling* (3rd ed.). Routledge.

Skinner, H., Steinhauer, P., & Sitarenios, G. (2000). Family Assessment Measure (FAM) and Process Model of Family Functioning. *Journal of Family Therapy*, *22*, 190-210. https://doi.org/10.1111/1467-6427.00146

Speranza, M., Guénolé, F., Revah-Levy, A., Egler, P. J., Negadi, F., Falissard, B., & Baleyte, J. M. (2012). The French version of the Family Assessment Device. *Canadian Journal of Psychiatry*, *57*(9), 570-577. https://doi.org/10.1177/070674371205700908

Stanton, M. (2009). The systemic epistemology of family psychology. In J. H. Bray & M. Stanton (Eds.), *Handbook of family psychology* (pp. 5-20). Wiley-Blackwell. https://doi.org/10.1002/9781444310238.ch1

Teachman, J., Tedrow, L., & Kim, G. (2013). The demography of families. In G. W. Peterson & K. R. Bush (Eds.), *Handbook of marriage and the family* (3rd ed., pp. 39-63). Springer. https://doi.org/10.1007/978-1-4614-3987-5_3

Trute, B., Hiebert-Murphy, D., & Levine, K. (2007). Parental appraisal of the family impact of childhood developmental disability: Times of sadness and times of joy. *Journal of Intellectual & Developmental Disability*, *32*(1), 1-9. https://doi.org/10.1080/13668250601146753

Tsamparli, A., Petmeza, I., McCarthy, G., & Adamis, D. (2018). The Greek version of the McMaster Family Assessment Device. *PsyCh Journal*, *7*(3), 122-132. https://doi.org/10.1002/pchj.218

Van Schoors, M., De Mol, J., Morren, H., Verhofstadt, L. L., Goubert, L., & Van Parys, H. (2018). Parents' perspectives of changes within the family functioning after a pediatric cancer diagnosis: A multi family member interview analysis. *Qualitative Health Research*, *28*(8), 1229-1241. https://doi.org/10.1177/1049732317753587

von Bertalanffy, L. (1968). *General systems theory*. Braziller.

Wendorf, C. A. (2002). Comparisons of structural equation modeling and hierarchical linear modeling approaches to couples' data. *Structural Equation Modeling*, *9*(1), 126-140. https://doi.org/10.1207/S15328007SEM0901_7

Widiss, D. A. (2016). Legal recognition of same-sex relationships: New possibilities for research on the role of marriage law in household labor allocation. *Journal of Family Theory and Review*, *8*(1), 10-29. https://doi.org/10.1111/jftr.12123

Woltman, H., Feldstain, A., MacKay, J. C., & Rocchi, M. (2012). An introuction to hierarchical linear modeling. *Tutorials in Quantitative Methods for Psychology*, *8*(1), 52-69. https://doi.org/10.20982/tqmp.08.1.p052

Yancey, A. K., Ortega, A. N., & Kumanyika, S. K. (2006). Effective recruitment and retention of minority research participants. *Annual Review of Public Health*, *27*(1), 1-28. https://doi.org/10.1146/annurev.publhealth.27.021405.102113