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ORIGINAL ARTICLE

Behavioral Treatment of Problem Behavior for an Adult with Autism Spectrum Disorder and Misophonia

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Abstract
Misophonia is characterized by an autonomic response (e.g., increased heart rate) that is elicited by certain innocuous or repetitive sounds, and individuals with misophonia may display an extreme, overt response commonly associated with rage, hatred, and a loss of self-control. In this investigation, we used a combined respondent and operant approach to treat problem behavior evoked by bodily sounds (i.e., coughing, sneezing, sniffling, and clearing throat) for an adult with autism spectrum disorder. The intervention produced immediate reductions of problem behavior and the effects of treatment maintained during progressively lean schedules of reinforcement. The results of this study will be discussed in light of past research, along with limitations, and future directions for research and clinical practice.

Keywords Autism spectrum disorder · Fixed-time schedule · Misophonia · Problem behavior

Misophonia is a neurological and behavioral condition characterized by strong physical and emotional responses that are caused by intolerance to specific auditory stimuli (Jastreboff and Jastreboff 2014; Moller 2011; Wu et al. 2014). Common triggers for misophonic responses include smacking of the mouth while eating, sniffling, sneezing, and clearing the throat. In response to these stimuli, individuals with misophonia may experience feelings of rage, hatred, and a loss of self-control that could significantly impact their lives as well as the lives of their significant others (Dozier 2015; Schwartz et al. 2011). Given how often one might encounter these sounds on a daily basis, finding an effective treatment for misophonia is imperative.

Relatively little is known about the etiology of misophonia based on a dearth of literature published on this topic (Bernstein et al. 2013). One theory suggests that stronger activation in specific neural areas, including the anterior insular cortex, which is responsible for

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processing emotional experience, and the default mode network, which detects stimuli in the environment that are personally relevant, contributes to the subjectively heightened experience (Brout et al. 2018). Trigger stimuli may be processed by these centers in the brain and make these otherwise neutral stimuli for most people highly salient and emotionally charged. Similarly, the network neural models suggest that stronger pathways between specific neural areas, such as those mentioned above, contribute to the misophonic experience. Another theory, described as a conditioned aversive reflex, suggests that misophonia develops through classical conditioning (Dozier 2015). According to Dozier and Morrison (2017), an unconditioned stimulus (e.g., sneeze) elicits an unconditioned response (e.g., startle reflex). The unconditioned response (i.e., startle reflex) serves as an unconditioned stimulus that elicits another unconditioned response (i.e., autonomic arousal). Through repeated pairings between the initial trigger stimulus (i.e., sneeze) and the unconditioned stimulus (i.e., startle reflex), the initial trigger stimulus (i.e., conditioned stimulus) elicits the conditioned response (i.e., autonomic arousal associated with emotional responding).

The lack of firm evidence for the etiology of misophonia makes treatment of this disorder difficult. Further, the absence of misophonia from the Diagnostic and Statistical Manual – Fifth Edition and ICD-10 compounds the issue for medical doctors and psychologists when they encounter patients with misophonia (Schwartz et al. 2011). Schwartz et al. reported there is no cure for misophonia, and few published empirical studies have evaluated treatments (Bruxner 2016). However, several published case studies may offer preliminary guidance for clinical practitioners who encounter patients with this condition (e.g., Bernstein et al. 2013; Schroder et al. 2017; Schroder et al. 2017). These studies reported some success based on individual or combined treatments that incorporate cognitive behavior therapy (CBT) and other talk-based therapies (e.g., acceptance and commitment therapy).

A commonality among the studies using talk-based therapies is the use of psychoeducation to discuss the relationship between one's thoughts, behavior, and physiology. For example, an individual with misophonia may think that someone who is smacking their mouth while eating is doing so "just to annoy" them. This thought reportedly triggers a behavioral response, such as confronting or avoiding the person making those sounds, along with physiological arousal associated with the sympathetic nervous system (e.g., increased heart rate and blood flow, and decreased digestion). However, by changing one's thoughts, a person can learn to overcome maladaptive behavior (e.g., yelling) associated with the trigger stimuli and engage in a socially acceptable response. In our review of case studies in which talk therapies were employed (noted above), all have reported significant improvement in misophonic symptoms.

Nevertheless, there are major limitations in the extant research on treatment for individuals with misophonia. First, the number of studies employing treatments for individuals with misophonia is limited. Only two studies (i.e., Dozier 2015; Dozier and Morrison 2017) employed a systematic pairing procedure in which the trigger sound was paired with some pleasurable stimulus. These studies also incorporated talk-based therapies and progressive muscle relaxation as part of the treatment package. Second, the homogeneity of the participants severely limits the generality of their findings to other populations (e.g., individuals with comorbid psychiatric disorders). None of the participants reported any personal or familial history of psychiatric illness and they were otherwise neurotypical. The exception is one study which included a subset of participants with comorbidities (Schroder et al. 2017). Schroder et al. employed CBT as the sole treatment approach. Third, all of the studies we reviewed included self-reported ratings of stress responses associated with trigger stimuli; self-reported data are prone to inaccuracies (van de Mortel 2008). Thus, the research literature offers little guidance for practitioners treating individuals with misophonia who also have autism spectrum disorder (ASD) and severely limited communication skills. The present study evaluated the effects of a treatment based on classical and operant conditioning on problem behavior triggered by bodily sounds exhibited by an individual with ASD. We used direct behavioral data to evaluate the effects of the treatment.

Method

Participant, Setting, and Materials

Andy was age 19, 170.18 cm tall, and weighed 97.52 kg. He was diagnosed with ASD by a neurologist at age 3, and was delayed in meeting his developmental milestones (e.g., walking and talking). Andy had a significant history of problem behavior evoked by bodily sounds. Triggers for problem behavior were coughing, sneezing, sniffling, and clearing one's throat. Andy engaged in multiple topographies of problem behavior. Aggression included hitting another person using an open or closed hand and throwing items at another person. Disruptive behavior included negative vocalizations (e.g., yelling and crying), banging on surfaces, and throwing items against the wall or to the floor. Self-injurious behavior included hitting his face or forehead repeatedly with one or both hands. At the time of this study, Andy was administered the following medications: divalproex 500 mg once daily, sertraline 100 mg once daily, trazodone 150 mg once daily, clonazepam 1.5 mg twice daily, and haloperidol 10 mg once daily. Andy did not administer any medication during his therapy appointments at the treatment facility. Andy had very limited adaptive skills, with scores in the low range for all areas of adaptive functioning according to the Vineland-3 (Hill et al. 2017). Specifically, Andy obtained the following standard scores (SS): 20 for Communication (range, 14 to 26), 20 for Daily Living Skills (range, 14 to 26), 20 for Socialization (range, 16 to 24). The Internalizing and Externalizing subdomains in the Maladaptive Behavior Index were rated to be High by his parents, with v-scale scores of 22 and 23, respectively; thus, indicating significant need for support.

Andy received a total of 15 h of direct applied behavior analysis (ABA) therapy on five days per week for six weeks at a treatment facility prior to starting treatment for bodily sounds. The treatment facility was a 130.06 sq. m two-story building in Southern California. Sessions occurred in a 6.1 m × 6.1 m room, furnished with tables, toys, chairs along the perimeter of the room, and decorations commonly found on the walls in school classrooms (e.g., cartoon pictures of animals and a calendar). Session materials included a table, chairs, edibles (i.e., Skittles cut in half and equivalent-sized fruit snacks per presentation,), data sheets, pen, and a timer.

Dependent Variables, Measurement, Interobserver Agreement, and Procedural Integrity

Aggression was defined as contacting another person with an open or closed hand from ≥15.24 cm and projecting an object not designed to be thrown a distance of >0.61 m that landed <0.61 m of another person. Disruptive behavior was defined as emitting a vocalization above conversational level using a negative tone, making contact with an inanimate surface from ≥15.24 cm such that the sound could be heard from ≥1.82 m, and projecting an object not designed to be thrown a distance of >0.61 m that landed >0.61 m from another person. Self-injury was recorded when Andy made contact with his own head from ≥15.24 cm using a closed fist. The frequency of problem behavior was recorded during 10-s intervals during each 5-min session. The total frequency of problem behavior in each session was aggregated and then expressed as a rate (i.e., responses per minute) by dividing the sum of problem behavior in each session by 5 (i.e., the total number of minutes in each session). A primary data collector (not the primary therapist) recorded problem behavior during all sessions. This data collector stood in an area of the room that was out of Andy's line of sight (e.g., across the room and behind him); this person cued the primary therapist when to present each edible and sound in the session.

A secondary data collector independently collected data during 20.1% of all sessions in the evaluation. This data collector sat unobtrusively at a table in another part of the room. Interobserver agreement (IOA) was calculated using the interval-by-interval method. An agreement was scored if the frequency of problem behavior was the same in each 10-s interval across each observer's data sheets. The percentage of agreement across both observers was calculated by adding the number of intervals with an agreement, dividing the sum by the total number of intervals in the session (i.e., agreements plus disagreements), and multiplying the quotient by 100. Mean IOA for sessions with Andy was 99% (range, 83% to 100%).

Procedural integrity was assessed by the primary data collector during all sessions in this evaluation, and by the secondary data collector during each session in which they collected data (i.e., 20.1% of all sessions). Data were collected in each 10-s interval; the interval was scored as being implemented correctly if the therapist did the following: (a) the bodily sound was emitted at the scheduled time, (b) the edible was presented at the scheduled time (i.e., 2 s before the bodily sound), and (c) the therapist withheld vocal statements during each 10-s interval. If a procedure was not relevant during any given interval, the data collector scored the therapist as having implemented the procedures correctly unless they committed some other error (e.g., providing any other demands that were not relevant to the session). If an error occurred within a given 10-s interval, the interval was scored as a minus for integrity and feedback was given to the therapist at the end of the 5-min session. Mean procedural integrity was 99% (range, 97% to 100%) during all sessions in the evaluation. Mean IOA for procedural integrity was 99.4% (range, 97% to 100%).

Procedure and Experimental Design

Preference Assessment A paired-stimulus preference assessment (Fisher et al. 1992) was conducted to obtain a hierarchy of Andy's preferences. A total of six items, including Skittles, M&M's, fruit snacks, Doritos, Bugles, and Lay's plain potato chips, were included in the assessment. During each trial, two items were presented simultaneously, and Andy was instructed to select one for consumption. Each item was presented an equal number of times and was paired with every other item twice. After all trials were completed, the total number of selections for each item was added. The items with the most selections were considered relatively more preferred compared to the other items in the assessment. Skittles and fruit snacks were selected the same number of times and were considered to be equally preferred by Andy; these stimuli were used for treatment described below.

General Procedure Sessions were 5 min in duration and included multiple presentations of four specific bodily sounds, including coughing, sneezing, sniffling, and clearing throat. Each session during both baseline and treatment was immediately preceded by a 2-min pre-session period in which Andy was instructed to sit at the table and the primary therapist talked with him about non-specific topics (e.g., activities completed prior to the session, plans for the rest of the day, an area of interest for Andy or the therapist, etc.). However, no demands, including questions, were presented to Andy during the pre-session period. If Andy vocalized during presession (e.g., "Mommy and daddy are on their way home,"), the statement was acknowledged (e.g., "that's right,") but the therapist did not elaborate on the statement further. The purpose of pre-session was to transition Andy to the table where the session would be conducted and for the therapists to gather any remaining materials needed to conduct the procedures (e.g., edibles and data sheets). In addition, it allowed a period of calm for Andy and ensured there was no carryover from other stimuli or events (e.g., interrupting a preferred activity) that may have evoked problem behavior before the treatment session began. If problem behavior occurred, the presession period was reset until 2 min elapsed without problem behavior.

The primary therapist initiated the treatment session immediately after presession following a discreet signal from the primary data collector about when to emit the bodily sound and whether an edible should be paired with the sound. During all sessions, each of the four bodily sounds were presented once in random order before they were repeated. In other words, all four sounds had to be presented once before a specific bodily sound could be presented again by the therapist; this allowed for roughly equal exposure to each bodily sound during each session. Bodily sounds were presented once every 20 s (unless otherwise specified). Between two and four treatment sessions occurred per therapy appointment.

Baseline During baseline, the bodily sound was presented every 20 s unless problem behavior occurred. If problem behavior occurred, the presentation of the next bodily sound was delayed by 30 s from the last instance of problem behavior. For example, if Andy displayed problem behavior 15 s after the presentation of the bodily sound, the next presentation of the bodily sound would occur 30 s after the instance of problem behavior. In addition, the therapist provided vocal attention (e.g., "don't hit yourself," or, "stop throwing things,") following each instance of problem behavior. Thus, escape and attention were contingent on each instance of problem behavior. If problem behavior did not occur during these baseline sessions, the bodily sounds were presented at the set schedule of once every 20 s. Andy did not have access to edibles during baseline sessions.

Treatment When the treatment session began, the primary therapist presented a bodily sound once every 20 s and withheld all comments regardless of problem behavior. Further, the primary therapist did not delay the presentation of the next bodily sound if problem behavior occurred. This resulted in a total of 15 presentations of bodily sounds per session. In addition, a proportion of the bodily sounds emitted by therapists in each session were paired with a highly preferred edible stimulus that was identified through the preference assessment. Specifically, an edible was presented 2 s before a bodily sound was presented along with a statement (e.g., "you can have a Skittle," or, "here's a Skittle,"). The size of the edible that was delivered was roughly equivalent to half of one Skittle.

Initially, treatment included a dense schedule of reinforcement by pairing every bodily sound with an edible (i.e., fixed-time [FT] 20 s). Specifically, in the FT 20 s schedule, each bodily sound was paired with an edible stimulus. However, the proportion of bodily sounds that were paired with an edible stimulus was reduced gradually and systematically over the course of treatment based on Andy achieving steady-state responding within a given condition. For example, in the FT 40 s schedule, an edible was presented every 40 s; however, the bodily sound was still presented every 20 s (i.e., the proportion of edibles that were paired with bodily sounds was reduced to roughly half in each session). If the rate of problem behavior began to increase within a given schedule of reinforcement, the density of the reinforcement schedule was increased to the last reinforcement schedule in which Andy displayed relatively less problem behavior (e.g., moving from a FT 20–40 s schedule to a FT 20 s). When the rate of problem behavior reduced to multiple sessions with no problem behavior, schedule thinning resumed to progressively lean schedules of reinforcement. See Table 1 for details about each treatment condition.

No FT Bodily sounds were presented every 20 s. There were no edibles paired with bodily sounds, and no programmed consequences for problem behavior.

The initial treatment evaluation was conducted using a reversal ABAB design (Baer et al. 1968).

Results

Results are depicted in Fig. 1. During baseline, problem behavior occurred at variable rates with an increasing trend, overall. Andy engaged in problem behavior at an average of 1.2 responses per minute (RPM; range, 0.2 RPM to 2.8 RPM) during this phase. When treatment was introduced, problem behavior was reduced to an average of 0.35 RPM (range,

Table 1 Presentation of Bodily Sounds and Edibles During Each Condition

Condition	Attention	Escape	Bodily Sound	Edible
Baseline	✓	✓	20 s (or 30 s after PB)	–
FT 20 s	–	–	20 s	20 s
FT 20–40 s	–	–	20 s	20 s or 40 s
FT 40 s	–	–	20 s	40 s
FT 60 s	–	–	20 s	60 s
No FT	–	–	20 s	–

FT = fixed time; PB = problem behavior

0 RPM to 1.4 RPM); problem behavior did not occur in three out of four sessions in this phase. The effects of treatment were verified by reintroducing baseline procedures and problem behavior immediately increased to an average of 1.27 RPM (range, 1.2 RPM to 1.4 RPM); problem behavior occurred at elevated and stable rates in this phase. Treatment was subsequently reintroduced and problem behavior did not occur; thus, the effects of treatment were replicated in this phase.

In the next treatment phase (i.e., ESC + ATT EXT, FT 20–40 s), problem behavior increased to an average of 0.52 RPM (range, 0 RPM to 1.8 RPM based on the data in the graph). As a result, the density of the reinforcement schedule was increased to FT 20 s and a decrease in problem behavior was observed over the course of this phase. In the last 20 sessions of this phase, problem behavior occurred at an average of 0.03 RPM (range, 0 RPM to 0.4 RPM). As a result, the process of schedule thinning was continued and the ESC + ATT EXT, FT 20–40 s condition was reintroduced. In this phase, problem behavior occurred at an average of 0.02 RPM (range, 0 RPM to 0.4 RPM), which was less than the rate observed the first time this phase was introduced.

The schedule of reinforcement was thinned further in the next treatment phase (i.e., ESC + ATT EXT, FT 40 s), and the rates of problem behavior increased. Specifically, problem behavior occurred at an average of 0.43 RPM (range, 0 RPM to 3.2 RPM) during the last 20 sessions in this phase. As a result, the schedule of reinforcement was increased to ESC + ATT EXT, FT 20–40 s and problem behavior did not occur in the last 20 sessions of this phase. Thus, ESC + ATT EXT, FT 40 s was reintroduced, and Andy engaged in an average of 0.02 RPM (0 RPM to 0.4 RPM) in this entire phase. Based on low rates of problem behavior, the schedule of reinforcement was advanced further to ESC + ATT EXT, FT 60 s, and Andy's problem behavior further reduced to an average of 0.01 RPM (0 RPM to 0.4 RPM). In the final treatment phase (i.e., ESC + ATT EXT, No FT) during which no edibles were included in the entire 5-min session, there were no instances of problem behavior.

Discussion

The results of this study showed that problem behavior exhibited by an adult with autism and misophonia can be successfully treated using ABA-based procedures. In

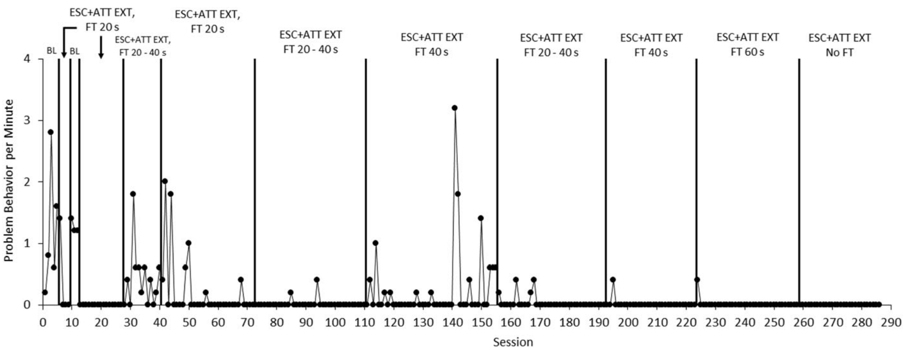

Fig. 1 Andy's problem behavior during the treatment evaluation and schedule thinning. ESC = Escape, ATT = Attention, EXT = Extinction, FT = Fixed time

this study, a treatment package comprised of the systematic exposure to bodily sounds, escape extinction, attention extinction, and pairing bodily sounds with a preferred stimulus (i.e., edibles) successfully eliminated problem behavior in a contrived context.

Past research on this topic included individuals with relatively advanced verbal repertoires who received various combinations of talk-based therapies (e.g., CBT) to address behavioral challenges reported by the participants themselves. Although CBT has been demonstrated to be successful in the treatment of numerous psychiatric conditions (Vigerland et al. 2016), a presumed prerequisite to participate in this type of therapy is a strong communicative repertoire. However, none of the published studies included participants with any reported communication deficits, such as was the case for our participant, so further research on the use of CBT with this population is warranted.

The participant in this study was an individual with severely limited communication skills and severe problem behavior (i.e., aggression, disruption, and self-injury) that more closely resembled the participants in McCord et al. (2001). In McCord et al., the researchers used a differential reinforcement of other (DRO) behavior procedure to treat problem behavior evoked by other sounds (e.g., alarms, insults, loud vocalizations, etc.). That is, preferred stimuli were delivered based on the absence of problem behavior during a given interval of time. In the present study, preferred stimuli (i.e., edibles) were paired with the presentation of trigger sounds (i.e., on a FT schedule), not on the absence of problem behavior, but based on Dozier's (2015) theory that misophonia develops as a conditioned aversive disorder. It is unclear whether a DRO or a FT schedule is a more effective or efficient approach to treating problem behavior triggered by bodily sounds, and future researchers and clinicians should systematically evaluate and compare both treatments for individuals with misophonia.

Another well-documented intervention in the treatment of problem behavior with individuals with ASD is differential reinforcement of alternative (DRA) behavior (Petscher et al. 2009). However, with DRA, the individual is taught to engage in a socially acceptable, alternative response that serves the same function of problem behavior. In the treatment of misophonia, this approach might require the individual to request others to "stop" making the bodily sounds instead of engaging in maladaptive behavior. This treatment approach was not employed in the present study because these behaviors (i.e., sneeze, sniffle, cough, and clearing the throat) cannot be completely suppressed by the person emitting the bodily sound. Thus, requesting individuals to stop coughing might be futile, because bodily sounds often occur involuntarily. However, the individual with misophonia could engage in an alternative response which may be deemed to be socially appropriate, such as offering a drink of water or saying "Bless you," to the individual clearing their throat or sneezing, respectively. Future studies should consider evaluating the efficacy of DRA in the treatment of misophonia.

It is unclear whether the bodily sounds that were presented in this study underwent any form of conditioning for Andy. Evidence to the contrary are based on the fact that problem behavior dropped out entirely after only one treatment session at a FT 20 s schedule. When the baseline procedures were reemployed in the third phase, problem behavior increased again to the initial baseline level. Although it is possible that the other components of the treatment package (i.e., attention and escape extinction) were the key components that produced behavior change, an alternative hypothesis is that the edible presented to Andy facilitated a process of reciprocal inhibition. Specifically, providing Andy the edible stimulus in close temporal proximity to the presentation of the bodily sound could have evoked a set of responses (i.e., picking up the edible to consume) that could have inhibited other responses, namely, problem behavior. Thus, problem behavior increased again during the reversal to baseline. Reciprocal inhibition has been implicated as the underlying mechanism for effectively treating neuroses, such as anxiety disorders (Wolpe 1995), and it is possible the effects of treatment in this study are so attributed.

Some limitations of this study should be noted. First, treatment sessions were conducted in a highly controlled context with stimuli correlated with the onset of the treatment sessions, such as sitting at a table with no instructional materials and a therapist beginning to talk about various topics. Thus, the onset of treatment sessions could have been relatively easily detected by Andy. Although Andy did not display problem behavior toward the end of treatment despite systematic exposure to bodily sounds (and the absence of edibles), problem behavior still occurred when these sounds were accidentally presented by our therapists outside of the programmed treatment sessions or when the sneeze of an individual in a neighboring office could be heard through the walls of the treatment center. Thus, a next step in treatment should be to systematically generalize treatment to extra-therapy contexts by programming common stimuli (Stokes and Baer 1977). For example, incorporating the treatment procedures into other activities with which Andy engages (e.g., while playing ball with others or while he is alone with a puzzle) could promote treatment effects across settings by helping establish stimulus control on Andy's suppression of problem behavior. The lack of generalization we observed is a well-documented challenge in the treatment of individuals with ASD (National Research Council 2001), and future studies should extend this study by addressing generalization.

It is also possible that Andy did not have sufficient exposure to our treatment package; thus, the suppression of problem behavior would not be expected when treatment challenges (e.g., therapists accidentally emitting a bodily sound outside of the treatment context) occurred. A period of five months had elapsed from the initial baseline session until the last treatment session. In their study, Wacker et al. (2011) waited an average of 10.25 months (range, 5 months to 18 months) before systematically presenting the first treatment challenge to participants who had received functional communication training (FCT). It was not until the third treatment challenge was presented (approximately 16 months after FCT was conducted) that the researchers began to observe no problem behavior for some of their participants.

Another limitation is the lack of experimental functional analysis to identify the function of Andy's problem behavior. A functional behavior assessment is crucial to the development of function-based interventions targeting the reduction of problem behavior. The baseline procedures in this investigation were developed based on the report and direct observations of how parents responded to Andy's problem behavior. That is, they reportedly tried to suppress any bodily sounds as best as they could in Andy's presence and also frequently commented on his problem behavior (e.g., "stop hitting"). Our treatment incorporated attention and escape extinction if problem behavior occurred, but also included the pairing of bodily sounds with edibles as an attempt to reduce the potentially aversive quality of bodily sounds perceived by Andy. It is unclear which variable, or combinations of variables, of this treatment package were responsible for behavior change. Future researchers should conduct functional analysis and/or component analyses to identify the critical variables responsible for behavior change.

Misophonia is a relatively understudied topic with clear social implications for the individuals afflicted with the condition as well as their significant others. Future researchers and clinicians should continue to systematically evaluate their treatment efforts with this population for the benefit of other practitioners who encounter patients with this condition.

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Compliance with Ethical Standards

Disclosure of Potential Conflicts of Interest All authors of this manuscript declare that they have no conflicts of interest.

Research Involving Human Participants Ethical approval: All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinski declaration and its later amendments or comparable ethical standards.

Informed Consent Informed consent was obtained from the guardian of the participant included in this study.

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