8-9-2019

Exploring the utility of brief functional analyses procedures for individuals with CHARGE syndrome

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By

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A Dissertation
Submitted to the Faculty of
Mississippi State University
in Partial Fulfillment of the Requirements
for the Degree of Doctor of Philosophy
in Educational Psychology (School Psychology Concentration)
in the Department of Counseling, Educational Psychology, and Foundations

Mississippi State, Mississippi

August 2019

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Title of Study: Exploring the utility of brief functional analyses procedures for individuals with CHARGE syndrome

Pages in Study 107

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A critical step in addressing problem behavior is identifying the function of problem behavior, or reason for engaging in the problem behavior, using functional analysis (FA). Individuals with CHARGE Syndrome engage in problem behaviors that vary across topographies and etiology (e.g., pain, anxiety, sensory concerns; Hartshorne et al., 2017). The literature has illustrated time and time again the effectiveness of these procedures across populations, settings, age groups, and topographies of behavior; however, no studies have been documented exploring the utility of FA procedures with individuals with CHARGE Syndrome. The current study completed brief functional analyses (Northup et al., 1991) with individuals diagnosed with CHARGE Syndrome who presented with problem behavior. Participants included individuals between the ages of 8 to 22 years old diagnosed with CHARGE Syndrome and presenting with problem behaviors. Results indicated that BFA procedures were successful in identifying the function of problem behavior with 4 out of 5 participants.

DEDICATION

While there are several people I can think of that inspired my dissertation, it would simply not have been possible without the generous families of children with CHARGE Syndrome that I encountered along the way. After attending my first CHARGE Syndrome conference, I was astounded by families' resilience, kindness, passion for helping their children achieve their full potential, and willingness to share their experiences, as well as their amazing children with me. Knowing that I wanted to make CHARGE and behavior the focus of my dissertation, I knew it would require the cooperation of families and that it would require effort on the part of both caregivers and the child. When presented with the tasks associated with my dissertation, participating families did not think twice and were more than helpful throughout the process. The dedication of CHARGE families to furthering research and raising awareness is inspiring and will continue to motivate me to complete research in this area. I cannot thank the families that participated in my dissertation enough for their time and effort. This work is dedicated to you.

ACKNOWLEDGEMENTS

Of the people I want to thank for making my dissertation possible, I can think of those who were both on and off of the front lines. First and foremost, I want to thank Dr. Dan Gadke and Dr. Kasee Stratton for their tireless effort in making what I originally thought would be a near impossible study, a reality. You were both always available to answer questions, help me problem solve, and lend a listening ear when the task at hand became overwhelming. I am so thankful for all of your guidance, support, and general understanding not only when it came to my dissertation, but throughout my entire graduate school journey.

Second, I would like to thank the other students in my graduate program – you all have always been such a wonderful source of support and I could not have completed this task without you. Specifically, I want to thank my peers who assisted me with data collection: Megan Anderson, Matthew Ferrigno, Keely McCulla, and Alex Clarke. I greatly appreciate your time and effort in assisting me with this project and absolutely could not have successfully completed this huge milestone without your help. You were all patient when we experienced issues that required problem solving and willingly offered your assistance and support throughout the process – I cannot thank you enough!

Lastly, I want to thank my family – specifically, my mom and dad. Thank you for always encouraging me to take the necessary steps to complete my goals and supporting me along the way, even from a distance. You were both always willing to listen and provide support in any

way you could. Finally, thank you for never failing to answer my late night phone calls and lending a listening ear.

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CHAPTER I

INTRODUCTION

The field of behavior analysis has long been oriented toward the goal of successful assessment and treatment of problem behaviors in individuals of all ages and diagnoses. In response to this goal, procedures referred to as functional analyses (FA) have been developed to assist in the accurate identification of the function of problem behaviors. By identifying the function of a problem behavior, appropriate interventions can be designed and implemented to reduce engagement in problem behavior. Over the years, FA procedures have been altered to meet the needs of the field (i.e., increasing efficiency, accuracy) and continue to be an area of extensive research. Efforts to extend FA procedures to different populations and behavioral presentations have been made; however, the utility of FA procedures for individuals with CHARGE Syndrome, the leading cause of congenital deafblindness (U.S. Department of Education, National Center on Deaf-Blindness, 2017), have yet to be explored.

Successfully intervening on problem behaviors is critical in order to reduce the likelihood of academic and social concerns and decrease the likelihood of negative outcomes, such as reduced academic achievement (Algozzine, Wang, & Violette, 2011), distress of the individual and those who interact with them (Dominick et al., 2007), tissue damage as a result of physical harm to the self or others, decreased ability to learn adaptive and appropriate behavior, and difficulty maintaining social relationships (Herzinger & Campbell, 2007). The use of FA methodology to identify functions of problem behavior (which then informs intervention) has

been shown to be effective for a variety of behaviors and populations (Iwata, Dorsey, Slifer, Bauman, & Richman, 1982/1994; Northup et al., 1991). Problem behavior was historically managed primarily through extinction and punishment procedures (Axelrod, 1987); however, with the development and growth of applied behavior analysis (ABA) professionals began to consider the function, or ultimate goal, of problem behavior when designing interventions (Carr, 1977).

Following the identification of functions of behavior by Carr (1977), research began to be conducted on methodologies that would lend themselves to the accurate identification of the functions of problem behaviors. Iwata et al. (1982/1994) first implemented the traditional FA methodology, which consisted of four 15-min conditions. These included: social disapproval, academic demand, unstructured play, and alone (Iwata et al., 1982/1994). While this method proved to be effective and useful, the traditional FA methodology required multiple replications of conditions along with lengthy condition times, which was a concern of many who were implementing them. Northup and colleagues (1991) implemented brief FA (BFA) procedures that would increase efficiency by decreasing session times to 5 or 10 min. Further, the BFA included different conditions (social attention, escape from a demand, tangible, and alone) than those used in traditional FA, as well as using a confirmatory analysis procedure to ensure accurate findings. Lastly, Hanley, Jin, Vanselow, and Hanratty (2014) aimed to further improve FA methodologies by allowing for single conditions to address multiple functions underlying a single problem behavior. Hanley and colleagues implemented preference assessments and functional interviews with caregivers through the interview informed synthesized contingency analysis (IISCA). The IISCA was designed to be more efficient, to assist in addressing instances

in which other FA methodologies produce little or no differentiation across conditions, or if multiple functions are hypothesized to interact to maintain the behavior.

While there has been a plethora of research conducted on the use of FA methodologies with common problem behaviors (e.g., self-injurious behavior, aggression) only recently have FA methodologies been used in the treatment of unique problem behaviors (e.g., inappropriate sexual behavior, rumination; Beavers, Iwata, & Lerman, 2013). Further, there is limited research on the utility of FAs in low incidence populations, such as individuals with severe intellectual disabilities or sensory impairments (Delgado-Casas, Navarro, Garcia-Gonzalez-Gordon, & Marchena, 2014; Magee & Ellis, 2000). The present study aims to examine the utility of FA methodology with individuals with CHARGE Syndrome, a rare genetic disorder that is the leading of cause of congenital deafblindness. The researcher aimed to target individuals with CHARGE Syndrome between the ages of 6 and 22 years who have had the majority of their necessary medical procedures completed and were within the maximum age to attend school.

Due to CHARGE Syndrome being a low incidence disability, this line of research is important in order to begin filling the gaps in the literature with regards to problem behavior of individuals with CHARGE Syndrome. CHARGE is a genetic condition that is characterized by the following medical features: coloboma of the eye, heart defects, atresia choanae, delayed (retardation) growth and development, genital hypoplasia, and ear anomalies/deafness (Blake et al., 1998; Blake & Prasad, 2006). A behavioral phenotype developed for individuals with CHARGE Syndrome includes the following: (a) low normal cognitive functioning, (b) very goal directed and persistent with a sense of humor, (c) socially interested but immature, (d) repetitive behaviors that increase under stress, (e) high levels of sensation seeking, (f) difficulty with self-regulation when under conditions of stress and sensory overload, and (g) difficulty shifting

attention or transitioning to new activities (Hartshorne, 2011). Further, sensory impairments experienced by these individuals may result in unique problem behaviors, such as self-stimulatory behaviors (Hartshorne et al., 2009). Further, individuals with CHARGE are often diagnosed with obsessive-compulsive disorder (OCD), attention deficit/hyperactivity disorder (ADHD), Tourette Syndrome, and autism spectrum disorder (ASD), which could result in comorbid problem behaviors.

Problem behaviors in individuals with CHARGE can manifest through physical, verbal, and nonverbal topographies and can be affected by concerns regarding sensory issues, anxiety, and pain (Hartshorne et al., 2017). The most complex potential influence on problem behavior is pain. Individuals with CHARGE syndrome undergo a multitude of medical procedures (Stratton & Hartshorne, 2018) that often lead to the experience of chronic pain (Nicholas, 2011; Stratton & Hartshorne, 2018). Chronic pain can lead to engaging in a variety of problem behaviors (Courtemanche, Black, & Reese, 2016; Symons & Danov, 2005) and an increase in internalizing concerns, such as anxiety (Nicholas, 2011). Further, the experience of chronic pain also brings into question whether automatic reinforcement, communicating the painful experience, or reactions of others in the environment are maintaining problem behavior.

By conducting this study with individuals with CHARGE Syndrome, the researcher aims to begin to fill the gap of understanding the level of effectiveness and utility of FA methodology in identifying the functions of problem behavior in individuals with unique problem behaviors and low incidence disabilities. Further, this study will explore the role of pain in the presentation of problem behaviors experienced by individuals with CHARGE Syndrome and investigate the utility and accuracy of contingency reversals in confirming results obtained from BFA procedures.

Research Questions

This study seeks to answer the following research questions.

Research Question #1: Can BFA procedures be used to detect functions of problem behaviors in children with CHARGE syndrome/multisensory impairments?

Research Question #2: Can a contingency reversal confirm and further validate the BFA procedures and findings?

Research Question #3: If the frequency of engagement in problem behaviors are undifferentiated across BFA conditions, was pain elevated on the CHARGE Non-Vocal Pain Assessment (CNVPA)?

CHAPTER II

LITERATURE REVIEW

A historical cornerstone of behavior analysis has been the assessment and treatment of problem behavior in individuals with a variety of developmental presentations. As a result of this pursuit, researchers have developed well-established procedures, known as functional analyses, in efforts to most effectively and efficiently determine why an individual may be engaging in a problem behavior and inform meaningful intervention. Since their inception, functional analyses have taken on many forms, have been used with a variety of populations, and have been used to treat a host of different problem behaviors. However, research in the areas of functional analyses continues to grow and evolve with the ever-changing needs of individuals with behavioral difficulties. Researchers need to remain diligent in their exploration of FA procedures with individuals with unique presentations of disabilities and behavioral concerns. One such example would be individuals with CHARGE syndrome, a genetic condition resulting in multiple anomalies and the leading cause of congenital deafblindness (U.S. Department of Education, National Center on Deaf-Blindness, 2017) To date, the literature is absent of behavioral analytic approaches to managing behavior excesses or increasing skills for individuals with CHARGE, including the use of functional analyses. Within the literature review, problem behavior will be discussed in terms of topographies, populations, and potential consequences. Second, methodologies, types, populations, and behavioral topographies that can be used for functional

analyses will be discussed. Lastly, individuals with CHARGE syndrome, characteristics, and research on behavior within this population will be addressed.

Problem Behavior

Problem behaviors can present in a variety of common topographies (e.g., self-injurious behavior, aggression, disruptions, vocalization, property destruction) and more unique, such as mouthing objects, rumination, inappropriate sexual behavior, and expelling/packing food (Beavers et al., 2013). Engaging in these behaviors could be disruptive to the individual themselves, others, or both. Problem behaviors can present in typically developing individuals, as well as those with varying disabilities and presentations. Examples of disabilities or disorders that are commonly associated with the presentation of problem behaviors include ADHD (e.g., Sibley, Altszuler, Morrow, & Merrill, 2014), autism spectrum disorder (ASD; e.g., Dominick et al.,, 2007), intellectual disabilities (e.g., Delgado-Casas et al., 2014), and typically developing individuals (e.g., LeGray, Dufrene, Sterling-Turner, Olmi, & Bellone, 2010).

If an individual is consistently engaging in problem behaviors, there is the possibility for several negative outcomes across multiple areas of functioning, including academic and social functioning. At school, negative outcomes may include decreased academic achievement (Algozzine et al., 2011) and reduction of engagement in instructional time (Alter, Walker, & Landers, 2013). Negative social outcomes associated with problem behaviors may include (a) negative relationships with adults (i.e., teacher frustration; Alter et al., 2013), (b) peers (i.e., isolation, bullying,; Teerlink, Caldarella, Anderson, Richardson, & Guzman, 2017), (c) distress of the individual and those who interact with them (Dominick et al., 2007), and (d) difficulty maintaining social relationships (Herzinger & Campbell, 2007). Other negative outcomes may

include tissue damage caused by physical harm to the self or others or decreased ability to learn adaptive and appropriate behavior (Herzinger & Campbell, 2007).

The field of ABA is rooted in behavioral theory, as developed by E.L. Thorndike, John B. Watson, and B.F. Skinner (Keller & Schoenfeld, 1950). Key developments in behavioral theory, such as The Law of Effect by E.L. Thorndike and operant conditioning originally described by B.F. Skinner in 1930, examined the relationship between specific consequences and behaviors and how those consequences strengthened or weakened a behavior. Further, Watson focused on observable events and objective data collection. With these findings, the field of ABA determined that the gold standard would require the use of the following: applied, behavioral, analytic, technological, conceptually systematic, and effective procedures that produce generalized outcomes (Baer, Wolf, & Risley, 1968). Therefore, while there is a possibility for negative outcomes due to engagement in problem behaviors, these outcomes can be avoided or remediated by identifying the function (or reason for engaging in a specific behavior) of a behavior and designing and implementing an intervention that maps onto the identified function (Hanley, Iwata, & McCord, 2003).

Prior to the introduction of ABA, extinction and punishment procedures were exclusively used to manage problem behaviors (Axelrod, 1987). However, when researchers began to hypothesize functions of behavior prior to implementing intervention, a shift in procedures used to decrease problem behaviors occurred. Carr (1977) originally posited specific functions of self-injurious behavior (SIB), which inspired additional studies to explore and identify functions maintaining other problem behaviors. Researchers identified the following functions: (a) attention (Carr & McDowell, 1980); (b) escape (Carr & Newsom, 1985); (c) sensory (Favell, McGimpsey, & Schell, 1982); and (d) tangible items (Derby et al., 1992). Following the

identification of common function categories, Iwata, Dorsey, Slifer, Bauman, and Richman (1982/1994) designed a methodology for pinpointing contingencies controlling an individual's problem behavior (Carr, 1994), which became known as functional analysis (FA).

Functional Analyses (FA)

The idea that problem behaviors serve certain behavioral functions resulted in the development of the first universally accepted FA methodology by Iwata et al. (1982/1994). Iwata and colleagues' (1982/1994) novel conceptualization has resulted in a shift in the understanding of and intervention development for problem behaviors (Dunlap & Fox, 2011). Prior to the introduction of FA procedures, practitioners relied on contingency management, or providing positive reinforcement for appropriate behavior and ignoring or punishment of unwanted behaviors, to manage problem behaviors. As previously stated, with the development of the field of ABA, the approach to addressing problem behaviors shifted from punishment and positive reinforcement only to considering both positive and negative sides of reinforcement and punishment. FA procedures and applications easily meet these guidelines, which is why FA has become a critical and commonly used instrument in intervention design (Beavers et al., 2013).

In broad terms, an FA is a procedure rooted in ABA that involves the manipulation of the environment and the way the interventionist responds to the target problem behaviors to identify specific environmental variables maintaining the behavior. This is also known as the function of the behavior. After identifying the function, an intervention that allows for the same function to be served for engagement in appropriate behavior is developed. For example, if an individual is currently receiving social attention following a loud inappropriate vocalization, an intervention could be developed in which there is an absence of social attention for emitting an inappropriate vocalization, but the individual receives social attention for having a quiet voice.

Since Iwata et al. (1982/1994) introduced what has become known as the 'traditional FA', the topography and procedures have evolved to include brief, trial based, and synthesized functional analyses, all of which have been applied to a variety of populations and problem behaviors.

Types of Functional Analyses

The traditional FA methodology, originally employed by Iwata and colleagues (1982/1994) for individuals engaging in self-injurious behavior, is the most commonly used, researched, and referenced type of FA (Beavers et al., 2013). The traditional FA includes the following four 15-minute conditions: social disapproval, academic demand, unstructured play, and alone. Prior to the analysis of behavior in any of the conditions, researchers first operationally defined target problem behaviors and trained staff on procedures and observations (recording occurrence or non-occurrence of a behavior using continuous 10 second intervals). During the social disapproval condition, toys were present in the room while the interventionist read or engaged in some other kind of task. Social attention in the form of concern/disapproval and brief physical contact was given to the child only when they engaged in one of the target problem behaviors – all other behaviors were ignored. This condition investigated whether or not the behavior was maintained by positive reinforcement via social attention. During the academic demand condition, cognitively appropriate academic demands were presented to the individual. In the absence of problem behavior, if the child did not respond to the demand within 5 s, the interventionist implemented a prompting hierarchy and social praise to ensure that the demand was completed. However, if the client engaged in a problem behavior, the demand and any social attention associated with the demand was removed for 30 s. The academic demand condition examined if problem behaviors were being maintained by negative reinforcement. The

unstructured play condition, which served as a control for the presence of others in the treatment room, required the presence of toys and interventionists, allowing the client to move about the room. Interventionists delivered social praise contingent upon absence of problem behavior, and ignored target problem behaviors. The alone condition required the client to be in the treatment room without access to toys or other individuals. If results of this condition included elevated problem behavior, it could serve as evidence that the problem behavior is being maintained by automatic reinforcement. Results ultimately indicated that each client's problem behaviors were repeatedly associated with one of the conditions, meaning that a specific social or physical manipulation resulted in the engagement of problem behavior.

Efforts to replicate the findings of Iwata and colleagues (1982/1994) were extended across settings, behaviors, ages, and diagnoses (Iwata et al., 1994; Mace, Lalli, & Lalli, 1991). Trahan, Donaldson, McNabney, and Kahng (2014) applied traditional FA procedures to individuals from infancy to mid- to late-adulthood (50+ years old) presenting with varying diagnoses including intellectual disability, genetic or medical conditions, and dementia, while others chose to assess individuals with developmental delay (Lambert, Bloom, Clay, Kunnavatana, & Collins, 2014). Further, behaviors other than SIB, such as bizarre speech (Trahan et al., 2014), inappropriate sexual behavior (Fyffe, Kahng, Fittro, & Russell, 2004), and nail biting (Woods et al., 2001) were targeted through the use of traditional FA procedures. Of these studies, all results identified a specific function and led to the design of an effective intervention (Fyffe et al., 2004; Woods et al., 2001).

A meta-analysis of FAs completed by Beavers and colleagues (2013) reported that traditional FA procedures were used most frequently due to the opportunity to present all conditions multiple times, allowing the researcher to identify patterns of behavior and confirm

hypotheses. However, while there are many useful and appropriate applications of traditional FA procedures, researchers began voicing concerns regarding necessity of extended analyses, exposure to conditions multiple times, and justification for a delay in treatment when faster procedures could be implemented (Hanley et al. 2003).

After years demonstrating the effectiveness of traditional FA procedures for function identification of problem behavior, Northup and colleagues (1991) set out to improve the efficiency of functional analysis procedures while still maintaining the accuracy of function identification by designing what is now known as the BFA. Participants of this study differ from those of Iwata and colleagues (1982/1994) in that their primary problem behavior was aggression. While the BFA maintains conditions from the traditional FA including social attention, escape from a demand, and alone, a condition specifically targeting the receipt of a tangible item was added in place of the unstructured play condition. Also, all conditions lasted between 5 and 10 min, rather than 15 min. Further, there were subtle differences in the conditions carried over from the traditional FA methodology. During Northup and colleagues' (1991) escape condition, the demands consisted of tasks other than academic demands (e.g., folding towels). A difference in the alone condition implemented by Northup and colleagues was that toys were available to the client and the condition served as a baseline measure. Lastly, the tangible condition consisted of the interventionist being present in the room and delivering a desired tangible item following engagement in a problem behavior while ignoring other responses or behavior. Another unique difference between the BFA and the traditional FA methodology is the presence of a contingency reversal in the BFA, which is used to confirm the hypothesized function of behavior. The contingency reversal is applied to the condition with the highest frequency of behavior and requires the specific contingency in place to be delivered (e.g.,

social attention, receipt of a tangible item) in the presence of a manding response that was taught to the client (e.g., saying "please"), rather than engagement in a problem behavior. Following the contingency reversal, the highest frequency condition was repeated as it was during the first implementation. For example, if the hypothesized function was a tangible item, the tangible would be presented to the child if they said "please", but removed if they engaged in a problem behavior. Ultimately, results of Northup and colleagues' (1991) were similar to results of Iwata and colleagues (1982/1991) in that all participants had one condition that resulted in more frequent aggressive behavior, indicating that a specific manipulation of the social or physical environment was the function of the behavior.

While Northup and colleagues (1991) used a mand training procedure for their contingency analysis, subsequent studies have implemented alternate contingency analysis procedures. For example, LeGray et al. (2010) used differential reinforcement of other behavior (DRO) or differential reinforcement of alternative behavior (DRA) procedures in their contingency analysis. The same order of conditions (contingency reversal, normal condition, contingency reversal) were implemented as in Northup and colleagues (1991); however, rather than delivering the desired reinforcer (social attention, escape, etc.) for a manding response, the desired reinforcer was delivered based upon the engagement in an 'other' behavior or absence of target problem behavior. For example, if the hypothesized function of the behavior was social attention, then social attention would be provided to the client upon the absence of problem behavior or engagement in prosocial behaviors.

In order to validate BFA procedures, researchers sought to replicate (Derby et al., 1992) and compare (Kahng & Iwata, 1999; Tincani, Castrogiavanni, & Axelrod, 1999) results of traditional FAs with BFAs. While several studies lent evidence towards findings between the two

procedures being inconsistent (Derby et al., 1992; Kahng & Iwata, 1999) and concern regarding increased false positives (Kahng & Iwata, 1999), other studies found that BFA procedures resulted in shorter assessment times and ultimately identified the same function as the traditional FA methodology (Wacker, Berg, Harding, & Cooper-Brown, 2004; Tincani et al., 1999). However, steps have been taken by researchers to address proposed shortcomings of the BFA procedures. For example, the varying options of contingency reversals including a DRO (LeGray et al., 2010) and a mini-reversal (Cooper et al., 1992) were implemented to determine if the identified function was correct.

In terms of the settings, populations, and behavioral concerns for which BFAs have been used, the list is fairly comprehensive. Settings have included the classroom (Boyajian, DuPaul, Handler, Eckert, & McGoey, 2001; Northup et al., 1991); home (Watson & Sterling, 1998; O'Reilly, Lancioni, King, Lally, & Dhomhnaill, 2000); vocational (Wallace & Knights, 2003); and university clinical settings (MacDonald, Wilder, & Dempsey, 2002; Northup et al., 1991). Examples of populations and problem behaviors BFAs have been applied to include typically developing individuals (Gardner, Spencer, Boelter, DuBard, & Jennett, 2012); ASD (Lyons et al., 2007); intellectual disability (Tincani et al., 1999); eye poking (MacDonald et al., 2002); SIB (Kahng & Iwata, 1999); post-meal rumination (Lyons et al., 2007); noncompliance (Gardener et al., 2012); and disruptive behavior (LeGray et al., 2010).

In summary, while BFAs have been used sparingly in the research and traditional FA procedures have been preferred (Beavers et al., 2013), many studies have used 5-10 minute sessions in their analyses. This indicates that a characteristic of both methodologies – repeated exposure to conditions in the traditional FA and efficiency of condition times in the BFA – is valued by researchers. Overall, research has aimed to increase the effectiveness of BFA

procedures (Cooper et al., 1992; LeGray et al., 2010), apply them to a variety of settings (Boyajian et al., 2001), individuals (Lyons et al., 2007), and problem behaviors (MacDonald et al., 2002; Kahng & Iwata, 1999), and use results to develop ultimately successful interventions (LeGray et al., 2010).

In further efforts to streamline FA procedures, Sigafoos and Saggers (1995) introduced a practical application of FA in the classroom referred to as the trial-based FA (TBFA). Participants included two males (10 and 12 years old) diagnosed with ASD and severe intellectual disability who presented with aggression in the form of hitting, pushing, biting, scratching, pinching, hair pulling, and spitting at/throwing objects at other individuals. Conditions of the TBFA included attention, tangible, task, and control trials and were completed a total of four times each across five days, resulting in a total of 20 discrete trials.

Each trial consisted of two parts, the test and control condition, which could last a maximum of 2 min. For example, during the first 60 s of the trial for attention the teacher would inform the student, “I’ll be right with you.” The teacher then turned away from the student and began completing another task for a maximum of 60 s. A partial interval recording method was used during this trial in that if the student engaged in aggression at all during the first 60 s, the teacher turned her attention to the student and spoke to them. Once the 60 s has lapsed or the student engaged in aggression, the second part of the trial began. During the second 60 s, the teacher provided the student with quality attention regardless of the presence or absence of aggression. Data were recorded as the presence or absence of aggression. Similarly, during the tangible trial, the teacher placed a desired snack or drink within view of the child and said, “You can have this in a minute,” beginning the first part of the trial. If the child engaged in aggression, they were provided with enough of the preferred tangible to occupy them for the full 60 s trial

following aggression. Lastly, during the task trial the teacher delivered pre-academic demands to the child by prompting them (verbally and physically) to engage in the activity every 10 s, if not already doing so. If aggression occurred during the first 60 s, the demand was removed for the next 60 s.

Several pros and cons to the TBFA have been identified through subsequent research. One positive of the TBFA includes efficiency – specifically, when traditional FAs and TBFAs have been compared, results have indicated that the TBFA takes 84.8% less time than traditional methods (LaRue et al., 2010). Further, the traditional FA and the TBFA indicated the same function of behavior in 80% of participants and did not require repeated reinforcement of problem behavior. Other positives of the TBFA include the ability to train other professionals in its implementation, and the various settings it can be conducted in outside of clinical settings (Larkin, Hawkins, & Collins, 2016). Further, several meta-analyses have found that the TBFA literature has made efforts to generalize TBFA procedures across professionals, using results to inform intervention design and implementation, varying session length/number of trials in an analysis, and setting (Rispoli, Ninci, Neely, & Zaini, 2014; Ruiz & Kubina, 2017). Alterations to the procedure have also been made by adding teacher interviews, pre-analysis observations, and contingency reversals (Lloyd et al., 2015). While it is clear that the TBFA can serve as a viable option for professionals practicing both outside of and in the clinical setting due to its flexibility, some of these positives have the potential to lead to undesirable outcomes, such as an increase in false positive or negative results and possible lack of procedural integrity as TBFAs are typically conducted by professionals outside of the field of ABA (Rispoli et al., 2014).

Hanley and colleagues (2014) further altered original FA methodology by designing the IISCA, which allows for the combination of conditions, as well as shorter condition times.

Participants in the study had a diagnosis of a pervasive developmental disorder or ASD and ranged from 3-11 years old. Problem behaviors of participants were varied and included noncompliance, difficulty with tolerance delay, and tantrums. The IISCA method begins with an open-ended functional assessment interview and an observation of the child to form hypotheses regarding functions of problem behaviors. Based upon results of the interview and observation, a functional analysis is designed to identify the function of problem behaviors. Each IISCA includes a test condition and a control condition. It is important to note that while the same conditions from previous FA methodologies (i.e., toy play/control, tangible, social attention, and escape), conditions can be combined or examined in isolation to address all possible functions in the IISCA methodology. Another aspect of the IISCA is the implementation of a treatment plan following the conclusion of the conditions. The treatment includes the following components: teaching functional communication responses as a replacement for problem behavior, increasing the complexity of functional communication responses, implementing delay and denial training, behavioral chaining of simple and complex responses, and generalizing treatment to everyday settings.

Since the introduction of the IISCA, various studies have aimed to extend the procedures to an increased number of individuals, as well as compare results to other FA procedures. Jessel, Hanley, and Ghaemmaghami (2016) compiled results from 30 replications of the IISCA with participants of varying age groups, diagnoses, settings, implementers, and behavioral topographies. Findings indicated increased engagement in problem behaviors during the synthesized condition and zero to low rates of problem behavior during the control condition across the majority of replications. However, several concerns have been raised regarding IISCA procedures, such as increasing the likelihood of responding during synthesized conditions and

requiring interventionists to develop complex interventions to target equally complex functions (Fisher, Greer, Romani, Zangrillo, & Owen, 2016).

Due to the differences between the IISCA and other FA methodologies and concerns presented with the procedures, researchers have aimed to investigate the ability of the IISCA to yield results similar to the traditional FA. Findings of these comparison studies appear to be inconsistent. For example, Fisher and colleagues (2016) identified the functions of participants' behavior to be explained by isolated functions (e.g., only social attention or only escape). Further, low levels of agreement were observed between the IISCA and traditional FAs. However, Slaton, Hanley, and Raftery (2017) found the majority of behaviors assessed in their study were maintained by synthesized functions, and therefore did not align with the findings of the traditional FA, which only examines isolated functions.

Research in functional analysis has continued to progress and move forward with attempts to improve the efficiency and effectiveness of FA methodology. Aspects of the FA that have been targeted to improve the efficiency and effectiveness of the FA include shortened condition times (Hanley et al., 2014; Northup et al., 1991) and combined conditions (Hanley et al., 2014). The variety of FA methodologies is useful in terms of identifying the best methodology to use with a particular client. For example, a methodology with shorter condition times may be appropriate for problem behaviors that could be potentially dangerous (i.e., self-injurious behavior).

Functional analyses in specific populations and behavioral topographies

Outside of changes in general methodology, other ways functional analysis research has continued to develop is in the application across low incidence populations and behavioral topographies. One low incidence population FAs have been applied to is severe intellectual

disabilities. Graff, Lineman, Libby, and Ahearn (1999) examined the utility of a traditional FA in a 6-year old child with ASD and severe intellectual disability and found inconclusive results. However, Delgado-Casas and colleagues (2014) completed an FA for four individuals with severe intellectual disabilities and found that FA methodology was successful in identifying the function of each participant's problem behaviors.

For individuals with hearing and/or vision loss, another low incidence concern, experimental analyses have been conducted, but not true FA methodologies. For example, one study used a descriptive analysis in a pre-school classroom with a young child with severe hearing and vision loss (Harding et al., 1999). Procedures for this analysis included three phases: descriptive analysis-structural (Phase 1), descriptive analysis-functional (Phase 2), and brief experimental analysis (Phase 3). During Phase 1, researchers observed the participant in their typical classroom environment and graphed their behavior across one to two weeks. During Phase 2, researchers collected data on the behavior of adults in the classroom and plotted child data along with adult data to determine the presence of antecedents and consequences of problem behavior. Lastly, during Phase 3 an analogue condition was chosen based upon hypothesized functions (isolated or synthesized functions could be tested). Results indicated differentiation between analogue conditions between the participant's appropriate and problem behaviors.

Magee and Ellis (2000) examined the effects of extinction on the object mouthing, object destruction, and aggression of a child with profound hearing loss and a moderate intellectual disability. Participants were exposed to conditions similar to those described by Iwata and colleagues (1982/1994) including alone, attention, play, and demand. During the alone condition, the participant was alone in the treatment room. During the attention condition, the target behavior resulted in attention, while all other behaviors were ignored. Researchers used both

spoken language and sign language to provide the participant with attention. During the play condition, the researcher interacted with the participant, but ceased following engagement in a target behavior. Lastly, during the demand condition, a demand was given every 10 s and removed (along with the presence of the researcher) following a target behavior. While these procedures are mostly consistent with those of a traditional FA, it is important to note that researchers only tracked the occurrence of one target behavior throughout the analysis and made the assumption that all other target behaviors were maintained by the same function. The target behavior that occurred most frequently was put on extinction. Results indicated a decrease in two of the three target behaviors once extinction was applied. While both of these studies were ultimately trying to decrease problem behaviors using a manipulation of the environment, it is important to note that tried and true FA methodologies were not employed in either study for individuals with hearing or vision concerns.

Comprehensive overview of the use of functional analysis

Recently a comprehensive meta-analysis summarized populations FAs have been completed with, behavioral topographies they were used for, and the type of FA procedures used (Beavers et al., 2013). The majority of FAs were conducted with children in inpatient hospital settings, followed by schools and outpatient clinics. More specifically, FAs were conducted primarily with individuals diagnosed with developmental disability (81.6%), ASD (37.3%), and typically developing individuals (21.5%). In terms of frequent behavioral topographies assessed by FAs, aggression, vocalizations, self-injury, property destruction, and disruption were among the most frequent. However, approximately one fourth of the studies included in the meta-analysis examined unique presentations of behavior, including licking, mouthing, or sniffing objects, problems associated with feeding (i.e., rumination, vomiting, gagging, expelling/packing

food), hyperventilating, disrobing, and inappropriate sexual behavior. Lastly, the most common FA procedure used was the traditional FA (86.1%), followed by the brief FA (12.7%) with 10 min sessions being used most often (41.8%), 5 min sessions (37.3%), and 15 min sessions (7%) across all procedures used.

Across results of the meta-analysis by Beavers and colleagues (2013), there are several findings that should be highlighted. First, while the majority of studies were conducted using traditional FAs, the most common session lengths were between 5 and 10 min. This could indicate that while researchers value the repeated exposure to the same condition, the efficiency of the method chosen is also of importance. Further, it is important to emphasize the overall lack of exploration into the use of FA methodologies with individuals with low incidence disabilities. CHARGE syndrome, a low incidence genetic disorder that is the leading cause of congenital deafblindness (U.S. Department of Education, National Center on Deaf-Blindness, 2017), is a population that could benefit from a better understanding of the effectiveness of functional analyses for individuals with sensory impairments.

CHARGE Syndrome

CHARGE syndrome, caused by a mutation of the CHD7 gene, occurs in approximately 1 in 10,000 births (Hartshorne et al., 2009) and is characterized by the following features used in the acronym: coloboma of the eye, heart defects, atresia choanae, delayed (retardation) growth and development, genital hypoplasia, and ear anomalies/deafness (Blake et al., 1998). CHARGE is currently diagnosed using Blake and colleagues (1998) criteria, which includes four major criteria (coloboma, choanal atresia, characteristic ear abnormalities, and cranial nerve dysfunction) and seven minor criteria (genital hypoplasia, developmental delay, cardiovascular malformations, growth deficiencies, orofacial cleft, tracheoesophageal-fistula, and characteristic

face). In order to receive a diagnosis of CHARGE syndrome, an individual must present with four major criteria or three major and three minor criteria. Further, individuals with CHARGE present with a spectrum of intellectual functioning ranging from normal intelligence to profound intellectual disability and a range of adaptive behaviors (Salem-Hartshorne & Jacob, 2005). This syndrome presently represents the largest population of individuals born as deafblind (U.S. Department of Education, National Center on Deaf-Blindness, 2017).

Research has been conducted to identify a behavioral phenotype for individuals with CHARGE and includes the following characteristics: low normal cognitive functioning, very goal directed and persistent with a sense of humor, socially interested but immature, repetitive behaviors that increase under stress, high levels of sensation seeking, difficulty with self-regulation when under conditions of stress and sensory overload, and difficulty shifting attention or transitioning to new activities (Hartshorne, 2011). Identified patterns have included behaviors typically associated with other disorders (i.e., ASD, ADHD, OCD, tic disorder, deafblindness), the ability to adjust to the environment and adapt to their own disabilities, a connection between behavior and concerns with self-regulation, and behavior used as communication (Hartshorne, Hefner, & Davenport, 2005). Researchers have attempted to confirm several of these behavioral observations. Graham, Rosner, Dykens, and Visootsak (2005) examined behavioral similarities and differences of individuals with CHARGE, Down Syndrome, Prader-Willi Syndrome, and Williams Syndrome. Results indicated that while individuals with CHARGE syndrome engaged in behaviors that were similar to those engaged in by individuals with ASD (e.g., socially withdrawn, need for structure/organization), ultimately, their engagement in these behaviors was hypothesized to be related to their visual and hearing impairments (Graham et al., 2005; Hartshorne et al., 2017; Smith, Nichols, Issekutz, & Blake, 2005). Further, language used by

individuals with CHARGE was not observed to contain features of language engaged in by individuals with ASD such as scripting or echolalia. Individuals with CHARGE have also been found to have difficulty sleeping due to concerns with their airways and initiating sleep (Hartshorne et al., 2009).

After considering this overview of behavioral characteristics of individuals with CHARGE syndrome, it is critical to delve deeper and gain a more comprehensive understanding of problem behavior in CHARGE. Problem behavior in CHARGE can present across a variety of topographies including physical (e.g., hair pulling, biting, pinching, self-injury), verbal (repetitive speech, yelling, complaining), or non-verbal (agitation, withdrawal, pacing, and invading personal space) behaviors (Hartshorne et al., 2017; Stratton & Hartshorne, 2018). Problem behaviors in CHARGE can be influenced by several variables including sensory issues, anxiety, self-regulation strategies (which can serve as a mitigating variable) and pain (Hartshorne et al., 2017; Stratton & Hartshorne, 2018). For example, sensory issues can lead to engagement in self-stimulatory behaviors, such as head banging or teeth grinding, that can lead to further degradation of sensory systems. Anxiety in individuals with CHARGE syndrome can produce both internalizing and externalizing concerns, such as negative thought patterns, anger, aggression, sleeping and eating disturbances, and physical complaints (Eugster, 2007).

While all variables influencing problem behaviors in CHARGE are important to consider, pain is a rather complex variable that can sometimes be difficult to understand or detect. Due to the variety of health variables, individuals with CHARGE syndrome are more likely to experience pain and require significant medical care from a multi-disciplinary team (Nicholas, 2011). Individuals with CHARGE frequently require intensive and invasive medical procedures, often averaging around a dozen medical surgeries (Stratton & Hartshorne, 2018). As a result of

their complexities individuals with CHARGE may experience chronic pain from the following: chronic recurrent otitis media and/or sinusitis, chronic constipation, feeding problems, cranial nerve anomalies, gastroesophageal reflux, muscle, hip and back pain, and sleep problems (Nicholas, 2011; Stratton & Hartshorne, 2018), as well as, falls from poor balance, medical procedures, long-term hospital stays, and recovery periods (Stratton & Hartshorne, 2018). Chronic pain becomes more difficult to manage when accompanied by a developmental disability and communication deficits, as sharing that they are in pain and gaining assistance to relieve pain is more difficult, due to communication limitations (Choi et al., 2017; Symons, Harper, McGrath, Breau, & Bodfish, 2009). Specifically, chronic pain can increase the likelihood of an individual engaging in aggression (Courtemanche et al., 2016), destructive behavior, self-injury (e.g., Courtemanche et al., 2016; Symons & Danov, 2005) and experiencing stress, anxiety, irritability, tension, and nervousness (Nicholas, 2011).

Automatic reinforcement has been described as engaging in a behavior purely because the behavior itself is reinforcing (Vaughn & Michael, 1982). While FAs and methods of identifying automatically reinforced behavior have been completed (Hagopian, Rooker, & Zarcone, 2015; Patel, Carr, Kim, Robles, & Eastridge, 2000; Querim et al., 2013), automatic reinforcement is the least understood function of behavior. Interestingly, while many of the problem behaviors associated with pain are often originally engaged in to receive automatic reinforcement or to communicate the experience of pain, over time, the individual's social environment and reactions of others around them may shape those behaviors. Therefore, because individuals with CHARGE experience chronic pain that could be initiating problem behaviors, it is critical to understand the role of pain and rule out automatic reinforcement before considering more common functions of behavior. Despite the fact that the behavioral presentation of

individuals with CHARGE is at the very least similar to individuals with different disabilities, it is surprising to note that their behavior presentation has yet to be explored through a behavioral analytic lens (e.g., functional perspective).

Current Study

While the use of FA methodologies has been explored in a variety of ways (i.e., ways to make methodologies more effective/efficient, effectiveness for different populations and behaviors), there are still gaps within those variables of FA research. The current study aims to address these gaps in the literature. Of specific interest is the effectiveness and accuracy of BFAs in identifying functions of problem behavior in a population that is both rare and has the potential to present with unique topographies of problem behavior.

In terms of this study, the BFA has been chosen for implementation for a variety of reasons based upon previous literature. While Beavers and colleagues (2013) indicated that the majority of studies in which FAs were conducted used traditional FAs, the condition times were almost always shortened to 5 to 10 min. Harding and colleagues (1999) implemented descriptive and experimental analyses with an individual with severe vision and hearing loss, which required the use of observations prior to making decisions regarding the conditions to be implemented. Further, Magee and Ellis (2000) implemented a modified traditional FA in that data for only one target behavior was monitored and all other behaviors were assumed to be maintained by the same function. What can be gathered from these studies is when assessing individuals with sensory impairments, procedures that reduce assessment time have previously been implemented and have been successful in achieving differentiation between test and control conditions. Further, it is important to note that while both studies including participants with sensory

impairments used variations of FA procedures, neither used the validated procedures as outlined in the literature.

Individuals with CHARGE syndrome will serve as the low incidence population being assessed as individuals with this syndrome are more likely to engage in unique problem behaviors due to multi-sensory impairments. There is currently no published research to date that has applied FA procedures to individuals with CHARGE syndrome. However, there have been several studies that have implemented modified FA procedures or specific parts of FA (i.e., functional interview) for low incidence disabilities that present with similar characteristics as individuals with CHARGE syndrome, such as Prader-Willi syndrome (Hall, Hustyi, Chui, & Hammond, 2014; Hustyi, Hammond, Rezvani, & Hall, 2013) and Down syndrome (Neil & Jones, 2015; Scheithauer, O'Connor, & Toby, 2015). Further, individuals with CHARGE syndrome are more likely to experience chronic pain, which can lead to engagement in problem behaviors that could be maintained by automatic or some other form of reinforcement (Nicholas, 2011). Since automatic reinforcement is less understood (Hagopian et al., 2015), it is important to consider this piece prior to hypothesizing more common functions of behavior. In summary, due to sensory impairments in this population as well as the multitude of medical procedures required, it is critical to determine appropriate methods for identifying functions of problem behavior and designing interventions for these individuals based on the hypothesized functions.

Research Questions

This study seeks to answer the following research questions.

Research Question #1: Can BFA procedures be used to detect functions of problem behaviors in children with CHARGE syndrome/multisensory impairments?

Research Question #2: Can a contingency reversal confirm and further validate the BFA procedures and findings?

Research Question #3: If the frequency of engagement in problem behaviors are undifferentiated across BFA conditions, was pain elevated on the CHARGE Non-Vocal Pain Assessment (CNVPA)?

CHAPTER III

METHODOLOGY

Participants and Setting

All of the following methods were approved by the Institutional Review Board (IRB) at Mississippi State University. Refer to Appendix A for the letter of approval from the IRB. Participants were recruited locally, nationally, and internationally; recruitment took place over the CHARGE Syndrome Foundation and Australasian CHARGE Association's webpage, Facebook pages for families and individuals with CHARGE Syndrome, email listservs, and recruitment flyers. Refer to Appendix B for recruitment materials. A total of 5 participants between 8 to 22 years of age participated in the procedures. This age group was chosen because this age group is representative of the population of children who most frequently participate in functional analyses (Beavers et al., 2013). Further, most children within this age range who have CHARGE have already had the majority of their necessary medical procedures conducted; therefore, minimizing confounding variables when conducting and analyzing data from FA. Further, by including individuals up to 22 years old, the top end of individuals who may still be enrolled in school were included. Exclusionary criteria included (a) individuals with complete sensory loss in hearing and vision; (b) dangerous behaviors (e.g., severe aggression); (c) individuals currently in residential care; and (d) non-English speaking individuals. Participants contacted researchers via email and completed the screening process detailed below prior to being identified as a participant. Parents of participants were compensated with (a) basic

feedback on the results of the BFA; (b) provided with behavioral resources; and (c) potential strategies to mediate their child's problem behavior. Lastly, participants and parents of participants were allowed to stop their participation at any time during the following procedures.

Data were collected at a university-based clinic andinternationally attended conferences. . Across all settings, a controlled environment, free of outside distractions was used (e.g., small room). Functional analyses and data collection took place in this controlled environment. The study included five participants (2 males, 3 females) between the ages of 8 and 22 years old (M = 13.67). Pseudonyms were used throughout the manuscript to protect client identity.

Screening

Demographic questionnaire. Due to the complex medical concerns of individuals with CHARGE syndrome, several screening processes were employed to rule out any potential conflicting variables and ensure the individual was an appropriate participant for the study. Participants responded to a call for recruitment posted on/received through one of the previously mentioned modes of recruitment. Following a response, the primary researcher contacted the potential participant's parent or guardian via phone/email. Parents/guardians were asked to complete a demographic questionnaire targeting questions regarding the following: (a) identifying information for the individual completing the questionnaire and the participant, (b) participant date of birth and gender, (c) age of CHARGE syndrome diagnosis, (d) professional who made the diagnosis, (e) information regarding any genetic testing completed and results, (f) CHARGE characteristics (e.g., coloboma, choanal atresia), (g) when the individual began walking, (h) description of the individual's gait, (i) any behavioral diagnoses (i.e., ASD, ADHD), (j) medications regularly taken, (k) levels of hearing and vision for both right and lefts ears and eyes, (l) sleep problems, and (m) the number of surgeries the individual has undergone. For

specific and comprehensive questions, please see the attached copy of the demographic questionnaire in Appendix C.

Harrison. Harrison was a 14-year-old Caucasian male who presented with the following characteristics of CHARGE Syndrome: choanal atresia, CHARGE middle and inner ear abnormalities, sensorineural hearing loss, vestibular problems, heart defects, cleft lip/palate, spine anomalies, and obsessive-compulsive/perseverative behavior. Harrison presented with moderate visual difficulty in both eyes and is completely deaf in both ears without his cochlear implants. Lastly, Harrison has undergone 20 surgeries.

Harrison's target behavior was noncompliance with wearing the external devices needed for his cochlear implants; however, this behavior was broken down into device removal and noncompliance. Device removal was operationally defined as any time Harrison brought his hand to one or both devices and touched them, began pulling the devices off of his head, or completely removed the devices from his head. Noncompliance was operationally defined as any time Harrison vocally said or signed "no" or "stop" or resisted having the devices placed back on his head (e.g., pushing the interventionist's hands away, blocking his own head).

Lizzy. Lizzy was an 8-year-old Caucasian female with the following characteristics of CHARGE Syndrome: coloboma of the eye, choanal atresia, anosmia, CHARGE outer, middle, and inner ear, vestibular problems, heart defects, and obsessive-compulsive/perseverative behavior. In terms of vision, Lizzy presented with significant difficulty seeing out of her right eye and some trouble seeing out of her left eye. Additionally, Lizzy had retinal detachment surgery in the winter of 2018. Lizzy presented with total deafness in her right ear and normal hearing in her left ear.

Lizzy's target behaviors included noncompliance, aggression towards objects, inappropriate vocalizations, eloping, and perseverative behavior. Noncompliance was operationally defined as failure to initiate compliance or attempts to complete a demand within three seconds or a vocal refusal (e.g., "I don't want to" or "no"). Aggression towards objects included throwing objects and slamming doors. Inappropriate vocalizations were defined as screaming/yelling, grunting, and comments delivered in a sarcastic tone that could be interpreted as disrespectful (e.g., "I'm not your servant"). Eloping included any attempt by Lizzy to leave the room or physically escape a situation (i.e., making moves to open the door, running away from the interventionist) without permission. Lastly, perseverative behavior was defined as asking the same question more than once after it had already been answered.

Simon. Simon was a 19-year-old Caucasian male with the following characteristics of CHARGE Syndrome: coloboma of the eye, anosmia, swallowing problems, facial palsy, CHARGE outer, middle, and inner ear, vestibular problems, heart defects, genital abnormalities, growth deficiency, and typical CHARGE face. Simon presented with moderate visual difficulty in both eyes and some trouble hearing out of both ears.

Simon's target behaviors included physical noncompliance, vocal noncompliance, and inappropriate vocalizations. Physical noncompliance was operationally defined as failing to physically initiate compliance within three s of a demand being issued. Vocal noncompliance included any time Simon verbally refused to engage in a behavior (e.g., "No, I don't want to", "That's not right"). Lastly, inappropriate vocalizations were defined as calling people inappropriate names (e.g., fool, clown, etc.)

Elsa. Elsa was an 11-year-old Caucasian female with the following characteristics of CHARGE Syndrome: coloboma of the eye, anosmia, facial palsy, CHARGE outer, middle, and

inner ear, sensorineural hearing loss, vestibular problems, typical CHARGE face, palm crease, spine anomalies, and obsessive-compulsive/perseverative behavior. Elsa presents with normal vision in both eyes (aside from her coloboma) and has much difficulty with hearing in both ears. Elsa received cochlear implants in the fall of 2016.

Elsa's target behaviors included aggression towards others, irritability, and perseverative behaviors. Aggression towards others was defined as punching, pinching, or hitting another person with an open hand. Irritability included any time Elsa clenched her fists tightly causing her body to shake as well as engaging in inappropriate vocalizations (i.e., "I just want to hurt you"). Lastly, perseverative behaviors were operationally defined as crying, verbal expressions (i.e., "can I call my mom", "I need to see my mom"), and asking the same question more than once. Each time the question or expression was repeated, it was counted as another instance of the behavior.

Hazel. Hazel was a 22-year-old Caucasian female with the following characteristics of CHARGE Syndrome: choanal atresia, facial palsy, CHARGE outer and middle ear, heart defects, kidney abnormalities, growth deficiency, spine anomalies, and obsessive-compulsive/perseverative behavior. Hazel presents with normal vision in both eyes and normal hearing in her left ear. However, Hazel has much difficulty hearing with her right ear.

Hazel's target behaviors included hand biting, inappropriate vocalizations, and hitting objects/items. Hand biting was operationally defined as Hazel bringing one or both hands to her mouth or bringing one or both hands to her mouth and enclosing her mouth around her hand (each occurrence of a hand being brought to her mouth was counted as one behavior). Inappropriate vocalizations included loudly screaming or squealing (each time a breath was taken, that was considered the end of one instance of the behavior). Lastly, hitting objects/items

was operationally defined as using one or both hands/fists to hit items around her (e.g., doors, walls). Table 1 provides an overview of participant information.

Table 1

Overview of Participant Information

Part.	Age	Race	Gender	Hearing	Vision	Target Behaviors
Harrison	14	C	M	Completely Deaf – Both Ears	Moderate Difficulty – Both Eyes	Hearing Aid Noncompliance
Lizzy	8	C	F	Total Deafness – R Ear; Normal Hearing – L Ear	Sig. Difficulty – R Eye; Some Difficulty – L Eye	Noncompliance, Aggression Towards Objects, Inappropriate Vocalizations, Elopement, Perseverative Behavior
Simon	19	C	M	Some Difficulty – Both Ears	Moderate Difficulty – Both Eyes	Physical Noncompliance, Vocal Noncompliance, Inappropriate Vocalizations
Elsa	11	C	F	Much Difficulty – Both Ears	Normal Vision, Except Coloboma – Both Eyes	Aggression Towards Others, Irritability, Perseverative Behavior
Hazel	22	C	F	Much Difficulty – R Ear; Normal Hearing – L Ear	Normal Vision – Both Eyes	Hand Biting, Inappropriate Vocalizations, Hitting Objects/Items

Descriptors for the level of hearing (e.g., completely deaf, much difficulty, etc.) and vision (significant difficulty, moderate difficulty) were sourced from the demographic questionnaire the participant's parents completed. Further, parents were asked to answer the questions based upon their child's level of hearing/vision based on any corrective measures. For example, if the child wears glasses and their vision is corrected using just glasses, they would say 'normal vision'.

Measures

Parents of participants were asked to complete several measures prior to their child's participation in the preference assessments and brief functional analysis procedures. These included CNVPA, the Reinforcer Assessment for Individuals with Severe Disabilities (RAISD), and the functional informed interview.

CHARGE Non-Vocal Pain Assessment (CNVPA)

Many complex medical conditions accompany a diagnosis of CHARGE and often have the potential to result in chronic pain and numerous medical procedures (Stratton & Hartshorne, 2018). Due to changes in overt behavior that can accompany acute or chronic pain, it is critical to rule out pain as an underlying variable acting upon the function of targeted problem behaviors. The CNVPA (Stratton & Hartshorne, 2012) was developed based upon parental input regarding behaviors of children with CHARGE syndrome while experiencing pain and was adapted from the Non-Communicating Children's Pain Checklist-Revised (NCCPC-R; Breau, McGrath, Camfield, & Finley, 2002) and the Pediatric Pain Profile (PPP; Hunt et al., 2007). Overarching domains that could indicate pain included in the CNVPA are vocal, social, facial, activity/challenging behaviors, and body and limbs/physiological factors. Specific observable behaviors that may indicate pain are included under each domain and rated on a 4-point likert scale from 0 (not at all) to 3 (a great deal). When interpreting the CNVPA, it is important to consider the baseline and pain ratings. Specifically, higher scores indicate a higher likelihood that the individual is experiencingpain.

Reinforcer Assessment for Individuals with Severe Disabilities (RAISD)

Due to the sensory impairments of individuals with low levels of hearing and vision, it will be important to consider the likelihood of unique reinforcers. The RAISD is a structured interview designed to compile a list of potential preferred stimuli developed by Fisher, Piazza, and Bowman (1996). The use of a structured interview to inquire about potential reinforcers has proven effective in producing a more potent set of reinforcers than unstructured questioning through the use of prompts and cues that assist in the identification of preferred items. The interview inquires about different types of stimuli including visual, auditory, taste, touch, and specific tangibles. Following the identification of any potential reinforcers in each area, the interviewer asks further questions regarding the stimulus and under what conditions the stimulus is most reinforcing. Reliability for this measure is reported as a measurement of interobserver agreement and resulted in the following average agreement coefficients for differing responses: (a) occurrence - 96%; (b) nonoccurrence – 96.9%; and (c) total 98.3% (Fisher et al., 1996). Measures of validity were not reported by the authors of the RAISD. Approximate time to complete the RAISD is 15-20 minutes.

Functional Informed Interview

Due to the time constraints of the data-collection setting, it was critical to have hypotheses regarding potential functions and a detailed description of problem behaviors prior to beginning the functional analysis. A meta-analysis completed by Beavers et al. (2013) indicates that there has been a recent increase in the number of FAs gathering and reporting data from supplementary assessments. Participants' parents were interviewed using a functionally informed interview developed by Hanley et al. (2014) for use in the IISCA. The interview consists of open-ended questions targeting relevant background information (language abilities, preferred

items, and play skills), description of problem behaviors, behavioral intensity and any potential risks associated with the targeted problem behaviors, patterns of behavior, antecedents, and consequences.

Forced-Choice Preference Assessment

Following the completion and consideration of items endorsed on the RAISD, researchers conducted a formal forced-choice preference assessment, as described by Fisher, Piazza, Bowman, Hagopian, Owens, and Slevin (1992). A total of six items were used and were presented two at a time. The items were presented in combinations so that each item was paired with the five other items once, resulting in a total of 14 pairs presented. Once presented with the two items, the researcher observed which item the participant approached first. The unapproached item was removed and the child was allowed to play with the approached item for five s. If the participant approached both items at once, the items were blocked. If the participant did not approach either stimuli within 5 s of their presentation, the participant was prompted to explore each item for 5 s. Following exploration, the items were again presented to the participant and the same procedure for approached and unapproached items as described above was followed. However, when neither item was approached within 5 s again, both items were removed and the next trial began. A copy of the data sheet used to complete the preference assessment is included in Appendix D.

Brief Functional Analysis (BFA)

The BFA was originally developed to address time and generalizability concerns associated with traditional FA procedures (Northup et al., 1991). Due to time constraints of the data-collection (a week-long conference), BFAs with 10-minute conditions were conducted. Ten

min conditions were the most common condition lengths used in FAs conducted between 2001 and 2012 (Beavers et al., 2013); therefore, 10 min conditions were used. Conditions that were conducted included social attention, demand, tangible, and control (play) conditions, as used by Boyajian and colleagues (2001). The implementation order of conditions was randomized for each participant (see a set of pre-randomized conditions for all six participants in Appendix E). Following the initial implementation of these four conditions, a contingency reversal using DRO was used to confirm the function of the participant's problem behavior (LeGray et al., 2010). Due to the single subject nature of BFA methodology, data obtained by the BFA was graphed as an alternating treatment design. Participants received approximately a 1 min break, or the length of time it took to transition between conditions and set up new materials, between conditions. However, the researcher determined if a longer break was needed due to increased emotional or behavioral responses. Further, there was the possibility that the BFA could be discontinued for the following reasons: (a) engagement in dangerous behavior, (b) heightened emotional distress, (c) illness, (d) medical emergency, or (e) any other reason the researcher or parent deemed necessary. Detailed descriptions of these conditions are provided below in the independent variable section.

Dependent Variables (Problem Behaviors)

Beavers et al. (2013) report that a large portion of the studies included in their meta-analysis included less frequently observed problem behaviors such as rumination (Lyons et al., 2007), expelling or packing of food (Patel, Piazza, Santana, & Vokert, 2002), inappropriate sexual behavior (Fyffe et al., 2004), and licking, mouthing, or smelling objects (Stichter, Sasso, & Jolivette, 2004). Individuals with CHARGE Syndrome may engage in what would be considered rare problem behaviors due to their sensory impairments, such as hearing aid

noncompliance (Markey et al., 2015). However, individuals with CHARGE are just as likely to engage in common problem behaviors such as aggression towards objects or people (hitting, kicking, shoving, punching, etc.); general noncompliance with demands; inappropriate vocalizations; SIB; and disruptions as their peers. Table 2 provides a list of dependent variables for this investigation. The dependent variable is reported as rate of combined target/problem behaviors and is calculated by dividing the total number of problem behaviors that occurred by the length of the condition (i.e., 10 minutes). For example, if the participant engaged in 9 target behaviors during the social attention condition, which lasted 10 minutes, 9 would be divided by 10 for a results of 0.9 problem behaviors per minute.

Table 2

List of Dependent Variables

Dependent Variables
Device Removal
Noncompliance
Physical Noncompliance
Vocal Noncompliance
Aggression Towards Objects
Aggression Towards Others
Inappropriate Vocalizations
Elopement
Perseverative Behavior
Irritability
Hand Biting
Hitting
Objects/Items

Independent Variables (BFA Conditions)

BFA conditions included control (play), social attention, demand, and tangible; each condition was 10 min in length (Boyajian et al., 2001; LeGray et al., 2010). Data collection for

the BFA was collected by a researcher on the corresponding data sheet (See Appendix E for an example of the BFA protocol and data sheet; Note: materials listed on the protocol are examples of items that could be used). Items for each participant were determined based upon the RAISD, functional interview, and forced-choice preference assessment. Each condition was broken down into 1-min intervals in which frequency of behaviors during that minute were recorded. The researcher was required to indicate the condition, date, client, observer, role as primary or rely data collection, and the number of the condition (i.e., first, second, third) on the datasheet. Researchers also had the opportunity to include any specific notes regarding the events during each minute interval. Operational definitions of participant target behaviors were included at the bottom of the datasheet.

Control (Play) condition. The control (play) condition lasted a total of 10 min. Materials present included a chair for the interventionist to sit in and the participant's most preferred items (determined by the results of the RAISD, functional interview, and forced-choice preference assessment), which were available at all times. The interventionist did not prompt the participant to play with preferred items and did not make requests or demands. Target behaviors were ignored while other behaviors received consequences including parallel play, communication, and praise. Specifically, the interventionist acknowledged the participant every 15 s contingent on absence of problem behaviors within the last 5 s. Praise (verbal or social) was provided every 5-10 s (e.g., "I like how you are playing with those toys", "nice job having a quiet voice").

Social attention condition. The social attention condition lasted 10 min, with a 2 min period prior to beginning the condition in which the participant had free access to high quality social attention. Materials present included a magazine/book, chair, and less preferred items. These items were determined based upon the results of the RAISD, functional interview, and

forced-choice preference assessment. During the first 2 min, the interventionist provided the participant with high quality social attention that was free from demands or requests. Less preferred items were also available during this time. At the end of the first 2 min, the interventionist sat in the provided chair and read a magazine/book while less preferred items were still available to the participant. For the remaining 10 min, if a target behavior occurred the interventionist provided brief social attention for each behavior (e.g., "please don't scream, other people can hear you"). All other behaviors were ignored, including appropriate requests or attempts to obtain attention.

Demand condition. The demand condition lasted a total of 10 min. Required materials included a table, two chairs, and any items associated with the demands. These items were identified based upon information gathered from the functional interview regarding the participant's language, cognitive/academic functioning as reported by the parent, and conditions under which the behavior is more likely to occur. For example, reading passages, flashcards, worksheets, etc. were necessary for academic demands. Examples of other materials included blocks or other items for the participant to pick up or give the interventionist upon demand. The participant and the interventionist were seated at a table and a demand was presented every 15-s. If compliance was not initiated, three-step guided compliance (verbal, gestural, and physical prompts) was used with 10 s between each topography of prompt.

When the participant engaged in target behaviors during the demand sequence, the interventionist said, "Okay, you don't have to" and removed demand materials. The participant was allowed a 30 s break from demands and the interventionist turned away without looking at the participant (i.e., providing no attention). If target behaviors persisted throughout the 30-s break, behaviors were ignored while the frequency continued to be tracked. The interventionist

continued to not look directly at the participant. Following the 30 s break, a new demand was issued. Engaging in other behaviors during the demand sequence resulted in verbal praise for complying following the verbal or gestural prompt. If other behaviors occurred during the 30 s break, these behaviors were ignored and the interventionist did not engage with the participant.

Tangible condition. The total time for the tangible condition was 10 min with a 2 min period prior to the session in which the participant was allowed free access to their most preferred item. Materials included a chair and the participant's most preferred item. This item was identified using data obtained from the RAISD, functional interview, and forced-choice preference assessment. During the first 2 min of the condition the participant received access to the preferred item. At the end of the first two min, the interventionist said "my turn" and removed the preferred item from the child. For the remaining 5 min, the interventionist stood or sat in the room while holding the toy. If a target behavior occurred, the interventionist said "okay" and returned the preferred item to the participant for 30 s, while providing no social attention or cooperative/parallel play. All other behaviors were ignored. If the child appropriately requested the toy, requests were ignored, but noted on the data sheet.

Contingency reversal. Following the completion of the preceding four trials, a contingency reversal was completed for the condition that resulted in the highest frequency of problem behaviors. The design of the contingency reversal was B-A-B, where B was the contingency reversal and A was the original condition that resulted in the most problem behaviors. During the contingency reversal a differential reinforcement of other behavior (DRO) was provided for the absence of target behaviors and withheld for engaging in target behaviors. The topography of the reinforcement was dependent upon which condition resulted in the most target behaviors. For example, if the supposed function of aggression was attention, attention

was provided for the absence of aggression while engaging in aggression was ignored. For this example, the A condition consisted of ignoring appropriate behaviors and providing attention for aggression. If the hypothesized function were a tangible item, the tangible item was available for the individual to use in the absence of aggression and was removed for a brief period if the individual engaged in aggression. Lastly, if the supposed function was escape, the individual was no demands were placed on the individual contingent upon the absence of aggression and demands were issued if the individual engaged in aggression.

Interventionist Training and Interobserver Agreement (IOA)

Four interventionists in a school psychology program were trained on the implementation of BFA conditions and observation techniques. Each research assistant was provided with a list of behaviors with operational definitions and watched a video of a child engaging in problem behavior. The research assistants were required to record the frequency of problem behaviors throughout the video and their observations were compared with a key indicating how many of each behavior were present. Research assistants were trained to 90% interobserver agreement (IOA). If a research assistant dropped below 90% IOA, they were retrained. IOA was collected by two researchers collecting data on the same participant concurrently in all conditions implemented during the FA. IOA will be calculated based upon 33.33% of conditions conducted. In order to ensure accurate identification of the function of the problem behavior IOA was calculated for each participant by calculating the frequency of agreements per minute (numerator) and dividing by the number of agreements plus disagreements (denominator). This number was then multiplied by 100. An average percentage of IOA across 33.33% sessions was also included in the results. Interobserver agreement will be reported for each participant in the results, as well as an overall average in the discussion.

Treatment Integrity

In order to train research assistants on treatment integrity, the same video used for training assistants on IOA was used. Research assistants were given a treatment integrity checklist and were required to record all steps of the treatment that were or were not completed during the implementation of the session. Researchers were considered to be adequately trained when treatment integrity reached 90%. Treatment integrity data was collected by a researcher using a treatment integrity datasheet during each condition of the BFA. Each condition was broken down into specific steps in a table and the researcher indicated whether the step was completed or not. The researcher collecting IOA was responsible for checking off each step of each condition during the BFA and prompting the primary researcher to complete steps if they did not. See Appendix E for an example of the treatment integrity sheet. Treatment integrity will be reported for each participant.

Procedure

Measure Completion

Following a response to the call for participants, potential participants were asked to complete the demographic questionnaire. If found to meet all criteria, the parents/guardians of the participant were contacted via phone/email to complete the RAISD.

Brief Functional Analysis

Several procedures were completed in person. Prior to completing BFA conditions, researchers conducted a forced-choice preference assessment with each participant. Following completion of the preference assessment, all necessary conditions of the BFA were conducted. The following data were collected during the BFA: (a) treatment integrity; (b) frequency of

behaviors across all 10 min conditions; (c) interobserver agreement; and (d) a CNVPA was completed during the BFA in order to track the presentation of pain-related behaviors across all conditions. After all original conditions were completed (tangible, attention, escape, toy play/control), contingency reversals were conducted for the condition(s) resulting in the highest frequency of target behaviors.

Data Analysis

Due to the single subject nature of the current study, analyses primarily included visual analysis. Visual analysis is regarded as the primary method for interpreting the effects of an intervention (Baer, 1977; Kratochwill et al., 2010; Wolery, Busick, Reichow, & Barton, 2010) and aids in determining the functional relationship between the intervention and fluctuations in the dependent variable (Parker, Hagan-Burke, & Vannest, 2007). Visual analysis results in low error rates (Rakap, 2015) and allows researchers and interventionists to make data-based decisions (Baer, 1977). Further, since this study is focused on within subject differences/treatment effects, and not between subjects treatment effects, graphs will not have the same scaling on the y-axis. This will aide in more accurate interpretation of individual participant data by making graphs easier to interpret visually. For example, if one participant engaged in a rate of 20 target behaviors per minute, and another engaged in a rate of two target behaviors per minute, graphing those two BFA results on the same scale (1-20) would not be conducive to making future treatment decisions for both participants.

CHAPTER IV

RESULTS

The purpose of the current study was to determine the utility of current BFA procedures to identify functions of problem behaviors in individuals with CHARGE syndrome as evidenced by differentiated engagement in problem behavior across conditions. Further, the study implemented a contingency reversal to determine if the contingency reversal is a good tool to confirm and validate the findings of BFAs. Lastly, participants were screened for engagement in behaviors that could indicate pain in individuals with CHARGE syndrome, as outlined in the CNVPA to determine if undifferentiated BFA results could be attributed to the individual experiencing pain.

The results are discussed in terms of all results for a single participant across (a) results of the BFA; (b) interobserver agreement; (c) treatment integrity; (d) CNVPA scores from days in pain and in-session scores.

Results by Participant

Harrison

Harrison's target behaviors included cochlear implant device removal and noncompliance. For full operational definitions, please refer to the participants and setting subsection in the methodology section. Results of the BFA (see Figure 1) indicated that Harrison's behavior was most differentiated in the social attention condition, as evidenced by a rate of 4.6 behaviors per minute. Following the implementation of the four conditions of the

BFA, a contingency reversal for social attention was completed. As a reminder, the contingency reversal was implemented in a B-A-B design, with B indicating the contingency reversal and A indicating the condition of the BFA that resulted in the highest frequency of combined problem behaviors. In Harrison's case, the contingency reversal (B) involved providing Harrison with high quality social attention contingent upon refraining from touching or removing his devices and engaging in noncompliance (i.e., refrained from saying/signing "no" or "stop"). During the contingency reversal, Harrison's behavior occurred at a rate of zero during the controls (B), and was again elevated during the BFA social attention procedures (A).

Average interobserver agreement across 33.33% of conditions completed for Harrison was 98.6% with 100% treatment integrity. Harrison's mother indicated that on a day when he does not feel well or is in pain, his score on the CNVPA would be 48. However, after the interventionist completed the CNVPA for Harrison's presentation during the BFA, his behavior resulted in a score of 4. When the scores from both CNVPAs are compared, it appears that Harrison was not experiencing pain at the time the BFA was conducted. Harrison's target behaviors during the BFA were differentiated across sessions.

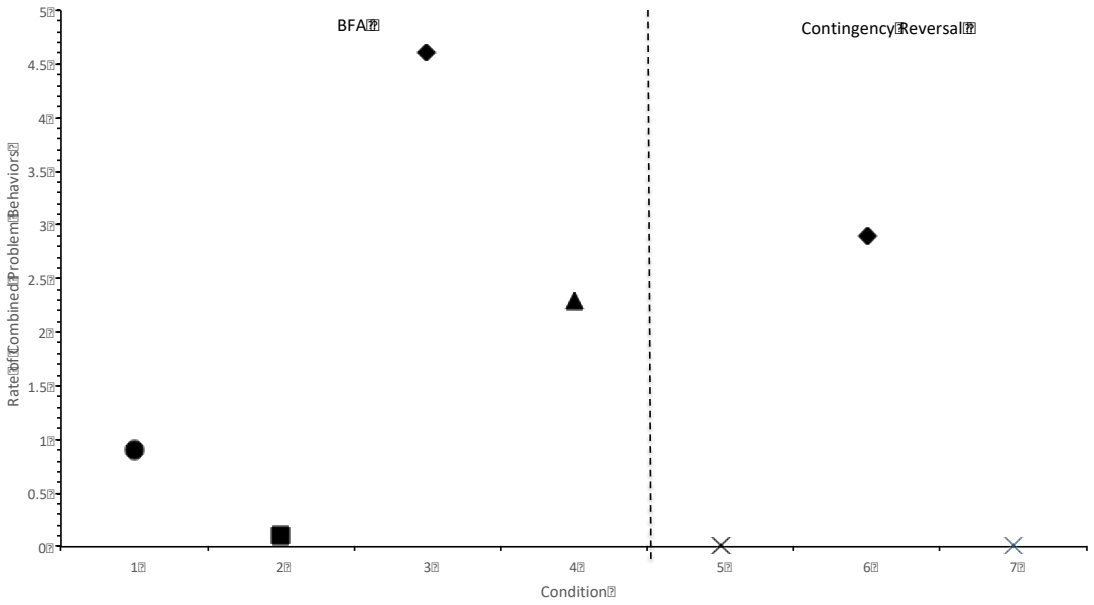

Figure 1. Results of Harrison's BFA

Rate of Harrison's problem behaviors per minute across all four conditions of the BFA and the contingency reversal are presented above. The square represent the control condition, the diamond represents the social attention condition, the circle represents the tangible condition, the triangle represents the demand condition, and the X represents the contingency reversal.

Lizzy

Lizzy's problem behaviors included noncompliance, aggression towards objects, inappropriate vocalizations, eloping, and perseverative behavior. Results of the BFA (see Figure 2) indicated that Lizzy's behavior was most differentiated in the tangible condition, as evidenced by a rate of 1 behavior per minute. In Lizzy's case, the contingency reversal (B) consisted of allowing Lizzy access to her most preferred item contingent upon her refraining from engaging in any of the previously identified problem behaviors (i.e., Lizzy was allowed to play with the tablet unless she engaged in target behaviors). During the contingency reversal, Lizzy's behavior occurred at a rate of zero during the controls (B), and was again elevated during the BFA tangible procedures (A).

Average interobserver agreement across 33.33% of conditions completed for Lizzy was 100% with 100% treatment integrity. Lizzy's parents indicated that on a day when she does not feel well or is in pain, her score on the CNVPA would be 40. However, after the interventionist completed the CNVPA for Lizzy's presentation during the BFA, her behavior resulted in a score of 2. When the scores from both CNVPAs are compared, it appears that Lizzy was not experiencing pain at the time the BFA was conducted. Lizzy's target behaviors during the BFA were differentiated across sessions.

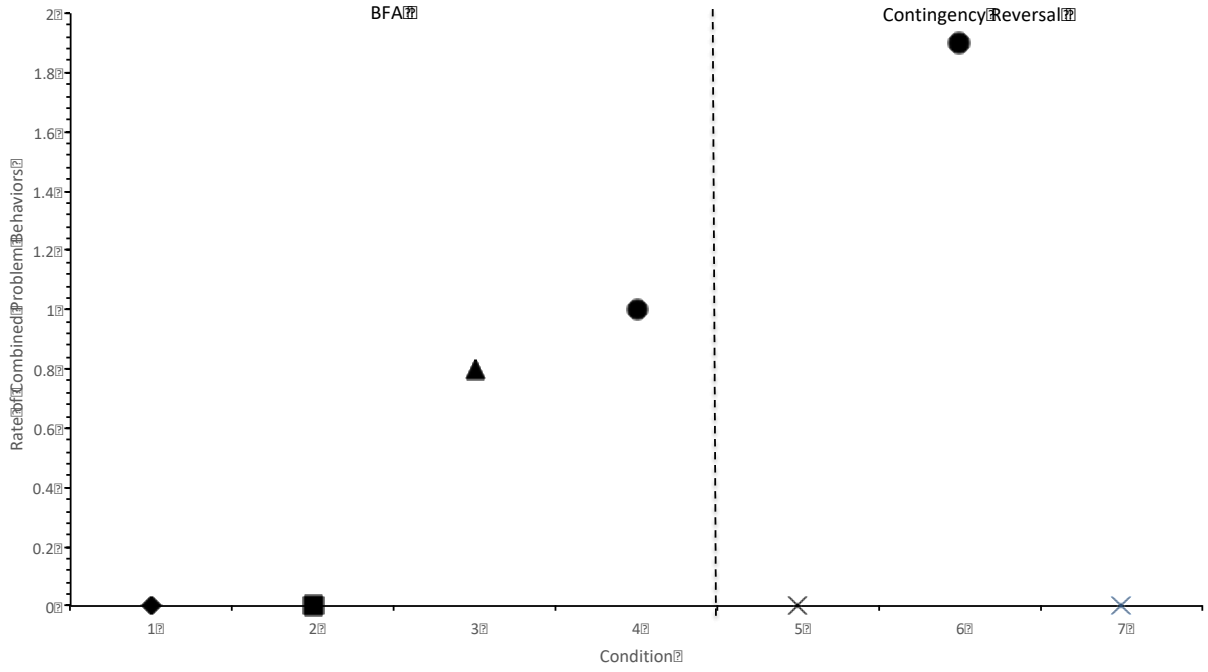

Figure 2. Results of Lizzy's BFA.

Rate of Lizzy's problem behaviors per minute across the conditions of the BFA are presented above. The square represents the control condition, the diamond represents the social attention condition, the circle represents the tangible condition, the triangle is represented by the demand condition, and the X represents the contingency reversal.

Simon

Simon's problem behaviors included physical noncompliance, verbal noncompliance, and inappropriate vocalizations. Conditions of the BFA did not elicit any of the identified target behaviors from Simon, resulting in no differentiation across conditions (see Figure 3). Due to no engagement in problem behavior, a contingency reversal could not be completed. Per the methodology, undifferentiated results mean that the BFA was not successful in identifying the function of problem behavior; therefore, a contingency reversal was not necessary as there was no hypothesized behavior to confirm.

Average interobserver agreement across 33.33% of conditions completed for Simon was 100% with 100% treatment integrity. Simon's mother indicated that on a day when he does not feel well or is in pain, his score on the CNVPA would be 29. However, after the interventionist completed the CNVPA for Simon's presentation during the BFA, his behavior resulted in a score of 5. When the scores from both CNVPAs are compared, it appears that Simon was not experiencing pain at the time the BFA was conducted. Simon's target behaviors during the BFA were undifferentiated across sessions as evidenced by no engagement in problem behavior, which indicate that Simon was likely not experiencing pain during this time. Results of the CNVPA during the session further suggested the absence of pain during the BFA.

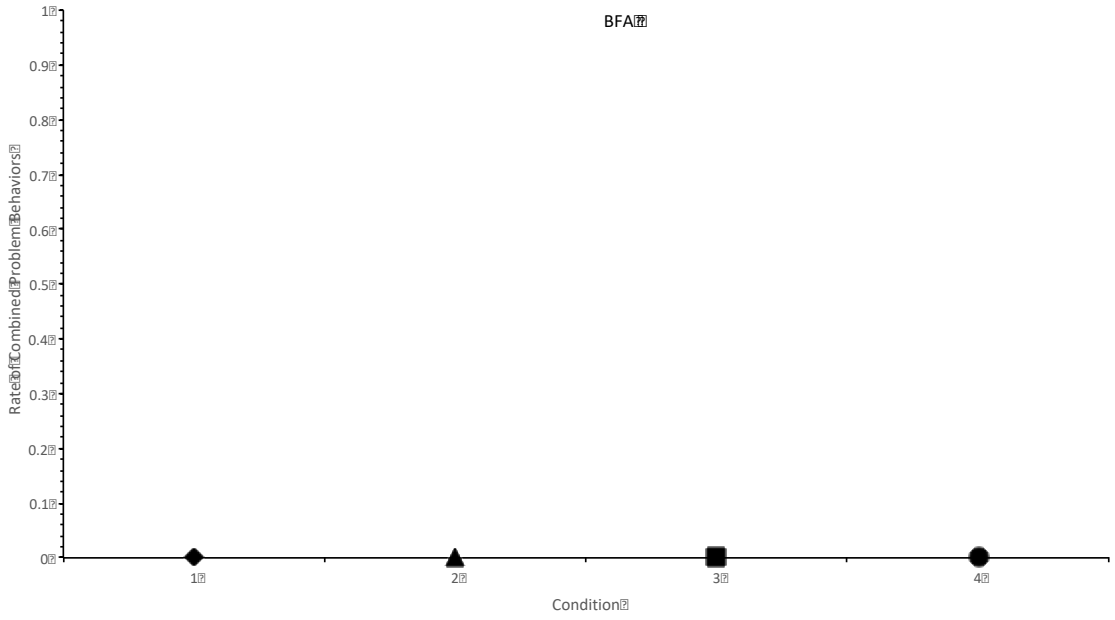

Figure 3. Results of Simon's BFA

Rate of Simon's problem behaviors per minute across the conditions of the BFA are presented above. The square represents the control condition, the diamond represents the social attention condition, the circle represents the tangible condition, the triangle is represented by the demand condition, and the X represents the contingency reversal.

Elsa

Elsa's problem behaviors included aggression towards others, irritability, and perseverative behavior. Results of the BFA (see Figure 4) indicated that Elsa's behavior was most differentiated in the social attention condition, as evidenced by a rate of 2.4 behaviors per minute. In Elsa's case, the contingency reversal (B) consisted of providing Elsa with high quality social attention (i.e., speaking in an animated tone, responding to attempts at conversation, engaging Elsa in conversation, brief physical touch) contingent upon her refraining from engaging in any of the previously identified problem behaviors. During the contingency reversal, Elsa's behavior occurred at rates of less than 0.2 behaviors per minute during the controls (B),

and occurred at a higher rate during the BFA social attention procedures (A; 0.8 behaviors per minute).

Average interobserver agreement across 33.33% conditions completed for Elsa was 93% with 100% treatment integrity. Elsa's mother indicated that on a day when she does not feel well or is in pain, her score on the CNVPA would be 37. However, after the interventionist completed the CNVPA for Elsa's presentation during the BFA, her behavior resulted in a score of 8. When the scores from both CNVPAs are compared, it appears that Elsa was not experiencing pain at the time the BFA was conducted. Elsa's target behaviors during the BFA were differentiated across sessions, therefore the in-session score on the CNVPA further confirms that Elsa's problem behaviors are likely maintained by environmental variables, rather than the experience of pain.

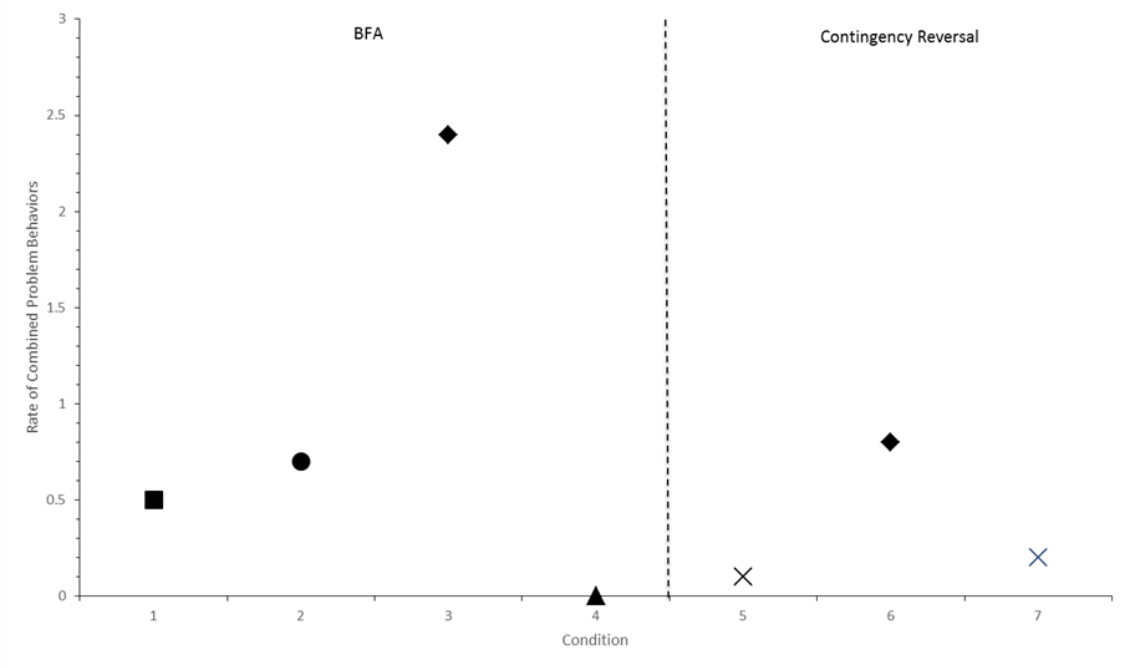

Figure 4. Results of Elsa's BFA.

Rate of Elsa's problem behaviors per minute across the conditions of the BFA are presented above. The square represents the control condition, the diamond represents the social attention condition, the circle represents the tangible condition, the triangle is represented by the demand condition, and the X represents the contingency reversal.

Hazel

Hazel's problem behaviors included hand biting, inappropriate vocalizations, and hitting objects/items. For full operational definitions, please refer to the participants and setting subsection in the methodology section. Results of the BFA (see Figure 5) indicated that Hazel's behavior was most differentiated in the social attention condition, as evidenced by a rate of 7.6 behaviors per minute. In Hazel's case, the contingency reversal (B) consisted of providing Hazel with high quality social attention (i.e., talking to Hazel, brief physical touch, looking at Hazel) contingent upon her refraining from engaging in any of the previously identified problem behaviors. During the contingency reversal, Hazel's results were undifferentiated as evidenced

by target behaviors occurring during both controls (B; 2.5 behaviors per minute and 0.7 behaviors per minute) and the BFA social attention procedures (A; 1.3 behaviors per minute).

Average interobserver agreement across the 33.33% of conditions completed for Hazel was 90% with 100% treatment integrity. Hazel's parents indicated that on a day when she does not feel well or is in pain, her score on the CNVPA would be 35. However, after the interventionist completed the CNVPA for Hazel's presentation during the BFA, her behavior resulted in a score of 16. When the scores from both CNVPAs are compared, it appears that Hazel may have been experiencing pain at the time the BFA was conducted.

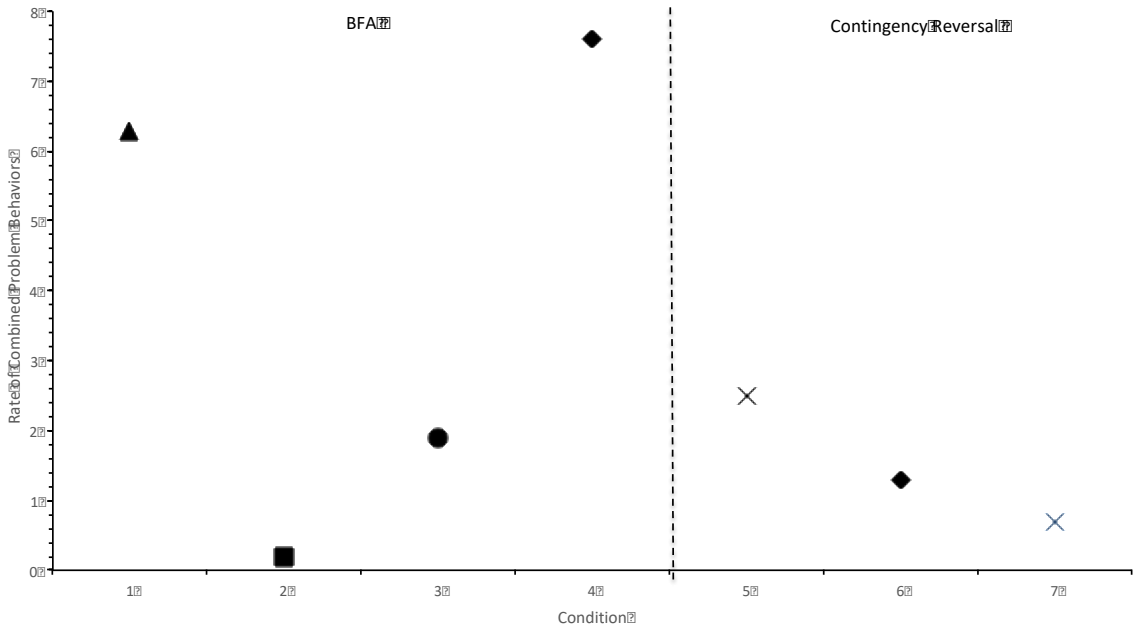

Figure 5. Results of Hazel's BFA.

Rate of Hazel's problem behaviors per minute across the conditions of the BFA are presented above. The square represents the control condition, the diamond represents the social attention condition, the circle represents the tangible condition, the triangle is represented by the demand condition, and the X represents the contingency reversal.

CHAPTER V

DISCUSSION

The use of FA procedures has been examined in various populations, including typically developing individuals (Gardner et al., 2012), individuals diagnosed with ASD (Lyons et al., 2007), and intellectual disability (Tincani et al., 1999). Further, FA procedures have been used to address a wide variety of problem behaviors (eye poking; MacDonald et al., 2002; SIB; Kahng & Iwata, 1999; post-meal rumination; Lyons et al., 2007; noncompliance; Gardener et al., 2012; and disruptive behavior; LeGray et al., 2010). However, there has been limited research into the utility of these procedures with individuals with hearing, vision, and other sensory impairments, such as CHARGE Syndrome. While studies have been completed using experimental analyses for individuals with hearing and/or vision loss/impairment (Harding et al., 1999; Magee & Ellis, 2000), true FA methodologies have not been employed with these populations to this point. Due to the noted behavioral concerns for individuals with CHARGE syndrome (Hartshorne et al., 2017), it is critical to explore the use of FA procedures with the goal of determining functions of problem behavior and using FA results to design and implement meaningful interventions. By understanding the utility of FA procedures in this population, problem behavior in individuals who present with hearing, vision, and other sensory impairments will be able to be addressed and treated in an efficient and effective manner. This will ultimately result in the avoidance or decreased potential negative outcomes of problem behavior.

The current study sought to determine if current BFA methodologies would be successful in identifying functions of problem behaviors in individuals with CHARGE syndrome. Further, for those FAs that resulted in differentiation across conditions, a contingency reversal was implemented to determine if the contingency reversal method can aide in confirming results of FAs. Lastly, due to the number of medical procedures and chronic pain experienced by individuals with CHARGE syndrome (Nicholas, 2011), the potential role of pain in the engagement in problem behavior was examined.

In order to answer these questions, participants and their parents completed several measures (i.e., demographic questionnaire, RAISD, a functional interview, and a forced-choice preference assessment) prior to implementation of the FA procedures in order to determine if the individual was appropriate for the study, preferred tangible items and activities, and detailed descriptions of target behavior. Further, these measures allowed researchers to set up potent conditions for each individual participant. Following completion of these measures, BFA methodologies and a contingency reversal as outlined by LeGray and colleagues (2010) was implemented and a CNVPA was completed based upon the participant's behavior during the BFA conditions. While results have been discussed on an individual participant basis, this chapter aims to discuss results of the study as a whole and how they relate to the research questions posed. Further, implications for future research will be discussed.

Overview of Findings

Identification of Problem Behavior

Due to the single subject nature of FA, visual analysis is the primary mode of interpreting results. Therefore, detection of the function of problem behavior is determined by differentiation of rate across conditions. That is to say, researchers or interventionists hope to see elevated

frequencies of problem behavior in one condition and low or no engagement in problem behavior in remaining conditions. Differentiation across conditions occurred for Harrison, Lizzy, Elsa, and Hazel, while Simon did not engage in problem behavior during any condition of the BFA.

For Harrison, the demand, tangible, and social attention conditions resulted in problem behavior. However, the social attention condition was clearly differentiated as evidenced by the rate of problem behavior between social attention and the other conditions differing by at least 2.3 problem behaviors per minute. Lizzy's FA resulted in problem behaviors occurring at fairly similar rates during the demand and tangible conditions; however, ultimately, the demand condition resulted in the highest rate of problem behavior. Elsa was observed to engage in problem behavior during the control, tangible, and social attention condition. The social attention condition resulted in the highest rate of behavior as evidenced by a difference of at least 1.7 behaviors per minute between the social attention condition and other conditions. Lastly, Hazel's FA resulted in problem behaviors across all four conditions, however, the demand and social attention condition resulted in substantially higher rates of behavior in the demand and social attention conditions. However, the highest rate exhibited by Hazel was elicited during the social attention condition. Overall, these results provide evidence for current BFA procedures having utility to detect the function of problem behaviors in individuals with CHARGE syndrome.

However, for the participant who did not engage in problem behaviors during any condition of the BFA (i.e., Simon), there could be several possible explanations. First and foremost, it is important to note that FA procedures used with other populations are not always successful in identifying a clear function. Reasons that could be cited in past research for a BFA's inability to identify a clear function of behavior have been conditions not being long enough to elicit the behavior (Derby et al., 1992), not repeating conditions (as evidenced by

sparing use of BFAs with only one exposure per condition; Beavers et al., 2013), and differences in FA outcomes depending on the individual who implements the conditions (English & Anderson, 2004; Huete & Kurtz, 2010). Results of studies comparing results of FAs implemented by familiar versus unfamiliar individuals can result in differing functional relationships (English & Anderson, 2004), no engagement in problem behavior, and lower rates of responding (Huete & Kurtz, 2010). Specifically, English and Anderson (2004) posited that caregivers might be discriminative stimuli for the individual's problem behavior. In the case of both participants with no differentiation across conditions, the majority of their problem behavior occurs with parents/caregivers, per parent report. For Simon, his physical noncompliance, vocal noncompliance, and inappropriate vocalizations occur primarily with his mother. While parent implemented FA procedures were outside the scope of this study, it would be beneficial to explore this possibility in future studies to determine if a function could be identified if an individual who could be serving as a discriminative stimulus implemented the conditions.

Lastly, in regards to the identification of problem behavior using FA methodologies in general, it is important to consider the implications of how we design each condition for individual participants. Often times, FA procedures may fail to detect a function of problem behavior due to lack of assessment of under what conditions are we most likely to elicit problem behavior. While each condition should be tailored to increase the likelihood of eliciting the problem behavior, it is often the case that the tangible condition is the only condition for which we take the time to do so – specifically, by completing preference assessments and other measures to ensure the potency of the tangible reinforcers (e.g., LeGray et al., 2010). , Roscoe, Rooker, Pence, Longworth (2009) completed a study in which they assessed for low probability and high probability demands and found that clearer FA results were derived when low

probability demands were issued during the FA. Results of this study lend support for the personalization for all conditions of an FA. For example, one could comlete a preference assessment specifically targeting different kinds of social attention in order to determine which is the most reinforcing. For the participants of the current study, it may have been beneficial to include assessments prior to the BFA that were specific to functions other than the tangible function. For example, we may have succeeded in eliciting problem behavior for Simon if we had tailored demands to lower probability demands (i.e., household chores) or ensured that the social attention provided was the most preferred version of social attention.

Contingency Reversal

In addition to determining if BFA procedures would produce differentiation in the frequency of behaviors across conditions, the current study sought further evidence to support the conclusions of the BFA procedures. Contingency reversals are used following FA conditions to either lend more support for a hypothesized function or indicate to the interventionist/researcher that there may be more than one function maintaining the behavior/other variables at play. In terms of the contingency reversal procedure, this was only completed when a participant's behavior was differentiated across conditions and a conclusion could be made regarding the function of the participant's target behaviors. When a researcher or interventionist is not specifically concerned with the efficiency of the FA procedures, it may be appropriate to repeat conditions to determine if problem behavior can be elicited during one of the conditions before determining that the FA was not conclusive and a contingency reversal is not needed. However, since the primary goal of the current study was to determine if the function of problem behavior for individuals with CHARGE could be identified using brief, rather than extended analyses, repeating conditions would have defeated the purpose of the first research

question. However, as discussed in the previous section, the functions of four of the six participants' problem behavior (i.e., Harrison, Lizzy, Elsa, and Hazel) were able to be identified using the BFA and contingency reversals were completed for those participants.

As a brief review, the contingency reversal was implemented in an A-B-A design with hopes that when "A", or when the participant was receiving the proposed function for engaging in any behavior other than the target behavior, was implemented the frequency of problem behavior would return to lower rates or drop to zero. Conversely, when "B" (repeated FA condition) is implemented, the researcher hopes to see target behaviors increase once again when the target behaviors are the only response being followed by the proposed function (i.e., escape from demands, social attention, or tangible item). Once "A" is repeated, it is hypothesized that if the proposed function is correct, the rate of problem behavior will once again return to lower rates or drop to zero. If differentiation between the "A" and "B" conditions is achieved, the contingency reversal will have been effective in confirming the results of the BFA.

In the cases of Harrison and Lizzy, the contingency reversal was successful in confirming the results of BFA. For both participants, the contingency reversal condition (A) resulted in no problem behaviors while problem behaviors returned to similar, if not higher levels, when the condition from the BFA was repeated (B). When the contingency reversal condition (A) was implemented with Elsa, she was observed to engage in low frequencies of problem behavior (between one and two behaviors). Following the reimplementation of the BFA condition (B) Elsa's problem behaviors were observed to increase again, although not to levels observed during the BFA. Lastly, Hazel's contingency reversal led to ambiguous results as evidenced by the highest frequency of problem behavior occurring during the first contingency reversal condition (A) and proceeding in a decreasing trend across the remaining BFA (B) and

contingency reversal (A) conditions. These results to not lend themselves to clear support of the hypothesized function obtained from Hazel's BFA. While the level of differentiation varied across the four participants that a contingency reversal was implemented for, ultimately, the contingency reversal supported the functions identified during the BFA conditions for three out of four participants.

CNVPA and the Role of Pain

The final research question of the current study addressed a variable that is relevant for all populations, but especially important to assess in individuals with CHARGE – the role of pain. Individuals with CHARGE syndrome experience chronic pain due to chronic constipation, feeding problems, sleep problems, cranial nerve anomalies gastroesophageal reflux, muscle, hip and back pain, and sleep problems (Nicholas, 2011; Stratton & Hartshorne, 2018), as well as, frequent medical procedures, increased likelihood of falls due to poor balance, and extended hospital stays and recovery periods (Stratton & Hartshorne, 2018). Research has indicated that in some cases the experience of pain can result in an increased likelihood of an individual engaging in a variety of problem behaviors (i.e, Courtemanche et al., 2016; Symons & Danov, 2005). Due to the relationship that has been exhibited in research between pain and problem behavior, it is critical to assess pain in order to rule out the possibilities that problem behaviors occur as a means to communicate pain or that pain serves as the underlying antecedent to engage in problem behavior.

Parents of participants were provided with the option to complete the CNVPA (Stratton & Hartshorne, 2012) for their child following the completion of the functional interview. Another CNVPA was completed by the researcher/research assistants based upon their observations of the participant during the BFA conditions. Parents were asked to think of a time

that their child had been in pain or did not feel well and complete the CNVPA based upon that time. The purpose of this was to provide the researcher with an idea of what the participant's score on the CNVPA would be comparable to if they were experiencing pain. Scores from the "pain day" and the BFA session CNVPAs were compared to aid in the determining if pain appeared to play a role in the participant's engagement in problem behavior. For the first four participants, scores on participant CNVPAs indicated that they were not experiencing pain during the implementation of the BFA and contingency reversals as evidenced by relatively low scores when compared with those completed by their parents based on days they have been experiencing pain.

However, in the case of Hazel, the score obtained on the CNVPA following the completion of the BFA and contingency reversal indicated that Hazel may have been experiencing pain ("pain day" = 35; BFA procedures = 16). Additionally, Hazel's varied frequencies of problem behaviors across both BFA conditions and the contingency reversal is further evidence that her engagement in problem behavior may not be entirely due to commonly manipulated environmental variables. In summary, the CNVPA provided a valuable numerical conceptualization of the likelihood that participants may or may not be experiencing pain during the procedures of the study and lent to the control of a critical variable for this population.

Implications

The current study resulted in evidence of preliminary support for the successful implementation of BFA procedures, contingency reversals, and the use of a pain scale to aid in ruling out a critical variable that has the potential to be tied to the individual's engagement in problem behavior. The results obtained from this study have several implications including support for the use of behavior analytic assessment procedures with individuals with CHARGE,

the potential for these procedures to be used in various settings, and successful use of pre-assessment measures to help researchers/interventionists tailor BFA conditions to an individual by including specific reinforcers and types of attention and demands.

Use of BFA with Individuals with CHARGE

While the publication of copious amounts of literature through the years has shown behavior analytic strategies to be successful with individuals of varying ages, diagnoses, and overall developmental presentations, little to no literature exists on how individuals with CHARGE syndrome respond to these principles. One of the most critical components of addressing problem behavior is identifying the function (Carr, 1977). After the function of problem behavior is known, an intervention that maps directly onto the function can be designed. While one could make the assumption that due to the successful use of FAs with various populations, FAs would also be successful with individuals with CHARGE. However, the presence of sensory impairments of both vision and hearing, as well as other medical complexities, difficulty sleeping (Hartshorne et al., 2009), chronic pain (Stratton & Hartshorne, 2018) in individuals with CHARGE syndrome have the potential to make identifying the function of problem behavior more difficult. The nature of FA methodology and its ability to be effective as it currently stands relies somewhat on an individual's ability to detect and experience certain changes and manipulations of the environment. For example, during the attention condition, it is critical that an individual is aware that another person is in the room and can hear or experience the attention that is being given to them contingent upon their problem behavior. Depending on the individual's level of hearing or vision, this may become more difficult and may require extensive modification of the condition; however, no modifications to conditions were made for this study.

The results of this study indicated that for this group of participants, current BFA methodologies were successful in identifying the function of problem behavior in the majority of participants, despite impairments in multiple sensory systems. Further, an added measure/variable to assess during the BFA relevant to this particular population was the use of the CNVPA to determine if pain was playing a role in the presentation of problem behavior. This measure was especially helpful when considering the results of participants whose BFA's did not yield differentiated results (i.e., Simon) or resulted in limited variation between conditions (Hazel). While the BFA alone was not successful in identifying a clear function of problem behavior for these participants (Simon and Hazel), it provided useful information in regards to the need for further exploration and manipulation of variables – specifically in the areas of the FA being implemented by a parent or familiar individual, repeating or extending the length of conditions, or gathering more information about the possible presence of pain.

Use of a Contingency Reversal as Means of Confirming BFA Results

Across FA literature, there are several methodologies that have been implemented to provide support for the results of FA procedures. Among these are repeating conditions (as in the traditional FA; Iwata et al., 1982/1994), confirmatory analysis (Tincani et al., 1999), and the contingency reversal (Northup et al., 1991; LeGray et al., 2010). While all of these have been successful in offering further support of functions of problem behavior identified during an FA, there are potential pros and cons to each. While repeatedly implementing conditions and observing similar patterns across each re-implementation is effective and allows for a confident hypothesis regarding the function of problem behavior, it is also time-consuming and may not be feasible for all situations. In regards to a confirmatory analysis, which involves the repetition of only the condition that resulted in the highest frequency of problem behavior (Tincani, 1999),

this procedure does look to replicate similar frequencies of behavior during the condition of the hypothesized function; however, this methodology does not allow for comparison against other conditions outside of those completed during the original FA.

While both of the previously discussed methods are useful in their own right, the contingency reversal supplies a middle ground between effectiveness, control, and efficiency. As previously discussed, a contingency reversal provides tightly controlled behavioral contingencies across several conditions. A contingency reversal is a quick and clear way to determine if the hypothesized function is the solitary function maintaining the behavior or if other functions/variables may be at play. For example, in the current study, contingency reversals for Harrison, Lizzy, and Elsa indicated that the function identified in the BFA was the maintaining function because little to no problem behavior was observed during the "A" conditions and an increase in problem behavior during the "B" condition. In the case of Hazel, her BFA results were variable; however, a contingency reversal was completed for the condition that resulted in the highest frequency of problem behaviors – social attention. During the contingency reversal, Hazel's problem behaviors were observed across all three conditions – most notably during the conditions in which Hazel was receiving attention contingent upon the absence of problem behavior. The results of this contingency reversal allowed for interventionists/researchers to come to a confident conclusion that multiple functions or other variables, such as chronic pain or painful experiences, may be playing a role in Hazel's engagement in problem behavior. Overall, the contingency reversal was demonstrated to be effective in confirming the results of the BFA or indicating the need for further exploration.

Feasibility of BFA Implementation

Over the years, professionals in the field of behavior analysis have supplied several alternatives to the traditional FA (Iwata et al., 1982/1994) that have lent themselves to increasing the convenience, efficiency, and effectiveness of implementation. In the past, the FA was viewed as a luxury that only those with ample amounts of time in a controlled clinical setting could afford (Axelrod, 1987); however, research on these alternative methodologies, including the BFA, have proved otherwise. BFAs have proved to be successful in a variety of locations including vocational (Wallace & Knights, 2003), home (Wacker et al., 2004), school/classroom (Boyajian et al., 2001; LeGray et al., 2010), and outpatient settings (Derby et al., 1992; Call, Wacker, Ringdahl, Cooper-Brown, & Boelter, 2004).

While it was not a direct research endeavor of the current study, when the setting used is combined with the positive results of the majority of BFAs conducted, it appears there is evidence in support of the versatility and adaptability of BFA procedures. The current study was conducted in a university-based outpatient clinic and in meeting rooms at a hotel being used for the 13th Biennial CHARGE Syndrome conference in Melbourne, Victoria, Australia organized by the CHARGE Syndrome Association of Australasia. As can be seen from the results of the study, BFA procedures were successful when implemented in this novel setting. This finding lends further evidence in support of previous studies that BFAs can be implemented in a variety of settings outside of strictly controlled settings without diminishing the potency of conditions.

Limitations

While the present study provided support for the use of FA procedures for individuals with CHARGE syndrome, several limitations with the current study exist. One limitation of the current study was that the three out of five participants were high functioning in terms of

expressive language. As previously stated, individuals with CHARGE syndrome present across a spectrum in terms of functioning in hearing, vision, language/communication, cognitive functioning, and medical concerns (Blake & Prasad, 2006). Therefore, the current study only provides evidence for this specific group of participants and results cannot yet be generalized to individuals with CHARGE Syndrome as a whole; however, it provides evidence that lends itself in the direction of support for the use of these procedures with individuals with CHARGE Syndrome.

Another limitation to the current study was the collection of CNVPA data for the 'sick' or 'in pain' day from parent memory, rather than actual observation. While this method was appropriate for the preliminary investigation of the utility of such a measure in conjunction with ABA assessment procedures, it may be beneficial to collect data regarding sick or in pain days in real time. This would help improve the validity of scores for days when an individual is in pain and would aide in reducing the possibility of inflated scores or incorrect recall of behaviors and indicators associated with the expression of pain for that particular child. Additionally, results could also be compared to 'baseline' measures that parents complete when the child is reported to have a day that appears to be pain-free.

In terms of data collection, the BFA was the first time that research assistants saw the individual's problem behavior in real time. While researchers had the information from the functional interview and operational definitions derived from information gathered during the interview, not having seen the individual engage in the problem behavior before may have contributed to some discrepancies in IOA, or agreement on what qualified as engagement in the behavior. Specifically, there was a range between 90-100%, with an average of 96.32% interobserver agreement across participants While time constraints did not allow for an

observation of each participant before the BFA, it may be helpful in future studies to incorporate a brief observation period or ask that parents/caregivers bring short videos depicting the target problem behaviors to ensure a general consensus between parents and researchers regarding what the target behavior is.

Lastly, as can be seen in the research, familiar versus unfamiliar individuals (Huete & Kurtz, 2010) can have an impact on the frequency of problem behaviors and ultimate outcomes of FA procedures. English and Anderson (2004) cited various reasons for these differences in responding including caregivers serving as a discriminative stimulus and the presence of a caregiver versus an experimenter altering the potency of a reinforcer. Due to the time constraints associated with data collection for the current study, the training of caregivers in the implementation of FA procedures for participants who did not show differentiation across conditions could not be completed.

Future Studies

While the results of the current study provide preliminary evidence of support for the use of BFA procedures along with contingency reversals as confirmatory measures, there is still extensive work to be done to further investigate this topic. Although the participants in the current study covered a large age range (8-22 years old) and were evenly split between males and females, larger participant sizes with varied presentations of CHARGE Syndrome in regards to cognitive functioning, levels of hearing and vision, age, and language should be included. Specifically, it will be important for future studies to examine the effectiveness of BFA procedures with individuals who present with more significant hearing and/or vision loss as these sensory impairments could have implications for the way the conditions need to be presented

(i.e., changing the kind of attention/way attention is delivered so the individual is aware of the person's presence, etc.).

Results from the current study indicated that the BFA alone may not be sufficient in detecting problem behavior in all individuals with CHARGE, as is true with other populations (e.g., the cases of Simon and Hazel). For participants like Simon, a traditional FA (Iwata et al., 1982/1994) with repeated and extended condition times or an IISCA (Hanley et al., 2014) may be necessary. Hazel's variable engagement in problem behavior across conditions could be beneficial in further teasing apart functions. Specifically, running conditions a second time, combining functions into one condition (as in done in the IISCA), or asking follow up interview questions of parents and caregivers could help in the design of conditions and ultimately the observation of differentiation across conditions. Further, in the case of Simon, who engages in problem behavior primarily towards or with familiar individuals (i.e., family members), it may be beneficial to explore the option of parent, caregiver, or a familiar interventionist to implement the FA to determine if a BFA alone would be successful in identifying a function of problem behavior. Overall, future studies should explore and compare the utility of different FA methodologies in individuals with CHARGE syndrome. Lastly, for participants who continue to demonstrate variable engagement in problem behavior, further exploration into other maintaining variables, such as pain, should be completed.

Lastly, while the current study did not seek to confirm the identified function through the implementation of an intervention and observed reduction of problem behavior, future studies should make this a primary goal. While methods such as the contingency reversal and the confirmatory analysis are helpful in confirming the function of problem behavior, the successful reduction of problem behavior following the implementation of a function-based intervention is

the best evidence that the results of an FA were correct. Additionally, researching the success of function-based interventions following the completion of an FA would provide support for the comprehensive utility of ABA practices in individuals with CHARGE Syndrome. Future studies could also examine generalization and maintenance of intervention results.

Summary

The purpose of the present study was to collect preliminary data regarding the utility of ABA practices in individuals with CHARGE Syndrome – specifically, the ability of BFA procedures in conjunction with contingency reversals to identify the function of problem behavior. Further, the current study aimed to investigate the possible presence of pain in participants with undifferentiated results following the completion of the BFA and contingency reversal, when appropriate. Results provide preliminary evidence in support of the effectiveness of BFA and contingency reversal procedures in identifying and confirming functions of problem behavior in individuals with CHARGE Syndrome. Finally, results indicated the possibility of pain being a mitigating factor in engagement in problem behavior for one participant.

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APPENDIX A

IRB APPROVAL LETTER

MISSISSIPPI STATE UNIVERSITY

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NOTICE OF DETERMINATION FROM THE HUMAN RESEARCH PROTECTION PROGRAM

DATE:	April 05, 2018
TO:	Daniel Gadke, Ph.D., Counseling Ed Psyc & Foundations, Alexander Clarke;Carlen Henington;Kevin Armstrong;Kasee Stratton-Gadke;Keely McCulla;Megan Anderson;Matthew Ferrigno
PROTOCOL TITLE:	Exploring the utility of brief functional analyses procedures for individuals with CHARGE syndrome
PROTOCOL NUMBER:	IRB-18-020
APPROVAL PERIOD:	Approval Date: April 05, 2018 Expiration Date: March 15, 2019

Under an expedited review procedure, the research project identified above was approved for one year on April 05, 2018 by the Mississippi State University Institutional Review Board (MSU IRB). The application qualified for expedited review under CFR 46.110, Category 7.

This memorandum is your record of the IRB approval of this study. Please maintain it with your study records.

Please note that the MSU HRPP accreditation for our human subjects protection program requires an approval stamp for consent forms. The approval stamp will assist in ensuring the HRPP approved version of the consent form is used in the actual conduct of research. **If applicable, you must use the stamped consent form for obtaining consent from participants.**

The MSU IRB approval for this project will expire on March 15, 2019. If you expect your project to continue beyond this date, you must submit an application for renewal of this HRPP approval. HRPP approval must be maintained for the entire term of your project.

If, during the course of your project, you intend to make changes to this study, you must obtain approval from the HRPP prior to implementing any changes. Upon becoming aware of an unanticipated problem that suggests participants or others are at greater risk of harm than was previously known or recognized, a problem report must be submitted to the HRPP as soon as possible, but always within 10 days. Serious problems must be reported verbally within one business day, in addition to the submission of the written Problem Report.

You are required to maintain complete records pertaining to the use of humans as participants in your research. This includes all information or materials conveyed to and received from participants as well as signed consent forms, data, analyses, and results. These records must be maintained for at least three years following project completion or termination, and they are subject to inspection and review by the HRPP and other authorized agencies.

Please notify this office when your project is complete. Upon notification, we will close our files pertaining to your project. Reactivation of the HRPP approval will require a new HRPP application.

If you have any questions relating to the protection of human research participants, please contact the HRPP by phone at 325.3994 or email irb@research.msstate.edu. We wish you the very best of luck in your research and look forward to working with you again.

Approval Period: April 05, 2018 through March 15, 2019

APPENDIX B

RECRUITMENT MATERIALS

Exploring the utility of brief functional analyses procedures for individuals with CHARGE syndrome

Participants will be recruited through a variety of means including recruitment at the following sources:

1. Facebook CHARGE Syndrome Pages
2. Yahoo! Listserv CHARGE Syndrome
3. Email recruitment letter
4. Mail recruitment letter
5. Recruitment flyers or poster at national/international CHARGE Syndrome Conferences

Recruitment Source: Social Media

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1. Facebook CHARGE Syndrome Pages
2. Yahoo! Listserv CHARGE Syndrome

Recruitment Materials

The Mississippi State University Bulldog CHARGE Syndrome Research Lab is currently conducting a research study to determine the utility of brief functional analyses (BFA) on individuals with CHARGE syndrome. Brief functional analyses are used to determine the function of problem behavior – in other words, what is causing an individual to engage in problem behavior? While this method has been used for individuals with high incidence disabilities, there is little to no research in those with low incidence conditions. Specifically, we are interested in whether or not BFA procedures can accurately identify functions of problem behavior. Further, we plan to compare scores on a pain measure to engagement in problem behavior across the analysis to consider if pain may be an underlying motivator to engage in problem behavior.

SO WHAT DO WE NEED FROM YOU?
First, we request that interested individuals respond to the call for participants and complete a demographic questionnaire (15-20 minutes) to determine appropriate fit for the study. The questionnaire will include questions regarding characteristics of CHARGE….Once an individual has been determined to be a good fit for the study they will be asked to do the following:

1) Reinforcer Assessment for Individuals with Severe Disabilities (RAISD) – completed via phone (30 minutes - Parent)
2) Functional Informed Interview – completed via phone (30 minutes - Parent)
3) Preference Assessment – Completed Face-to-Face (20-30 minutes – Child)
4) Brief Functional Analysis – Completed Face-to-Face (1 hour – Child)

WHO CAN PARTICIPATE?

Individuals between the ages of 6-22 years old with a diagnosis of CHARGE (clinical or genetic diagnosis). Exclusionary criteria include:

1) Individuals with complete hearing AND vision loss
2) Individuals engaging dangerous behaviors (e.g., aggression, self-injurious behavior)
3) Individuals currently in residential care
4) Non-English speaking individuals

Please contact Dr. Daniel Gadke or Dr. Kasee Stratton, Assistant Professors and Licensed Psychologists, if you are interested in participating or would like to inquire about any further information: dgadke@colled.msstate.edu or kstratton@colled.msstate.edu

Recruitment Source: Mail Letter

1. Email recruitment letter
2. Main recruitment letter

Recruitment Letter

DATE

Dear Parent/Caregiver:

We hope this [letter/email] finds you and your family well. Over the years, I have been presented with multiple questions and concerns regarding the presentation of problem behaviors in individuals with CHARGE syndrome. As a result of these concerns, we are requesting your participation in a research study, **Exploring the utility of brief functional analyses procedures for individuals with CHARGE syndrome.**

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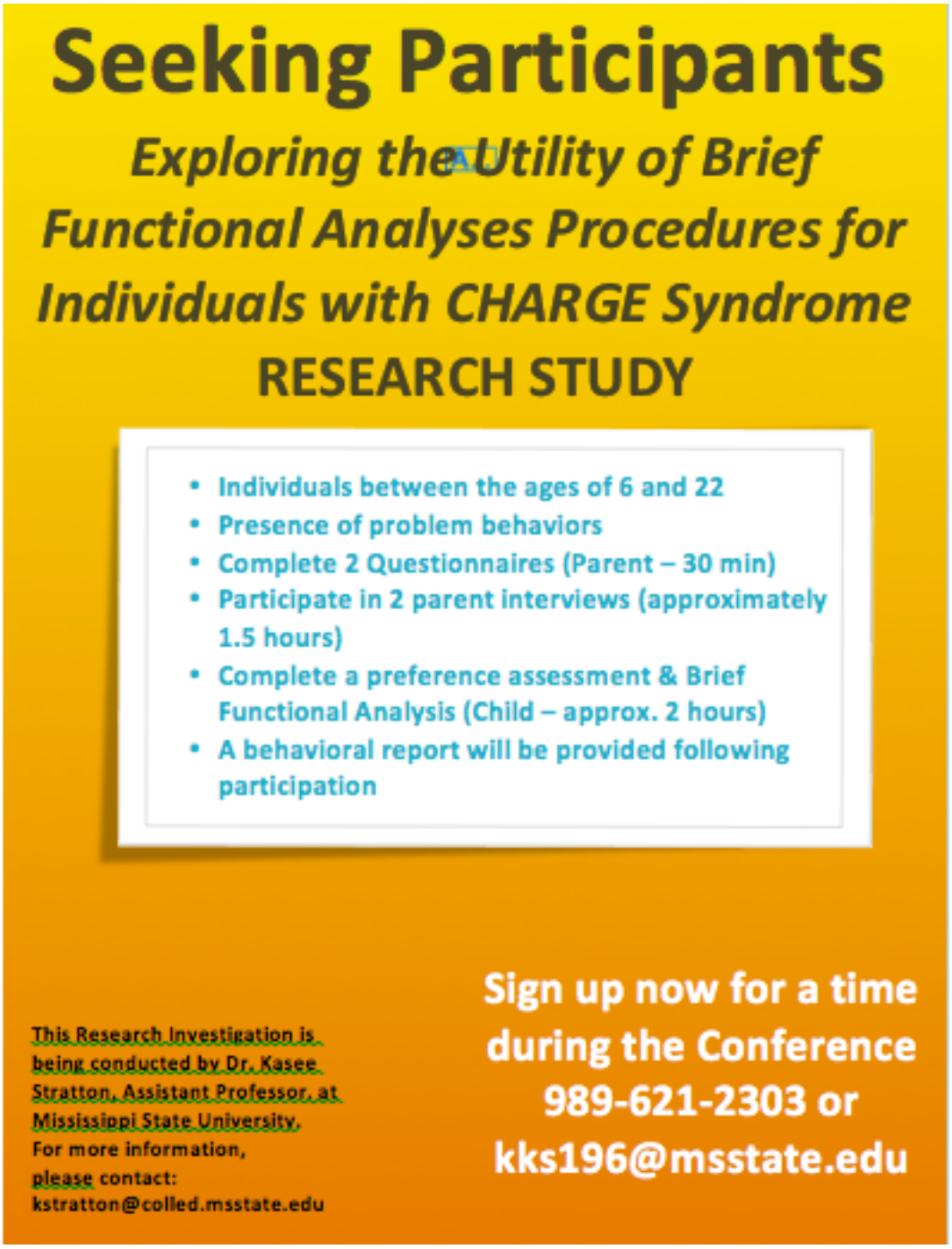
All the best to you and your family,

Hailey Ripple, M.S.
Doctoral Candidate
Mississippi State University
her156@msstate.edu

Recruitment Source: Conference

1. Conference presentation/display (see attachment "Conference Recruitment Flyer")


Seeking Participants
Exploring the Utility of Brief Functional Analyses Procedures for Individuals with CHARGE Syndrome
RESEARCH STUDY
Individuals between the ages of 6 and 22
Presence of problem behaviors
Complete 2 Questionnaires (Parent – 30 min)
Participate in 2 parent interviews (approximately 1.5 hours)
Complete a preference assessment & Brief Functional Analysis (Child – approx. 2 hours)
A behavioral report will be provided following participation
Sign up now for a time during the Conference
989-621-2303 or
kks196@msstate.edu
This Research Investigation is being conducted by Dr. Kasee Stratton, Assistant Professor, at Mississippi State University.
For more information, please contact:
kstratton@colled.msstate.edu

APPENDIX C

DEMOGRAPHIC QUESTIONNAIRE

Brief Experimental Analyses and CHARGE Syndrome
Demographics Sheet

1. Are you the child's? (Please Circle Below)

   MOTHER FATHER GUARDIAN OTHER (please specify) _____

2. Child's Name: _____

3. Child's Date of Birth (Month/Day/Year):_____/_____/_____

4. Child's Gender (Please Circle)

   MALE FEMALE

5. At what age was your child diagnosed as having CHARGE? _____ years old

6. Who made the diagnosis of CHARGE? (e.g., geneticist, ENT, pediatrician)

7. Please indicate the date and nature of your child's most recent surgery:
   _____

8. Please indicate the date and nature of your child's most recent illness:
   _____

GENE TESTING:

8. Has your child been tested for the CHD7 gene mutation? _____ YES _____ NO

   a. If yes: Did you child test positive or negative for the mutation?
   _____ Positive _____Negative

b. When was your child tested? (Month/Year) _____/_____

CHARGE Characteristics: (please check all that apply)

	Check all that apply	**Characteristic**	**Description**
	Example: X	Child has CHARGE Syndrome	
8.		Coloboma of the eye	Coloboma of the iris, retina, choroid, macula or disc (not the eyelid); microphthalmos (small eye) or anophthalmos (missing eye): CAUSES VISION LOSS

9.		Choanal atresia or stenosis	The choanae are the passages that go from the back of the nose to the throat. They can be narrow (stenosis) or blocked (atresia). It can be unilateral (one-sided) or bilateral (both sides), bony or membranous.
10.		Anosmia (missing or decreased sense of smell)	Cranial Nerve I- missing or decreased sense of smell
11.		Swallowing problems	Cranial Nerve(s) IX/X - Swallowing difficulties, aspiration
	Check all that apply	**Characteristic**	**Description**
12.		Facial Palsy	Cranial Nerve VII - Facial palsy (one side or both)
13.		CHARGE outer ear	Short, wide ear with little/no lobe, "snipped off" helix (outer fold), inner fold which is discontinuous with tragus, triangular concha, floppy often stick out
14.		CHARGE middle ear	Malformed bones of the middle ear (ossicles): CAUSES CONDUCTIVE HEARING LOSS
15.		CHARGE inner ear	Malformed cochlea (Mondini defect); small or absent semicircular canals: CAUSE HEARING LOSS AND BALANCE PROBLEMS
16.		Sensorineural Hearing Loss	"Nerve loss"
17.		Vestibular Problems	Balance problems
18.		Frequent Middle Ear Infections	
19.		Heart Defects	Can be any type, but many are complex, such as tetralogy of Fallot
20.		Cleft lip +/- cleft palate	Cleft lip with or without cleft palate, cleft palate, submucous cleft palate
21.		TE (Tracheosophageal) fistula	Espphageal atreaisa, Trancheo-espphageal fistula (TEF), H-shaped TEF; connection between wind pipe and esophagus)
22.		Kidney Abnormalities	Small kidney, missing kidney, misplaced kidney, reflux
23.		Genital Abnormalities (Hypoplasia)	*Male*: small penis, undescended testes *Female*: small labia, small or missing uterus *Both*: lack of puberty without hormone intervention
24.		Growth deficiency	Growth hormone deficiency Other short stature
25.		Typical CHARGE Face	Square face w/ broad prominent forehead, arched eyebrows, large eyes, prominent nasal bridge with square root, thick nostrils, prominent nasal columella (between the nostrils), flat midface, small mouth, occasional small chin, larger chin with age. Facial asymmetry even without facial palsy
26.		Abdominal Defects	Umbilical hernia, omphalocele
27.		Palm crease	Hockey-stick palmar crease
28.		Spine Anomalies	Scoliosis, kyphosis, hemivertibrae
29.		Obsessive-Compulsive Behavior or Perseverative Behavior	Perseverative behavior in younger individuals, obsessive compulsive behavior (OCD) in older individuals

30.		Other	Please describe:
31.		Other	Please describe:

32. At what age (in years) did your child start to walk? If not yet walking, put a check here: _____

My child began to walk at ____ years.

33. I would describe my child's walking ability as *(Please Circle):*

NORMAL/GOOD AWKWARD GAIT/ ASSYMETRIC WOBBLY/UNSTEADY/POOR

34. Please indicate any diagnoses given to your child for her/his behavior (e.g., autism, ADHD):

_____

__

35. What medications and herbal supplements is your child taking on a regular basis?

_____

_

36. To the best of your knowledge, how well does your child see? (with glasses or contact lenses, if used)

(Circle number of ONE choice in each column)

LEFT	RIGHT	
1	1	NORMAL VISION
2	2	SOME TROUBLE SEEING
3	3	MODERATE DIFFICULTY
4	4	MUCH DIFFICULTY
5	5	TOTALLY BLIND

37. To the best of your knowledge, how well does your child hear? (with hearing aids or other hearing devices, if used)

(Circle number of ONE choice in each column)

LEFT	RIGHT	
1	1	NORMAL HEARING
2	2	SOME TROUBLE
3	3	MODERATE DIFFICULTY
4	4	MUCH DIFFICULTY
5	5	TOTALLY DEAF

38. Does your child have problems with sleep? (Please Circle) YES NO

39. How many surgeries has your child had? _____ surgeries

APPENDIX D

FORCED CHOICE PREFERENCE ASSESSMENT PROTOCOL AND DATA SHEET

Forced Choice Preference Assessment Protocol

General Instructions

- Identify 6 items to be used throughout the preference assessment
- Items will be presented 2 at a time (each being paired with the other 5 items once), resulting in a total of 14 pairings.

Instructions for Responses Following Presentation of items

- Participant approaches one of the two items:
    - Unapproached item removed, participant is allowed to play with the approached item for five seconds

- Participant approaches both items at once:
    - Items are blocked

- Participant does not approach either stimuli within 5 seconds of presentation:
    - Participant is prompted to explore each item for 5 seconds
    - Following exploration, items are presented again
    - If participant approaches one of the items, the procedure described above will be used
    - If neither item is approached a second time after five seconds of exposure, both items will be removed and the next trial will begin

Forced Choice Preference Assessment Data Sheet

Student Initials:_____ Interventionist:_____
Date:___/___/_____
Primary/Reliability

Item 1:

1	**2**	**1**	**3**	**1**	**4**	**1**	**5**	**1**	**6**

Item 2:

2	**3**	**2**	**4**	**2**	**5**	**2**	**6**

Item 3:

3	**4**	**3**	**5**	**3**	**6**

Item 4:

4	**5**	**4**	**6**

Item 5:

5	**6**

Item 1:

Item 2:

Item 3:

Item 4:

Item 5:

Item 6:

Problem Behaviors:

BEHAVIOR 1:

APPENDIX E

BFA MATERIALS: PRE-RANDOMIZED CONDITIONS, PROTOCOL, DATA SHEET, AND TREATMENT INTEGRITY

Pre-Randomized Conditions

Participant: #001
Order of BFA Conditions
1. Tangible
2.Control
3. Social Attention
4. Demand

Participant: #002
Order of BFA Conditions
1. Control
2. Social Attention
3.Tangible
4. Demand

Participant: #003
Order of BFA Conditions
1. Social Attention
2. Control
3. Demand
4. Tangible

Participant: #004
Order of BFA Conditions
1. Social Attention
2. Demand
3. Control
4. Tangible

Participant: #005
Order of BFA Conditions
1. Control
2. Tangible
3. Social Attention
4. Demand

Participant: #006
Order of BFA Conditions
1. Demand
2. Control
3. Tangible
4. Social Attention

Brief Functional Analysis
Session Description/Protocol

Date: MM/DD/YYYY
Client: CLIENT INITIALS
Purpose: To identify variables that maintain the patient's problem behaviors. Target behaviors that will receive consequences are: **LIST TARGET BEHAVIORS HERE**
Behaviors that will not receive consequences are: **LIST BEHAVIORS THAT MAY OCCUR BUT ARE NOT SUBJECT OF FA (IF THERE ARE ANY)**

General procedure: Each session will be 10 minutes in length and will be conducted in a treatment room.

TOY PLAY (10 min)
Materials: chair, preferred toys:
Stimulus conditions: Preferred items will be available. Therapist sits in a chair. (Do not prompt the patient to play with toys and do not make requests/demands.)
Consequences for target behaviors: ignore
Consequences for other behaviors: If the patient initiates play or communication, the therapist should interact with him/her or engage in parallel play (do not engage in rough and tumble play). Therapist should attend to the patient every 15-s, as long as targeted inappropriate behavior has not occurred within 5 seconds. Provide 5-10 seconds of verbal and social praise, e.g., "Nice playing with the toys", "Good sitting!"

DEMAND (10 min)
Materials: table, 2 chairs
Demands:
Stimulus conditions: The patient and therapist are seated at the table. Therapist presents demands using 3-step guided compliance consisting of sequential verbal, gestural, and physical prompts. A new prompt is given every 15 seconds with 10 seconds between the verbal, gestural, and physical prompts.
Consequences for target behaviors:
- During the DEMAND sequence: Say "Okay, you don't have to," while removing demand materials. Turn away and do not directly look at client. Do not issue more demands for 30 seconds
- During 30-second ESCAPE period: Ignore and do not look at client. Continue to score target behaviors during the escape period (discuss with Case Manager whether to score these behaviors on a separate key).
- After 30 seconds of escape, present a new demand.

Consequences for other behaviors:
- During the DEMAND sequence: Provide verbal praise for compliance following the verbal or gestural prompt.
- During 30-second ESCAPE period: Do not attend to any problem behaviors and do not interact with the patient.

Out of Seat Behavior: Discuss with Case Manager whether demands will continue to be presented even if the patient leaves the table or whether he/she will be physically guided to the table to work.

SOCIAL ATTENTION (12 min)

Materials: magazine, chair, less preferred toys:

Stimulus conditions: For first 2 minutes, high quality attention is provided (no data collected during this time). Remaining 10 minutes, therapist sits in chair reading a magazine. Toys are present in the room.

Consequences for target behaviors: Brief social attention (e.g., "Don't do that! You'll hurt yourself.") Therapist should attend to each targeted behavior.

Consequences for other behaviors: Ignore all other behaviors

Note: Notify your Case Manager if the patient attempts to obtain attention appropriately (e.g., lightly taps therapist or verbally requests attention).

TANGIBLE (12 min)

Materials: chair, preferred items:

Stimulus conditions: 2 minutes prior to session, the patient is allowed to play with the preferred toy (no data are collected at this time). Once session begins, the therapist sits or stands in the room holding the toy.

Consequences for target behaviors: Therapist says "Okay" and returns the toy to the patient for 30 seconds. Therapist provides no social attention and does not play with the patient.

Consequences for other behaviors: Ignore all other behaviors

Note: Ignore appropriate requests for toys, however, notify Case Manager if this occurs.

Sessions:

Date	Session #	Condition Name	Reliability (check if yes)	Analyzed	Compared
	001				
	002				
	003				
	004				
	005				
	006				
	007				
	008				
	009				
	010				
	011				
	012				
	013				
	014				
	015				

	016				
	017				
	018				
	019				
	020				
	021				
	022				
	023				
	024				
	025				

FUNCTIONAL ASSESSMENT KEY ASSIGNMENTS:

KEYS	BEHAVIOR	DESCRIPTION
1	Self-injury	
2	Aggression	
3	Disruption	

Session Materials:
Social Attention:
Demand:
Toy Play:
Tangible:

BFA Data Sheet

Client: _____Observer: _____ Primary/Rely Trial #: ____ Date: ___/___/_____
Condition:_____

Time	Bx 1	Bx 2	Bx 3	Bx 4	Bx 5	Bx 6	Toy Contact	Comments
1							Y / N	
2							Y / N	
3							Y / N	
4							Y / N	
5							Y / N	
6							Y / N	
7							Y / N	
8							Y / N	
9							Y / N	
10							Y / N	
Total								

Bx1	Definition KEY
Bx 2	
Bx 3	
Bx 4	

BFA Treatment Integrity Data Sheet

Toy Play	Trial #:	Date:
Materials present:	Preferred toys	Y N
Step #	Task	Check if occurred
1	Timer set with intervals app for 10 min with 1 min intervals	
2	If target behaviors occurred, the interventionist ignored such behaviors	
3	If the patient initiates play or communication, the therapist should interact with him/her or engage in parallel play	
4	Interventionist should attend to the patient every 15-s, as long as targeted inappropriate behavior has not occurred within 5 seconds. Provided 5-10 seconds of verbal and social praise	
5	No demands were made of the child during this sequence	

Demand Condition	Trial #:	Date:

Materials present:	Items for demands	Y N
Step #	Task	Check if occurred
1	Timer set with intervals app for 10 min with 1 min intervals	
2	Interventionist issues demands to client every 15 seconds	
3	If target behaviors occurred, the interventionist says “okay you don’t have to” allows the client to escape for 30 seconds before issuing new demand	
4	If the client engages in other behaviors that are not included in the target behaviors (i.e., non-compliance), the interventionist should implement 3 step-guided compliance, with 10 seconds between verbal, gestural, and physical prompts.	
5	If the patient initiates play or communication, the therapist should ignore	

Attention Condition	Trial #:	Date:
Materials present:	mid level preferred toys, magazine or book for interventionist	Y N
Step #	Task	Check if occurred
1	Timer set to run for 2 minutes prior to start of sequence	
2	Client plays with mid level toy while receiving high quality attention (no data are collected at this time)	
3	Timer set with intervals app for 10 min with 1 min intervals	
4	If target behaviors occurred, the interventionist provides brief attention (i.e., “Sally, please don’t get upset and hit or scream”)	
5	If the patient initiates play or communication, the therapist should ignore	
6	No demands were made of the child during this sequence	

Tangible Condition	Trial #:	Date:
Materials present:	Preferred toys	Y N
Step #	Task	Check if occurred
1	Timer set to run for 2 minutes prior to start of sequence	
2	Client is allowed to play with the preferred toy	
3	Timer set with intervals app for 10 min with 1 min intervals	
4	If target behaviors occurred, the interventionist returns the toy to the client for 30 seconds	
5	If the patient initiates play or communication, the therapist should ignore	
6	No demands were made of the child during this sequence	