# TELEHEALTH TRAINING OF CAREGIVERS TO INCREASE POSITIVE AIRWAY PRESSURE (PAP) MACHINE USE IN ADULTS WITH DOWN SYNDROME

by

Emma Jean Walker, B.G.S.

MASTER'S THESIS

Presented to the Faculty of

The University of Houston-Clear Lake

In Partial Fulfillment

Of the Requirements

For the Degree

MASTER OF ARTS

in Behavior Analysis

THE UNIVERSITY OF HOUSTON-CLEAR LAKE

MAY, 2022

TELEHEALTH TRAINING OF CAREGIVERS TO INCREASE PAP MACHINE USE IN ADULTS WITH DOWN SYNDROME

by

Emma Jean Walker

APPROVED BY

_____

Dorothea Lerman, Ph.D., Chair

_____

Jennifer Fritz, Ph.D., Committee Member

_____

Sarah Lechago, Ph.D., Committee Member

RECEIVED/APPROVED BY THE COLLEGE OF HUMAN SCIENCES AND HUMANITIES:

_____

Samuel Gladden, Ph.D., Associate Dean

_____

Glenn Sanford, Ph.D., Dean

## Dedication

I dedicate this thesis to my parents, Wayne and Dianne, and my sisters, Abby and Izzy. My parents taught my sisters and I to celebrate differences and treat others with compassion from a young age. This shared value led me to persue a career working with individuals with a variety of neurodiverse backgrounds to achieve their goals. My families' love and support has been constant throughout this process. I cannot thank you all enough.

## Acknowledgements

I would like to express my utmost gratitude for my mentor, Dr. Dorothea Lerman, for her support and guidance throughout this project. I also thank Dr. John Berens and Dr. Fidaa Shaib at Baylor College of Medicine for their assistance with participant recruitment, data collection, and administrative tasks. Thank you to Dr. Greg Young who assisted with project development. I would also like to thank Jennifer Nguyen and Ashley Richmond for their assistance with data collection. I could not have completed this project without these individuals' help.

ABSTRACT

TELEHEALTH TRAINING OF CAREGIVERS TO INCREASE PAP MACHINE USE IN ADULTS WITH DOWN SYNDROME

Emma Jean Walker
University of Houston-Clear Lake, 2022

Thesis Chair: Dorothea Lerman, Ph.D.

A common sleep disorder among individuals with Down syndrome is obstructive sleep apnea (OSA). The most common treatment of OSA is Positive Airway Pressure (PAP) therapy. However, adherence to PAP therapy prescriptions is low. The high prevalence of OSA and low adherence with PAP therapy emphasized the need for an intervention to increase PAP machine use in individuals with DS. The following study evaluated a caregiver-implemented behavioral intervention aiming to increase PAP machine use in adults with DS using a multiple baseline across participants design. Participants in this study included three adults diagnosed with DS and OSA. The researchers utilized behavioral skills training via videoconferencing software to train the caregivers to implement the intervention. The intervention included the use of graduated exposure to the PAP therapy (i.e., slow progression of steps leading up to 4 hours of PAP machine use), differential negative reinforcement (i.e., longer breaks following compliance,

shorter breaks following noncompliance with the graduated exposure step), contingent positive reinforcement (i.e., rewards following compliance with the graduated exposure step) and noncontingent positive reinforcement (i.e., access to a leisure item during intervention sessions). This caregiver-implemented behavioral intervention was effective at increasing PAP machine use for all participants. The results of this study serve as preliminary results for the effectiveness of this behavioral intervention when implemented by caregivers.

TABLE OF CONTENTS

LIST OF TABLES

LIST OF FIGURES

# CHAPTER I:

# INTRODUCTION

Down syndrome (DS) is the condition indicated by the extra copy of chromosome 21 out of the 23 chromosomes humans have in their DNA (National Down Syndrome Society, n.d.). This genetic abnormality affects the development of an individual producing the common characteristics associated with DS such as small stature, low muscle tone, developmental delays, and learning disabilities. Individuals with DS also have predisposing factors to certain sleep problems. Factors such as midfacial hypoplasia, obesity, and poor muscle tone lead obstructive sleep apnea (OSA) to be prevalent in individuals with DS (Lee et al, 2018). Sixty-five percent to one hundred percent of adults with DS also have OSA, whereas 5-7% of typically developing adults are diagnosed with OSA (Gimenez et al, 2021). OSA is a sleep breathing disorder that occurs when an individual's airway is intermittently obstructed as the muscles in their upper throat relax during sleep. As the muscles relax, the airway closes and stops breathing causing brief arousals during sleep. These arousals can lead to high blood pressure, excessive daytime fatigue, heart problems, and impairments in cognitive functioning (Sawyer et al., 2019).

## Positive Airway Pressure (PAP) Therapy

The primary therapy for adults with OSA is positive airway pressure (PAP) therapy (Rotenberg et al, 2016). PAP therapy is often referred to as the "golden standard" of OSA treatment. PAP therapy treats symptoms of OSA via a machine that delivers pressurized air through the tubes attached to the mask. This positive pressure ensures that the individual's airway remains open and unobstructed when used during sleep. While PAP therapy is effective at alleviating OSA symptoms, this benefit is contingent on adherence to the recommended use of the PAP machine (Gimenez et al, 2021). PAP therapy adherence is widely referred to as 4 or more hours of PAP use at night in both

research and practice (Sawyer, et al, 2019). Adherence to PAP therapy has been found to reduce sleepiness during the day, normalize sleep architecture, lower blood pressure, improve cardiovascular health, and decrease many other side effects associated with OSA (Lance, 2020). Alternatively, less than 4 hours of PAP use at night is associated with risks of cardiovascular morbidity and mortality and increased impairments caused by OSA such as daytime fatigue (Sawyer, et al, 2019).

**PAP Therapy Adherence**

Although PAP therapy is the primary treatment for OSA, PAP therapy has been described as inconvenient and uncomfortable (Rotenberg et al, 2016). Several modifications have been made to the PAP machine with the goal of increasing machine portability, decreasing the sound of the machine, and increasing the comfortability of the mask, but adherence remains low. The percentage of adherence to PAP machine prescriptions, from 2011 to 2015, varied from 35% to 87% among those in the general population (Lance, 2020). Insufficient PAP therapy use is reported for almost one-third of typically developing adults with OSA (Gimenez et al, 2021).

Adherence to PAP therapy is equally as low in individuals diagnosed with DS compared to the typically developing population (Kang et al, 2019). However, individuals with DS may experience additional barriers that typically developing adults may not encounter. Individuals diagnosed with DS and OSA may have additional sensory or behavioral concerns that deter healthcare providers from recommending PAP therapy in the first place (Gimenez et al, 2021). Additionally, individuals with OSA and DS may require additional caregiver support to accurately set up, use, and maintain their PAP machine. The need for a caregiver to assist with proper PAP machine use can interfere with the feasibility of PAP therapy. It is widely recognized that the main limitation to PAP therapy is low adherence required to experience benefits (Rotenberg et al, 2016).

This emphasizes the need for research-based interventions to promote adherence to PAP therapy.

**Literature Review**

The number of empirically supported interventions aiming to increase PAP therapy adherence is scarce, but on an increasing trend (Weaver & Sawyer, 2010). Interventions to improve PAP therapy adherence can be categorized as supportive, educational, cognitive behavioral or a mixed strategy intervention. Currently, most interventions fall under the supportive category. Supports can vary in modality such as phone calls, physical documents, or follow-up appointments and are used to promote adherence to PAP therapy. However, pilot studies show no difference in adherence among participants receiving supports and the control group at two months (DeMolles et al., 2004; Stepnowsky et al., 2007). Overall, evaluations of education as an intervention to improve PAP therapy adherence show very little influence on PAP therapy adherence in patients with OSA (Weaver & Sawyer, 2010).

Several studies have examined cognitive behavioral interventions aiming to increase PAP therapy adherence. One cognitive-behavioral intervention included the use of individualized information about OSA, symptoms, treatment relevance, goal setting, symptom changes, trouble shooting, and goal refinement (Aloia et al, 2001). The individualized intervention was reported to influence self-efficacy and decision making to promote PAP therapy adherence. Differences in adherence among the intervention group and control group were not observed in the 1-week or 4-week follow up appointment. Upon the 12-week follow up appointment, the experimental group showed 3 more hours of PAP therapy than the control group. This supports the claim that cognitive behavioral interventions can be effective at increasing PAP adherence. Ultimately, research suggests that the most effective interventions for increasing PAP adherence are those that

incorporate multiple strategies (e.g., support, education, and cognitive behavioral interventions) to support the multidimensional nature of PAP adherence (Weaver & Sawyer, 2010).

Sawyer et al (2019) evaluated the effectiveness, feasibility, and acceptability of a multiple strategy intervention on PAP adherence. They created a social-cognitive individualized intervention to increase PAP adherence in typically developing adults diagnosed with OSA. The researchers conducted the intervention in four phases: pre-diagnosis, post-diagnosis, post-PAP titration, and the first week of at home PAP therapy. All intervention phases took place at the sleep center or was delivered via telephone (during the at-home PAP phase) and was implemented by a registered nurse. During the pre-diagnosis phase, participants were provided an educational video created by the American Sleep Apnea Association. During this phase, researchers guided the participants to select two to five social support resources, reasons the individual initiated sleep intervention, and goals for diagnosis and treatment. The post-diagnosis phase included a discussion of limitations regarding their OSA diagnosis, a review and continued revision of the goals for treatment, the PAP therapy introduction, mask-fitting, demonstration for application of mask, and the introduction of titration. A titration is an overnight sleep study conducted at the sleep clinic. The titration is used to determine the type of PAP therapy (e.g., CPAP, BiPAP) necessary for the patient. During the post-titration phase, the researchers reviewed the participant's bedtime routine and how to incorporate PAP therapy, barriers to PAP use and solutions on how to overcome them, risks for nonadherence, and the individual's personal goals. In the final phase (the first week of home PAP therapy), the researchers initiated daily contacts via telephone for the first two days. Finally, the researcher made a follow-up phone call the fifth day of PAP therapy at home. Phone contacts involved the discussion of concerns or barriers to PAP

therapy, a review of risks for nonadherence, and identifying the individual's social support resources. To show experimental control, the researchers compared the intervention group to the "usual care" (control) group. The participants randomly selected to experience "usual care" received the current standard practice for the diagnosis and treatment of OSA in adults. These practices included providing the patient with informational brochures regarding OSA, standard diagnostic testing, PAP therapy prescription, and contact information for the sleep center.

Results of this study indicated that the individualized social-cognitive intervention was a feasible and acceptable intervention (Sawyer et al, 2019). However, there was no significant difference in PAP therapy use between the intervention group and "usual care" group at 1 week, 1 month, or 3 months following the completion of the study. This treatment package is one of the few interventions aimed to increase adult PAP adherence that has been experimentally evaluated. While this study offers useful information on the initial steps to promote PAP adherence, it did not address already existing nonadherence.

Research on behavioral interventions to increase PAP adherence, specifically in individuals with DS, is extremely limited. Koontz et al (2003) developed a behavioral intervention aimed to increase PAP adherence in children. Participant's diagnoses varied. Of the 20 participants, five were diagnosed with either attention-deficit/hyperactivity disorder (ADHD) or DS. Trained researchers implemented the initial treatment sessions at a university-associated rehabilitation clinic before training the caregiver to implement the same procedures in the clinic and at home. The intervention consisted of contingent positive reinforcement, graduated exposure, pairing, and escape extinction. The researchers used contingent positive reinforcement in the form of praise and a tangible "prize" at the end of each session if the participant complied with PAP use. The researchers delivered tokens or stickers throughout the session as an immediate positive

reinforcer for younger participants or participants more developmentally delayed. These participants were allowed to exchange their tokens or stickers for a prize after the session. The researchers gradually increased the requirement for reinforcement until the participant willingly prepared for PAP machine use, assisted with placing the PAP mask on their face, and tolerated the mask and pressure until the individual fell asleep or the session was concluded. The researcher implemented graduated exposure by presenting the PAP machine and mask one step at a time in the order in which the stimuli fell in the PAP therapy sequence. The researcher combined these procedures (i.e., contingent positive reinforcement and graduated exposure) with pairing by providing the child access to a distracting and enjoyable activity (e.g., music, story, or movie) during the PAP therapy sessions. The researcher also utilized escape extinction in response to participant's noncompliance. Thus, when the participant attempted to remove the PAP machine mask or get up from the reclined chair, the researcher or caregiver physically blocked the removal of the mask or block the participant from getting up and vocal statements regarding noncompliance were ignored.

Koontz et al. (2003) found the intervention to be effective at increasing PAP therapy use for 75% of participants. The authors suggested that compliance with PAP therapy is influenced by the fit of the equipment, caregiver's consistent use of reinforcement, and the caregiver's correct implementation of the escape extinction component. Results indicated that this treatment package may be effective in increasing PAP adherence in children with and without intellectual and developmental disabilities (IDD). The authors emphasized the importance of utilizing behavior analysis principles and procedures to increase PAP therapy adherence.

The intervention evaluated by Koontz et al (2003) has not yet been systematically replicated with adults or in non-medical settings. The interventions described above have

been primarily implemented by researchers with little caregiver involvement. Behavioral interventions primarily implemented by caregivers may provide benefits that researcher-implemented interventions may lack. Caregivers have unlimited access to the individual that may allow the intervention to occur in the natural setting (i.e., during the bedtime routine) and many times throughout the day (Leaf, 2017). Among many other benefits of caregiver-implemented interventions, it also provides the caregiver an opportunity to play an active role in the individual's progress toward a goal. For caregivers to effectively implement a behavioral intervention, they must be extensively trained. This involves a therapist using evidence-based training procedures to teach the caregivers how to implement the intervention. The delivery of this training can take many forms based the nature of the training, the setting and modality. For example, trainings may take place in-person at a clinic, in-person in the home, or via telehealth in various environments. The current study evaluated the effectiveness of telehealth caregiver training on an intervention targeting PAP machine use in adults with DS.

The importance and value of telehealth training has increased since the start of the COVID-19 pandemic. Individuals are now able to continue to their healthcare services through video- and/or audio-conferencing software (i.e., Zoom™). Telehealth has been used in the field of applied behavior analysis for roughly two decades (Pollard et al, 2021). Pollard and colleagues evaluated the outcomes of transitioning from in-person services to telehealth caregiver training to reach more participants without increasing the frequency of in-person contact. The results proved that behavior analysts can effectively train caregivers and technicians to implement behavioral interventions via telehealth and maintain or increase target behaviors. Pollard and colleagues also found data to suggest that caregivers can effectively implement treatment programs for adaptive living skills when coached by a behavior analyst.

The current study evaluated the telehealth training of caregivers to implement a behavioral intervention targeting PAP machine use at night. Utilizing caregiver training via telehealth provided access to bedtime routines without requiring a therapist to be physically present. Conducting behavioral interventions in the home also avoids issues of generalization of behaviors from the clinical environment to the natural environment as the intervention can be implemented in the natural environment where the behavior should occur. Having caregivers implement this intervention in their respective homes would take advantage of the caregiver's unlimited access to the individual in their natural environment, decrease expenses related to travel to a clinic for the intervention, and increase accessibility to interventions for PAP adherence.

While there are many benefits to caregiver-implemented behavioral interventions, caregivers may not feel comfortable implementing certain behavioral procedures due to their desire to honor their child's personal autonomy and/or their own physical limitations. For example, caregivers may have difficulty implementing escape extinction with older individuals in the natural environment without hands-on assistance from a professional. Modifying the treatment components described by Koontz et al. (2003) may be a potential solution to this issue. For example, only using escape extinction as a last resort may increase the practicality and acceptability of caregiver-implemented interventions for increasing PAP adherence.

The purpose of this study was to extend the findings of Koontz et al. (2003) by utilizing telehealth training for caregivers of adults with DS and OSA to implement a behavioral intervention aimed to increase PAP adherence. The study aimed to evaluate the efficacy and feasibility of the intervention and to assess whether caregivers could effectively implement the treatment package in the home with the participant.

# CHAPTER II:

# METHOD

## Participants and Setting

Participants included three adults diagnosed with DS. Eligible participants were referred to the researcher by physicians and sleep medicine doctors at a university-based, interdisciplinary medical clinic for adults with intellectual and developmental disabilities. Eligible participants included individuals who (a) were prescribed a PAP machine, (b) typically fell asleep within 30 min (without PAP machine), (c) had a reported history of noncompliance with PAP machine use, (d) had a web-enabled device with a camera and adequate internet connection for videoconferencing sessions, and (e) were willing to meet one to three times per week for the duration of the study. The researchers excluded individuals with a comorbidity of DS and autism spectrum disorder or who engaged in severe problem behavior due to safety concerns.

Rory was a 23-year-old man diagnosed with DS and OSA. Rory resided with his parents. His primary caregiver involved in the study was his mother. Rory's mother purchased the PAP machine, but Rory reportedly never used the machine effectively. Rory's mother identified the primary barrier to effective PAP machine use was Rory refusing to wear the mask.

Ross was a 28-year-old man diagnosed with DS and OSA. Ross resided with his parents. His primary caregiver involved in the study was his mother. Ross's PAP machine was rented through a medical device company. Ross had rented the PAP machine for 1 month prior to the beginning of the study, but it was removed from the home due to the compliance requirement of health insurance companies. If a patient does not use the PAP machine for 4 hours or more per night during the first 90 days of possession, the insurance company has the right to remove the PAP machine from the

individual's home. At the beginning of the study, Ross did not have a PAP machine in their possession. Ross's mother identified the primary barrier to effective PAP machine use was Ross removing the mask after 30 min to 1 hour of PAP use.

Aliza was a 24-year-old woman diagnosed with DS and OSA. Aliza resided with her parents. Her primary caregiver involved in the study was her mother. Aliza rented her PAP machine from a medical device company. Aliza had previously rented a PAP machine prior to the study, but it was removed due to not meeting the health insurance compliance requirement. At the beginning of the study, Aliza did not have a PAP machine in their possession. Aliza's mother identified the primary barrier to effective PAP machine use was Aliza only tolerating the PAP machine for short periods of time at a lower pressure than prescribed by the doctor.

The researcher held all sessions via HIPAA-compliant videoconferencing software (Zoom™). Sessions were recorded for data collection purposes. Baseline sessions, night intervention sessions, and night terminal goal probes were conducted in the participant's bedrooms while they were laying down in bed. During the day intervention sessions, participants had the option to select where in their home they would like to use their PAP machine. Rory selected the living room for the location of intervention sessions during the day, and Ross and Aliza selected their bedrooms.

Videoconferencing sessions occurred one to three times per week. During participation in the study, Rory's caregiver had a family matter that interfered with conducting sessions regularly. Therefore, sessions did not occur one to three times per week, as desired. During the day intervention sessions, there were multiple two-week periods of time in which sessions would not occur due to this matter. For the first half of the day intervention sessions, sessions occurred one time per week. In the second half of day intervention sessions, sessions occurred two times per week. Upon the transition to

night sessions, sessions occurred two to three times per week, as desired. On average, all participants had two to three video conferencing sessions per week.

**Response Measurement, Interobserver Agreement, and Procedural Integrity**

The researchers collected data on multiple dimensions of the participant's behavior, including the duration of PAP machine use, PAP tolerance steps completed, and compliance with PAP graduated exposure steps. The researchers collected data on the PAP tolerance steps completed and compliance with the PAP graduated exposure steps via direct observation. Duration of PAP use was recorded using direct observation and via PAP machine downloaded data.

Data on participant's completion of PAP tolerance steps (Table 1) were collected for all baseline sessions and terminal goal probes and calculated into a percentage of PAP tolerance steps completed during the session. The completion of a step was defined as the participant tolerating the step for the entire duration without engaging in vocal or physical refusal. The percentage of PAP tolerance steps completed in a baseline session or terminal goal probe was calculated by dividing the number of PAP tolerance steps completed by the number of PAP tolerance steps presented and multiplying by 100.

Table 1:

*PAP Tolerance Steps*

| Step # | Tolerance Step |
|---|---|
| 1 | Mask on face for 30 s |
| 2 | Mask strapped on for 2 min |
| 3 | Tubing attached for 2 min (machine not on) |
| 4 | Machine turned on for 2 min |
| 5 | Machine turned on for 1 hour |
| 6 | Use PAP machine for a minimum of 4 hours at night |

Data on the participant's compliance with the PAP graduated exposure steps (Table 2) were collected during all intervention sessions. Compliance with a graduated exposure step was defined as tolerating the step within 5 s to 10 s of the caregiver's attempt to present the PAP mask/machine without engaging in vocal or physical refusal. The researchers calculated the percentage of compliance with the graduated exposure steps by dividing the number of graduated exposure steps the participant complied with by the total number of graduated exposure steps presented in an intervention session and multiplying by 100. The number of graduated exposure steps presented varied in the day intervention sessions from one step to 21 steps presented in each session. The number of graduated exposure steps in an intervention session was based on how many steps could be presented within a one-hour session.

Table 2:

*PAP Graduated Exposure Steps*

| Step # | Graduated Exposure Step |
|---|---|
| 1 | Mask within one foot of the participant for 2 min |
| 2 | Mask only on participant's hand for 3 s |
| 3 | Mask only on participant's elbow for 3 s |
| 4 | Mask only on participant's shoulder for 3 s |
| 5 | Mask only on participant's cheek for 3 s |
| 6 | Mask only on participant's nose for 3 s |
| 7 | Mask only on participant's nose for 10 s |
| 8 | Mask only on participant's nose for 15 s (straps on but not fastened) |
| 9 | Mask only on participant's nose for 30 s (straps on but not fastened) * |
| 10 | Mask and straps only on participant's nose for 30 s (straps on and fastened) |

| Step # | Graduated Exposure Step |
|---|---|
| 11 | Mask and straps only on participant's nose for 60 s (straps on and fastened) |
| 12 | Mask and straps only on participant's nose for 2 min (straps on and fastened) * |
| 13 | Mask, straps, and tubing on participant's face for 30 s |
| 14 | Mask, straps, and tubing on participant's face for 60 s |
| 15 | Mask, straps, and tubing on participant's face for 2 min * |
| 16 | Mask, straps, and tubing are connected to the machine (not on) for 30 s |
| 17 | Mask, straps, and tubing are connected to the machine (not on) for 60 s |
| 18 | Mask, straps, and tubing are connected to the machine (not on) for 2 min * |
| 19 | PAP machine use for 30 s |
| 20 | PAP machine use for 60 s |
| 21 | PAP machine use for 2 min * |
| 22 | Increase from 2 min of PAP use to 1 hour of PAP use<br>a) 2 min to 10 min increasing by 1 min<br>b) 10 min to 30 min increasing by 5 min<br>c) 30 min to 1 hour increasing by 10 min |
| 23 | 1 hour of PAP use at night * |
| 24 | Increase from 1 hour of PAP use to 4 hours of PAP use at night (increasing by 15 min) |
| 25 | 4 hours of PAP use at night * |
| | *Indicates step is equivalent to a PAP tolerance step |

The researchers also tracked noncompliance (i.e., physical and/or vocal refusal). A vocal refusal was defined as any communicative response indicating the individual would like to escape from the PAP machine (e.g., "No mask." or "No. Stop."). A physical refusal was defined as blocking the application of the PAP mask/machine and/or the removal of the PAP mask/machine. Mask removal was defined as the participant

removing the mask one inch or more from their face for 2 s or more. This excluded when the participant adjusted the mask or took a drink as long as the mask was replaced within 20 s. If the mask was not replaced within 20 s, it was considered noncompliance.

Duration of PAP use was recorded via direct observation until the participant fell asleep using the PAP machine. Following the participant falling asleep with the PAP machine on, data on the duration of PAP use were automatically recorded and downloaded from the PAP machine. For Ross and Aliza, the collaborating sleep clinic physicians obtained the PAP machine report and sent it to the researcher. Rory's caregiver connected the PAP machine data to a smart phone application to report the duration of PAP use for twelve intervention sessions. For all other intervention sessions, the researcher used direct observation or PAP machine downloads to record the duration PAP machine use for Rory. The primary observer recorded the duration of PAP machine use for baseline sessions, intervention sessions, and terminal goal probes.

**Interobserver Agreement**

A second independent observer collected data on completion of PAP tolerance steps during baseline and terminal goal probes for the purpose of calculating interobserver agreement (IOA). Point-by-point IOA was calculated by dividing the number of agreements in a specific session by the number of agreements plus disagreements in the session and multiplying by 100 to obtain a percentage of reliability. The researcher then averaged the IOA percentages for each session to obtain an average IOA for each response measurement. An agreement was defined as two observers recording the same response to a PAP tolerance step. A disagreement was defined as two observers recording different responses to the PAP tolerance step. IOA on the PAP tolerance steps completed was calculated for 50% of Rory's baseline sessions, 40% of Ross' baseline sessions, and 33% of Aliza's baseline sessions. IOA on the PAP tolerance

steps completed during baseline sessions for all participants was 100%. IOA on the PAP tolerance steps completed was calculated for 25% of terminal goal probes for all participants. IOA for the PAP tolerance steps completed during terminal goal probes was 100% for all participants.

A second independent observer collected data on compliance with PAP graduated exposure steps during intervention sessions for the purpose of calculating IOA. Point-to-point IOA was calculated using the same method as mentioned previously. An agreement was defined as two observers recording the same response to a PAP graduated exposure step. A disagreement was defined as two observers recording different responses to the PAP graduated exposure step. IOA for compliance with PAP graduated exposure steps was calculated for 27% of Rory's intervention sessions, 30% of Ross' intervention sessions, and 33% of Aliza's intervention sessions. IOA for compliance with graduated exposure steps was 100% for all participants.

The researcher used point-to-point IOA to calculate reliability. Total IOA was calculated using the same method as mentioned previously. An agreement was defined as two observers recording the same duration of PAP machine use within 30 s of one another. For example, if the primary observer recorded 3 min and 45 s of PAP use and the secondary observer recorded 3 min and 23 s of PAP use, it would be considered an agreement. A disagreement was defined as two observers recording different durations of PAP machine use of 30 s or more. For example, if the primary observer recorded 8 min of PAP use and the secondary observer recorded 6 min and 15 s of PAP use, it would be considered a disagreement. IOA on the duration of PAP use was calculated for 40% of Ross' baseline sessions and 25% of Aliza's baseline sessions. Rory did not use the PAP machine during baseline sessions. IOA on the duration of PAP use was 100% for Ross and Aliza. IOA on the duration of PAP machine use was calculated for 27% of Rory's

intervention sessions, 30% of Ross's intervention sessions, and 33% of Aliza's intervention sessions. IOA on the duration of PAP machine use during intervention sessions was 100% for all participants. IOA on the duration of PAP machine use was calculated for 25% of terminal goal probes for all participants. IOA on the duration of PAP machine use during terminal goal probes was 100% for all participants.

**Procedural Integrity**

Data on caregivers' correct implementation of the intervention were collected during all observed intervention sessions for the purposes of determining procedural integrity. Correct implementation for day intervention sessions was defined as (a) the participant having access to the noncontingent reinforcer prior to the presentation of the PAP mask or turning the PAP machine on, (b) the caregiver stating the target graduated exposure step before presenting the PAP mask or turning the PAP machine on, (c) removing the PAP mask within 15 s of the completed graduated exposure step or noncompliance, (d) providing the contingent reinforcer within 15 s of the completed graduated exposure step, (e) providing a 90-s to 2-min break following compliance before presenting another graduated exposure step, and (f) providing a 30-s to 45-s break following noncompliance before presenting another graduated exposure step. Table 3 lists the caregiver instructions for the correct implementation of the day intervention sessions.

Table 3:

*Caregiver Instructions (Day Intervention Sessions)*

| Step # | Instruction |
|---|---|
| 1 | Provide the participant the noncontingent positive reinforcer (i.e., leisure item) before presenting the PAP mask (Graduated exposure steps #1-18) or before turning on the PAP machine (Graduated exposure steps #18-22) |
| 2 | State the current target step before presenting the PAP mask (Graduated exposure steps #1-18) or before turning on the PAP machine (Graduated exposure steps #18-25) |
| 3 | Remove the PAP machine within 15 s of the end of the graduated exposure step or noncompliance |
| 4 | Provide the contingent positive reinforcer (i.e., reward or token) within 15 s of the end of the completed graduated exposure step |
| 5 | Provide a 90-s to 2-min break following compliance with the graduated exposure step before beginning another session |
| 6 | Provide a 30-s to 45-s break following noncompliance with the graduated exposure step before beginning another session |

Correct implementation for night intervention sessions was defined as (a) the participant having access to the noncontingent reinforcer prior to turning on the PAP machine for a minimum of 30 min and a maximum of 45 min, (b) the caregiver stating the current graduated exposure step prior to turning on the PAP machine, (c) removing the PAP mask within 15 s of the completed graduated exposure step (if participant is awake at the end of the step) or noncompliance, and (d) providing the contingent positive reinforcer within 15 s of the end of the graduated exposure step (if the participant is awake at the end of the step) or the following morning (if the participant is asleep at the end of the step). Table 4 lists the caregiver instructions for correct implementation of the night intervention sessions. An incorrect implementation was defined as the caregiver

failing to engage in the any of the previously mentioned behaviors during an intervention session.

Table 4:

*Caregiver Instructions (Night Intervention Sessions)*

| Step # | Instruction |
|---|---|
| 1 | Provide the participant the noncontingent positive reinforcer (i.e., leisure item) before turning on the PAP machine |
| 2 | State the current target step before turning on the PAP machine |
| 3 | Remove the PAP machine within 15 s of the end of the graduated exposure step (only if participant is awake at the end of the step) or noncompliance |
| 4 | Provide the contingent positive reinforcer (i.e., reward or token) within 15 s of the end of the completed graduated exposure step (if the participant is awake at the end of the step) or the following morning (if participant is asleep at the end of the step) |

The researcher calculated procedural integrity by dividing the number of steps the caregiver engaged in correctly by the number of steps the caregiver had the opportunity to engage and multiplying by 100. The researcher calculated IOA to determine the reliability of the measurement of procedural integrity. Point-to-point IOA was calculated using the same method as mentioned previously. An agreement was defined as two observers recording the same caregiver response during a given step of the caregiver instructions (Table 3 and Table 4). A disagreement was defined as two observers recording different caregiver responses during a given step of the caregiver instructions. IOA for procedural integrity was collected for 27% of Rory's, 30% of Ross's, 33% of Aliza's intervention sessions. IOA for procedural integrity was 92% for Rory and 100% for Ross and Aliza.

## Procedure

### Intake Appointment

The researcher conducted an intake appointment with each participant's primary caregiver. During this appointment, the researcher confirmed the participant's eligibility and the caregiver and participant's consent to participate. The researcher also obtained information regarding the participant's normal bedtime routine, history of PAP therapy, and other potential barriers to PAP therapy (e.g., chronic congestion, sleeping position). The researcher confirmed with each caregiver that they had received training on PAP machine use and routine maintenance prior to beginning the study.

### Preference Assessment

The researcher coached the caregiver to conduct a multiple stimulus without replacement preference assessment (DeLeon & Iwata, 1996) to systematically determine preferred items and/or activities to be used as the noncontingent positive reinforcer (i.e., leisure item/activity) and the contingent positive reinforcer (i.e., reward) used during intervention sessions and terminal goal probes.

A multiple stimulus without replacement (MSWO) preference assessment is an assessment created by DeLeon and Iwata (1996). During this assessment, the researcher instructed the caregiver to present the items in an equidistant array and instruct the participant to "pick one." After an item was selected, the participant had the opportunity to engage with the item for 1 min before the selected item was set to the side for the remainder of the assessment. The remaining unselected items were then presented again, and the participant was instructed to "pick one." This process was repeated until the participant had selected every item in the array or chose not to select any of the remaining items. Following the full assessment, the researcher created a hierarchy of preference for the items presented. The most preferred item from the MSWO was used as the contingent

positive reinforcer and the second or third most preferred item was used as the noncontingent reinforcer during intervention sessions.

Rory selected the iPad as his most preferred item and the "Ben 10" interactive toy as his second most preferred item. According to the results, the iPad should have been used as the contingent reinforcer. However, due to this participant's constant access to the iPad in his natural environment, the researcher selected the iPad as the noncontingent positive reinforcer and the "Ben 10" interactive toy as the contingent positive reinforcer. The researcher made this adjustment due to the caregiver's concern of evoking disruptive behavior due to the removal of the iPad that would interfere with sessions. After two intervention sessions, the "Ben 10" interactive toy was replaced with a fake dollar as the contingent positive reinforcer at the caregiver's request. The researcher instructed the caregiver to provide Rory a "dollar" following the progression to a new graduated exposure step (during day intervention sessions). The caregiver provided the participant the opportunity to exchange five "dollars" for a meal from a fast-food restaurant of his choice. Following the transition to night intervention sessions and night terminal goal probes, the noncontingent positive reinforcer was changed from the iPad to playing music on his smart speaker to promote sleeping while using the PAP machine. However, after five night intervention sessions, Rory requested the noncontingent positive reinforcer no longer be used. Rory's caregiver also began providing Rory a meal from a fast-food restaurant of his choosing the following day after an intervention session in which Rory complied with the graduated exposure step instead of using the fake "dollars" upon transitioning to night intervention sessions.

Ross selected Berenstain Bears books as his most preferred items and marching band videos as his second most preferred item. The researcher selected marching band videos as the noncontingent reinforcer. However, Ross' caregiver shared that Ross'

favorite treat were milkshakes. Therefore, due to this information, the researcher asked Ross if he would rather earn fake "dollars" to exchange for milkshakes or to earn reading his Berenstain Bears books. Ross reported that he preferred to earn the fake dollars. The researcher instructed the caregiver to provide Ross a "dollar" following compliance with the graduated exposure step. The caregiver provided the participant the opportunity to exchange three "dollars" for a milkshake from a fast-food restaurant of his choice. Following the transition to night intervention sessions and night terminal goal probes, the noncontingent positive reinforcer was changed from marching band videos to his caregiver reading books for 30 min to 45 min at the beginning of each session to promote sleeping while using the PAP machine. The contingent positive reinforcer remained the same.

Aliza selected her microphone as her most preferred item and her iPad as her second most preferred item. The researcher selected the iPad as the noncontingent positive reinforcer and the microphone as the contingent positive reinforcer. Following the transition to night intervention sessions and night terminal goal probes, the noncontingent positive reinforcer (iPad) was only provided for 30 min to 45 min at the beginning of each session to promote sleeping while using the PAP machine. The contingent positive reinforcer was also changed from the microphone to a fake "dollar" following compliance with the graduated exposure step. The caregiver provided Aliza the opportunity to exchange eight "dollars" for a meal from a fast-food restaurant of her choice. After reaching graduated exposure step #24 (i.e., 1 hour of PAP use at night), Aliza's caregiver requested that Aliza earn two "dollars" upon compliance with the graduated exposure step.

**Baseline Sessions**

The researcher trained the caregiver on the baseline procedures by providing written instructions, a visual model, and role-playing the procedures with the caregiver during videoconferencing sessions. The caregiver's responding had to meet a mastery criterion (100% correct responding for one role-play scenario) before implementing baseline sessions with the participant. Prior to the first baseline session, the researcher ensured the participant had followed a consistent bedtime routine for a minimum of 1-week. A consistent bedtime routine was defined as going to sleep within 30 min of the same time each night for a minimum of 1 week. This information was used to rule out an inconsistent bedtime routine as a barrier to PAP use.

The researcher observed all baseline sessions remotely during the participants' regular bedtime. During the first baseline session, the caregiver presented each PAP tolerance step shown in Table 1 by vocally stating the instruction (e.g., "First, we are going to place the mask on your face for 30 s.") before presenting the PAP mask/machine until the participant engaged in vocal or physical refusal or until the caregiver had presented all PAP tolerance steps. Following vocal or physical refusal, the researcher instructed the caregiver to terminate the baseline session by removing all PAP machine parts. Following the first baseline session, the caregiver only presented the PAP tolerance steps the participant did not complete in the previous baseline session. For example, during Ross' first baseline session, he completed PAP tolerance steps #1 (i.e., mask on face for 30 s) through #5 (i.e., 1 hour of PAP use). Thus, in the subsequent baseline sessions only tolerance step #6 (i.e., 4 hours of PAP use) was presented.

Baseline sessions were conducted until stable responding was observed (i.e., two or more data points at a stable trend or level) for the percentage of PAP tolerance steps completed and duration of PAP use. The researcher used the baseline data to determine a

starting point for the PAP graduated exposure steps in the intervention sessions. Rory had two baseline sessions. Ross had five baseline sessions. Aliza had four baseline sessions.

**Intervention Sessions**

The researcher trained the caregiver on the intervention procedures by providing written instructions, a visual model, and role-playing with the caregiver during videoconferencing sessions. The caregiver's responding had to meet a mastery criterion (100% correct responding for one role-play scenario) before implementing intervention sessions with the participant.

Intervention components included continuous access to a highly preferred item not contingent on any behavior (i.e., noncontingent positive reinforcement), slow progression through broken down PAP tolerance steps (i.e., PAP graduated exposure steps), rewards contingent on compliance with graduated exposure steps (i.e., contingent positive reinforcement) and longer breaks when the participant complied with the graduated exposure step and shorter breaks when the participant did not comply with the graduated exposure step (i.e., differential negative reinforcement). The number of graduated exposure steps presented during day intervention sessions varied based on the duration of the PAP graduated exposure step(s). Day intervention sessions were a maximum of 1 hour. Only one graduated exposure step was presented during night intervention sessions.

Each intervention session began with the caregiver providing the participant access to the noncontingent positive reinforcer (i.e., leisure item), as determined by the preference assessment. The participant had continuous access to this item throughout the day intervention sessions. After providing access to the leisure item, the caregiver presented the participant with a graduated exposure step (Table 2) by stating the target step prior to presenting the PAP mask or turning on the PAP machine.

During the first intervention appointment, the caregiver began by presenting the graduated exposure step that aligned with the participants' responding in baseline sessions. The researcher determined the initial graduated exposure step by identifying the tolerance step(s) the participant completed in the final baseline session, identifying the equivalent PAP graduated exposure step, and selecting the graduated exposure step that preceded the equivalent PAP graduated exposure step. For example, if the participant completed PAP tolerance step #1 (i.e., mask on face for 30 s) through #4 (i.e., 2 min of PAP use), the initial graduated exposure step was PAP graduated exposure step #20 (i.e., 1 min of PAP use). The initial graduated exposure step was step #1 (i.e., PAP mask within 1 foot of the participant) for Rory, step #22b (i.e., 18 min of PAP use) for Ross, and step #22a (i.e., 7 min of PAP use) for Aliza.

When targeting graduated exposure step #1 (i.e., PAP mask within 1 foot of participant) through #21 (i.e., 2 min of PAP use), the subsequent intervention sessions began with the caregiver's presentation of the last graduated exposure step with which the participant complied in the previous intervention session. If the participant was compliant with the step, the caregiver targeted the next step in the graduated exposure hierarchy. If the participant was noncompliant with the step, the caregiver was instructed to present the same step until the participant's responding met the criteria to move to the next step or to move back a step. Criterion to move to the next graduated exposure step was the participant complying with a given graduated exposure step for three consecutive attempts. Criterion to move back a graduated exposure step was the participant engaging in noncompliance with a given graduated exposure step for three consecutive attempts.

For intervention sessions targeting graduated exposure step #22 (i.e., increase from 2 min to 1 hour of PAP use) through step #25 (i.e., 4 hours of PAP use at night), the first step presented in succeeding intervention sessions was the next graduated exposure

step, if the participant was compliant with the final graduated exposure step in the previous intervention session. If the participant was noncompliant with the final graduated exposure step in the previous intervention session, the same graduated exposure step was presented again until the participant's responding met the criterion to move onto the next graduated exposure step or move back a step. Criterion to move to the next graduated exposure step was the participant complying with a given graduated exposure step. Criterion to move back a graduated exposure step was the participant engaging in noncompliance with a given graduated exposure step for three consecutive attempts.

Differential negative reinforcement was used in the form of longer breaks following compliance and shorter breaks following noncompliance with the graduated exposure step. The researcher instructed the caregiver to provide the participant a break from the PAP machine/parts for a minimum of 90 s and a maximum of 2 min contingent on the participant's compliance with the graduated exposure step before presenting another graduated exposure step. The caregiver also provided the participant a contingent positive reinforcer (i.e., reward) when the participant was compliant with the graduated exposure step. If the participant engaged in vocal or physical refusal, the caregiver removed the PAP machine/parts for a minimum of 30 s and a maximum of 45 s before presenting another graduated exposure step. Breaks following compliance or noncompliance was excluded during the final graduated exposure step presented in an intervention session and intervention sessions with only one graduated exposure step presented (i.e., night intervention sessions).

Upon completion of graduated exposure step #21 (i.e., 2 min of PAP use), the researcher progressed from 2 min of PAP use to 1 hour of PAP use by increasing the target duration of PAP use by 1 min upon compliance until targeting 10 min of PAP use

(i.e., PAP tolerance step #22a). Upon compliance with 10 min of PAP use, the researcher increased the target duration of PAP use by 5 min upon compliance until targeting 30 min of PAP use (i.e., PAP tolerance step #22b). Upon compliance with 30 min of PAP use, the researcher increased the target duration of PAP use by 10 min upon compliance until targeting 1 hour of PAP use (i.e., PAP tolerance step #22c). Upon compliance with 1 hour of PAP machine use during a day intervention session or a day terminal goal probe, the researcher transitioned to nighttime intervention sessions.

Night intervention sessions occurred after the participants' normal bedtime routine while the participant was lying in bed. During night intervention sessions, the caregiver provided the noncontingent positive reinforcer (i.e., leisure item) prior to turning on the PAP machine. The participant had access to the leisure item for the first 30 to 45 min of the night intervention session. After the first 30 to 45 min, the leisure item was put away to promote sleeping while using the PAP machine for the rest of the graduated exposure step. The caregiver presented the graduated exposure step by stating the target duration of PAP use prior to turning on the PAP machine. If the graduated exposure step was longer than 1 hour and 30 min, the researcher ended the video call after 1 hour and 30 min or when the participant fell asleep using the PAP machine. When this occurred, the researcher relied on the PAP machine data to record the duration of PAP use and determine compliance with the graduated exposure step. When targeting 1 hour and 30 min or less of PAP use, the caregiver provided the contingent positive reinforcer (i.e., reward) within 15 s of the completed step. When targeting more than 1 hour and 30 min of PAP use, the caregiver provided the contingent reinforcer the following morning. If the participant engaged in noncompliance at any time during the night intervention session, the caregiver was instructed to allow the participant to remove the PAP mask/machine and not provide any additional attention regarding the PAP

machine/therapy. Differential negative reinforcement in the form of breaks from the PAP machine was not used during night intervention sessions.

The researcher included a stringent criterion for the inclusion of escape extinction. Escape extinction is a more intensive intervention component that involves the caregiver continuously prompting following noncompliance with a graduated exposure step, as opposed to providing a 30-s break following noncompliance. This criterion was six consecutive graduated exposure steps with noncompliance before escape extinction was implemented. If any of the participants were to require extinction, the researcher would have trained the caregiver to use a least-to-most prompting strategy. The caregiver would have prompted using a verbal instruction plus a model prompt (i.e., showing the participant the application of the PAP mask) before physically assisting with the application of the PAP mask. However, none of the participants' responding met this criterion or required the use of extinction.

The researcher was present via videoconferencing software during night intervention sessions and night terminal goal probes for a maximum of 1 hour and 30 min. However, the researcher ended the video call upon the participant falling asleep with the PAP machine to promote natural PAP therapy use. When the graduated step was longer than 1 hour and 30 min in duration (i.e., graduated exposure steps #24 and #25) or when the participant fell asleep, the rest of the data (i.e., duration of PAP use) was automatically recorded by the PAP machine. Based on the machine-recorded data, the researcher determined if the participant complied with the PAP graduated exposure step. The researcher relied on caregiver report for the delivery of reinforcement contingent on compliance with the graduated exposure step when the videoconferencing session ended before the graduated exposure step was complete.

**Independent Intervention Sessions**

Upon the transition to night intervention sessions, the researcher trained the caregiver to conduct graduated exposure sessions independent of the researcher. The researcher instructed the caregiver to conduct no more than three independent sessions per week on the days or nights there was not videoconferencing sessions. During these independent sessions, the caregiver followed the written instructions provided by the researcher and targeted the last step the participant was compliant with in the previous videoconferencing session. For example, if the participant was compliant with using the PAP machine for 40 min in the previous intervention session, the caregiver implemented independent sessions targeting 40 min of PAP use until the next videoconferencing session with the researcher present. All independent intervention session procedures remained the same as night intervention sessions with the researcher. The caregiver informed the researcher when an independent session was conducted, and the researcher obtained the machine downloaded data for data collection purposes. This component of the intervention was not required for participation. Rory's caregiver conducted eight independent sessions, Ross's caregiver conducted two independent sessions, and Aliza's caregiver did not conduct independent sessions.

**Terminal Goal Probes**

The researcher conducted a probe of the terminal goal after every four intervention sessions and upon the transition from day intervention sessions to night intervention sessions. The purpose of the terminal goal probes was to assess generalization of compliance from targeted graduated exposure steps to untargeted graduated exposure steps in the form of longer PAP use duration in a terminal probe than in the previous intervention session. In other words, the researcher was evaluating if the participant's responding in a terminal goal probe would allow the researcher to skip some

of the graduated exposure steps. Based on the participants compliance during terminal goal probes, the researcher identified the equivalent graduated exposure step and advanced to the graduated exposure step that proceeded it. For example, if the participant was targeting 2 hours of PAP use during night intervention sessions and during the terminal goal probe the participant complied with 2 hours and 25 min of PAP use, the researcher would target 2 hours and 24 min of PAP use. If the participant tolerated a shorter duration of PAP use in the terminal goal probe than in the preceding intervention session, the researcher continued to progress through the graduated exposure steps in the following intervention sessions based on compliance in the previous intervention session, as described previously (See Intervention Sessions).

Upon the participant complying with 1 hour of PAP use in an intervention session or a day terminal goal probe, the researcher initiated a night terminal probe session following the participant's bedtime routine to determine the first graduated exposure step for night intervention sessions. The researcher was present during all terminal goal probes via videoconferencing software.

During terminal goal probes, the caregiver provided the participant access to the noncontingent positive reinforcer (i.e., leisure item) before presenting the PAP mask/machine. The caregiver presented the tolerance step that corresponded with the terminal goal for the day intervention sessions or night intervention sessions. For day intervention sessions, the terminal goal was 1 hour of PAP machine use (i.e., tolerance step #5). For night intervention sessions, the terminal goal was 4 hours of PAP use at night (i.e., tolerance step #6). If the participant was compliant with the day terminal goal (i.e., 1 hour of PAP use), the caregiver provided the contingent positive reinforcer before ending the session. If the participant was compliant with the night terminal goal (i.e., 4 hours of PAP use at night), the caregiver provided the contingent positive reinforcer the

following morning. If the participant was noncompliant with the day or night terminal goal, the caregiver did not provide the contingent positive reinforcer.

**Social Validity**

Following the completion of the study, the researcher delivered two modified versions of the Treatment Acceptability Rating Form (TARF) to the participating caregivers via email (Reimers and Wacker, 1988). One version of the modified TARF was for the caregiver about the acceptability and feasibility of the intervention and satisfaction of the results (See Appendix A). The other version of the modified TARF was for the participant but implemented by the caregiver about the participants' satisfaction of the format of the meetings with the researcher and the caregiver-implemented sessions (See Appendix B). The caregiver presented each question and the answer options in the TARF to the participant before recording the participant's response. No participants have reached this portion of the study at this time.

**Experimental Design**

A single subject, nonconcurrent, multiple baseline across participants design was used to demonstrate experimental control and evaluate the effectiveness of this intervention. A changing-criterion design was embedded in the intervention phase of this study by systematically increasing the response effort (i.e., the duration of PAP use) required to receive the contingent positive reinforcer (i.e., reward).

# CHAPTER III:

# RESULTS

Results of this study will be depicted using visual analysis via line graphs and described in this section. Data collection is still ongoing for all participants. However, the results thus far are shown below.

## Rory

Figure 1 depicts Rory's percentage of PAP tolerance steps completed during baseline and terminal goal probes in the top panel and the results of the participant's percentage of compliance with PAP graduated exposure steps in the bottom panel

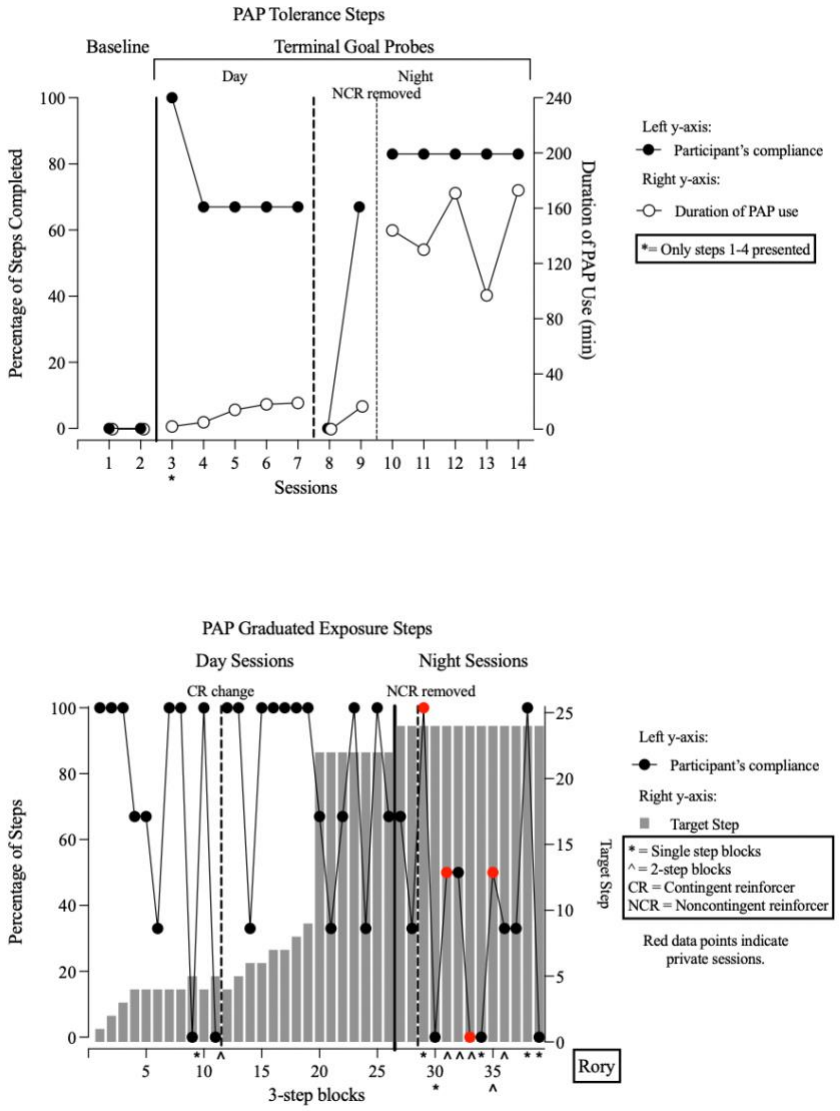


*Figure 1:*
*Rory's PAP Tolerance Steps and PAP Graduated Exposure Steps graphs*

The top panel of the figure depicts the percentage of PAP tolerance steps Rory completed during baseline sessions and terminal goal probes. The x-axis depicts sessions. The left y-axis depicts the percentage of steps completed and the right y-axis depicts the duration of PAP use in minutes. The right y-axis ranges from zero to 240 min to represent the 4 hours (i.e., 240 min) terminal goal. Denoted by the closed circles is Rory's compliance and denoted by the white circles is the duration of PAP use. Asterisks

indicate only tolerance steps #1 through #4 (i.e., 2 min of PAP use) were presented due to a scheduling conflict.

During both baseline sessions, Rory engaged in noncompliance upon the delivery of the first tolerance step (i.e., Mask on face for 30 s). Therefore, there were no completed tolerance steps and no PAP use in baseline sessions. The researcher began with graduated exposure step #1 (i.e., mask within 1 foot) in the first intervention session. The bottom panel of the figure depicts Rory's compliance with the PAP graduated exposure steps during day and night intervention sessions. The x-axis depicts the graduated exposure steps in 3-step blocks. This means each value on the x-axis represented three graduated exposure steps unless indicated otherwise. The left y-axis depicts the percentage of steps, and the right y-axis depicts the target graduated exposure step. The right y-axis ranges from zero to 25 to represent the 25 graduated exposure steps. Denoted by the black circles is Rory's compliance and denoted by the gray bars is the target graduated exposure step. Asterisks indicate the block consisted of only one graduated exposure step. Arrows indicate the block consisted of two graduated exposure steps. When referring to block numbers, the researcher is discussing the PAP graduated exposure steps graph.

Rory's initial intervention session began with the presentation of the first graduated exposure step (i.e., mask within 1 foot). Rory's percentage of compliance during the first eleven 3-step blocks (i.e., blocks #1-11) was high before gradually decreasing and maintaining at high variability. After observing the varying levels of compliance, Rory's caregiver requested to replace the contingent positive reinforcer from a "Ben 10" interactive toy to earning fake "dollars" to exchange for a meal at a fast-food restaurant of his choosing. When targeting PAP graduated exposure steps #1 through #21, the contingent reinforcer (i.e., "dollar") was provided following compliance with a PAP

graduated exposure step for three consecutive attempts. When targeting PAP graduated exposure steps #22 through #25, the contingent reinforcer was provided following compliance with each graduated exposure step. Following this change, Rory's percentage of compliance remained at high levels (92% average) with low variability (i.e., blocks #12-19).

The first terminal goal probe was conducted following Rory's first four intervention sessions (see top panel of figure). During this terminal goal probe (i.e., session #3), only PAP tolerance steps #1 through #4 were presented due to a scheduling conflict. During this terminal goal probe, Rory tolerated 2 min of PAP use. The researcher conducted another terminal goal probe following this to assess Rory's compliance with the day terminal goal (i.e., 1 hour of PAP use). During this terminal goal probe (i.e., session #4), Rory tolerated 5 min of PAP use (i.e., 67% of the tolerance steps completed). Therefore, the researcher jumped ahead to targeting graduated exposure step #22a (i.e., 4 min of PAP use) in the following intervention session (i.e., block #20). Rory's compliance with the PAP graduated exposure steps decreased (55% average) and showed moderate variability during blocks #20 through #22.

During the third terminal goal probe (i.e., session #5), Rory tolerated 14 min and 16 s of PAP use. Based on Rory's compliance, the researcher continued with intervention sessions by jumping ahead graduated exposure steps and targeting graduated exposure step #22b (i.e., 13 min of PAP use, increasing by 5 min). Rory's compliance with the PAP graduated exposure steps increased to moderately high percentages (75% average) with high variability blocks #22 through #25.

Due to researcher error, the next terminal goal probe occurred six intervention sessions after the previous terminal goal probe, instead of after four intervention sessions. During this fourth terminal goal probe (i.e., session #6), Rory complied with less PAP

machine use (i.e., 18 min and 6 s of PAP use) than in the previous intervention session (i.e., 53 min of PAP use). The researcher continued with intervention sessions by presenting the same graduated exposure step as in the previous intervention session (i.e., 53 min of PAP use). After observing compliance with 53 min of PAP use in the intervention sessions, the researcher conducted another terminal goal probe to assess if words to describe the terminal goal affected Rory's compliance. During the fifth terminal goal probe (i.e., session #7), the researcher instructed the caregiver to state the terminal goal was "60 minutes" instead of "one hour." However, Rory complied with less PAP machine use (i.e., 19 min and 5 s of PAP use) than in the previous intervention session (i.e., 53 min of PAP use) again. Therefore, the researcher continued with the next intervention session targeting 53 min of PAP use (i.e., step #1 of block #26). Upon compliance with 1 hour of PAP use in block #26, the researcher transitioned to night sessions by conducting a night terminal goal probe. During the first night terminal goal probe (i.e., session #8), engaged in noncompliance before the machine was turned on. Therefore, the terminal probe session was ended.

Upon the transition to night intervention sessions, the noncontingent positive reinforcer was changed from the iPad to music on his interactive speaker. However, following the second graduated exposure step in block #28, the noncontingent reinforcer was no longer provided at Rory's request. He shared that he preferred to sleep with the PAP machine instead of listening to music. Rory's caregiver also began providing Rory a meal from a fast-food restaurant of his choosing the following day after an intervention session in which Rory complied with the graduated exposure step instead of using the fake "dollars".

During the first night intervention session, Rory's caregiver presented graduated step #23 (i.e., 1 hour of PAP use at night) due to Rory completing 1 hour of PAP use

during the last day intervention session (i.e., graduated exposure step #22c). Rory's compliance in the first two session blocks following the transition to night intervention sessions (i.e., block #27 and #28) maintained at 67% compliance. Following the first four night intervention sessions, the researcher conducted another terminal goal probe. During this terminal goal probe (i.e., session #9), Rory complied with 15 min and 27 s of PAP use (i.e., 67% of tolerance step completed). In the following night intervention session, the researcher continued with the graduated exposure step based on the previous intervention session (i.e., 1 hour and 45 min of PAP use).

Following the removal of the noncontingent reinforcer (i.e., music on the interactive speaker) at Rory's request, his compliance maintained at high variability. However, this could be due to the variation in graphing conventions for blocks #29-36. These blocks were graphed in one and two-step blocks to separate the videoconferencing intervention sessions from the independent intervention sessions. Figure 2 will provide further information on Rory's compliance with the graduated exposure steps in the form of duration of PAP use during intervention sessions which may help clarify the trends in Rory's compliance.

In the first independent intervention session (i.e., block #29), Rory's caregiver presented graduated exposure step #24 (i.e., 2 hours of PAP use at night). Rory complied with 2 hours and 11 min of PAP use. Rory surpassed the graduated exposure step (i.e., 2 hours of PAP use) due to him sleeping when the target step ended. During the next terminal goal probe (i.e., session #10), Rory complied with longer PAP use (i.e., 2 hours and 24 min of PAP use) than in the previous intervention session (i.e., 2 hours and 15 min of PAP use). Therefore, the researcher jumped ahead to targeting 2 hours and 23 min of PAP use in the following intervention session. Rory's compliance in the session blocks following the first independent session (i.e., blocks #30-#34) decreased (20% average)

with moderate variability. Due to Rory's noncompliance with the graduated exposure step #24 (i.e., 2 hours and 38 min of PAP use) for three consecutive sessions, the researcher instructed the caregiver to present the previously targeted step (i.e., 2 hours and 23 min of PAP use) during the next intervention session.

During the next terminal goal probe (i.e., session #11), Rory complied with 2 hours and 23 min of PAP use (i.e., 83% of tolerance steps completed). Following this, Rory's compliance increased (66% average) in the next two blocks (i.e., blocks #35 and #36). During the next terminal goal probe (i.e., session #12), Rory complied with 2 hours and 51 min of PAP use (i.e., 83% of tolerance steps completed). Thus, the caregiver targeted 2 hours and 50 min of PAP use (i.e., graduated exposure step #24) during the next video conferencing intervention session. In the following terminal goal probe (i.e., session #13), Rory tolerated 1 hour and 37 min of PAP use. Therefore, the researcher continued with the progression of the steps and targeted 3 hours and 5 min of PAP use in the following session. In blocks #35 and #39, Rory's percentage of compliance slightly increased to 43% average compliance with PAP graduated exposure steps. In the next terminal probe (i.e., session #14), Rory tolerated 2 hours and 53 min of PAP use. In the most recent intervention session, Rory's caregiver conducted an independent intervention session targeting 3 hours and 20 min of PAP use. Rory complied with 2 hours and 15 min of PAP use. Therefore, in succeeding sessions, the researcher will continue with intervention sessions targeting 3 hours and 20 min of PAP use.

Figure 2 depicts Rory's duration of PAP machine use during intervention sessions. The x-axis depicts sessions. The y-axis depicts the duration of PAP use in minutes. The y-axis ranges from zero to 240 min to represent the 4 hours (i.e., 240 min) terminal goal. Denoted by the closed circles is Rory's compliance duration and denoted by the solid dash is the target graduated step duration.

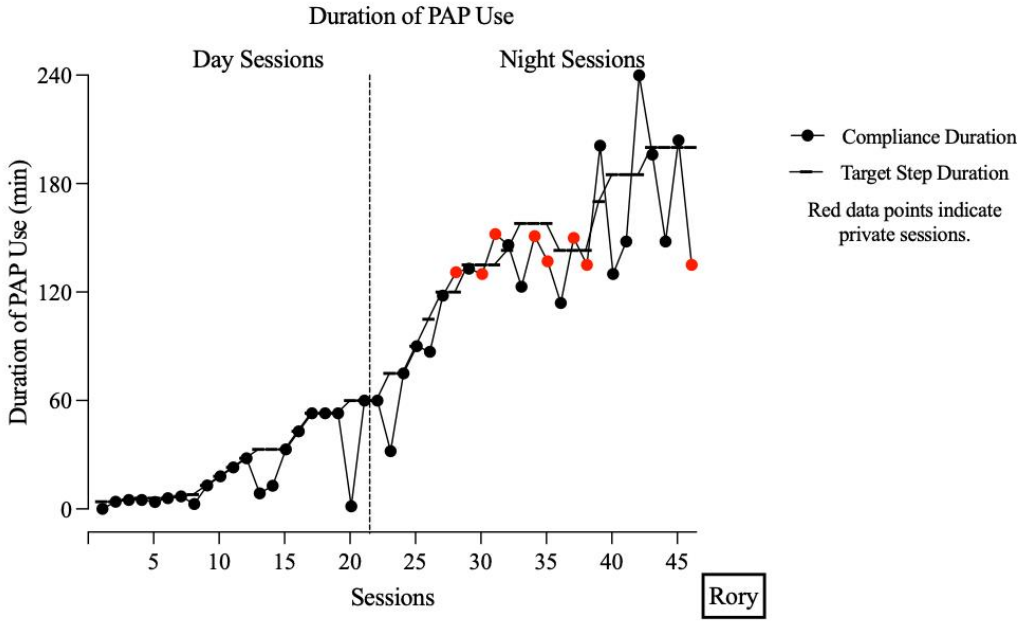


*Figure 2:*
*Rory's Duration of PAP Use*

Session #1 on Figure 2 represents block #20 in the PAP Graduated Exposure Step graph (i.e., bottom panel of Figure 1). Rory's compliance duration remained at nearly the same level as the target step duration until session #13. Rory engaged in noncompliance after 8 min and 35 s in session #13 and after 12 min and 50 s in session #14. In session #15 through #19, Rory's compliance met the target step duration. In session #20, Rory engaged in noncompliance after 1 min and 27 s of PAP use before meeting the target step duration (i.e., 1 hour of PAP use) in session #21. In the first session following the transition to night sessions (i.e., session #22), Rory's compliance remained at the same level as the target step duration before engaging in noncompliance in the following session (i.e., session #23). Session #24 though session #32 compliance remained at nearly the same level as the target duration. However, during sessions #33, #34, and #35, Rory did not comply with graduated exposure step #24 (i.e., 2 hours and 38 min of PAP use).

Therefore, the researcher instructed the caregiver to move back to the previous targeted graduated exposure step (i.e., 2 hours and 23 min of PAP use). Following this change (i.e., sessions #36-38), Rory's compliance occurred at variable levels with meeting the target duration in session #35. The target duration increased after session #36 due to Rory's high level of compliance (i.e., 2 hours and 51 min of PAP use) in the preceding terminal goal probe. In session #39, Rory surpassed the target duration (i.e., 3 hours and 21 min of PAP use) due to him sleeping at the end of the target duration. In session #41, Rory's compliance duration increased to 4 hours and 25 min. Thus, Rory's compliance met the terminal goal for the first time. In sessions #39 through #45, Rory's duration of PAP use remained moderately variable.

**Ross**

Figure 3 depicts Ross' percentage of PAP tolerance steps completed during baseline and terminal goal probes in the top graph and the participant's percentage of compliance with PAP graduated exposure steps in the bottom graph.

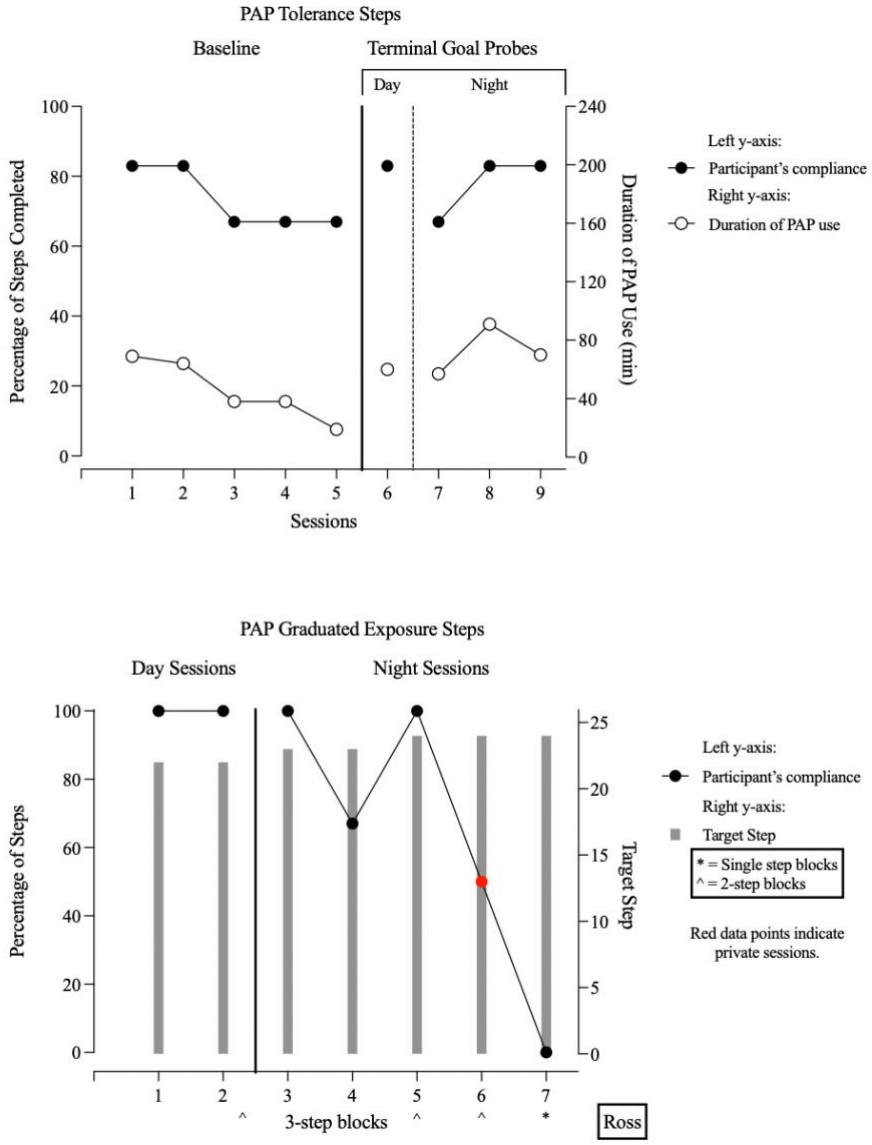


*Figure 3:*
*Ross' PAP Tolerance Steps and PAP Graduated Exposure Steps graphs*

The top panel of the figure depicts the percentage of PAP tolerance steps Ross completed during baseline sessions and terminal goal probes. The x-axis depicts sessions. The left y-axis depicts the percentage of steps completed and the right y-axis depicts the duration of PAP use in minutes. The right y-axis ranges from zero to 240 min to represent the 4-hour (i.e., 240 min) terminal goal. Denoted by the closed circles is Ross'

compliance and denoted by the white circles is the duration of PAP use. When referring to session numbers, the researcher is discussing the PAP tolerance steps graph.

During the first two baseline sessions (i.e., session #1 and #2), Ross tolerated 1 hour and 9 min of PAP use and 1 hour and 4 min of PAP use, respectively (i.e., 83% of tolerance steps completed). In sessions #3 and #4, Ross tolerated 38 min of PAP use (i.e., 67% of tolerance steps completed). In session #5, Ross tolerated 19 min of PAP use (i.e., 67% of tolerance steps completed). Therefore, the researcher targeted 18 min of PAP use in the first intervention session.

The bottom panel of the figure depicts Ross' compliance with the PAP graduated exposure steps during day and night intervention sessions. The x-axis depicts the graduated exposure steps in 3-step blocks. This means each value on the x-axis represented three graduated exposure steps unless indicated otherwise. The left y-axis depicts the percentage of steps, and the right y-axis depicts the target graduated exposure step. The right y-axis ranges from zero to 25 to represent the 25 graduated exposure steps. Denoted by the black circles is Ross' compliance and denoted by the gray bars is the target graduated exposure step. Asterisks indicate the block consisted of only one graduated exposure step. Arrows indicate the block consisted of two graduated exposure steps. When referring to block numbers, the researcher is discussing the PAP graduated exposure steps graph.

Ross' caregiver presented graduated exposure step #22b (i.e., 18 min of PAP use) in the first intervention session (i.e., block #1). Ross was 100% compliant with the graduated exposure steps presented in the first four intervention sessions (i.e., blocks #1 and #2). During the first terminal goal probe (i.e., session #6), Ross tolerated 1 hour of PAP use (i.e., 83% of tolerance steps completed). Therefore, the researcher transitioned to night sessions by initiating a night terminal goal probe. During this probe (i.e., session

#7), Ross tolerated 57 min of PAP use (i.e., 67% of tolerance steps completed). Thus, the following night intervention session (i.e., block #3) targeted 56 min of PAP use (i.e., graduated exposure step #23).

Upon the transition to night sessions, the noncontingent positive reinforcer was changed from marching band videos to his caregiver reading books to him for the first 30 to 45 min of the session. In the first three night intervention blocks (i.e., blocks #3 through #5), Ross' compliance remained at high percentages with little variability. During the second night terminal goal probe (i.e., session #8), Ross tolerated 1 hour and 31 min of PAP use (i.e., 83% of tolerance steps completed). This was a shorter duration of PAP use than in the previous intervention session.

Due to researcher error, Ross' caregiver presented graduated exposure step #24 (i.e., 1 hour and 36 min) in the following intervention session. The correct graduated exposure step was 1 hour and 51 min of PAP use, because in the previous intervention session (i.e., step #1 in block #4), Ross complied with 1 hour and 36 min of PAP use (i.e., graduated exposure step #24). In the following intervention session (i.e., step #2 in block #4), Ross did not complete the 1 hour and 36 min of PAP use. Therefore, Ross' caregiver presented this step again in the following session (i.e., step #3 in block #4). In block #5, Ross complied with 100% of the graduated exposure steps.

During the next terminal goal probe (i.e., session #9), Ross tolerated 1 hour and 10 min of PAP use. In block #6, Ross' caregiver conducted the first independent session targeting 2 hours and 6 min of PAP use. Due to Ross sleeping when the end of the target duration was over, his duration of PAP use was 2 hours and 43 min. Therefore, compliance in this block was 100%. In the second independent intervention session, Ross' caregiver targeted 2 hours and 6 min of PAP use, and Ross tolerated 1 hour and 51 min of PAP use. Due to Ross complying with 2 hours and 6 min of PAP use in the first

independent intervention session, the researcher targeted 2 hours and 21 min in the following videoconferencing intervention session. However, during this session, Ross complied with only 1 hour and 36 min of PAP use.

Figure 4 depicts Ross' duration of PAP machine use during intervention sessions. The x-axis depicts sessions. The y-axis depicts the duration of PAP use in minutes. The y-axis ranges from zero to 240 min to represent the 4 hours (i.e., 240 min) terminal goal. Denoted by the closed circles is Ross' compliance duration and denoted by the solid dash is the target graduated step duration.

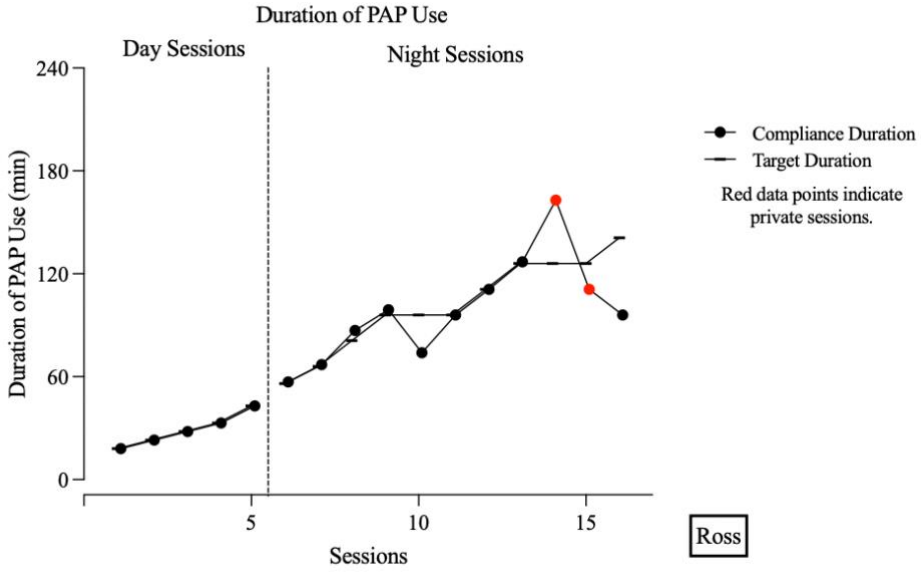


*Figure 4:*
*Ross' Duration of PAP Use*

Session #1 on Figure 4 represents block #1 in the PAP Graduated Exposure Step graph (i.e., bottom panel of Figure 3). Ross' duration of PAP use matched the target duration from session #1 through session #9. In session #10, Ross complied with 1 hour and 14 min of PAP use, but the target duration was 1 hour and 36 min of PAP use. Therefore, in the following session, the caregiver presented the same graduated exposure

step again (i.e., 1 hour and 36 min of PAP use). In sessions #11 to #13, Ross' duration of PAP use matched the target duration. In session #14, Ross tolerated 2 hours and 43 min of PAP use due him sleeping past the target duration of PAP use. In sessions #15 and #16, Ross complied with 15 min and 30 min less than the target duration, respectively.

**Aliza**

Figure 5 depicts Aliza's percentage of PAP tolerance steps completed during baseline and terminal goal probes in the top panel and her percentage of compliance with PAP graduated exposure steps during intervention sessions in the bottom panel.

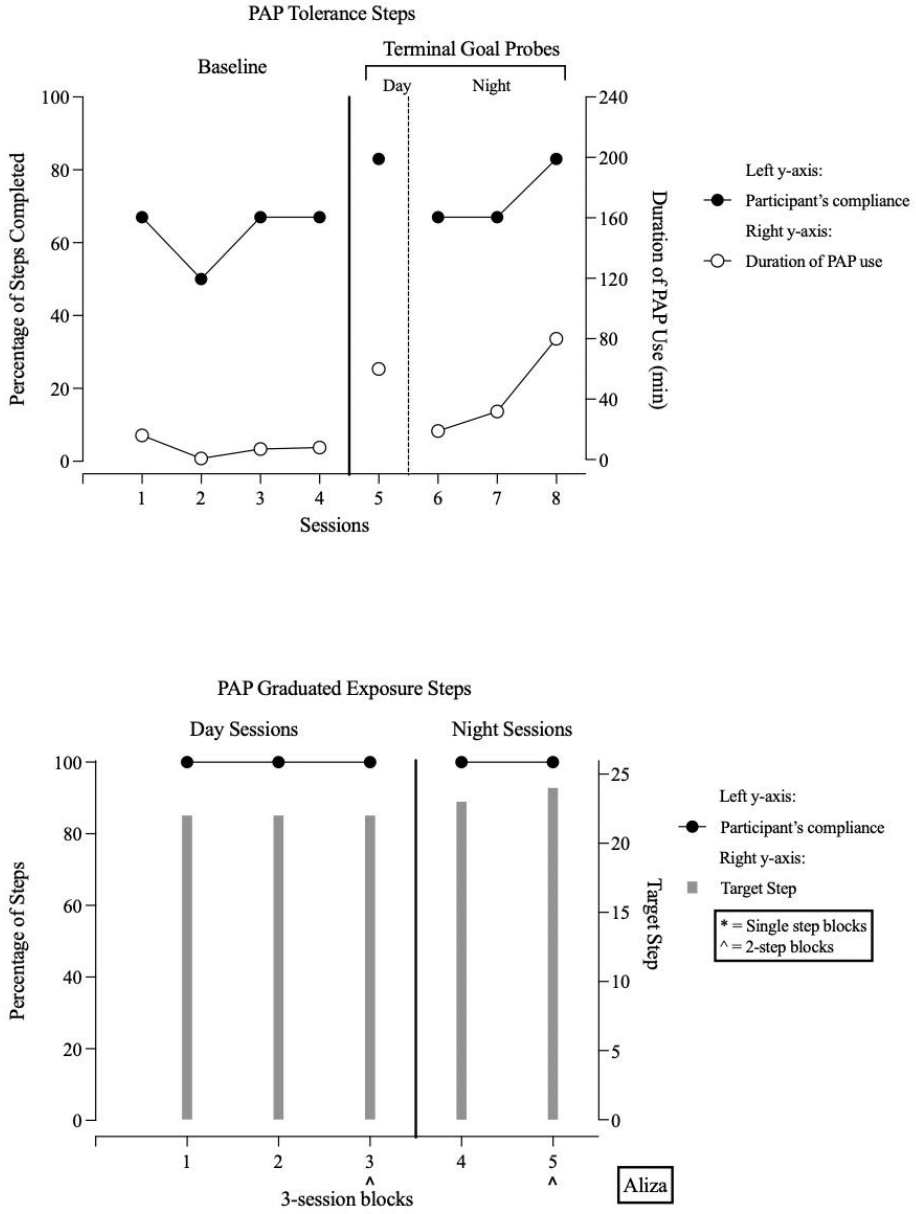


*Figure 5:*
*Aliza's PAP Tolerance Steps and PAP Graduated Exposure Steps graphs*

The top panel of the figure depicts the percentage of PAP tolerance steps Aliza completed during baseline sessions and terminal goal probes. Graphing conventions remain the same as described previously in Figure 3. When referring to session numbers, the researcher is discussing the PAP tolerance steps graph (i.e., top panel).

During the first baseline session (i.e., session #1), Aliza tolerated 16 min of PAP use (i.e., 67% of tolerance steps completed). In session #2, Aliza tolerated 46 s of PAP use (i.e., 50% of tolerance steps completed). In sessions #3 and #4, Aliza tolerated 7 min and 8 min of PAP use, respectively (i.e., 67% of tolerance steps completed). Therefore, the researcher targeted 7 min of PAP use in the first intervention session.

The bottom panel of the figure depicts Aliza's compliance with the PAP graduated exposure steps during day and night intervention sessions. The x-axis depicts the graduated exposure steps in 3-step blocks. This means each value on the x-axis represented three graduated exposure steps unless indicated otherwise. The left y-axis depicts the percentage of steps, and the right y-axis depicts the target graduated exposure step. The right y-axis ranges from zero to 25 to represent the 25 graduated exposure steps. Denoted by the black circles is Aliza's compliance and denoted by the gray bars is the target graduated exposure step. Asterisks indicate the block consisted of only one graduated exposure step. Arrows indicate the block consisted of two graduated exposure steps. When referring to block numbers, the researcher is discussing the PAP graduated exposure steps graph.

In the four intervention sessions (i.e., blocks #1-3), Aliza's compliance with graduated exposure steps remained at 100%. Following these sessions, the researcher initiated a day terminal goal probe. During this session (i.e., session #5), Aliza tolerated 1 hour of PAP use (i.e., 83% of tolerance steps completed). Therefore, the researcher transitioned to night sessions by initiating a night terminal goal probe. During this session

(i.e., session #6), Aliza tolerated 19 min of PAP use (i.e., 67% of tolerance steps completed). Due to a machine pressure change following this session, the researcher initiated another night terminal goal probe to ensure the change did not affect Aliza's compliance. During this second night terminal goal probe, Aliza tolerated 31 min and 52 s of PAP use. Aliza was more compliant (i.e., 31 min of PAP use) in the night terminal goal probe than in the preceding day intervention session (i.e., 30 min of PAP use). However, due to Aliza's compliance in the previous day intervention session (i.e., 30 min of PAP use), the researcher decided to present the next graduated exposure step in the first night intervention session, instead of presenting the same target duration again (i.e., 30 min of PAP use). Thus, the caregiver to present graduated exposure step #22c (i.e., 40 min of PAP use) in the first night intervention session.

Upon the transition to night intervention sessions, the noncontingent reinforcer (i.e., iPad) was only used for the first 30 to 45 minutes of the session. The contingent positive reinforcer also changed from the microphone (as used in day sessions) to a fake "dollar." Aliza's caregiver provided the opportunity to exchange the "dollars" for a meal at a fast-food restaurant of her choice upon earning eight "dollars." Following this transition, Aliza's percentage of compliance remained at 100% for the following four session blocks (i.e., blocks #4 and #5). Following session block #4, Aliza's caregiver requested that Aliza earn two "dollars" compliance with the graduated exposure step when targeting 1 hour or more of PAP use. Aliza's compliance with PAP graduated exposure steps have remained at 100%. In the third night terminal probe session (i.e., session #8), Aliza tolerated 1 hour and 20 min of PAP use (i.e., 83% of tolerance steps completed). Therefore, in the following intervention session (i.e., step #2 in block #5), the researcher targeted 1 hour and 30 min of PAP use at night.

Figure 6 depicts Aliza's duration of PAP machine use during intervention sessions. The x-axis depicts sessions. The y-axis depicts the duration of PAP use in minutes. The y-axis ranges from zero to 240 min to represent the 4 hours (i.e., 240 min) terminal goal. Denoted by the closed circles is Aliza's compliance duration and denoted by the solid dash is the target graduated step duration.

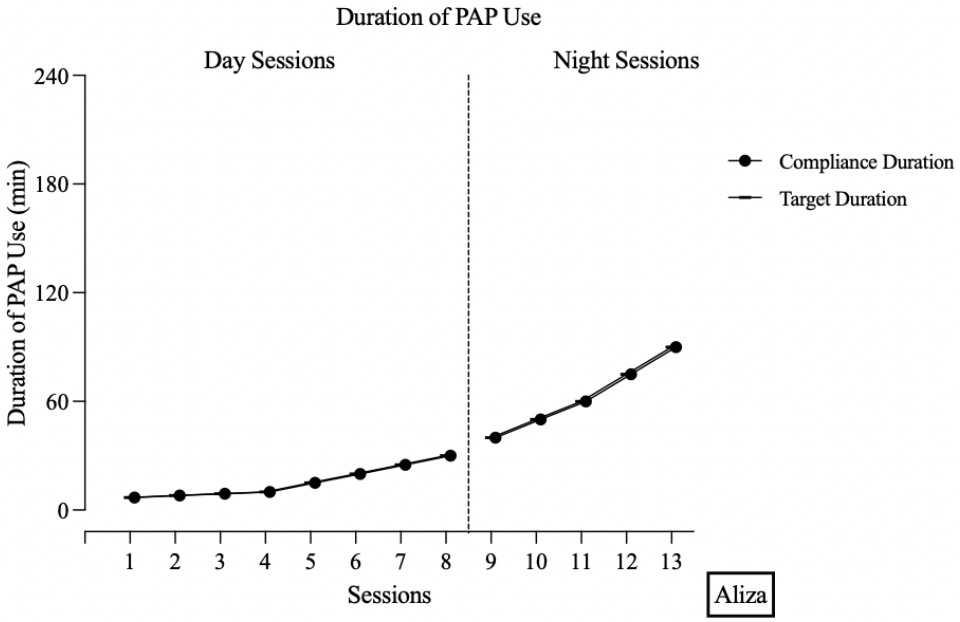


*Figure 6:*
*Aliza's Duration of PAP Use*

Session #1 on Figure 6 represents block #1 in the PAP Graduated Exposure graph (i.e., bottom panel of Figure 5). Aliza's duration of PAP use matched the target duration from session #1 through session #8. Following the transition to night sessions (i.e., sessions #9 through #12), Aliza's duration of PAP use remained the same as the target duration.

## Procedural Integrity

Procedural integrity was collected to determine the accuracy of the caregiver's implementation of the intervention procedures. Results of each caregiver's procedural integrity during intervention sessions are depicted below. While data on removing the mask within 15 s of the completed graduated exposure step or noncompliance was collected, this step is not included in the results due to the researcher observing it rarely being applicable. This component was not applicable majority of the time because all participants demonstrated the ability to remove the PAP mask independently of their caregiver's assistance. The researcher relied on caregiver report for the delivery of reinforcement contingent on compliance with the graduated exposure step when the videoconferencing night session ended before the graduated exposure step was complete. Therefore, this component (i.e., contingent reinforcement) was not depicted in the procedural integrity graphs for these sessions.

Figure 7 depicts Rory's caregiver's procedural integrity during intervention sessions. The x-axis depicts the graduated exposure steps in 3-step blocks. This means each value on the x-axis represented three graduated exposure steps unless indicated otherwise. The y-axis depicts the percentage of steps the caregiver correctly implemented the intervention step. Denoted by the black circles is the delivery of the instruction for the graduated exposure step. Denoted by the white circles is the 90-s to 2-min break following compliance. Denoted by the black squares is the delivery of the contingent reinforcer within 15 s of compliance. Denoted by the white squares is the 30-s to 45-s break following noncompliance. Denoted by the red circles is the delivery of the NCR prior to the presentation of the PAP mask or turning on the machine. Asterisks indicate the block is made up of one graduated exposure step. Arrows indicate the block is made up of two graduated exposure steps.

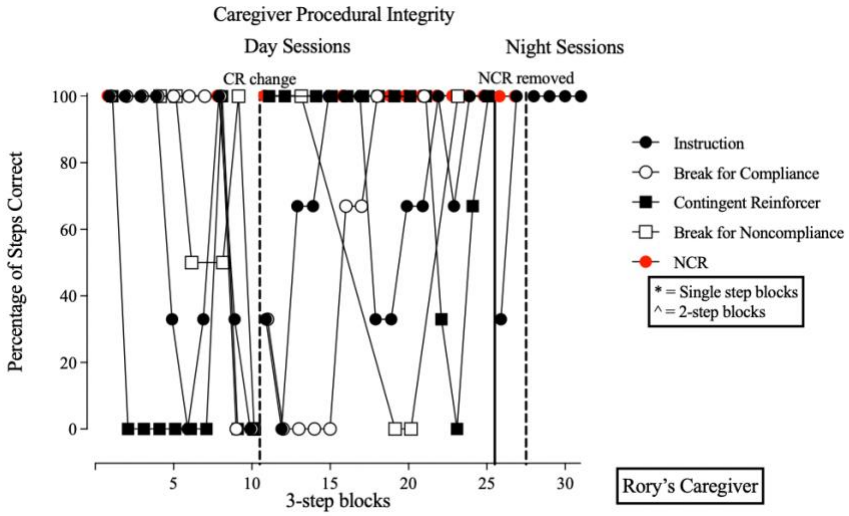


*Figure 7:*
*Rory's Caregiver's Procedural Integrity*

Rory's caregiver's procedural integrity was highly variable throughout the day intervention sessions. In all day sessions, Rory's caregiver accurately provided Rory the noncontingent reinforcer before presenting the PAP mask or turning on the PAP machine. During blocks #2-7, Rory's caregiver did not accurately deliver the contingent reinforcer following steps for compliance due to Rory having continuous access to the contingent reinforcer. In blocks #5 and #7, Rory's caregiver delivered the instruction prior to presenting the PAP mask for 33% of the graduated exposure steps. In session block #6 and #8, Rory's caregiver correctly provided a break contingent on noncompliance 50% of the steps in that block, as Rory did not comply with two of the steps in the block. In blocks #9 and #10, Rory's caregiver's percentage of steps correct decreased. These are the same blocks that Rory engaged in variable responding as seen in Figure 1.

Following block #10, the contingent reinforcer was changed from the "Ben 10" interactive toy to a fake "dollar" following the progression to a new step. Following this change (i.e., block #11), Rory's percentage of steps in which the contingent reinforcer was delivered accurately increased to 100%. However, the percentage of steps correct for the delivery of the instruction and the break for compliance decreased to 0% in session

block #11. Rory's caregivers correct implementation of the break for compliance remained at 0% in session blocks #11 through #15 due to the caregiver failing to respond to the caregiver's prompt that the break was not over yet and presenting a new graduated exposure step prior to 90 s. In session blocks #16 through #18, Rory's caregiver's correct delivery of a break following compliance increased to 100%. Rory's caregiver's correct delivery of the instruction prior to presenting the PAP mask increased from 0% in block #11 to 67% in block #13 and #14, and 100% in block #15. In blocks #18 and #19, Rory's caregiver accurately delivered the instruction 33% of steps. This correct responding increased in blocks #20 and #21 to 67% before reaching an average of 75% in blocks #22 through #25.

Rory's caregivers' delivery of a 30-s to 45-s break following noncompliance decreased from 100% in block #15 to 0% in block #19 and #20 due to his caregiver continuing to prompt compliance following vocal refusal and providing a short break than 30 s. In session block #24, the correct implementation of this step increased to 100%. In session block #22, Rory's caregiver's correct implementation of providing the contingent reinforcer decreased to 33%, then 0% in block #23. This was due to the caregiver providing the contingent reinforcer longer than 15 s after compliance. In block #24, correct implementation of this step increased to 67%, then 100% in block #25.

Following the transition to night sessions (i.e., block #26), breaks for compliance and noncompliance were no longer provided, as each session only contained one graduated exposure step. Therefore, following the completion of the graduated exposure step the caregiver provided the contingent reinforcer and the session was over. The delivery of the contingent reinforcer was no longer recorded, as the reinforcer was delivered the following morning. The researcher relied on caregiver report for this component for the remaining sessions.

Rory's caregiver provided the noncontingent reinforcer in 100% of the steps in block #26 and #27. Before beginning step #1 in block #28, Rory requested the noncontingent reinforcer are no longer be provided, as he preferred to sleep while using his PAP machine. Following the removal of the noncontingent reinforcer, the caregiver' implementation of the instruction was at a 91% correct on average in the four three-step blocks.

Figure 8 depicts Ross' caregiver's procedural integrity during intervention sessions. The graphing conventions remain the same as Figure 7.

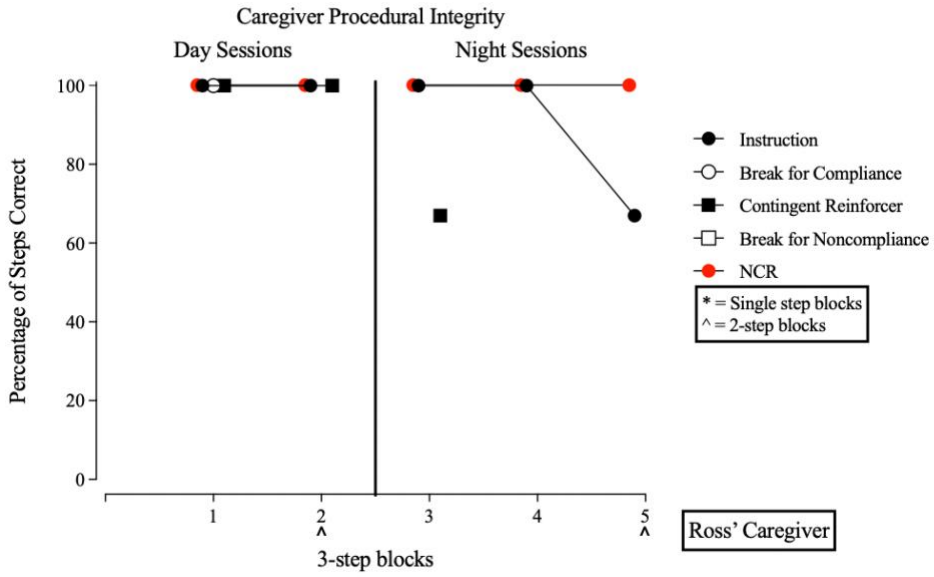


*Figure 8:*
*Ross' Caregiver's Procedural Integrity*

Ross' caregivers' procedural integrity remained at 100% for all intervention components during the day sessions (i.e., block #1 and #2). Upon the transition to night sessions, Ross' caregiver's delivery of the contingent reinforcer was over 15 s following the end of the graduated exposure step. Therefore, in block #3, Ross' caregiver's percentage of steps correct for the contingent reinforcer was 67%. Following this block, the researcher no longer observed the delivery of the reinforcer due to the participant falling asleep and the reinforcer was delivered the following morning. The researcher relied on caregiver report for this component for the remaining sessions. In block #3 and #4 the caregiver correctly provided the instruction and the noncontingent reinforcer. In block #5, Ross' caregiver did not provide the instruction before turning on the PAP machine. Thus, the instruction component decreased to 67% correct.

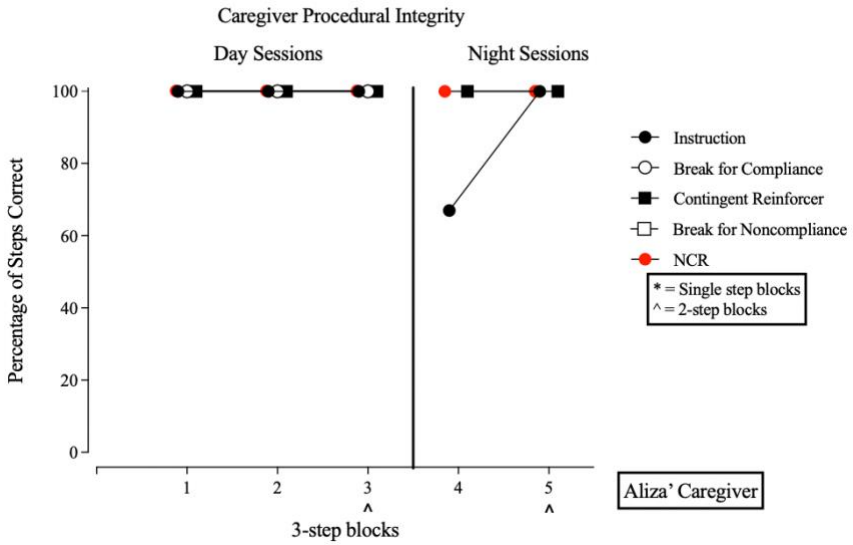


*Figure 9:*
*Aliza's Caregiver's Procedural Integrity*

Aliza's caregivers' procedural integrity remained at 100% for all intervention components during the day sessions (i.e., block #1 through #3). Following the transition to night intervention sessions, Aliza's caregiver delivered the instruction after turning on the PAP machine. Therefore, Ross' caregiver implemented the instruction component 67% of steps in block #4. In block #5, this increased to 100%. All other components remained at 100% correct.

# CHAPTER IV:

# DISCUSSION

Data on the participants' percentage of tolerance steps completed, percentage of compliance with graduated exposure steps, and compliance with PAP use demonstrate the intervention was successful in increasing PAP machine use for all three participants. At the beginning of participation, Rory did not tolerate the PAP machine being near him In the most recent three intervention sessions, Rory used the PAP machine for 2 hours and 42 min at night on average. At the beginning of participation, Ross tolerated 19 min of PAP use. Currently, he uses the PAP machine for 2 hours and 3 min at night on average, as seen in the three most recent intervention sessions. At the beginning of participation, Aliza tolerated 8 min of PAP use and currently uses the PAP machine for 1 hour and 15 min at night on average, as seen in the most recent three intervention sessions.

While these participant's data have not yet met the terminal goal (i.e., 4 hours of PAP use for three consecutive nights in a row), they do offer information on the efficacy of the caregiver-implemented intervention and serve as evidence to support the prediction that with additional sessions, all participants will reach the terminal goal. This study is still in progress to evaluate this prediction.

## Rory

As seen in Figure 1, Rory's PAP use was not established at the beginning of the study. In other words, Rory refused to have the PAP mask and machine near him prior to the intervention. Throughout the day intervention sessions, Rory's compliance with the graduated exposure steps occurred at high levels with moderate variability. However, Figure 2 demonstrated that while Rory's compliance with the steps were variable, his duration of PAP use steadily increased throughout the study.

Rory's case differed from the other two participants in the duration of participation, the variability of participant's compliance and caregiver's procedural integrity, and the removal of the noncontingent reinforcer in night sessions. Rory's compliance with the tolerance steps in baseline resulted in day intervention sessions to begin with graduated exposure step #1 (i.e., PAP mask within 1 ft). Thus, the number of day intervention sessions for Rory was over four times more than for Ross and Aliza, as their initial graduated exposure step was #22 (i.e., 18 min and 7 min of PAP use, respectively). During participation in the study, Rory's caregiver had a family matter that interfered with conducting sessions regularly. This along with the initial graduated exposure step and varying levels of compliance led to Rory's participation spanning just over a year.

Rory's compliance with PAP graduated exposure steps throughout the intervention was highly variable. Rory's caregiver's correct implementation of the intervention also occurred at variable rates. Figure 10 depicts Rory's PAP Graduated Exposure Steps graph in the top panel and Rory's Caregiver's Procedural Integrity graph in the bottom panel to analyze the trends in these data. These are the same graphs as displayed in the results section and graphing conventions remain the same as mentioned previously.

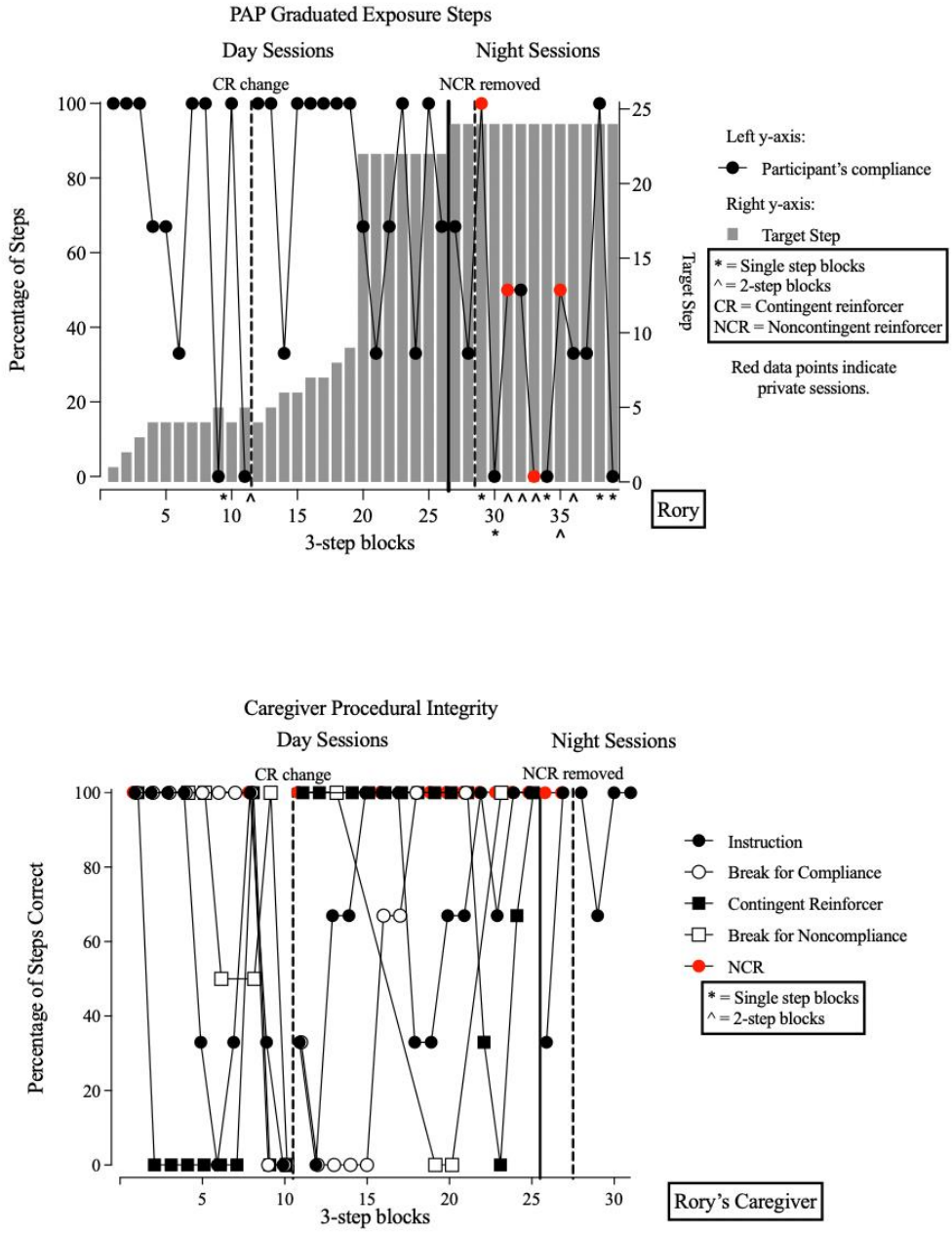


*Figure 10:*
*Rory's PAP Graduated Exposure Steps and Rory's Caregiver's Procedural Integrity graphs*

The 3-step blocks align in both graphs until block #28. Following this block, Rory's caregiver conducted a independent intervention session and procedural integrity was not collected during these sessions. Rory's caregiver did not correctly provide the

contingent reinforcer for blocks #2 through #7. Rory's compliance decreased at variable levels during these blocks, as well. This suggests the absence of salient reinforcement for compliance with a graduated exposure step affects the percentage of compliance with the graduated exposure steps.

Upon the contingent reinforcer change, Rory's caregiver correctly provided the contingent reinforcer and Rory's compliance increased. During block #12, Rory's caregiver's correct delivery of the instruction and break for compliance decreased to 0% of steps. In session block #13, Rory's compliance decreased to 33% of steps. This may suggest that the repeated absence of the instruction prior to a graduated exposure step may have affected compliance. It is also possible the correct duration for a break for compliance decreasing to 0% in blocks #12 through #15 could have affected Rory's compliance, as well. Rory's compliance and Rory's caregiver's procedural integrity occurs at highly variable percentages in blocks #20 though #25. Conclusions regarding Rory's variable compliance evoking Rory's caregiver's procedural integrity or vice versa cannot be drawn from these data, as there are extraneous variables outside of the researcher's knowledge that could have affected this variability, as well. However, the ultimate concern is whether Rory's duration of PAP use increased due to the implementation of this intervention. As seen in Figure 2, it is currently at 3 hours and 16 min. Therefore, this intervention was effective at increasing Rory's PAP machine use.

Data on Rory's compliance and Rory's caregivers' procedural integrity offer insight on how deviation from intervention procedures can occur, yet still produce significant behavior change. However, because Rory's caregiver's procedural integrity was highly variable throughout the intervention, the extent to which the intervention components were responsible for the behavior change observed in Rory may be challenged. Upon further evaluation of the errors in correct implementation of the

intervention, the researcher identified failure to remove the contingent reinforcer after a break for compliance, providing shorter breaks following compliance, and vague instructions were the most common errors. Thus, Rory's caregiver engaged in the steps of the intervention, but did not meet the criteria for a correct response. For example, in blocks #12 through #15, Rory's caregiver engaged in 0% of steps correctly when providing a break for compliance. However, this was due to his caregiver providing a 1-min break instead of a 90-s to 2-min break following compliance. Thus, it is safe to conclude that the intervention components were responsible for the increase in Rory's PAP machine use. This is because the errors Rory's caregiver made were not due to the complete omission of intervention component or the addition of other components. These data also suggest that the criteria for correct implementation of the intervention may not require the stringent criteria to be effective.

The intervention components differed for Rory from the other participants due to the removal of the noncontingent reinforcer. Rory requested that the interactive speaker no longer play music during night intervention sessions as he would prefer to sleep. Thus, the researcher decided to remove this component. Upon the removal, Rory's compliance with graduated exposure steps occurred with moderate variability, but Rory's compliance with PAP use continued to increase from 1 hour and 58 min to 3 hours and 16 min. While the removal of the noncontingent reinforcer may have affected Rory's compliance with the PAP graduated exposure steps, it could have also been affected by the increasing requirement to earn the contingent reinforcer. Therefore, it is likely Rory's change in compliance percentages following this change was affected by multiple variables. The researcher did not find this change in Rory's percentage of compliance concerning due to the consistent increase in Rory's duration of PAP use.

The researcher had concerns regarding the accurate withholding of contingent reinforcement (i.e., fast food meals) following sessions in which Rory did not comply with the target duration of PAP use. The researcher continued to remind the caregiver of the importance of providing the contingent reinforcer only following compliance with the graduated exposure step. However, on two different occasions, the researcher observed the caregiver discard trash from Rory's highly preferred fast-food restaurant during sessions. The researcher ensured that Rory had the choice of which fast-food restaurant to exchange his "dollars" for to maintain motivation to earn the contingent reinforcer. Thus, even if Rory had occasionally had noncontingent access to fast-food, motivation to earn and exchange the contingent reinforcers maintained because Rory could select different fast-food to exchange the contingent reinforcers for. This demonstrates the importance of the caregiver's feasibility and willingness to withhold reinforcers and should be considered when selecting contingent reinforcers in future research.

Lastly, the process in which the machine recorded data were obtained by the researcher differed in Rory's case. Rory's caregiver purchased the PAP machine. Therefore, the data were not automatically accessible by the collaborating sleep physicians. To obtain the machine recorded data, Rory's caregiver had to either send the researcher a picture of the machine smartphone application or take the PAP machine into the clinic for a manual download. Occasionally, the researcher had to rely on caregiver report until machine recorded data were accessible due to either errors in the smart phone application or the caregiver's delay to bring the machine into the clinic. When the researcher did not have access to these data, Rory's caregiver report did not reliably account for the accurate duration of PAP use. This limited the researcher's ability to accurately progress (or regress) through the graduated exposure steps, because the target graduated exposure step was determined by the participant's responding in the previous

session. Due to these errors, there were three intervention sessions in which the researcher increased the graduated exposure step when Rory's accurate duration of PAP machine use did not warrant the advancement. Therefore, this intervention may not be as effective and feasible without immediate access to the machine recorded data. This is serves as a potential limitation on the feasibility of this intervention.

**Ross**

Ross' percentage of completed PAP tolerance steps and percentage of compliance with the PAP graduated exposure steps demonstrated the intervention was effective at increasing PAP machine use from baseline sessions to the most recent graduated exposure session. Ross' caregiver implemented the intervention at high integrity throughout the intervention. The high percentages of Ross' compliance and Ross' caregiver's procedural integrity serve as evidence to support that this caregiver-implemented intervention was effective and feasible.

It is important to mention Ross' prior history with participation in this study. Ross began participation in this study 8 months prior to the first recorded baseline session. However, the first baseline sessions conducted informed the researcher that Ross' PAP machine use already met the terminal goal of the intervention (i.e., 4 hours of PAP use for three consecutive nights). Therefore, Ross did not initially qualify to participate in this study. Ross' caregiver initiated contact with the researcher a few months later to share that Ross no longer used the PAP machine effectively. Thus, the researcher conducted new baseline sessions, as seen in Figure 3, and Ross' active participation began. This raises some important considerations regarding the terminal goal for this study.

As seen in Ross' case, there is a possibility that upon three consecutive nights of effective PAP use (i.e., 4 hours or more) a participant's duration of PAP use can regress

to unacceptable levels. This raises a concern that the terminal goal does not suffice as a criterion of mastery for effective PAP use. The researcher chose to set the terminal goal at three consecutive nights due to the consistency of this criteria observed in practice. However, the purpose of the terminal goal was to serve as a criterion to determine effective PAP use, but Ross' behavior served as proof that this may not be the case. Perhaps increasing the terminal goal to 4 hours of PAP use for five consecutive nights would better predict longer term adherence to PAP use. Future research should consider modifying the terminal goal to account for this concern.

### Aliza

Aliza had the fewest number of intervention sessions. While her participation in the study was not long, there were already improvements in Aliza's PAP use. Aliza tolerated the PAP machine for very short periods of time at the beginning of the study. Aliza currently uses the PAP machine for 1 hour and 15 min at night. She also began falling asleep for short periods of time. This provided encouraging support that the intervention was effective at increasing Aliza's PAP use.

## General Discussion

Obstructive sleep apnea is extremely prevalent in adults with Down syndrome (DS) (Gimenez et al, 2021). While Positive Airway Pressure (PAP) therapy is proven to be effective at treating sleep apnea, adherence to PAP therapy prescriptions is concerningly low in the general sleep apnea population (Lance, 2020). Adults with DS may experience additional barriers that prohibit effective PAP therapy (e.g., sensitivity to the pressure, caregiver assistance required) and further decrease the likelihood of PAP therapy adherence. The current study evaluated the efficacy of a caregiver-implemented behavioral intervention aiming to increase PAP therapy in adults with DS. This study extended the findings of Koontz and colleagues (2003) by evaluating the intervention

with a new population (i.e., adults) and a different modality (i.e., telehealth training and caregiver-implemented). A strength of the current study is the heterogeneity of these participants in terms of their initial levels of PAP tolerance. Rory's initial level of tolerance was much lower (i.e., did not tolerate the mask near him) than Ross' and Aliza's (i.e., 19 min and 8 min, respectively). This intervention has demonstrated significant improvements for all participants regardless of initial tolerance levels.

A limitation of the current study was the schedule of the terminal goal probes. Terminal goal probes were used to advance through graduated exposure steps when the participant demonstrated compliance with the untargeted graduated exposure steps in a terminal goal probe. The researcher chose to conduct these probes every four sessions and upon the transition to night sessions. This schedule was determined based on the consideration of how often a terminal goal probe could be conducted without the participant experiencing aversive effects from setting an unobtainable goal too frequently and the desire for consistency across participants. On average, participants had two to three sessions per week. Thus, terminal goal probes were conducted roughly every other week. While this schedule offered consistency across participants and low frequency of probes, it was not sensitive to the participants compliance in the preceding intervention sessions. As mentioned before, compliance observed in terminal goal probes was used to advance through the untargeted graduated exposure steps if the participant's compliance was more than in the preceding intervention session. However, when a participant engaged in noncompliance in the session(s) prior to the terminal goal probe, it was unnecessary to evaluate compliance with the terminal goal, because the participant demonstrated noncompliance already. Thus, a more sensitive schedule of conducting a terminal goal probes would be to probe upon a set number of sessions with compliance. For example, if the terminal goal probe schedule only allowed for terminal goal probes to

be conducted after the participant was compliant with five graduated exposure steps, it would minimize the number of unnecessary terminal goal probes and the number of sessions the participant does not contact reinforcement. This schedule would also enhance efficiency of the intervention.

Another limitation of the current study was the use of a token economy without the researcher systematically establishing the tokens as secondary reinforcers (i.e., training the participant to use a token economy). Instead, the researcher relied on the caregivers' verbal explanation of the system to the participant. A token economy is a reinforcement system that allows for a secondary reinforcer to be provided and later exchanged for a primary or terminal reinforcer. In this study, the secondary reinforcer was the fake "dollars" provided to the participants upon compliance with the graduated exposure step. The terminal reinforcer for Rory and Aliza was a meal from a fast-food restaurant of their choosing. The terminal reinforcer for Ross was a milkshake from a fast-food restaurant of his choosing. The number of fake "dollars" the participants were required to earn in order to have the opportunity to exchange for the terminal reinforcer (e.g., fast food meal or milkshake) was determined prior to the first session in which the "dollars" were used. This criterion was based on how frequently the caregiver was willing to provide the reinforcer and the cost of the terminal reinforcer in the natural environment. The researcher recommended the caregivers require a minimum of three "dollars" and a maximum of eight "dollars" based on the actual financial cost of these items. For example, the researcher predicted a milkshake cost about five dollars. Therefore, with assistance from the researcher, Ross' caregiver decided to require three "dollars" for a milkshake. Rory's caregiver decided to require five "dollars" for a fast-food meal. Aliza's caregiver decided to require eight "dollars" for a fast-food meal.

Rory and Aliza's caregiver's both requested to change these criteria, so the participant had the opportunity to earn the terminal reinforcer (i.e., fast food meal) more frequently. Rory's caregiver requested that upon the transition to night sessions, that the "dollars" no longer be used and rather, Rory earned the terminal reinforcer for complying with the graduated exposure step during night sessions. Prior to this change, Rory earned one "dollar" for every step he complied with and had to earn five "dollars" to receive the terminal reinforcer. Aliza's caregiver did not change the number of "dollars" required to exchange for a fast-food meal. Instead, Aliza's caregiver requested that for any graduated exposure step targeting more than 1 hour of PAP use, Aliza earned two "dollars" instead of one. This was due to the increase in effort required to meet the increasing PAP use goal and earn the reinforcer (i.e., "dollar"). Throughout the intervention, Ross earned one "dollar" for each graduated exposure step with compliance.

While the lack of systemic training on token economies is a limitation to the current study, it was observed that the token economy (i.e., "dollars" and terminal reinforcer), along with the other intervention components, was still effective at increasing PAP use in all participants even without the explicit training of the token economy. Future research should remedy this limitation by explicitly establishing a token economy with a consistent reinforcement schedule.

Future areas of research should also evaluate the necessity of the specific intervention components via component analysis. While the current study provides evidence that the intervention is still effective following the removal of differential negative reinforcement (i.e., longer breaks for compliance, shorter breaks for noncompliance), a more in-depth analysis of each component may offer insight to the necessary components for success. Research could also evaluate the efficacy of the intervention conducted through direct implementation by the therapist directly

conducting the intervention without the caregiver's involvement. However, the benefits of the caregiver-implemented approach might outweigh the interest in a researcher-implemented approach, especially for a behavior (i.e., PAP use) that occurs during a private time (i.e., while sleeping).

The current study systematically evaluated the effects of a caregiver-implemented intervention on PAP machine use in adults with DS. The results suggested the intervention is effective at increasing the duration of PAP therapy. Due to the ongoing nature of this study, conclusions regarding the effectiveness of increasing PAP therapy to the terminal goal cannot yet be drawn. However, the steady increase in all participants PAP machine use throughout the intervention serve as promising evidence to support the intervention efficacy. This intervention offered the caregiver a hands-on approach to assisting their son or daughter towards a behavior change for a health-related goal. It also serves as evidence to suggest that this intervention may be effective without the use of escape extinction. This study also accounts for the validity of the measurements and procedures, but also included a social validity measure for both caregiver and participant. This provides insight on the social significance (i.e., feasibility and acceptability) of the intervention and components.

REFERENCES

Aloia, M. S., Di Dio, L., Ilniczky, N., Perlis, M. L., Greenblatt, D. W., & Giles, D. E. (2001). Improving compliance with nasal CPAP and vigilance in older adults with OAHS. *Sleep & breathing = Schlaf & Atmung*, *5*(1), 13–21. https://doi.org/10.1007/s11325-001-0013-9

DeMolles, D. A., Sparrow, D., Gottlieb, D. J., & Friedman, R. (2004). A pilot trial of a telecommunications system in sleep apnea management. *Medical care*, *42*(8), 764–769. https://doi.org/10.1097/01.mlr.0000132353.99209.fe

Garmendia, Farré, R., Ruiz, C., Suarez Girón, M., Torres, M., Cebrian, R., Saura, L., Monasterio, C., Negrín, M. A., & Montserrat, J. M. (2021). *Telemedicine Strategy to Rescue CPAP Therapy in Sleep Apnea Patients with Low Treatment Adherence: A Pilot Study*. https://doi.org/10.3390/jcm10184123

Giménez, Altuna, M., Blessing, E., Osorio, R. M., & Fortea, J. (2021). Sleep Disorders in Adults with Down Syndrome. *Journal of Clinical Medicine*, *10*(14), 3012–. https://doi.org/10.3390/jcm10143012

Kang, E. K., Xanthopoulos, M. S., Kim, J. Y., Arevalo, C., Shults, J., Beck, S. E., Marcus, C. L., & Tapia, I. E. (2019). Adherence to positive airway pressure for the treatment of obstructive sleep apnea in children with developmental disabilities. *Journal of Clinical Sleep Medicine*, *15*(06), 915–921. https://doi.org/10.5664/jcsm.7850

Koontz, K. L., Slifer, K. J., Cataldo, M. D., & Marcus, C. L. (2003). Improving pediatric compliance with positive airway pressure therapy: The impact of behavioral intervention. *Sleep*, *26*(8), 1010–1015. https://doi.org/10.1093/sleep/26.8.1010

Lance, M.D., C. (2020, February 24). *Increasing Adherence to PAP Therapy for Patients with Sleep Apnea*. Cleveland Clinic.

https://consultqd.clevelandclinic.org/increasing-adherence-to-pap-therapy-for-patients-with-sleep-apnea/

Leaf, J.B., Leaf, R., McEachin, J. *et al.* Advantages and Challenges of a Home- and Clinic-Based Model of Behavioral Intervention for Individuals Diagnosed with Autism Spectrum Disorder. *J Autism Dev Disord* **48,** 2258–2266 (2018). https://doi.org/10.1007/s10803-017-3443-3

Lee, C. F., Lee, C. H., Hsueh, W. Y., Lin, M. T., & Kang, K. T. (2018). Prevalence of obstructive sleep apnea in children with Down Syndrome: A meta-analysis. *Journal of clinical sleep medicine: JCSM: official publication of the American Academy of Sleep Medicine*, *14*(5), 867–875. https://doi.org/10.5664/jcsm.7126

Mayo Foundation for Medical Education and Research. (2020, July 28). *Sleep apnea*. Mayo Clinic. Retrieved March 15, 2022, from https://www.mayoclinic.org/diseases-conditions/sleep-apnea/symptoms-causes/syc-20377631

National Down Syndrome Society. (2021). *What is Down Syndrome?* https://www.ndss.org/about-down-syndrome/down-syndrome/

Reimers, T. M., & Wacker, D. P. (1988). Parents' Ratings of the Acceptability of Behavioral Treatment Recommendations Made in an Outpatient Clinic: A Preliminary Analysis of the Influence of Treatment Effectiveness. *Behavioral Disorders*, *14*(1), 7–15. https://doi.org/10.1177/019874298801400104

Rotenberg, B., Murariu, D., & Pang, K. (2016). Trends in CPAP adherence over twenty years of data collection: A flattened curve. *Journal of Otolaryngology-Head and Neck Surgery*, *45*(1), 43 https://doi.org/10.1186/s40463-016-0156-0

Sawyer, A., King, T., Weaver, T., Sawyer, D., Varrasse, M., Franks, J., Watach, A., Kolanowski, A., & Richards, K. (2019). A Tailored Intervention for PAP Adherence: The SCIP-PA Trial. *Behavioral Sleep Medicine*, *17*(1), 49–69. https://doi.org/10.1080/15402002.2016.1276018

Stepnowsky, C. J., Palau, J. J., Marler, M. R., & Gifford, A. L. (2007). Pilot randomized trial of the effect of wireless telemonitoring on compliance and treatment efficacy in obstructive sleep apnea. *Journal of medical Internet research*, *9*(2), e14. https://doi.org/10.2196/jmir.9.2.e14

Weaver, & Sawyer, A. M. (2010). Adherence to continuous positive airway pressure treatment for obstructive sleep apnoea: implications for future interventions. *Indian Journal of Medical Research (New Delhi, India: 1994)*, *131*(2), 245–258.

APPENDIX A:

CAREGIVER TREATMENT ACCEPTABILITY RATING FORM (TARF)

**Treatment Acceptability Rating Form:** Caregiver

1. How likely is this intervention to make permanent improvements in your son/daughter's PAP machine adherence?

   ___ Not likely

   ___ Unsure

   ___ Very likely

   Other comments:

2. How satisfied are you with the results of the intervention?

   ____ Not satisfied

   ____ Indifferent

   ____ Satisfied

   Other comments:

3. How feasible were the **daytime** intervention sessions (i.e., 30-min to 1-hour sessions, 1-3 times per week)?

   ____ Not feasible

   ____ Indifferent

   ____ Feasible

   Other comments:

4. How feasible were the **nighttime** intervention sessions (i.e., 30-min to 1-hour sessions, 1-3 times per week)?

   ____ Not feasible

   ____ Indifferent

____ Feasible

Other comments:

5. How feasible were the intervention components (i.e., instruction, reinforcement for compliance, less/no reinforcement for noncompliance) to implement?

   ____ Not feasible (if so, please specify which component(s) were not feasible in the comments)

   ____ Indifferent

   ____ Feasible

   Other comments:

6. How acceptable were the intervention components (i.e., instruction, reinforcement for compliance, less/no reinforcement for noncompliance) to implement?

   ____ Not acceptable (if so, please specify which component(s) were unacceptable in the comments)

   ____ Indifferent

   ____ Acceptable

   Other comments:

7. How satisfied are you with the delivery of the intervention (i.e., all remote via Zoom, caregiver-implemented sessions, researcher-assisted)?

   ____ Not satisfied (Would prefer in-person, researcher-implemented sessions)

   ____ Not satisfied (Would prefer in-person caregiver trainings, but remote caregiver-implemented sessions)

   ____ Indifferent

| |
|---|
| ____ Satisfied<br><br>Other comments: |
| 8. Please provide any other feedback below. |

APPENDIX B:

PARTICIPANT TREATMENT ACCEPTABILITY RATING FORM (TARF)

| **<u>Treatment Acceptability Rating Form:</u>** Participant |
| --- |
| 1. Did you enjoy meeting with *researcher's name* to work on using your PAP machine?<br>___ Yes<br><br>___ No<br><br>___ No response |
| 2. Did you enjoy working with your mom on using your PAP machine?<br>___ Yes<br><br>___ No<br><br>___ No response |
| 3. Will you use your PAP machine to sleep at night?<br>____ Yes<br><br>____ No<br><br>____ Unsure<br><br>____ No response |