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A parent-report scale of behavioral inhibition: Validation and application to preschool-age children who do and do not stutter

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In Memoriam: Dr. Edward “Warren” Lambert (1945–2017)

On January 13, 2017, the scientific community at Vanderbilt University lost Dr. Lambert, an outstanding, highly respected and much loved colleague, mentor, researcher, and teacher. To paraphrase Ernest Hemingway, *every man’s life ends but it is the details of how he lived that distinguish one man from another*. Dr. Lambert was a brilliant scientist with a long list of scholarly achievements but what distinguished Warren most was his wisdom, integrity, humility, and dedication to his colleagues and his students. He was a gentleman, a true scholar, and an individual with the rare gift of inspiring and enriching the lives of all he worked with and/or mentored. We are all the better from learning from, knowing, working, and laughing with him. Dr. Lambert is dearly missed, but will be long-remembered and not soon forgotten.

Highlights

- The *Short Behavioral Inhibition Scale* (SBIS) – developed for use with preschool-age CWS and CWNS - has good internal consistency and test-retest reliability.
- Preschool-age CWS, compared with their CWNS peers, are more apt – on average and at the extremes – to exhibit significantly higher behavioral inhibition tendencies.
- For preschool-age CWS, behavioral inhibition – as measured by the SBIS - is significantly associated with stuttering frequency, stuttering severity, stuttering-related consequences/reactions, and speech-associated communication attitudes (for CWS older than 4 years of age).
- The *Short Behavioral Inhibition Scale* (SBIS), a brief (i.e., 5-item) parent-report questionnaire of behavioral inhibition, may be considered for inclusion as a part of a comprehensive fluency diagnostic.
- Children who score 13 or lower (i.e., in the low 15th percentile of the SBIS distribution) can be reasonably assumed to exhibit behavioral inhibition tendencies.

Abstract: Purpose: This two-part (i.e., Study 1, Study 2) study investigated behavioral inhibition (BI) in preschool-age children who do (CWS) and do not (CWNS) stutter. The purpose of Study 1 was to develop the Short Behavioral Inhibition Scale (SBIS), a parent-report scale of BI. The purpose of Study 2 was to determine, based on the SBIS, differences in BI between CWS and CWNS, and associations between BI and CWS's stuttering frequency, stuttering severity, speech-associated attitudes, and stuttering-related consequences/reactions.

Method: Participants in Study 1 were 225 CWS and 243 CWNS with the majority of them being included in Study 2. In Study 2, a speech sample was obtained for the calculation of stuttering frequency and severity, and the parents of a subset of CWS completed the Communication Attitude Test for Preschool and Kindergarten Children Who Stutter (Vanryckeghem & Brutten, 2007), and the Test of Childhood Stuttering Disfluency-Related Consequences Rating Scale (Gillam, Logan, & Pearson, 2009).

Results: Study 1 analyses indicated that SBIS is a valid and reliable tool whose items assess a single, relatively homogeneous construct. In Study 2, CWS exhibited greater mean and extreme BI tendencies than CWNS. Also CWS with higher, compared to CWS with lower, BI presented with greater stuttering frequency, more severe stuttering, greater stuttering-related consequences, and more negative communication attitudes (for CWS older than 4 years of age).

Conclusion: Findings were taken to suggest that BI is associated with early childhood stuttering and that the SBIS could be included as part of a comprehensive evaluation of stuttering.

Keywords: Assessment; childhood stuttering; behavioral inhibition

1. Introduction

1.1. Behavioral inhibition (BI): General overview and motivation for its empirical study in childhood stuttering

From early infancy through adulthood, individuals show remarkable variability in the way they react to, interact with, and operate within their environment. Such differences in the intensity and frequency of reactions to stimuli, together with attempts to regulate them are associated in part with individuals' *temperament*. Although, as suggested by Rothbart (2011), temperament is biologically or constitutionally based, its phenotype is influenced or molded by the complex and continued interplay between genetic and environmental factors (Buss & Plomin, 1984).

One temperamental construct that has received considerable attention since its introduction by Kagan and his colleagues (Garcia Coll, Kagan, & Reznick, 1984; Kagan, Reznick, Clarke, Snidman, & Garcia Coll, 1984; Kagan, Snidman, & Arcus, 1998) is *behavioral inhibition* (BI). As described by Rubin, Hastings, Stewart, Henderson, and Chen (1997), BI refers to the tendency to process and react to unfamiliar/novel stimuli (people, objects, social situations) with behavioral signs of cautiousness, fear, restraint, wariness, and withdrawal. Children with strong BI tendencies are typically hypervigilant in novel or uncertain situations, highly alerted to novel stimuli, more likely to present patterns of negative affectivity and often described as shy, quiet, and reticent (Lonigan, Vasey, Phillips, & Hazen, 2004). Also, they are less likely to initiate interactions and often withdraw from social situations and activities (Rubin, Burgess, & Hastings, 2002).

Furthermore, the literature is replete with findings that BI is a prominent risk factor for the development of (social) anxiety in childhood and adolescence pointing to a possible common underlying cause (Clauss & Blackford, 2012; Degnan, Almas, & Fox, 2010; Fox, Henderson, Marshall, Nichols, & Ghera, 2005). For example, Chronis-Tuscano and colleagues (2009) reported that infants who had been consistently rated by their mothers as highly behaviorally inhibited from infancy to childhood had a 4-fold increased odds for a social anxiety disorder diagnosis in adolescence. Another longitudinal study reported that 13-year-old adolescents who had been classified as behaviorally inhibited at age 2 had significantly higher rates of anxiety than their peers who had been classified as uninhibited (Schwartz, Snidman, & Kagan, 1999). Also, Biederman and colleagues (2001) found that the rate of social anxiety disorder is significantly higher in behaviorally inhibited than noninhibited children. Indeed, considerable evidence supports the notion that BI and anxiety are connected to one another. Of particular note, is the fact that BI and anxiety share a number of behavioral as well as psychophysiological traits, for example, overly sensitive threat detection system, preferential allocation of attention towards negative stimuli, an avoidant coping style, and overactive amygdalar responding to threatening stimuli (Pérez-Edgar & Fox, 2005).

Thus, the study of the role of BI relative to childhood stuttering is motivated by a growing literature suggesting that older children, adolescents, and adults who stutter are at a higher risk for (social) anxiety disorders than individuals who do not stutter (Blood, Blood, Maloney, Meyer, & Qualls, 2007; Craig & Tran, 2014; Iverach et al., 2016; McAllister, Kelman, & Millard, 2015; Mulcahy, Hennessey, Beilby, & Byrnes, 2008; Smith, Iverach, O'Brian, Kefalianos, & Reilly, 2014). Behavioral inhibition's moderate

continuity/stability from toddlerhood to adulthood (Degnan & Fox, 2007; Pedlow, Sanson, Prior, & Oberklaid, 1993; Pfeifer, Goldsmith, Davidson, & Rickman, 2002), coupled with its strong link to the development of anxiety disorders (Chronis-Tuscano et al., 2009; Hirshfeld-Becker et al., 2008), could be taken to suggest that BI, at least in some young children who stutter (CWS), may potentially contribute to the presence and/or development of anxiety in later childhood and/or adulthood.

Furthermore, BI seems to be related to classical conditioning with individuals scoring high on BI self-report scales being more sensitive to associative learning and more prone to forming associations between stimuli compared to individuals scoring low on BI. Specifically, individuals high in BI have been shown to acquire stimulus–response associations (e.g., in eye blink conditioning experiments) faster than non-inhibited individuals and for those associations to be more resistant to extinction (Catuzzi & Beck, 2014; Caulfield, McAuley, Servatius, 2013; Caulfield, VanMeenen, Servatius, 2015; Holloway, Allen, Myers, & Servatius, 2014; Myers et al., 2012). Thus, one might speculate, that CWS who are high in BI are at greater risk for relatively rapid conditioning to negative internal (e.g., reaction to their own speech disfluencies) and/or external stimuli (e.g., listeners looking away from the child who stutters when he or she stutters). Such reactions on the part of the child who stutters may, in turn, exacerbate the frequency and/or severity of his or her stuttering and be a contributing factor to the development of associated non-speech behaviors (e.g., head turning, breaking eye contact with listeners, etc.; see Conture & Kelly, 1991 for empirical data pertaining to such non-speech behaviors as well as speculation regarding the possible origins of such behaviors).

Despite the above observations, it is interesting that BI has received minimal, research attention with regard to early childhood stuttering (Choi, Conture, Walden, Lambert, & Tumanova, 2013). To address this gap in our knowledge base, it was the general purpose of the present study to empirically study the role that BI may play in early childhood stuttering in relatively large samples of CWS as well as a control group of normally fluent peers. Specifically, the study attempted to determine differences in BI, if any, between preschool-age CWS and children who do not stutter (CWNS) as well as associations, if any, between CWS's BI and their overt and covert stuttering symptoms (e.g., stuttering frequency and severity). However, before further coverage of the details regarding the present study, a brief overview of BI-related temperamental traits in relation to childhood stuttering is provided to help motivate the present empirical study as well as support the importance of studying this specific temperamental trait in CWS. Specifically, the overview will mainly focus on empirical findings that pertain to differences in temperamental constructs (shyness, negative affectivity, emotional reactivity) reported to be related to BI in children.

1.2. Relation of BI to childhood stuttering: General overview

To provide context for the above discussion of BI, it should be noted that various multifactorial models of stuttering (e.g., Conture et al., 2006; Conture & Walden, 2012; Smith & Kelly, 1997; Smith & Weber, 2017) support the notion that emotional processes may be one important contributor to childhood stuttering. Such theoretical speculation is consistent with the present authors' attempt to determine how behavioral inhibition is associated with childhood stuttering. Although children may demonstrate behavioral inhibition in several different ways, one common way is for them to be sensitive or

reactive to environmental input and stimuli.

The notion that a sensitive/highly reactive temperament may constitute a risk factor for the onset of stuttering in some preschool-age CWS had been proposed and discussed prior to it being empirically studied (Conture, 1991; Glasner, 1949; Peters & Guitar, 1991; Starkweather, 2002). More recently, Guitar (2019) posited that CWS tend to be more sensitive/reactive to unfamiliar people and situations and are more easily aroused by environmental stimuli than CWNS. Furthermore, a sensitive or inhibited temperament, as Guitar has suggested, may lead CWS to be more hesitant to speak, particularly when encountering new situations, new tasks and/or novel conversational partners.

Behavioral inhibition, therefore, is likely to be associated with sensitivity to one's internal and external surrounds. If this is the case, children who tend to be behaviorally inhibited may notice very small changes or differences in their environment. Such hypersensitivity to change, difference and novelty, at least some of the time, may contribute to less than adaptive reactions to environmental change.

Empirical evidence based on parent-report questionnaires of temperament (*Behavioral Style Questionnaire [BSQ]*; McDevitt & Carey, 1978; *Dutch version of Child Behavior Questionnaire [CBQ-D]*; Van den Bergh & Ackx, 2003; *Short Temperament Scale for Children [STSC]*; Prior, Sanson, & Oberklaid, 1989) indicate that CWS, when compared to CWNS, are (a) more sensitive, anxious, fearful, and introverted (Fowlie & Cooper, 1978), (b) less likely to approach unfamiliar situations and people (Fowlie & Cooper, 1978; cf. Kefalianos, Onslow, Ukoumunne, Block, & Reilly, 2014; Reilly et al., 2009), (c) more emotionally reactive and less able to shift attention away from

emotionally arousing stimuli (Karrass et al., 2006), (d) more negative in quality of mood (Eggers et al., 2010; Wakaba, 1998; cf. Lewis & Goldberg, 1997), and (e) more vulnerable and sensitive (Oyler, 1996b). More recently, Kraft, Ambrose, and Chon (2014) found, with such findings replicated by Kraft, Lowther, and Beilby (2019), that CWS's ability to self-regulate and modulate their emotional reactions is significantly and negatively related to the severity of their stuttering. In other words, these findings indicate that CWS with lower self-regulatory skills exhibit greater stuttering severity.

Also, Ambrose, Yairi, Loucks, Seery, and Throneburg (2015) studied the temperament (among other variables) of 19 children with persistent stuttering using the parent-report questionnaire *Children's Behavior Questionnaire Short Form* (Putnam & Rothbart, 2006) and found that they were rated by their parents as more negative in temperament than their peers who recovered from stuttering ($n = 35$) and their normally fluent controls ($n = 37$). However, in a prospective community-based study employing the *Short Temperament Scale for Children* (STSC; Prior et al., 1989) at the age of 2 years, no significant difference in the temperamental trait "approach" was reported between a group of 67 children who recovered and a group of 34 children who persisted in stuttering (Kefalianos et al., 2017) at the age of 2 years.

In addition to parent-report based tests of temperament, Schwenk, Conture, and Walden (2007) measured the reaction of preschool-age CWS and CWNS to auditory stimuli (i.e., unexpected movement noise aperiodically emanating from a remotely controlled camera) and reported that CWS were significantly more vigilant to those background or environmental sounds than CWNS. Interestingly, it has been found that young BI children display heightened reactivity to novel auditory stimuli and an

exaggerated tendency to engage automatic orienting responses to them (Henderson, Pine, & Fox, 2015). Thus, Schwenk et al.'s findings that CWS, when compared to CWNS, are more reactive to and less able to quickly habituate to auditory environmental stimuli, could be taken to suggest that CWS are more apt to present with higher BI than their CWNS peers. Related to such a suggestion, Ntourou, Conture, and Walden (2013) employed an emotion (frustration) eliciting experimental paradigm. These authors reported that CWS were more emotionally reactive than their fluent peers, as evidenced by increased levels of exhibited negative affect.

Recently, Zengin-Bolatkale, Conture, Key, Walden, and Jones (2018) reported that young CWS, when compared to their CWNS peers, while viewing arousing pictures of negative valence, exhibited greater emotional reactivity, as measured by the late positive potential (LPP), an electrophysiological index of emotional processing. Likewise, skin conductance data obtained during a stressful speaking task revealed that CWS with persisting stuttering exhibited higher sympathetic arousal than recovered CWS (Zengin-Bolatkale, Conture, Walden, & Jones, 2018). Finally, results from Boey's (2012) large sample-size epidemiological and phenomenological study of young CWS in Belgium indicated that highly emotionally reactive CWS tend to have more severe stuttering, characterized by a higher percentage of vowel prolongations and more intense physically concomitant behaviors.

Results from the aforementioned parent-report questionnaire, behavioral observation, psychophysiological, and epidemiological studies do not provide conclusive proof of an association between BI and childhood stuttering; however, such findings are generally consistent with the BI construct. These previous findings, while supportive of

possible differences in BI between CWS and CWNS, would appear to need greater empirical support resulting from investigations designed to specifically investigate the BI construct. To the authors' knowledge, the only previously reported empirical study specifically designed to investigate BI and stuttering is that of Choi and colleagues (2013)

Choi et al measured BI by using a protocol/methodology developed by Kagan, Reznick, and Gibbons (1989) to study BI in children. To study the possible association between BI and childhood stuttering, Choi et al. used the Kagan et al (1989) measure or index of BI, that is, the latency (in seconds) to the child's 6th spontaneous comment during an experimenter-child conversational interaction. Using Kagan et al's index of BI, Choi et al reported that preschool-age CWS, compared to CWNS, were more likely to exhibit high BI and that CWS with high, compared to CWS with low, BI exhibited more stuttered disfluencies. Choi et al.'s findings are consistent with Guitar's (2019) notion that at least some CWS have a constitutional bias for "inhibited temperament" (i.e., highly reactive and/or sensitive towards change, novelty or differences in their environment).

Of course, the Choi et al. (2013) study is but one empirical investigation of BI in CWS and their CWNS peers, which suggests that a good deal remains unknown about the possible relation of BI to developmental stuttering. Furthermore, although the Kagan et al index of BI used by Choi et al has been used relatively frequently in the literature and developed by the researchers who introduced the construct of BI, it is not easily applied to and/or widely disseminated in clinical practice. Therefore, it would be of some help to develop a valid, reliable and time-efficient, user-friendly assessment tool for determining

BI tendencies in young children.

1.3. Relation of BI to childhood stuttering: The current investigation (Study 1, Study 2)

Thus, it was the general goal of this two-part (i.e., Study 1 and Study 2) empirical investigation to further study the possible relation between BI and childhood stuttering and develop and apply a clinically-friendly, parent report measure, the *Short Behavioral Inhibition Scale* (SBIS), to determine possible BI tendencies in preschool-age CWS and CWNS. Specifically, the general purpose of Study 1, to be described in greater detail immediately below, was to develop the SBIS and test its validity and reliability.

Subsequently, it was the general purpose of Study 2 to apply the SBIS to relatively large samples of young CWS and CWNS and determine possible differences in BI between CWS and CWNS as well as possible associations between CWS's BI and measures of stuttering (e.g., stuttering frequency and severity). Further elaboration of Study 2 hypotheses will be described in detail below after presentation of Study 1 and its findings.

2. Study 1: Development and validation of SBIS

2.1. Purpose

As mentioned above, it was the purpose of Study 1 to construct and validate the SBIS. The development of the SBIS was motivated, in part, by the fact that other parent-report questionnaires, such as the aforementioned BSQ and CBQ, constitute more global assessments of temperament. As such, tests like the BSQ and CBQ, which are excellent in their own right, were not designed to measure BI as described and operationalized by Kagan and his colleagues. Thus, the development of a short, easy-to-score test that is specifically focused on the construct of BI, taking the form of a questionnaire such as the

SBIS, was deemed important to further investigate the possible association between BI and childhood stuttering.

2.2. Generation of questionnaire items

The SBIS was initially developed by the second author (Oyler, 1996a), based on Kagan and colleague's research on the characteristics of BI in young children (Kagan, 1984; Kagan, Reznick, & Snidman, 1987). Oyler's review of Kagan's work yielded 41 words/phrases describing BI (e.g., retreats immediately, hesitant to talk to people, stays close to parent) and the descriptors were then categorized in seven groups according to their similarity. For example, descriptors grouped in the category "withdraws" were "retreats immediately," "withdraws," "retreats," and "avoids contact with novel objects." Subsequently, a statement that depicted the descriptors was written for each category with the resulting seven statements being randomly allocated within the initial 7-item SBIS.

Next, the questionnaire was given to 10 parents of normally fluent preschool-age children (i.e., two males and three females with a minimum of an undergraduate degree, and one male and four females with a minimum of a master's degree in Education). These parents were asked to (a) rate the questionnaire items in terms of "readability and clarity" using a 5-point scale (1 = not liked, 2 = somewhat liked, 3 = average/okay, 4 = liked, 5 = liked very much), and (b) circle words/phrases that could be improved upon and write individual suggested revisions. The second author made minor changes in the wording of the items according to raters' feedback and produced the draft version of SBIS which contained seven items. The 7-item SBIS evolved to the current 5-item one through the analyses described below.

2.3. Participants

To evaluate the structure of the scale and its internal consistency, the 7-item SBIS was given to the parents of 243 CWNS and 225 CWS between 2;10 and 6;3 years of age with a mean of 49.15 months ($SD = 9.84$). Other psychometric properties (i.e., test-retest reliability and validity) were assessed with subsamples, not the full sample. All participants were paid volunteers naïve to the purposes and methods of the study and were part of an ongoing series of empirical studies (e.g., Choi et al., 2013, 2016; Clark, Conture, Walden, & Lambert, 2015; Coulter, Anderson, & Conture, 2009; Ntourou, Conture, & Walden, 2013) examining the relation between speech-language processes, emotional variables, and developmental stuttering in young children. The protocol was approved by the Institution Review Board at Vanderbilt University, Nashville, Tennessee. For each participant, parents signed informed consent, and children gave assent.

Requirements for inclusion in the study were that children score at or above the 16th percentile rank on one or more of the following norm-referenced speech and language tests: (a) the *Peabody Picture Vocabulary Test* (PPVT-III or PPVT-IV; Dunn & Dunn, 1997, 2007), (b) the *Expressive Vocabulary Test* (EVT or EVT-2; Williams, 1997, 2007), (c) the *Test of Early Language Development - Third Edition* (TELD-3; Hresko, Reid, & Hamill, 1999), (d) and the “Sounds in Words” subtest of the *Goldman–Fristoe Test of Articulation* (GFTA or GFTA-2; Goldman & Fristoe, 1986, 2000). Also participants were monolingual native speakers of American English, and had no known or reported history of neurological, hearing, developmental, attentional, emotional, academic, and/or intellectual problems.

A child was considered a CWS if he/she(a) exhibited three or more stuttered disfluencies (sound/syllable repetitions, monosyllabic whole-word repetitions¹, and sound prolongations) per 100 words of conversational speech (based on a 300-word conversational sample during a child-examiner play interaction) (Conture, 2001), and (b) a total overall score of 11 or above (a severity equivalent of at least “mild” for preschool children) on the *Stuttering Severity Instrument* (SSI-3 or SSI-4; Riley, 1994, 2009).

A child was considered a CWNS, if he/she (a) exhibited two or fewer stuttered disfluencies per 100 words of conversational speech (based on a 300-word conversational sample obtained through child-examiner play interaction), and (b) received a total overall score of 10 or below (a severity equivalent of less than “mild” for preschool children) on the SSI-3.

2.4. Results

2.4.1. Questionnaire Structure – Factor Analysis

The total number of participants ($N = 468$) was divided into two samples ($n = 234$, $n = 234$) that were used for *exploratory* and *confirmatory* factor analyses respectively. The two samples comprised comparable numbers of CWS and CWNS participants and comparable boy to girl ratios. The participants in the *exploratory* analysis sample were 112 CWS (30 girls, 82 boys), and 122 CWNS (55 girls, 67 boys).

¹ According to the SSI-3 manual (Riley, 1994, p. 4) “Repetition of one-syllable words may be stuttering if the word sounds abnormal (shortened, prolonged, staccato, tense, etc.); however, when these single-syllable words are repeated but are otherwise spoken normally, they do not qualify as stuttering using the definition just stated” (Riley, 1994, p. 4). Thus, only perceptually “abnormal (shortened, prolonged, staccato, tense, etc.)” single-syllable whole-word repetitions were counted as stuttered disfluencies. In the present study, perceptually effortless, non-tense repetitions of single-syllable whole words—such as those produced for emphasis (e.g., the child says, “it was a big, big dog,” while gesturing how large the dog was)—were not counted as stuttered or nonstuttered disfluencies. Perceptually non-effortful, non-tense repetitions of single-syllable whole words were not counted as stuttered nor as nonstuttered disfluencies and were excluded from the fluency data.

Similarly, the *confirmatory* analysis sample comprised 113 CWS (30 girls, 83 boys), and 121 CWNS (57 girls, 64 boys).

2.4.1.1. Exploratory factor analysis

Exploratory factor analysis was conducted - using principal components as the method of initial factor extraction - to examine the factor structure of the questionnaire (based on the $n = 234$ EFA participants). Inspection of the scree plot (see Figure 1) and the commonly-cited Kaiser-greater-than-1 criterion (Kaiser, 1960), according to which factors with eigenvalues greater than 1 are retained, pointed to a single-factor solution for SBIS.

The single-factor solution was taken to suggest that all questionnaire items can be regarded as assessing a single, relatively homogeneous dimension of BI. Table 1 presents the factor loadings for the seven items of the questionnaire. According to Comrey and Lee (1992), loadings greater than .71 are considered excellent, .63 very good, .55 good, .45 fair, and .32 poor. Thus, as shown in Table 1, the last two items (6 and 7) have poor loadings².

2.4.1.2. Confirmatory factor analysis

Confirmatory factor analysis was conducted to determine the adequacy of the factor structure of the exploratory factor analysis (Brown, 2015; Kline, 2015). To assess

² The structure model of the original 7-item scale was also statistically examined separately for all CWS ($n=225$) and all CWNS ($n=243$). For the CWNS group, results indicated that based on the Kaiser-greater-than-1 criterion the SBIS had a unitary construct (single factor), with the factor loadings of the first five SBIS items ranging from 0.79 to 0.49. Factor loadings for SBIS items 6 and 7 were 0.35 and 0.22, respectively. Likewise, for the CWS group, this additional EFA extracted a single factor with factor loadings for the same first five SBIS items ranging from 0.83 to 0.36. Factor loadings for SBIS items 6 and 7 were 0.27 and 0.21, respectively. To determine the equivalence of the CWS and CWNS factor structures, the authors employed the Tucker's congruence coefficient (ϕ) (Lorenzo-Seva & ten Berge, 2006). Results of the congruence analysis indicated that the two factors (one obtained from CWNS and the other from CWS) were highly similar or functionally equivalent ($\phi = 0.994$).

model fit, the chi-square test statistic (χ^2), the standardized root mean squared residual (SRMR), the Tucker–Lewis index (TLI), the comparative fit index (CFI), and the root mean square error of approximation (RMSEA) were used (based on the $n = 234$ CFA participants) (Jackson, Gillaspay, & Purc-Stephenson, 2009).

Results from all these indices revealed that the model with only the first five items loading on a single factor ($\chi^2 = 3.32$, $df = 4$, $p = .51$; SRMR = .02; TLI = 1.00; CFI = 1.00; RMSEA = 0.00) provided a better fit to the data than the alternative model in which all seven items load on a single factor ($\chi^2 = 43.48$, $df = 14$, $p < .001$; SRMR = .06; TLI = .90; CFI = .93; RMSEA = 0.09). Furthermore, the Cronbach's alpha coefficient of internal consistency of the 5-item questionnaire was .81, slightly higher than the .76 reliability coefficient of the 7-item questionnaire.

Results from both exploratory and confirmatory factor analyses supported the use of a shorter 5-item SBIS version. Thus, subsequent analyses in both Study 1 (i.e., psychometric properties) and Study 2 are based on the 5-item rather than the initial 7-item questionnaire.

2.4.2. Psychometric properties

2.4.2.1. Internal Consistency and Reliability

The Cronbach's alpha coefficient of internal consistency based on the entire sample ($N = 468$) was .80. Given that behavioral inhibition is considered to be a temperamental trait, it is expected to be relatively stable over time (e.g., Asendorpf, 1994; Kagan, Reznick, & Snidman, 1988). Thus, an index of behavioral inhibition should possess adequate temporal stability (or test-retest reliability). To assess the test-retest reliability of the SBIS, parents of 76 (46 CWNS, 30 CWS) out of the 468 participants

completed the SBIS questionnaires at two time points separated by approximately 8 months ($M = 8.28$, $SD = 0.79$).

Results based on the Pearson's r and the intraclass correlation coefficient (two-way random effects absolute agreement model) (McGraw & Wong, 1996; Shrout & Fleiss, 1979) for test-retest reliability were $.79$ ($p < .001$) and $.88$ ($p < .001$), respectively. Also difference in scores between the first time point ($M = 18.54$, $SD = 3.77$) and second time point ($M = 19.03$, $SD = 3.98$) was not statistically significant ($p = .10$). Thus, results of *reliability* measurement support the *internal consistency* and *test-retest reliability* of the SBIS.

2.4.2.2. Validity

2.4.2.2.1. Concurrent validity

To examine concurrent validity of the 5-item SBIS questionnaire, SBIS data of 53 participants were compared with indicators of BI obtained from direct behavioral observation by Choi and her colleagues (Choi, Conture, Walden, Lambert, & Tumanova, 2013). As described above, Choi et al. used a method developed by Kagan and colleagues, researchers who introduced the term BI, which involves the measurement of the latency to children's 6th spontaneous comment during an interaction with unfamiliar adults in a laboratory setting. The selection of this variable as an index of BI is based on prior research suggesting that it successfully discriminates between children high and children low in BI (e.g., Kagan, Reznick, & Gibbons, 1989; Kagan, Reznick, Snidman, Gibbons, & Johnson, 1988).

Results of concurrent validity measurement indicated a significant negative correlation between the latency to the 6th spontaneous comment and the present study's

total SBIS score (*Spearman's rho* = $-.31$, $p = .022$). Such results indicate that participants who exhibit longer latencies exhibit lower SBIS scores (with lower SBIS scores indicating higher BI). Thus, this finding appears to support the *concurrent* validity of the SBIS questionnaire.

2.4.2.2.2. Convergent validity

To assess convergent validity the SBIS data of 391 participants were correlated with their score on the subscale *approach/withdrawal* of the *Behavioral Style Questionnaire* (BSQ; McDevitt & Carey, 1978), a scale that has been shown to be related to BI (Izard, Schultz, Fine, Youngstrom, Ackerman, 1999-2000). Parents of these participants completed the BSQ after the SBIS during their visit at the research lab. As described by Anderson, Pellowski, Conture, and Kelly (2003), *approach/withdrawal* refers to the nature of initial responses to new stimuli—people, situations, places, foods, toys, etc. The BSQ's internal consistency (0.80) and the test-retest reliability (0.94) of the BSQ's *approach/withdrawal* subscale are good (Carey, 1997, p.13). High scores on the *approach/withdrawal* scale indicate reluctance to approach novel situations and/or people.

Results of *convergent* validity measurement indicated significant negative correlations between total SBIS score and BSQ *approach/withdrawal* ($r = -.68$, $p < .001$). Thus, high scores on *approach/withdrawal* indicating children's tendency to withdraw were related to low scores at the SBIS, with the latter indicating higher BI. These findings support the *convergent* validity of the SBIS questionnaire.

2.4.2.2.3. Divergent validity

Divergent validity refers to the absence of an association between constructs that should be theoretically unrelated (DeVellis, 2012). To assess SBIS's divergent validity the SBIS data of 391 participants were correlated with their score on the subscale *persistence* of the BSQ, a subscale that relates to the child's ability to continue an activity in the face of obstacles and distractions.

Results of *divergent* validity measurement indicated a lack of relationship between BSQ and SBIS ($r = -.02, p = .76$) supporting the *divergent* validity of the SBIS.

2.5. Discussion

The purpose of Study 1 was to develop a brief, simple to administer, and reasonably easy to interpret parent-report BI questionnaire for young CWS and CWNS with satisfactory psychometric properties. This led to the 5-item SBIS. As described above, data obtained from administration of the SBIS to a large number of preschool-age children were subjected to several analyses to determine the SBIS's psychometric properties.

First, exploratory and confirmatory factor analyses supported a single-factor model, indicating a uniform construct underlying the questionnaire. Second, to determine the *reliability* of SBIS internal consistency and test-retest reliability were assessed. Internal consistency refers to the general agreement between the items that purport to measure the same construct and the composite score. High internal consistency, usually expressed in Cronbach's alpha (α), is essential because it "speaks directly to the ability of the clinician or the researcher to interpret the composite score as a reflection of the test items" (Henson, 2001, p. 178).

Cronbach's α is regarded satisfactory if the value is greater than .7 (Nunnally, 1978; Peterson, 1994) and its value greatly depends on the number of items in the scale/test (Lord & Novick, 1968). Cortina (1993) demonstrated that increasing the number of items of a scale from 6 to 18 resulted in an increase of α from .72 to .88. Thus, the α (.80) for the 5-item SBIS appears acceptable. Likewise, the SBIS test-retest reliability appeared to be satisfactory in the degree of stability over an 8-month span, as indexed by high Pearson's r (.79) and intra-class correlation coefficient (.88) (Cicchetti, 1994).

Finally, the *validity* of the SBIS, or the degree to which it measures what it is designed to measure, was determined by how strongly SBIS scores were correlated with direct observation of BI behavior in children. Specifically, children who took longer (after controlling for the influence of duration of instances of stuttering and unintelligible utterances were removed from the epoch during which latency was measured) to produce their 6th spontaneous comment during an interaction with unfamiliar adults in an experimental setting—one index of BI (Kagan, Snidman, & Arcus, 1998)—had lower SBIS scores. This relation was statistically significant but of moderate magnitude (Cohen, 1988). The size of this relation could be partially attributed to the relatively small sample size ($n = 53$). However, considering the moderate correlations reported by various studies that compare parent-report and direct behavioral observations (Ponitz, McClelland, Matthews, & Morrison, 2009; Smith-Donald, Raver, Hayes, & Richardson, 2007), the moderate correlation between SBIS scores and Kagan's index of BI appears typical for such analyses. Furthermore, the validity of SBIS was supported by significant correlation between SBIS scores and parental ratings of withdrawal.

Overall, the SBIS seems to function well for a brief parent report measure. Thus, it would seem to be appropriate to consider for use in both clinical and research settings. One of its strengths is that it is based on a relatively large sample size of both CWS and CWNS in addition to the fact that it appears to exhibit acceptable psychometric properties. The convergent validity of the SBIS, however, might have been further strengthened if parents had been asked to complete an additional BI questionnaire, such as the *Behavior Inhibition Questionnaire* (BIQ; Bishop, Spence, & McDonald, 2003). Although the BIQ's psychometrics appear robust, it includes items related to "talking," which the SBIS does not, suggesting the possibility that BIQ findings – if applied to or used with CWS - may be influenced by the children's stuttering and/or other speech-language difficulties. Whatever the case, the relative abilities of the BIQ and SBIS to index BI in young CWS and their CWNS peers remains unclear and must await further empirical study.

3. Study 2: Application of SBIS to the study of BI relative to childhood stuttering

3.1. Purpose

The purpose of Study 2 was to employ the SBIS to determine between-group differences (CWS vs. CWNS) in BI and associations between CWS's BI and their stuttering frequency, stuttering severity, speech-related communication attitudes, and stuttering-related consequences. Specifically, Study 2 tested the following four hypotheses:

Hypothesis 1: CWS, when compared to CWNS, have higher BI as indexed by lower mean SBIS score, as well as greater numbers of CWS than CWNS in the high BI group (lower 15% of the SBIS distribution).

Hypothesis 2: CWS's BI is significantly related to their frequency of stuttered disfluencies and stuttering severity as measured by the *Stuttering Severity Instrument* (SSI-3 or SSI-4; Riley, 1994, 2009)

Hypothesis 3: CWS's BI is significantly related to their self-reported communication attitudes as measured by the *Communication Attitude Test for Preschool and Kindergarten Children Who Stutter* (KiddyCAT; Vanryckeghem & Brutten, 2007)

Hypothesis 4: CWS's BI is significantly related to their reaction to their stuttering as measured/indexed by the *Disfluency-Related Consequences parent rating scale* from the *Test of Childhood Stuttering* (TOCS; Gillam, Logan, & Pearson, 2009).

3.2. Participants

Study 2 included participants from Study 1 who had completed and scored at or above the 16th percentile rank on all four speech-language tests (TELD, PPVT, EVT, GFTA) and who had not been part of the only other known study that examined the role of BI in childhood stuttering (Choi, Conture, Walden, Lambert, & Tumanova, 2013). Thus, participants in Study 2 were 179 preschool-age CWS (49 girls, 130 boys) and 198 preschool-age CWNS (91 girls, 107 boys) between 3;0 and 6;3 years of age with a mean of 47.54 months ($SD = 9.40$) for CWS and 51.01 months ($SD = 10.24$) for CWNS. Table 2 presents speech, language and fluency characteristics (i.e., Mean [M], Standard Deviation [SD]) of the two talker groups (i.e., CWS and CWNS).

Racial information was available for 131 CWS (73.18%) and 130 CWNS (65.65%). Most CWS participants were identified by their parents as White (78.62%), followed by Black/African American (15.27%), multiracial (5.34%) and Asian (0.76%).

Similarly, 85.38% CWNS were White, 10% Black/African American, 3.85% multiracial, and 0.77% American Indian.

The Hollingshead Four-Factor Index of Social Position (Hollingshead, 1975) was used to describe participants' family's socioeconomic status (SES). This index takes into account both parents' educational level, occupation, and marital status. Computed SES scores range from 8 to 66, with a higher score indicating higher SES status. Of the 377 total participants, SES information was available for 147 CWS and 150 CWNS (78.79% of the total participants). For this subset of the total participants, CWS had a mean social position score of 45.34 ($SD = 10.88$) and CWNS had a mean of 45.58 ($SD = 11.01$), a non-significant difference, $F(1, 295) = .04, p = .846$.

The parent(s) of all participants filled out the SBIS questionnaire. However, KiddyCAT (Vanryckeghem & Brutton, 2007) and TOCS *Disfluency-Related Consequences* (Gillam, Logan, & Pearson, 2009) data were available from a subset of CWS participants, 91 (50.84% of total; 25 girls, 66 boys) and 45 (25.14% of total; 12 girls, 33 boys) respectively.

3.3. Dependent variables

3.3.1. Behavioral inhibition

Behavioral inhibition was measured by means of the *Short Behavioral Inhibition Scale* (SBIS), a brief, parent-report questionnaire of BI for preschool-age children (see Study 1 on SBIS's development and validation). The SBIS was completed by the participants' parent(s) in the presence of a research assistant, usually a speech-language pathologist, who was available to answer any questions about the statements on the questionnaire. This assessment tool consists of five items assessing children's inhibited

behaviors such as withdrawal from unfamiliar people and responses to novel situations (see Appendix for the 5-item SBIS form). Each item presents both ends of the assessed behavior continuum (e.g., approach vs. withdrawal) and is scored on a 5-point Likert type scale. Thus possible scores on the 5-item SBIS range from 5 to 25 with lower scores indicating higher BI.

3.3.2. *Stuttering frequency*

Stuttering frequency was measured by means of a conversational sample of 300 words elicited from each participant (both the CWS and CWNS participants). This sample was obtained during an approximately 15-30 minutes loosely structured play-based interaction between each participant and a researcher trained in eliciting conversational samples and “on-line” speech disfluency counts. Disfluency and word count data were recorded in real-time on the Vanderbilt University’s Disfluency Count Sheet (Conture, 2001).

Each participant’s frequency of stuttered disfluencies (SD) was calculated by dividing the total number of stuttered disfluencies by the total number of words spoken (300 words). The types of speech disfluencies considered by the present authors as SDs are described above as well as in detail elsewhere (e.g., Clark, Conture, Walden, & Lambert, 2013; Tumanova, Conture, Lambert, & Walden, 2014). Intraclass correlation coefficients using the absolute agreement criterion, were computed to assess interjudge agreement for the measurement of SDs. Four trained coders independently identified SDs for 33 participants from video-recorded speech samples. The average measure of intraclass correlation coefficients for identification for SDs was .99, $p < .001$, a value far exceeding the popular criterion of .70 (Yoder & Symons, 2010).

3.3.3. Stuttering severity

Stuttering severity was measured by means of the total SSI-3 or SSI-4 score (Riley, 1994, 2009). The total SSI-3/SSI-4 total score is based on information from three components: (a) stuttering frequency, (b) duration of the three longest stuttering events, and (c) rating of observed physical concomitants.

3.3.4. Speech-related communication attitude

Participants' (i.e., children's) speech-associated attitudes were measured by the KiddyCAT. The Kiddy-CAT was administered to 91 CWS by an adult examiner (Vanryckeghem & Brutten, 2007). This self-report, 12-item, binary (yes/no) scale determines preschool-age CWS's communication attitudes towards speech difficulty (Clark, Conture, Frankel, & Walden, 2012). Scores range from 0 to 12, with higher scores indicating more negative attitudes towards their speech.

3.3.5. Stuttering-related consequences

To assess whether BI is associated with stuttering-related consequences, the parents of 45 CWS parents completed the *Disfluency-Related Consequences* rating scale from the TOCS (Gillam, Logan, & Pearson, 2009). The developers of this scale describe it as a scale comprised of "nine statements about the ways in which the child copes with and reacts to speech disfluency, as well as the ways in which others react to the child's disfluent speech." (Gillam et al., 2009, p.62). All but 1 of the 9 *Disfluency-Related Consequences* statements refer to the extent to which the child who stutters exhibits associated secondary behaviors (i.e., avoidance behaviors) and negative emotions in reaction to stuttering. Only the last of the nine statements focuses on others' reactions to the child's stuttering. The internal consistency of the *Disfluency-Related Consequences*

rating scale for preschool-age children is .85 and scores range from 0 to 27, with higher scores indicating greater stuttering-related consequences.

3.4.Results

3.4.1. *Descriptive characteristics of the two talker groups*

Although all participants presented with language abilities within the typical range, a multivariate analysis of variance (MANOVA) indicated significant between-group (i.e., CWS vs. CWNS) differences at the Bonferroni-adjusted alpha level of 0.012, on all language measures: PPVT, $F(1,370) = 12.85, p < .001$; EVT, $F(1,370) = 6.89, p = .009$; and TELD, $F(1,370) = 6.41, p = .012$. In contrast, the between-group difference in GFTA scores was not statistically significant, $F(1,370) = .73, p = .389$. The two talker groups also differed significantly in age ($z = -3.44, p < .001$) and results of a chi-square test indicated a significant difference in gender between CWS and CWNS, $\chi^2 = 13.91, df=1, p < .001$.

3.4.2. *Hypothesis 1: Differences in BI between CWS and CWNS*

3.4.2.1. *Mean differences analyses*

Prior to inferential statistical analyses of possible differences in BI between young CWS and their CWNS peers (Hypothesis 1), normality of the SBIS data was verified by visually inspecting the distribution of the standardized residuals. Also the Levene's test revealed that the assumption of homogeneity of variance was met, $F(1, 375) = 1.57, p = 0.210$. Furthermore, correlational analyses were conducted to examine the relation between the main dependent measure (SBIS score) and different talker group descriptive characteristics (chronological age, SES, vocabulary skills, language skills) to identify potential covariates for inclusion in the analysis of variance. As shown in Table 3, none

of the descriptive characteristics were significantly related to SBIS scores so they were not included in subsequent analyses. To determine overall between-group (CWS vs. CWNS) differences in BI (as measured by their mean SBIS score), a generalized linear model (GLM) with talker group (CWS, CWNS) and gender (male, female) as fixed factors was constructed. Effect sizes are reported as partial eta squared (η_p^2), with values close to .01, .06, and .14 being interpreted as small, medium, and large in magnitude respectively (Cohen, 1988).

Findings supported Hypothesis 1 (see Figure 2). Specifically, results indicated that CWNS and CWS differed significantly in mean SBIS scores (CWS: *estimated marginal mean, EMM* = 17.04, *standard error, SE* = 0.34, CWNS: *EMM* = 18.43, *SE* = 0.29), $F(1, 373) = 9.58, p = .002, \eta_p^2 = .02$. Also, for the combined CWS and CWNS SBIS scores, girls' mean SBIS score (*EMM* = 17.26, *SE* = 0.36) was significantly lower than that of boys (*EMM* = 18.21, *SE* = 0.27), $F(1, 373) = 4.43, p = .036, \eta_p^2 = .01$. However, no interaction effect of gender and talker group, $F(1, 373) = .42, p = .519$ was confirmed.

3.4.2.2. Extreme scores analysis regarding Hypothesis 1

Our further analyses regarding Hypothesis 1 were consistent with analytical methodology suggested by Kagan et al. (1989) and followed by others (e.g., Gest, 1997; Kertes et al., 2009), who empirically studied children at the extremes of BI positing that the BI temperament style is manifested more clearly in the extremes. Specifically, the present authors employed a chi-square test analysis to assess whether there was a relation between talker group and *high* (lower 15% of the SBIS score distribution, SBIS score equal to and/or lower than 13) versus *low* BI groups (upper 15% of the SBIS score

distribution, SBIS score equal to and/or higher than 22). In essence, where there more CWS than CWNS in the high BI group. As shown in Table 4, results indicated that there were more CWS with high BI and fewer CWS with low BI when compared to their CWNS peers, $\chi^2 = 7.69$, $df = 1$, $p = .006$.

3.4.3. Hypothesis 2: Relation between CWS's BI and their stuttering frequency and stuttering severity

To determine the possible relation between CWS's BI (as indexed by their SBIS scores) and their stuttering frequency and severity, two separate GLM models were used. For both models, frequency of stuttered disfluencies (stuttering frequency) and SSI-3/SSI-4 score (stuttering severity) were the dependent variables and SBIS score the predictor variable. Given that measures of stuttering frequency and severity follow a non-normal distribution (Jones, Onslow, Packman, & Gebski, 2006; Tumanova, Conture, Lambert, & Walden, 2014), the GLM analyses used an identity link function to fit stuttering frequency and severity to a gamma distribution.

Findings supported Hypothesis 2. Specifically, results indicated that CWS's SBIS score was a significant predictor of CWS's stuttering frequency (Wald $\chi^2 = 28.74$, $df = 18$, $p = .049$) and stuttering severity (Wald $\chi^2 = 38.84$, $df = 18$, $p = .003$). Thus, CWS with higher BI, as indexed by the SBIS, exhibited greater stuttering frequency and severity during a child-examiner conversation.

3.4.4. Hypothesis 3: Relation between CWS's BI and their attitudes towards communication

To determine the possible relation between CWS's BI (as indexed by their SBIS score) and their communication attitudes (as indexed by their Kiddy-CAT score), a GLM

with KiddyCAT as the dependent measure, SBIS as the predictor variable, and SSI-3/SSI-4 as a covariate to control for the effect of stuttering severity was employed. Results indicated that SBIS was not a predictor of CWS's KiddyCAT scores, Wald $\chi^2 = 19.76$, $df = 16$, $p = .231$.

However, the above Kiddy-CAT findings, the present authors post-hoc hypothesized, might have been influenced by the fact that chronological age impacts CWS's KiddyCAT scores (Groner, Walden, & Jones, 2016; Guttormsen et al., 2015) and that young CWS's awareness of stuttering, while present in *some* 3-year-old CWS, dramatically rises between 4- and 5 years of age (Ambrose & Yairi, 1994; Ezrati-Vinacourf, Platzky, & Yairi, 2001). To test this post-hoc hypothesis – that chronological age impacts the relation between CWS's BI (i.e., SBIS scores) and their communication attitudes (i.e., Kiddy-CAT scores) - the present authors analyzed the relation between CWS's BI and communication attitudes for CWS older than 4 years of age ($n = 42$) and CWS younger than 4 years of age ($n = 49$) separately, while controlling for the effect of stuttering severity.

This post-hoc hypothesis was confirmed, Specifically, results indicated that SBIS was a predictor of KiddyCAT scores for CWS between 4;0 and 6;3 years of age, Wald $\chi^2 = 27.74$, $df = 14$, $p = .015$, but, not for 3 year-old CWS, Wald $\chi^2 = 23.62$, $df = 15$, $p = .072$. Thus, the initial finding that BI was not significantly related to Kiddy-CAT scores appears to have been driven by the 3 year-old CWS participants. In short, Hypothesis 3 was partially supported, in that BI is related to CWS's expressed attitudes toward speech/communication, but only for preschool-age CWS older than 4 years of age.

3.4.5. *Hypothesis 4: Relation between CWS's BI and their stuttering-related consequences.*

To determine the possible relation between CWS's BI (as indexed by their SBIS), and their reaction to their stuttering (as indexed by their parent's TOCS Disfluency-Related Consequence scale), a GLM model was employed. For this model, TOCS Disfluency-Related Consequence scale was the dependent measure, SBIS the predictor variable, and the overall SSI-3/SSI-4 score a covariate to control for the effect of stuttering severity. Given the non-normal distribution of the dependent measure (Tumanova, Choi, Conture, & Walden, 2018), the GLM analysis used a log link function to fit the dependent measure to a gamma distribution.

Findings supported Hypothesis 4. Specifically, results indicated that SBIS was a significant predictor of CWS's TOCS Disfluency-Related Consequence scale score, Wald $\chi^2 = 32.92$, $df = 14$, $p = .003$. In other words, CWS with high, when compared to those with low, BI exhibited a higher score on the TOCS Disfluency-Related Consequence scale. Thus, BI seems to be associated with disfluency-related consequences in preschool-age CWS.

4. Discussion

4.1. Difference in BI between CWS and CWNS (Hypothesis 1)

Our first hypothesis was confirmed (i.e., CWS exhibit greater mean and extreme BI tendencies than CWNS). In other words, CWS, as a group, based on their SBIS scores, appeared more behaviorally inhibited than their typically fluent peers.

Specifically, CWS presented with significantly lower mean SBIS scores (i.e., greater BI tendencies) than their CWNS peers, with an associated small effect size. Furthermore,

there were significantly more CWS, than CWNS in the high BI group. There are at least two possible interpretations of these findings, as will be discussed immediately below.

On the one hand, the effect size of the difference in mean SBIS between CWS and CWNS was small. This implies some degree of overlap between CWS and CWNS, even though the mean SBIS scores between these talker groups was significantly different. This overlap in CWS and CWNS's SBIS distributions impacts the strength to which one might suggest that BI is the sole province of childhood stuttering. Such a suggestion, the authors would note, has not been made in the present article.

On the other hand, even though the effect size of the between-group difference in mean SBIS was small, there were significantly more CWS with high BI and fewer CWS with low BI than CWNS. This finding is consistent with that of Choi and colleagues (2013). And, it will be recalled, there was no overlap in participants between the Choi et al and the present study.

Given that CWS exhibited significantly greater mean and extreme BI tendencies than CWNS, small effect size notwithstanding, it is interesting to consider the mounting evidence that older children and adults who stutter are at a higher risk for (social) anxiety disorders than children/adolescents and adults who do not stutter (e.g., Blood, Blood, Maloney, Meyer, & 2007; Craig & Tran, 2014; Iverach et al., 2016; McAllister, Kelman, & Millard, 2015; Mulcahy, Hennessey, Beilby, & Byrnes, 2008; Smith, Iverach, O'Brian, Kefalianos, & Reilly, 2014). Is early childhood behavioral inhibition related to later childhood/adolescence/adulthood social anxiety in individuals who stutter? The present authors certainly cannot answer to this question, but some cautious discussion appears warranted.

Behavioral inhibition's moderate stability from childhood to adulthood (Degnan & Fox, 2007) raises the possibility that some young CWS likely maintain this behavioral tendency as they grow older, a tendency that has been identified as a prominent risk factor for developing (social) anxiety (Clauss & Blackford, 2012; Fox, Henderson, Marshall, Nichols, & Ghera, 2005), into later childhood and beyond. Therefore, it could be suggested, that anxiety is not solely a consequence of stuttering (e.g., Craig & Tran, 2006; Reilly et al., 2009), but also a possible contributing antecedent to aspects of the disorder such as social anxiety (given that children's temperament is present from birth). Although this hypothesis is plausible, empirical testing to support or refute such speculation must await future investigations.

Investigations of such speculation would seem to have potential for increasing our understanding of the role of emotions in stuttering by systematically studying associations between more "dispositional" (i.e., BI) and "environmental" (e.g., peer relations) variables. For example, one possible line of investigation might involve the study of both trait-like/dispositional variables of emotional development (e.g., BI), emotional processes (e.g., attentional bias to threat) and their association/interaction with environmental variables (e.g., peer relations). Specifically, whether these trait-like and environmental variables, singularly or in combination, impact the developmental trajectories of overt (e.g., stuttered syllables) and covert (e.g., avoidance behaviors) stuttering behaviors. Understanding the dynamic interplay over time of emotional processes, environmental factors, and stuttering behaviors would not only shed light on the developmental trajectories of childhood stuttering, but also on the role of emotional variables in long-term outcomes in CWS.

4.2. BI and stuttering frequency and severity for CWS (Hypothesis 2)

Our second hypothesis was confirmed (i.e., BI is associated with stuttering frequency and stuttering severity). Present findings regarding Hypothesis 2 are consistent with Choi and colleagues (2013) report that CWS with high, when compared to CWS with low BI exhibit greater stuttering frequency. The present study's finding of a relation between BI and stuttering frequency appears consistent with some psycholinguistic models of stuttering, for example, the *vicious cycle hypothesis* (Arnstein, Lakey, Compton, & Kleinow, 2011; Bernstein Ratner & Wijnen, 2006; Vasic & Wijnen, 2005). Such speculation suggests that speech disfluencies result from a hypervigilant/hyperfunctioning monitoring system of the internal speech plan (Civier, Tasko, & Guenther, 2010; Lahat et al., 2014). So, although, it may be argued, too *little* monitoring of the internal speech plan may be problematic, so might too *much* monitoring, with the ideal degree of monitoring, or "sweet spot," perhaps residing between these two extremes. Interestingly, there is empirical support for the notion that emotion influences the internal speech plan. Specifically, Van Lieshout, Ben-David, Lipski, and Nmasivayam's (2014) found that emotional stress impacted both the preparatory and the execution stages of speech motor control in adults who stutter. These speculations and empirical evidence are intriguing, but must await further investigations to determine their precise contributions to childhood stuttering.

Interestingly, empirical evidence in the field of psychology suggests that children high in BI are hypervigilant and present with enhanced error monitoring (McDermott et al., 2009). Thus, given the apparent confluence among present findings, available theories regarding stuttering, and findings from psychology, it may be suggested that CWS higher

in BI, compared to those with lower in BI, exhibit greater frequency of disfluencies due to their tendencies towards higher error monitoring and lower threshold for detection of errors. If true, such monitoring and detection characteristics may contribute to more frequent interruptions and repairs of encoding errors, with such interruptions and repairs manifesting themselves as speech disfluencies.

With regard to stuttering severity, the aforementioned relation between BI and stuttering frequency may partly account for the significant correlation between BI and stuttering severity. Perhaps, one might speculate, this significant correlation between BI and stuttering severity suggest that BI is associated with a third variable, physical tension, with increases in physical tension contributing to more frequent, longer speech disfluencies and associated non-speech behaviors. Consistent with such speculation, Kagan, Reznick, and Snidman (1987) reported that children high in BI exhibit increased physical tension, particularly in laryngeal muscles, when they speak in unfamiliar or threatening situations. Perhaps, CWS with greater BI tendencies exhibit increased physical tension when talking with an unfamiliar adult (e.g., the experimenter). Consequently, such increased muscle tension may contribute to longer *duration* of stuttered disfluencies and/or greater physical tension leading to greater stuttering severity. Consistent with such speculation, Kazenski et al. (2014) reported that social-communicative challenging speaking conditions were associated with increased laryngeal tension for CWS with more severe stuttering (with the latter, based on present findings, associated with greater BI tendencies).

4.3. BI and communication attitudes for CWS (Hypothesis 3)

Our third hypothesis was partially confirmed (i.e., BI is related to CWS' self-reported communication attitudes, but only for CWS older than 4 years of age). The lack of association between KiddyCAT and SBIS in 3 year-old CWS might indicate that in the early preschool-age years BI does not impact CWS's perceptions/attitudes towards their communication. Rather, it is only in later years as stuttering awareness increases, that temperament may color, condition, impact or shape CWS's communication attitudes. Alternatively, one could argue that not many 3 year-old CWS are aware of their stuttering which renders the assessment of the relation between BI and communication attitudes in that age group problematic.

In older preschool-age CWS, BI was found to be related to self-reported communication attitudes, with CWS high in BI exhibiting more negative attitudes towards their speech and communication abilities than CWS low in BI. Consistent with this finding, greater degree of introversion is significantly correlated with greater negative communication attitudes in adults who stutter (Stipdonk, Liefink, Bouwen, & Wijnen, 2014).

Although considerable evidence from studies conducted in North America and Europe (Clark, Conture, Frankel, & Walden, 2012; Novšak Brce & Vanryckeghem, 2017; Ntourou, Marousos, Paphiti, Furlas, & Vanryckeghem, 2016; Vanryckeghem, Brutten, & Hernandez, 2005; Vanryckeghem, De Niels, & Vanrobaeys, 2015; Węsierska & Vanryckeghem, 2015) indicate that preschool-age CWS exhibit greater negative communication attitudes than their fluent peers, it is unclear what biological or constitutional (if any) factors contribute to the development of such negative communication attitudes in young CWS. To the present authors' knowledge, the current

study is the first to document the role of an apparent biological/constitutional-related characteristic/attribute (i.e., BI) associated with negative communication attitudes in young CWS.

To provide some support for how BI may impact negative communication attitudes in older preschool-age CWS, the authors would like to cite Escalona's (1968) notion of *effective experience*. According to this idea, "events in children's lives are experienced only as they are filtered through the individual child's nervous system, so that an environmental event is not the same for all." (Rothbart, 2011, p. 30). This notion suggests that children experience their environment differently due to their unique temperamental/idiosyncratic attributes. Based on present findings, it seems reasonable to suggest, that behaviorally inhibited children, more often than not, confront novel communication situations with avoidance, caution, distress, hesitation, and physiological signs of emotional arousal. These tendencies or proclivities, one may further suggest, provide a "filter" or lens through which more behavioral inhibited children perceive and interpret the quality and valence of their communication and social interactions. Thus, high BI CWS may be more likely to negatively or less than positively evaluate such interactions.

If the above speculation is true, older preschool-age CWS with high BI may experience greater distress and more negative evaluation of their communication and/or social abilities and competence than CWNS and/or CWS with less inhibited temperament. This may be especially the case when dealing with the less than positive consequences of stuttering. For example, from a very young age, typically fluent children have been observed to appraise stuttering negatively and interrupt, ignore, mock,

and walk away from CWS, who are more likely to be “victims of bullying” than CWNS (Davis, Howell, & Cooke, 2002; Ezrati-Vinacour, Platzky, & Yairi, 2001; Langevin, Bortnick, Hammer, & Wiebe, 1998; Langevin, Kleitman, Packman, & Onslow, 2009).

4.4. BI and disfluency-related consequences for CWS (Hypothesis 4)

Our fourth hypothesis was confirmed (i.e., BI is related to CWS’s reaction to their stuttering as measured by parents’ responses to the TOCS Disfluency-Related Consequences scale). Specifically, CWS with high, when compared to low, BI were rated by their parents as displaying stronger reactions to their stuttering. This finding may be taken to suggest that CWS with greater BI tendencies are not only reactive to change/novelty in their environment, one of the hallmarks of individuals with a BI temperament, but they are also reactive to and less tolerant of their speech disfluencies. It may be speculated that such reactions take many forms. For example, such reactions may present as non-speech behaviors (e.g., upper lip raising, Conture & Kelly, 1991) as well as “emotional” reactivity associated with fluency breakdowns, events seeming part of what the TOCS Disfluency-Related Consequences may be measuring.

Interestingly, Tumanova, Choi, Conture, and Walden (2018) empirically studied the association between preschool-age children’s reaction to their stuttering, measured by the same tool (TOCS Disfluency-Related Consequences scale), and their mean length of utterance (MLU) during an unstructured interaction with an unfamiliar examiner. Tumanova et al. reported that children with high scores in TOCS Disfluency-Related Consequences scale exhibited shorter MLU and suggested that this finding may reflect children’s attempt to minimize stuttering by truncating their verbal output. Interestingly, shy/reticent children have been reported to exhibit shorter MLU than talkative children

when interacting with strangers (Van Kleeck & Street, 1982). Thus, an alternative account of Tumanova et al.'s finding (i.e., CWS's shorter MLU related to their greater reaction to stuttering) may be more related to aspects of their CWS's temperament than their attempt to truncate their MLU to be more fluent.

4.5. General Discussion: Theoretical and clinical implications

Present findings are part of the growing body of evidence regarding the possible contributions of temperamental and emotional processes to childhood stuttering. Although a relation between BI and stuttering is confirmed by our study's results (and consistent with Choi et al.'s 2013 findings regarding BI and stuttering), the mechanism/process that underlie this relation remains unclear. Some might propose that BI, in conjunction with other processes (e.g., motoric, speech-language, etc.) contributes to the onset and development of stuttering (i.e., BI \rightarrow stuttering). Still others could view BI and stuttering as bidirectionally influencing each other through complex, dynamic processes (i.e., BI \leftrightarrow stuttering). For example, the production of stuttered disfluencies might trigger, for a hypervigilant, high BI child, behavioral and physiological reactions. These reactions, could, in turn, lead to more frequent and/or severe speech disfluencies. Alternatively the association between BI and stuttering could be explained by a common third-order variable such as enhanced error monitoring (as suggested above), which may contribute to the production of stuttered disfluencies and BI behavioral tendencies.

Although it is challenging to determine which, if any, of these theoretical perspectives best accounts for the relation between BI and stuttering, the finding that BI appears to be associated childhood stuttering for at least some CWS would seem worthy of clinicians' attention. For example, during a diagnostic evaluation of an apparently high

BI CWS, the clinician may want to ensure that ample time is available for the child to warm up so that he/she is reasonably comfortable interacting with the clinician (and gaining a more representative sample of the child's speech and language and hence instances of speech disfluencies). Doing so, the present authors believe, may increase the frequency of more spontaneous comments and decrease the frequency of more brief responses to examiner's questions. Such brief responses, which may be less than representative of the central tendency of the length of the child's utterances. These shorter utterances may, in turn, contribute to a lower frequency of stuttered disfluencies, a frequency that is less than reflective of the child's true central tendency of stuttering frequency (related to such speculation, Sawyer & Yairi, 2006, reported on the impact of sample size on the measurement of speech disfluencies in children).

Although not directly studied in the present study, one might speculate that CWS with high BI tendencies may be more prone than CWS with low BI to react to their own speech disfluencies. Specially, a reaction that exacerbates and/or maintains the frequency and severity of their disfluencies. It may also be speculated that hi BI CWS, when compared to low BI CWS, may be more apt to interpret their speech disfluencies as challenges to their communicative abilities, an interpretation leading them to try to avoid and/or escape their disfluencies. Of course, such speculations are just that and must await future investigations to determine their applicability to the onset and development of childhood stuttering.

Thus, the SBIS and present findings based on its application to childhood stuttering provide a preliminary observational window through which clinicians and researchers may begin to view the possible relation between BI and stuttering in young

children. It remains unclear, of course, whether further viewing will determine if BI meaningfully contributes to a variety of variables possibly associated with childhood stuttering. For example, negative communication attitudes, overly stressful reactions to the negative consequences of stuttering, avoidance/escape behavioral reactions to stuttering, exacerbation of (sub)clinical motoric, speech or language difficulties, and so forth. What is clear, however, is that future empirical investigation of these possible associates of childhood stuttering – regardless whether results are significant or not - will further our understanding of the possible role that emotion plays in the onset and development childhood stuttering. A role that is often discussed but just as often disregarded.

5. Caveats

The principal component analysis indicated that SBIS items related to fear and reactivity had poor loadings and that a model without these items provided a better fit to the data. However, fear and reactivity have some degree of overlap with the construct of BI. Thus, the SBIS is less than a robust instrument for determining fear and reactivity components of BI, a limitation that should be noted by prospective users of SBIS. Also, some users might find the scoring of the test counterintuitive since higher scores indicate lower, rather than higher BI.

Also, present SBIS results are based on parental responses, not unlike some findings resulting from other instruments used to test CWS, for example, the TOCS. Regardless, some clinicians as well as researchers may question the wisdom of relying on parental responses rather than, for example, more direct measurement of BI or BI-related

behaviors like those of Choi et al (2013). Such questions are well taken but must await answers resulting from future research employing more direct measures. For the present, however, we would like to note Henderson and Wachs's (2007) suggestion that although "parent report measures do contain some subjective parental components, available evidence indicates that these measures also contain a substantial objective component that does accurately assess children's individual characteristics." (p. 402).

It should be noted that various speech and language measures differed between CWS and CWNS (e.g., the PPVT), but there was no apparent correlation between these measures and participants' SBIS scores. Perhaps non-standardized measures of speech and language (e.g., syntactic complexity of language production, lexical diversity), rather than scores on standardized tests, may reveal a significant relation to the SBIS measure of BI. Thus, there may be some value, in future research, to employ non-standardized measures of speech and language to determine whether such processes impact SBIS' measure of BI in young children who do and do not stutter.

6. Conclusion

Behavioral Inhibition (BI) is an aspect of temperament characterized by low threshold of limbic arousal and thus relatively intense behavioral and affective reactions to differences, novelty and change. A child exhibiting high BI is more apt to show avoidance, initial restraint, reticence and caution when encountering different, novel or unfamiliar people, situations or stimuli.

To empirically test suggestions in the literature that BI is associated with childhood stuttering, the present authors developed and administered the SBIS, a short (5-item) parent-report questionnaire, to a relatively large sample of young CWS and CWNS

to determine possible between- and within-group differences in BI. Present findings support the reliability as well as validity of the SBIS for the study of BI in young children who do and do not stutter. Such findings also support, based on the SBIS, the notion that preschool-age CWS exhibit greater mean and extreme behavioral inhibition tendencies than their typically fluent peers. In addition, present results provide data-based insights into the relation between young children's BI, stuttering frequency, stuttering severity, and reactions to stuttering. Specifically, CWS with high BI, when compared to children with lower BI, exhibited stronger reaction to their stuttering, and greater stuttering frequency and severity. Also for CWS 4 years of age and older, BI is associated with self-reported attitudes towards communication with CWS with high BI reporting greater negative communication attitudes than CWS with lower BI.

Among various explanations for the present findings, one of seeming interest to both research into and treatment of childhood stuttering, is the possibility that CWS exhibiting BI tend to be more sensitive to environmental change, difference and novelty as well as possibly exhibit a lower threshold for error detection. Such tendencies, in turn, may exacerbate the frequency and severity of CWS's stuttering as well as associated behaviors. Whether further research supports or refutes such an explanation as well as the possibility that BI tendencies impact stuttering frequency, severity and associated behaviors, BI does seem to be related to childhood stuttering, for at least some children. However, at least at present, the precise mechanism/process that underlies this relation remains unclear, with further clarification requiring further empirical study.

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Bios

Katerina Ntourou is an Assistant Professor at the University of Oklahoma Health Sciences Center. Her research interests include the role of executive functioning, emotional processes, and temperament in childhood stuttering.

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Tedra Walden is a Professor of Psychology and Professor of Hearing and Speech Sciences at Vanderbilt University. She is interested in early social and emotional development of young children, particularly those with developmental disabilities such as autism, developmental stuttering, Down syndrome, and other intellectual disabilities.

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Figure Captions

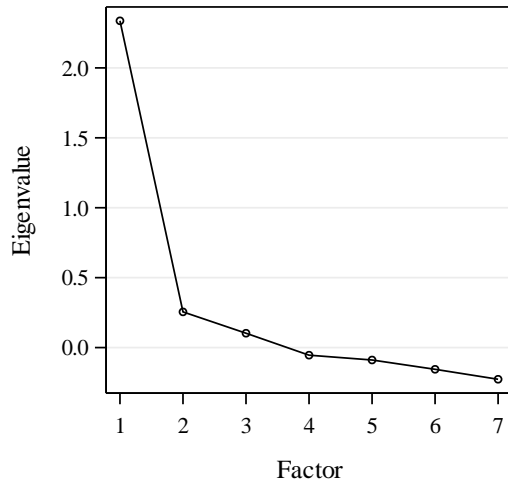


Figure 1: Study 1. Scree Plot for the Exploratory Factor Analysis. A single factor seems to represent the SBIS items because of the marked “elbow” after the first factor.

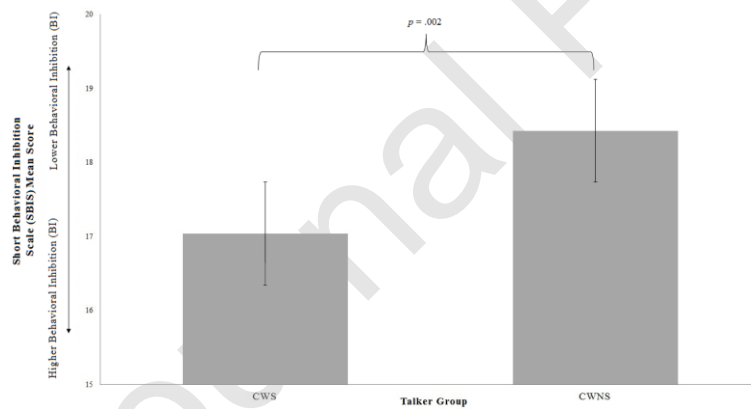


Figure 2: Study 2. Mean scores of the 5-item Short Behavioral Inhibition Scale (SBIS) for children who do (CWS) and do not (CWNS) stutter. Error bars represent plus or minus 1 standard error. Note: Lower SBIS scores indicate higher behavioral inhibition tendencies.

Table 1 *Study 1. Factor Loadings based on Exploratory Factor Analysis (Principal Component Analysis) for the 7 Items of the Short Behavioral Inhibition Scale (SBIS) Questionnaire*

	Factor Loading
Retreats/Approaches unfamiliar people/objects (Item 1)	.75
Stays close/Easily separates from parent (Item 2)	.64
Slow/Quick to warm up (Item 3)	.80
Stops/Continues play/vocalization when unfamiliar person approaches (Item 4)	.63
Stays alone/Engages in group (Item 5)	.43
Heightened fears/No unusual fears (Item 6)	.32
High/Low reactivity (Item 7)	.22

Note: n = 234 (112 children who stutter, 122 children who do not stutter)

Table 2 Study 2. Means (*M*) and Standard Deviations (*SD*) for Norm-referenced Speech-language Tests, and Speech Fluency Measures for Children who Stutter (CWS; *n* = 179) and Children who do not Stutter (CWNS; *n* = 198).

	CWS		CWNS		
	<i>M</i>	(<i>SD</i>)	<i>M</i>	(<i>SD</i>)	<i>p</i>
<i>Speech-language measures (standard score)</i>					
PPVT	108.68	(12.49)	113.36	(12.67)	<.001
EVT	112.45	(11.81)	115.68	(11.91)	<.01
TELD spoken language	114.51	(14.94)	118.12	(12.57)	.010
GFTA	108.36	(9.21)	109.20	(9.75)	.389
<i>Speech fluency measures</i>					
Percent Stuttered disfluencies	8.73	(5.77)	1.12	(.72)	<.001
SSI total score	18.27	(5.55)	6.32	(2.28)	<.001

Note. Peabody Picture Vocabulary Test (PPVT-III/PPVT-IV; Dunn & Dunn, 1997, 2007); Expressive Vocabulary Test (EVT/EVT-2; Williams, 1997, 2007); Test of Early Language Development (TELD-3; Hresko, Reid, & Hamill, 1999); Goldman-Fristoe Test of Articulation (GFTA-2; Goldman & Fristoe, 2000); ; Percent Stuttered Disfluencies = Number of Stuttered Disfluencies per 100 spoken words; Stuttering Severity Instrument (SSI-3/SSI-4; Riley, 1994, 2009).

Table 3 Study 2. *Pearson Product-Moment Correlations between Short Behavioral Inhibition Scale (SBIS) scores and Chronological Age, Socioeconomic Status (SES), and Norm-referenced Speech-language Tests for Children who Stutter (CWS) and Children who do not Stutter (CWNS).*

	SBIS			
	CWS		CWNS	
	<i>Pearson Correlation</i>	<i>p</i>	<i>Pearson Correlation</i>	<i>p</i>
Chronological Age (in months)	.016	.835	.048	.505
Socioeconomic Status (SES)	.159	.065	.022	.785
<i>Speech-language measures (standard score)</i>				
PPVT	.096	.201	.042	.561
EVT	.035	.637	.051	.476
TELD spoken language	.053	.480	-.014	.844
GFTA	-.011	.884	.060	.406

Note. Peabody Picture Vocabulary Test (PPVT-III/PPVT-IV; Dunn & Dunn, 1997, 2007); Expressive Vocabulary Test (EVT/EVT-2; Williams, 1997, 2007); Test of Early Language Development (TELD-3; Hresko, Reid, & Hamill, 1999); Goldman-Fristoe Test of Articulation (GFTA-2; Goldman & Fristoe, 2000)

Table 4 *Study 2. Crosstabulation of talker group (CWS vs. CWNS) and high (lower 15%, SBIS score equal to and/or lower than 13) versus low BI (upper 15%, SBIS score equal to and/or greater than 22) group, with expected values italicized in parentheses.*

	Talker group			
Low/High BI group	CWS	CWNS	χ^2	
Low BI (<i>SBIS score equal to and/or greater than 22</i>)	32 (<i>40.9</i>)	52 (<i>43.1</i>)	7.51**	
High BI (<i>SBIS score equal to and/or lower than 13</i>)	42 (<i>33.1</i>)	26 (<i>34.9</i>)		

Note. ** = $p < .01$

Appendix

Short Behavioral Inhibition Scale

Elizabeth Oyler DeFranco, Ph.D., CCC-SLP & Katerina Ntourou, Ph.D., CCC-SLP

Name: _____ Date of _____

Birth: _____

Date of Evaluation: _____ Age: _____ Sex: M F Respondent: Mother

Father

Below is a list of personal traits or characteristics that describe children. Please circle the number that best describes your child compared to other children the same age. For each item, please circle one number from the 1 to 5.

1)	Retreats immediately from unfamiliar people or objects	OR	Approaches people and objects		
	<u>1</u> usually retreats	<u>2</u> retreats somewhat	<u>3</u> average	<u>4</u> approaches somewhat	<u>5</u> approaches easily

2)	Stays close to the parent	OR	Easily separates from parent		
	<u>1</u> difficult to separate	<u>2</u> hesitant to separate	<u>3</u> average	<u>4</u> separates easily	<u>5</u> separates very easily

3)	Takes a period of time to warm up and interact with unfamiliar people	OR	Quickly warms up and interacts with unfamiliar people		
	<u>1</u> long time to warm up	<u>2</u> Somewhat hesitant to warm up	<u>3</u> average	<u>4</u> Approaches fairly easily	<u>5</u> approaches and warms up very easily

4)	Stops playing and vocalizing when unfamiliar person approaches	OR	Continues playing and vocalizing when unfamiliar person approaches		
	<u>1</u> stops	<u>2</u> quieter and hesitant	<u>3</u> average	<u>4</u> plays and notices	<u>5</u> plays and is unaffected by one's approach

5)	Stays alone and away from other children or caregiver/teacher when in a group	OR	Engages and easily mixes with children or caregiver/teacher when in a group		
	<u>1</u> isolates	<u>2</u> quieter and hesitant	<u>3</u> average	<u>4</u> mixes fairly easily	<u>5</u> mixes very easily

Note to User of the SBIS: The 5- item SBIS is talker- group neutral, allowing its findings to be applied to both young children who do (CWS) and do not (CWNS) stutter. Thus, determination of whether a child stutters or not should be based on standard- of-practice behavioral, diagnostic, observational etc. testing SBIS should be used only as *augmentation* of standard- of- practice testing and neither replace nor substitute for standard for standard- of- practice means for classifying children as CWS or CWNS.