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I, Kendra Deanne MacLeod, hereby submit this original work as part of the requirements for the degree of: Doctor of Philosophy

in Psychology

It is entitled: Evaluating Adherence to Continuous Positive Airway Pressure Therapy in Children with Sleep-Disordered Breathing

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Committee Chair signature: Steven Howe, Ph.D.
Evaluating Adherence to Continuous Positive Airway Pressure Therapy in Children

with Sleep-Disordered Breathing

A dissertation submitted to

Division of Research and Advanced Studies
of the University of Cincinnati

in partial fulfillment of the requirements
for the degree of

DOCTOR OF PHILOSOPHY

In the Department of Psychology
Of the College of Arts and Sciences

2009

by

Kendra D. MacLeod

M.A., University of Cincinnati, 2005

Committee: Steven Howe, Ph.D. (chair)
Kelly Byars, Psy.D.
Laura Nabors, Ph.D.
Robert Stutz, Ph.D.
ABSTRACT

The purpose of this study was to systematically examine the experience of caregivers and their children who were using continuous positive airway pressure (CPAP) to treat sleep-disordered breathing (SDB) and to examine barriers to treatment adherence through multiple perspectives (i.e., caregiver, child and medical team). The study also aimed to provide recommendations to improve care received by families being served by the Sleep Disorders Center (SDC). The family sample consisted of 15 caregivers and their children who were using CPAP as a treatment for SDB. The medical team sample included nine clinicians who were responsible for treating the children with SDB. Procedures consisted of caregiver interviews, child and medical team focus groups and supplementary data. Interviews and focus groups were analyzed using the constant comparative method for producing emergent-grounded theory. In summary, the results of this study demonstrated that adhering to CPAP is especially challenging in children and interventions designed to promote CPAP adherence need to be family-based, ongoing and multi-faceted. Given the potential for high emotional costs to CPAP use for both the parent and child, care needs to focus on maximizing the benefits of CPAP use.
ACKNOWLEDGEMENTS

I am grateful to the Sleep Disorders Center staff at the Cincinnati Children’s Hospital Medical Center (CCHMC), as they made it possible for me to complete this project. It also bears mentioning that this study received support from the original research fund division from Behavioural Medicine in Clinical Psychology at CCHMC.

To Professor Howe, I owe an enormous debt of gratitude. He stepped in when I needed direction and saw my potential. He was a constant support during times that were challenging. His knowledge and humour was much appreciated (even the bad Canadian jokes). He has enriched my thinking and enhanced my competency as a researcher.

I am incredibly appreciative of Dr. Byars’ involvement. Without his willingness to mentor me, I could not have undertaken this study. His expertise and guidance was instrumental throughout the entire process. He was generous with his time and wisdom and he has had a crucial influence on my development as a clinical psychologist.

Professor Nabors read this dissertation and raised pertinent questions that helped me enhance the final document. I also want to thank Professor Stutz for his insightful comments. My committee was incredibly supportive throughout the entire process.

On a closing note, I never imagined that I would enter the work force the same year my father retired. I would not have gotten through the obstacles I faced without the invaluable foundation and encouragement from my family and friends. My Uncle Ken deserves specific mentioning as his computer skills were a major asset in preparing for my defense presentation (who knew that a Ph.D. would not prepare you for navigating YouTube). Over the course of my graduate career I have been fortunate to develop
everlasting friendships that have been vital to my success. I am grateful for the cultural and training opportunities that the United States has provided me.
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Introduction

Sleep-Disordered Breathing

Sleep-disordered breathing (SDB) is common and leads to serious morbidity during childhood (O’Brien et al., 2004). The spectrum of SDB ranges from persistent primary snoring to repetitive episodes of complete or partial blockage of airflow seen in obstructive sleep apnea (OSA; O’Brien et al., 2004). Habitual snoring has been reported to occur in 3-12% of the general pediatric population (Redline et al., 1999) and OSA has an estimated prevalence of 1-3% (O’Brien et al., 2004). Airway obstruction that characterizes SDB leads to sleep fragmentation and gas exchange abnormalities in affected children (Mindell, Owens, & Carskadon, 1999). If left untreated, children with SDB may experience impaired school functioning, behavioral difficulties such as inattention, aggression and hyperactivity, neurocognitive deficits such as poor executive functioning and memory, and medical morbidity such as hypertension and hormonal problems (O’Brien et al., 2004; Owens, Opipari, Nobile, & Spirito, 1998; Streisand & Efron, 2003).

Adenotonsilar hypertrophy--enlarged tonsils and adenoids--is the most common etiology for SDB in children (Streisand & Efron, 2003). However, there are other common mechanisms for SDB including structural anomalies that result in upper airway narrowing and decreased neuromuscular tone that results in a floppy upper airway (Streisand & Efron, 2003). Thus, children with craniofacial anomalies and neurological disorders that affect upper airway openness are at an elevated risk for SDB. Tonsillectomy and adenoidectomy (T&A) is the first-line treatment for pediatric SDB when the condition is attributed to adenotonsilar hypertrophy (Streisand & Efron, 2003).
However, not all children experience symptom relief after surgery (Streisand & Efron, 2003). In addition, post-surgical complications associated with T&A (e.g., hemorrhage, respiratory decompensation, anesthetic complications, pain) have been observed in up to 32% of pediatric cases (Ruboyianes & Cruz, 1996). Furthermore, there is an increasing prevalence of children developing SDB associated with obesity rather than enlarged adenoids or congenital malformations of the mouth and throat (Streisand & Efron, 2003). Thus, alternatives to surgical treatment of pediatric SDB are needed.

**Efficacy of Continuous Positive Airway Pressure**

Continuous positive airway pressure (CPAP) is the most common treatment choice for adults suffering from SDB (Sullivan, Issa, Berthon-Jones, & Eves, 1981) and has been shown to lead to improved cognitive and psychiatric functioning, decreased daytime sleepiness, and improved cardiovascular functioning (Beebe, Groesz, Wells, Nichols, & McGee, 2003; Montserrat et al., 2001). CPAP is generated by a blower attached to a mask or nose piece and serves to stent the airway open during sleep and thus prevents apneic episodes (Massa, Gonsalez, Laverty, Wallis, & Lane, 2002). The Food and Drug Administration has approved CPAP for domestic use in children. Recent research supports the efficacy of CPAP as a treatment for SDB in children; polysomnographic (PSG) data collected after the start of CPAP treatment shows improvements in sleep parameters and parental report indicates improvements in daily functioning (Kirk, & O’Donnell, 2006; Marcus et al., 2006; Rains, 1995). Unfortunately, despite such promising treatment outcomes, many children have difficulty tolerating and adhering to CPAP (Kirk, & O’Donnell, 2006; Marcus et al., 2006; Rains, 1995).
**Adherence to CPAP**

In the first study to use objective measures to evaluate adherence rates in children using CPAP (aged 2 to 16), approximately one-third of the participants dropped out before six months and the average hours used per night was only 5.3 (Marcus et al., 2006). Another study examining effectiveness of CPAP followed children of all ages from initial titration to a year post titration (Massa et al., 2002). The study reported that 74% of the children in their sample were able to successfully tolerate CPAP (Massa et al., 2002). However, once CPAP was tolerated 44% of children were not able to establish long-term use (Massa et al., 2002). Similarly, O’Donnell and colleagues (2006) reported that 82% of the children in their sample (ranging from infants to adolescents) were able to successfully tolerate CPAP. Yet, for over half of the children, the process took considerable time, ranging from 9 to 295 days (O’Donnell, Bjornson, Bohn & Kirk, 2006). Furthermore, of the 82% who were able to tolerate CPAP, 12% had needed intensive psychological support to achieve success and 20% were not able to establish long-term use despite substantial support provided by the clinical team managing the children’s medical treatment for SDB (O’Donnell et al., 2006). Finally, a retrospective chart review of patients aged 7 to 19 followed by a pediatric sleep clinic documented “good adherence” (i.e., defined by meter readings of > 4 hours use per night and 5 or more days use per week) in 70% of their patients (Uong, Epperson, Biathlon, & Jeffe, 2007). High adherence in this particular sample may be attributed to a comprehensive education program, a high level of support, and regular follow-up that was standard of care in the sleep program (Uong et al.).
There is a paucity of systematic research examining specific barriers to CPAP treatment in children (Kirk & O’Donnell, 2006). Anecdotal observations are the basis for commentaries regarding barriers to CPAP adherence that have been published in studies examining the efficacy of CPAP therapy in children (Marcus et al., 2006; Massa et al., 2002; McNamara & Sullivan, 1999; Pallombini, Pelayo, & Guillemiault, 2004). Common reported barriers associated with initial difficulty tolerating CPAP were related to mask fit and nasal discomfort (Massa et al., 2002). One study examining adherence rates and effectiveness of CPAP in 30 children with OSA who ranged from less than one year of age to 19 years old, did document barriers; however, it focused specifically on physical side-effects and did not examine additional correlates of adherence found for other medical treatments (Massa et al., 2002). For instance, knowledge about treatment, beliefs about health, parental monitoring, motivation, locus of control, medical visits, perception of illness severity and age (Rapoff, 1999).

Interestingly, research suggests reported physical complications have been minimal (Kirk & O’Donnell, 2006; Massa et al., 2002) and in two studies no children who successfully tolerated CPAP at the beginning discontinued due to physical side-effects from the treatment (Massa et al., 2002; O’Donnell et al., 2006). Kirk and O’Donnell (2006) support these findings; they stated that physical complications in the children they had treated with CPAP, representing all age groups, had been minimal, especially when care was taken in the beginning to adequately fit the mask and maximize comfort. They did find that if the mask did not fit at the initiation of CPAP then the chance of the child displaying good adherence in the future was minimal (Kirk & O’Donnell, 2006). Furthermore, there is also some evidence that barriers to adherence in
the first few weeks are often different than the barriers experienced by families after a longer period of CPAP use (Marcus et al., 2006). For instance, equipment and mask difficulties may be more of a barrier in the first few weeks where as parental monitoring and motivation may be barriers associated with long term use (Marcus et al.).

**Interventions to Increase CPAP Adherence**

There are a few preliminary studies that have examined interventions to improve CPAP adherence in children. Unfortunately, these studies are limited by the lack of robust data regarding barriers to CPAP adherence in children. In addition, findings from these studies have limited generalizability (Rains, 1995). In one study examining the efficacy of CPAP, when the initial titration was not accepted by the child, a home behavioral intervention was offered involving progressive introduction of the CPAP from parents using a structured protocol (e.g. use mask alone during the day while playing until child can wear it without stress then encourage child to fall asleep with it, then introduce pressure; Massa et al., 2002). The home behavioral intervention was successful in increasing adherence for only 4 out of 17 children who were not able to initially tolerate CPAP (Massa et al., 2002). The reasons identified included parent refusal, children being lost to follow-up, and complications from comorbid medical conditions (Massa et al., 2002). Importantly, the study found lower adherence rates in children between the ages of one and five because of refusal to wear the mask and difficulty tolerating air pressure (Massa et al., 2002). Consistent with these findings, Kirk and O’Donnell (2006) also reported age to be an important determinant of mask and pressure toleration. They found that children less than 12 months and older than 6 years were more likely to tolerate CPAP during their first exposure. Furthermore, a study examining
compliance rates in children aged 6 months to 18 years found age to be a significant factor in number of nights used and hours of use per night as compliance decreased with increasing age and was lowest among children between the ages of 13 to 18 (O’Donnell et al., 2006). Thus, the most salient barriers to CPAP adherence may vary with age.

Achieving proper mask fit and pressure is essential for successful CPAP treatment; however this can be challenging in children due to more rapid rates of physical growth and difficulties obtaining correct mask sizes (Palombini et al., 2004). Thus, the importance of regular clinic visits and PSG studies to re-evaluate OSA symptoms is even more important in children (Kirk & O’Donnell, 2006; McNamara & Sullivan 1999; Palombini et al., 2004). Unfortunately, there is no current universal standard for follow-up. One study examined the experience of pediatric doctors treating OSA in nine different sleep centers through a questionnaire (Marcus et al., 1995). They found that of 94 patients identified, 21 needed modifications of pressure levels after first titration PSG due to patient growth (Marcus et al., 1995). Yet, the results from the questionnaire revealed marked variability in treatment protocols for each center, highlighting the need for the development of consistent standards that incorporate regular long-term follow-up (Marcus et al., 1995).

A recent article about maximizing adherence in children recommended the following standards for follow-up PSG’s: every 3-4 months in children less than one year, every 6 months for 1 to 3 year olds, annually for 3 to 8 year olds and every 18 to 24 months for children older than 8 (Kirk & O’Donnell, 2006). The authors discussed the importance of providing the child and their parents with an initial positive experience to CPAP treatment. Thus, if a child is resistant to CPAP they recommend an individually
tailored behavior plan that not only allows the family to be reinforced for adherence behavior at home but also provides the parents with skills to provide reinforcement for their child (Kirk & O’Donnell, 2006). In the children (including infants to adolescents) who received behavioral interventions, a median time of 68 days from diagnosis was needed before they were ready to initiate CPAP in their first titration study. Given the consistently documented difficulties with adherence and toleration of CPAP, Kirk and O’Donnell (2006) strongly encourage sleep clinics to identify a CPAP coordinator to keep the family connected to the sleep physicians, and to provide a high level of ongoing support for families (such as providing assistance with purchasing equipment, helping with the transition to home, and being available to address problems).

Several other researchers have also identified the need to target the parent as well as the child (e.g., McNamara & Sullivan, 1999; Rains, 1995). For example, one study examined effectiveness of CPAP therapy to treat OSA in children younger than one year and found that parental nonadherence was associated with treatment discontinuation (McNamara & Sullivan, 1999). The study also reported that parents who were adherent often stated that their efforts (i.e., checking mask during night) were worthwhile as they noticed improvement in their infant’s daytime behavior (McNamara & Sullivan, 1999). Similarly, O’Donnell and colleagues (2006) found that parents’ attitudes and beliefs affected compliance rates in children from infancy to adolescence. In those families who were not able to establish long-term use, parents stated reasons such as feeling overwhelmed, not seeing the need for CPAP, or feeling uncomfortable with supporting a treatment that was so undesirable to their child (O’Donnell et al., 2006). Rains (1995) developed a behavioral intervention to increase CPAP adherence in children between the
ages of 6 to 12 years old and found that parent training in problem-solving and positive reinforcement were necessary for success. Finally, a group of researchers who examined the effectiveness of a multi-component behavioral training program in increasing adherence to CPAP in four pre-school aged children who either had a developmental delay, behavioral problems or serious physical health difficulties declared that long-term success was dependent on parental variables (Slifer et al., 2007). They stated that without parental dedication and belief in treatment, the likelihood of adherence to CPAP therapy in children is diminished (Slifer et al., 2007). Caregiver training (e.g., behavioral management, education) was seen as a crucial component of their intervention protocol (Slifer et al., 2007). Thus, the results from these studies highlight the necessity of helping parents to understand the benefits of CPAP and providing them with the knowledge and skills appropriate for their child’s development level (McNamara & Sullivan 1999).

**Theoretical Framework for Understanding Pediatric Adherence**

Increasing adherence to CPAP in children is likely to require substantial support. Because initial tolerance and long-term use seem to be problematic, interventions will need to focus on removing barriers related to both of these factors, such as improving follow-up procedures and mask fit, as well as examining personal, developmental, parental and environmental factors. Given the complexity of children’s adherence, broader theoretical frameworks are being developed to incorporate multiple systems and variables. For instance, De Civita and Dobkin (2004) recently proposed a broader model for understanding pediatric adherence and operationally defined adherence as “the manifestation of multiple treatment-related behaviors prescribed by a medical team, which is influenced by development and contextual characteristics, shaped by disease,
and interpreted by the caregiver and individual child” (p. 158; De Civita & Dobkin, 2004). This definition captures the fact that many personal, environmental and disease related variables influence pediatric adherence and the interaction of these variables is multidimensional and dynamic (De Civita & Dobkin, 2004). Thus, similar to a family systems approach, they suggest that adherence needs to be understood through the ongoing interactions between the caregiver, child, and health care team, which they refer to the triadic partnership.

Understanding a dynamic, multidimensional construct is difficult as no single source can capture all aspects of it and many factors need to be examined. According to this model, a child’s adherence will fluctuate with changes in disease course, environment (e.g., divorce, family’s resources, contact with medical team) and developmental level (De Civita & Dobkin, 2004). There is some evidence that adherence declines over time for both chronic and acute illnesses (Rapoff, 1999). Additionally, in the past, adherence was conceptualized as categorical and one-dimensional, meaning that children were either adherent or nonadherent and that the rate of adherence was consistent across and within treatments. However, current research has provided evidence for multiple variables and barriers that contribute to adherence (De Civita & Dobkin, 2004). For example, families may participate in “adaptive nonadherence” (Deaton, 1985 in Rapoff, 1999) where they balance maintaining a quality of life with managing the health needs of the ill child by varying adherence levels based on effect, cost, inconvenience, discomfort, and time (Rapoff, 1999).

De Civita and Dobkin’s (2004) inclusion of developmental level especially demonstrates the complexity of understanding adherence in children, as the concept of
development includes many elements such as cognitive ability, social interactions, emotional resources and physical skills which are constantly changing (De Civita & Dobkin, 2004). For example, most children progress from infancy, where they are completely dependent on the parent to be responsible for adhering to the treatment regimens, to adolescence, where roles and responsibilities are adjusted as they acquire more skills (De Civita & Dobkin, 2004). In fact, adolescents appear to be less adherent than younger children, which may be explained largely by the changing of roles (La Greca, 1990). Parents are the most knowledgeable about treatment regimens and when adolescents become more responsible for their treatments parents may become less involved (La Greca, 1990). Furthermore, the combination of changes in physical growth, hormones, mood, perceptions of risks, problem solving and decision making skills, importance of peers and parent-child relationship create specific challenges to understanding adherence behaviors in adolescents (Krasennegor, Epstein, Johnson, & Yaffe, 1993). Thus, poor adherence cannot be solely related to refusal of a treatment regimen as other more complex factors such as developmental level, the parent-child relationship and interactions between the family and medical staff need to be considered (De Civita & Dobkin, 2004).

Purpose of Study

In conclusion, interventions to improve CPAP adherence in children are greatly needed; however there is limited research to help guide the development of such interventions (Marcus et al., 2006). Furthermore, increasing adherence to CPAP therapy in children appears complex and designing successful interventions most likely requires targeting several barriers at different levels (e.g., child and their environment). Therefore,
the purpose of this study was to systematically examine the experience of caregivers and their children who were using CPAP therapy to treat SDB and to examine barriers to treatment adherence through multiple perspectives and systems (i.e., caregiver, child and medical team). In addition, the study aimed to provide data that would improve care received by families being served by the Sleep Disorders Center (SDC) at Cincinnati Children’s Hospital Medical Center (CCHMC).

Method

Participants

Caregivers and their child who initiated CPAP for PSG confirmed SDB were recruited from October 2006 to April 2007 at CCHMC. Eligible children were starting CPAP for the first time and met at least one of the following two criteria for SDB: 1) an apnea hypopnea index (AHI) of greater than 1.00 or 2) more than 10% of sleep is spent with carbon monoxide levels greater than 50. Children also needed to have been between the ages of 2 and 17 years at the time of participation. Children were excluded if they had a tracheotomy. Twenty eligible families were identified during recruitment; however, two families refused because they could not commit to the follow-up caregiver interview and to the child focus group scheduled for the spring. Three families also consented and then withdrew prior to any data collection; reasons included the family’s decision not to use CPAP anymore, the child becoming ill, or the parent becoming ill. Thus, the total family sample consisted of 15 caregivers (including a father, a grandmother, an aunt and twelve mothers) and their children who were using CPAP as a treatment for SDB. The children ranged from 4 to 17 years ($M = 13.1, SD = 3.42$) with 47% being African American and 67% being male. All children had OSA with 40% being severe (AHI > 30), 47%
moderate (AHI = 15 to 30) and 13% mild to moderate (AHI = 5 to 15). There were 67% of children who had additional medical conditions (e.g., seizures, asthma, hypertension). Furthermore, 53% who were diagnosed with obesity and 33% with a developmental delay.

The medical team responsible for treating the children also participated in the study. There were a total of nine clinicians who took part in the focus group, including a visiting physician, three attending physicians, two fellows, two nurses and one nurse practitioner.

Procedure

All families who were referred to the SDC at CCHMC for SDB were considered for this study. Eligible families were determined by examining the results of all overnight diagnostic PSG studies completed at the SDC. Families were approached for participation in the study when they attended appointments at the SDC, usually when they received the results from their diagnostic PSG. If the family expressed interest, parental written consent and verbal assent for children ages 6 to 8 or written assent for children ages 9 to 17 were obtained. The first caregiver interview was also scheduled at this time. Caregivers were provided $20 cash for each interview that they completed and children were provided with a $20 gift certificate from a retail discount store for their participation in the focus group. Data were collected at three times. Procedures consisted of interviews, focus groups and supplementary data (see Table 1). The research was approved by the CCHMC and UC institutional review boards.
Table 1

Summary of Data Collection Procedures

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<tr>
<th>Data Collection Procedure</th>
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<td>Child Focus Group</td>
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<td>Objective Adherence Information</td>
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^ After at least three nights of CPAP use and within the first three weeks of starting CPAP.
* Within three to four months of beginning CPAP
~ The point at which the entire sample has been recruited (after April 2007 and before August 2007).

Caregiver Semi-Structured Interviews

Individual semi-structured interviews were conducted at two times with caregivers. The intervals between interviews were chosen based on evidence that initial barriers to adherence (i.e., within first few weeks) are often different than the barriers experienced by families after a longer period of CPAP use (Marcus et al., 2006).
Both the Time 1 and 2 interviews were conducted at CCHMC and audio taped for subsequent analysis. The interviews followed a broad topical outline with open-ended questions developed from relevant literature on SDB, the CPAP process, pediatric adherence and information gathered from a previously conducted retrospective chart reviews. The interview outlines were reviewed by all the members of the research team, as well as an additional health professional who was knowledgeable with pediatric sleep disorders and their treatment to increase the validity and appropriateness of questions (see Appendix A and B). I conducted all interviews.

The Time 1 caregiver interview took an average of 56 minutes ($SD = 19$) and occurred after at least three nights of using CPAP and within three weeks of initiating CPAP ($M = 14$ days; $SD = 3.8$). The interview at Time 2 took an average of 33 minutes ($SD = 6$) and occurred within three to four months of beginning CPAP ($M = 104$ days, $SD = 12.7$). All caregivers, except for one who could not be located for follow-up, completed both the Time 1 and 2 interviews (93%).

**Focus Groups**

*Child Focus Groups.* Several focus groups with the children examined their experiences of being treated with CPAP and identified barriers to adherence. Based on recommendations in the literature describing the use of focus groups in pediatric populations, children under 6 were not included, nor were children with a developmental delay that would impede their ability to interact in a group setting or communicate feelings and opinions (Downey, Perkin, & MacQuarrie, 2000). Thirteen of the 15 children were eligible to participate in a child focus group. However, one eligible adolescent male was not able to be contacted as his family was lost to follow-up. Of the
two children who were ineligible; one was under the age of six and the other did not have adequate language skills. Based on the demographics of the sample (i.e., age, gender and developmental level) and the needs of the families, there were seven child focus group sessions conducted. Of the seven sessions, four were run with only one child and the facilitator. These smaller sessions were scheduled to accommodate families who expressed interest in their child having the opportunity to discuss their CPAP experience, but could either not attend the larger group sessions due to a schedule conflict (i.e., male age 16 and female age 15 with a developmental delay) or did not have a larger demographically similar group (i.e., female age 16 and male age 11 who had a developmental delay). Of the larger focus group sessions, one involved two adolescent females ages 14 and 17 who both had a developmental delay. The other two groups involved adolescent males who were 12 to 15 years old with one having four participants and the other having two. Thus, every child, except for the adolescent male who was lost to follow-up, participated in a focus group session.

I facilitated all focus groups. Parents were not present during the sessions to increase productivity (Downey et al., 2000). However, parents were asked to stay in the waiting room for the duration of the group so that if their child should feel uncomfortable the facilitator would be able to find the parent. The facilitator also offered time, prior to beginning the focus groups, for parents to express any concerns or ask any questions. No children or parents expressed discomfort about the process. Focus groups followed an agenda, which began with a review of the meaning of confidentiality, the voluntary nature of the study, and what the children could expect, all of which were discussed in developmentally appropriate language. The agenda also included the introduction and
discussion of group rules, visually displayed. The rules included only use first names, wait until other person stops talking before they can talk, and if uncomfortable with something let the facilitator know. In addition, broad developmentally appropriate questions were prepared to help facilitate group discussion such as draw a picture of their CPAP machine or describe how they use it, express what this experience has been like, what would they say to another child who was initiating CPAP, and what could CCHMC do to help make things easier. Finally, the focus group agenda also included a debriefing session at the end where the children could discuss their thoughts and feelings related to participating in the project (see Appendix C). For children in the smaller focus groups, the facilitator answered questions about the experience of other children using CPAP. All groups were audio taped for subsequent analysis.

Medical Team Focus Group. A focus group was used to examine the experience of the medical team who were treating the children using CPAP. The focus group was held during one of the medical team’s weekly conference times. Members of the medical team were informed two weeks in advance so that they could have the option of not attending the focus group if they felt uncomfortable with participating. Written consent was obtained from each participant.

Similar procedures to the child focus groups were used. The session was audio taped and an agenda was followed that included broad questions and a debriefing at the end. The medical team completed a “treatment barriers exercise” that involved the team generating as many barriers to children using CPAP as they could and subsequently rating the barriers from most to least impactful. Next, the team members proposed solutions or interventions for overcoming the treatment barrier in order to increase
adherence. Finally, the team was engaged in a discussion about the services provided to children prescribed CPAP. The treatment team was asked to discuss their perceptions regarding what processes and procedures were working well and what the SDC could do better when treating families (see Appendix D).

**Supplementary Data**

*Clinician rating form.* The clinician responsible for the care of each participating family was asked to complete a rating form at two different time points corresponding with the caregiver interviews. At Time 1 the clinician was asked to predict the family’s overall level of adherence on a 7-point Likert-type rating scale (i.e., no use to very good), that was defined at each point based on the average number of hours used each night and nights used per week (i.e., very poor is less than two nights per week with less than 5 hours per night on average). This form also asked the clinician to list the reasons for their predictions (i.e., weaknesses or strengths of the family; see Appendix E). At Time 2 the clinician was asked to rate the level of current overall adherence on the same rating scale. The form also required the clinician to answer whether the level of adherence had changed over the course of treatment and if there was a change, state the direction. In addition, the clinician was asked to check off any barriers from a list. There was also space to add any additional barriers not provided (see Appendix E). All rating forms were completed by a nurse practitioner, except in one case where the families’ care was being managed by a sleep medicine fellow.

**Chart Review**

A chart review was completed for each child to gather information on demographics (i.e., age, gender, race), disease related characteristics (i.e., OSA severity),
other diagnoses (i.e., health conditions and developmental delay) as well as dates and types of services provided by the SDC.

**Objective Adherence Information**

Most CPAP machines have an electronic counter which provides the number of hours it has been used. Caregivers were asked to provide the number at both the initial and follow-up interviews.

**Analysis**

**Interviews and Focus Groups**

When determining the appropriate method of data analysis it is best to examine the purpose of the study and the questions that the researcher aims to answer (Strauss & Corbin, 1998). The purpose of this study was to systematically examine the experience of caregivers and their children who were using CPAP therapy to treat SDB and to examine barriers to treatment adherence through multiple perspectives. In addition the study aimed to provide data that would help design effective interventions to promote CPAP adherence and to improve care received by families being served by the SDC at CCHMC. This study would be one of the first to systematically examine the experience of caregivers and their children who are using CPAP.

Qualitative methodology is the best choice when minimal information is available regarding a specific area of study, as questions can be clarified and a higher level of detail can be obtained (Heary & Hennessy, 2002). Furthermore, qualitative methodology does not rely on pre-set categories by the researcher, as participants are not forced to choose how they feel by selecting from a limited amount of options (Weiss, 1998). They
have the opportunity to freely express their perceptions, which allows for the possibility of discovering new or unique information regarding an area of study (Weiss, 1998). In addition, quantitative measures are static and usually only provide the *what* of a behavior (Heary & Hennessy, 2002). They typically do not provide the *how* and *why* (Heary & Hennessy, 2002). Thus, qualitative methods are better suited for studies such as this one that are concerned about developing theory rather than testing it, about service delivery, and about identifying specific barriers for future interventions because the salient components involve understanding the process or experience perceived by the families rather than measuring a quantity or intensity of a behavior (Denzin & Lincoln, 2005; Straus & Corbin, 1998; Weiss, 1998).

Finally, sampling procedures for quantitative measures usually require large sizes and random assignment so they can use inferential statistics and generalize the findings to other families (Nastasi & Schensul, 2005). Unlike quantitative methodology, smaller sample sizes are appropriate, even desirable, when working with qualitative methodology (Weiss, 1998). This is because sampling methods are guided by the research question and by efforts to represent the population in their normal context (Nastasi & Schensul, 2005; Strauss & Corbin, 1998). Thus, randomness and large samples are not necessary as the purpose of the sampling is to obtain an in-depth understanding rather than a generalization (Nastasi & Schensul, 2005; Strauss & Corbin, 1998).

Based on these considerations, it was determined that the constant comparative method for producing emergent-grounded theory would be used to analyze the semi-structured caregiver interviews as it is a qualitative technique that provides systematic procedures for building theory and examining experiences (Glaser, 1992; Glaser &
Data analysis was carried out separately for the initial and follow-up interviews, with comparisons between emergent themes made later. Each transcript was read twice before all caregiver statements were condensed and coded. Coded statements were systematically compared to identify categories, and those that occurred repeatedly within a single transcript were considered emergent themes. In keeping with the constant comparative method for producing emergent-grounded theory, the same systematic process was reiterated for analysis of each transcript (Glaser, 1992). Once all transcripts were analyzed, the categories and themes from each transcript were systematically compared to those from subsequent transcript(s). While new themes could arise in any individual transcript, only those themes supported by data in successive transcripts were maintained and allowed to expand. As the continuous process of coding and comparing occurred across and within transcripts, group themes emerged. The sorting, coding, categorizing, and constant comparison was documented so an audit trail could be reviewed by another member on the research team for a validity check (Glaser, 1992; Glaser & Strauss, 1967). NVivo, a software program for handling qualitative data, was used to assist with the coding and comparison process.

Methods similar to that described for the individual interviews were also used to analyze the data for the focus groups. Emergent themes were analyzed separately for each focus group and then compared.

Supplementary Data

Descriptive statistics (e.g., frequencies and percentages) were used to summarize and analyze the supplementary data including the objective adherence data downloaded from CPAP machines, information obtained from the clinician rating forms (i.e.,
number/type of barriers, clinician ratings of adherence), demographic information (i.e., age, gender, race), disease related characteristics (i.e., OSA severity), diagnoses (i.e., other medical conditions) and types of services obtained by the SDC (i.e., number of follow-up appointments).

Results

Caregiver Interviews and Child Focus Groups

Theme 1: Adherence is Difficult

Adherence to CPAP in children with sleep-disordered breathing is fundamentally difficult. Almost all children and their parents reported marked adjustment difficulties and a fairly high level of discomfort when using CPAP. At the initial interview one caregiver stated “it is a struggle every night trying to get him to use it and some nights we no more than leave the room and he has it off.” Another parent described the first night as “very challenging, because my son took it off, was crying and throwing a fit and I couldn’t get him to go back to sleep unless I just left it off.” Furthermore, some parents still reported adjustment difficulties at the follow-up interviews that were at least three months later. For example, one caregiver of an adolescent female stated that “she does not like the mask feeling and she still got to get used to it. I mean she ain’t scared of it, she will put it on her face, but it is seems like she still got to get used to it.” Another parent noted that “I think she is warming to the idea although I just think you know if we can just get it to stay on all night, that will be a miracle.”

In fact, several caregivers reported feeling surprised at how difficult the therapy seemed to be for their children. One mother declared “my daughter has never had any problems with anything before. We have been through quite a few surgeries and medical
things and nothing has been as big of a deal compared to this.” In addition, many children reported that using CPAP was harder than they had expected it to be and that it was a long ongoing process. After wearing CPAP for three months, one adolescent male stated “I don’t think I will ever get used to it.” Furthermore, both parents and children often identified experiencing a high level of frustration related to CPAP use. All children in the focus groups agreed that the most frustrating aspect of CPAP therapy was being “uncomfortable” and “having difficulty sleeping.” Parents also reported lack of sleep for themselves. Moreover, most caregivers endorsed that a high level of patience, effort and energy was needed by them throughout the adjustment period. For these reasons, caregivers often described the CPAP experience as “stressful,” “a lot of worry,” and “negative.” During an initial interview one parent described her role throughout the process:

   Oh! Lot of cajoling, a lot of “you know we have to do this,” “you know you have to wear this because it will really help make you feel better and you will have so much more energy at school.” “You will feel so much better.” “It will help you really get a good night’s sleep.” It is just she finally relented and you know wore it, but I think for the first couple of nights my husband and I took turns getting up and checking on her and I think it took us a couple of nights, you know, we would go in and whether it came off or it fell off or she took it off I don’t know, but we did have to put it back on.

Another caregiver expressed his involvement at the follow-up interview:
I am being patient and consistent with him, that is all I can do, just to get him to do it. Eventually I am sure he will do it on his own. It is just getting him comfortable you know and once I can get him comfortable and settled then I am sure everything will work out all right.

Finally, when parents and children were asked to give advice to other families who were initiating CPAP, their statements consistently highlighted the difficult nature of using CPAP. For example, phrases were used such as “stick with it,” “keep trying,” “try it and see,” “long process,” “family time commitment,” “expect battles and periods of resistance,” “be prepared for ups and downs,” “time will tell,” and “it will take time and patience.” During a follow-up interview one parent recommended to “be prepared, as it is not just going to be something they are going to put on and be jolly, expect resistance.”

**Theme 2: Adherence Fluctuates and is Multi-Dimensional**

CPAP adherence in children has **two components and is not static.** The findings highlight the importance of including both number of nights used and hours used each night when defining good CPAP adherence. Both children and parents reported that using CPAP all night is much more challenging than using it every night. Many parents stated at the follow-up interview (3-4 months after initiating CPAP) that their children were not able to use their CPAP all night. Similarly, many children reported that they started out each night wearing the mask but then removed it before morning. Some took it off because it became too uncomfortable while others did not want to replace the mask after getting up (e.g., washroom) as it would take longer to fall back to sleep. The following description from a caregiver captures the multi levels of CPAP adherence:
It’s keeping it on that is the problem. And I think lately he has been going to sleep with it on. The problem is keeping it on all night and if we just find something that would help him keep it on all night then I think it will be much better. I mean, my husband goes in there before he is going to sleep to make sure to see if he got it on, but the problem is, he is just not keeping it on all night once he goes to the bathroom.

In addition to having two components, adherence is not constant. A child’s level of adherence can change over the course of CPAP treatment in either direction. For instance, some children who previously had established high levels of adherence experienced difficulties after a cold, a vacation, or pressure change. Furthermore, fluctuations in motivation to use CPAP, whether in the parent or child, were frequently noted to impact adherence. One parent described changes in her son’s adherence levels over the course of treatment “it has been ongoing, he was real good about it at the beginning every night and never forgets. Of course I was in there every night putting it on him. Now he is real lacksadaisy about it.”

**Theme 3: Two Key Challenges to CPAP Adherence**

*Child adjustment and parent monitoring were endorsed as the most common and biggest challenges to CPAP adherence.* Physical side effects, cost and regimen complexity were not frequently specified as challenges. Given the high level of discomfort reported, it is not surprising that many parents stated the biggest challenge of CPAP therapy is getting their child to use the machine on a consistent basis. Similarly, many children reported that “getting used to it” was the most difficult challenge when using CPAP, making consistent use arduous. At the follow-up interview parents were still
using phrases such as “she has just got to work with it and get used to it,” “he is still having a hard time using it, he still is having some difficulty going to sleep.” When reflecting on her child’s adjustment difficulties one parent stated:

I think it is just one of those things that over time hopefully it will get better. She will finally get to point where she will say “okay this damn thing is here to stay, I might as well get used to it.” I don’t know, maybe, you know maybe not, but I don’t know, I guess time will tell.

Parents also reported barriers to providing a high level of monitoring, especially over time. For instance, some parents noted their own sleep deprivation, increased tension between them and their child and exhaustion of their emotional resources contributed to feelings of wanting to give up. In addition, many parents reported that it was difficult to set limits and encourage their child to use CPAP when they seemed to be so uncomfortable and frustrated. For example, one parent commented “to see your son going through the challenges and the frustration, that has been the hardest. And you know you don’t want to be arguing with him at midnight about the machine.” Another caregiver expressed “I have ran out of energy the last couple of weeks. I have invested a lot. I thought I had invested plenty of time.” In addition, one parent noted “it is hard for me, a lot of times I end up going to bed before she does because I am just so tired.” Finally, one caregiver stated “I would say the biggest challenge of CPAP therapy has been my husband and I dropping the ball.”

Parents and children who viewed CPAP use as something that had to be done regardless of the discomfort and effort seemed to experience more success. Parents who approached the therapy in this way, often compared CPAP to a “job” or “chore.” For
example, one caregiver explained “it is a job, you got one job but it is like you got another job all over again. You know you got to do this for your daughter, so it is just like your second job. There is no choice.” Those children in the focus groups who achieved higher levels of adherence declared that to be successful one had to make a commitment and “you have to make the decision to use it.” In contrast, families who perceived CPAP as optional seemed to be less successful. One parent reported that “I think if it were a question of life and death, I don’t make as much of an effort as I should perhaps because of that, because it isn’t.”

**Theme 4: Parental Involvement is Essential**

*Ongoing persistence and monitoring from parents is important for the success of CPAP use, regardless of the child’s age.* Children in the focus groups reported that for the implementation of CPAP to be successful, even for older children, there needs to be a high level of parental monitoring and support. This is because it is difficult to be motivated to use a therapy that is so uncomfortable. In fact, one adolescent commented that the parent’s role is to “help with everything in the beginning.” Similarly, many parents, including those of older children, reported that if they did not monitor or support their child in using CPAP, their child would not use CPAP at all. For example, one caregiver noted “I think probably if it wasn’t for me asking him those times like I do it probably wouldn’t have been even three times a week that he sleeps with it.” Another parent described the role he plays in his adolescent son’s treatment:

> I said, “Are you using it?” and he said, “Yeah, I am using it” and then I waited a couple of days or so and then I went in to check on him, just you know to see if everything is all right, as I usually do check on him, and I
noticed the mask wasn’t on, so I woke him up. I said, “Why ain’t you got your mask on.” He said, “Well, I am not comfortable, I can’t sleep with it on.” I said, “But you need to keep trying you know and maybe you just need to get used to it.” He said, “It is uncomfortable.” I said, “We will just try and put it back on.” I went out of there and left him alone, waited another couple of days again and went in and checked him again. He was doing the same thing. I just talked to him. I just waited till the next day and sit down and talk to him, you know, “Why aren’t you wearing the mask,” and he says, “I am just not comfortable.” So I just wanted to take him back and try to get him fitted for another mask. Hopefully, that will work you know or just keep going until we get the right one, that is all I can do.

In fact, many parents who provided substantial monitoring and support felt this increased their child’s level of adherence. For instance, when one parent reflected on reasons for her child’s success she stated:

Obviously, it’s a no-brainer. It’s the parental support. I think the parent has to be really involved in CPAP therapy for it to be successful or at least to have a good chance of being successful.

Caregivers described providing support through “checking,” “enforcing,” “encouraging,” and “motivating.” For example, one parent noted that she “helps him to understand that this is for him for his health and this is something that he got to do. He might not like it, but he got to do it.” Other parents made similar comments such as “keeping him
interested and keeping him aware of his health” and “convincing my son that this is good for him.”

Furthermore, some parents at the follow-up interview reported that their child’s level of adherence would likely be higher if they were more involved or noted that adherence levels increased when they became more involved.

It was not getting used when we left it up to her. Definitely a parental push to make sure that it’s getting done was needed. We are still working on, you know, some things but it is better than it was and I think that is an improvement.

Another caregiver stated:

I think the biggest problem right now is, I think between my husband and I, we just have to get better about making sure that she’s, you know, she has got it on, because it is not one of those things it appears that is going to happen on its own, that she will take the initiative. I just don’t think she is.

Finally, parents often reported that they needed to provide the highest level of monitoring and support when initiating CPAP therapy and when new challenges presented themselves throughout the process. For instance, most parents reported that they had to initially sleep in their child’s room or have their child sleep in their room.

Another caregiver described how her son’s motivation for using CPAP varied over time requiring her to provide additional emotional encouragement by “relaying positivity and being more supportive when he is going through those frustrated times, those valleys.”

Finally, one parent shared the approach she felt caregivers should use when beginning CPAP therapy with a child of any age:
I think there comes a point where you are the parent and you know what is best, so you got to put your foot down. The kid only has so much flexibility and then it is kind of like okay, but this is the way it is and this is what you have to do.

Theme 5: Perceived Treatment Efficacy is Important

Parent and child report of a positive experience with CPAP therapy was largely related to perceived treatment efficacy. Few children in the focus groups endorsed an overall positive experience with using CPAP. However, those that did stated it was related to a noticed improvement in their health such as losing weight, a reduction in headaches or having more energy. Furthermore, many parents reported that perceived benefits of using CPAP was the “most important thing” or “most helpful” in facilitating adherence. One caregivers’ description of her experience at the follow-up interview demonstrates the impact believed treatment efficacy can have on attitude towards CPAP therapy:

It will take a lot of effort; you know, keep coming back and forth to the doctor but it is worth it, it is positive. I see him, you know, talking more and interacting with his family more, not staying off to himself. He is asking questions, laughing, getting in a conversation, you know, whereas before he would just stay off to himself in his room, sit there looking like he could barely hold his eyes open.

Adding further support for the importance of perceived treatment efficacy, many families reported their feelings towards CPAP therapy fluctuated with perceived effectiveness. For instance, some parents reported a neutral or negative feeling about
CPAP at the beginning but then experienced positive feelings when their child showed improvements in their symptoms. One parent reflected on the change in her emotions over the course of treatment:

Encourage your child to use it the proper way and to give it some time and realize that if he or she uses it the proper way that he or she will benefit from it. They will improve in their behavior, in their sleep, in their studies, in their overall personality, social skills will develop better, so that is what I think. CPAP has become positive to me.

On the other hand, some parents described feeling positive and hopeful when first initiating CPAP therapy; however, they became frustrated and discouraged when their child experienced ongoing difficulties adjusting and minimal benefits to their health. Furthermore, children and parents who noted a neutral or negative overall experience often stated this was because there were no perceived benefits or changes in symptoms. In fact, both parents and children who did not see health advantages at the follow-up interview (3-4 months) where often questioning whether the frustration and time commitment required for CPAP was worth it. Several children in the focus groups endorsed “I do not see why I have to use it as it is not helping.” One parent commented:

Yeah, the most important thing would be for him to wear it every night, for us to see results. Since we are not seeing results I think the importance, my son doesn’t see the importance of wearing the machine. If he saw results like, “Oh well I feel a lot better.” I think he would have no problem with me reminding him, but since he is not really seeing anything, he is thinking it is a waste of time.
Of importance, many families could not identify any benefits within the first two weeks of using CPAP, which was likely due to the fact that good adherence levels were yet to be reached. For example, one caregiver reported:

Well at this point it is kind of hard to say because she is not keeping it on all night, so I haven’t really been able to see what the benefits are. I did hear from one of the teachers this past week at school that she did seem to be tired and I said well we are still working the kinks out with this, so it may take us a while. You know, I guess in my mind I had hoped it would have been on all night and we would have gotten the benefits immediately and I think if we have it on all night every night we would have got the benefits immediately, but we were having a hard time with that.

In addition, at the initial interview many parents responded with comments such as “too soon to tell,” “don’t know,” “can’t say,” “I know it would be effective if I can get him to use it,” “it has got benefits if you can get your kid to sleep with it,” and “I believe it can be a lot more effective if he uses it properly”.

Theme 6: Confusion about Treatment Benefits

Many families were confused or unclear about expected outcomes and benefits of CPAP use. In the study sample, there was much variation in presenting symptoms of SDB and the impact on daily functioning. Some families reported significant daytime fatigue while other children were more asymptomatic and families were not sure how to perceive effective treatment. For instance, one parent stated:

I mean, they say she has severe sleep apnea, but I have never, I mean I have slept with her like on vacation and stuff and I don’t hear her, you
know wake up or anything like that. She has no daytime fatigue or
gasping.

Most of the children in the focus groups stated that they did not know what to expect with CPAP use or what improvements they should see in their health. Many could not articulate why using CPAP was important or what would happen if they did not use CPAP. In addition, many had questions regarding how long they would have to use CPAP therapy. Moreover, some families reported inaccurate benefits. For instance one child had insomnia and thought using CPAP would help him fall asleep quicker but was disappointed when he learned it was more difficult to fall asleep.

Of note, both children and parents often expressed that they received adequate information about using the actual CPAP machine but not enough about how adhering to CPAP is a long ongoing process and benefits may not be immediate. During the focus groups, children also voiced a need to receive more details about realistic expectations for improvements in their health and when these changes would begin. Further limiting the flow of information, few families shared their experiences or observed benefits with each other over the course of treatment. For example, at the follow-up interview some parents noticed positive changes in their child’s mood or behavior but did not say anything. In addition, other caregivers were unsure if their child experienced benefits from using CPAP as the topic had never been discussed. The ensuing comment by a caregiver at the follow-up interview highlights the poor communication within many families about CPAP therapy:

I don’t know the answers to what you are asking. I am going to have to ask him some questions just for my own understanding such as “How well
have you really adjusted to that?” “Do you notice any change in your energy level, in your concentrating?” “How do you feel now that you have been using this?” and “Do you notice any change in the way you are feeling?”

**Theme 7: Good Family Satisfaction with the SDC**

*Overall, families had a positive experience regarding the services they received through the SDC at CCHMC.* Almost all parents expressed a positive experience and satisfaction with care at the initial interview. For example, statements included “I don’t want to change anything I think they are doing their job,” “they have been very responsive to my needs,” “they are knowledgeable and helpful in solving problems,” “they know what they are doing,” “they have been very accommodating” and “I am grateful and comfortable with their services.” The few unsatisfactory comments that were mentioned surrounded the amount of education provided by the SDC. One caregiver stated “they should have explained it to us, they should have had a CPAP machine, showed it to us and told us what to expect, they didn’t do too good of a job getting us ready for it.” Similarly, another parent commented that “they should let you try it out in the office once you are told that you will need it, so it won’t be a shocker of exactly what you are getting and what you are going to be doing.” Finally, some children in the focus groups thought a “warning” would be helpful that stated “children using CPAP experience struggles that come and go.”

Although experiences were still fairly positive at the follow-up interview (3 to 4 months later) some parents expressed concerns about the timeliness of services and the
level of follow-up. In relation to service delivery, one parent noted “sometimes the appointments are a little bit way out” and another stated:

I think that is my biggest gripe is that it just seemed to have taken so long to get from A to B. It just seems like between the doctor ordering the test, getting the test done, reading the test, getting back to the doctor, taking the next step, it just seems to be an excruciatingly slow process.

The level of parent reported satisfaction with follow-up by the SDC was mixed. Some were disappointed; one parent said “probably a little bit more phone contact or either sending out a letter saying it has been couple of months and I just like to talk to you.” However, others were satisfied as one caregiver noted “they definitely follow through because when I forget to make an appointment, they definitely get a hold of me,” while another stated “they don’t let things slide by.”

**Theme 8: Limited Bond with the SDC**

Many families did not seem to be strongly connected to the SDC, despite reporting positive interactions with staff. Few parents knew who was treating their child’s SDB or how to contact them. For example, one caregiver stated “Everybody has been very nice. It is just that I am having trouble with names because we are seeing so many different people and so many different departments.” In fact, caregivers often referred to the connection they had with other medical professionals that have been involved with their children for many years. Furthermore, when asked to describe their relationship with the SDC, some parents responded by saying that they had not had enough contact with people from the SDC to describe their interactions as a relationship. For instance, caregiver statements included “I don’t know them really, we haven’t really
had too much conversation,” “I have only talked to them five times, so I mean I don’t really have a relationship,” and “I don’t think they really know him.” Of note, those families who reported having regular follow-up through the SDC seemed to feel a stronger sense of connection. For example, one parent described her relationship as “you are part of that team and you have information to give and they have information to give.”

Theme 9: Follow-up is Challenging

Consistent follow-up is challenging in this population. Despite significant efforts by the SDC, most parents reported minimal or no follow-up at the time of the 3 to 4 month interview. Some parents reported confusion related to follow-up and were not sure if they had another appointment or who was supposed to initiate contact. Many families indicated that they did not have a follow-up appointment scheduled because they were waiting for the SDC to contact them. For instance, “they were supposed to call me right?” “don’t know, maybe we could check on that one before I go?” and “I think they were supposed to follow-up with me but I’m not sure” were common responses. Conversely, other caregivers acknowledged contact by the SDC but attributed poor follow-up due to negligence on their end. One parent stated “No. You know, she called me too. I need to call her.” Another responded:

No. Wait a minute. I think they called me. They called me last week or was it this week? They called me one of these weeks and before I could call them back or whatever, my son accidentally erased the message, so I got to call them back. I think she does have an appointment with them, but I can’t remember what day it was.
Finally, another caregiver declared:

The nurse called me and left a message about maybe a week and a half ago and I had the paper and I had the number, she left the number well you know, when they called, but I don’t know if it is in my saved messages or not. If it is, then I am going to call her when I am at home. If not, I am going to see if I can just call over to get her name. I know her first name, but I hope there is not more than one nurse with her first name.

Of interest, most of the parents who acknowledged poor follow-through, noted that their children had multiple medical problems and appointments, making it difficult for them to keep track and manage scheduling.

**Theme 10: SDC has No Standardized Care for CPAP Use**

There is no standardized care/protocol for families initiating CPAP to treat their SDB through the SDC at CCHMC. Large variation was observed in families reported experiences. For instance, the time between receiving a diagnosis of SDB, obtaining a CPAP machine and having the titration study was very inconsistent. The average time between the diagnostic sleep study and titration sleep study was 90 days ($SD = 61.7$), with a range from 1 to 230 days. Variability was also noted with the frequency of contact and type of education that was provided to each family.

*Medical Team Focus Group*

**Barriers Exercise**

*Barrier 1: Great disruption to routine.* The team felt that CPAP use is “extensive” and requires much involvement from the family such as taking care of the machine. However, they also believe that current demands already placed on the child (e.g.,
physical and mental) as well as the child’s personality factor into how much of a change in routine is required and what personal resources are available for the change. Interventions that were suggested to address this barrier included: 1) support groups for children, 2) frequent follow-up with the SDC, 3) initiate CPAP gradually, 4) provide psychology support services to families having difficulties, 5) help increase motivation through education, 6) offer extra assistance through the sleep laboratory, and 7) make available options such as hypnotherapy or home desensitization before initiating treatment.

**Barrier 2: Discomfort.** The team recognized that many aspects of CPAP therapy can be uncomfortable such as the mask itself, the constant air pressure or feeling claustrophobic. Furthermore physical complications such as nose bleeds, dry nasal passages, tactile sensitivity, or sinus infections can occur. To address these issues, the team suggested: 1) gradual initiation of CPAP, 2) nasal steroids, 3) regular PSG’s to optimize air pressure, 4) using ramping feature on machines (starts with lower air pressure and advances to optimal setting over time), 5) ensuring good mask fit at beginning and considering of custom made masks from Europe, 6) scheduling follow-up soon after starting, 7) using humidification feature on machines, and 8) efficiently treating infections.

**Barrier 3: Lack of comprehension by the family.** The team observed that many of the families had minimal understanding of CPAP therapy, particularly about the process, the resources involved, and the changes needed for CPAP to be successful. To deal with this, the team recommended standardization of the family education process. The information should be ongoing and include reinforcement components at subsequent
appointments that occur on a frequent basis. Education materials should include more practical demonstrations ("show and tell") and need to be customized to literacy level of the parents. An evaluation aspect should also be added so that a family’s knowledge base can be tested. Contact information for support staff should be clear so families know who to call when they experience problems. In addition, a health nurse should be available for home visits when necessary. Finally, it would be helpful to have a website available to families that provided helpful information and education.

**Barrier 4: Parental expectations of therapy involvement.** More specifically the medical team believed parents had limited knowledge of the motivation, investment, and level of commitment that is required by them. To improve parents’ knowledge the team felt the clinic needed to offer an extensive office visit prior to the initiation of CPAP where substantial education could be provided about SDB complications, the importance of CPAP and the level of investment that is required. During this visit, short and long term goals should be clearly stated and unrealistic expectations discussed. Furthermore, frequent contact is required so that trust can be established between the family and the treating team.

**Barrier 5: Variability in parenting skills.** The medical group noted that the current level of structure in the child’s life (e.g., sleep hygiene) is important to consider. For instance, in families where parents struggle to set limits, implementing CPAP therapy is likely to be challenging. The team proposed that frequent follow-up in the home by a nurse or social worker would be helpful for these families. They also expressed that family education, support groups and the involvement of extended relatives may be helpful.
Barrier 6: Insurance and equipment difficulties. The group thought that the wait time between when the child was diagnosed with SDB and when they received their CPAP machine was often too long. It was suggested that the CCHMC could provide the CPAP machine, or at least “loaner equipment” to ensure standardization and efficiency. If not possible, the medical team proposed that frequent contact with the family during the waiting period might help along with explaining the time line to families at the beginning of the process. Finally, the team acknowledged the importance of submitting proper documentation of medical needs to insurance companies in a timely fashion.

Service Related Discussion

What does the SDC currently do well when treating children with CPAP? The medical team believed they have a strong multi-disciplinary approach and collaborate well with other departments within the hospital such as psychology and otolaryngology. Furthermore, they can offer a variety of quality services by having a specialized nurse (e.g., education) and sleep lab (e.g., interface fitting and sleep studies). They also thought that the center is committed to improve services and to strive for excellence through activities such as research. In addition, the team thought staff were well trained, knowledgeable and highly skilled to manage complex cases. Finally, the group felt that the quantity of patients they can serve through the team approach is an asset.

What does the SDC need to do better when treating children with CPAP? Although they reported many strengths, the team felt the center needs to develop a standardized care plan that includes: 1) improved education for families prior to initiating CPAP (e.g., clinic demonstrations of the equipment and the use of different mediums to provide information), 2) home visits to families, and 3) making the period of time
between CPAP prescription, receiving the machine and doing the titration study more consistent across families. Furthermore, while they would like to see standardization of protocols they would also like to see more personalized family care when problems occur. In addition, the group stated that they would like to expand their collaborative approach to care to include nutrition and cardiology. Finally, the team reported that they would like to increase their understanding of follow-up difficulties and general barriers to CPAP adherence.

**Supplementary Data**

**Objective Adherence Data**

Only 47% of families (7 out of 15) were able to provide the number of hours used as recorded by the CPAP machine at the initial and follow-up interviews. Some machines do not have the capability of recording the hours used. It is unclear, however, whether this occurred for those parents who could not provide the hours or if it was related to difficulties understanding how to obtain the required information. For those parents who reported the recorded use, the average hours at the initial interview was 18.9 ($SD = 39.4$; range 5.8 to 153.5 hours). The average recorded hours used at the follow-up interview was 295.9 ($SD = 675.0$; range 17.9 to 2098.0 hours). Refer to Table 2 for further information.
Table 2

Summary of Supplementary Data

<table>
<thead>
<tr>
<th>Participant</th>
<th>Clinician Rating Form</th>
<th>Objective Adherence Data</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Predicted Adherence</td>
<td>Current Adherence</td>
</tr>
<tr>
<td></td>
<td>Adherence Consistent?</td>
<td></td>
</tr>
<tr>
<td>Male age 15</td>
<td>Very Good</td>
<td>?</td>
</tr>
<tr>
<td>Female age 16</td>
<td>Average</td>
<td>Very Poor</td>
</tr>
<tr>
<td>Male age 11</td>
<td>Average</td>
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</tr>
<tr>
<td>Male age 16</td>
<td>Above Average</td>
<td>?</td>
</tr>
<tr>
<td>Female age 14</td>
<td>Poor</td>
<td>?</td>
</tr>
<tr>
<td>Male age 15</td>
<td>Above Average</td>
<td>Very Poor</td>
</tr>
<tr>
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<td>Average</td>
<td>?</td>
</tr>
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<td>Male age 12</td>
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</tr>
<tr>
<td>Female age 17</td>
<td>Good</td>
<td>Average</td>
</tr>
<tr>
<td>Male age 15</td>
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<td>Average</td>
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<td>?</td>
</tr>
<tr>
<td>Male age 12</td>
<td>Average</td>
<td>?</td>
</tr>
</tbody>
</table>

Very Poor = Uses CPAP less than two nights per week with on average LESS than 5 hours per night
Poor = Uses CPAP less than two nights per week with on average MORE than 5 hours per night
Average = Uses CPAP 3 to 5 nights per week with on average LESS than 5 hours per night
Above Average = Uses CPAP 3 to 5 nights per week with on average MORE than 5 hours per night
Good = Uses CPAP 6 to 7 nights per week with on average LESS than 5 hours per night
Very Good = Uses CPAP 6 to 7 nights per week with on average MORE than 5 hours per night
? = Not Available
Clinician Rating Form

Initial clinician rating form. All children were predicted to use CPAP therapy to some degree. The majority of children (73%) were predicted to display adherence levels of Average and below. Thus most children were expected to use CPAP equal to or less than 3 to 5 nights per week and less than 5 hours per night. Only 13% were expected to use CPAP for 6 to 7 nights a week (see Table 2).

Clinician rationale for predicting Very Poor and Poor adherence levels were: 1) cognitive limitations/minimal understanding of caregiver, 2) several no-shows for clinical appointments, 3) adherence to past treatments has been poor, 4) child has severe developmental delay, and 5) history of nasal congestion. The reasons for Average adherence or better included: 1) good clinic follow-up, 2) family appears motivated/invested, 3) supportive family, 4) child has daytime sleepiness so will feel better on CPAP, 5) long history of successful adherence to medical regimens, and 6) child adjusted well to CPAP in overnight sleep study.

Follow-up clinician rating form. The clinicians could not rate current adherence levels for 60% of families (9 out of 15) because the child was not seen for follow-up by the SDC. Ratings that could be completed were based on parent and child self-report of how many nights CPAP was used per week and on average how many hours each night. Of the six families that attended follow-up appointments, 33% reported Very Poor use (i.e., less than two nights per week with an average less than 5 hours per night), 33% reported Average use (i.e., 3 to 5 nights per week with an average less than 5 hours per night) and 33% reported Above Average or better (i.e., 3 to 5 nights per week with an average or more than 5 hours per night). No families indicated Very Good use (6 to 7
nights per week with an average of more than 5 hour per night). Although the number of nights varied, all but one family stated their child used their CPAP less than five hours per night (see Table 2).

Similar to the initial clinician rating form, the treating clinician was asked to provide reasons for poor adherence or sub-optimal adherence. Reasons included: 1) sinus problems and recurring upper respiratory infection, 2) reported decreased use over time with no clear explanation, 3) refusal to wear mask related to anxiety/fear, and 4) mask removal at night.

Finally, all families who followed-up reported a change in adherence over the course of treatment. Three families noted that adherence levels decreased over time and three stated adherence levels increased over time. Given that adherence levels are not constant throughout CPAP use, it is difficult to predict adherence levels. In fact, of the six families who followed-up, clinicians were not accurate in any of their initial predictions. Three families achieved better adherence and three families reported worse adherence levels (see Table 2).

Discussion

Consistent with other studies, the results show that it is fundamentally challenging for children who are using CPAP therapy to achieve and sustain optimal adherence (e.g., Massa et al., 2002; O’Donnell et al., 2006). In fact, both the families and treating clinicians who participated in this study believe that for CPAP to be successful a large amount of effort is required from the child, caregiver and medical team. Prior to this research project, the most salient barriers and deterrents to consistent CPAP use in children were yet to be systematically investigated. Thus, minimal information was
available for guiding clinicians interested in developing interventions to promote improved adherence in the pediatric population. Based on the findings from this study, I propose several recommendations for promoting CPAP adherence in children. Most importantly, it will be crucial for the SDC and other treatment centers to develop standardized protocols that incorporate several necessary components.

**Recommendations for Standardized Protocol**

*Initial Clinical Interview*

The inclusion of an in-depth family clinical interview that precedes the initiation of CPAP is essential for several reasons. First, the medical team can establish the rapport that is required when dealing with adherence issues. Discussing adherence levels can be daunting for many families because of fear that they will be blamed or seen negatively when treatment goes poorly (Rapoff, 1999). Thus, families must feel secure and safe before they are likely to openly communicate about challenges and seek out the medical team for problem solving support. If fact I agree with other researchers (e.g., Kirk and O’Donnell, 2006) who recommend that each family should have an assigned contact person who is readily available for ongoing support.

Second, relevant information can be learned to assist the medical team in identifying families who may be at greater risk for difficulties in the initial stages of implementing CPAP therapy. For instance, perceptions of SDB severity and symptoms, current family stressors, and past experiences with medical regimens can be investigated. Although factors influencing adherence levels fluctuate, a preliminary analysis can help detect early obstacles. Many established medical regimens have attempted to identify *risk profiles* to increase the accuracy of adherence assessments (Rapoff, 1999). For example,
it has been observed that children and families who have less knowledge about their cancer and cystic fibrosis treatment tend to be less adherent (Rapoff, 1999). Although research pinpointing a risk profile for children using CPAP therapy is in infancy, the findings suggest that families using CPAP are at increased risk for poor adherence when: 1) children are more asymptomatic, 2) families perceive less illness severity and display limited knowledge about SDB and CPAP therapy and 3) parents demonstrate poor skills in monitoring or limit setting. Thus, based on information gathered in the initial interview, treatment protocols can be personalized for each family. For example, some families may only need an educational pamphlet describing helpful parenting strategies for monitoring, disciplining, prompting, and offering incentives whereas others may need training sessions.

Education and Preparation

It is paramount that education be provided to both the child and the caregiver on an ongoing basis. However, it is particularly important to offer information about SDB and the process of CPAP therapy prior to initiating treatment. More specifically, instruction should focus on realistic expectations concerning treatment outcomes and symptom improvement. Parent and child expectations for using CPAP therapy should be ascertained so that unrealistic views can be corrected. For example, it is important for families to understand that almost all children experience adjustment difficulties and discomfort when using CPAP. Furthermore, immediate improvement in SDB symptoms may not occur until CPAP is being used on a consistent basis, which may take time. In addition, parents and children need to be prepared for the challenges that are likely to come and go over the course of treatment. The precedent should be set that regular
follow-up is a necessary aspect of CPAP treatment so that ongoing problems can be discussed and resolved. A standardized education protocol should also include a demonstration of the CPAP machine given that home care companies differ in the quality of explanations that are provided. Thus, over the course of treatment education should be interactive and a combination of verbal discussion, written handouts and modeling utilized.

Finally, it is imperative that both parents and children understand their roles and responsibilities for implementing CPAP. For instance, caregivers need to be aware that much involvement is needed from them, regardless of their child’s age. Parents have an impact on their child’s health beliefs and behaviors at all developmental levels (Rapoff, 1999). In fact, parents and adolescents likely need more assistance in negotiating roles and preparing for the use of CPAP therapy. Medical team’s prescribing CPAP in a pediatric population need to be aware of the unique challenges associated with adolescence and adherence to complex medical regimens. Furthermore, given that most children use CPAP long-term, families will need assistance in modifying parent and child roles to correspond with changes in development.

Ongoing Follow-up and Risk Assessment

Our findings support that it is important to incorporate regular phone or clinic follow-up on an ongoing basis so that adherence can be monitored and the relationship between the family and medical team can be maintained. It is recommended that risk assessments be conducted on a regular basis so that additional support can be provided when needed. Furthermore, frequent follow-up can help capture times of success, providing opportunities for families to receive positive reinforcement. It is also important
to document the factors contributing to good adherence so that they can be referenced if
the family experiences difficulties in the future. In addition, having information about
factors that facilitate adherence can assist with developing preparation and prevention
protocols. Consistent with our recommendations, Uong and colleagues (2008) found a
70% adherence rate in their sample of children and hypothesize that continuity of care,
regular follow-up and ongoing education by a sleep specialist strongly contributed to
their high success rates.

According to adherence theories such as the Children’s Health Belief Model
(Bush & Iannotti, 1990) many families engage in a cost-benefit analysis where perceived
benefits and perceived barriers are compared. Given the high level of discomfort
reported by most children and the level of time and energy that is required by parents the
perceived barriers can be high when implementing CPAP therapy (Bush & Iannotti,
1990). This is especially true if adjustment difficulties and a high level of parent
involvement persists over time. Hence, it is essential to monitor the cost-benefit ratio for
children using CPAP and to focus effort on minimizing the barriers and maximizing the
benefits. For instance, it is important to remove as much discomfort as possible by
ensuring proper mask fit, using the most effective air pressure, having a humidifier, and
utilizing the ramping feature.

Importantly, our results as well as previous studies examining other intensive
treatment regimens show patient and caregiver perceived efficacy as the most important
perceived benefit for maintaining good adherence (Rapoff, 1999). Furthermore, a study
investigating predictors of poor quality of life in overweight children and adolescents
found parental perceived symptoms of OSA correlated with quality of life where as more
objective polysomnographic data did not (Carno, Anson, Kraus, Black, Short, & Connolly, 2008). Thus, it is vital to help each family clearly identify and operationalize their child’s SDB symptoms prior to initiating CPAP. Furthermore, each family should have systematic methods for measuring changes in symptoms so treatment efficacy can be monitored (e.g., teacher rating of daytime fatigue, or child self-rating of energy on likert scale). Moreover, it is crucial to facilitate communication regarding changes in symptoms within families and as part of regular follow-up.

Having said this, it is likely that families of children who are more asymptomatic will need extra support, especially over time. For example, interventions directed at increasing motivation may be needed such as positive incentives for children and extra education for parents about the long-term health benefits of CPAP. Parent or child support groups may also be helpful for this population. In addition, for families that have limited resources it may be beneficial to implement CPAP in stages so that perceived barriers are reduced (Bush & Iannotti, 1990). For example, a single working parent may wish to begin using CPAP only on weekends.

_Diagnosis, Titration and Prescription Process_

To ensure optimal circumstances for promoting adherence it is necessary to have a process that is similar for all families. For instance, the time between receiving a CPAP machine and having the titration study completed should be minimized so that children are using the most effective air pressure. Furthermore, it is highly recommended that only CPAP machines that provide objective data about blower usage be prescribed to families. Many parents, especially of older children, were guessing about their child’s level of adherence. Thus, having access to electronic counters on the machines allows parents to
have an automatic and continuous monitoring tool. Recent technological improvements in CPAP blower meters have led to the availability of units that not only measure blower usage time (i.e., whether the machine is turned on and left running), but also estimates actual usage through sensors that can detect resistance from the user’s breathing. Such measurement could promote greater independence with self-monitoring in adolescents. In addition, it is important that medical clinics have a mechanism for objectively documenting and monitoring adherence levels on a consistent basis. Being able to access comparable objective assessment data across patients will not only improve clinical care but would create opportunities for clinical research. Integrating this objective measurement tool into the treatment will likely be the standard of care in the future.

Collaboration

Given that follow-up in this population is challenging, but is a critical component to success, creative strategies are likely needed. Many children who have SDB also have multiple medical issues that require contact with several different health care professionals. Thus, collaborating with other health care professionals who already have a strong therapeutic alliance with families may improve monitoring of adherence as well as reduce the burden that many families experience with frequent hospital visits.

Program Evaluation

To facilitate the growth of knowledge about children using CPAP, it is recommended that the SDC and other treatment centers include ongoing methods for evaluating the effectiveness of their programs and interventions. For instance, family satisfaction and adherence outcomes are important areas to assess.
Future Directions

It is clear that families implementing CPAP need substantial support. The demands on both the child and parent often exceed internal and external resources, leading to stress and increased negative parent-child interactions. This is even more so for families experiencing high levels of stress prior to initiating CPAP (e.g., financial). Thus, additional research is required to clarify the most effective methods for medical professionals to support both the needs of parents and children while facilitating positive coping. Given that maintaining long-term adherence in pediatric populations is complex, future studies will also need to further investigate the factors contributing to fluctuations in adherence for children using CPAP. Although parent and child perceived efficacy was found to be an important factor in facilitating ongoing adherence, other personal and environmental variables leading to adherence fluctuations in either direction need to be explored using a more diverse population.

The Transactional Model of Stress (Lazarus and Folkman, 1984) may be a valuable framework to guide future research, as it has been applied to various chronic illness groups in attempts to better understand coping and adherence (e.g., Lee & Poole, 2005). The model has gone through several evolutions, including contributions from Thompson and Janigian (1988). A major premise of this model is that the child’s personal factors (e.g., beliefs, personality) interact with their environment (e.g., caregiver-child relationship) to influence their cognitive appraisal or meaning of their illness and treatment (Lee & Poole, 2005). The outcome of the cognitive appraisal in turn influences the child’s emotions, coping reactions and adherence levels (Lee & Poole, 2005). Children appraise their situation on an ongoing basis and coping occurs when the child
views their illness or treatment as stressful and perceives their current resources/support as inadequate (Lee & Poole, 2005). There are a broad range of coping mechanisms and only some will promote adherence. For instance, a child can engage in avoidant coping where they decide to not wear the machine as a means to deal with the stress. According to the Transactional Model of Stress, if parents become overly burdened with demands then they are less likely able to provide the necessary support for their children to have a positive appraisal of CPAP treatment (Lee & Poole, 2005). Thus, when developing interventions, it is important to consider the impact on stress levels for both parents and children. For instance, children who participated in the study expressed that a peer support group would be beneficial. However, given that parents of children with SDB often have multiple appointments, parent and child online chat groups may be more successful in reducing overall stress, promoting positive parent-child interactions and increasing adherence.

Study Limitations

There were several limitations of the study. The purpose of qualitative research is to obtain an in-depth understanding of experiences, rather than to generalize. Thus, it is not problematic for the study aim that all of the participating children, except two, were between the ages of 11 to 17 and all but two of the females had a developmental delay. However, future research will need to confirm the generalizability of the results to the experiences of other families using CPAP. Another weak point of the study involved the composition and timing of the child focus groups. Unfortunately, the demographics of the sample (i.e., age, gender and developmental level) did not work out evenly, leaving some children without a similar group. Furthermore, unlike the caregiver interviews, the child
focus groups were conducted at only one time. In addition, a drawback of the study design is that important demographic variables were not collected for both the family and medical team samples. For instance, years of experience working in the field for medical team members and family social economic status were not obtained. Another weakness involved the clinician rating forms. Although, clinician rationale for predicted adherence levels was obtained, the information was limited. It would have been beneficial to follow-up with each clinician and obtain a more in depth understanding of their reasoning. For instance, “supportive family” was often listed as a reason for good adherence but little is known about what constitutes a family being supportive. Similarly, although follow-up was found to be challenging in this population, factors contributing to the difficulty were not examined in detail (e.g., accessibility to health care). Given the importance of good follow-up future research will need to examine this area in greater depth.

Summary

In summary, this study demonstrates that adhering to CPAP is especially challenging in children and interventions designed to promote CPAP adherence will need to be family-based, ongoing and multi-faceted. Furthermore, given the potential for high emotional costs to CPAP use for both the parent and child, care will need to focus on maximizing the benefits of CPAP use. Although the findings provide some insight into factors impacting and facilitating adherence, future studies are needed to expand on this work, specifically regarding the effectiveness of interventions and care developed based on the recommendations.
References


Appendix A: Interview Guide for Initial Caregiver Interview

**Introduction:**

During this interview, I want to talk with you about you and your family’s experiences regarding your child/adolescent’s sleep-disordered breathing (SDB) and about your experience with the Sleep Disorders Center at CCHMC.

Let’s start by talking about your child/adolescent’s sleep-disordered breathing. There are many different sleep disorders. Some children/adolescents may have more than one.

Can you tell me what specific sleep-breathing your child/adolescent has?
How well do you think you understand what this disorder is (s)?
What has it been like for you and your family since you found out about your child/adolescent’s SDB?
How severe do you feel your child/adolescent’s disorder (s) is?

I. **Diagnosis/Knowledge/Referral Process**

Who referred your child/adolescent to the Sleep Disorders Center at CCHMC?
What were the reasons for your child/adolescent being referred (i.e., symptoms or concerns)?
When did you find out that your child/adolescent had SDB?
Who told you that your child/adolescent had?
What did they tell you about it (i.e., treatment, symptoms)?
What did they have to do to determine that your child/adolescent had a SDB?
How did you feel when you were told your child/adolescent had SDB?
What happened after your child/adolescent was diagnosed with SDB?
How was it decided that your child/adolescent would start continuous positive airway pressure therapy (CPAP)?
Were other treatment options discussed or tried prior to the start of CPAP (i.e., tonsillectomy)?
How well do you think you understand what CPAP therapy is?
How does CPAP help treat the symptoms of your child/adolescent’s SBD?
Do you know what pressure settings your child/adolescent was prescribed?
What did you feel when you were first told about CPAP therapy?
How many times did your child/adolescent have to come to CCHMC and stay overnight for reasons related to their SDB?
What was the purpose of each?
How did these overnight visits go (i.e., behavior and emotions)?
What was the process like for obtaining approval from your insurance company for starting CPAP therapy?
Is there anything else about the referral or diagnosis process that I should know about?
Now that I know about the process leading up to your child/adolescent starting CPAP I want to know about what happened when your child/adolescent actually began CPAP therapy.

II. Initiation of CPAP Therapy

When did your child/adolescent first start CPAP therapy?
How long was this after it was decided that your child/adolescent would use CPAP?

What instructions or information were you given about using CPAP (i.e., when should your child/adolescent use CPAP, cleaning of machine, follow-up with medical team, what to do if problems occur with equipment or with child/adolescent tolerating the therapy)?
Who gave you this information?

When did you first see what a CPAP machine looked like?
Was it the same time as your child/adolescent?

What was your reaction to seeing the device?
Your child/adolescent’s reaction?

Describe what the first night was like when your child/adolescent used CPAP.
What was your child/adolescent’s reaction (i.e., behaviors and emotions)?
Depending on your child/adolescent’s age or abilities, s/he may need a lot of help with using CPAP.

How much help does your child/adolescent need with using CPAP therapy?
Who is responsible for providing this help?

Many families face challenges, or things that get in the way, when trying to follow a health treatment, have you and your child/adolescent encountered any challenges to using CPAP therapy?
If so, what happened when these challenges occurred (i.e., try to problem solve, seek help from the Sleep Disorders Center)?
Are the people treating your child/adolescent’s SDB aware of these challenges?

Is there anything that would have made things easier (i.e., information, support)?

Currently, how many nights has your child/adolescent used CPAP?
Out of these nights, what is the longest amount of time your child/adolescent has used CPAP?
What is the shortest amount of time?
On average, how long has your child/adolescent used CPAP per night?

If I were to ask your child/adolescent, what do you think s/he would say about say about CPAP therapy?
Has your family had to make any changes since your child/adolescent started CPAP (i.e., routine)?
What does your child/adolescent’s CPAP therapy cost your family financially?
Are there any negatives to your child using CPAP therapy to treat their SDB (i.e., physical side-effects)?
How much do you think your child/adolescent should be involved in the CPAP therapy?
Do you think this should change as your child/adolescent gets older?
Would you say that your first experience with CPAP has been positive?
If so, what made it a positive experience?
Do you have a follow-up visit scheduled with someone who is treating your child/adolescent’s SDB?

Now that I know about how your child/adolescent was diagnosed and what it has been like beginning CPAP therapy, I would like to talk about your experience with the services you have received from the Sleep Disorders Center throughout this process.

III. Experience with treatment from Sleep Disorders Center at CCHMC

When was your first contact with someone from the Sleep Disorders Center?
How many times since then have you had contact (i.e., appointments or phone calls)?
How would you describe your relationship with the people who are treating your child’s SDB?
Would you change anything about your interactions with them?
Are you satisfied with the level of care that you have received regarding the treatment of your child/adolescent’s SDB?
Based on your experiences, is there anything that could be improved?
If so, how would you recommend this be done?
Have the people treating your child/adolescent been responsive to any questions, concerns or problems that you may have had?
Do you feel like you can ask questions regarding your child/adolescent’s care?
If you could change anything about the process or services you have received from the Sleep Disorders Center, what would it be?
Based on your experiences, what are the strengths of the Sleep Disorders Center?
How much control do you feel you have over your child/adolescent’s SDB treatment?
How would you describe the role you have played in your child/adolescent’s treatment?

Finally, I would like to finish the interview by asking you some questions about your overall experience with your child/adolescent’s SDB and CPAP therapy.
IV. Overall Experience

How has your family been affected by your child/adolescent’s SDB?
By the use of CPAP therapy?
Now that you have started CPAP therapy, how do you feel about it?
Is this different from when you were first told about it?
Have you noticed any changes in your child/adolescent since s/he started CPAP (i.e., behavior, mood, daytime, nighttime)?
Is there anything that you wished you would have been told about CPAP therapy or your child/adolescent’s SDB that you were not?
How effective do you feel CPAP therapy has been in addressing the symptoms of your child/adolescent’s SDB?
Sometimes following a health treatment can affect the quality of life of the child/adolescent and their family. Has CPAP therapy impacted you or your child/adolescent’s quality of life in any way?
Do you have any current concerns about your child/adolescent’s health?
Your child/adolescent’s treatment?
What would you tell other parents who just had a child/adolescent diagnosed with SDB and were going to treat it using CPAP?
What has been most helpful throughout this process?
Overall, what has been the biggest challenge to using CPAP therapy to treat your child/adolescent’s SDB?
Of all the things we talked about today, what do you think is the most important, or the area that stands out the most clearly?
Appendix B: Interview Guide for Follow-up Caregiver Interview

Introduction:

It has been about 3 months since our first interview. Today, I would like to obtain an update from you about how your child/adolescent’s treatment of his/her Sleep-disordered breathing (SDB) has been going.

Last time we talked your child/adolescent had been on CPAP for _____ days. You also told me that your child/adolescent was using CPAP at a pressure of _______. Sometimes the prescribed pressure changes over the course of SDB treatment.
   Has the pressure changed for your child/adolescent?
   If so, what was the change?

Before we get into specific questions, how have things been going since I last talked with you?

I. Use of PAP Therapy

I would like you to describe a typical night regarding your child/adolescent starting with getting ready for bed to waking up in the morning (i.e., what time did s/he go to bed, did s/he use CPAP, if so did anyone have to help with the CPAP therapy, how many hours did s/he use it)?

Now I would like you to describe an atypical night regarding your child/adolescent’s sleep routine.

Depending on your child/adolescent’s age or abilities, s/he may need a lot of help with using CPAP.
   How much help does your child/adolescent need with using CPAP therapy?
   If so, who is responsible for providing this help?

You might remember that we talked about how many families face challenges, or things that get in the way, when trying to follow a health treatment, have you and your child/adolescent encountered any challenges to using CPAP therapy?
   If so, what happened when these challenges occurred (i.e., try to problem solve, seek help from the Sleep Disorders Center)?
   Are the people treating your child/adolescent’s SBD aware of these challenges?

Is there anything that would have made using CPAP easier (i.e., information, support)?

In the last seven days how many nights has your child/adolescent used CPAP?
   Out of these nights, what is the longest amount of time your child/adolescent has used CPAP?
   What is the shortest amount of time?
   On average, how long did your child/adolescent use CPAP per night over this time period?
In the last month how many nights has your child/adolescent used CPAP?
   On average, how long did your child/adolescent use CPAP per night over the last month?
Has your child/adolescent’s use of CPAP been the same for the majority of nights since starting (i.e., different on weekends v.s. weekdays)?
   If not, what has contributed to the variation?
   If so, what do you feel has contributed to this consistency?
If I were to ask your child/adolescent, what do you think s/he would say about say about CPAP therapy?
Has your family had to make any changes since your child/adolescent started CPAP (i.e., routine)?
What does your child/adolescent’s CPAP therapy cost your family financially?
Are there any negatives to your child using CPAP therapy to treat their SDB (i.e., physical side-effects, inconvenience)?
How much is your child/adolescent currently involved in using CPAP therapy?
   Do you think this should change as your child/adolescent gets older?
Would you say that your experience with CPAP has been positive?
   If so, what has made it a positive experience?
   If not, what would have needed to be different to make it a positive experience?

Now that I have obtained an update on what it has been like using CPAP therapy, I would like to obtain an update about your experience with the services you have received from the Sleep Disorders Center throughout this process.

II. Experience with treatment from Sleep Disorders Center at CCHMC

Who is currently treating your child/adolescent’s SDB?
   How often do you have contact with this person (i.e., appointments or phone calls)?
How would you describe your current relationship with the people who are treating your child/adolescent’s SDB?
   Would you change anything about your interactions with them?
Are you satisfied with the level of care that you have received regarding the treatment of your child/adolescent’s SDB?
Based on your experiences, is there anything that could be improved?
   If so, how would you recommend this be done?
Have the people treating your child/adolescent been responsive to any questions, concerns or problems that you may have had?
   Do you feel like you can ask questions regarding your child/adolescent’s care?
If you could change anything about the process or services you have received from the Sleep Disorders Center, what would it be?
How much control do you feel you have over your child/adolescent’s SDB treatment?
How would you describe the role you have played in your child/adolescent’s treatment?

Now that your child/adolescent has been using CPAP for a longer period of time, I would like to ask you the same questions I asked you at the end of your first interview regarding your overall experience with your child/adolescent’s SDB to see if anything has changed.

III. Overall Experience

How has your family been affected by your child/adolescent’s SDB?
   By the use of CPAP therapy?
Now that your child/adolescent has been on CPAP therapy for a longer time, how do you feel about it?
   Is this different from when you were first told about it?
Have you noticed any changes in your child/adolescent since s/he started CPAP (i.e., behavior, mood, daytime, nighttime)?
Is there anything that you wished you would have been told about CPAP therapy or your child/adolescent’s SDB that you were not?
How effective do you feel CPAP therapy has been in addressing the symptoms (i.e., physical and behavioral) of your child/adolescent’s SDB?
Sometimes following a health treatment can affect the quality of life of the child/adolescent and their family. Has CPAP therapy impacted you or your child/adolescent’s quality of life in any way?
Do you have any current concerns about your child/adolescent’s health?
   Your child/adolescent’s treatment?
What would you tell other parents who just had a child/adolescent diagnosed with SDB and were going to treat it using CPAP?
What has been most helpful throughout this process?
Overall, what has been the biggest challenge to using CPAP therapy to treat your child/adolescent’s SDB?
Of all the things we talked about today, what do you think is the most important, or the area that stands out the most clearly?
Appendix C: Child Focus Group Agenda

**Note:** Questions, explanations and rapport building activities will be modified so they are developmentally appropriate for each age group.

1. **Introduction and Rapport Building**
   - Facilitator will introduce herself.
   - Each child will be asked to decorate a name tag using only their first name. Then, each child will be asked to say their first name, say how old they are, what grade they are in and something that they like and dislike.

2. **Explanation of Purpose and Ground Rules**
   - Facilitator will explain the meaning of confidentiality, the voluntary nature of the study and what the children can expect in developmentally appropriate language.

   **Example of Possible Script:** I want to thank you for coming. Today, I want to learn about what it is like for children to use CPAP therapy such as what they like and dislike about it. I will be asking the group some questions over the next hour, there are no right or wrong answers so say what you think or feel. I hope everyone will be able to talk as what each of you thinks and feels is important to me. If there is something that you want me to know but you can’t say out loud I will be giving each of you some paper that you can write it on and I can pick it up at the end of the group. If at anytime you feel like not doing this anymore, please let me know and you can stop. I will keep what you say secret. Just like I am going to keep what you say secret, you should also keep what the other children say secret. When you leave today you may want to tell your parents what was said and that is okay, just don’t use the other children’s names. Any questions?

   - Review ground rules: only use first names, raise your hand before talking, only one person can talk at one time, no side conversations, say what you think and feel, no right or wrong answers, hope everyone talks as want to hear what everyone has to say.

   - Facilitator will explain that the group will be audiotaped so that things are not missed but that it will not be shared with parents or doctors.

3. **Possible Questions for General Group Discussion**
   - What is this thing (will have a CPAP machine on a table)?
     Draw me a picture of what yours looks like/Describe what yours looks like.
What does it do?
How come people want you to use it?
When do you have to use it?
What happens when you use it?

- What has it been like having to use this machine?
- Many children who use this often say there are things that “get in the way” of them using it, tell me the things that have “gotten in your way”?
- What are your likes and dislikes about using this?
- How has your life/health been affected by using this?
- What does your family think about you using this?
- Has anything changed by you using this?
- What are some ways that people at CCHMC could have made this easier?
- What would you tell another child who was going to be using this for the first time?

4. Debriefing and Wrap-up

- Facilitator will summarize and reflect back what she heard and correct any misunderstandings.
- Each child will have the opportunity to make a last statement about their CPAP experience or about anything that was discussed.
- Each child will also be given an opportunity to share what their experience was like to participate in the focus group.
Appendix D: Medical Team Focus Group Agenda

1. Introduction, Explanation of Purpose and Ground Rules
   - Facilitator will explain agenda and that participation is voluntary
   - Review ground rules: only one person can talk at one time, no side conversations, say what you think and feel
   - Facilitator will explain that the group will be audiotaped so that things are not missed but that confidentiality will be maintained.

2. Barriers to CPAP Exercise
   - Group will be asked to generate as many barriers to adherence in children using CPAP as they can. Once listed then the group will have to rate each in their level of importance.
   - Once the group has identified the top 10 barriers they will be asked how interventions could address each of these barriers.
   - Emergent themes from the caregiver interviews may also be presented for discussion (i.e., reaction, elaboration).

3. Service Related Discussion
   - What are the challenges to prescribing CPAP for children?
   - What do you feel the Sleep Disorders Center does well when treating children with CPAP?
   - What do you feel the Sleep Disorders Center needs to do better?

4. Debriefing and Wrap-up
   - Facilitator will summarize and reflect back what she heard and correct any misunderstandings.
   - Each staff member will have the opportunity to make a last statement about anything that was discussed.
Appendix E: Clinician Rating Forms

**CPAP Adherence Study: Initial Contact**

Date: _______________  Participant ID #: _______________

Diagnoses:

1) ____________________  2) ____________________
3) ____________________  4) ____________________

Give pressure setting prescribed: ____________________

Based on your experience with this family, circle your prediction of the family’s overall level of adherence with CPAP using the following definitions to guide your decision:

- **No Use**
  - Never uses CPAP.

- **Very Poor**
  - Uses CPAP less than two nights per week with on average LESS than 5 hours per night.

- **Poor**
  - Uses CPAP less than two nights per week with on average MORE than 5 hours per night.

- **Average**
  - Uses CPAP 3 to 5 nights per week with on average LESS than 5 hours per night.

- **Above Average**
  - Uses CPAP 3 to 5 nights per week with on average MORE than 5 hours per night.

- **Good**
  - Uses CPAP 6 or 7 nights per week with on average LESS than 5 hours per night.

- **Very Good**
  - Uses CPAP 6 or 7 nights per week with on average MORE than 5 hours per night.

(0) (1) (2) (3) (4) (5) (6)
No use  Very Poor  Poor  Average  Above Average  Good  Very Good

In the space below please provide the rational for your decision:

_______________________________________________________________________________________
_______________________________________________________________________________________
_______________________________________________________________________________________
_______________________________________________________________________________________
_______________________________________________________________________________________
_______________________________________________________________________________________
_______________________________________________________________________________________
CPAP Adherence Study: Follow-up

Date: ___________________  Participant ID #: ______________

Give current pressure setting: ______________________________

Based on your experience with this family, circle the family’s overall level of current adherence with CPAP using the following definitions to guide your decision:

**No Use**
Never uses CPAP.

**Very Poor**
Uses CPAP less than two nights per week with on average LESS than 5 hours per night.

**Poor**
Uses CPAP less than two nights per week with on average MORE than 5 hours per night.

**Average**
Uses CPAP 3 to 5 nights per week with on average LESS than 5 hours per night.

**Above Average**
Uses CPAP 3 to 5 nights per week with on average MORE than 5 hours per night.

**Good**
Uses CPAP 6 or 7 nights per week with on average LESS than 5 hours per night.

**Very Good**
Uses CPAP 6 or 7 nights per week with on average MORE than 5 hours per night.

(0) No use (1) Very Poor (2) Poor (3) Average (4) Above Average (5) Good (6) Very Good

Has this level of Adherence been consistent over the course of treatment? ___Yes ___No

If checked “NO”, please explain: ____________________________________________________________

□ Fit of mask □ Skin breakdown □ Oral secretions □ Sinus problems

□ Change of pressure □ Anxiety/Fear □ Aspiration risks □ Other: ___________

□ Mask removal at night □ Refusal to wear mask □ Social Stigma □ Other: ___________

□ Sleeps at multiple houses □ Insurance Difficulties □ Eye irritation □ Other: ___________

Additional comments: __________________________________________________________

______________________________________________________________________________

______________________________________________________________________________