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Dual Diathesis-Stressor Model of Emotional and Linguistic Contributions to Developmental Stuttering

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Abstract

This study assessed emotional and speech-language contributions to childhood stuttering. A dual diathesis-stressor framework guided this study, in which both linguistic requirements and skills, and emotion and its regulation, are hypothesized to contribute to stuttering. The language diathesis consists of expressive and receptive language skills. The emotion diathesis consists of proclivities to emotional reactivity and regulation of emotion, and the emotion stressor consists of experimentally manipulated emotional inductions prior to narrative speaking tasks. Preschool-age children who do and do not stutter were exposed to three emotion-producing overheard conversations—neutral, positive, and angry. Emotion and emotion-regulatory behaviors were coded while participants listened to each conversation and while telling a story after each overheard conversation. Instances of stuttering during each story were counted. Although there was no main effect of conversation type, results indicated that stuttering in preschool-age children is influenced by emotion and language diatheses, as well as coping strategies and situational emotional stressors. Findings support the dual diathesis-stressor model of stuttering.

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Keywords

Developmental stuttering; Emotion; Emotion regulation; Disfluency

Stuttering, which disrupts ongoing speech, can be a life-changing disorder. The disorder is typically characterized by repetitions of sounds, syllables, and monosyllabic words, and by sound prolongations. The frequency, duration, type, and severity of these speech disfluencies vary greatly across speaking situations (Bloodstein and Bernstein-Ratner 2008). Such variability requires explanations that account for between- as well as within-person variability.

Stuttering is a developmental disorder that usually begins between 2.5–6 years of age (e.g., Conture 2001; Mănsson 2000; Yairi and Ambrose 1999). It affects about three times as many boys as girls and has a lifetime incidence of nearly 5%. Of those affected, 70–80% discontinue without formal treatment (e.g., Yairi and Ambrose 1999), though the reasons for this are unknown. For the remaining children (i.e., about 1% continue to stutter after age 6). the negative impact of stuttering can be significant and life-long, affecting academic, emotional, social, and vocational achievements (Conture 1996; Yairi 1997). For adults, stuttering has been linked to depression (Santostefano 1960), depressive mood (Tran et al. 2011), and anxiety and somatic complaints (Treon et al. 2006). Children who stutter have been described as more emotionally reactive, angry/frustrated, fearful, cautions/reserved and less adaptable (e.g., Anderson et al. 2003; Eggers et al. 2010; Embrechts et al. 1998; Karrass et al. 2006), as well as exhibiting poor attention regulation (e.g., Felsenfeld et al. 2010) and high risk for bullying (Blood and Blood 2007). Thus, there is a need to identify factors that initiate/cause, exacerbate or perpetuate stuttering. Furthermore, it is important to study young children who stutter (CWS) who are close to the onset of stuttering, and contrast them to children who do not stutter (CWNS), to disentangle factors that contribute to stuttering from subsequent consequences of stuttering.

Emotion-Language Dual Diathesis Stress Process Model of Stuttering

Stuttering has been related to speech-language (e.g., Anderson and Conture 2004; Anderson, et al. 2005; Hall et al. 2007; Ntourou et al. 2011) and psychological factors (e.g., Alm 2004; Bloodstein and Bernstein-Ratner 2008; Craig el al. 2003; Johnson et al. 2010). However, there has been relatively little intersection between speech-language pathology and psychological approaches to emotion, stress and coping (ef., Brutten and Shoemaker 1967; Treon 2010).

We approach stuttering in terms of a diathesis-stress process in which “stress activates a diathesis, changing the potential of predisposition into the presence of psychopalhology” (Monroe and Simons 1991, p 406). Variable stressors may “activate” relatively stable diatheses (individual differences), leading to disruptions in fluent speech and language. Conture and Walden (in press) proposed a *Dual Diatheses-Stressor* framework (DD-S). in which diatheses and stressors of emotion and speech-language processes contribute to childhood stuttering (Fig. 1). The emotion diathesis consists of relatively stable proclivities for emotional reactivity and regulation. Emotional stressors are variable features of situations that elicit emotion. The language diathesis comprises relatively stable processes of speech-language planning and production. Language stressors are situational requirements for effective communication.

Both linguistic and emotional challenges can act as stressors in particular situations. For example, when linguistic requirements are high (e.g., perceived or real need to achieve accurate, rapid conversation), they may challenge the child’s developing speech-language

system. Likewise, when emotional requirements are high (e.g., novel situations of high importance to the speaker), they impose emotional stress and may tax existing resources. Language and emotional stressors may be present concurrently (e.g., public speaking situations or getting to know someone new), These variable situational stressors can operate adversely on existing language and emotional diatheses to impact the child's attempts to initiate or maintain fluent communication, The present study is the authors' first direct test of the overall DD-S model including linguistic and emotional factors simultaneously.

The emotional components of the model are conceptualized in terms of two aspects: reactivity and regulation. *Emotional reactivity* refers to the tendency to experience frequent and intense emotional arousal (Kagan 1997). *Emotion regulation* involves processes of initiating, maintaining, or modulating the occurrence, intensity, or duration of feelings and emotion-related physiological processes (Thompson 1994). Emotional reactivity and regulation each have dispositional as well as situationally-responsive components. The model predicts that emotional reactivity, emotion regulation and their joint effects influence the frequency and severity of stuttering in preschool-age children.

Emotion and Language Contributions to Developmental Stuttering

Emotion and Stuttering

Regarding the emotion diathesis, childhood stuttering has been associated with emotional temperament (e.g., Anderson, et al. 2003; Eggers, et al, 2010; Embreehts et al, 1998; Karrass et al. 2006; ef. Reilly et al. 2009). Some recent models (e.g., Conture et al. 2006; Riley and Riley 2000) include temperamental emotional factors in a broad perspective on stuttering (see Smith and Kelly 1997 for review). Evidence links emotional reactivity to stuttering (e.g., Embreehts et al, 1998), and our own findings link stuttering temperamental proclivities both to emotional reactivity and to stuttering (e.g., Karrass et al. 2006). Karrass et al. (2006) focused on group differences between CWS' and CWNS' temperament, but did not link those differences directly to instances of stuttering. The study was limited by its correlational design, which did not address direction of causality. Furthermore, temperamental differences, which are cross-situationally stable by definition, cannot readily account for within-child variations in stuttering across situations.

Findings pertaining to *diagnosed groups* may not map directly onto *instances* of stuttering. To address that limitation, Arnold et al. (2011) studied preschool-age CWS and CWNS exposed to emotion-eliciting background conversations followed by narratives, reasoning that emotions immediately prior to speaking would impact speech Findings indicated that CWS who engaged in more frequent and longer-lasting regulatory strategies while speaking stuttered less than CWS who used fewer regulatory strategies. Johnson et al. (2010) observed preschool-age CWS and CWNS in a disappointing gift procedure assessing children's use of display rules that specify when one should mask emotional expressions (e.g., Cole 1986; Kieras et al. 2005; Saarni 1984). Children received a desirable or disappointing gift prior to a free-play conversation. CWS had more negative emotional expressions than CWNS after receiving the undesirable gift Furthermore, CWS stuttered more after receiving the desirable than disappointing gift, suggesting that emotions prior to and during speaking contribute to childhood stuttering even in situations involving positive emotion.

Coping and Stuttering

We explored a role in developmental stuttering for how children deploy behavioral coping strategies. The study of stress and coping focuses mainly on decreasing negative emotion, but gave rise to the more general notion of emotion regulation, processes of increasing and decreasing both negative and positive emotion, (Gross 1998). Coping behavior integrates

both linguistic and emotional facilities into overt responses to a negative emotion. Indeed, understanding the coordination of linguistic and emotional facilities into coping strategies is nascent, leading us to explore five strategies measured by a parental report instrument described below. The present study thereby makes a first investigative foray into children's coping strategies in relation to stuttering.

Language and Stuttering

Regarding the speech-language diathesis, vulnerabilities in speech-language planning and production may be associated with childhood stuttering (Hall et al. 2007; Ntourou et al. 2011; ef, Nippold 1990). CWS have been reported to show difficulty in phonological encoding, (e.g., Byrd et al. 2007; Sasisekaran et al. 2006), and lexical and syntactic processes (e.g., Anderson and Conture 2004; Tsiamtsiouris and Cairns 2009; ef., Häge 2001; Hennessey et al. 2008). CWS ate also more likely to have language discrepancies between linguistic domains such as receptive versus expressive language (Anderson, et al. 2005; Coulter et al, 2009). In the present study, based on previous findings, we hypothesized that poor language development would be associated with increased stuttering. Since stuttering manifests as an expressive language disorder, we also studied unevenness in expressive language as our operationalization of language discrepancies, and whether such unevenness would be associated with increased stuttering.

The Present Study

The present study used an experimental method to evaluate the DD-S model with respect to stuttering during a story-telling task. The method was adapted from studies of the effects of overheard anger on children in families with marital distress. Overheard anger increases arousal and distress in toddlers (Easterbrooks et al, 1994), preschool (Cummings 1987; Cummings et al, 1989) and school-age children (Davies et al. 1996). In the present study children listened to auditory recordings of three "overheard" conversations (OC) between two adults (happy, angry and neutral). We expected these variations in situational emotional stressors to impact stuttering immediately afterward. This study design addresses the direction of effects between emotion and stuttering behaviors, that is, whether there is a causal link between prior induced emotion and subsequent stuttering.

We operationalized the concepts in the DD-S framework as follows: 1) the emotion diathesis consisted of scores focusing on negative affect and emotion regulation as reported by parents; (2) situational emotional stressors were created via the three overheard conversations described above and below and 3) the language diathesis comprised several widely-used assessments of language development and a measure of test scatter on an expressive language scale as an index of unevenness of expressive development. Finally, we assessed parent reports of children's use of various coping strategies.

In addition to testing the fit of the overall DD-S model, hypotheses about components of the model were: 1) Temperamental proclivities to emotional reactivity increases stuttering and emotion regulation decreases stuttering in CWS and reactivity and regulation will interact, 2) CWS and CWNS are differentially impacted by experimentally manipulated emotional overheard conversations as compared to neutral ones, 3) Observed emotional reactivity and regulation during speaking are associated with stuttering in CWS, and 4) Lag in language development is associated with increased stuttering in CWS and greater unevenness of expressive language is associated with increased stuttering. We explored the role of coping strategies in stuttering.

Method

Participants

Nineteen CWS (mean age=46.5 mo, range 37–60 mo; 13 males) and 22 CWNS (mean age=49.3 mo, range 37–59 mo; 9 males) monolingual English speakers were included in the final data corpus (additional excluded children are noted below). All were Caucasian except one African-American CWNS. Participants were paid volunteers recruited through a free local monthly parent magazine, Tennessee State birth records or the Vanderbilt Bill Wilkerson Hearing and Speech Center No CWS had received treatment for stuttering. The protocol was approved by the Vanderbilt University IRB. Informed consent by parents and assent by children were obtained. All included children passed a pure tone hearing screening (1 was excluded for failing to pass the hearing screening).

A child was assigned to the CWS talker group if he or she had: (a) three or more within-word sound/syllable repetitions, sound prolongations, broken words and/or monosyllabic whole-word repetitions per 100 words of conversational speech (Bloodstein, 1995; Conture 2001) and (b) a score of II or higher (severity of at least “mild”) on the Stuttering Severity Instrument-3 (SSI-3; Riley 1994). A child was assigned to the CWNS talker group if he/she (a) exhibited two or fewer sound/syllable repetitions, sound prolongations, broken words and/or monosyllabic whole-word repetitions per 100 words of speech (Conture and Kelly 1991; Zebrowski and Conture 1989) and (b) received a score of 10 or lower (severity of less than “mild”) on the SSI-3. Three children were excluded because they could not be classified into one of the two groups.

To avoid confounds with clinically significant speech-language concerns, children were excluded for scoring below the 16th percentile on the Peabody Picture Vocabulary Test-3rd Edition (PPVT, Dunn and Dunn 1997), the Expressive Vocabulary Test (EVT, Williams 1997), Test of Early Language Development-3 (TELD, Hresko et al. 1999) or “Sounds in Words” subtest of the Goldman-Fristoe Test of Articulation-2 (Goldman and Fristoe 2000), Two children were excluded based on this criterion.

Procedure

Participants were tested twice. The first session included standardized tests of speech and language, bilateral pure tone hearing screenings and an unstructured conversation with an experimenter, These were used to classify talker group and assess whether speech, language, and hearing were within normal limits. During a second session 1–2 weeks later, participants were fastened in a standard child car seat mounted in a child-size replica of a “Jeep.” A 19-inch flat-panel monitor served as the “windshield” on which visual stimuli were displayed and auditory stimuli were presented through speakers on each side of the monitor. A Powerpoint slideshow presented pages from picture books during a familiarization period immediately prior to “overhearing” an emotionally lenored conversation and for the narrative task. Children were videotaped during the overheard conversations and narrative. Thus, each condition included a pre-narrative stimulus presentation, one overheard conversation, and a narrative task.

Pre-narrative Stimulus Presentation—Illustrations from four textless picture books by Mercer Mayer were shown on the monitor: *Frog, Where Are You?* (1969)), *A Boy, a Dog and a Frog* (1967), *Frog on his Own* (1973) or *Boy, a Dog, a Frog and a Friend* (1971). The child was told, “Look at the pictures for the story you will tell later.”

Overheard Conversations—To create emotional arousal, children “overheard” three 1–2-minute audiotaped conversations between two adult female actresses, Each conversation

was presented in counterbalanced order with the perception to the child that it occurred in the next room. Three scripts were developed and each was enacted with happy, angry, or affectively neutral vocal tone and prosody. Twenty-seven children heard conversations from one set of actresses and 14 heard a new set of actresses. Validation samples (repotted in Arnold et al. 2011) indicated that adults and children perceived each conversation in line with the intended emotion.

Narrative Task—Following each OC the child was encouraged to tell a story cued by illustrations from one of the picture books. Children were prompted with "What do you see happening here?" or "What are they doing?"

Measures

Temperamental Emotion—During the first session, parents completed items from the Children's Behavior Questionnaire (CBQ; Rothbart et al. 2001). One scale measured Emotion Regulation and was an additive composite of two CBQ scales: Falling Reactivity (alpha=0.79) and Inhibitory Control (alpha=0.76). The Negative Affect scale comprises a coherent set of items (alpha=0.72) from 4 CBQ scales (reverse-scored items indicated with '(R)'. From the Fear scale we used "Is afraid of the burglars or the "boogie" man," and "Is not afraid of the dark," (R). From the Sadness scale we used, "Tends to become sad if the family's plans don't work out," "Seems to feel depressed when unable to accomplish some task," and "Becomes upset when loved relatives or friends are getting ready to leave following a visit." From the Anger scale we used. "Gets quite frustrated when prevented from doing something s/he wants to do," and "Gels angry when s/he can't find something s/he wants to play with." From the Discomfort scale, we used. "Is quite upset by a little cut or bruise," "Is not very upset at minor cuts or bruises" (R), and "Hardly ever complains when ill with a cold" (R). All items were rated on 7-point scales from "extremely true" to "extremely untrue."

Emotional Reactivity During the OC and Narrative—Positive emotion (e.g., smiles), negative emotion (e.g., frowns), and movement (e.g.. kicking) were coded from digital video recorded during the second session, during both the OC and the first three minutes of narrative. Positive and negative emotion were rated separately by one of four coders on 5-point scales at I-sec intervals. Instances of movement were scored as present or absent during each interval. Seconds were aggregated into a total duration for positive and negative emotion factors. Coders were trained to a minimum reliability of 0.80 and for a random 20% subset emotions were rated by all four coders with pairwise reliabilities between 0.76 and 0.96.

Principal components analysis indicated that while *listening* during the OC, movement factored positively with positive emotion and separate from negative emotion. This is consistent with Watson and Tellegen (1985), who proposed that positive and negative emotion arc separate dimensions. However, while *speaking*, positive and negative emotion factored together inversely and separate from movement, consistent with Russell and Carroll (1999), who proposed that positive and negative emotion represent two ends of a single dimension. Each factorization was used to index observed emotional behavior during the OCs and while narrating.

Emotion Regulation During the OCs and Narratives—Four behaviors coded from video during each OC and the first three minutes of each narrative assessed emotion regulation: (1) social looks at the experimenter, (2) self-stimulation without visual attention to the self (e.g., pulling an ear lobe), (3) self-stimulation with visual attention (e.g., touching fingers while looking at them), and (4) looks away from the computer screen (Buss and

Goldsmith 1998; Grolnick et al. (1996). Each was rated by one of five coders as present or absent in each I-sec interval. Seconds were aggregated into a total duration and converted to percentage of time each behavior occurred. Coders were trained to a minimum reliability of 0.80 and for a random 20% subset regulation was rated by all five coders, with pairwise reliabilities from 0.83 to 0.99. The final score reflected the maximum of the four percentages for each participant.

Coping Strategies—During the first visit, parents completed the Child Coping Scale (CCS, described in Eisenberg et al. 1993), which rates children's coping behaviors on 7-point scales indicating the likelihood that the child would engage in each of 13 types of coping in 3 anger/frustration scenarios and one general context (no scenario). A total of 42 items were combined as described by Eisenberg et al. (1993) into five classes of coping strategies: aggression (e.g., hitting, alpha=0.86), venting (e.g., crying, alpha=0.89), social support seeking (e.g., getting help from a teacher, alpha=0.88), distancing (e.g., walking away, alpha=0.81) and instrumental problem solving (e.g., fixing something broken, alpha= 0.73).

Language Skills—Receptive language was measured using the TELD3-REC and the PPVT-III. Expressive language skill was measured with the EVT and TELD3-EXP Standard scores on the four were averaged for a score of language performance. Unevenness in expressive language consisted of within-test scatter on the TELD3-EXP, a more comprehensive measure of language ability than the EVT, which focuses more on vocabulary. Scatter was operationalized as the presence/absence of multiple basal runs of correct items separated by incorrect items.

Mean Length of Utterance (MLU)—MLU was calculated as the mean number of morphemes per utterance dining the first session free play conversation with the experimenter.

Fluent and Disfluent Words During the Narrative—Total Words during narratives was a count of fluent and disfluent words. Stuttering-Like Dis fluencies (SLD) was a count of sound-syllable repetitions (e.g., "I b-b-broke the glass"), single-syllable word repetitions (e.g., "I broke the-the-the glass"), audible sound prolongations (e.g., "I bbbroke the glass") and inaudible sound prolongations (i.e., silently holding the articulatory position for the stop phase of a stop-plosive consonant, e.g., "I broke the … glass"). Revisions were the number of stops and restarts with changes in content involving one or more words (e.g., "The boy is… The dog is…").

Attitude Toward Speaking—During the first visit, children were administered the KiddyCAT Communication Attitude Test for Preschoolers and Kindergarteners (Vanryckeghem, and Brutten 2007), which discriminates between CWS and CWNS, regardless of age or gender (e.g., Clark et al. 2011), The KiddyCAT consists of 12 statements read aloud by the examiner, to which children respond with 'yes' or 'no' indicating what they think about their speech (e.g., "I like-the way I talk."). Scores for the 12 items are summed.

The Hollingshead Four-Factor Index of Social Position—(Hollingshead 1975) classified social-economic status (SES) based on parent report.

Results

Descriptive Statistics

Overall and group means for CWS and CWNS are in Table 1, CWS stuttered more (consistent with the criteria for assignment to groups) and spoke fewer total words in the narrative. CWS also were rated lower in observed regulatory behaviors during the narrative. No other group differences were found (e.g.. age, SES), including no main effects of group on temperamental proclivities to negative reactivity or emotion regulation.

Modeling Approach

Mixed model analysis assessed stuttering during the three narratives. In addition to the emotional and linguistic parameters repotted individually below, the model included live variables known to be linked to stuttering. Boys exhibited significantly more stuttering than girls, F (1,83.14)=12.47. p<0.00l. Age was not associated with stuttering frequency. As expected, as the number of words spoken in a narrative increased, so did the number of stutterings, F(1,84.69)=50.12, p<0.0001. Greater MLU overall was associated with less stuttering, F(1,77.35)=5.87, p<0.02. Greater self-reported concern about one's own speaking (KiddyCAT) was associated with less stuttering, F(1,75.35)=20.03, p<0.000l.

We approached modeling as follows. We first tested the significance of the overall model, followed by parameters of the model in five groups: (a) emotional diathesis parameters (CBQ derived scales), (b) emotional stressor parameters (overheard conversation conditions within subjects), (c) emotional behavior (behavior coding of emotion and emotion regulation), (d) language diathesis (tests of language development and uneven expressive language (scatter on the TELD3-EXP), and (e) coping (CCS scales). For each group, we assessed effects associated with individual parameters in the context of the complete model by evaluating change in model likelihood (Verbeke and Molenberghs 2000) when each group was added to a (nested) model containing all parameters except those in the group being evaluated. Doing so assured that each group of parameters yielded a significant improvement in fit, that is, each group of parameters contributed a significant increase in explanatory specificity to the overall model.

Test of the Overall DD-S Framework

To test whether both linguistic and emotional factors are simultaneously implicated in stuttering, we evaluated the aggregate of parameters. Emotional and linguistic factors. taken together, did significantly explain variations in stuttering across children. That is, the model including all parameters was a better fit than the empty (null) model, $\chi^2(42)=100.78$, p<0.000 1, CAIC corrected (Verbeke and Molenberghs 2000). Talker Group (CWS vs. CWNS) was included in the model to evaluate differences in effects between talker groups (interactions with Group). The effect for Talker Group unconditioned by covariates (the simple effect) was significant, F(1,65.81)=59.75, p<0.000l. It is noteworthy that the other predictors more fully accounted for variance in stuttering, reducing the main effect of talker group to non-significance, p>0.4

Emotional Diathesis

We tested the DD-S framework's prediction that temperamental proclivities for emotional reactivity and regulation—as reported by participants' parents—play a role in stuttering. Findings indicated that they do, extending our prior findings (e.g., Arnold et al. 2011; Johnson et al, 2010; Karrass et al. 2006). For all children (CWS and CWNS) there was an interaction between parent-reported negative emotion and regulation on stuttering. When higher proclivity to negative emotion co-occurred with high emotion regulation there was less stuttering for all children, F (1,83.39)=8.31, p<0.005. est. β=−0.157, p<0.0001. In

addition, the main effect for greater proclivity to regulate was marginally associated with more stuttering, $F(1,84.64)=2.95$, $p<0.09$, est. $\beta=0.099$, $p<0.09$. This model had significantly better fit than the model containing all other parameters except participants' proclivities to react and regulate, $\chi^2(3)=52.31$, $p<0.0001$. These findings support the DD-S model in that the diatheses of emotional reactivity and regulation significantly predicted frequency of stuttering.

Emotional Stressor

We tested whether there was an effect of emotional tenor (neutral/happy/angry) of the prior OC on stuttering in the narrative that followed. There was no main effect or interaction of OC Tenor and Talker Group. We then tested whether emotional tenor might have had a weaker effect, such that carryover from the first OC overshadowed effects of subsequent OCs. This was the case. There was an interaction of Talker Group and whether the first OC was emotional in tenor or not, $F(1,89.10)=13.54$ $p<0.0005$, CWS stuttered significantly *more* during all three narratives when the first OC was positive or negative (EMM=16.6) versus neutral (EMM=11.6), $F(1,90.59)=5.67$, $p<0.02$. CWNS stuttered significantly *less* during all three narratives when the first OC was emotional, $F(1,84.18)=6.46$, $p<0.02$, EMMs=0.9 for emotional vs 6.1 for neutral. This model had significantly better fit than a model with all other parameters except the presence or absence of emotional tenor during the first OC, $\chi^2(2)=29.27$, $p<0.0001$, suggesting that the impact of prior emotion on subsequent stuttering can linger for quite some time.

Overt Emotional and Emotion Regulatory Behavior

We tested whether concurrent emotional reactivity and emotion regulation play a role in stuttering. Findings indicate that they do; their inclusion resulted in a model with significantly better fit than a model with all other parameters except participants' reactive and regulatory behavior while speaking, $\chi^2(6)=16.00$, $p<0.02$. For CWS only, negative emotion while speaking was associated with more stuttering, est. $\beta=0.585$, $p<0.0001$. Such negative emotion could, of course, result from the act of stuttering, rather than being an antecedent contribution to stuttering. However, findings cast doubt on this possibility because for CWS only, during speaking there was an interaction of exhibited negative emotion with emotion regulation. More regulatory behavior in the context of greater negative emotion was associated with less stuttering, $F(1,80.48)=13.12$, $p<0.001$, est. $\beta=-0.579$, $p<0.001$.

Diathesis of Language

We tested whether differences from age norms in expressive and receptive language development might be associated with stuttering. There was no effect of language development on stuttering (as indicated by the average standard score on two receptive and two expressive indicators of language performance), $p>0.9$. This is inconsistent with our prediction that variations in language skill would contribute to stuttering in preschool-age CWS.

We also tested whether unevenness in expressive language skill might reflect a vulnerability associated with stuttering. Findings suggest that it does so as predicted, uneven expressive language was associated with more stuttering for all children, $F(1,84.43)=9.065$, $p<0.005$. Including this effect resulted in a model with significantly better fit than that of a model containing all other parameters except presence or absence of marked unevenness in expressive language skills, $\chi^2(2)=40.96$, $p<0.000l$. This finding supports the hypothesis that unevenness in expressive language may act as a diathesis for stuttering.

Coping Strategies (CCS)

We tested our provisional hypothesis about parent-reported proclivities for their child to use different behavioral coping strategies (aggressing, venting, social support seeking, problem solving and distancing) being associated with stuttering. Including these effects resulted in a model with a significantly better fit than the model containing all other parameters except parent-reported coping strategies, $\chi^2(7)=66.03$, $p<0.0001$. All five coping strategies were significantly associated with stuttering, though not entirely as provisionally predicted. For all children, *more* stuttering was associated with greater parent-reported distancing/withdrawal. $F(1,85)=7.43$, est. $\beta=0.109$, $p<0.01$ and social support seeking $F(1,86,45)=17.88$, est. $\beta=0$ 199, $p< 0.0001$. For all children, *less* stuttering was associated with greater instrumental problem solving, $F(1,80.87)=11.10$, est. $\beta=-0.169$, $p<0.005$. There was a significant contrast between talker groups for the association between a proclivity to vent (cry) and stuttering, $F(1,76.55)=9.35$, $p<0.005$. For the CWS, greater proclivity to venting was associated with less stuttering, est. $\beta=-0.216$, $p<0.01$, whereas for the CWNS, it was associated with more stuttering, est. $\beta=0.194$, $p<0.05$. Likewise, the two talker groups differed in terms of the association between proclivity to aggression and stuttering, $F(1,82.70)=4.00$, $p<0.05$: Though the estimated βs for neither group were significant, for the CWS, greater proclivity to aggression was associated with more stuttering whereas for the CWNS it was associated with less stuttering, Thus, coping strategies were related to stuttering.

Discussion

The Dual Diathesis-Stress (DD-S) framework (Conture and Walden in press) attempts to represent why stuttering predictably occurs more in certain situations, The DD-S framework specifies contributions to stuttering from both internal diatheses, or predisposing factors, and situational stressors in emotion and language. The present study extends prior work that separately examined linguistic (e.g., Coulter et al. 2009) and emotional processes (e.g., Arnold et al. 2011; Johnson et al. 2010; Karrass et al. 2006). Findings from this study support the overall DD-S framework, showing roles in stuttering for emotional and speech-language diatheses, emotional stressors and coping strategies.

Emotion Diatheses

We expected that proclivities to negative reactivity and to lower emotion regulation would be associated with more stuttering. This prediction fits the commonplace view that negative emotional reactivity is problematic, whereas regulation is salutary. Although there were no main effects of either negative emotion or emotion regulation on stuttering, we found the expected interaction between negative arousal and emotion regulation. Thus, a simple explanation—reactivity is “bad” and regulation is “good”—might sometimes be applicable to the diagnosis of stuttering, but less so to stuttering in particular situations. Despite fewer observed regulatory behaviors in the CWS group during speaking, for both talker groups less stuttering was observed for children described by their parents as typically exhibiting higher levels of both negative emotion and regulation, perhaps reflecting a balance in which children react and recover as they adjust to situations.

This interaction of reactivity and regulation was also found in the relation between CWS’ exhibited reactivity/regulation and stuttering. CWS who exhibited more negative emotion without concomitant regulation exhibited stuttered more, consistent with Brutten and Shoemaker’s (1967) speculation. In contrast, CWS who exhibited both more negative emotion and more regulatory behavior stuttered less. This last finding is subtle, yet important, inasmuch as it runs contrary to the notion that emotional processes are not causally related to stuttering, but instead reflects a reaction to stuttering (Alm 2004). Even if

all the negative emotion during the narrative resulted from stuttering, we also found that emotion regulation reduced stuttering. Coupled with finding that unregulated negative emotion during speaking was associated with more stuttering, the notion that emotional processes are importantly part of the causal nexus of developmental stuttering is supported.

In our findings, for the production of fluent utterances, the proclivity to negative emotion and regulation appear to act in concert. To the extent that coordination of emotional processes is successful in adapting each individual to her or his changing situation (Frankel and Ray 2000; Thompson 1994), the individual may be enabled to communicate more fluently in her or his current social situation.

Situational Emotional Stressors

Findings suggest that situational stressors giving rise to emotions are associated with occurrences of stuttering. The presence or absence of emotional tenor in the first OC affected how much all children stuttered in the three subsequent narratives combined. Intriguingly, CWNS and CWS exhibited this effect in opposite directions. Consistent with our speculation, CWS stuttered more when the first OC was emotional, suggesting that emotional arousal in itself, whether positive or negative, disrupts fluency for CWS. Furthermore the effect persisted, even when the child experienced intervening events that might have been expected to after his or her emotions (see Anderson, et al. 2003).

Counter-intuitively, however, CWNS stuttered more in all three narratives when the first conversation overheard was unemotional. In this experiment, variations in prosodie features of the overheard speech connoted positive or negative emotion or lack of either. Perhaps, for CWNS, a first conversation that was prosodieally flat and lacking emotion made the situation more disconcerting or unusual than a first conversation with emotional content. Anecdotally, the neutral conversation was sometimes perceived to be “strange.” Young children may be accustomed to more prosodic variation and a lack thereof, particularly in a university laboratory, may have heightened their uncertainty. Of course, frequency of stuttering did not reach clinical levels for CWNS, and the emotion manipulation did not turn them into “stutterers;” rather, they are typical 3- to 5-year-old speakers who sometimes stutter within normal limits.

Language Diathesis

The DD-S model specifies that language diatheses contribute to stuttering, however we found that overall language development relative to age mates was not related to frequency of stuttering, inconsistent with current predictions and some previous findings (e.g., Ntourou et al. 2011; cf, Nippold 1990, 2004). It should be noted that we excluded participants who scored normatively in the 16th percentile or below to avoid confounding language disorder with stuttering, thus, the range of language scores was restricted.

Greater intra-subtest scatter for expressive language predicted more stuttering, consistent with previous reports of a relation between stuttering and unevenness in language (e.g., Anderson et al. 2005; Coulter et al. 2009). Scatter reflects inconsistent response sequences in which easier items are failed but a substantial consecutive (basal) string of subsequent harder items are correct. Godber et al. (2000) suggested that such “performance gaps” in intellectual tests are a measure of cognitive inefficiency. By extension, children who stutter may have expressive language inefficiencies (Anderson and Conture 2004; Tsiamtsiouris and Cairns 2009). Perhaps when searching for a suitable means to express themselves, preschool-age children who stutter struggle to quickly and efficiently formulate their linguistic plan.

Test scatter may also reflect variations in arousal or attention (Lezak 1995). Given that CWS have been reported to have increased emotional arousal and/or difficulty regulating arousal, it is possible that these children are distracted from speaking by unregulated arousal, by excessive regulatory effort or both. Bosshardt (2006) reported that people who stutter are likely to have difficulty focusing or shifting attention on non-language tasks, consistent with other findings (Eggers et al. 2010; Felsenfeld et al. 2010; Heitman et al. 2004). Another possibility is that CWS with scatter may not fully commit attention to the task. Worthy of further investigation—perhaps using a more refined quantitative measure—intra-subtest scatter, a new operationalization of language unevenness that awaits replication, may reflect a variety of problems in regulating attention, broadly construed.

Coping Strategies

We explored parents' reports of their children's proclivities to use five different coping strategies and found that all five coping strategies comprising the CCS scales were associated with stuttering frequency. Three strategies were associated with *less* stuttering: Instrumental coping (for all children), aggression (for CWNS) and venting (for CWS). Two strategies were associated with *more* stuttering for all children: Social support seeking and distancing, Significant effects for parental reports of all five types of coping are consistent with the suggestion that stuttering is related to coping. However, the present findings indicate that not all coping strategies positively impact stuttering. Since strategies like support seeking and distancing are associated with more rather than less stuttering, we need to better understand the various types of coping and their relation to childhood fluency. Future studies are needed to explicate the role of various coping strategies on stuttering.

Limitations

This was not an investigation of mechanisms. As suggested above, different types of regulation play different roles in developmental stuttering, some beneficial, some less so—an issue that goes beyond the present DD-S model, into theories about underlying mechanisms. DD-S is a process model and as such, mechanisms for each process are not specified. Further research needs to be informed, not only by our present DD-S process model but also by a theory of how different processes work together. Such a theory of mechanisms would include, for example, explanations of how coping strategies coordinate emotional and linguistic facilities to influence stuttering, sometimes for better and sometimes for worse.

An additional limitation is that overheard conversations between unfamiliar people did not produce the clear effect expected. These conversations may be less evocative than, for example, gruesome scenes of accidents or amputations (inappropriate for use with children). Yet overheard conversations belong to a class of emotion inducers that happen every day, and thus seem to afford good generalizability. We used them to elicit mild-to-moderate emotion because we expected to see effects of the hypothesized processes more clearly than if children experienced very strong emotion. Although a carry-over effect from the first conversation influenced stuttering for all children, there were no effects of the OCs for the within-subjects condition. It is unclear whether similar conversations between familiar persons or some other emotion induction may accentuate or mask effects reported in this study, and replications using other emotion inductions arc needed.

Concluding Remarks and Future Directions

The present study was motivated by a theoretical framework whereby diatheses and stressors of both language and emotion are important predictors of childhood stuttering (Conture and Walden in press). As such, it is the first to empirically assess linguistic and emotional processes simultaneously, along with behavioral coping strategies, in preschool-

age children who do and do not stutter. As a first such test of the model, all results should be interpreted cautiously until replicated. An important future direction is to study CWS longitudinally to investigate whether children who discontinue stuttering differ from those who do not and whether changes in emotional and linguistic capabilities precede or are concurrent with changes in stuttering.

Importantly, this study's findings for the multi-faceted DD-S framework are strong enough to offset the shared method variance problem in stuttering research—group membership (CWS/CWNS) is substantially defined by the primary outcome, stuttering frequency. Results showed a marked shift in variance explained, away from group membership and toward diatheses and stressors of language, emotion and coping. The present results suggest that fluent speech may be best facilitated by efficiently-regulated emotions. When this coordination is disrupted a child may be at risk for stuttering. Moreover, the disruptions may take many forms. Reactivity can be under-regulated, which may be the case in the present study for CWS who display relatively greater negative emotion while narrating without displaying concomitantly greater regulation. Reactivity might be over-regulated and emotion would be dampened. Some forms of regulation, like instrumental coping, may use less linguistic resources and so reduce stuttering, whereas others, like social support seeking, may use more linguistic resources in communicating, and may exacerbate stuttering. Ultimately, untangling the complex findings will likely require investigation of underlying mechanisms, and how and when they are recruited.

In sum, present findings are consistent with the perspective that childhood stuttering is not solely a disorder of speech and language, but also has emotional components that may be associated with psychological co-morbidities that impact the quantity and quality of this childhood communication disorder.

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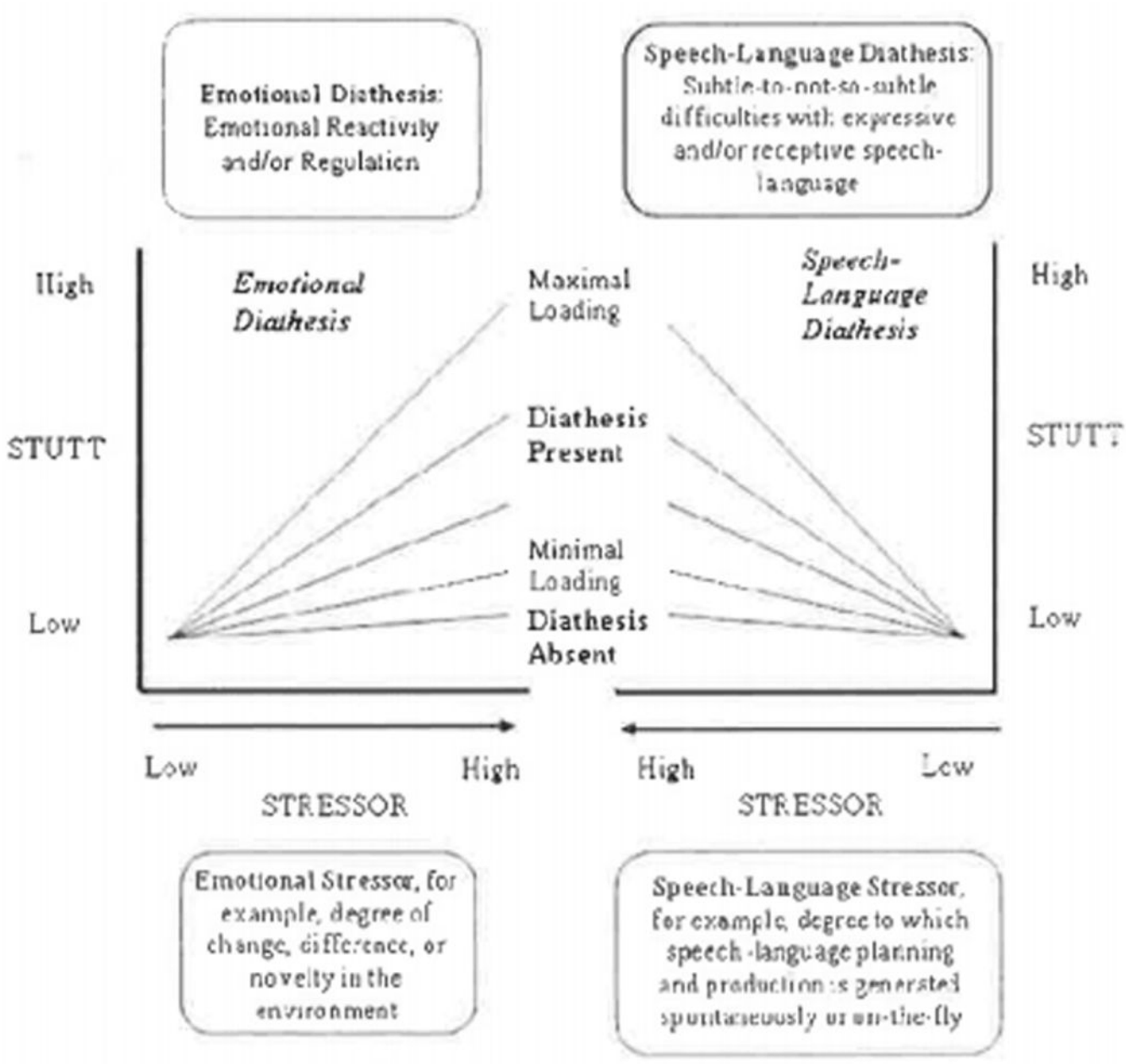

Fig 1.
Dual diathesis-stressor model of stuttering from Conture and Walden (in press)

Table 1

Means (M) and standard deviations (SD) for all children and CWS and CWNS groups

	All		CWS		CWNS		Sig. Δ(cws, cwns)
	M	**SD**	**M**	**SD**	**M**	**SD**	
Individual Differences							
Age (months)	47.95	7.55	46.47	7.97	49.23	7.10	*n.s.*
SES	42.71	10.99	40.03	7.26	45.22	13.35	*n.s.*
Coping Strategies (CCS—parent report)							
Coping By Distancing	2.22	0.97	1.96	1.03	2.44	0.88	*n.s.*
Instrumental Coping	3.62	1.27	3.30	1.10	3.91	1.36	*n.s.*
Coping By Support Seeking	4.85	1.24	4.74	1.33	4.95	1.19	*n.s.*
Coping By Aggression	2.74	1.35	2.63	1.72	2.84	0.96	*n.s.*
Coping By Venting	4.06	1.25	4.07	1.48	4.05	1.05	*n.s.*
Emotional Temperament (CBQ—parent report)							
Negative Affect	3.87	0.99	3.86	1.10	3.88	0.78	*n.s.*
Emotion Regulation	4.92	0.65	4.93	0.66	4.90	0.64	*n.s.*
Language Skills							
Language Skill (PPVT+EVT+TELD)	111.1	9.64	109.3	11.11	112.73	8.09	*n.s.*
Repeated Measures							
Emotion							
OC Emotion Factor 1: Positive & Movement	0.00	1.00	−0.10	0.81	0.08	1.14	*n.s.*
OC Emotion Factor 2: Negative	0.00	1.00	−0.04	1.00	0.04	1.01	*n.s.*
OC Emotion Regulation	0.92	0.08	0.91	0.08	0.93	0.08	*n.s.*
Narrative Emotion Factor 1: Valence	0.00	1.00	0.22	1.02	−0.19	0.95	*n.s.*
Narrative Emotion Factor 2: Arousal	0.00	1.00	−0.09	1.10	0.07	0.90	*n.s.*
Narrative Emotion Regulation	0.38	0.20	0.34	0.18	0.42	0.21	0.03
Speech							
Total Words	279.5	97.94	262.5	96.21	293.91	97.79	0.0001
Total SLDs	8.89	9.95	15.16	11.61	3.58	2.76	0.0001