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Parent routines for managing cystic fibrosis in children

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Abstract

Management of cystic fibrosis (CF) is burdensome and adherence is often suboptimal. Family routines are associated with adherence and health outcomes in other disease populations. Few studies have examined routines in CF. The study's aim was to describe parent experiences developing and utilizing CF care routines. Semi-structured interviews with a convenience sample of 25 parents of children under 13 years of age with CF were analyzed using phenomenological analysis. Three domains emerged: parent experiences developing a routine, support systems facilitating maintenance of routines, and challenges with maintaining care routines. Parents found routines difficult to establish, used trial and error, encountered barriers, and found support helpful to manage care demands. Some parents chose to deviate from their routine. Providing anticipatory guidance to promote the use of care routines and strategies to manage potential challenges may facilitate use of routines and improve CF management.

Keywords

routines; cystic fibrosis; treatment adherence; parents

Optimal management of cystic fibrosis (CF) is complex and time-consuming. The daily regimen for CF typically begins in the morning and ends at bedtime and may require an investment of over an hour a day on the part of both parent and child. Components of the treatment regimen often include taking oral and nebulized medications, doing chest physiotherapy, engaging in regular exercise, and eating and drinking to reach increased fat and calorie recommendations. It may also include taking supplemental enzymes, insulin, receiving parenteral nutrition, and completing sinus care. Chest physiotherapy, or airway clearance treatments, are typically prescribed twice daily for 15-30 minutes each, depending

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Informed Consent: All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2000. Informed consent was obtained from all patients for being included in the study.

on the method used, and may be increased to four times daily if the child shows signs of acute illness. These treatments loosen mucous from the lungs, and with the aid of special “huff coughs,” move it to the upper airways from which it can be expelled. Failure to do airway clearance contributes to plugged airways with loss of pulmonary function. Depending on the child's age, motor skills and preferences, the treatment may be accomplished through chest percussions (in which parents physically clap the child's back and sides in a particular pattern), or by wearing a vest that mechanically vibrates the child, or handheld devices which send vibrations into the lungs when a child breathes into them. Regardless of the method, parental presence is usually required for children, whether to actually perform the treatment on the child's body, to prepare the equipment and clean it, or to supervise older children completing these tasks independently.

Knowledge of the recommended care practices for a pediatric chronic illness is an important initial step to optimizing health. After education about the recommended treatment regimen, the family's ability to organize and complete daily tasks impacts quality of life for children and caregivers as well as adherence to the recommended care regimen. For example, a study in childhood asthma found quality of life to be related to the degree of burden associated with the complexity and time needed to manage the asthma routine. Parents reported that management was burdensome when asthma care was more complex and were more likely to have trouble reminding the child to take medications (Fiese & Wamboldt, 2000). While these relationships have been examined in asthma specifically, it is reasonable to hypothesize that similar processes and experiences may be present in CF given the illness is managed within the family context, and the relative increased complexity and burden of the care recommended for CF.

Reported rates of adherence to CF recommendations indicate a great need to understand and address barriers to adherence. In a study of children ages 6-13 years who have CF, adherence rates for airway clearance ranged from 51-74% of the recommended treatment frequency (Modi et al., 2006). Adherence rates for completing recommended aerosolized medications ranged from 48-82% and taking enzymes was between 27-90% (Modi, et al., 2006). The ranges reported by Modi and colleagues represent data collected by multiple methods, including self-report, Daily Phone Diary, and objective measures (electronic monitoring and pharmacy refills) and highlight the limitations inherent in studies which utilize a single means of measuring adherence. Single-measure studies typically fall within these ranges. For example, the study by Zindani and colleagues reports adherence rates obtained from electronic monitoring for vitamin intake of 63% and 66% for nebulized dornase alfa (Zindani, Streetman, Streetman, & Nasr, 2006). Moreover, only 12% to 16% of children are estimated to be achieving calorie intake recommendations (Mackner, McGrath, & Stark, 2001). Parents have reported several barriers to treatment adherence in CF, including forgetting, child oppositional behaviors, and time management. Children with CF have reported oral barriers such as medication taste and swallowing difficulties. Trends were noted in the relationship between barriers and adherence, including negative correlations between the number of treatments and adherence, and the number of parent-reported barriers and adherence to inhaled medications (Modi & Quittner, 2006).

Problem-solving interventions have been used in pediatric populations to increase treatment adherence to specific health behaviors such as taking medications and monitoring blood sugars (DeLambo, Ievers-Landis, Drotar, & Quittner, 2004). DeLambo and colleagues also found that a more supportive family relationship was associated with reports of better adherence to airway clearance treatments and aerosolized medications (DeLambo, et al., 2004). Given that managing CF can involve engaging in various health-related behaviors throughout the day, development of a daily routine that successfully merges the many treatment-related tasks with other family and daily activities is key to effective management of the illness.

Routines involve communication, commitment, and continuity. Parents first communicate to their children their expectations about the “whats, hows, and whens” of tasks that need to be completed. Commitment is demonstrated by completion of the task, and signals importance of the task to the child. Repetition of the task or procedure allows for continuity and consistent practice with similar processes and expectations, and sets the stage for children to be more responsive to structure. Children consequently develop the skills to better manage the disease. Notably, the presence of routines can serve as a protective factor when there is a disruption in the family system, whether related to management of a chronic illness or another acute stressor.

Another protective factor for some families is their use of spirituality. Parents of children with chronic diseases use spirituality to cope (Dollahite, Marks, & Olson, 1997; Grossoehme, Ragsdale, Wooldridge, Cotton, & Seid, 2010). Spirituality has also been related to pediatric treatment adherence (Grossoehme, Cotton, et al., 2013; Park & Nachman, 2010). We have been unable to locate research investigating the use of routines within the family context to manage CF in children and adolescents. Given the complexity of the daily CF regimen, there is a potential benefit to better understanding how routines can improve adherence and quality of life for children and their families. We are therefore limited in our empirical knowledge of how routines are used, how families develop them, and their impact on treatment management. The aim of this study was therefore to conduct a secondary data analysis to describe parent experiences with developing and utilizing CF care routines.

Method

Participants

This study is a secondary analysis of qualitative data collected during a mixed-methods study examining psychosocial and spiritual factors relating to parental treatment adherence in CF, which has been presented elsewhere (Grossoehme, Cotton, et al., 2013; Grossoehme, Szczesniak, et al., 2013). That primary study was approved by the Institutional Review Board at a Midwestern academic pediatric medical center. Eligibility criteria for that primary study included: 1) having a child at least three months post-CF diagnosis and 2) the child was ages 3 months to 13 years, which has been defined as the period of time when parents may be assumed to bear up to 80% of treatment responsibility (Drotar & Ievers, 1994). Exclusion criteria included the inability to speak English. The accredited CF Center follows 225 children and 108 were eligible based on age at the time of this study. A total of

83 parents were enrolled in the primary study of treatment adherence and completed questionnaires. In addition, a convenience subsample of 25 sequentially approached parents were recruited to also participate in a telephone interview about beliefs related to treatment completion (Berant, Mikulincer, & Florian, 2003; Boss, Dahl, & Kaplan, 1996; Charmaz, 2006; Eddy et al., 1998). No parent who was approached about the additional telephone interview declined to participate, and the interview participants were the first 25 who enrolled in the larger study. During the primary study's analytic phase, data related to parental use of treatment routines were frequently observed. Since that lay outside the scope of the primary study's focus on psychosocial and spiritual factors relating to treatment adherence, the present secondary analysis of the interview data (only) was undertaken.

Procedure

After obtaining consent, parents participated in a semi-structured telephone interview that included questions about daily CF treatments. The semi-structured interview was developed based on previous research (Ekedahl & Wengstrom, 2007; Grossoehme, Ragsdale, Snow, & Seid, 2011). The interview guide was intended to collect data on general parental coping and spiritual coping, as well as parents' perceptions of how spirituality might relate to their child's disease or treatment regimen. It should be noted that parental spiritual beliefs were not a prerequisite for participation, nor did all participants articulate spiritual beliefs. As with most qualitative studies, the open-ended nature of the questions elicited a variety of responses. The interview guide is included as an Appendix. Interviews were conducted on the telephone by the first author, digitally recorded, transcribed by a commercial medical vendor, and verified for accuracy by the first author. Telephone interviews are frequently utilized for sensitive topics such as substance use and sexual behaviors as they reduce demand characteristics and socially-desirable responses that can occur in in-person interviews (Jaya, Hindin, & Ahmed, 2008; Midanik, Greenfield, & Rogers, 2001). Treatment adherence is also a sensitive topic with pressure to provide socially-desirable answers (Modi, et al., 2006). Previous experience of the first two authors and numerous other studies have found phone interviews to promote rapport to elicit systematic information and additional clarification through the use of follow-up questions ("probes").

Analysis

Phenomenological methodology was used to guide data analysis and interpretation of findings. Phenomenology assumes that all knowledge is socially constructed and that common, everyday statements are epistemologically significant and may have different meanings for different people (Boss, et al., 1996). This methodology permits improved clinician sensitivity to patient and caregivers' experiences. A primary aim of phenomenological analysis is to present an accurate statement of the participants' experience and understand and describe the essential meaning of this experience. Understanding the specific experiences of multiple individuals allows empirically-gained knowledge to guide clinical care and public policy improvement (Polkinghorne, 1989).

All authors had experience in coding narrative data and CF disease management (Filigno et al., 2012; Grossoehme, et al., 2010; Grossoehme et al., 2012; Grossoehme, et al., 2011). Interview transcripts had previously been entered into NVivo 10.0, a qualitative analysis

software that was used in the primary study to organize and store the data (“NVivo qualitative data analysis software,” 2012). The first stage in the coding procedure was for the first author read to through all interviews. Additionally, part of that first step, the second author read through a randomly selected ten percent in order to obtain a general knowledge of data patterns (i.e., meaningful words, phrases, or sentences) aligned with the *a priori* research question within the interviews related to the use of routines. During that process, they independently generated a list of key words used by participants which related to the development, use, or absence of treatment routines. Prior to the investigators' review of the transcripts, it was agreed that any items which appeared on both lists would be retained and consensus would be reached on the inclusion or exclusion of all other items. Next, the list of key search terms was entered into NVivo 10.0 (NVivo qualitative data analysis software, 2012) to identify all occurrences of the key terms in the interview transcripts. The search terms included the following terms: routine, ritual, and treatment. These terms were entered individually into NVivo's Query/Text Search function, which functions similarly to the “find” function in word processing software. The Query/Text Search function returns a list of all documents in which the term appears and the number of occurrences in each document. Each document can be opened and each occurrence of the term is highlighted. In addition, statements which had previously been coded during the primary study and placed in categories named, “What helps get treatments done” and “What makes it hard to get treatments done” were also extracted. Most of the data previously coded and placed in either category were response to interview questions, “What's it like trying to get your child's recommended treatments in every day?”, “What has helped you make it through this time”, and “What has made it difficult for you to get through?” This previously coded content was specifically located and reviewed because the information in these categories was what suggested that treatment routines may be an important potential area for further empirical examination. It also allowed the authors to further verify that the NVivo search terms used in this secondary analysis identified all relevant statements appropriate for further examination.

During the second phase of coding, with all potentially relevant data extracted from the primary study's data, all authors independently reviewed the set of initial codes from the qualitative data extracted by the NVivo software several times in order to build familiarity with the data. Next, each author independently analyzed the set of initial codes and created an expanded interpretation for each data extract. Authors independently categorized the codes into clusters based on similarity of meaning, hereby referred to as “themes” and provided theme names that broadly described the overarching behavioral and psychological processes.

During the third phase of coding, all authors participated in a series of meetings to reach consensus on developing a list of primary themes discussed by participants regarding use of routines in CF care. During these group meetings authors presented their list of independently-derived themes, and discrepancies regarding the proposed themes were resolved by consensus. Typical discrepancies were differences of opinion as to whether a particular theme was sufficiently broad enough to be designated a theme, if a theme was absent from another author's list, and if a code adequately supported the proposed theme.

Discussion included reviewing data extracts (and the actual interview when more context was needed), interpreting codes, and continued until there was unanimous agreement on themes. Themes were selected because they named a concept articulated by a significant number of interviewees.

All identified themes were included in the final analysis. Finally, domain labels were developed to organize themes and provide a final meaningful higher-order framework reflecting participants' experiences with the use of routines.

Results

Twenty five parents (68% mothers) completed interviews averaging 32 minutes in length. The mean age of the children (52% female) was 7.2 years (SD=4.4). Demographic and clinical data are presented in Table 1. The number of pulmonary exacerbations was obtained by chart review, based on whether the child's pulmonologist diagnosed an exacerbation (Britto et al., 2002; Fuchs et al., 1994). Three main domains emerged from the interview themes: a) Parent experience developing a routine, b) Systems of support facilitate maintenance of a care routine, and c) Challenges to maintaining care routines. See Table 2 for domains and domain-specific themes. See Table 3 for memorable quotations that support identified domains and themes.

Domain 1: Parent experience developing a routine (see Table 3, quotes 1.1—1.17)

Major theme: Establishing a routine was difficult at first—Parents described various experiences and difficulties they encountered in establishing routines. Parents reported feeling overwhelmed shortly after their child's diagnosis with the burden of care required each day. While some families experienced challenges due to unfamiliarity with the general use of routines, others treated CF care like other daily tasks. For example, one parent conceptualized their child's CF care as similar to other necessary daily care tasks such as tooth-brushing. The majority of parents reported their current routine was the result of experimenting with different strategies, including reordering tasks and schedules to complete CF treatments in a manner that worked for them.

Major theme: Parents chose to deviate from the recommended care routine—Some parents chose to deviate from the care recommended by their child's pulmonologist either by doing fewer or more treatments. One family opted not to do the child's treatment on Sundays, in observance of a Sabbath (a day of rest) for religious reasons. Notably, two parents opted to do more treatments per day than was recommended, hoping this would improve their child's health.

Major theme: Use of a routine was influenced by learning from the positive and negative consequences of following routines and completing or missing treatments—One parent allowed her son to skip a treatment for him to learn from the natural negative health consequences of missing a treatment (i.e., difficulty breathing and potential hospitalization) with the aim of increasing his motivation for treatment adherence. Parents found conflict regarding treatments and burden decreased when the routine was more consistent because the child and parent expected the elements and order of the routine.

Parents also described children being more cooperative with treatments when they were allowed to watch TV and use electronic devices during treatments as an incentive or a distraction.

Major theme: Acknowledgement and acceptance that daily routine is going to be different because of CF treatment demands—Parents reported that the daily routine required more planning and structure to accommodate CF care demands than it would if they did not have a child with CF. Several families reported that because they prioritize CF treatments, they often arrive late to, or miss family events.

Domain 2: Systems of Support facilitate maintenance of a care routine (See Table 3, quotes 2.1-2.9)

Major theme: Support within the family system—Five aspects of family support emerged as significant. First, parents discussed their important leadership role in establishing the consistency, necessity, and schedule for their child's CF treatment completion. Second, they also discussed the establishment of treatment goals and plans, and being flexible when appropriate to promote treatment completion. Third, parent teamwork facilitated treatment completion and involved communication, shared responsibility, and alternation of treatment supervision. Fourth, the knowledge and behavior of the child with CF facilitated treatment routines when the child knew the “what, when, and how” of their treatment. Some children took the initiative and reminded parents when it was time for treatments. Finally, sibling involvement facilitated routines when they provided distraction and companionship activities during airway clearance treatments. One parent referred to the family as a “team.”

Major theme: Support outside of the family system—The presence of an in-home respiratory therapist was identified as a facilitator to creating a daily routine and teaching children about airway clearance techniques. Parents described the value of their workplaces allowing for flexible work schedules, including reducing effort to half-time, to accomplish daily CF care. One family was supported by an extended family member with CF who advised establishing and maintaining a strict routine immediately after diagnosis.

Domain 3: Challenges to maintaining care routine (See Table 3, quotes 3.1-3.11)

Major theme: Child factors—Parents identified child-related barriers to routines that occurred across developmental stages. Parents described the child developing independence from the parent and “saying no” to treatments as challenges. Child resistance to treatments arose as the child became older, when treatment times conflicted with their ability to play outside the home with peers, or limited their spontaneity. In addition, children had increased awareness of feeling different than peers due to having CF as they became older. Inconsistent child eating patterns were also reported as a challenge for parents.

Major theme: Parent factors—Parent barriers to treatment routines included being overwhelmed by the burden of daily CF care and their own inexperience caring for a child with a chronic illness. They also discussed the unpredictability of CF and the difficulty with

completing additional treatments when their child was acutely ill. Finally, one parent reported that she had difficulty waking up in the morning to start the daily CF care routine.

Major theme: Family factors—Three family themes emerged. These included the complexity of caring for more than one child, having a busy family life, and how CF can add complexity and sometimes disruption to family plans.

Discussion

To our knowledge this is the first study to describe parent experiences developing and using daily care routines to manage CF. The sample was unique in that it included children across several developmental time periods, including infancy through adolescence. Given the complexity and time-intensive demands associated with the effective management of CF, the development and use of daily CF care routines has the potential to aid families in planning and organizing the day to include illness-related tasks, academic, social, and family activities. In addition, the organization afforded by a routine allows for a start and end point for treatment-related tasks and decreases the likelihood the event consumes more time than needed, therefore decreasing perceived treatment burden.

Results from this investigation highlight several valuable areas for clinical intervention as well as future research. First, many families in this study reported that establishing a routine was difficult at first. For some families, this may be their first time being a parent and have little to no practice with developing and following a consistent daily routine. A “daily routine” is truly an abstract concept until the schedule is sufficiently practiced to become a “routine.” Some parents will need practical and concrete assistance setting up a visual schedule that includes all recommended treatments and other priorities such as sleep and schoolwork.

Moreover, many families have no experience managing the demands of a chronic illness, especially one as demanding as CF care. Once a schedule of tasks is created, families will need to practice following the schedule to determine what works, what does not work, and how to best prioritize tasks and activities. Our sample discussed this occurring through “trial and error.” Moreover, some parental decisions regarding daily care will be based on values, such as religious beliefs and parents wanting to teach the child about consequences of non-adherence, and are important to understand when providing recommendations to promote use of routines.

Second, it is important to understand sources and quality of social support including, but not limited to, family, friends, and the care team to manage CF care demands. Social support has long been documented as a key protective factor in the effective management of a chronic illness, and in CF specifically (DiMatteo, 2004). Within the immediate family, parents identified parental teamwork and leadership to identify treatment goals, develop and maintain schedules, and ensure completion as key factors. To this end, Fiese and Wamboldt (2000) discuss the importance of role assignment, including clear designation and expectations for who in the family is responsible for what component of disease management. As children learn more about their illness and practice increased independent

self-management, they will assume greater responsibility for their care. Siblings were also noted to have an important contribution to care management, supporting the notion that CF is a “family affair” and that more effective management may be promoted by all family members being a part of the daily care routine as facilitators.

The quality of the parent-child relationship is especially important for providers to attend to given that routines consist of both the specific practices of the routine, and the emotional connections between family members associated with the routine. Fiese and colleagues (2005) found that when parents and children reported poorer quality of life, there was less of an emotional investment in the asthma care routine in addition to more reported “emotional drain.” Therefore, the amount of burden, or strain, parents experience related to the daily demands of CF care, distinct from engaging in the routine, may reflect the emotional investment the family has in the routine. The emotional connection between parents and children is a powerful source of reinforcement, and can be used to encourage children to complete treatments that are part of the CF routine. Moreover, there is likely to be less parent-child conflict over adherence when children engage in regular care routines.

Findings from this study, in conjunction with previous research on routines to manage pediatric asthma, suggest that multidisciplinary CF care would be enhanced by providing families with anticipatory guidance and active intervention regarding how to develop and follow a daily routine to manage CF care (Espina, Ochoa de Alda, & Ortego, 2003). Our data support the recommendations offered by Fiese and Wamboldt regarding how to support families in order to plan routines including: 1) providers assisting the family in identifying the key necessary aspects of care, 2) working with the family to identify who is primarily responsible for each activity, and 3) planning for how to integrate the plan into the family's daily activities (Fiese & Wamboldt, 2000). Parents can “indirectly” educate their children about the importance of CF care by ensuring it is a priority, is consistently done, and treatment completion is accurately reported to the CF team during visits. “Direct” education includes ongoing developmentally-appropriate conversations about the various aspects and importance of CF management. Integrating anticipatory guidance about the benefits of routines, offering assistance with creating an initial schedule that can be practiced, and problem-solving potential barriers is therefore an important aspect of overall CF team-based care.

Clinical interventions would specifically include setting treatment goals, how to maximize shared responsibility between caregivers (when there is more than one), and adequate follow-up to ensure support and progress towards goals. Fiese and colleagues (2000) discuss that effective management of a chronic illness must be flexible in order to adapt and meet the needs of the developing child and family system. Once a routine is followed more consistently, the family will be more successful with assimilating new elements into the routine that are associated with disease progression. Assessment of the daily routine and its effect on adherence is warranted when suboptimal adherence is reported or is assumed to be responsible for suboptimal health outcomes. Finally, other sources of support beyond the CF care team such as arranged in-home services (i.e, in-home respiratory therapist) and telephone or electronic communication with other patients may assist with improving routines to fit within the family system and can be facilitated by CF teams as appropriate.

Barriers to routines noted by families included emotional and behavioral factors in children and their parents. Parents find it difficult when their children start feeling “different” than peers without CF and are managing challenging child behavior such as treatment refusal. Parents reported feeling overwhelmed by the burden of daily CF care, challenged by needing to engage and model responsible behavior themselves (i.e., waking to an alarm) to ensure that CF care is completed, and how to manage the complexity of family life with illness management. Once barriers to following the routine and treatment recommendations are identified they can be addressed using empirically-supported interventions such as problem-solving and motivational interviewing (DeLambo, et al., 2004; Duff, 2003). There is also potential promise in the use of web-based or electronic applications to aid families in self-monitoring following a routine once it is developed.

While quantitative and qualitative approaches have many clear differences, both analytic methods rely on apriori hypotheses to guide method, limited bias in data collection and analysis, and appropriate data interpretation. The richness and the fullness by which qualitative data lead to and explain a theoretical model are of significant priority within the qualitative approach. Developing a model from rich descriptions of a phenomenon requires the investigator to creatively and rigorously synthesize a theory from seemingly disparate pieces of data. Qualitative studies have been justifiably critiqued for lacking methodological rigor. Morse discussed the importance of “saturation” for ensuring a rigorous, valid qualitative model (Morse, 1995). She defined saturation as “data adequacy” and this has come to represent the point in the analytic process when “no new information is obtained.” Within this study, given no themes were excluded from the final analysis during consensus agreement it appears that saturation was sufficiently reached within the sample size, and that the inclusion of additional interviews would not have added additional critical information to the model.

While the concept of saturation is certainly an important aspect in evaluating the rigor of qualitative studies, it has been more recently critiqued because of its subjective nature. Charmaz proposed four criteria for assessing when sufficient data has been collected for interpretations to be valid (Charmaz, 2005). To increase the methodological rigor in this study, our final model was also reviewed against the criteria proposed by Charmaz. These criteria include: a) credibility, b) originality, c) resonance, and d) usefulness (Charmaz, 2005). First, credibility (i.e., validity) of the data was checked. These novel qualitative findings in the management of CF are in line with previous research investigating the use of routines in other pediatric chronic illness, including asthma. In addition, logical links exist between the initial aim (parents will describe the use of treatment routines) and the subsequent themes that were identified. Parents identified that routines were easier with practice, social support was an important facilitator, and barriers existed within the family system that impacted following the routine. Second, the data are original as our work extends the understanding of the use of routines in CF as well as offers future directions for further investigation. Third, the data has resonance, portraying the experience of routine development and maintenance by parents of children with CF. The final model's clarity would offer other CF parents insight about developing and maintaining treatment routines. Finally, the data are useful in that interpretations are immediately applicable in everyday

clinical care, and address basic and important processes, such as facilitating and removing barriers to using routines.

Limitations of the study include the use of cross-sectional design which limits our understanding of the evolution of family routines as the child ages. Moreover, treatment approaches to address barriers to use of a routine would notably vary depending on a child's age and this study included children from several developmental periods. Participants self-selected to participate and were from a single center, thereby limiting generalizability. Although telephone interviews can yield high-quality data, and may be preferable for interviews which may include sensitive topics, they are not without limitations. It is impossible to obtain nonverbal data and additional contextual data is only obtained by using interview probes to elicit more information. While every step was taken to minimize these issues, they are inherent in such methodology.

In spite of these limitations, important conclusions can still be drawn. Parent experiences were in line with previous research, documenting the initial challenges after diagnosis, parental stress, challenging child behavior, and how busy families are as barriers to CF management. This study also highlights important areas of potential clinical empirically-supported intervention. Expanding similar enquiry to multiple CF centers, and examining how routines serve as a protective factor for maintaining quality of life, decreasing burden, and optimizing adherence is warranted. Future investigations would also benefit from examining the influence of increased use of technology in health care delivery and changes in the health care landscape on quality of life and adherence, given the financial realities that influence families' ability to access or receive the necessary support in and out of the home. As technology advances, communication between CF patients and their care teams may increase in frequency, ease, and effectiveness, leading to more timely and economical interventions to improve health outcomes.

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Appendix

Semi-structured interview guide

Domain	Question	Probe
Introduction	Hello, my name is <i>N.</i> , and I am conducting the interviews for this project. Thank you for agreeing to participate in this interview to help us understand more about factors related to completing all of your child's recommended home treatments for his/her CF. We have recruited parents whose children have had CF for different lengths of time. Some parts of this interview may be uncomfortable for you because having a child with a chronic disease is stressful and can be hard to think about, and some of the topics are not things that people often talk about—such as their religious beliefs and practices. Please remember that you can ask me any questions any time, choose not to answer a question, or stop the interview. I am audiotaping our conversation so that I can focus on listening to you and not have to take notes. After we're done, the recording will be transcribed, and all of the names will be removed to protect you and your child's privacy. Any questions before we begin?	
Coping with CF	What was it like for you when your child was diagnosed with CF? How has life changed since that time? What's it like trying to get your child's recommended treatments in every day?	(probe: listen for emotion words, strategies for coping, etc)
Oblique reference to religion	What gives you a sense of hope or optimism? What helped you make it through this time? What has made it difficult for you to get through?	
	Is religion or spirituality important to you as you cope with your child's CF?	If "YES", ask, "How much strength or comfort do you get from your religion/spirituality right now? (All that you need; somewhat less than you need; much less than you need; none at all)" If "NO", ask, "Has there been a time when religion/spirituality was important to you?"
	We know that sometimes people use ways to help their child cope that were not recommended by their doctors. Have you/your child ever used any of the following to cope with your child's CF? Yoga Prayer Massage Meditation Herbal supplements Chiropractic Acupuncture Aromatherapy	For each item endorsed "yes", probe with (1) did the child find it helpful? And (2) did you discuss with your child's pulmonologist?
Spirituality	How much control do you feel you have over CF/life in general? Has your faith or beliefs changed since your child was diagnosed? Do you see yourself: working together with God as a partner? taking control over what you can, and give the rest up to God? expecting God to take control/take your worries away? praying/pleading for a miracle or to make things come out okay? making sense of the situation without relying on God?).	Does your family "have" CF or does CF "have" your family?
Faith and adherence	Do you see any relationship between your religious beliefs and how you take care of your body/your child's treatment? Do you see any relationship between your religious beliefs and getting care at the clinic or hospital (or PCP)?	probe around motivations, sanctification of the body, etc

Domain	Question	Probe
Closing	This is the end of the question we had planned to ask. Thank you very much for your time and willingness to talk with me about these topics. Is there anything that you'd like to add that you didn't have the chance to tell me about? Do you have any questions for me? Good-bye.	

References

- Berant E, Mikulincer M, Florian V. Marital satisfaction among mothers of infants with congenital heart disease: the contribution of illness severity, attachment style, and the coping process. *Anxiety, Stress & Coping*. 2003; 16(4):397–415.10.1080/10615580031000090079
- Boss, P.; Dahl, C.; Kaplan, L. The use of phenomenology for family therapy research. In: Sprenkle, DH.; Moon, SM., editors. *Research methods in family therapy*. New York, NY: Guilford Press; 1996. p. 83-106.
- Britto MT, Kotagal UR, Hornung RW, Atherton HD, Tsevat J, Wilmott RW. Impact of recent pulmonary exacerbations on quality of life in patients with cystic fibrosis. *Chest*. 2002; 121:64–72.10.1378/chest.121.1.64 [PubMed: 11796433]
- Charmaz, K. Grounded theory in the 21st century. In: Denzin, NK.; Lincoln, YS., editors. *The SAGE handbook of qualitative research*. Thousand Oaks, CA: Sage Publications; 2005. p. 507-535.
- Charmaz, K. *Constructing Grounded Theory*. Thousand Oaks, CA: SAGE Publications; 2006.
- DeLambo KE, Ievers-Landis CE, Drotar D, Quittner AL. Association of observed family relationship quality and problem-solving skills with treatment adherence in older children and adolescents with cystic fibrosis. *Journal of Pediatric Psychology*. 2004; 29(5):343–353.
- DiMatteo MR. Social support and patient adherence to medical treatment: a meta-analysis. *Health Psychology*. 2004; 23(2):207–218. [PubMed: 15008666]
- Dollahite DC, Marks LD, Olson MM. *Faithful Fathering in Trying Times: Religious Beliefs and Practices of Latter-Day Saint Fathers of Children with Special Needs*. 1997
- Drotar D, Ievers CE. Age differences in parent and child responsibilities for management of cystic fibrosis and insulin-dependent diabetes mellitus. *Journal of developmental and behavioral pediatrics*. 1994; 15(4):265–272. [PubMed: 7798372]
- Duff AJ. Cultural issues in cystic fibrosis. *Journal of Cystic Fibrosis*. 2003; 2(1):38–41. [PubMed: 15463845]
- Eddy ME, Carter BD, Kronenberger WG, Conradsen S, Eid NS, Bourland SL, Adams G. Parent relationships and compliance in cystic fibrosis. *Journal of Pediatric Health Care*. 1998; 12(4):196–202. [PubMed: 9832734]
- Ekedahl M, Wengstrom Y. Nurses in cancer care--stress when encountering existential issues. *European Journal of Oncology Nursing*. 2007; 11(3):228–237.10.1016/j.ejon.2006.09.005 [PubMed: 17134941]
- Espina A, Ochoa de Alda I, Ortego A. Dyadic adjustment in parents of daughters with an eating disorder. *European Eating Disorders Review*. 2003; 11:349–362.10.1002/erv.530
- Fiese BH, Wamboldt FS. Family routines, rituals and asthma management: a proposal for family-based strategies to increase treatment adherence. *Families, Systems & Health*. 2000; 18(4):405.
- Filigno SS, Brannon EE, Chamberlin LA, Sullivan SM, Barnett KA, Powers SW. Qualitative analysis of parent experiences with achieving cystic fibrosis nutrition recommendations. *Journal of Cystic Fibrosis*. 2012; 11:125–130.10.1016/j.jcf.2011.10.006 [PubMed: 22112934]
- Fuchs HJ, Borowitz DS, Christiansen DH, Morris EM, Nash ML, Ramsey BW, Wohl ME. Effect of aerosolized recombinant human DNase on exacerbations of respiratory symptoms and on pulmonary function in patients with cystic fibrosis. *New England Journal of Medicine*. 1994; 331(10):637–642.10.1056/NEJM199409083311003 [PubMed: 7503821]
- Grossoehme DH, Cotton S, Ragsdale J, Quittner AL, McPhail G, Seid M. "I honestly believe God keeps me healthy so I can take care of my child": Parental use of faith related to treatment

- adherence. *Journal of Health Care Chaplaincy*. 2013; 19(2):66–78.10.1080/08854726.2013.779540 [PubMed: 23593948]
- Grossoehme DH, Ragsdale J, Wooldridge JL, Cotton S, Seid M. We can handle this: parents' use of religion in the first year following their child's diagnosis with cystic fibrosis. *J Health Care Chaplain*. 2010; 16(3-4):95–108.10.1080/08854726.2010.480833 [PubMed: 20658424]
- Grossoehme DH, Ragsdale JR, Cotton S, Meyers MA, Clancy JP, Seid M, Joseph PM. Using spirituality after an adult CF diagnosis: cognitive reframing and adherence motivation. *Journal of Health Care Chaplaincy*. 2012; 18(3-4):110–120.10.1080/08854726.2012.720544 [PubMed: 23094612]
- Grossoehme DH, Ragsdale JR, Snow A, Seid M. We were Chosen as a Family: Parents' Evolving Use of Religion when Their Child has Cystic Fibrosis. *Journal of Religion and Health*. 2011:1–12.10.1007/s10943-011-9477-5
- Grossoehme, DH.; Szczesniak, R.; Dimitriou, S.; Dodd, C.; Britton, L.; Chini, B.; Seid, M. Parental adherence: spiritual, religious and psychosocial influences. Paper presented at the North American Cystic Fibrosis Conference; Salt Lake City, UT. 2013.
- Jaya Hindin MJ, Ahmed S. Differences in young people's reports of sexual behaviors according to interview methodology: a randomized trial in India. *American Journal of Public Health*. 2008; 98(1):169–174.10.2105/AJPH.2006.099937 [PubMed: 18160677]
- Mackner LM, McGrath AM, Stark LJ. Dietray recommendations to prevent and manage chronic pediatric health conditinos: adherence, intervention, and future directions. *Journal of Developmental and Behavioral Pediatrics*. 2001; 22(2):130–143.10.1097/DBP.0000000000000012 [PubMed: 11332783]
- Midanik LT, Greenfield TK, Rogers JD. Reports of alcohol-related harm: telephone versus face-to-face interviews. *Journal of Studies on Alcohol and Drugs*. 2001; 62(1):74–78.
- Modi AC, Lim CS, Yu N, Geller D, Wagner MH, Quittner AL. A multi-method assessment of treatment adherence for children with cystic fibrosis. *Journal of Cystic Fibrosis*. 2006; 5(3):177–185.10.1016/j.jcf.2006.03.002 [PubMed: 16679071]
- Modi AC, Quittner AL. Barriers to treatment adherence for children with cystic fibrosis and asthma: what gets in the way? *Journal of Pediatric Psychology*. 2006; 31(8):846–858.10.1093/jpepsy/ksj096 [PubMed: 16401680]
- Morse JM. The significance of saturation. *Qualitative Health Research*. 1995; 5:147–149.
- NVivo qualitative data analysis software. QSR International Pty Ltd; 2012.
- NVivo qualitative data analysis software. Vol. 10.0. QSR International Pty Ltd; 2012.
- Park J, Nachman S. The link between religion and HAART adherence in pediatric HIV patients. *AIDS Care*. 2010; 22(5):556–561.10.1080/09540120903254013 [PubMed: 20397076]
- Polkinghorne, DE. Phenomenological research methods. In: Valle, RS.; Halling, S., editors. *Existential-phenomenological perspectives in psychology*. New York, NY: Plenum Press; 1989. p. 41-60.
- Zindani GN, Streetman DD, Streetman DS, Nasr SZ. Adherence to treatment in children and adolescent patients with cystic fibrosis. *Journal of Adolescent Health*. 2006; 38:13–17.10.1016/j.jadohealth.2004.09/013 [PubMed: 16387243]

Table 1

Clinical and demographic characteristics of the sample

	Parent		Child	
	N	(%)	N	M (SD)
Gender				
Female	17	(68)	13	(52)
Ethnicity				
Caucasian (Non-Hispanic)	23	(92)		
African American	2	(8)		
Age				7.2 (4.4)
Pulmonary Exacerbations ^a				0.92 (1.3)

^aNumber of pulmonary exacerbations in 12 months prior to the study.

Table 2
Summary of Consensus Major Themes for Three Core Domains

Domain 1: Parental experience developing a routine	
1	Establishing a routine was difficult at first
2	Parents chose to deviate from the recommended care routine
3	Use of routine was influenced by learning from the consequences of following routines and completing or missing treatments
4	Acknowledgement and acceptance that daily routine is going to be different because of CF treatment demands
Domain 2: Systems of support facilitate maintenance of a care routine	
1	Support within the family system
2	Support outside of the family system
Domain 3: Challenges to maintaining a care routine	
1	Child factors
2	Parent factors
3	Family factors

Table 3
Memorable Quotations from Parent Interviews

Domain 1: Parent experience developing a routine
<p>Major theme: Establishing a routine was difficult at first</p> <p>1.1 At the beginning, it was like oh my gosh, how are we going to get it all in one day?</p> <p>1.2 But it was more hard for us to try to get into a routine because we weren't used to it.</p> <p>1.3 It's just normal. It's like brushing your teeth.</p> <p>1.4 So...the general outline was probably prescribed, the details of how we made it work was much more from a trial and error standpoint...</p> <p>1.5 And it's easier for me to get ready for work before I do his treatments...things like that probably kind of evolved...</p> <p>Major theme: Parents chose to deviate from the recommended care routine</p> <p>1.6 I did make the decision we don't do treatments on Sunday...a day always set aside, church, ministry-related things and rest.</p> <p>1.7 My husband and I decided to give her three treatments a day...I know her medicine only lasts for six to eight hours. I'm not willing for her to be unprotected eight hours during the day.</p> <p>Major theme: Use of routine was influenced by learning from the positive and negative consequences of following routines and completing or missing treatments</p> <p>1.8 I said, you know what's going to happen if you don't do your treatment. And he said, "well, I'll be OK, mama"...Well, the next morning, he wanted his treatment.</p> <p>1.9 The most important thing is the breathing treatment because if we don't do that, we're back in the hospital...</p> <p>1.10 ...we have seen that he can get sick pretty quickly...the downside of him being in the hospital is it's really, really, really difficult for us...</p> <p>1.11 She knows there's no compromise...same exact time...it's very easy.</p> <p>1.12 The more we keep him on a schedule, like every day at 10 am this is what we do; every day at noon this is what we do, the easier it is on us and him.</p> <p>1.13 When he is on his vest, he will sit quietly and just watch TV.</p> <p>1.14 ...our son really likes Baby Einstein™ videos so those are his treats. When he gets to do his chest percussions, he gets to watch those.</p> <p>Major theme: Acknowledgement and acceptance that daily routine is going to be different because of CF treatment demands</p> <p>1.15 The amount of...structure we need to our days is considerably more than what we probably would have had without it [CF]...</p> <p>1.16 We ran late constantly to things.</p> <p>1.17 We've not gone to family events.</p>
Domain 2: Social support as a facilitator to maintaining a care routine
<p>Major theme: Support within the family system</p> <p>2.1 I try to keep it on a routine, you know, we do this at this time and that seems to help her.</p> <p>2.2 ...her breathing treatments are not negotiable. It will happen...</p> <p>2.3 Yeah, you know, it's just we had a routine where my husband, would do the morning and then I would do the night.</p> <p>2.4 So, it really kind of helped that we knew what each other was doing, the treatments and that way, we can manage it into our schedule...</p> <p>2.5 ...when we say it's time for your vest, he gets ready for it because he'll go next to the breathing machine...</p> <p>2.6 ...usually they watch TV together [during their treatments]...it's kind of our evening routine...there's certain shows they like to watch... something we do every night as a family.</p> <p>Major theme: Support outside of the family system</p> <p>2.7 So, a respiratory therapist was able to come out and help with the treatment and that really, really helped a lot and helped to get established and...</p>

2.8 ...my job and my role, it provides me a fair amount of flexibility.

2.9 ...my first cousin also has it [CF]...so they would tell us, to start early and make sure everything is done

Domain 3: Challenges to maintaining a care routine

Major theme: Child factors

3.1 ...he going to the age now where he starts to fight some of his treatments and say no because no is his favorite word.

3.2 When she was a baby it was easier because...she had no choice...Now, she's older, she wants to be adventurous and play.

3.3 I think that's hard...they get out and see other kids, and will be like...not everybody else is doing all this stuff that I have to do.

3.4 It's an ongoing struggle and every day is different. One day she will eat so well and it's perfect...and then the next day she'll just graze and...won't eat a lot of anything.

Major theme: Parent factors

3.5 It was overwhelming, it was stressful, it was just...yeah, it was hard.

3.6 ...every day is something new...we had all bowel issues...and since he was a year old, he flip-flopped, it turned respiratory.

3.7 I have a trouble getting up early in the morning and so I have to get up earlier in the mornings to do it.

Major theme: Family factors

3.8 ...frankly, the presence of siblings, that's probably the thing that makes it the most difficult.

3.9 She plays sports. She's always on the go.

3.10 He likes to go camping, so we have to find somebody with a generator that we can do his vest with...Or...be close enough to come home to do it...at a cub scout camp, ...one of us has to be with him...to make sure he stays hydrated...

3.11 When she's sick, everybody's life has to stop and we stay home and we have to do the breathing treatments...