Treatment of Chronic Breath-holding in an Adult with Severe Mental Retardation: A Clinical Case Study

Derek D. Reed & Brian K. Martens

We describe a clinical case study surrounding the behavioral assessment and operant treatment of, an adult with severe mental retardation who engaged in chronic breath-holding. In this clinical case, previous neurological and medical testing had ruled out biological bases for the individual's breath-holding. A functional behavioral assessement suggested that breath-holding was likely maintained by contingent staff attention. Treatment analysis consisted of the following conditions: baseline, physical redirection, fixed-time schedule of attention, differential reinforcement of other behavior, and a treatment package. Ultimately, a treatment package consisting of physical redirection and differential reinforcement of other behavior was found to be the most viable treatment option, reducing rates of breath-holding by 50%.

Breath-holding spells, or BHS, are uncommon in children, observed in only 4.6% of the childhood population (DiMario, 2001). Individuals demonstrating breath-holding episodes may experience cyanosis and, in some cases, death resulting from complications related to loss of consciousness (Paulson, 1963). To date, no prevalence information is available concerning the occurrence of breath-holding spells in adults. Childhood occurrences of breath-holding typically dissipate before the age of 2, even when no treatment has been implemented (MedlinePlus, 2005). The U. S. National Library of Medicine (MedlinePlus, 2005) suggests that children typically engage in breath-holding as a response to fear, trauma, or some other startling event. Other research has also suggested that breath-holding in adults may result from other psychological conditions such as somataform disorder (Inagaki, et al., 2001).

While the U. S. National Library of Medicine alludes to a psychological component (i.e., emotion) of breath-holding, recent research suggests an additional biomedical explanation. Specifically, Bhat and colleagues (2007) found 35 cases of anemia in a sample of 59 children exhibiting breath-holding spells. Both the anemic and non-anemic breath-holding children were given oral iron therapy for 12 weeks. Results of the study indicated a 77% reduction for the anemic breath holders, with only a 29% reduction for the non-anemic breath holders, suggesting a possible medical explanation for at least some cases of breath-holding.

Author Note
We thank Frances M. Szczech for her assistance throughout the duration of this study.

While these aforementioned psychological and medical conditions offer insight into what may have caused the initial breath-holding episodes, they do not explain the maintenance of breath-holding when these conditions are either (a) absent in the individual, or (b) ameliorated due to previous treatment. A plausible hypothesis for the maintenance of breath-holding episodes under such conditions may be that initial occurrences of breath-holding were reliably followed by certain social consequences in the individual's environment. Under this philosophical framework, the breath-holding individual may not necessarily be cognizant of his/her reasons for engaging in this behavior, but it may have been shaped or conditioned over time. In recent years, some researchers have begun speculating that breath-holding, in at least some individuals, may be under operant control (Kern, Mauk, Marder, & Mace, 1995; Richman, Lindauer, Crosland, McKerchar, & Morse, 2001; Singh, 1979). Thus, given the effective use of behavioral technologies, the operant relationships governing the occurrence of breath-holding may be modified with simple consequence-based procedures and/or antecedent manipulations.

One of the first documented operant treatments for breath-holding was published by Singh in 1979. In this study, a 15 month old boy presented with severe breath-holding, which maintained even after medical and psychodynamic approaches to treatment. Singh's operant approach consisted of aromatic ammonia presented to the boy contingent upon breath-holding episodes. Following implementation of this procedure, the breath-holding behavior quickly reduced to zero levels and maintained over an additional year of observations. Despite this procedure's clear effect, its application in applied settings may be met with skepticism and

low social validity due to its sole reliance on punitive consequences. Moreover, Singh's procedure did not reinforce or promote appropriate breathing, which would likely be more effective for long-term maintenance.

Recent advances in behavioral technology have allowed clinicians to identify the possible maintaining functions of aberrant behavior. One such approach has been the use of systematic manipulations of environmental events conducted within a multi-element research design to quickly identify function while retaining experimental control during the assessment process (i.e., functional analysis; Iwata, Dorsey, Slifer, Bauman, & Richman, 1982/1994). In an example of function-based treatment for breath-holding, Richman, Lindauer, Crosland, McKerchar, and Morse (2001) describe the successful use of functional analysis and treatment analysis to treat a 16-year-old male diagnosed with mental retardation. Similar to Singh's (1979) example, medical or biological explanations were ruled out given the participant's health status. Richman and colleagues (2001) demonstrated that the boy's breath-holding occurred during the alone and ignore conditions of a functional analysis, suggesting that behavior was maintained by nonsocial reinforcement (e.g., physical sensations) rather than caregiver attention. Moreover, caregiver reports suggested that reprimands appeared to be effective at managing this behavior at home. Thus, when a reprimand was paired with differential reinforcement of other behavior (DRO) using physical activities as reinforcers, breath-holding reduced to near zero levels. This example demonstrates the advantages of pairing a punishment technique to decrease breath-holding with a reinforcement-based procedure to promote an appropriate functional alternative to the target behavior. In addition, through an inclusion of caregiver recommendations, these authors were able to design a socially valid treatment.

Kern, Mauk, Marder, and Mace (1995) also demonstrated the advantages of using functional analysis as an assessment tool in the formulation of treatment components for a 7-year-old girl diagnosed with Cornelia-de-Lange syndrome and mental retardation. Again, no medical explanations could account for the persistence of the breath-holding episodes. Subsequent functional analysis suggested that breath-holding occurred most often when contingent attention was applied. Given this hypothesis, treatment consisted of ignoring the breath-holding, delivering attention on fixed schedules, and teaching the participant functional communication for appropriate requesting of attention. Treatment analysis concluded that this intervention package reduced breath-holding to near zero levels.

The present case study leads the reader through the entire problem-solving process in a case of chronic breath-holding in an adult with mental retardation. Specifically, this case study will highlight the use of both indirect and direct functional assessment techniques to guide problem identification and treatment conceptualization. In addition, this study describes the methodology behind our treatment analysis to arrive at a data-based decision regarding intervention selection for breath-holding episodes.

Case Study

Participant

The participant in this case study was a 38-year-old woman named Jane. Jane presented with severe mental retardation, scoring in the below average range in adaptive behaviors as compared to norms on the Vineland Adaptive Behavior Scale (Sparrow, Balla, & Cichetti, 1984). All attempts at administering cognitive assessments were unsuccessful due to her low functioning status. Jane engaged in occassional self-injury, with topographies ranging from head banging, hand-biting, and gouging of her ears. Jane was referred for an acute consultative relationship with the first author due to an increasing level of breath-holding. Staff reported that Jane's breath-holding had been occurring for over three years, and occassionally resulted in fainting and injury. Previous neurological assessments had determined that no medical or biological bases were contributing to the breath-holding episodes.

Setting

Jane resided in residential home with 11 other consumers with disabilities ranging from Down's Syndrome to acquired brain inury. At any given time, three to four staff were on duty in the residential home. All assessments and interventions were conducted in this setting.

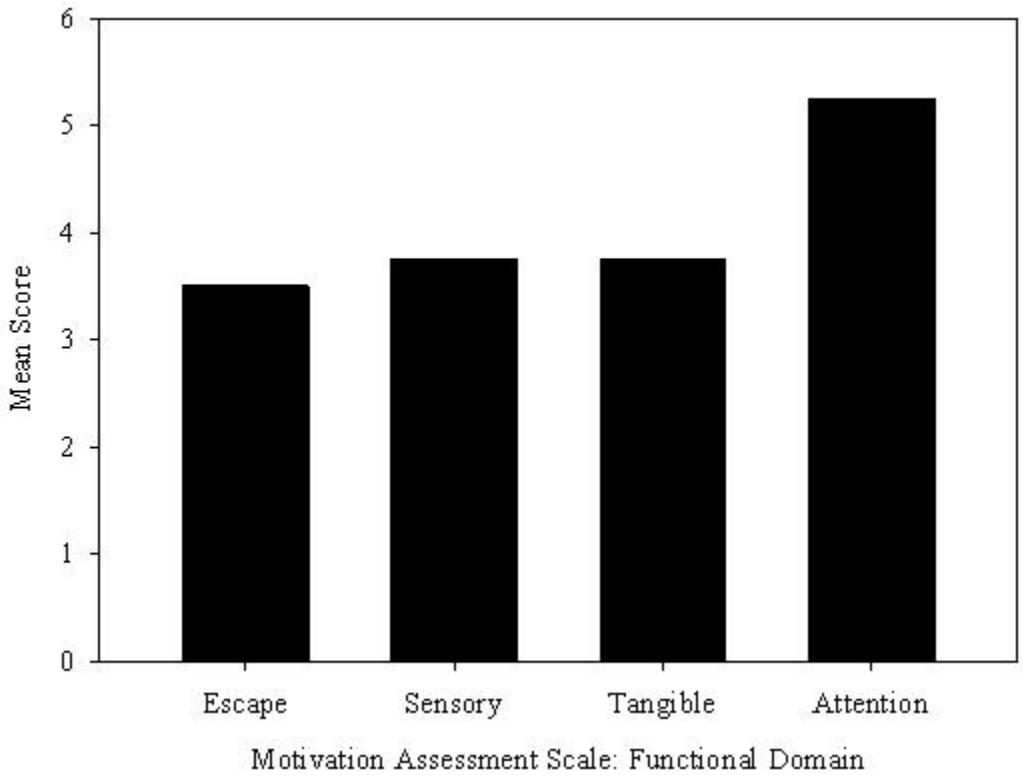

Figure 1: Results of the Motivation Assessment Scale. Mean scores were derived by averaging the scores from each of the four items on each respective functional domain.

Indirect Functional Behavioral Assessment

Problem identification interview (PII). A problem identification interview (PII; Bergan & Kratochwill, 1990) was conducted in order to address the scope of the residential staff's concerns regarding Jane's breath-holding. Specifically, two staff members with over one year of direct care experience with Jane participated in the interview. These staff reported that Jane needed constant attention, with problem behavior or breath-holding occurring after 2-3 minutes of ignoring. Staff indicated that Jane's breath-holding had never occurred (to their knowledge) when she was in an unsafe environment, such as on the stairway or in the shower. However, staff feared that significant injury may occur if breath-holding resulted in fainting in either situation.

As part of the PII, staff worked conjointly with the consultant to arrive at an operational definition of Jane's breath-holding behavior. Interestingly, Jane engaged in a ritualized behavior of slowly placing her hands over her ears when beginning to hold her breath. Thus, the definition agreed upon was "the holding of both palms and/or hands up to ears, raising elbows towards the ceiling, eyes fixed in one direction, long inhalation without exhalation, with stiffening of muscle tone throughout body." Staff estimated that Jane's breath-holding occurred at a rate of 1.25 times per hour.

When asked to identify potential antecedents to Jane's breath-holding, staff hypothesized the following: (1) lack of attention, (2) busy times, such as cooking, transitions, morning routines, etc., (3) transitioning back into the group home after returning from the day program, and (4) when staff were around her. Potential consequences were hypothesized to be (1) staff attention, and (2) sensory experiences. At the time of the PII, Jane's behavioral support plan indicated that staff should lightly touch Jane's stomach and blow a puff of air into her face when she began breath-holding. The rationale behind this plan was that these events elicited laughing in Jane, thereby disrupting the breath-holding episode. However, staff reported dissatisifaction with this plan due to the increasing rates of breath-holding they were observing. Staff indicated that their goal for change was a significant reduction in breath-holding to manageable rates. Ideally, they wished for zero levels of breath-holding, but indicated that any reduction would be viewed as a success.

Motivation Assessment Scale (MAS)

In an effort to further elucidate hypothesized functions of Jane's breath-holding, the Motivation

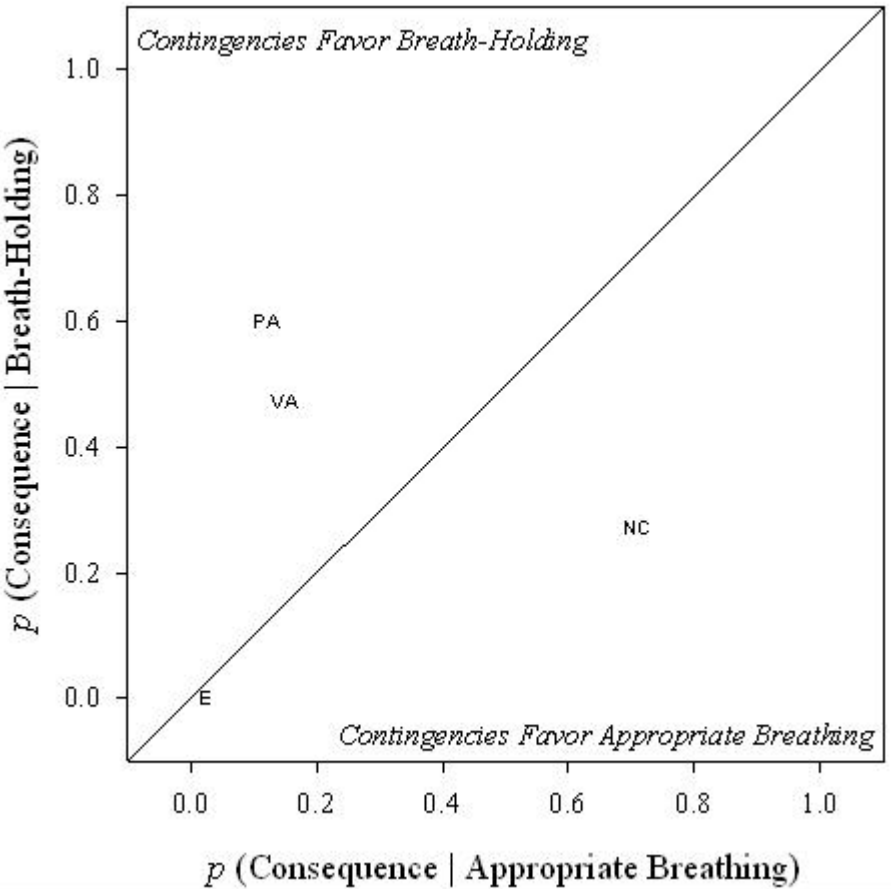

Figure 2: Operant contingency space analsysis of Jane's breath-holding. Conditional probabilities of physical attention (PA), verbal attention (VA), escape (E), and no overt consequences (NC) given both breath-holding and appropriate breathing are presented on the ordinate and abscissa, respectively. Each conditional probability pairing datum point on the figure is depicted by the respective consequential event's abbreviation.

Assessment Scale (MAS; Durand & Crimmins, 1989) was administered to both staff members who participated in the PII. The MAS is a 16-item likert-type scale, which asks the informant to indicate how likely the client is to engage in the problem behavior during different antecedent and consequential events (ranging from 0 [never] to 6 [always]). The 16 items are equally distributed across four function domains (i.e., sensory, escape, attention, tangible), with higher scores in a given domain indicating more confidence in that domain serving as the function of the client's behavior. Figure 1 depicts the mean scores obtained from the staff with regards to Jane's breath-holding. Specifically, Jane's staff scored escape as the least probable function (mean score: 3.50), with sensory and tangible tied for the second probable function (mean score: 3.75 each). The most probable function was attention, with a mean score of 5.25. These data corroborate the verbal report of the staff during the PII.

Direct Functional Behavioral Assessment

Temporal analysis

Twenty minute observerations using frequency counts targeting Jane's breath-holding were conducted during (1) Jane's arrival to home from the day program, (2) while dinner was being prepared, and (3) after dinner, in response to the staff's report that Jane's breath-holding occurred most during busy times in the home. Results of these observations suggested that Jane's breath-holding occurred at near-equal rates across the three temporal classes. Specifically, breath-holding was observed at rates of 12.0, 13.8, and 12.8 breath-holding episodes per hour for the three observation periods, respectively. These rates were substantially higher than those estimated by staff during the PII.

Contingency space analysis. A contingency space analysis (Eckert, Martens, & DiGennaro, 2005; Schwartz, 1989) was conducted to determine the degree of contingency (see Martens, DiGennaro, Reed, Szczech, & Rosenthal, in press) for consequential events associated with Jane's breath-holding, based upon conditional probabilities of behavior-consequence pairings. Specifically, three twenty minute observations using a 15-sec partial interval recording system were conducted between the transition home from work and dinner. As per the continengcy space analysis, behavior-consequence pairings were conducted for both the occurence and non-occurence of breath-holding episodes. Coded consequences for the contingency space analysis included withdrawal-of-command (i.e., escape),

other (no overt consequence observed), verbal attention, and physical attention (i.e., touch). Attention was differentiated between verbal and touch to take into account the protocol used as part of her behavior support plan (BSP) for touching Jane's stomach during a breath-holding episode. The results of this contingency space analysis are presented in Figure 2.

Within a contingency space analysis, data points falling above the diagonal line suggest that the consequential event favors the occurrence of the problem behavior (i.e., the maintaining function). Data points falling below the diagonal line suggest that those consequential events favor appropriate/desired behaviors. In the present case study, Jane's breath-holding appeared to be maintained by both forms of attention (physical attention and verbal attention). On the contrary, Jane's appropriate breathing was primarily ignored (i.e., no observed consequence) by her assigned staff. Due to the lack of demands directed to Jane, there were very few opportunities for escape. The data from this direct functional behavioral assessment hypothesizing attention as the maintaining function for Jane's breath-holding corroborates both the PII and the MAS. Unfortunatley, given the contingent (see Martens et al., in press), rather than dependent, properties of the attention data points, these findings suggest that Jane's BSP was not being followed with high degrees of integrity. That is, since Jane's BSP indicates that every instance of breath-holding should be interupted with a touch to the stomach, we would expect the conditional probability of touch to be 1.00. The data in the present analysis suggests that this procedure was omitted during approximately 50% of instances of Jane's breath-holding.

Conclusions of the Functional Assessment

Based upon both indirect and direct functional behavioral assessment techniques, the following conclusions were made regarding Jane's breath-holding behavior: (1) Jane had learned contingencies relating attention to breath-holding, (2) breath-holding was maintained by both verbal and physical attention, due in part to her behavior support plan, (3) no overt contingencies were in place for appropriate breathing, and (4) a functionally equivalent treatment plan may reverse these contingencies, thereby favoring appropriate breathing.

Treatment Analysis

Based upon the concerns of the staff, as well as the findings and conclusions derived from the functional assessment, a brief experimental analysis of treatments (see Martens, Eckert, Bradley, & Ardoin, 1999) was conducted to quickly identify an appropriate procedure to reduce Jane's breath-holding episodes. Specifically, the individual treatment components identified for analysis were a phsyical redirecton procedure, a fixed-time schedule of attention, and a differential reinforcement of other behavior (DRO) procedure. All sessions were 10 minutes in duration. Observations were done using a 15-sec partial interval recording system. A multiple treatment reversal design was utilized, using sequential applications of treatment components with abridged data series and a mini-withdrawal, as per BEA methodology (Martens, et al., 1999). This design element was selected due to its ability to quickly identify viable treatment options without complete sacrifice of experimental control.

Baseline

During baseline conditions, the consultant observed staff interactions with Jane while keeping the staff blind to the results of the functional assessment. Specifically, staff were instructed to interact with Jane as they naturally would. Results from this condition are presented in the first phase of Figure 3, as well as the fifth phase as a one session mini-withdrawal of treatment. During baseline conditions, percentages of intervals with breath-holding ranged from 10.0 to 12.5 in phase one, and was 17.5 during the mini-withdrawal.

Physical redirection (PR)

During the physical redirection conditions, the consultant would approach Jane from behind, and gently guide her hands down upon any instance of Jane taking a long inhalation and raising her hands to her ears. No eye contact or verbal attention was provided during this procedure. This condition was selected in an effort to minimize attention to her breath-holding, while maintaining her personal safety. The physical redirection phase is presented as the second phase in Figure 3, with percentages of intervals with breath-holding ranging from 2.5 to

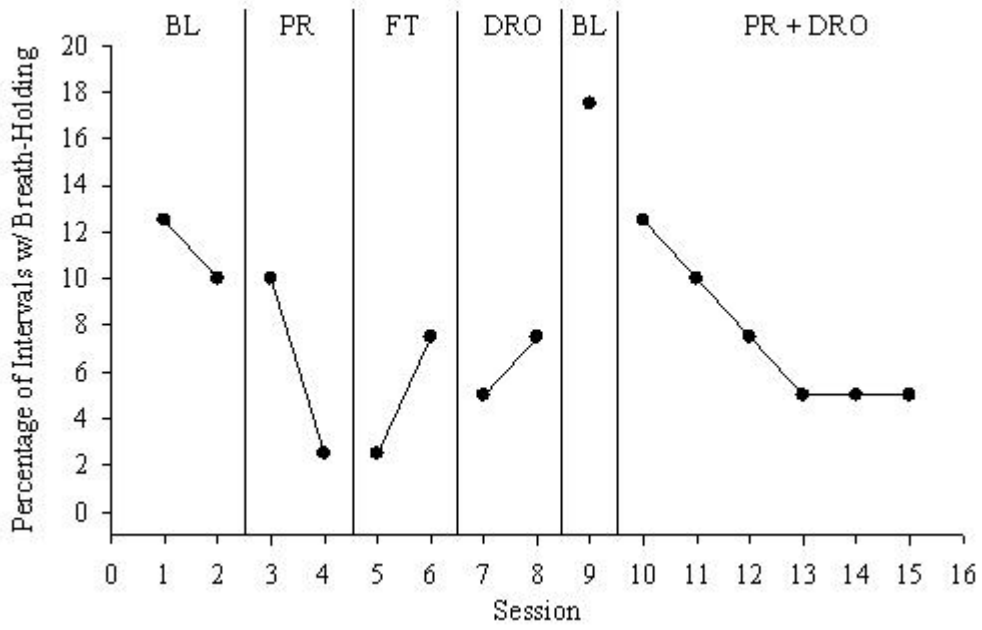

Figure 3. Number of 15-second intervals with occurrences of breath-holding for Jane, in each of the five study conditions. BL = baseline; PR = physical redirection; FT = fixed-time schedule (72-sec); DRO = differential reinforcement of other behavior (30-sec whole-interval); PR + DRO = treatment package of physical redirection plus differential reinforcement of other behavior (30-sec whole-interval).

10.0 demonstrating a clear reduction in the target behavior.

Fixed-time schedule of attention (FT)

Results from the direct observations suggested that staff interacted with Jane approximately once every 90-sec. Based upon this baseline rate, a fixed-time schedule of 72-sec (FT:72) was selected to provide attention approximately 20% sooner than was provided during baseline interactions. However, to limit incidental reinforcement (Vollmer, Ringdahl, Roane, & Marcus, 1997), a 5-sec absence of breath-holding was required before attention was provided. Attention came in the form of both verbal and physical forms. Verbal attention was kept to neutral non-directive statements regarding the immediate context of the environment (e.g., "Jane, the sky looks pretty dark outside right now"). Physical attention came in the form of the consultant reaching out to Jane and firmly holding her hand while verbal attention was delivered. Instances of breath-holding were ignored during this condition. However, if the episode was deemed dangerous (due to signs of cyanosis), Jane's behavior support plan was to be followed. There were no instances of dangerous episodes during any of these session.The FT condition was selected in order to examine the effectiveness of a dense schedule of reinforcement, which we hypothesized would decrease any active motivation operations (e.g., long periods of time without social interaction) which previously increased the likelihood of breath-holding.

Results of the FT condition are presented as the third phase in Figure 3, with percentages of breath-holding ranging from 2.5 to 7.5. These data suggest a decrease from baseline, but no clear advantage from the PR condition.

Differential reinforcement of other behavior (DRO)

A DRO condition was selected for analysis in an attempt to teach Jane that appropriate breathing would be reinforced, while breath-holding would not result in any further attention. Specifically, Jane was provided with contingent verbal praise (e.g., "You're doing a great job being safe") and physical attention (similar to FT condition) for 30-sec of continuous breathing. All instances of breath-holding were ignored, unless deemed dangerous. At no point during any DRO sessions did a dangerous episode occur. The DRO condition is depicted in Figure 3 as the fourth phase of the design, with percentages of breath-holding ranging from 5.0 to 7.5. These data suggest a stable reduction from BL, but no superiority over either the PR or FT conditions with regards to reduction of breath-holding.

Physical redirection + differential reinforcement treatment package (PR + DRO).

A treatment package combining the physical redirecton and differential reinforcement procedure was implemented following a mini-withdrawal. These treatments were selected for the package due to their successful reduction of breath-holding episodes, and ability to keep Jane safe while promoting appropriate breathing within a functional perspective through the inclusion of contingent verbal and physical attention. Moreover, staff indicated preference for these treatments due to ease of implementation, suggesting some indication of social validity. Results from the package are depicted in the sixth phase of the design in Figure 3. Suprisingly, levels of breath-holding were initially high during the first two sessions of the treatment package, with percentage of intervals of 12.5 and 10.0, respectively. While initially relatively high in comparison to other treatment condition, these sessions indicated an immediate marked decrease from the mini-withdrawal levels of breath-holding. Over the next four sessions, breath-holding levels reduced to stable levels of 5.0 percent of intervals. In sum, the treatment package reduced breath-holding to half the percentage of intervals demonstrated in the initial baseline.

Discussion

Breath-holding is a rare but serious behavior which presents challenges to clinicians due to its various forms of etiology and maintaining variables. Moreover, while a fair amount of research has attempted to understand the variables surrounding the occurrence of breath-holding in children, little work has been done regarding this phenomenon in adult populations. Fortunately, recent research has demonstrated that operant approaches may prove to be a viable treatment option in the reduction of breath-holding occurring in the absence of medical conditions (Richman et al., 2001). Specifically, techniques such as functional assessment may now afford clinicians with a target appropriate alternative response sharing the same behavioral functions as breath-holding.

The present case study utilized both indirect and direct functional assessment techniques. Throughout each assessment, attention was found to be the likely function of the breath-holding behavior. Such corroboration amongst assessment techniques increased our confidence in the hypothesis that the contingent delivery of attention by staff for Jane's breath-holding was the likely maintaining function. In retrospect, we believe that the topography of Jane's breath-holding which involved slowly lifting her arms and elbows and covering her ears while engaging in breath-holding was further evidence of an attention component. Specifically, staff began attending to Jane's ear-covering due to its easy visual cueing. Jane may have learned that ear-covering elicited more attention than breath-holding alone, because holding one's breath is not readily observable by outsiders.

Upon targeting attention as the likely maintaining function, we systematically evaluated the effects of various interventions which were previously documented as effective treatment options for breath-holding. Through the use of a brief experimental analysis, we quickly identified the most practical, yet effective, treatment which staff could carryout upon the conclusion of the consultative relationship. By incorporating staff's concerns of Jane's safety through the inclusion of the physical redirection condition, we likely increased the social validity of our treatment, which could impact the integrity to which the plan is followed by the staff implementing it.

Due to the acute nature of the consultative relationship described in this study, several limitations exist, and appropriate caveats to interpretation must be presented. First, due to confidentiality issues related to the residential home, no other independent observers were available to collect reliability or treatment integrity data. Second, due to time constraints we were only able to collect two observational probes per treatment condition. It is unknown whether or not breath-holding would have continued descending during the physical redirection condition, or whether or not it would continue ascending to higher rates during the FT and DRO conditions. Indeed, continuing data collection until steady state responding was observed would be ideal to best understand the effects of these treatment conditions. Third, long-term follow-up data were not collected in order to evaluate the maintenance of this treatment over time. Fourth, we were unable to conduct analog functional analysis conditions due to the safety risks associated with Jane's levels of breath-holding and because of

feasibility issues surrounding data collection in a residential home. Finally, no standardized or systematic social validity assessments were conducted.

While these limitations compromise the experimental rigor of our analyses, we believe our approach to case conceptualization and treatment analysis may serve as a springboard for other clinicians interested in treating pervasive breath-holding behavior, or researchers interested in continuing to refine treatment approaches. Specifically, future research should evaluate appropriate analog functional analysis conditions for breath-holding behavior in natural settings, as well as ways to incorporate participant choice and preference in the selection of treatment conditions (e.g., concurrent-chains schedules) to further promote social validity.

References

Bergan, J. R., & Kratochwill, T. R. (1990). Behavioral consultation and therapy. New York: Plenum Press.

Bhat, M. A., Ali, W., Mohidin, K., & Sultana, M. (2007). Prospective study of severe breath holding spells and role of iron. Journal of Pediatric Neurology, 5, 27-32.

DiMario, F. J. (2001). Prospective study of children with cyanotic and pallid breath-holding spells. Pediatrics, 107, 265-269.

Durand, V. M., & Crimmins, D. B. (1988). Identifying the variables maintaining self-injurious behavior. Journal of Autism and Developmental Disorders, 18, 99-117.

Eckert, T. L., Martens, B. K., DiGennaro, F. D. (2005). Describing antecedent-behavior- consequence relations using conditional probabilities and the general operant contingency space: A preliminary investigation. School Psychology Review, 34, 520-528.

Inagaki, T., Horiguchi, J., Mizuno, S., Kanata, K., Miyaoka, T., Hiruta, H., et al. (2004). Breath- holding spells in somatoform disorder. International Journal of Psychiatry in Medicine, 34, 201-205.

Iwata, B. A., Dorsey, M. F., Slifer, K. J., Bauman, K. E., & Richman, G. S. (1994). Toward a functional analysis of self-injury. Journal of Applied Behavior Analysis, 27, 197–209. (Reprinted from Analysis and Intervention in developmental Disabilities, 2, 3–20, 1982)

Kern, L., Mauk, J. E., Marder, T. J., & Mace, F. C. (1995). Functional analysis and intervention for breath holding. Journal of Applied Behavior Analysis, 28, 339-340.

Martens, B. K., DiGennaro, F. D., Reed, D. D., Szczech, F. M., & Rosenthal, B. D. (in press). Contingency space analysis: An alternative method for identifying contingent relations from observational data. Journal of Applied Behavior Analysis.

Martens, B. K., Eckert, T. L., Bradley, T. A., & Ardoin, S. P. (1999). Identifying effective treatments from a brief experimental analysis: Using single-case design element to aid decision making. School Psychology Quarterly, 14, 163-181.

MedlinePlus. (2005). Breath holding spell. Retrieved February 29, 2008, from http://nlm.nih.gov/medlineplus.

Paulson, G. (1963). Breath holding spells: A fatal case. Developmental Medicine & Child Neurology, 5, 246-251.

Richman, D. M., Lindauer, S. D., Crosland, K. A., McKerchar, T. L., & Morse, P. S. (2001). Functional analysis and treatment of breath holding maintained by nonsocial reinforcement. Journal of Applied Behavior Analysis, 34, 531-534.

Schwartz, B. (1989). Psychology of learning and behavior. New York: W. W. Norton & Co.

Singh, N. N. (1979). Aversive control of breath-holding. Journal of Behavior Therapy and Experimental Psychiatry, 10, 147-149.

Sparrow, S. S., Balla, D. A., & Cicchetti, D. V. (1984). Vineland Adaptive Behavior Scales: Interview Edition, Survey Form Manual. Circle Pines, MN: American Guidance Service.

Vollmer, T. R., Ringdahl, J. E., Roane, H. S., & Marcus, B. A. (1997). Negative side effects of noncontingent reinforcement. Journal of Applied Behavior Analysis, 30, 161-164

Derek D. Reed, M.S., BCBA
320 Rindge Ave #101
Cambridge, MA 02140
dreed.syr@gmail.com

Brian K. Martens, Ph.D.
Department of Psychology
430 Huntington Hall
Syracuse University
Syracuse, NY 13244
bkmarten@syr.edu