



Identifying Factors that Facilitate Treatment Adherence in Cystic Fibrosis: Qualitative Analyses of Interviews with Parents and Adolescents

Christina J. Nicolais¹ · Ruth Bernstein¹ · Estefany Saez-Flores¹ · Katherine A. McLean² · Kristin A. Riekert³ · Alexandra L. Quittner⁴

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Abstract

Cystic fibrosis (CF) is a progressive, genetic disease affecting multiple organ systems. Treatments are complex and take 2–4 h per day. Adherence is 50% or less for pulmonary medications, airway clearance, and enzymes. Prior research has identified demographic and psychological variables associated with better adherence; however, no study has extensively identified facilitators of treatment adherence (e.g., adaptive behaviors and cognitions) in a sample of parents and adolescents. Forty-three participants were recruited from four CF centers as part of a larger measurement study. Participants included 29 parents (72% mothers; 72% Caucasian) and 14 adolescents (ages 11–20, 64% female, 71% Caucasian). Participants completed semi-structured interviews to elicit barriers to adherence. However, facilitators of adherence naturally emerged, therefore indicating need for further exploration. Interviews were audiotaped, transcribed and content-analyzed in NVivo to identify those behaviors and beliefs that facilitated adherence, using a phenomenological analysis. Frequencies of these themes were tabulated. Nine themes emerged, with individual codes subsumed under each. Themes included social support, community support, organizational strategies, “intrinsic characteristics,” combining treatments with pleasurable activity, flexibility, easier or faster treatment, prioritizing treatments, and negative effects of non-adherence. Results demonstrated the importance of identifying strategies that positively affect adherence. Interventions that are strength-focused, build on prior success, and utilize positive models generated by those who have successfully integrated CF treatments into their lives are more likely to be efficacious.

Keywords Cystic fibrosis · Facilitators · Adherence · Adolescents · Parents

Cystic fibrosis (CF) is a progressive genetic disease affecting over 30,000 individuals in the United States and over 70,000 worldwide. National implementation of newborn screening and medical advances in treatments have contributed to a rapid increase in lifespan. As of 2015, over half of individuals with CF are at least 18 years of age, up

from 29% in 1986 (Cystic Fibrosis Foundation, 2016). CF is a complex illness that impacts multiple organs, including the respiratory, digestive, and reproductive systems (Ratjen et al., 2015). Because of its widespread effects, treatments are complex and require 2–4 h each day (Sawicki & Goss, 2015). Adherence is poor and has been estimated at 50% or less for pulmonary medications (Quittner et al., 2014).

Multiple studies have identified factors associated with poor adherence; barriers, such as forgetting, time management difficulties, treatment burden, social and academic demands, limited perceived health benefit, fatigue, and stigma/embarrassment have been well documented (George et al., 2010; Modi & Quittner, 2006). However, identification of both positive and negative influences that impact adherence is needed to improve adherence behaviors. This is critical because of the negative consequences linked to poor adherence: depression, higher health care costs, reduced

✉ Christina J. Nicolais
nicolaisc@miami.edu

¹ Department of Psychology, University of Miami, 5665 Ponce de Leon Blvd, Coral Gables, FL 33146, USA

² Department of Pediatrics, Rush University Medical Center, Chicago, IL, USA

³ Johns Hopkins Adherence Research Center, Johns Hopkins School of Medicine, Baltimore, MD, USA

⁴ Miami Children’s Research Institute, Miami, FL, USA

quality of life, increased pulmonary exacerbations and intravenous antibiotic use, more frequent hospitalizations, and earlier mortality (Briesacher, Quittner, Fouayzi, Zhang, & Swensen, 2011; Eakin, Bilderback, Boyle, Mogayzel, & Riekert, 2011; Quittner et al., 2014; Smith, Modi, Quittner, & Wood, 2010).

Treatment Facilitators in Individuals with Cystic Fibrosis

Despite the challenges of adhering to the CF treatment regimen (Quittner et al., 2014), patients anecdotally report that some treatment-related behaviors and practices are helpful in completing their treatments. Two studies have examined these factors in CF. Sawicki, Heller, Demars, and Robinson (2015) interviewed 18 youth and their parents about barriers to, and facilitators of, treatment adherence. Treatment facilitators included focusing on the importance of treatments, strong relationships with care teams, treatment routines, and transferring responsibilities from parents to adolescents. This study identified mostly behavioral and environmental facilitators of treatment, with limited identification of relevant attitudinal and cognitive factors. In addition, patients in this study attended a single clinic, limiting generalizability of the results. A similar study of 25 adolescents and young adults with CF at a single center identified facilitators, such as a good clinic team, support and reminders, perceived benefits of treatments, ease of completion, having routines, distractions and rewards for treatments, and guilt for not completing treatments (George et al., 2010). This study also focused primarily on behavioral and environmental facilitators, with limited attention to attitudes and beliefs. Additionally, this study combined adolescents and adults for their analyses, and did not interview parents. Therefore, it is difficult to apply these results to adolescents, for whom parents are still central to daily disease management. Thus, no studies to date have fully examined adaptive treatment-related behaviors, including attitudes and cognitions, in a sample of parents and adolescents with CF.

Importantly, intrinsic factors (e.g., optimism, finding meaning, confidence in the healthcare team, sense of personal control, acceptance) have been associated with positive outcomes, such as improved quality of life and social and emotional functioning (Abbott, Hart, Morton, Gee, & Conway, 2008; Findler, Shalev, & Barak, 2014; Sawicki, Sellers, & Robinson, 2011). However, none of these have been specifically associated with adherence. In studies focused on CF, perceived competence (e.g., self-efficacy, belief in one's ability to achieve success; Bandura, 1997) has been associated with better quality of life in adults (i.e., better emotional, social, and role functioning; Mitmansgruber et al., 2016; Wahl, Rustøen, Hanestad, Gjengedal, & Mourn,

2005). Self-efficacy has also been associated with better academic achievement in adolescents with CF (Grieve et al., 2011) and predicts better adherence (Parcel et al., 1994). More recently, self-efficacy was shown to moderate the relationship between depressive symptoms and adherence (Hilliard, Eakin, Borrelli, Green, & Riekert, 2015).

To date, studies of treatment facilitators have focused primarily on behaviors and daily routines. This narrow focus has limited our understanding of what facilitates and promotes adherence. To broaden our understanding of the facilitators of adherence, we identified adaptive adherence-promoting behaviors, attitudes, and cognitions elicited during semi-structured interviews with parents and adolescents with CF.

Methods

Participants

Participants were adolescents with CF and their parents, treated at one of four CF care centers: Chicago, Baltimore, and two in South Florida. Inclusion criteria for adolescents were as follows: (a) a primary diagnosis of CF and (b) age between 11 and 20 years. Participants were excluded if they: (a) had a lung transplant, (b) had a developmental disability or delay that would prevent them from completing the interview, (c) were non-English speaking, or (d) were the sibling of a participant already enrolled in the study. Recruitment goals were 12 adolescents and 24 parents of adolescents with CF. This included parents of non-participating adolescents ($n = 12$), thus, half of the parents were to be caring for adolescents that were included in the study and half were to be parents of non-participating adolescents. This oversampling of parents was done in the original study to enable both examination of agreement in reports between adolescents and their parents and to minimize the over-representation of certain points of view. Recruitment goals were also set at sample sizes which the investigators anticipated would allow for achievement of thematic saturation for barriers to treatment adherence (the aim of the original study). At three sites (Chicago and South Florida) clinic team members identified and screened participants for eligibility and interest. At the Baltimore site, the research team screened patients for eligibility and contacted them using an opt-out procedure. Interested patients were contacted in person during a clinic visit or by phone. Sampling was completed to ensure saturation of intended data. Of the 48 eligible participants, two parents declined due to scheduling conflicts and three participants were not reachable after completing consent. No other eligible participants declined participation. Health data were abstracted from the participants' electronic medical records.

The Institutional Review Board at each site approved the interviews and electronic chart review.

Demographic Information

Participants completed a self-report demographic questionnaire prior to the interview, which included age, gender, race/ethnicity, education, living situation, work/school status, household income, and insurance type. Parents also reported their relationship to the patient, marital status, and work status. Complete sample demographics are in Table 1, with sufficient detail to characterize the sample and inform extrapolation of results to other samples.

Interview

Patients and parents completed a semi-structured interview (sample items in Table 2). The interview was designed as the open-ended interview stage in the development of a measure of barriers to adherence. Questions were informed by themes elicited during two previous studies (McLean, Alpern, Madan, Riekert & Quittner, 2015; BALANCE, 2014). The questions were modified for greater clarity after the first five participants, though content elicited did not change. Three pre-doctoral graduate students, a graduate-level research assistant, and a post-doctoral fellow, supervised by two clinical psychologists conducted all interviews. The study team (consisting of psychology graduate students, a graduate-level research assistant, and two clinical psychologists) at each site reviewed each other's interview transcripts and participated in weekly phone calls to ensure that interviews were completed uniformly. Interviews were conducted in a private, patient exam room, or by phone, and lasted approximately 60 min. All interviews were audio-recorded and transcribed. Participants received \$50 for participating.

Data Analysis

Interviews for this study were conducted with the intention of eliciting barriers to treatment adherence; however, in the process of conducting interviews, patients naturally endorsed and discussed facilitators of treatment adherence. Therefore, the data discussed here are a separate and secondary set of qualitative analyses, which were conducted to examine these facilitators more fully. A qualitative approach is ideal for this study because little is known about what facilitates treatment adherence in adolescents with cystic fibrosis. Naturally endorsed patient perspectives are therefore a necessary starting point for identifying these factors.

During the first phase of this secondary data analysis, three pre-doctoral students in clinical psychology independently reviewed each transcript and, using a phenomenological approach, identified facilitating factors, defined as

concepts that adolescents or parents believed to positively influence adherence. Coding was conducted using NVivo Pro v. 11 (QSR International). Next, the pre-doctoral students compiled a list of facilitating factors identified and coded three transcripts each. The coders met as a group to determine standard definitions for each factor and ensure uniform use of each factor. Disagreements were resolved through collaborative discussions and definitions of factors were adjusted as appropriate. This list was then used to re-code each transcript. At this point, the study team compared the factors to one another to categorize them into broader themes and analyzed their occurrence. Clinical psychologist investigators oversaw the coding process, reviewing pre-doctoral graduate student work at each step.

Results

Tabulations of the frequencies and percentages of facilitation codes are reported in Table 3. They are organized into nine themes (section headings), with specific codes (named by the authors and in quotations below) falling under each major theme.

Social Support

“Parent support,” was the most highly endorsed facilitator of doing treatments. Specifically, parents and adolescents mentioned that tangible parental support (e.g., carrying medications, putting together nebulizers, washing equipment, completing manual chest physiotherapy, and setting out pills) made it easier to complete treatments. For example, one parent stated:

“... medication has to be mixed. It doesn't come pre-mixed so I have to measure out a mil. of Amikacin and four mils. of, um, saline, but it's really no big deal...I probably shouldn't [set up her equipment] because of her age, but I still do.” (mother, 20yo, F)

Adolescents reported finding such tangible support helpful. One adolescent said, “...my mom helps me...she plugs in the things and ... puts on the vest and then she turns it on.” (14yo, M) Support also came in the form of encouragement or comfort. For instance, a mother described the emotional support she provides during treatment times, saying “In the evenings, often I'll bring my laptop, sit on the end of her bed and I'll do my reporting from work. So we're in the same room just hanging out together.” (mother, 15yo, F)

Peer support was also identified as a facilitator (“social support”). Parents and adolescents reported that peers frequently supported teens by encouraging healthy behaviors and prioritization of treatments. For example, one parent stated “...[her friends] know that she needs to take her

Table 1 Participant demographics

Adolescents (<i>n</i> = 14)		
	<i>M</i> (<i>SD</i>)	<i>n</i> (%)
Age	15.89 (2.6)	
Gender		
Female		9 (64.3)
Male		5 (35.7)
Race/ethnicity		
Caucasian		10 (71.4)
African American		2 (14.3)
Hispanic/Latino		1 (7.1)
Asian/Pacific Islander		1 (7.1)
Housing status		
Live with my parents		7 (50.0)
Live with family members		1 (7.1)
Live in a college dorm		1 (7.1)
Insurance type		
Private through employer		10 (71.4)
Private, individually purchased		1 (7.1)
Public		2 (14.3)
FEV ₁ % predicted	80.2 (19.3)	
BMI	20.7 (3.5)	
CFRD	1 (7.1)	
Pancreatic Insufficient	13 (94.1)	
Hospitalizations		
0		70.6%
1+		29.4%
Patient data of participating parent (<i>n</i> = 29)		
	<i>M</i> (<i>SD</i>)	<i>n</i> (%)
Age	14.5 (2.8)	
Gender		
Female		21 (72.4)
Male		8 (27.6)
Race/ethnicity		
Caucasian		22 (75.9)
African American		2 (6.9)
Hispanic/Latino		4 (13.8)
Asian/Pacific Islander		1 (3.4)
Work status		
Working full-time		Mothers, 14 (48.3) Fathers, 20 (69.0)
Working part-time		Mothers, 5 (17.2) Fathers, 3 (10.3)
Not working		Mothers, 9 (31) Fathers, 0
Insurance type		
Private through employer		20 (69.0)
Private, individually purchased		2 (6.9)
Public		7 (24.1)
FEV ₁ % predicted	83.1 (20.7)	
BMI	20.7 (4.3)	

Table 1 (continued)

Patient data of participating parent (<i>n</i> = 29)		
	<i>M</i> (<i>SD</i>)	<i>n</i> (%)
CFRD	1 (3.4)	
Pancreatic insufficient	26 (89.7)	
Hospitalizations		
0		66.7%
1+		33.3%
Parents (<i>n</i> = 29)		
	<i>M</i> (<i>SD</i>)	<i>n</i> (%)
Relation to patient		
Mother		21 (72.4)
Father		7 (24.1)
Grandmother		1 (3.4)
Race/ethnicity		
Caucasian		21 (72.4)
African American		2 (6.9)
Hispanic/Latino		5 (17.2)
Asian/Pacific Islander		1 (3.4)
Age		
21–30		1 (3.4)
31–40		4 (13.8)
41–50		12 (41.4)
51–60		10 (34.5)
61–70		2 (6.9)
Marital status		
Single		1 (3.4)
Married		22 (75.9)
Separated		2 (6.9)
Divorced		2 (6.9)
Income		
Under \$20,000		2 (6.9)
\$20,000–59,999		6 (20.6)
\$60,000–99,999		6 (20.6)
\$100,000–139,999		6 (20.6)
\$140,000–179,999		2 (6.9)
Over \$200,000		3 (10.3)

Table 2 Sample interview items

What is setting up your [treatment] like for you/your teen?
Who is around when you/your teen does their treatments, if anyone? How about when you/he/she cleans the equipment afterwards?
What are your easiest treatments for you/your teen to get done? What makes it easy for you/them? [If response is “pills”] Other than pills, what are the easiest treatments to get done?
In what way do the following (e.g., friends, family stress, finances) affect your/your teen’s ability to follow their treatment plan? How do you/your teen work around these challenges? What has been helpful?
Do you/your teen feel comfortable telling other people about their CF? If so, who do you/does your teen feel comfortable talking about their CF with? Who do you/your teen NOT feel comfortable discussing it with?
Everyone has times in their lives when they are really good about following their regimen and times when it’s hard to get all of their treatments done. Think about a time when getting all of your/ your teen’s treatments done was fairly easy. What was going on?

Table 3 Code endorsement within themes (Parent $n=29$, Adolescent $n=14$)

Theme	Parent n (%)	Adolescent n (%)
Social support		
Parent support	26 (86.67)	12 (85.71)
Social support	18 (60.00)	9 (64.29)
Community support		
Good relationship with care team	25 (83.33)	9 (64.29)
Insurance company cooperation	13 (43.33)	1 (7.14)
School cooperation	11 (36.67)	2 (14.29)
Pharmacy easy work with	7 (23.33)	0
Organizational strategies		
Routine	24 (80.00)	7 (50.00)
Equipment or treatment location	18 (60.00)	7 (50.00)
Organization	16 (53.33)	2 (14.29)
Clustering treatments	13 (43.33)	4 (28.57)
Technology for reminders	5 (16.67)	1 (7.4)
Theme: intrinsic characteristics		
Independence	21 (70.00)	6 (42.86)
Acceptance and disclosure	10 (33.33)	10 (71.43)
Enjoys treatment	10 (33.33)	2 (14.29)
Positive beliefs about treatment efficacy	8 (26.67)	4 (14.29)
Motivated when sick	4 (13.33)	1 (7.14)
Motivation	3 (10.00)	5 (35.71)
Responsibility to family	0	1 (7.14)
Combining with pleasurable activity		
Extracurricular activities as exercise	19 (63.33)	9 (64.29)
Multi-tasking	15 (50.00)	6 (42.86)
Flexibility		
Flexibility in responsibilities	12 (40.00)	11 (78.57)
Flexibility in treatment schedule	7 (23.33)	8 (57.14)
Easier or faster treatment		
Treatment is quick	16 (53.33)	10 (71.43)
Treatments or technology improvement	11 (36.67)	4 (28.57)
Prioritizing treatment		
Prioritizing treatments	9 (30.00)	5 (35.71)
Negative effects of non-adherence		
Negative effects of non-adherence	10 (33.33)	2 (14.29)

medication with her meals... they're encouraging her to make sure she eats at lunch time, and that sort of thing." (father, 12yo, F) Adolescents endorsed similar experiences: "...my friend will be—the eye for me... so just in case I get—I pass out or something, she'll be able to get my inhaler..." (15yo, F) Thus, parents and adolescents perceive social support as an important positive influence on adherence behaviors.

Community Support

"Community support" captures positive relationships with institutions, which were perceived to increase adherence: "good relationship with care team," "insurance company cooperation," "school cooperation," and "pharmacy easy to work with."

"Good relationship with care team" was the second most frequently endorsed code by parents; however, it ranked lower among the adolescent codes (only the 6th most frequently endorsed). For parents, the relationship with the CF care team served two primary functions. First, parents reported that the care team provided education about fitting treatments into daily life (e.g., how to complete treatments while traveling, explaining the disease, and education on the function of specific treatments). One parent highlighted how helpful the pulmonologist was in educating her daughter about the value of adherence: "... [The pulmonologist is] the one that has really been instrumental in explaining to [Patient] how it works and what's going on and showing her the CAT scan and answering very candid questions directly head on." (mother, 15yo, F) Parents also mentioned that treatment completion was more feasible when providers quickly answered their calls, called in prescriptions, scheduled appointments, and advocated on their behalf with insurance companies. One parent stated, "...I go to [nurse practitioner] and she...not once ever told me 'No' or 'I don't know.' [She] makes sure she calls me back before the day is over ... I love my, um, my CF staff who's uh, taking care of my baby." (mother, 11yo, F) Another parent commented on the helpfulness of the team in managing insurance difficulties: "[CF center]...will help you fight, they know the ropes, they'll write the letters, they're very prompt um about following up on things ... then just you know not giving up, not taking no for an answer." (mother, 17yo, F) Therefore, our interviews with parents demonstrated that care teams play an essential role in facilitating treatment adherence.

In addition to the team, other community support was helpful for families. Parents endorsed the importance of schools, insurance companies, and pharmacies; however, adolescents rarely acknowledged these factors, which further emphasizes the role parents play in supporting their adolescents. Parents appreciated it when they were responsive and flexible, making access to medications much easier [e.g., "Our health insurance is wonderful." (father, 14yo, M); "...any issues I have I call [the pharmacy] and they're right there." (mother, 11yo, F)]

Organizational Strategies

Families reported using "organizational strategies" to maintain treatment materials, and establish the timing and order of treatments. This theme included "routines," "equipment

or treatment location,” “organization,” “clustering treatments,” and “technology for reminders.” “Routines” were highly endorsed by parents and moderately endorsed by adolescents. This code represented that integrating treatments into daily schedules ensured their completion. For example, one parent explained that their adolescent’s treatments are completed after dinner every night: “We eat dinner and we do the dishes and we do the treatment right after that.” (mother, 15yo, F) Another explained that treatments are completed right after waking in the morning and right before going to bed every night: “We don’t really let anything get in the way, honestly. Um we just...it’s your standard, get up, this is what you do. This is what you do before you go to bed.” (mother, 11yo, F) Similarly, parents explained that having routines related to the order of these multiple treatments was also helpful:

Mother, 15yo, F: She’ll do her HyperSal and then I’ll go back with her Pulmozyme and just kind of check in between. I’ll set a timer and then when the vest goes off, even if I’m in another room, I know that it’s time to do her TOBI.

Another organizational strategy families used was keeping treatments nearby, accessible, and visible (“equipment or treatment location”). For example, many parents and adolescents reported that taking treatments with them increased the likelihood of their completion [e.g., “she always... has enzymes with her” (mother, 17yo, F)]. Having treatments in specific, convenient locations was also frequently mentioned:

12yo, M: Well, I keep it set up in a specific...in the corner...right next to me. So, and it’s like by the extension cord so I can plug it in. So all I have to do in the morning is just plug it up and get right to it.

Families also used management systems to organize treatments (“organization”). Parents reported that adolescents were more likely to complete treatments when parents used organizational systems for medication refills [e.g., “I have a spreadsheet with all the different meds and I manage that” (mother, 15yo, F) and the medications themselves [e.g., “I have everything lined up in the-, in the ‘frigerator and in the kitchen so when we wake up in the morning...we start off our process.” (mother, 11yo, F) Families also used “clustering,” which was coded when a participant simultaneously completed multiple treatments to shorten treatment time [e.g., “while he’s doing his nebulizer, he can do his vest treatment” (mother, 19yo, M)].

Intrinsic Characteristics

“Intrinsic characteristics” are those attitudes and emotional states that participants endorsed as facilitators of treatment

completion. These included “independence,” “acceptance and disclosure,” “enjoys treatment,” “positive beliefs about treatment efficacy,” “motivated when sick,” “motivation,” and “responsibility to family.” “Acceptance and disclosure” was the third most frequently endorsed facilitator for adolescents, but was more rarely endorsed by parents. This code represented patient acceptance of their disease and disclosure to others. For many adolescents, the ability to disclose their disease to others enabled them to complete treatments in front of their peers (e.g., in their dorm room, at social events, while friends were at their home, in the lunch room). For example, one adolescent stated: “...basically at 7 o’clock I go to church youth group... I wouldn’t be going to bed until 10 on a school night. It was always a pain so I am able to do my vest while I’m there.” (15yo, M)

Adolescents explained that acceptance and disclosure of their disease was important. For example: “I store [treatments] in my room. I just kind of have them wherever there’s an outlet...my friends know everything about me so it just doesn’t bother them...I talked to [my boyfriend] about it, he’s totally fine with it...” (19yo, F)

Parents highly endorsed “independence” as a facilitator. This code captured treatment-related behaviors that adolescents can manage independently [e.g., “...at this point, she’s able to...fill her nebulizer cup. She can manage all of her equipment, and do her treatments on her own.” (father, 13yo, F); “...I do it all by myself... usually like in the morning like I am on my own because my parents are at work...” (19yo, M)]. These independent attitudes and behaviors had often emerged recently, and were typically accompanied by parental support.

The “intrinsic characteristics” theme also captured attitudes commonly assumed to increase treatment adherence. For example, “positive beliefs about treatment efficacy,” which is belief that treatments work to reduce symptoms, was endorsed by about 25% of parents and teens [e.g., “Yeah, so she knows that it’ll help so she’ll, she takes them” (mother, 18yo, F); “I know I have to do them to stay healthy” (16yo, M)]. Similarly, “motivation” represented an intrinsic drive to complete treatments [e.g., “she’s usually pretty good with uh- minimal encouragement, and getting done what she needs to get done.” (father, 17yo, F)], and “motivation when sick” represented the tendency to adhere to treatments more during exacerbations [e.g., “then I get sick, and then I’m like oh okay, well I’m going to do it now.” (15yo, F)].

Combining with Pleasurable Activity

Parents and adolescents reported that engaging in pleasurable activities while completing treatments facilitated their completion. For example, completing their exercise during extracurricular activities made fitting in regular exercise easier [“exercise as physical activity”; e.g., “...]

now we're doing running, and she actually likes it. So it's nice when they like it, you don't have to make her do it." (father, 11yo, F)]. Multitasking represented a similar concept in which treatments were paired with pleasant or productive activities. One adolescent stated: "I don't mind doing the vest, because...I can do [it] whenever. When I'm watching TV or just hanging out, whatever, I'll do my vest." (16yo, M) Parents also endorsed multitasking in a positive way [e.g., "He goes and sits on the couch in our living room where he's got the TV and, you know, games, etc. And can, you know, take his meds and sit there and do his treatments" (mother, 19yo, M)]. Thus, parents and teens agreed that incorporating pleasant activities during treatment increased adherence.

Flexibility

Parents and adolescents endorsed both "flexibility in responsibilities" and "flexibility in treatment schedules" as positive influences. "Flexibility in responsibilities" was represented by adolescent statements that treatments were easier when non-treatment responsibilities were lessened. For adolescents, this was most commonly endorsed when they did not have school (e.g., summers, weekends, cancelled school days) or extracurricular activities. One adolescent exemplified this, saying:

16yo, M: All days in the summer is pretty easy. Because I wasn't...required to do anything. So I would just wake up, do them, get them done with, and just have the rest of the day to bullsh** and do whatever... summer makes everything easier. No stress, no nothing. It's just nice.

Similarly, another adolescent explained that treatments were easiest to complete "...when it was like a big hurricane, when like I couldn't go to the park and like the internet was like really down and I couldn't like go on the computer so I wasn't really distracted a lot and I got to do everything." (14yo, M) Although parents endorsed this code less frequently, they spoke about it similarly, saying that treatments were easier to complete when "...we didn't have activities in the afternoon, be it flute lessons, her tutoring...her church. If there was no other life involved..." (mother, 15yo, F)

Adolescents also endorsed the value of "flexibility in the treatment schedule" which allowed them to enjoy other activities by adjusting their treatment times [e.g., "... if I know I'm not going to have enough time to do them that night, I might try to wake up a little bit earlier and do them in the morning." (16yo, M)]. Parents endorsed this facilitator significantly less often.

Easier or Faster Treatment, Prioritizing Treatments, and Negative Effects of Non-adherence

Three final themes emerged. Parents and adolescents shared that time and efficiency in completing treatments made adherence easier ("quick treatments"; e.g., albuterol, pills). Parents and adolescents also reported that innovations in treatment modalities that reduce time (e.g., development of dry powder formulations of antibiotics and a nebulizer that also completes airway clearance) facilitate adherence. In addition, "prioritizing treatments," which captures the family's tendency to make treatments the key focus of their time (rather than other enjoyable activities, obligations, or life events) and "negative effects of non-adherence," which recognized that avoiding doing these treatments has negative, physical consequences (e.g., digestive symptoms when skipping enzymes) were also discussed.

Discussion

This study was the first to extensively examine behavioral and attitudinal facilitators of treatment completion in parents and adolescents with CF. We aimed to identify adaptive behaviors, attitudes, and cognitions that parents and adolescents reported as having positive influences on treatment completion. Data were obtained using a qualitative coding method applied to semi-structured interviews with parents and adolescents.

A number of noteworthy conclusions can be drawn from these findings. First, both parents and adolescents perceived that social support increased treatment adherence. At the code level, they both reported that parents offered tangible (e.g., treatment set-up, carrying medications) and emotional (e.g., presence during treatments, encouragement) support. This result aligns with studies showing that parental monitoring and supervision are associated with better treatment adherence (Modi, Marciel, Slater, Drotar, & Quittner, 2008). Interestingly, little tangible support was obtained from peers, but peers were a critical source of emotional support (e.g., reminders, encouragement). Prior studies have reported similar patterns (e.g., Barker, Driscoll, Modi, Light, & Quittner, 2012).

Parents, and adolescents to a lesser degree, also reported that treatment-related support from community institutions was important. The CF care team was the most frequently endorsed type of community support, with the team providing mostly tangible types of assistance (i.e., managing insurance issues, prescribing refills, pre-authorizations for medications) as well as basic education about CF and the purpose of various treatments. Many members of the care team (e.g., pulmonologist, social worker, nurse practitioner) provided this type of support, and participants identified specific types

of support received from different providers, indicating that each member of the team contributed uniquely to their adherence. Two existing qualitative studies of individuals with CF (George et al., 2010; Sawicki et al., 2015) found similar results. Thus, our findings are in line with existing research, but extend the results to a geographically diverse parent and adolescent population. Our results suggest that future research should focus on the extent to which health care providers affect adherence, identifying the best ways to deliver this type of support. Given how busy CF centers are, this type of research could identify how to optimize support provision when resources are limited.

Parents and adolescents also perceive benefits in using various organizational approaches. The most frequent organizational strategies included routine, convenient location of treatments, and maintaining an organization system. Although other studies have reported that routines facilitate treatment adherence (George et al., 2010; Sawicki et al., 2015), our finding that families use specific “equipment or treatment location” strategies is unique. Specifically, we found that simply keeping treatments in accessible and visible locations increases perceptions of adherence.

Existing research has examined adaptive attitudes and cognitions in individuals with CF (e.g., optimism, acceptance, self-efficacy); however, few studies have examined their relationship to treatment completion. Unique to this study is our finding that patients report a number of “intrinsic characteristics,” or attitudes and cognitions that patients perceived to improve their adherence. “Independence” and “acceptance and disclosure” were two “intrinsic characteristics” that seemed to capture constructs that are similar to self-efficacy, which has been widely associated with adherence (e.g., Hilliard et al., 2015). Furthermore, disclosure, which is similar to this study’s identified code of “acceptance and disclosure,” has also been empirically associated with greater independence and better adherence and health outcomes (Borschuk et al., 2016). Thus, our study extends these findings by identifying a qualitative link between disclosure to peers, a sense of acceptance, and treatment adherence.

Interestingly, parents frequently reported “independence” as a positive influence on adherence. Both parents and adolescents also endorsed “parent support” as a treatment facilitator. Although this simultaneous desire for parent involvement and greater independence may seem contradictory, our findings suggest that families aim to integrate parental support with greater independence. This “combination strategy” should be potentially integrated into interventions that aim to both increase adherence and promote the transition to adult care (Madan, Alpern, & Quittner, 2014). In a previous study, this pattern emerged around age 15, with adolescents completing their treatments relatively independently; however, because parental support was withdrawn, rates of adherence

were poor and this led to parents becoming re-involved in treatments by age 16–17 to ensure their completion (Modi et al., 2008). This delicate balance between parental involvement (using positive support strategies) and the promotion of adolescent independence was mentioned frequently by both parents and teens. Existing research suggests this can be best managed through a gradual transition of task responsibility from parents to adolescents, implemented in stages (e.g., hold the mouthpiece, then turn on the equipment, then fill the medicine cup, then clean equipment; Madan et al., 2014).

As previously mentioned, having structured “routines” was an important facilitator for parents. This strategy has been identified in previous studies (George et al., 2010; Sawicki et al., 2015), however in our study, adolescents *more frequently* mentioned “flexibility in responsibilities” and “flexibility in doing treatments.” Although structured routines and flexibility seem contradictory, some families had adopted a good balance. Specifically, having “routines” describes a typical daily pattern of activities in which other activities are fit in around treatment times and “flexibility” denotes a preference for having fewer outside activities, and moving treatment times around to accommodate them. Importantly, our findings contrast those of studies showing better adherence on school days and during time-pressured situations (Ball et al., 2013). Thus, while adolescents may believe that their adherence is better on days when they are less busy, their reports may be biased by a decreased sense of stress.

This study has clear clinical and research implications. First, these findings demonstrate the importance of asking patients not only “what’s getting in the way,” which is the typical question asked in the clinic setting, but also asking “what’s working?” By doing so, clinicians can reinforce behaviors that are compatible with good adherence to ensure that they continue. Second, the treatment-facilitating behaviors and attitudes identified in this study could be incorporated into clinical interventions to improve adherence. This could be accomplished using a positive deviance approach (Marsh, Schroeder, Dearden, Sternin, & Sternin, 2004), which posits that we can identify behaviors used by individuals already achieving unexpected, good outcomes (high adherence) despite high risk (high treatment burden), and encourage their use in patients with worse outcomes (poor adherence). Future studies should evaluate the efficacy of this approach. Given that the focus of our study was on adolescents, these findings may be particularly informative for interventions targeting this developmental period.

Although this study has many strengths, it is not without limitations. First, the interview we used was designed to elicit barriers to treatment adherence. While this offers support for the facilitators that were mentioned “organically” in the interview, it also has implications for the sample. Namely, sample size was determined to be sufficient

due to saturation achieved for barriers to treatment adherence rather than facilitators. Second, as with all interview methods, participant responses may have been influenced by a social desirability bias. Third, all participating adolescents ($n = 14$) had parents who participated in the study, but all remaining parents ($n = 15$) did not have a participating adolescent. Thus, parents were over-represented and may have inflated the frequencies of some facilitators. This also prevented us from examining congruence between parent and adolescent perceptions of treatment facilitators. However, this recruitment strategy enabled us to include additional fathers and mothers who would have been excluded using a dyadic approach. Fourth, our participants were mostly female. Therefore, the facilitators identified in this study may be more relevant to females than to males. Finally, although our participants were asked to identify facilitators of treatment adherence, we cannot confirm that those facilitators actually increased adherence, given that adherence behaviors were not measured. Therefore, our results should be interpreted in this context.

Conclusions

This study identified behaviors, attitudes, and cognitions that parents and adolescents with CF perceived to facilitate adherence. We identified a number of behaviors and attitudes that may be helpful in completing this time-consuming, complex regimen. The next step would be to incorporate them into interventions that seek to promote better adherence, and interventions facilitating transition to independent care, and evaluate their efficacy. Specifically, clinicians working in CF centers may be able to focus on patient strengths and build on prior patient successes in integrating CF treatments into daily life. Our results suggest that such an approach is most likely to lead to effective behavior change.

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Compliance with Ethical Standards

Conflict of interest The authors Christina J. Nicolais, Ruth Bernstein, Estefany Saez-Flores, Katherine A. McLean, Kristin A. Riekert, and Alexandra L. Quittner declare that they do not have any conflict of interest.

Ethical Approval All procedures were in accordance with the ethical standards of the institutional research committees and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed Consent Informed consent was obtained from all individual participants included in the study.

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