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Quality of life in youth with obstructive sleep apnea syndrome (OSAS) treated with continuous positive airway pressure (CPAP) therapy

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Abstract

Objective/Background—Youth with OSAS have poor QoL which resolves somewhat following treatment. One mitigating factor in improved QoL following treatment may be adherence to the CPAP protocol, which presents a barrier to most youth. This study explored relations between CPAP adherence and QoL in youth with OSAS.

Participants—We recruited 42 youth-caregiver dyads in which youth between the ages of 8–16 years were diagnosed with OSAS and required CPAP use as part of their treatment plan.

Methods—Following diagnosis of OSAS requiring treatment with CPAP therapy, caregivers completed baseline measures of OSAS-specific QoL. The OSAS-specific QoL domains assessed included sleep disturbance, physical symptoms, emotional distress, daytime function, and caregiver concern. Families received routine CPAP care for three months, after which caregivers again completed measures of OSAS-specific QoL. Adherence data were collected from smartcards within the CPAP machine after three months of treatment.

Results—15 youth were adherent to CPAP therapy and 10 were not adherent. CPAP adherent youth demonstrated significant changes in two domains of OSAS-specific QoL when compared to non-adherent youth; decreased sleep disturbance and decreased caregiver concern.

Conclusions—CPAP adherence appears to be associated with positive changes in OSAS-specific QoL domains. It will be important for future research and clinical work to examine strategies for improving CPAP adherence in youth with OSAS.

INTRODUCTION

In individuals with obstructive sleep apnea syndrome (OSAS), periodic obstruction of the upper airway interferes with normal respiratory gas exchange. This results in apneic or

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hypopneic events of decreased oxygen saturation (American Academy of Sleep Medicine, 2001; American Thoracic Society, 1996) followed by subclinical sleep arousals to return to normal breathing. Although protective by nature, this mechanism leads to fragmented, poor quality sleep. Untreated OSAS can result in undesirable physiological, behavioral, and cognitive sequelae including cardiorespiratory impairment (Hunt and Brouillette, 1982; Amin et al., 2002; Ugur et al., 2008), growth retardation (Marcus, Carroll, Koerner, Hamer, Lutz and Loughlin, 1994; Bonuck, Parikh, and Bassila, 2006), daytime sleepiness, hyperactivity and behavioral disturbances (Ali, Pitson and Stradling, 1993; Melendres, Lutz, Rubin, and Marcus, 2004), learning and memory deficits (Lal, Strange and Bachman, 2012) and increased risk taking and injury risk (Avis, Gamble and Schwebel, 2015; Treager, Reston, Schoelles and Phillips, 2009).

OSAS affects approximately 2–4% of American youth, including 25–40% of obese youth (Lumeng and Chervin, 2008, Marcus et al., 2012; Redline et al., 1999). Recent calls pertaining to patient centered outcomes research emphasize the need to understand health as a “state of physical, mental, and social well-being,” highlighting the centrality of quality of life (QoL) in health outcome research (PCORI Strategic Plan, 2013). Available research suggests youth with OSAS have poorer overall QoL compared to their healthy peers (Georgalas, Tolley, and Kanagalingam, 2004). Further, it has been reported that poor QoL is related to the negative impact of OSAS on physical health outcomes (e.g., negative health perceptions, increased bodily pain, and poor physical functioning) and psychosocial functioning (e.g. mood disturbance, poor academic performance) (Baldassari, Mitcheel, Schubeter and Rudnick, 2008).

Treatment may improve QoL for youth with OSAS. Adenotonsillectomy is generally considered the first line of treatment for youth diagnosed with OSAS, and research reveals significant improvements in QoL following surgical treatment of OSAS via adenotonsillectomy (Costa and Mitchell, 2009; Garetz et al., 2015; Franco, Rosenfeld and Rao, 2000; Mitchell, 2007), especially in the domains of sleep disturbance, caregiver concerns, and physical suffering (Garetz et al., 2015). However, surgical treatment is not effective or appropriate for all patients; in particular, OSAS persists in 60–88% of obese youth after adenotonsillectomy (Mitchell, 2007; Bhattacharjee et al., 2010). Given the high pediatric obesity rate, more youth are requiring treatment beyond surgery for their OSAS (American Academy of Pediatrics, 2002; Beebe et al., 2007). As part of routine care, continuous positive airway pressure (CPAP) therapy is one such treatment.

Available evidence is limited but suggests CPAP therapy may increase the QoL of youth with OSAS. Daytime functioning, as conceptualized by laboratory measures of attention, objective school performance, and self-reported daytime sleepiness, all improve with CPAP use (Marcus et al., 2012; Sanchez, Martinez, Miro, Bardwell, and Buella-Casal, 2009). Further, in one study that did not control for adherence to CPAP therapy, improvements in QoL were reported in youth over three-months of CPAP therapy (Marcus et al., 2012).

Pediatric adherence cut-offs to CPAP are not standardized, but available evidence confirms that adherence to CPAP therapies in youth is generally poor (Marcus et al., 2012; Marcus et al., 2006), with typical nightly use among youth around three to four hours a night across

multiple studies (Marcus et al., 2006). Determining exactly which usage criteria best differentiates adherence from non-adherence is an issue that continues to be studied and debated. In adults, 4 hours of CPAP use for 70% of nights is generally accepted as a cut-off for adherence, although several studies demonstrate that increased CPAP use results in more positive outcomes (Sawyer, Gooneratne, Marcus, Ofer, Richards, and Weaver, 2011). However, youth may require longer mean duration of nightly use (i.e., >4 hours) given that they often require more sleep than adults and because OSAS disease processes vary across the lifespan (Choi et al., 2010). Previous studies completed in youth have suggested that an average of >4 hours of CPAP use per night for at least 50% of nights represents acceptable criteria given documented difficulties with adherence (O'Donnell, Bjornson, Bohn, and Kirk, 2006; Uong, Epperson, Bathon, and Jeffe, 2007).

Health behavior theory suggests practitioners may facilitate increased CPAP adherence in their patients if the patients perceive the psychosocial value of using CPAP (Rosenstock, 1974). In particular, higher outcome expectancies for CPAP use have been associated with increased adherence, at least in adults (Olson, Smith, Oei, and Douglas, 2008). If patients are counseled to recognize the QoL benefits they may gain from using CPAP, and begin to experience such improvements, it might offer a therapeutic strategy to increase adherence among patients. Thus, this study examined the relationships between CPAP adherence and QoL. We hypothesized youth adherent to their CPAP treatment would experience greater improvements in their quality of life in comparison to participants who were non-adherent.

METHODS

Participants

As part of a parent study (Avis, Gamble, and Schwebel, 2015; Lynch and Avis, 2015), 42 dyads of youth (ages 8–16 years) and their caregiver were recruited from the Pediatric Sleep Disorders Center at Children's of Alabama, where they were assessed for sleep disorder with standard overnight polysomnography. Inclusion criteria included a diagnosis of OSAS by a trained sleep technician and confirmation by a licensed sleep medicine clinician, plus a treatment plan recommending continuous positive airway pressure (CPAP) therapy. Exclusion criteria included: 1) cognitive or physical disabilities (e.g., intellectual disability, blindness), 2) comorbid medical or neurologic conditions, and 3) antipsychotic medication use. Exclusionary criteria were selected given their potential to negatively impact CPAP adherence. The study protocol was reviewed and approved by the Institutional Review Board at the University of Alabama at Birmingham.

Protocol

Following the informed consent and assent processes, the caregiver completed a demographics questionnaire and baseline measures of OSAS-specific QoL (details below). Next, the family initiated routine care using CPAP therapy, which included: 1) CPAP education by the CPAP Care Coordinator in the sleep center, 2) meeting with a home durable equipment representative who fitted the youth for an appropriate face mask and provided the CPAP machine to the youth, and 3) after a week of adjusting to the new regimen, a return

medical visit for an overnight sleep study to titrate the CPAP machine to the optimal pressure.

Three months after the titration study, the family returned for a second research visit, which included caregiver completion of the OSAS-specific QoL measure a second time and CPAP machine smartcard download. CPAP pressure was not altered during the study period, however at the three-month visit, CPAP settings were reassessed to ensure continued CPAP effectiveness.

Polysomnography

All participating youth met International Classification of Sleep Disorders-2 diagnostic criteria for OSAS (Marcus et. al, 2006) based on diagnostic assessments that included nocturnal polysomnography (NPSG) using Sandman 9.2 technology software (Embla, Broomfield, CO) and thorough clinical evaluation from attending board-certified sleep specialists early in the morning after the overnight NPSG. Standard pediatric scoring was used for respiratory events (Sanchez, Martinez, Miro, Bardwell, and Buela-Casal, 2009). Diagnosis of OSAS was defined as AHI>1.5 per hour (Marcus et al. 1992; Uliel, Tauman, Greenfeld, and Sivan, 2004; Witmans, Keens, Davidson Ward, and Marcus, 2003).

Measures

Demographics—Parents reported the youth's age, race, and gender plus household income, and maternal age and education level.

Quality of life—The OSA-18 is an 18 item measure of the impact of OSAS symptoms on a youth's quality of life (Strocker, Carrer, and Shapiro, 2005). Responses are scored on a 1–7 point scale from “none of the time” to “all of the time,” with higher scores indicating poorer quality of life. Subscales on the OSA-18 include: sleep disturbance (i.e., choking or gasping sounds while sleeping; restless sleep), physical symptoms (i.e., frequent colds; mouth breathing), emotional distress (i.e., mood swings; aggressive or hyperactive behavior), daytime function (i.e., excessive drowsiness; poor attention span), and caregiver concern (i.e., concern that your child is not getting enough air; worry about your child's general health). This measure is valid in pediatric populations with OSAS (Franco, Rosenfeld, & Rao, 2000). Caregiver proxy-report test-retest reliability by item ranged from $r = 0.74$ to $r = 0.93$ (Franco, Rosenfeld, & Rao, 2000).

Adherence—Objective adherence data was gathered through downloadable smartcards located in the CPAP machine after three months of treatment. These smartcard adherence trackers (Resmed Inc., San Diego, CA) recorded the number of minutes per night the CPAP machine was powered on with the mask in place. Use of smartcards has been reported as a more reliable and accurate means of tracking adherence to CPAP in comparison to self-report (Schwab et al. 2013). Consistent with previous pediatric studies, the minimal criteria for defining adherence to CPAP therapy included an average of >4 hours of use per night for at least 50% of nights (O'Donnell, Bjornson, Bohn, and Kirk, 2006; Uong, Epperson, Bathon, and Jeffe, 2007). This conservative definition of adherence, which accounts for both mean hours of nightly CPAP use as well as frequency of use, has been established as a

clinical and empiric benchmark of CPAP adherence as previously indicated by Uong and colleagues (2007).

Statistical Analysis

We first considered participant adherence to the prescribed treatment regimen and divided the sample into those patients who were adherent to the CPAP protocol versus those who were not. Next, descriptive statistics were computed to describe the sample. Percent change variables for OSA-18 scores were calculated to reflect the change in scores from baseline to three-months on therapy. Because percent change measures were significantly skewed and kurtotic, non-parametric Mann-Whitney U tests were used to compare the adherent and non-adherent groups. Effect sizes for the Mann-Whitney U tests are also provided using the formula $r = Z / \sqrt{N}$, such that .1 is a small effect, .3 a medium effect, and .6 a large effect (Fritz, Morris, and Richler, 2012). All statistical analyses were completed using SPSS Statistical package, version 22 (IBM Corp, Armonk, NY).

RESULTS

Of the 42 participant dyads recruited into this study, 2 had greater than 50% missing data due to technical errors, 14 were lost to follow up, and 1 youth had a technical error in the CPAP monitor's adherence recording. This left a sample of 25 youth-caregiver dyads for analysis. The sample included 16 boys and 9 girls; all participant caregivers were mothers. Statistical comparison of participant dyads with complete data and those excluded from the final analysis on demographic characteristics, baseline questionnaire data, and diagnostic polysomnography results yielded no significant differences.

Fifteen youth were classified as adherent and 10 as not adherent. Table 1 reports demographic information, baseline QoL results, and polysomnography variables for the adherent and non-adherent subsamples. The two subsamples were similar on most characteristics, with a few exceptions: the adherent participants were significantly younger and had lower age and gender-adjusted BMIs than the non-adherent participants. Additionally, the caregivers of the non-adherent group expressed significantly higher baseline OSA-18 scores on the caregiver concern subscale compared to the adherent group.

Baseline to three-month follow up percent change scores for the total OSA-18 score and the 5 subscale scores (sleep disturbance, physical symptoms, emotional disturbance, daytime function, and caregiver concern) served as the primary dependent variables. Results revealed that the percent change in the OSA-18 total score did not differ significantly between the CPAP adherent and non-adherent groups ($p = .10$). It should be noted, however, that the effect size for this difference was medium in magnitude ($r = 0.35$). Two significant differences were observed when examining the OSA-18 subscale scores. Compared to CPAP non-adherent youth, those who were adherent demonstrated significantly greater percent changes (i.e., decreases) in the domains of sleep disturbance (adherent group mean rank = 10.00 and non-adherent group mean rank = 16.67, $U = 30.00$, $z = -2.24$, $p = .03$) and caregiver concern (adherent group mean rank = 10.27 and non-adherent group mean rank = 16.22, $U = 34.00$, $z = -2.00$, $p = .05$). These effects were also medium in size ($r = 0.46$ and $r = 0.41$, respectively). CPAP adherent and non-adherent youth did not significantly differ on

the physical symptoms, emotional disturbance, or daytime function subscales (p 's > .10). The effect sizes for these differences ranged from small to medium, as shown in Table 2.

DISCUSSION

A primary strength of this study was the objective measurement of adherence through the CPAP machine's smartcard. This is because previous studies have reported overestimation of CPAP use when relying only on parental report (Marcus et. al., 2006). The ability to obtain consistent, objective data on the rate of CPAP use provides increased confidence in the results. Furthermore, this study bolsters the current base of research regarding outcomes of CPAP therapy and rates of CPAP adherence.

Our results suggest youth with OSAS may experience less sleep disturbance and less caregiver concern with adherent use of CPAP therapy as indicated by subscales of the OSA-18. Despite significant changes in these subscales, we did not observe a significantly different change in overall QoL on the OSA-18 total score between CPAP adherent and non-adherent youth, which was contrary to our primary hypothesis. One explanation for this may be a lack of statistical power, given that the effect size for this difference was medium. The subscales on which significant differences were observed may be a reflection of domains that are most impaired in youth with OSAS; and therefore may have been more significantly impacted by CPAP therapy than other subscales. Additionally, findings revealed that CPAP adherent youth were more likely to be younger and have lower BMI than those participants who were non-adherent. These findings are consistent with preliminary studies of CPAP adherence in children, which have found age and BMI to be inversely related to adherence (Sawyer et al., 2011). Possible explanations for these differences include greater parental involvement in younger participants, and possibly an unfavorable response to CPAP in overweight youth as observed in adults (Kim et al., 2013). It will be important for future research efforts to more thoroughly examine how factors such as age/development and BMI interact with CPAP adherence to affect QoL outcomes.

Our findings indicate the potential clinical importance of educating families about the health benefits of CPAP adherence with a focus on specific aspects of quality of life, such as improved sleep and reduced caregiver worry regarding their child's health, that may result from increased adherence to CPAP treatment. While patient and family education alone is unlikely to produce significant improvements in adherence (Sawyer et al., 2011), it remains standard of care and may influence decisions regarding CPAP adherence. It is notable that our results found greater CPAP use was associated with positive changes in the domains that previous literature has shown to be most impaired in youth with OSAS, parental emotional factors and physical health outcomes (Baldassari, et al., 2008). Thus parent/patient education regarding the benefits of CPAP should be combined with behavioral interventions that have been shown to be effective for improving CPAP adherence in children (Koontz, Slifer, Cataldo, and Marcus 2003). Such an integrated approach may promote optimal quality of life and CPAP adherence among children with OSAS and their caregivers.

Several study limitations warrant mentioning. First, this study included a relatively small sample size. Lack of power due to this small sample size likely prevented us from detecting

the small and medium effects observed in this study, particularly the medium effect size ($r = 0.35$) for the difference between overall quality of life (OSA-18 total score) in CPAP adherent versus CPAP non-adherent youth. We believe it is important to include this effect size in order to inform future studies on this topic. Second, we experienced a high dropout rate (33%) from baseline to three months of therapy, although examination of available data suggested the lost and retained participants were similar on demographic and health characteristics. This dropout rate is consistent with the dropout rate in another study on CPAP adherence (Marcus, 2006), which posited that dropout rates may actually be a reflection of non-adherence. Third, we followed participants for just three months. A longer follow-up period may have yielded different results in either or both groups. Fourth, youths with cognitive or physical disabilities, comorbid medical or neurological conditions, and those using antipsychotic medications were excluded. Therefore, the positive changes in QoL with increased CPAP adherence observed in this study may not generalize to other populations. Finally, there was no sham or placebo group; all youth enrolled were prescribed CPAP treatment. Therefore, the gains seen by the current participants could have been a result of other unmeasured factors, such as time and development.

Additional unmeasured factors could account for the low CPAP adherence found in this sample. These may include cultural opinions, level of family support, family organization or disorganization, the youth's understanding of their health, and other specific youth-based belief patterns. Future research should address the impact of these broader psychosocial and contextual factors on youth CPAP adherence. Demographic and health aspects of BMI and age may warrant additional exploration in their relations to CPAP adherence. Additionally, it will be important to examine the dose response experienced by children prescribed CPAP to determine optimal adherence levels for CPAP therapy in youth.

In summary, this study found medium effect sizes for overall improvement in quality of life, caregiver concern, and symptoms of sleep disturbance in youth adherence to CPAP versus those who were not adherent. Future research addressing this topic will likely need to include larger sample sizes in order to replicate and expand upon the medium-sized effects found in this study. Furthermore, additional research may consider the dose response of pediatric CPAP therapy and developing therapeutic interventions to improve adherence, possibly focusing on the youth-caregiver relationship.

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Table 1.**Baseline Characteristics of Children with OSAS**

Characteristic	CPAP Adherent (N = 15)	CPAP Non-Adherent (N = 10)	χ^2 or t	p
Demographics				
Youth Sex: % male	66.70	60.00	0.12	0.73
Youth Age: mean (SD), in years	10.53 (2.17)	13.50 (2.32)	3.26	0.01*
Youth Race: % white	46.70	40.00	0.11	0.74
Youth BMI: mean (SD)				
Age, gender adjusted	30.79 (8.86)	44.15 (12.62)	3.12	0.01*
BMI z-score	2.04 (1.05)	2.58 (0.53)	1.77	0.15
Household income: (%)				
Below \$20,000	28.60	33.30	0.06	0.81
\$20,000–39,000	35.70	55.60	0.88	0.35
\$40,000–59,000	28.60	0.00	3.11	0.08
Greater than \$60,000	7.10	11.10	0.11	0.74
Mother's education: (%)				
HS diploma or below	21.40	40.00	0.97	0.32
Some college	50.00	50.00	0.00	1.00
College degree	28.60	10.00	1.22	0.27
Pre Intervention OSA-18				
Sleep Disturbance	16.20	19.60	1.78	0.09
Physical Symptom	16.53	15.60	−0.27	0.79
Emotional Disturbance	11.73	11.00	−0.29	0.77
Daytime Function	13.80	14.60	0.42	0.68
Caregiver Concern	16.13	21.70	2.53	0.02*
OSA-18 Total Score	72.20 (14.19)	82.50 (15.95)	1.69	0.10
Polysomnography Data				
Total Sleep Time	455.66 (67.91)	444.00 (27.18)	−0.51	0.61
Sleep Efficiency %	83.83 (13.22)	88.95 (4.09)	1.40	0.18
AHI	13.15 (19.58)	10.45 (7.62)	−0.41	0.68
OSAS Severity: % Severe	33.33	50.00	1.33	0.52

Note: OSAS = Obstructive sleep apnea syndrome; BMI = Body Mass Index

*
p < .05

Table 2.

Percent Change OSA-18 Outcomes: Mann-Whitney U Test

Variable	Adherent N= 15		Non-Adherent N= 10		U	z	Effect Size
	Percent Change	Mean Rank	Percent Change	Mean Rank			
Sleep Disturbance	-63.47	10.00	-33.84	16.67	30.00 *	-2.24	0.46
Physical Symptom	-53.53	10.00	-40.74	13.67	39.00	-1.30	0.28
Emotional Disturbance	-34.90	10.68	-23.38	12.94	44.50	-0.79	0.17
Daytime Function	-55.53	10.00	-27.35	15.11	35.00	-1.77	0.37
Caregiver Concern	-55.67	10.27	-32.39	16.22	34.00 *	-2.00	0.41
OSA-18 Total Score	-53.45	10.06	-37.70	15.67	39.00	-1.70	0.35

*
p < .05