



Published in final edited form as:

Sleep Med. 2023 July ; 107: 179–186. doi:10.1016/j.sleep.2023.04.022.

Caregiver Experiences Helping Children with Down Syndrome Use Positive Airway Pressure to Treat Obstructive Sleep Apnea

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Author Contributions: MSX, MNN, WE, FKB, KB, SI, AD, JMD, CH, NB, IET contributed to the conception of this study; all authors contributed to its design. RB, SH, MNN, and WE contributed to data acquisition. MN, WE contributed to analysis of data. MSX, MNN, WE, FKB, KB, SI, AD, JMD, CH, NB, IET contributed to interpretation of data. All authors contributed to drafting the article for important intellectual content and have provided approval of the version to be published.

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Work was completed at Children's Hospital of Philadelphia, Cincinnati Children's Hospital Medical Center, and University of Pennsylvania.

All authors have reviewed and approved the manuscript.

Nonfinancial Disclosures: None.

Clinical Trial Registration: Investigating the Experience of Living with Down Syndrome and Obstructive Sleep Apnea Syndrome: <https://clinicaltrials.gov> (NCT04124471)

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Abstract

Background/Objective: While positive airway pressure (PAP) is an efficacious intervention for the treatment of obstructive sleep apnea syndrome (OSAS) in children with Down syndrome (DS), implementation and consistent use can be difficult. Caregiver perspectives and experiences using PAP are described with the aim of informing clinical practice.

Methods: Qualitative semi-structured phone interviews were conducted with 40 caregivers (i.e., mothers) of children with DS and OSAS treated with PAP for at least 6 months. Content analysis was used to identify themes associated with adherence and non-adherence.

Results: Respondents indicated variability in caregiver experience with the adoption of PAP and observed benefits of PAP. Varied experiences were attributed to several themes including accessing supplies, interactions with the medical team and equipment company, and patients' unique needs and behaviors, including the child's willingness and ability to adapt to PAP, sensory sensitivities, keeping the mask on all night, and differences in daytime behavior. Many families reported that desensitization with a reward system and trust within the caregiver-patient relationship were helpful. Caregiver suggestions for improving PAP adherence for families of children with DS included improving communication with the medical team and medical equipment company, emphasizing patience, using visual supports, and social support and education for extended family.

Conclusions: Although family experiences varied, several actionable strategies by both the medical team and families emerged for improving the experience of and adherence to PAP in children with DS.

Keywords

Down syndrome; pediatric; obstructive sleep apnea syndrome; positive airway pressure; adherence

1.0 Introduction

Down syndrome (DS) is the most common genetic chromosomal disorder causing intellectual disability in children, occurring in 1 per 800 births¹⁻³. DS is also associated with developmental delays, behavioral difficulties^{4,5}, a shorter life expectancy and early-onset Alzheimer's disease^{6,7}. Common comorbidities include hypothyroidism, obesity, and the obstructive sleep apnea syndrome (OSAS)⁴.

Compared to the prevalence of OSAS of approximately 2% in typically developing children⁸, the prevalence in children with DS is markedly higher^{9,10} with studies reporting prevalence between 45 and 55%⁹⁻¹¹. Children with DS are also at increased risk of having

persistent OSAS following surgical treatment^{9–11}. The predisposition of children with DS to OSAS and persistent OSAS following surgery is often attributed to craniofacial features (midface hypoplasia, glossoptosis), hypotonia, comorbid obesity, and hypothyroidism¹². Untreated OSAS is associated with cognitive and behavioral deficits in typically developing children and may exacerbate deficits observed in children with DS^{13–18}.

Positive airway pressure (PAP) is an efficacious treatment for pediatric OSAS; however, implementation is challenging and long-term adherence is variable^{19–23}. PAP adherence in children is multifactorial and has not been sufficiently studied, particularly in those with intellectual disabilities, including DS^{21, 22, 24–33}. Much of the research on PAP adherence in children with DS focuses on potential demographic predictors (e.g., age, race), disease characteristics (e.g., severity of OSAS, medical comorbidities), or device characteristics (e.g., mode, pressures, leak)^{20, 34}. It has been suggested that adherence challenges are often encountered at the initiation of PAP in children with DS, but that long-term adherence can be achieved with time, intensive support, and sometimes repeated trials over time^{22, 29, 30, 35}. There has been less emphasis, however, on understanding the process or experience of using PAP for children and their families^{36, 37}.

Children with DS may face similar PAP implementation issues as typically developing children, such as incorporating PAP into their routine; getting used to the sensation of the mask, headgear, and pressure; and the need for caregiver support. However, they may also encounter difficulties related to neurodevelopmental differences that may deter caregivers and healthcare providers from pursuing PAP, such as learning differences, sensory and behavioral concerns, and the inability to understand the need for PAP or communicate reasons for not wanting to use PAP (e.g., what is uncomfortable)^{5, 30}. Further, in addition to the burden of putting equipment on each night and the common experience of discomfort wearing a mask with pressurized air being administered while sleeping, common side-effects include nasal and ocular symptoms, skin pressure sores or dermatitis, and aerophagia. The unique experience of caregivers helping their children with DS adapt to and use PAP has not been explored; therefore, the primary objective of this study was to identify caregiver perspectives regarding the implementation and maintenance of treatment of OSAS with PAP in children with DS with the aims of informing clinical practice and developing a structured intervention for promoting adherence.

2.0 Methods

Caregivers (i.e., parents or legal guardians) were recruited from two large pediatric medical centers with sleep centers that included children with DS. Procedures were approved by the IRB of record for both sites). To obtain consent, the study was reviewed over the phone and verbal consent was provided by caregivers prior to the interview. A signed copy of the consent was mailed to families. In order to ensure a range of experiences and responses, caregivers were recruited if their children's PAP use met either of the following adherence criteria defined *a priori* to interview: "low adherence," at least 1 attempt within at least 3 months OR wearing PAP 50% of nights OR wearing PAP 2 hours on nights used in the 30 days prior to the interview; and "high adherence," wearing PAP 60% of nights AND wearing PAP for 4 hours/night on nights used for the 30 days prior to the

interview. PAP mode, including CPAP, bi-level PAP, or AutoPAP were all included given the hypothesis-generating nature of this study. While AutoPAP has not been well-studied in children, it is widely used clinically, including in those with DS³⁸; therefore, those on AutoPAP were not excluded.

Qualitative semi-structured phone interviews were conducted by a single trained researcher (M.N.) with caregivers of children with DS and OSAS. Children with DS were aged 6-18 years, and prescribed PAP for at least 6 months. Two children of Asian descent were adopted to the same family and the caregiver could not discriminate between the experiences of implementing PAP with both children. The interview guide (Appendix A) was developed based on the previous research and on authors' (MSX, KB, IET) clinical experience. The interview focused on caregivers' experiences with initiating and maintaining PAP with their child and aimed to elicit suggestions for enhancing adherence. Audio recordings of the interviews were transcribed by an outside service and sent to the Mixed Methods Research Lab (MMRL) at the University of Pennsylvania. The transcripts were then uploaded to NVivo 12 Plus, a qualitative analysis software (QSR International Pty Ltd., released 2018) for coding and analysis. A conventional approach to content analysis was used³⁹. Using an emergent content analysis approach⁴⁰, transcripts were reviewed for key ideas and concepts from the data. These concepts were then utilized to develop a book of themes (i.e., codes), with definitions and rules for application. Two investigators (M.N. and W.E.) applied the codebook to the transcripts from both sites and periodically refined the codebook based on inter-rater reliability tests to during the analysis (final $\kappa=.79$). Coded data were summarized, sorted into categories, and examined for factors associated with adherence and non-adherence, with demographics, and with practices associated with implementation of PAP. Demographics between adherent and non-adherent groups were compared using t-tests for parametric or Fisher's exact test for non-parametric variables. A p-value <0.05 was used as the criterion for statistical significance.

3.0 Results

Forty caregivers at two sites (N=20 at each site) of 41 children were interviewed. All caregivers were mothers. Table 1 presents demographic information by adherence category. Twenty-two participants met the high adherence criteria and 19 met the low adherence criteria. There were no differences in demographics by adherence except for PAP mode. All children had a titration PSG prior to the interview. Interviews lasted an average of 28 minutes. Median income and other demographic variables did not relate to a meaningful difference in themes in our sample.

In the following section, headings represent identified themes and subheadings represent subthemes. Further details expanding upon each theme, as well as examples of verbatim lay language caregiver statements that best represent the theme are also presented. Appendix B provides additional examples of quotes from families.

3.1 Orientation to PAP

3.1.1 Timing of initial PAP education—Two different experiences regarding initial OSAS and PAP education emerged: families who reported receiving comprehensive

detailed information about implications of OSAS for their child's health and how to help them incorporate PAP at the time of prescription, and those who received more detailed information at subsequent appointments. Some caregivers in the former group felt overwhelmed at the amount of information discussed during the initial appointment. The latter group described attempting to use the device at home with minimal guidance, then receiving further instruction and advice after an initial trial period. Caregivers reported that more detailed information about PAP, guidance, and strategies for helping their child become accustomed to PAP were relayed by sleep physicians and psychologists.

3.1.2 Type of educational resources/supports—Caregivers appreciated educational supports, specifically handouts that provided detailed information about OSAS, PAP devices, important resources, and recommendations tailored for each child's needs and behaviors. Some parents reported that these educational supports helped them to integrate new clinical information and treatment recommendations. In addition, some parents supplemented treatment team-provided education and support with information from other sources including online communities, independent support groups, and from other parents with the shared experience of helping their child adapt to PAP.

3.1.3 Role of the medical team—Caregiver sentiment regarding the level of support received from the medical team during PAP initiation varied widely. Some felt well-informed and equipped to introduce PAP. Others felt uninformed about OSAS and its consequences for their child, about PAP and how it worked, and were unsure of how to get additional information. They also reported feeling extremely concerned and overwhelmed about the lack of information because it had been communicated that their child's condition was severe. The latter group described wanting more detailed and explicit instruction about PAP, different settings, risks of not using it appropriately, and mask options.

“But when you initially go to the appointment, they sit down, and they explain everything to you. They are very thorough with explaining, showing the math, any questions, doing it over. Then they even tell you how to clean it. Show you how to clean it, tell you all the details. [I wrote it down in the] back of the book, change the filter, clean the hoses out, because it's a lot with the machine itself to keep it clean.”

“No, we didn't [receive any education at the time CPAP was prescribed]. I mean as far as the hospital telling us what the diagnosis was and this is what the risk was, how we can resolve it, that's basically what we had. We then came back home and did more study online ourselves.”

3.2 Strategies for Introducing PAP

Experiences initiating PAP were highly variable. Some children accepted the treatment after one or two nights, while others struggled to maintain an hour of usage each night years after initiation. Families reported that their medical team most commonly recommended introducing PAP to their child gradually by first allowing the child to explore/examine the mask, wearing the mask on their face without air pressure, and having a family member (e.g., caregiver; sibling) model wearing a PAP mask.

3.2.1 Patience—Caregivers highlighted the importance of patience throughout the process of adapting to the device. They stressed the value of allowing the child to become accustomed to the device at their own pace. Pairing exposure to the mask with a distracting activity, like watching television, was reported as a good method for helping children acclimate to wearing it during the day. Frequently, caregivers reported after successful introduction of PAP, they quickly integrated PAP into the child’s nightly bedtime routine.

Reassurance from the medical team that the process of adjusting to nightly PAP use could take several months was suggested as a way care could be improved for other families.

“I think you just have to be patient with them, patience is definitely I think is the key ... my advice to most parents again, you just have patience with them just figure out a way that will make it fun, try to say, ‘Hey, this is fun, this will help you breathe better,’ just interact with them to where it makes it fine.”

3.2.2 Visual Aids—Important advice from caregivers included use of visual aids to help children understand PAP functioning and benefits. Since not all children were able to understand verbal descriptions, visual aids (e.g., visual schedule) and storytelling tools (e.g. social stories⁴¹) to help relay the information were described as useful. Videos of children wearing PAP and a familiarity with someone who uses PAP were highlighted as successful strategies for promoting successful PAP initiation. Caregivers recommended using flip books, flash cards, or inventing a social story picture book depicting a child with DS using PAP as ways to help their child better understand.

“I think children born with Down syndrome are more visual. So, I feel like, once they see things, I think having experience of letting them discover for themselves. The video, now that I think of it, we did that early. Doing the video and kind of being hands-on with it I think is probably more helpful. That would be just to make sure that you have individuals there for ‘em I think helps along with talking to them about it. [Laughs] Letting them discover some things for themselves to get comfortable with it instead of pushing something on them.”

Some caregivers endorsed using positive/non-threatening imagery and imaginative play as a strategy to elicit cooperation and enhance their child’s adjustment to PAP. Examples included decorating and naming PAP devices or referring to them as special characters or friends and/or pretending to be an elephant while wearing the mask and tubing. This incremental approach, potentially spanning over several months before the child reached what the caregiver deemed an acceptable level of PAP use, was described as a successful strategy for introducing PAP.

“We just do our normal bed routine. Every night we do the same thing. You brush your teeth, you take your medicine, you get in bed, you put the mask on, you put the fan on, just a normal routine always would help us till we start putting it on, but it’s going on through the night is the problem, for us, after he wasn’t scared of it anymore.”

3.2.3 Mutual understanding between caregiver and child—Caregivers reported that mutual understanding within the caregiver-patient relationship was important in the

introduction of PAP. A key aspect was to ensure that the child understood the reason the parent was asking them to wear the device. Caregivers described trying many different strategies to help their child understand the purpose of using PAP as described in other sections of this manuscript. A small number of caregivers also mentioned the importance of their child's trust in their medical care team as an adherence factor.

“She trusts us in a sense like, that's how we've always approached everything with her. Whether she's getting a shot or a vaccination or whatever it is that she's doing, that even if it's unpleasant, it's kind of like, ‘We only do things that are going to help you. This is helping you. We wouldn't do this to you if you didn't need it.’ Those kinds of things. From the time she was little ‘til now, we still talk to her about, ‘These are things that help you. And this is why it helps you.’ Put in terms that she understands. She had that build up trust in us, I feel like.”

Interestingly, most caregivers expressed that if given the option, they would not introduce PAP any differently to their child. In general, families felt that their approach was the best they could offer their child based on their own situation and the child's unique characteristics and needs. Most caregivers preferred using positive reinforcement with their child rather than an authoritative approach. Positive reinforcement strategies ranged from praise to elaborate contests and stories. However, other caregivers reported that any reward technique was futile in introducing PAP.

3.3 Nighttime Adherence

Caregivers reported that once PAP was incorporated into their child's routine, the main adherence-related challenge was ensuring the mask was not removed or displaced during sleep. Caregivers perceived that the masks of “rough sleepers” would fall off during the night or the mask was uncomfortable or disruptive to their child's sleep. Caregivers' interventions to facilitate PAP use through night ranged from setting an alarm every hour to check that the mask was still in place to designing a special system of pillows to support the PAP device and mask for a child who preferred to sleep on their side.

“And then at the beginning, there were lots and lots of checks. Fortunately, she was going to bed, you know, earlier than my husband and I, so we could go in, put it back on her, put it back on her, put it back on her.”

3.4. Barriers to PAP Adherence

3.4.1 Mask and airflow sensitivity—Sensory sensitivity and reactivity to auditory and tactile stimuli such as air blowing on the child's face emerged as key factors for PAP tolerance. Some children were frightened of the sounds made by PAP, particularly of those from leaks and mask displacement. Other children had intolerance for anything touching their head or face. In some cases, these were significant impediments to their child's adherence.

“She was not keen on it at all. She doesn't like things on her face and around her head and it wasn't necessarily the texture thing, it's just a feeling thing. She doesn't like rain, she doesn't like showers, she doesn't like anything touching her head. So

that was difficult and challenging. Some days it was just enough for her to even put the CPAP machine on her face.”

3.4.2 Finding the right mask—Ill-fitting and uncomfortable masks were frequently cited as one of the biggest barriers to adherence. Parents described a lengthy, frustrating process of trial and error that involved trying various types and sizes of masks to find the most tolerable. Due to insurance regulations, some families had to wait for the mask to break or to be discontinued before receiving an alternative interface.

“Once we got the machine working, she had grown, and the mask was not fitting. That goes to all this that I was saying, the mask was uncomfortable because she had grown. The headgear was uncomfortable because she had grown, children grow very fast. By the time we got a machine that was working and a headgear that was the right size and settings that are comfortable, that’s been a huge process.”

“He used to do a total face mask and the face mask was *mask brand*, so we had ended up changing from the *mask brand* to *mask brand* because, as he’s grown, his forehead has become more flat and the mask is more rounded to go around a typical child’s head ... so I would have to really sit down and I had to figure out how to get the mask into a flatter shape which caused marks on his face. So, we had to find a different mask and they would say, ‘Well use the nasal one.’ I said, ‘He won’t use the nasal one, he will not because he has air rushing out of his mouth that freaks him out.’ If I know this, then we stay away from that. I tried to get them to try different ones. They do reluctantly because they wanted him to come in to get fitted and ‘I’m like, well they fit him at [Children’s Hospital], so there’s no point in doing it twice if we found one that fits.’ Then they, in the orders, they say they don’t have orders for our supplies and then the insurance doesn’t pay.”

3.4.2 Logistics and supplies—Caregivers expressed trouble regularly accessing new supplies when needed, including tubing, filters, masks, and cleaning products. These delays were partly due to insurance regulations, or the result of low regional access to device suppliers and parts distributors. Caregivers also reported that the time and energy required to order supplies and cleaning the equipment was significant. Caregivers made few distinctions between interactions with their child’s care team, device or supplies distributors, and insurance companies. However, caregivers conveyed that the main problem with access was an intersection of these entities. Caregivers who described access issues frequently resorted to their own, sometimes homemade interventions, when they felt unsupported by the medical system.

3.5 Child characteristics

Child characteristics, including the idiosyncratic needs and behaviors of each child were described as key factors related to successful PAP introduction. Children who were described as generally compliant, motivated by rewards, or who easily adjusted to change were more frequently deemed successful at using PAP.

“She didn’t like it. She didn’t want to do it. But kids with Down syndrome are very responsive to habit-forming. That’s what we did is we just made it a habit. We just did, like I said, a little bit more each time. She’s a people person. She just wants to please us. She wore it, but lots of times she would complain that it was uncomfortable, but she doesn’t have enough verbal skills to say in what way. That’s the hard part as well.”

“It was a little negative because I know that [Child’s Name] being a down syndrome kid he doesn’t have a long attention span, meaning that if something goes on his face or with anytime he has something it’s a short lived situation meaning that he is not going to keep it on at all times, it has to be like, hey, I have to give you something like a snack or something to say if you keep it on all night or whatever. So it was like a give and take situation right now. I think it was important, but I know for him being at that stage of his life, he wasn’t going to keep it on for a duration to try to get a clear reading.”

3.6 Suggestions and Advice for Families

Caregivers offered many recommendations for introducing PAP to other families initiating PAP treatment for their child with DS. Appendix B presents additional examples. Caregivers emphasized a focus on the importance of visualization tools as part of education and preparation for PAP initiation. Caregivers emphasized the importance of focusing on the benefits of PAP and positively reinforcing their child’s compliance. Caregivers also mentioned that approaching introduction in a deliberately routinized, incremental way was helpful in setting the child up for later success.

“I think that maybe if there could be some system where you could encourage parents to have lots of positive reinforcement and fun like play game or something where ‘Let’s see how long you can wear it, if you can beat your score from last night,’ or something like that. That really helped us anyway ‘cause [Child] was then more excited to wear it. Thought more positively instead of almost like a punishment.”

“The original schedule they gave us to build her up to using the air and getting used to it, getting her to fall asleep with the air on, all of that was great and it worked really well. And I think for a child who struggles even wearing masks, it would help even more. [Child] was way ahead ‘cause she was already okay wearing the mask. But where we’re struggling is just the keeping it on all night.”

3.6.1 Social support—Caregivers emphasized the importance of social support, particularly for the family member responsible for helping the child use PAP each night. Primary caregivers reported often being solely responsible for supervision and supporting their child with DS using PAP and that it would be beneficial if multiple family members, especially those who help care for the child, were more actively involved with PAP initiation and promotion. For some families, this meant having other caregivers, including parents and siblings, assist in the desensitization process, introduction play, or cleaning of the device. Other caregivers reported concerns related to extended family members and social contacts having poor understanding of the importance of PAP for their child.

“My husband looks at it like that’s something that I’m dealing with, so basically, I was in the ER with my [family member] all night and so he didn’t put it on her. He said, ‘Well, that’s your thing.’ Like if I’m not here, it’s going to be your thing. But maybe just making sure that the families are all on the same page.”

4.0 Discussion

This is the first study to the best of our knowledge to elicit the perspectives of parents of children with DS on the experience of initiating and adherence to long term PAP use. In contrast with a previous study that reported parent perspectives via questionnaire completed by caregivers of children with obesity and sleep disordered breathing³⁶, the current study utilized nuanced qualitative methods assessing caregiver viewpoints of a marginalized population, children with DS. Responses from caregivers indicated substantial variability in their experience in the adoption of PAP to treat OSAS. Most caregivers reported generally positive or neutral experiences with their child’s medical care team throughout the process, although negative experiences pointed to potential opportunities for improvements in care. Results confirmed that implementing PAP is challenging^{42, 43}, and families underscored the interplay of medical, technical, behavioral, interpersonal and logistical challenges supporting a previously proposed socio-ecological framework as a conceptualization for PAP adherence⁴⁴.

Studies have shown that support from the medical team is essential in facilitating initiation and long-term adherence to PAP in children^{36, 45}. In this study, families reported difficulty navigating the overall healthcare system. The challenges expressed and opportunities for improvement include a better understanding of the roles of the medical team and the durable medical equipment company, and overall improved communication of both entities with the family. Research on other chronic conditions, such as asthma indicates that a perceived partnership and shared decision-making with the medical team often improves adherence to medical recommendations^{46, 47}. Moreover, a recent study reporting parental experiences of having a child with DS and sleep difficulties highlights the need for medical teams to actively support families in strategies to reinforce healthy sleep or treat sleep problems⁴⁸. Further, a qualitative study of parents of adolescents with DS revealed that parents experience desperation about the acceptance of the current standard of care for the treatment of OSAS, leading them to seek out information and guidance from social media (e.g., Facebook sites for parents of children with DS) rather than from the medical team⁴⁹. Based on the communication challenges expressed, families would benefit from a partnership with their medical team in which expectations and roles are clearly defined.

To facilitate frequent and efficient communication and partnership with the medical team, leveraging technology to improve communication, such as secure text messaging, direct messaging members of the team through the electronic medical record, (EMR) and providing feedback to families from PAP modem downloads via telephone or messaging systems, may enhance communication and partnership. While caregivers did not express a specific desire for text messaging or EMR messaging, these methods are being increasingly utilized by caregivers and the healthcare system. Utilizing technology has recently been

found to be beneficial to adults who are prescribed PAP but has not yet been systematically studied in children^{50, 51}.

Varied experiences [revealed by caregivers in this study] highlighted the need for continuous support to families^{29, 35, 45, 52}. Caregiver reports varied based on each individual child's level of willingness and ability to adapt to PAP and caregivers' ability to incorporate PAP procedures into the home environment. Some parents expressed distress and feelings of being overwhelmed about first introducing PAP and then maintaining its long-term usage, whereas others reported ease in implementing PAP because the child took to it without difficulty. Some described initial challenges that eased over time, especially those with younger children. This reported caregiver experience is of interest as previous research has indicated that overall, adherence decreases with age, with children being more adherent than adolescents⁴³. The reason for this discrepancy is unclear. It is possible that children with DS are diagnosed with OSAS earlier than typically developing peers, such as late infancy or toddlerhood, which can be a challenging developmental period to implement PAP. It is also possible that behavioral plans are more effective in children with DS at older ages compared to typically developing peers due to neurodevelopmental differences. Lastly, it has also been proposed that multiple trials of implementation of PAP over time yields better adherence than a single attempt³⁵. Children with DS tend to have persistent OSAS and likely several attempts of implementing PAP over time, thereby eventually improving adherence with age.

Research demonstrates that children, including those with developmental differences, can improve their adherence to PAP over time^{29, 35, 52}. Initiating PAP with a balanced approach, such as presenting the benefits of PAP but also acknowledging that it is not a crisis or emergency may support families better in adopting a slow, consistent, and steady PAP implementation strategy. Consistent with this, caregivers suggested that the treatment team should clarify that PAP adherence may develop gradually as families can better balance their own expectations if they understand the potential trajectory of use. This can also prevent feelings of being overwhelmed, pressured, or being a "bad parent". Caregivers stated the significance of having the treatment team describe that observable changes such as improved attention, decreased sleepiness, or improvements in fatigue may be subtle, and it does not mean that PAP is not working or that they are not doing a good job at using it. Further, explaining that other long-term benefits, such as learning, cardiovascular health, or metabolism will likely not be directly observed by caregivers or felt by the child was deemed useful.

Children with DS tend to have a relative strength in visuo-spatial processing^{5, 53}, and frequently have expressive and/or receptive language delays. Children with DS often have difficulty with changes in routine and transitions⁵⁴ emphasizing the importance of repetition and consistency. Caregivers referenced these issues in their recommendations for promoting adherence to PAP. For example, the healthcare team may draw upon the caregiver's expertise with their children and consider utilizing visual schedules, charts, videos of children with DS preparing for and wearing PAP at night, and/or a PAP social story as visual support tools to explain why PAP is needed. Other caregiver suggestions that can enhance adherence include systematic desensitization and positive reinforcement; and reward systems such as a token economy, morning prize bag, or "PAP fairy" (e.g., similar to tooth fairy) who visits

during the night and leaves a prize under the child's pillow if the PAP is still on. Caregivers also indicated that using their child's imagination to "demedicalize" the device such as inventing a special character for the machine, pretending that the machine was their friend, naming the machine or decorating it also helped facilitating its use and making it the PAP experience more positive.

It is apparent from caregiver interviews that matching the appropriate implementation plan to the specific child and family is essential to successfully implementing PAP with a family-centered approach⁵. For example, children with DS are commonly described as willful, impulsive, and frequently noncompliant, and refusal behavior may contribute to nonadherence⁵⁴. However, other children with DS are described as easy-going, routine-/rule-governed "people pleasers," which may improve adherence. Caregivers expressed the need for social support, particularly from other family members and extended family, like other families supporting children with special medical needs^{55, 56}. One barrier families expressed was difficulty in helping extended family and social circles understand the significance of PAP treatment. Some of these apprehensions may be alleviated by providing parents resources to help those outside of the immediate family unit understand PAP and OSAS; and a foundation in which to access additional supports, such as peer support groups⁵⁷.

Caregivers did not comment during interviews about the mode of PAP therapy used possibly because they typically have experience with only one mode of PAP. However, PAP mode was statistically different by adherence group and may impact a child with DS's ability to adapt to PAP. Previous research has not consistently identified PAP mode as a predictor of adherence to PAP in children⁴³. Future research in children with DS prescribed PAP should explore this finding further with larger samples sizes powered to evaluate this potential relationship.

There are several limitations of this study, particularly related to the diversity of the sample. All caregivers interviewed were mothers. This was not intentional, however, and future research should include perspectives from fathers, other caregivers, and members of the family involved in caring for children with DS and OSAS and/or family members who also use PAP. We also acknowledge that the mothers that agreed to participate may have been particularly motivated and may not necessarily represent all mothers of children with DS, thereby contributing to selection bias. Additionally, while there was representation from African American and Caucasian families, illustrative of the institutions in which interviews were conducted, other racial and ethnic groups were not well represented. Cultural contributors likely exist for different populations. Further, this study did not include non-English speaking families, contributing to selection bias and limiting generalization of results. Future research should conduct similar interviews performed with a greater range of racial and ethnic populations, as well as include translating interviews and conducting them with non-English speakers. We also did not have information about functional impairment and/or cognitive abilities of the children. However, we believe that those interviewed are representative of a general DS population seen at our institutions. Finally, our sample was drawn from academic pediatric settings with specialized sleep programs in cities in the U.S. How fully these findings apply to children and families in other care settings merits further investigation.

In summary, families’ experiences vary based on the child’s needs and behaviors, caregiver beliefs and approach to initiating new activities, social support, learning strengths or preferences, and interactions with the medical team that includes health insurance and equipment companies. Families will likely benefit from a structured, clearly defined interdisciplinary approach to PAP orientation and implementation that utilizes visual aids such as visual charts, social stories and videos specifically directed to individuals with DS that families can reference at home or share with the extended family during the implementation and desensitization process. With these enhancements to clinical care, the health and well-being of children with DS and their families can be optimally supported.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

Acknowledgements

We would like to acknowledge and give thanks to all the families who participated in the conduct of these interviews.

Financial Disclosures:

Dr. Xanthopoulos reports grant support from the National Institutes of Health/ National Heart Lung and Blood Institute (NIH/NHLBI) (R21 HD101003), outside of the submitted work. Dr. Barg reports grant support from NIH/National Institute of Mental Health (NIMH) (P50MH113840; R01MH108548; P30MH097488), NIH/NIDDK (K24AI146137), Commonwealth of Pennsylvania Department of Health, and Patient-Centered Outcomes Research Institute (PCORI), outside of submitted work. Dr. Byars reports grant support from NIH/ NHLBI (R01HL147915), outside of submitted work. Dr. Esbensen reports grant support from NIH/National Institute of Child Health and Human Development (NICHD) (R61HD100934, R01HD093754, R01HD099150, R01HD093654), outside of submitted work. Dr. Ishman reports grant support from the NIH/National Institute of Dental and Craniofacial Research (NIDCR) (U01DE029750), and Department of Defense (12662677), outside of the submitted work. Dr. Meinzen-Derr reports grant support from the NIH/National Institute on Deafness and Other Communication Disorders (NIDCD) (R01DC0185550), NIH/National Center for Advancing Translational Sciences (NCATS) (UL1TR001425), Center for Disease Control (CDC) (DD19-001-U01 DD001279, 5U19DD001218-02-00/19-3776), and Department of Defense (AR170121), outside of the submitted work. Dr. Tapia reports grants support from NIH/NHLBI (K01 HL130719, R01HL152454, R21HD101003), outside of the submitted work.

Role of Sponsors:

The funders did not have a role in the conduct of the study; in the collection, management, analysis, or interpretation of data; or in the preparation of the manuscript. The views expressed in this article are those of the authors and do not necessarily represent the views of the NIH.

Funding:

This work was supported by the National Institutes of Health (5R61HL151253)

Abbreviations:

DS	Down syndrome
MMRL	Mixed Methods Research Lab
OSAS	obstructive sleep apnea syndrome
PAP	positive airway pressure

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Highlights:

- Caregiver experiences starting PAP with their child with DS were heterogeneous
- Accessing supplies, mask fit, and comfort emerged as barriers to adherence
- Challenges communicating with the medical team and equipment company were a barrier
- Patient unique needs and behaviors emerged as themes influencing PAP experience
- Use of patience, visual aids, family education, and behavior plans were recommended

Table 1.

Demographics by high adherence and low adherence

	High Adherence N= 22	Low Adherence N= 19	p
Age at Screening, years, M (SD)	13.3 (3.4)	12.0 (4.2)	0.32
Gender (Female), N (%)	17 (77.3)	11 (57.9)	0.31
Race			
Caucasian, N (%)	16 (72.7)	13 (68.4)	0.42
Black/African American, N (%)	4 (18.2)	6 (31.6)	
Asian, N (%)	2 (9.1)	0 (0)	
Type of PAP, N (%)			
CPAP	8 (36.4)	4 (21.1)	0.035
BLPAP	1 (4.6%)	7 (36.8)	
AutoPAP	3 (59.0)	8 (42.1)	
Site			
Site 1, N (%)	11 (50)	9 (47.4)	1.0
Site 2, N (%)	11 (50)	10 (52.6)	
Caregiver Characteristics	N=22	N=18	
Age (years), M (SD)	46.9 (8.9)	46.3 (7.7)	0.82
Gender (Female), N (%)	22 (100)	18 (100)	1.0
Race			
Caucasian, N (%)	18 (81.8)	13 (68.4)	0.32
Black/African American, N (%)	4 (18.2)	6 (31.6)	
Median Income, M (SD)	73533.7 (26571)	71259.4 (28326.4)	0.80

CPAP, continuous positive airway pressure; BLPAP, bi-level positive airway pressure; AutoPAP, auto-titrating positive airway pressure.

Non-Parametric tested with Fisher's exact test

Parametric with t-test

Note: The number of children and number of caregivers differ by one because two children were adopted by the same family and the caregiver could not discriminate between the experiences of implementing PAP with both children so both children's information is reported but one caregiver provided information.