

Core Cognitive Behavioral Therapy Skills in Williams Syndrome

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LIST OF TABLES

Tables	Page
1. Demographics.....	9
2. CBT Task Descriptive Statistics.....	16
3. Correlate Descriptive Statistics.....	17

LIST OF FIGURES

Figures	Page
1. Study Design.....	14

Introduction

A large body of research indicates that individuals with intellectual and developmental disabilities (IDD) are at increased risk for psychopathology compared to the general population (Deb, Thomas, & Bright, 2001; Brereton, Tonge, & Einfeld, 2006). Estimates of prevalence rates of psychiatric disorders range widely in this population, from as low as 10% (Borthwick-Duffy, 1994), to over 70% (Gustason, 1985; Mattila et al., 2010). Such variable rates are attributed to differences in ascertainment, psychiatric nosologies and measures of psychopathology. However, several recent, well-controlled studies in the US and abroad converge on prevalence rates in the 30-40% range (Cooper, Smiley, Morrison, & Allan, 2007; Dekker & Koot, 2003; Einfeld et al., 2006; Emerson, 2003; Emerson & Hatton, 2007; Molteno, Molteno, Finchilescu, & Dawes, 2001; Reiss, 1990; Rutter, Tizard, Yule, Graham, & Whitmore, 1976, Tsakanikos et al., 2006).

These studies point to high rates of internalizing problems such as anxiety and depression as well as high rates of externalizing issues such as inattention, hyperactivity, and behavior problems. The prevalence rates of some of these problems are similar to those in the general population while others, such as stereotypies, self-injury and aggression, are clearly elevated in those with IDD.

While longitudinal studies of psychopathology in IDD are rare, one recent report indicates that rates of psychopathology tend to decrease as children with IDD enter adolescence (de Ruiter, Dekker, Verhulst, & Koot, 2007). However, rates of externalizing problems and inattention generally decrease more rapidly than do rates of internalizing problems such as anxiety, depression, and other mood disorders (Einfeld et al., 2006). Moreover, problems with anxiety and mood persist throughout adulthood, and generally abate only in older adults with IDD (Cooper, Smiley, Morrison, Williamson, & Allan, 2007). These age trends underscore the need for increased attention to both research and practice towards mood and anxiety disorders in individuals with IDD.

Specific IDD Conditions

Beyond the generally increased prevalence of mood and anxiety disorders in IDD versus typically developing populations, certain types of disabilities or syndromes confer an even greater risk for these internalizing problems. Historically, research on psychopathology in those with IDD did not generally differentiate among etiologies, instead analyzing data by level of cognitive impairment. However, with advances in genetics and the advent of interdisciplinary research in IDD, it is now abundantly clear that different causes of IDD are associated with distinctive behavioral phenotypes and profiles of psychopathology. These phenotypes are best viewed in probabilistic terms, including an increased vulnerability to different psychiatric disorders or proneness to specific maladaptive and other behaviors (Dykens, 1995). Progress has thus been made in characterizing distinctive psychopathologies in Down, fragile X, 22q deletion, Prader-Willi, Smith-Magenis and other genetic syndromes. Here, we focus on one cause of IDD that confers markedly increased risk for anxiety -- Williams syndrome.

Williams Syndrome (WS) is a relatively rare neurodevelopmental disorder caused by a deletion of approximately 26 genes on the long arm of chromosome 7 (7q11.23) (Hillier et al., 2003). Prevalence of the syndrome is estimated at between 1 in 7500 and 1 in 20,000 live births (Stromme, Bjornstad, & Ramstad, 2002; Wang et al., 1997). Individuals with WS typically possess mild to moderate IDD, and their neurocognitive profile is characterized by strengths in facial recognition, language, and verbal short-term memory, with significant weaknesses in visual-spatial abilities. Behaviorally, individuals with WS are often described as hyper-social and friendly, showing high levels of empathy for others. Early anecdotal accounts of those with WS often mentioned their apparent fears and anxious behaviors, and more formal studies over the last decade have confirmed that anxiety and phobias are indeed pervasive aspects of the WS phenotype.

Dykens (2003) described fears and anxiety in a sample of children and adults with WS compared to a matched IDD group of mixed etiology in a set of three studies. According to parental reports on the Fear Survey Schedule for Children-Revised (FSSC-R; Ollendick, 1983), individuals with WS scored significantly higher

than those with other IDs on all five factors of the FSSC-R (fears of failure and criticism, the unknown, injury and death, animals, and medical fears). These fears tended to increase with age. In a second study, the author assessed fears from child report data, finding that WS individuals reported a wider range of fears than controls. Frequently endorsed fears in the WS syndrome group included fear of thunderstorms, loud sounds, and high places.

In a third study, Dykens (2003) assessed presence of diagnosable DSM-III anxiety disorders by administering the anxiety disorder domain of the Diagnostic Interview Schedule for Children to parents (DICA-R; Reich, Shayka, & Taibleson, 1991). 18% of the WS sample met criteria for Generalized Anxiety Disorder (GAD), as extrapolated from rates of Overanxious Disorder (the DSM-III corollary to GAD) and 35% met criteria for specific phobia. However, 50-61% of participants showed signs of anticipatory or generalized anxiety and as many as 94% showed subclinical phobia. Normative declines in fears of the unknown or “spooky things” did not occur with age.

In a study of adults with WS, Cherniske and colleagues (2004) found that thirteen out of eighteen were previously diagnosed with moderate or severe anxiety, and three more displayed mild subclinical symptoms. Echoing the results of Dykens (2003), they found that phobia was the most common diagnosis, followed by GAD.

Leyfer and colleagues (2006) used a structured parent interview, the Anxiety Disorders Interview Schedule for DSM-IV Parent version (ADIS-IV Parent; Silverman and Albano, 1996), to measure prevalence of DSM-IV psychiatric diagnoses in 4-16 year old children with WS. They found high rates of specific phobia (54%), and lower rates of GAD. They also found notable age effects with regards to GAD, as GAD diagnoses were significantly associated with advancing age. None of the subjects aged 4-6 received a diagnosis of GAD, while 14% of the 7-10 year old group met criteria, as did 33% of children aged 11-16. There was no relationship between phobia diagnosis and age. These trends indicate that phobia may be a stable aspect of the WS phenotype, and that generalized anxiety symptoms may worsen throughout development.

Einfeld, Tonge, & Rees (2001) administered the Developmental Behavior Checklist (DBC; Einfeld and Tonge, 1995) to parents or caregivers of youth and adults with WS three times over an 8-year period. They found that compared to an epidemiological control group with nonspecific IDD, those with WS scored significantly higher on the anxiety domain of the DBC at initial assessment. These effects were maintained at follow-up five years after the initial assessment. At a third time point eight years after initial assessment, however, anxiety subscale scores decreased significantly for the WS group, and were similar to controls (Tonge & Einfeld, 2003). Thus, anxiety persisted across development, with mixed findings in the sample at older ages.

Woodruff-Borden and colleagues (2010) measured the longitudinal course of clinically significant levels of anxiety in children and adolescents with Williams syndrome aged 4-13 years at initial assessment using the ADIS-IV parent version. At initial assessment, 60% of the sample had a diagnosable anxiety disorder, with phobia and GAD being the most common. A full 82% had an anxiety disorder at some point during the study, with 62% of this group (51% of the total sample) showing chronic, persistent anxiety, defined by the authors as having an anxiety disorder diagnosis in at least 80% of assessments after the initial diagnosis. 72% of individuals with a diagnosis developed an additional anxiety disorder at subsequent assessments, and the odds of receiving a diagnosis did not differ by age. Content of phobias also tended to remain relatively stable with increasing age.

In brief, despite different sample sizes, age groups, or diagnostic tools, extant studies have consistently indicated that anxiety is a persistent and salient feature of WS. Further, this anxiety is often clinically significant and diagnosable.

Interventions

Despite elevated prevalence rates of anxiety and other internalizing disorders in IDD populations, there is a paucity of research on psychosocial interventions targeting psychopathology in these groups, and no intervention work focusing on WS specifically (Hatton, 2002). Instead, most interventions in IDD populations rely on psychotropic medications, with high rates of polypharmacy, low

quality of psychiatric care and fragmentation of mental health services or systems of care (Holden & Gitleson, 2004; Lott et al., 2004; Lewis, Lewis, Leake, King, & Lindemann, 2002).

Beyond the reliance on pharmacological approaches, the lack of data on psychosocial approaches is also attributed to perception by therapists and researchers that any concomitant mental health problems in IDD are merely an extension of the broader cognitive impairment and thus not reflective of psychiatric disorders. This bias, known as diagnostic overshadowing (Reiss, Levitan, & Szyszko, 1982) persists despite the fact that recent research indicates that criteria for psychiatric diagnoses require little to no modification in mild-to-moderate IDD (Mason & Scior, 2004; Fletcher, Loschen, Stavrakaki, & First, 2007). Further, self-report from individuals with IDD is generally accurate and valid enough to reliably diagnose internalizing disorders (Hurley, 2006; McBrien, 2003; Lindsay, Michie, Baty, Smith, & Miller, 1994).

Moreover, in the past practitioners may have assumed that individuals with IDD did not possess the requisite skills to succeed in psychotherapy. This so-called 'therapeutic disdain' of performing psychotherapy with IDD individuals has limited the availability of psychotherapy for these populations (Bender, 1993). As such, having a low IQ (< 70 or 85) is typically an exclusionary criterion from studies examining therapeutic efficacy. Despite many descriptive studies on the phenomenology of psychopathologies in IDD, diagnostic overshadowing and therapeutic disdain persist to the point where mental health treatment in IDD still lags significantly behind treatment in the general population (Krahn, Hammond, & Turner, 2006). While behavior analytic approaches aimed at reducing specific maladaptive behaviors in the IDD population are commonly used, other therapeutic approaches have not garnered as much research attention, including widely used cognitive-behavioral therapies.

Cognitive-Behavioral Therapy

Since psychology's cognitive revolution of the 1970s, cognitive behavioral therapy (CBT) has become a dominant psychotherapy modality for treatment of a

wide range of psychopathology across the lifespan. CBT has been shown to be particularly effective in treating mood and anxiety disorders (Dreissen & Hollon, 2010; Hofmann & Smits, 2008; Kendall et al., 1997), yet few controlled trials have examined its efficacy for treating mood and anxiety problems in IDD.

While there are many different forms of CBT, they all revolve around the concept that thoughts and beliefs mediate emotions and behavior, a concept known as the cognitive model (Beck, 1995). Following this model, the content of thoughts and beliefs, rather than specific aspects of an event or situation, determine an individual's affect. Affect can be altered positively through identifying and challenging negative thoughts and beliefs. Similar to the general population, research shows that depression and anxiety in people with IDD are also associated with negative cognitive styles, indicating that the cognitive model is indeed applicable to individuals with IDD (Nezu, Nezu, Rothenberg, Dellcarpini & Groag, 1995).

CBT treatments for mood and anxiety disorders teach important problem solving and coping skills designed to address the core symptoms of these disorders. These skills are not only aimed at cognitions, but also extend to awareness of physiological reactions to thoughts and emotions, and implementation of physical relaxation techniques. CBT also incorporates behavioral concepts such as operant conditioning. While the cognitive components of CBT can be abstract, its effective use in children shows that it can be successfully adapted for developmental level, and that adult cognitive abilities are not necessarily prerequisites for achieving a positive treatment response in CBT (Kendall et al., 1997).

Readiness for CBT

Suitability and readiness for CBT depends heavily on an individual's understanding of the cognitive model, or that thoughts mediate emotions. Furthermore, comprehension of the cognitive model requires the presence of several fundamental skills. Most basically, the individual must be able to identify emotions, and differentiate between thoughts, emotions, and behaviors. Beyond recognition of thoughts, emotions, and behaviors as distinct constructs, CBT

requires at least some understanding of the causal connection between thoughts and emotions. Indeed, CBT success among typically developing individuals with mood and anxiety disorders was correlated with greater understanding of these basic skills prior to therapy (Safran, Segal, Vallis, Shaw & Samstag, 1993).

A handful of recent studies have evaluated readiness for CBT in people with IDD. Taken together, these studies indicate that individuals with IDD do possess certain deficits in core CBT skills (Dagnan & Chadwich, 1997; Sams, Collins, & Reynolds, 2006). Within-group correlations consistently showed that greater cognitive ability, especially verbal ability, was related to better performance on CBT tasks (Joyce, Globe, & Moody, 2006). Perhaps the most consistent finding across the literature is that individuals with IDD have trouble grasping and applying the concept of cognition as a mediator between an event and an emotion (Dagnan, Chadwich, & Proudlove, 2000).

However, one study found that training improves performance on a cognitive mediation task (Bruce et al., 2010). Further, compared to tasks involving cognitions, individuals with IDD showed relative strengths in assessments of myriad other core CBT skills, such as understanding emotional states, linking events to emotions, and discriminating between thoughts, feelings, and behaviors.

Each study examining CBT readiness in IDD has been conducted with mixed IDD groups made up of varying etiologies. This is important with regards to determining the generalizability of CBT interventions across large swaths of the IDD population. However, CBT interventions are generally conducted one-on-one or in small groups, where individual differences among clients can strongly influence the direction and content of therapy. Focusing research on distinct syndromes associated with IDD would allow the field to parse out syndrome-specific characteristics with regards to CBT skills. This knowledge would be highly valuable to clinicians in helping to determine whether CBT might be appropriate for a given client with IDD, and what a prudent treatment plan might be considering their phenotype. Given the elevated anxiety and strong verbal ability commonly found in WS, intervention via CBT in this population appears indicated and promising. Thus,

WS presents an ideal opportunity to investigate the phenomenology of core CBT skills and ability to learn these skills in a discrete population.

Methods

Design

This study used a randomized, independent group design to assess the effect of training on two tasks assessing two core CBT skills: discrimination between thoughts, feelings, and behaviors; and understanding cognitive mediation. Participants were randomized into either the skills training group or a control group where participants received education on musical instruments. CBT skills were assessed in all participants at three time points: at baseline (Time 1), following the administration of the training and control interventions (Time 2), and at follow-up 3-6 months following the baseline assessment (Time 3).

Participants

Participants were 30 individuals (22 males and 8 females) with Williams Syndrome who were selected to attend a weeklong residential music camp at Vanderbilt University. Participants ranged in age from 17-41 years old, with a mean age of XX (SD). Fifteen participants were randomized into the intervention group and 15 into the control group. Each group was comprised of 11 males and 4 females. See Table 1 for descriptive statistics of demographic data.

Table 1. Demographics

	Sample Size	Gender (M:F)	Mean Age (SD)	Mean FSIQ (SD)	Prior Therapy (Yes:No)
Intervention	15	11:4	23.93 (7.10)	73.20 (19.89)	4:11
Control	15	11:4	26.60 (7.59)	69.40 (12.90)	7:8

Inclusion and Exclusion Criteria

Participants were selected to attend the camp based on a confirmed diagnosis of Williams syndrome. No strict exclusion criteria regarding comorbid

psychopathology or medication usage were implemented, though the participants were screened to ensure that they could meet the expectations of a residential summer camp program. Therefore, this was not a truly random sample. There were also no geographical limitations and camp attendees were culled on a national level across the United States. All camp attendees were included in this study.

Measures

Demographics Questionnaire. Parents completed a questionnaire that provides background information about the participant's WS diagnosis, hearing loss, age, gender, family income, education, current medications, and history of psychotherapy.

Child Behavior Checklist (CBCL; Achenbach, 2001). The CBCL asks parents to rate 112 problem behaviors and contains an Internalizing Domain (anxious/ depressed, somatic complaints, withdrawn subdomains), Externalizing Domain (non-compliant and aggressive behavior subdomains) and three additional subdomains (social, thought, attention, and other problems). Raw scores were used in data analyses.

Vineland Adaptive Behavior Scales, Second Edition (VABS-2; Sparrow et al., 1984). The Vineland identifies the performance of behaviors required for person or social selfsufficiency. It yields standard scores for an Adaptive Behavior Composite, and Communication, Daily Living Skills, and Socialization subdomains.

Multidimensional Anxiety Scale for Children 2nd Edition (MASC 2; March et al., 1997). The MASC 2 is a brief assessment of anxiety designed for research and screening purposes. It yields a total anxiety score, as well as subscale scores assessing the following domains: separation anxiety/phobias, generalized anxiety, social anxiety, humiliation/rejection, performance fears, obsessions and compulsions, physical anxiety symptoms, panic, tense/restlessness, and harm avoidance. Raw scores were used in data analysis.

Kaufman Brief Intelligence Test, Second Edition (K-BIT2; Kaufman & Kaufman, 2004). The KBIT-2 is a brief intelligence test designed for research and screening purposes that provides a verbal intelligence score, nonverbal intelligence score, and full-scale IQ. The verbal scale consists of two subtests that assess vocabulary knowledge and the ability to identify terms from a short description, respectively. The nonverbal score is derived from one subtest that tests the ability to complete picture matrices. This test has been used successfully in research with IDD populations.

The Thought-Feeling-Behavior Task (TFB Task; adapted from Quakley, Reynolds, & Coker, 2004; Sams et al., 2006; Bruce et al., 2010). This task was originally designed to measure the ability to discriminate between thoughts, feelings, and behaviors in children without IDD, and was subsequently adapted for use in IDD. For the present study, several items were further adapted to make them better suited for an American sample, and also to avoid some of the salient fears in WS (e.g., story about a thunder storm). In order to maximize variability in analyses, additional items were also added to the task that matched the reading level and approximate difficulty of the established items. The original task included 18 total sentences and the adapted version includes 36. There were an equal number of stories containing positive and negative affect.

A trained administrator read six short stories to the participant. Each story contains three sentences that either represents a thought, feeling, or behavior respectively. An example follows:

Sarah is going on a trip to the beach. Sarah is very excited (feeling). Sarah hoped that she could have an ice cream there (thought). Sarah put a dollar in her purse to buy the ice cream (behavior).

Following each reading of the complete story, the thinking, feeling, and behavior sentences, are again read aloud individually. After each sentence is presented, the participant is asked to identify which category the sentence belongs to. Presentation order of the stories is randomized, and the order of the sentences within the stories

is counterbalanced in order to ensure that participants cannot make responses based on a pattern. Two different forms of the task were used during pre-test and post-test in order to avoid practice effects. These forms were pilot-tested in order to ensure equivalence in task difficulty and affective valence across forms. The maximum score on this task is 36, or 1 point for each sentence.

Cognitive Mediation Task (adapted from Dagnan et al., 2000; Doherr, Reynolds, Wetherly, & Evans, 2005). This task assesses the participant's ability to link thoughts and feelings and recognize that thoughts mediate between events and emotions. The task was adapted from previous research as earlier studies only asked participants to provide a feeling. In the adapted version, items were included that asked participants to provide both a cognition and an emotion.

For each item, participants are asked to imagine themselves in a particular situation. Then, they are provided with either a feeling or a thought. If provided a feeling, the participant is asked to supply an appropriate thought that may have mediated between the situation and the feeling. If the participant is provided with a thought, they are asked to supply an appropriate feeling that may have arisen given the situation and thought provided. Participants are then asked to explain why they chose the response that they did. For example:

Missing feeling:

"Imagine that you are going away for a while. You say goodbye to your family."

You think: 'I can't wait for my vacation.'

How do you think you would feel if you thought, 'I can't wait for my vacation?'

Why do you think you would feel (insert answer)?"

Missing cognition:

"Imagine that you are going away for a while. You say goodbye to your family.

You feel excited.

What thought might you have had that would make you feel excited?"

The test consists of 8 items in whole. Items are balanced between positive or negative tone (4 each), and between those requiring participants to provide a thought versus a feeling (4 each). Participants receive 1 point for each item if it is judged that their response indicated understanding that cognition plays a mediating role between situation and emotion based upon the explanation of their response. Two different forms of the task were used during pre-test and post-test in order to avoid practice effects. Half of the items from the pre-test and half of the items from the post-test were randomly selected to be used during the follow-up assessment.

Experimental and Control Interventions

The experimental intervention is a manualized procedure that is designed to train participants' ability to discriminate between thoughts, feelings, and behaviors, as well as ability to recognize the mediating role of cognition. This intervention was adapted from previous research (Bruce et al., 2010) in order to be appropriate for an American sample of adults/adolescents with WS. The intervention permits more focused instruction if the participant appears to have difficulty grasping the concepts being taught. Repetition of concepts, miming emotions, eliciting personal examples from the participant, and visual cues are all utilized to enhance learning.

The control intervention consisted of a manualized music education training focusing on musical instruments from cultures around the world. The structure matched that of the training intervention, but did not address any of the content contained in the training intervention. These intervention and control conditions were delivered in a single, individual session that was approximately 30 minutes long.

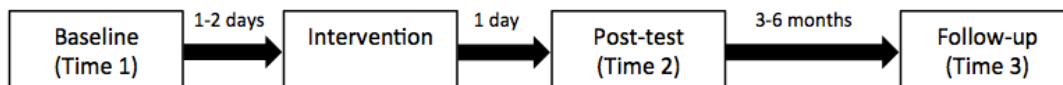
Procedure

Participants were randomized into either the experimental or control condition. Participants' caregivers completed the demographic information prior to Time 1. At Time 1, participants completed the KBIT-2, TFB Task, and Cognitive

Mediation Task. Participants received the interventions 1-2 days following the Time 1 assessment. Each participant completed their Time 2 assessment 1 day following their respective intervention.

Tasks were administered by trainees in clinical psychology or special education, with the exception of some follow-up assessments that were conducted by a trained, supervised undergraduate research assistant. Assessors were blind to participant's group status. Follow-up at Time 3 occurred approximately 3-6 months following Time 1 via phone interview. See Figure 1 for a visual representation of the study design.

Figure 1. Study Design



Results

Demographics and Psychotherapy Usage

There were no significant differences between groups on demographic factors such as IQ, age, gender, or prior participation in therapy. Therefore, covarying these factors in subsequent analyses was not necessary. Overall, 11 out of 30 participants (36.7%) were currently receiving or previously participated in psychotherapy.

Distribution of Task Data

Q-Q plots were generated for each of the experimental tasks. Visual inspection of these plots indicated that the data were not normally distributed. The plots displayed an S-shaped curve, indicating skewness. Specifically, data were positively skewed. Log transformations were not successful in normalizing the data. In order to account for this deviation from normality, nonparametric tests were used in data analyses whenever possible.

Performance on Thought Feeling Behavior Task

In order to assess ability on the TFB task baseline scores were compared to the null hypothesis of scores that would be expected if subjects performed at chance level. In order to account for the violation of normality a one-sample Wilcoxon signed rank test was performed rather than a one-sample T test. Results indicate that participants scored significantly above chance on this measure ($p < .001$).

Effect of Training

A Friedman's test was conducted in order to assess effects of training across groups and across time. These analyses revealed that there was no significant main effect of time and no significant group by time interaction effect. These results are true both when including only T1 (baseline) and T2 (post-intervention) and also when including T1, T2, and T3 (follow-up). There was no relation between scores on

the follow-up measure and length of time from intervention to follow-up. See Table 2 for descriptive statistics by group for CBT tasks.

Table 2. CBT Task Descriptive Statistics

	TFB Pre	TFB Post	TFB Follow	CM Pre	CM Post	CM Follow
Intervention	29.33 (8.86)	28.80 (7.91)	28.92 (7.99)	5.60 (2.06)	6.13 (1.68)	6.33 (1.56)
Control	30.13 (6.17)	30.67 (4.89)	29.40 (6.12)	6.07 (0.88)	6.8 (0.94)	6.47 (1.19)

Effects by Item Type

Responses on the TFB task were analyzed by type (thought vs. feeling vs. behavior) in order to determine if there was some systematic bias in which response option participants selected. No significant difference were found in responses provided by type. Specific Cognitive Mediation task items were analyzed based on whether the participant was asked to provide a thought or an emotion as a response. Using the whole sample at baseline, results indicate that there is no significant difference in participant’s ability to provide an appropriate thought versus an appropriate emotion.

Correlational Analyses

Correlational analyses found that verbal IQ was significantly positively related to all measures assessing CBT readiness at all time points (r range = .37 to .65; p range = .000 to .043). Nonverbal IQ was also significantly positively correlated with all assessments of CBT readiness save for the CM post-test (r range = .38-.53; p range = .002-.038). Prior therapy experience was not correlated with performance, nor were age or gender. Performance was not correlated with level of anxiety as measured by the MASC and CBCL, with level of depression/withdrawal as measured by the CBCL, or with level of communication, daily living, or social skills as measured by the Vineland. See Table 3 for descriptive statistics by group for these correlates.

Table 3. Correlate Descriptive Statistics

	MASC Total	VABS Comm	VABS Daily	VABS Social	CBCL Intern	CBCL Extern
Intervention	51.13 (19.17)	62.43 (14.15)	64.50 (7.76)	71.64 (9.81)	6.14 (4.59)	5.21 (5.70)
Control	53.92 (18.11)	53.57 (13.59)	58.93 (10.70)	68.50 (8.36)	7.00 (5.59)	2.67 (2.61)

Discussion

This experimental, controlled study is the first of its kind to investigate the effect of training on CBT skills in a discrete IDD population. Results did not support the main study hypothesis that a brief training module would increase ability in core CBT skills when compared to a control intervention. These results, however, do not rule out the possibility that those with WS can indeed take advantage of cognitive-behavioral therapies, and that they can be trained in CBT readiness skills if necessary prior to therapy. Indeed, the flip side of these results is that many participants already had the requisite skills for CBT, and that readiness training should be provided on an individualized, as-needed basis.

This study had a small sample size, but data indicate that the failure to achieve training effects is not likely to be rectified solely by the inclusion of more subjects. Instead, a number of other possible explanations likely influenced study results. First, this study was adapted from and attempted to replicate results from a similar study assessing CBT readiness in an IDD group of mixed etiology (Bruce et al., 2010). The training effects found by Bruce and colleagues (2010) may be partly explained by the fact that the authors excluded individuals from their study who scored over a certain threshold on CBT skill measures. This practice allowed the authors to utilize data only from those individuals who had room for improvement in their skills, thereby reducing possible ceiling effects. The positively skewed nature of the CBT skills data collected in the current study indicates the presence of a ceiling effect. Thus, it is quite possible that the baseline skill of the individuals assessed in this study was high enough to limit room for variability and improvement post-intervention.

The finding that the whole group scored significantly above chance on the TFB task supports the assertion of high baseline skills in study participants. Further, such well-developed skills may be associated with some selection bias and the lack of a truly random participant sample. This study was conducted during a sleep-away summer camp. Individuals with WS are vetted before being accepted into this program, and must have the functional abilities to thrive in this setting. Thus, their

insight and functioning level are likely to be higher than individuals with WS sampled randomly from the population.

The lack of a training effect may also be related to measurement issues. The assessments used to measure CBT readiness in this study were adapted slightly from measures used successfully in past research. However, these measures are not standardized, have not undergone factor analysis, and have not been subject to analyses that would elucidate their psychometric properties. Due to practical constraints, the adapted measures used in this study were piloted on individuals with forms of IDD unrelated to WS. So, while item difficulty and overall measure feasibility were addressed prior to conducting the study, this piloting could not be carried out with the population of interest. Given these constraints on measurement, it is possible that the null result may be accounted for in part by measurement error.

Despite these null results, this study uncovered a number of noteworthy findings. Previous research on CBT skills in IDD populations has found that participants tend to perform more poorly on items requiring them to provide thoughts or cognitions as responses versus emotions. This discrepancy was not present in the current study, which could indicate that individuals with WS possess a distinct CBT readiness profile. Further research directly comparing individuals with WS to various other groups would need to be conducted to test this possibility.

The high performance level of the sample overall is noteworthy in itself. Many individuals achieved perfect or near perfect scores on one or both measures, and individual item responses often evidenced sophisticated social and emotional cognition. For example, one item presented participants with a situation where a group of friends walks by without noticing the participant and the participant feels happy. One individual noted that a possible mediating cognition in this situation might be that the group of friends didn't notice him due to being busy attempting to integrate a new member, and that he was happy because new friendships were being formed. This example hints at an ability to reframe a potentially negatively valenced situation, a skill that is crucial for success in CBT.

The strong positive correlation of IQ with CBT readiness scores is in line with existing literature (e.g. Bruce et al. (2010); Joyce, Globe, & Moody, 2006). It should

be noted, however, that IQ accounted for only 14-42% of the variance in performance on CBT tasks. Performance was not related to other correlates utilized in this study, including presence of behavior problems/psychopathology, adaptive abilities, and previous participation in psychotherapy. Further research on this topic could investigate other potential correlates of CBT skill (i.e. executive functioning, theory of mind, parenting factors) in order to determine what variables other than IQ may make an individual a good candidate for CBT. The lack of a discrepancy in performance between cognitive vs. emotional items runs counter to previous research, and raises the possibility that the profile of CBT skills is different in WS as compared to IDD in general.

Over one third of the study sample had previously or was currently engaged in psychotherapy. Presenting problems were similar to those that might be expected from any population, such as trouble with anxiety, adjustment problems following a move or a parent's divorce, and trouble with attention and concentration. The relatively high rate of psychotherapy usage in this sample is noteworthy, but difficult to put in context, as little information exists regarding psychotherapy usage in WS and IDD populations in general. The 2013 National Survey on Drug Use and Health found that 14.5% of the general population received mental health services over the course of one year, but such recent public health data does not exist for IDD as a whole or for specific IDD syndromes. The field would benefit from more pointed population-based studies assessing psychotherapy usage in IDD populations, including presenting problems, treatment modality used, and profession of healthcare provider (social worker, psychologist, psychiatrist, etc.). Such an understanding of the nature of mental health service usage in IDD would allow for more effective treatment recommendations based on the readiness for CBT literature.

While the primary hypotheses of this study were not supported, future directions for research on CBT readiness in IDD are clear. Subsequent studies would benefit from the development of a more psychometrically sound battery of assessment measures, further refinement and expansion of training paradigms, and more broad replication of findings amongst a variety of IDD populations. The

baseline skill and history of psychotherapy usage in the whole sample were noteworthy findings, and may point to a clinical world where psychotherapy in IDD has developed largely unchecked by scientific rigor. Increasing the empirical support base for therapy readiness and efficacy of treatment in IDD would allow practitioners, patients, and caregivers to make more informed decisions regarding their care and ensure that the services provided offer the maximum possible benefit for those in need.

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