Attentional Regulation in Young Twins With Probable Stuttering, High Nonfluency, and Typical Fluency

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Purpose: Using a sample of 20,445 Dutch twins, this study examined the relationship between speech fluency and attentional regulation in children. A secondary objective was to identify etiological overlap between nonfluency and poor attention using fluency-discordant twin pairs.
Method: Three fluency groups were created at age 5 using a parent questionnaire: (a) probable stuttering (PS; $N = 826$; 4.0%), highly nonfluent (HNF; $N = 547$; 2.7%), and typically fluent (TF; $N = 19{,}072$; 93%). Multiple scales assessing attention, primarily self-regulation/inhibition, were obtained from both parents when children were ages 5 and 7 and from teachers when children were age 7.
Results: When compared with the TF controls, both the PS and HNF children received higher (i.e., more problematic) scores on parental attention ratings at both ages ($p < .002$). Effect sizes were moderate for both groups. Teacher and parent ratings were generally comparable. The discordant co-twin analyses suggested that nonfluency and attention were influenced by potentially overlapping genetic and shared environmental factors.
Conclusions: The liability to express both high nonfluency and problems with self-regulation/inhibition may arise from a common set of pathogenic mechanisms. This supports emerging models of stuttering, which propose that poor fluency may be part of a broader network of impaired self-regulatory processes.

KEY WORDS: stuttering, nonfluency, attention, twins

Recently, there has been increased interest in topics addressing the intersection between unstable fluency and attentional self-regulation in young children diagnosed with stuttering. Although most children who stutter (CWS) do not have frank attentional disorders, there is growing evidence that, relative to their fluency-normal peers, young CWS are more likely to be perceived by parents as having some attentional difficulties (Anderson, Pellowski, Conture, & Kelly, 2003; Guitar, 2006; Karrass et al., 2006). Among the attention-related problem behaviors that have been reported for CWS are high distractibility and difficulties with or slow shifting of focus (Embrechts, Ebben, Franke, & van de Poel, 2000; Karrass et al., 2006; Monfrais-Pfauwadel & Lacombe, 2002; Riley & Riley, 2000; Schwenk, Conture, & Walden, 2007). Moreover, a positive association between having ever stuttered and having an attention-deficit disorder has been reported in two retrospective studies of adults (Alm & Risberg, 2007; Biederman et al., 1993). Although these studies have provided suggestive evidence for a relationship between speech fluency and attentional regulation, no studies have provided a complete explanation for how these processes may intersect and whether they may be causally related.

The Development of Attentional Control

Attention is a complex neuropsychological construct. Research has suggested that what we typically call "attention" encompasses three related but separable subsystems: (a) orienting, (b) alertness, and (c) selective and executive attention (Berger, Kofman, Livneh, & Henik, 2007). Orienting and alerting systems develop early and can be measured within the first weeks and months of life. In contrast, the more anterior neural systems required for selective and executive attention (specifically, the anterior cingulate cortex [ACC] and the lateral prefrontal cortex [LPFC]) develop more gradually throughout childhood (Berger et al., 2007; Rueda, Posner, & Rothbart, 2005) and appear to continue developing into adolescence (Davidson, Amso, Anderson, & Diamond, 2006). It appears that executive attention functions begin the consolidation process between 24 and 36 months of age. Between the ages of 3 and 5 years, significant improvements in a variety of executive attentional tasks have been reported (Berger et al., 2007; Posner & Rothbart, 2000). For this reason, these ages are viewed as the most developmentally active period of executive attentional development (Rueda et al., 2005). These are also the ages at which stuttering is most likely to emerge (Guitar, 2006; Yairi & Ambrose, 2005).

Rueda and colleagues (2005) provide a concise definition of *executive attention,* calling it "the network involved in the volitional and controlled aspect of the attention system" (p. 576). Current models (e.g., Posner & Raichle, 1994) suggest that there are three integrated and measurable mechanisms that make up *cognitive attention* (also called *executive attention*): error detection and correction, inhibition, and the resolution of cognitive conflicts, as measured through Stroop and Flanker tasks. Each of these mechanisms is believed to interact with one another and with related internal systems (e.g., temperament) to support the emergence and stabilization of a broader ability known as *self-regulation,* which has been described by Berger and colleagues (2007) as "the ability to monitor and modulate cognition, emotion, and behavior, to accomplish one's goals and/or to adapt to the cognitive and social demands of specific situations" (p. 257) and by Rothbart and Bates (as cited in Berger et al., 2007) as the capacity to modulate a reactive temperament by "engaging behavioral strategies and exerting effortful self-control" (p. 265). Self-regulation is believed to be governed by a group of monitoring mechanisms, in both the cognitive and emotional domains. Because executive attention is involved in inhibitory control, helps to determine strategies for problem solving, and is directly associated with self-monitoring processes, it is considered by some (e.g., Berger et al., 2007; Posner & Rothbart, 2000) to be the central player in determining how "well-regulated" an individual will be. As with many complex traits, there is individual variation in self-regulatory and self-monitoring processes among preschool children (Posner & Rothbart, 2000; Reuda et al., 2005). Although the causal mechanisms underlying these variations have not been established, there is some evidence from twin studies of both emotional and cognitive self-regulation that genetic factors are involved to a moderate degree (Fan, Wu, Fossella, & Posner, 2001; Goldsmith, Buss, & Lemery, 1997).

The Relationship Between Attentional Regulation and Speech Fluency

Speech fluency (apart from stuttering) as a construct has been discussed for many decades by both speech-language pathologists and linguists (see Bloodstein & Ratner, 2008, and Yairi, 1997, for reviews of this literature). Although a number of definitions of speech fluency exist, one of the most concise was offered by Starkweather (1987), who defined *speech fluency* as "the ability to talk with normal levels of continuity, rate, and effort" (p. 12).

The precise variables that determine how customarily fluent a person will be have not yet been established. However, at a general level, we know that in order to produce speech that is relatively free of unwanted repetitions, speech blocks, unnecessary interjections, and false starts, one must be able to detect these speech intrusions, inhibit or correct them either prior to or during their occurrence, and monitor ongoing speech to minimize their production (Levelt, 1983). Some individuals, who would likely be perceived as exceptionally fluent speakers, perform these tasks with a high level of proficiency across speaking contexts, even when speaking under conditions of stress or emotional arousal. Conversely, individuals who display a relatively chronic inability to "gate" these unwanted speech intrusions would likely be perceived as highly nonfluent speakers. Presently, there is no evidence examining the relationship between speech fluency and executive attention in children or adults who are exceptionally fluent versus those who have chronically unstable fluency. However, if such a relationship exists, it would be reasonable to hypothesize that individuals who are exceptionally fluent speakers would possess more well-developed executive attention and self-monitoring skills than those with chronic fluency instability.

Recent models of stuttering development have alluded to the role that executive attention and other self-regulatory processes may play in the emergence of stuttering (Conture et al., 2006; DeNil, 1999; Hubbard Seery, Watkins, Mangelsdorf, & Shigeto, 2007; Smith & Kelly, 1997; Starkweather, Gottwald, & Halfond, 1990). In one recent model, for example, Conture and colleagues (2006) suggested that children who develop stuttering may inherit a biological predisposition to become nonfluent, with some (as yet unknown) proportion also inheriting—presumably independently—a "high-risk" temperament. This reactive temperament is not viewed as directly causing the child to stutter. Rather, it is argued that poor self-regulatory control sets the stage for fluency difficulties by interacting negatively with the many intrinsic and extrinsic factors that are believed to increase the liability for stuttering in constitutionally vulnerable children. Given this background, whether or not a given child begins to stutter or persists in stuttering becomes a function of the complex exposures experienced by that child during developmentally sensitive periods.

To date, no studies have been performed that have examined attentional (or emotional) self-regulation skills in children across the fluency continuum. In contrast, several recent studies have examined whether young CWS display atypical attentional skills and/or have attentional disorders. Most of these studies have used parent report instruments to assess attention, with a minority of studies combining parent report and some form of direct observation. In one such study, Riley and Riley (2000) used a combination of parent report on the Burks Behavior Rating Scales (Burks, 1976) and clinician judgment to determine the prevalence of an attention disorder in a sample of 50 CWS between the ages of 3;0 (years;months) and 9;11 who were examined prospectively. Two findings from this study are of particular interest. First, these investigators concluded that approximately one quarter of their CWS (26%) met their criteria for the presence of a significant attentional deficit. Moreover, among several candidate predictor variables that they examined, the presence of pretreatment attentional problems was found to be the single best predictor of poor treatment outcome at follow-up; that is, children in this sample who stuttered and had poor attention were found to be significantly less likely than those with adequate attention to have a positive treatment outcome at 24–48 months posttreatment, regardless of factors such as pretreatment stuttering severity.

Karrass and colleagues (2006) asked parents of 65 preschool-age children with stuttering and 56 control children to complete the Behavior Style Questionnaire (McDevitt & Carey, 1978), a parent report instrument that measures emotional reactivity, emotional regulation, and attention. Results of this study revealed that the children who stuttered were rated by parents as being significantly more reactive, significantly less able to regulate their emotions, and significantly poorer in attention regulation than children who did not stutter, even after controlling for gender, age, and language abilities.

In a study of particular relevance to the present investigation, Embrechts et al. (2000) asked the parents of 38 Dutch children who stuttered and 38 control children aged 3–8 years to complete the Children's Behavior Questionnaire (Rothbart, Ahadi, Hershey, & Fisher, 2001). This scale places items into 15 temperament categories, including fearfulness, shyness, and sadness. For 10 of the 15 categories, the CWS did not differ significantly from their age-matched peers with normal fluency. There were, however, highly significant group differences on the remaining five categories: activity level (higher for CWS), impulsivity (higher for CWS), attentional focusing (lower for CWS), inhibitory control (lower for CWS), and perceptual sensitivity (lower for CWS). These findings are important because it did not appear that parents of the CWS in this study rated their children as having problematic temperaments or problem behaviors in an "across-the-board" manner. Rather, the concerns that these parents reported were thematically coherent and focused upon perceived difficulties in self-regulation/inhibition, perceptual focus, and attention.

In a study focusing on adults with and without stuttering, Alm and Risberg (2007) asked 32 Swedish adults who stutter and 23 adults with normal fluency to complete a retrospective self-report instrument of attention problems during childhood. Results revealed significant group differences in reports of childhood attention-deficit/hyperactivity disorder (ADHD) symptoms ($p < .02$), with stuttering subjects receiving significantly higher symptom scores. More specifically, these investigators found that 41% of the adults who stutter received an attention score that was "above the maximum score" received by any member of the control group, although most stuttering subjects still fell outside the score range required for a retrospective clinical diagnosis of ADHD.

Limitations of Current Evidence

Although findings relating attention to stuttering are intriguing, several important issues remain to be addressed. Across studies, attention data were obtained from only one observer—usually, the mother. For childhood attention in particular, it has been recently documented that obtaining data from multiple raters and/or contexts (e.g., from both parents and teachers) enhances confidence in the measurement validity of that phenotype (Derks, Hudziak, van Beijsterveldt, Dolan, & Boomsma, 2006). Moreover, these prior data were all collected at only one sampling point, with no repeated measures taken to ensure stability.

The related problems of rater blinding and rater bias are shared by all of the studies that have relied upon parent report in clinically referred samples of CWS. Parents who completed the rating scales used to describe temperament and attention in these studies were clearly not blinded to the fluency status of their children, nor were the investigators who interviewed the parents and interacted with the subjects. In addition to problems inherent with failure to blind, there are potentially serious concerns about rater bias across these studies. As reported by Hauner, Shriberg, Kwiatkowski, and Allen (as cited in Hubbard Seery et al., 2007), parents of children who present with any diagnosed speech or language problem, including stuttering, are likely to report (perhaps, overreport) the presence of a wide array of temperamental difficulties, including "(over)-sensitivity, anxiety, distractibility, neuroticism, withdrawal, and difficulty in adaptability" (Hubbard Seery et al., 2007, p. 208). One approach to safeguard against such bias is to select children from nonclinical (e.g., population-based) samples. Although this type of selection is logistically complex, the advantage is that parents who respond to questions in multiple health and developmental domains will generally be less sensitized to any one particular deficit area when the data are collected.

Finally, as has been well articulated by several researchers (cf. Alm & Risberg, 2007; DeNil, 1999; Hubbard-Seery et al., 2007; Smith & Kelly, 1997; Yairi, 2007), there is a pressing need for scientists to develop explanatory models that can describe the mechanism(s) underlying these complex relationships for stuttering. For example, in the present context, it is possible that both unstable fluency and poor self-regulation of attention might be expressions of a common underlying deficit with shared genetic background. This latter speculation would result in a number of predictions about executive deficits that would be testable and might lead to novel insights about how fluency control fits into a broader neurobiological spectrum of regulatory disorders (Comings, 1994; Comings et al., 1996).

Objectives of the Present Study

The present study was designed to provide additional information about the relationship between fluency and attention in a unique sample of children who were not identified a priori as having either fluency or attentional disorders. Specifically, in this investigation, we obtained reports of fluency and multiple measures of attention from parents (and, for two measures, from teachers) of a large cohort of twins (N = 10,683 pairs) ascertained from the Netherlands Twin Registry (NTR).

Measures of fluency and attention were collected via questionnaire from mothers when children were 5 years of age, and measures of attention were obtained from both parents and from teachers when the twins were 7 years of age. Children were placed into one of three fluency groups—probable for stuttering (PS); highly nonfluent (HNF); and control, or typically fluent (TF)—based on responses to this questionnaire, which asked about fluency behaviors (among many other concerns). The primary objective of this study was to determine if children in the PS and HNF groups performed significantly differently from one another and from age-matched children with typical fluency on measures of attention. A secondary objective was to address the question of potential etiological overlap between nonfluency and attention using the discordant co-twin control method (described later).

Based on prior research, we predicted that children with probable stuttering would display elevated scores on measures of attentional problems at a higher frequency than children with typical fluency abilities. We did not, however, expect to see mean scores on these measures that would be elevated enough to place most of the CWS into the clinically impaired range. Because no comparable studies, to date, have selected subjects specifically on the basis of high nonfluency, we did not have past research to guide our predictions. However, because we view speech fluency as a quasi-continuously distributed threshold trait (Felsenfeld, Kubarych, Aggen, Martin, & Neale, 2005), similar to the liability threshold model for language disability proposed by Dollaghan (2004), we hypothesized that the highly nonfluent children would occupy an intermediate position between the PS and TF groups; that is, we anticipated that the ratings from this group would fall significantly above the ratings of the TF children but below the ratings obtained by the PS children on many of our measures of attentional behaviors. Finally, because we hypothesized that both disrupted fluency and difficulties with attention may arise from a shared underlying deficit in executive functioning that is genetically mediated, we predicted that our discordant co-twin control analyses would reveal the presence of significant shared genetic liability between the fluency and attentional phenotypes.

Method

Subjects

The data presented in this article were derived from a longitudinal study using the NTR, which is maintained by the Department of Biological Psychology at the Free University in Amsterdam (Bartels et al., 2007; Boomsma et al., 2006). From 1987 onward, the NTR has recruited families with twins a few weeks or months after birth. Currently 40%–50% of all multiple births are registered by the NTR.

Surveys were mailed to parents of twins around the 5th birthday of the pair. After a procedure of mailing reminders to nonresponders and (if financial resources were available) contacting persistent nonresponders by phone, a response rate of 66% was obtained. Families whose addresses were not available were included in the nonresponse group. In the present study, data were obtained from questionnaires returned for 20,445 children from birth cohorts 1989 to 1999. A twin pair was excluded when one member of the pair had a handicap or a disease that interfered with normal daily functioning (n = 256 pairs). For the majority of subjects (77.6%), the mother independently completed the fluency questionnaire items. The remaining fluency questionnaires were completed by fathers (4.5%), by both parents (17.7%), or by others (0.2%). For convenience, we hereafter refer to all of these questionnaires collectively as *parental questionnaire responses*.

For 795 same-sex twin pairs, zygosity was based on blood group (n = 12) or DNA polymorphisms (n = 783). DNA and blood collection was done in families who took part in various experimental and laboratory studies being performed through the NTR (Boomsma et al., 2006), including studies that investigated the genetics of cognition, brain function, and structure in children; ADHD; and the genetics of twinning. For the remaining same-sex twins, zygosity was based on questionnaire items dealing with similarity in physical characteristics (hair color, eye color, face color, facial appearance) and frequency of mistaking one twin for another by parents, relatives, and strangers (Goldsmith, 1991; Rietveld et al., 2000). The classification of zygosity was based on a predictive discriminant analysis, relating the questionnaire items to zygosity based on blood/DNA typing in a group of same-sex twin pairs (for a detailed description of the procedure, see Rietveld et al., 2000). Complete data on zygosity questions and on genetic markers of blood profiles were available for 768 twin pairs. According to this analysis, the zygosity was correctly classified by questionnaire in nearly 95% of the cases. If zygosity information was missing at age 5 years, zygosity status was determined by items from questionnaires sent at other ages. For 33 twin pairs, zygosity could not be established, and these pairs were omitted from the genetic analyses.

Procedures

Classification into PS and HNF groups. Children were classified as HNF or as PS on the basis of parental responses to six fluency items. These items asked the mother to evaluate "how often" the following behaviors occurred during the child's typical conversational speech: (a) repeating part of a sentence (e.g., a phrase); (b) slowly repeating whole words in a sentence; (c) rapidly repeating whole words in a sentence; (d) repeating part of a word (e.g., a syllable); (e) experiencing blocks at the beginning or middle of a word; and (f) prolonging a sound within a word. Each question was presented with a model or a verbal example by the interviewer and was rated using a 5-point scale (1 = *never,* 2 = *rarely,* 3 = *sometimes,* 4 = *often,* and 5 = *very often*). Items 1–3 are nonfluency behaviors that are observed in varying degrees in typically developing children of this age (Ambrose & Yairi, 1999; Guitar, 2006; Yairi & Clifton, 1972). In contrast, Items 4–6 correspond to the classic triad of core stuttering behaviors (part-word repetitions, blocks, and sound prolongations) that are traditionally used as diagnostic indicators of incipient stuttering in English (Ambrose & Yairi, 1999; Pellowski & Conture, 2002; Yairi & Ambrose, 2005) and in Dutch (Boey, Wuyts, Van de Heyning, De Bodt, & Heylen, 2007).

In the present study, children were classified as PS if they were reported to display one or more of the three core behaviors (Items 4–6) *often* or *very often.* Children were classified as HNF if they were reported to display at least two of the three nonfluent behaviors (Items 1–3) *often* or *very often* and reportedly produced the three core behaviors infrequently (*never, rarely,* or *sometimes*). Subjects who met our criterion for PS could also display concomitant nonfluency (i.e., they could have met our criterion for both fluency-affected groups). This occurred for 41% of the children (342 of 826) in the PS group. It should be noted that information about whether any of these children had received an independent (clinical) diagnosis of stuttering was not obtained.

Children whose fluency was considered "typical"—in that they did not meet our criterion as either PS or HNF—were classified as TF controls. A large percentage of these children, approximately one third, already exhibited highly stable fluency at this age, with mothers reporting that all six of the questionnaire items about speech nonfluency essentially "never occurred." A descriptive overview of the classification scheme is provided in Table 1.

Validity of the fluency classification system. The fluency status of children in this study was determined on the basis of responses to six behavioral items obtained when children were age 5 years. This classification procedure is consistent with other recent population-based studies of communication disorders in which the direct assessment of all study subjects (for the present study, more than 20,000 children) is not feasible (see, e.g., Bishop, Laws, Adams, & Norbury, 2006; DeThorne et al., 2006; Spinath, Price, Dale, & Plomin, 2004). For stuttering, it has been recognized for some time that mothers typically offer reliable reports about the presence of stuttering-like dysfluencies in their young children (Curlee, 1999; Yairi & Lewis, 1984). Moreover, it has been found that mothers' concerns about the presence of stuttering correlate highly with the diagnosis of incipient stuttering by professionals (Curlee, 1999; Johnson et al., 1959; Yairi & Ambrose, 2005). Ideally, the descriptive fluency items used in the present study should be validated against other measures of fluency outcome, such as self-report and/or therapy records. However, the items used in this study can be viewed as measuring parental

Table 1. Criterion for fluency group membership.

Fluency group	Description	Criterion
PS	Children are reported to display one or more of the three "core" behaviors of beginning stuttering.	Mother reports that at least one of the following core stuttering behaviors occurs "often" or "very often": 1. Rapidly repeats part of a word (e.g., muh-muh-muh-may I go?"). 2. Blocks at the beginning or middle of a word (example: "I . . . go to school"). 3. Prolongs a sound within a word (example: "I go to sssssssschool.")
HNF	Children are reported to exhibit fluency that is chronically unstable but does not contain frequent occurrences of the core stuttering behaviors described above. These children's speech would be described as sounding disorganized, fragmented, and marked by the unnecessary repetition of words and phrases.	Mother reports that at least two of the following behaviors occur "often" or "very often": 4. Repeats a part of a sentence (example: "And then he..and then he came home"). 5. Slowly repeats a word in a sentence (example: "I. . .I. . .I go to school"). 6. Rapidly repeats a word in a sentence (example: "III go to school"). AND ➢ The three core stuttering behaviors described above are reported to occur "never," "rarely," or "sometimes."
TF	Children demonstrate speech fluency that is typical for age.	Approximately one third of these children (34%) had received ratings of "never" on all six of the fluency items.

Note. PS = probable stuttering; HNF = highly nonfluent; TF = typically fluent.

perceptions of fluency difficulties that were of sufficient concern to be noted as unusual on a health survey.

Measures of attention. At age 5 years, parents independently completed a short (42-item) version of the Deveruex Child Behavior scale (DCB; Spivak & Spotts, 1966; Van Beijsterveldt, Verhulst, Molenaar, & Boomsma, 2004). The DCB asks the parent to rate his or her child's behavior over the preceding 2 months using a 5-point scale (1 = *never,* 5 = *very frequently*). The five-item Attention subscale from this survey was selected for analysis. This subscale includes items such as "is distracted by others," "does not attend to an activity," and "jumps from one activity to another."

At age 7 years, data on attention problems and (hyper)activity as rated by both parents and the teacher were obtained for all children, irrespective of their fluency status. Parental reports of these problems were measured with the Child Behavior Checklist/Ages 4–18 (CBCL/4-18; Achenbach, 1991a) and by the short form of the Conners' Parent Rating Scale—Revised (CPRS–R; Conners, Parker, Sitarenios, & Epstein, 1998). The CBCL/4-18 is a standardized questionnaire that consists of 113 items and measures the frequency and intensity of behavioral and emotional problem behaviors exhibited in the past 6 months in 4- to 18-year-old children. The items are rated on a 3-point scale: 0 = *not true,* 1 = *somewhat or sometimes true,* and 3 = *very often or often true*. The Attention Problem scale of the CBCL/4-18 consists of 11 items, including "can't concentrate," "is impulsive," and "can't sit still." The retest reliability of the Attention Problems subscale is .92, and the internal consistency is .86. The cross-informant agreement between maternal and paternal reports for the Attention Problems subscale is .73.

The CPRS–R is another standardized parent report scale that is used to assess problem behaviors that have occurred in the past month (Conners, 2001; Conners, Parker, et al., 1998). Items are rated on a 4-point scale (0 = *not true at all,* 3 = *very much true*). The short version contains 27 items (28 items for teachers) and includes a Cognitive Problems/Inattention subscale (6 items); a Hyperactivity subscale (6 items; 7 items for teachers); and an ADHD index (12 items). Psychometric information for the parent-report subscales of the CPRS–R is provided in the test manual (Conners, 2001). Results for the subscales used in this study are as follows: internal consistency/retest reliability coefficients for inattention are .92/.73, for hyperactivity are .87/.85, and for ADHD are .93/.72. In addition, the Attention subscales of the CBCL/4-18 and the CPRS–R have recently been shown to correlate (r = .45–.77) with *Diagnostic and Statistical Manual of Mental Disorders* (4th ed.; DSM-IV; American Psychiatric Association, 1994) diagnoses of ADHD that are derived from a structured diagnostic interview (Derks et al., 2008).

We asked teachers to rate children's behavior problems at age 7 years using the Teacher's Report Form (TRF; Achenbach, 1991b) and the Conners' Teacher Rating Scale—Revised (CTRS–R; Conners, Sitarenios, Parker, & Epstein, 1998). The scales of the TRF and the CTRS–R are similar to those described for the CBCL/4-18 inventory and the CPRS–R in their focus on attention and activity. When interpreting the teacher ratings, it is important to recall that teachers provided these ratings as a matter of course for all study subjects. Accordingly, the subjects for this particular study were not identified as participants in a study of speech fluency, and fluency concerns were never discussed with the teacher.

Internal consistency/retest reliability, respectively, for the relevant subscales of the CTRS–R are as follows: inattention, α = .89/.92; ADHD, α = .94/.80; and hyperactivity, α = .92/.72. The correlation between the parent and teacher ratings is modest but significant for all three scales: inattention, α = .47; ADHD, α = .49; and hyperactivity, α = .37. The psychometric coefficients for the Attention subscale of the TRF are as follows: internal consistency, α = .79, and retest reliability, α = .87.

The individual items included on the Attention and Activity subscales used in the present study (both parent and teacher forms) can be found in the Appendix.

Analyses

Between-group analyses. For each measure, raw scores were converted to standardized scores (t scores; $M = 50$, $SD = 10$). The t scores were then compared across all three groups using the SPSS MIXED procedure. The MIXED procedure is so named because it refers to the utilization of a mixed model, which permits the inclusion of both nested and fixed factors. Because twin data consist of nonindependent observations (co-twin scores are nonindependent), this procedure "nests" individuals within twin pairs. In our model, Attention scores were the dependent variables, and Fluency Group (PS, HNF, TF) and Gender were included as fixed factors. When the F value for a subscale reached significance ($p < .01$), post hoc tests were subsequently performed using the TEST command (which performs univariate post hoc comparisons) of the MIXED program. Because multiple comparisons were performed, between-group significance was set at a conservative level of $p \leq .01$.

Discordant co-twin control analyses. To address the possibility of some shared genetic association between fluency and problem behaviors, two analyses using fluency-discordant twin pairs were performed, following guidelines described in Cederlof, Friberg, and Lundman (1977) and Kendler et al. (1993). In the present study, *discordant twin pairs* were defined as those in which one member of the pair was classified as positive for presence of a fluency problem (either PS or HNF), and the other member of the pair (the co-twin) was classified as TF. Briefly, the rationale for discordant co-twin analyses is as follows: If shared genetic or environmental factors underlie the expression of potentially related problem behaviors such as poor fluency and poor attention, then twins who themselves do not exhibit Problem A (e.g., poor fluency) would still be at an elevated risk for the expression of the associated Problem B (e.g., poor attention) if their co-twin was affected with Problem A. If shared genes are relatively more important than shared environment in these cross-phenotype associations, then we would expect that identical (monozygotic [MZ]) twins will resemble one another more closely than will fraternal (dizygotic [DZ]) twins, considering MZ twins share more genetic material (100% for MZ vs. 50% for DZ).

Results

Prevalence

The overall prevalence rate that we obtained for PS was 4.0% (826 of 20,445), which is consistent with published estimates of the population prevalence of stuttering in children of this age (Guitar, 2006). The ratio of males:females affected found in this study was 1.5:1, which is also consistent with past epidemiological research of young children who stutter (Yairi & Ambrose, 2005). In the present study, the overall prevalence of the nonfluency phenotype was 2.7% (547 of 20,445), with a male:female ratio of 1.6:1. Because this is the first study to report population estimates for nonfluency, there are no data against which these obtained values can be compared. The prevalence values obtained from this large population sample for all three fluency subgroups are presented in Table 2.

Attention Measures

To examine the relationship among nonfluency, stuttering, and attention in this sample, t scores obtained from subjects in the two fluency-affected groups at ages 5 and 7 years on the attention measures were compared with one another and with the large group of TF children. Initially, maternal and paternal ratings were examined separately. However, because there were no significant differences between mean maternal and paternal scores for any of the attention measures, the parental scores

Table 2. Prevalence by fluency group.

Fluency group	Frequency	Percent of sample	Gender ratio (M:F)
TF	19,072	93.3	0.95:1
PS	826	4.0	1.5:1
HNF	547	2.7	1.6:1
TOTAL *N*	**20,445**		

Note. There were 249 potential subjects (1.2% of the eligible sample) who were excluded because of missing data.

were subsequently averaged for each subject, resulting in a midparent *t* score. (This was done to reduce the number of statistical comparisons.) Results of these analyses are presented in Table 3.

As can be seen in Table 3, results of the omnibus analyses for the attention domain were significant for all measures at ages 5 and 7 years. Thus, as we had hypothesized, children identified as PS received significantly higher (more problematic) scores than did TF subjects on all midparent measures of attention and hyperactivity at both ages ($p < .002$ for all measures).

Of particular interest in the present investigation were the attention scores received by children in the HNF group. Our hypothesis that this group would receive Attention ratings that were at an intermediate level between the PS and TF children was not confirmed by the midparent data. Instead, the HNF children were observed to perform more similarly to the PS children than we had expected. As had been found for the PS children, subjects in the HNF group were rated as having more attentional deficits and more hyperactive tendencies than the TF children at ages 5 and 7 years on all measures of attention and activity ($p < .002$ for all comparisons).

Table 4 displays results for the attention measures that were provided by teachers of children at age 7 years. Children in the PS group were rated by teachers as having significantly more attention problems than the controls (TRF Attention Problems and CTRS–R Attention Problems subscales; $p < .01$) but were not considered to be significantly more hyperactive. Teacher ratings for the HNF children were more mixed. The HNF children were rated by teachers as having more problems on one attention measure (CTRS–R Attention Problems; $p < .01$) but not the other (TRF Attention Problems). As was observed for the PS group, the HNF children were not significantly different from the TF subjects on either measure of hyperactivity. Children in the PS and HNF groups once again performed similarly to one another when teachers provided the ratings, with one exception: Teachers rated the PS children as having significantly more attention problems than the HNF children on the TRF Attention Problems subscale.

Magnitude of Group Differences

In order to describe the practical magnitude of the mean differences between the groups, effect size values (denoted as *d*) were obtained for each of the attention measures. To obtain this value, we subtracted the mean midparent attention *t* score of the TF group from the mean midparent scores of the PS and HNF groups, respectively, for each measure. This product was then divided by the *SD* of the TF group to yield an effect size.

Cohen (1988) has provided conventions for interpreting the magnitude of effect sizes: Each can be considered small (d = .10–.30), medium or moderate (d = .40–.60), or large (d = .70–1.00). To provide some reference for these values, a moderate effect size of .50, for example, would indicate that the difference between the means of the two groups (e.g., the PS vs. the TF group) is about one half of an *SD* (Portney & Watkins, 1993). According to Portney and Watkins (1993), *medium* or *moderate effect sizes* are those that are usually "large enough to be visible to the naked eye" (p. 652). More concretely, effect sizes in the moderate and large ranges have been found to correlate highly with judgments of clinically significant outcomes or group differences in various specialties of medicine (Farone, 2008). Table 5 presents the effect size (*d*) values and Cohen interpretation for all measures of attention in the present study.

As shown in Table 5, effect sizes were in the moderate range for all measures for both fluency-affected

Table 3. Midparent *t* scores (*M*s and *SD*s) for the five attention measures by fluency group.

	TF group			PS group			HNF group			
Measure	*N*	*M*	*SD*	*N*	*M*	*SD*	*N*	*M*	*SD*	*F*
Age 5 years										
DCB Attention Problems	19,033	49.7	9.0	822	54.0[b]	10.4	546	54.1[b]	10.0	101.7[a]
Age 7 years										
CBCL/4-18 Attention Problems	12,219	49.8	9.4	482	54.5[b]	11.1	332	55.3[b]	11.2	79.7[a]
CPRS–R Attention Problems	5,927	49.8	9.4	214	53.8[b]	11.5	177	54.0[b]	11.1	14.0[a]
CPRS–R ADHD	5,948	49.8	9.4	214	54.1[b]	11.2	178	54.3[b]	11.0	16.6[a]
CPRS–R Hyperactivity	5,954	49.8	9.3	215	53.9[b]	12.2	178	53.3[b]	10.9	20.1[a]

Note. DCB = Devereux Child Behavior scale; CBCL4-18 = Child Behavior Checklist/Ages 4–18; CPRS–R = Conners' Parent Rating Scale—Revised.

[a]*F* value is significant at $p < .001$. [b]Affected group is significantly different from controls at $p \le .002$.

Table 4. Teachers' *t* scores (*M*s and *SD*s) for the attention measures for the three fluency groups at age 7 years.

	TF group			PS group			HNF group			
Measure	*N*	*M*	*SD*	*M*	*M*	*SD*	*N*	*M*	*SD*	*F*
TRF Attention Problems	4938	49.8	9.9	184	55.1[b,c]	11.6	132	50.4	10.6	10.9[a]
CTRS–R Attention Problems	3750	49.7	9.7	127	55.8[b]	12.3	104	53.1[b]	12.5	18.0[a]
CTRS–R ADHD	3821	49.8	9.9	138	53.3	11.3	107	51.4	12.0	2.66
CTRS–R Hyperactivity	3826	49.9	9.9	138	52.5	11.4	107	50.7	11.2	1.6

Note. TRF = Teacher's Report Form; CTRS–R = Connors' Teacher Rating Scale—Revised.

[a]*F* value is significant at $p < .01$. [b]Fluency-affected group is significantly different from TF group at $p < .01$. [c]PS group is different from HNF group at $p < .01$.

groups, with the exception of the hyperactivity index for the HNF group, where the effect size was small. Overall, the two fluency-affected groups received attention ratings that placed them approximately one-half of an *SD* above the mean of the TF children, with a range of .38 to .58 *SD* units.

To further explore the practical magnitude of the present findings, the percentage of children who received midparent *t* scores at or above 65 was obtained for each subject group for all attention measures. A *t* score of 65 was selected because it is 1.5 *SD*s above the mean of the normative group and is often considered to be a cutoff value for a clinical referral. These proportions were subsequently used to obtain an odds ratio (OR) for each attention measure. Briefly, an OR is a relative measure of risk that indicates how much more likely it is that someone who is "positive" for a factor under study (in this case, is a PS or an HNF speaker) will manifest a particular outcome (a *t* score of 65 or higher on an attention measure) when compared with someone who is "negative" for that factor (i.e., is a TF speaker). Typically, ORs are expressed as values greater than 1.0. For example, when comparing the outcome of two independent groups, an OR of 2.70 would suggest that members of one group were found to be approximately 2.7 times more likely than members of the alternate group to experience a given outcome under study.

The percentage of children in each of our fluency groups who received a midparent *t* score of 65 or greater on the attention measures, and the associated OR for each measure, are presented in Table 6.

Table 6 reveals that most children in the PS and HNF groups (over 80%) received attention scores that fell below a threshold criterion for clinical concern. However, when compared with the TF children, a higher proportion of both PS and HNF children did exceed this threshold. ORs indicated that on average, PS and HNF children were about 2.5 times more likely than the TF children to have received *t* scores of 65 or greater on one or more of the attention measures (range of 1.97–2.80 times more likely). Interestingly, neither the PS nor HNF children were found to be significantly more likely than the TF children to receive a clinically elevated *t* score on the CPRS–R Hyperactivity subscale. This suggests that, to the extent that clinically concerning attention behaviors are present for children in either fluency-affected group, the problems are more likely to reflect poor focus and concentration than chronic overactivity.

Discordant Co-Twin Control Analyses

To address the possibility of some shared genetic association between fluency and attention, two analyses using fluency-discordant twin pairs were performed. For the first analysis, we identified (a) all of the *fluency-concordant pairs,* defined as those in which both twins had typical fluency, and (b) all *fluency-discordant pairs,*

Table 5. Effect size values comparing the PS and HNF groups with the TF group for the five attention measures.

Attention measure	Effect size (PS vs. TF)	Cohen interpretation	Effect size (HNF vs. TF)	Cohen interpretation
DCB Attention Problems	.48	Moderate	.50	Moderate
CBCL Attention Problems	.50	Moderate	.58	Moderate
CPRS–R Attention Problems	.43	Moderate	.45	Moderate
CPRS–R ADHD	.46	Moderate	.48	Moderate
CPRS–R Hyperactivity	.44	Moderate	.38	Small

Table 6. Percentage of children in each group with midparent *t* scores of 65 or greater on measures of attention and associated odds ratios displaying relative risk.

Attention measure	TF group	PS group	Odds ratio (95% CI)	HNF group	Odds ratio (95% CI)
DCB Attention Problems	6.4	16.1	2.80* (1.10–7.13)	15.9	2.77* (1.08–7.04)
CBCL Attention Problems	7.4	17.2	2.60* (1.06–6.31)	17.8	2.71* (1.11–6.55)
CPRS-R Attention Problems	8.4	18.2	2.42* (1.03–5.67)	15.3	1.97 (0.82–4.70)
CPRS-R ADHD	7.7	18.7	2.75* (1.15–6.57)	16.9	2.43* (1.01–5.87)
CPRS-R Hyperactivity	8.1	16.3	2.20 (0.92–5.28)	16.3	2.20 (0.92–5.28)

Note. CI = Confidence interval.

$^*p < .05$.

defined as those in which one twin had normal fluency and the other twin was "fluency-affected" (either PS or HNF). For the concordant TF pairs, the first-born member of the pair was designated as the *index twin.*

Our interest was in examining the maternally rated attention scores received by the TF twin in these two groups. (Maternal scores rather than midparent scores were used in these analyses because they were more complete and therefore maximized the sample size.) If shared genes and/or the shared environment are implicated in the associations between fluency and attention, then we would expect the mean attention scores of the TF twins in the discordant pairs to be higher than the scores received by the TF index twins in the concordant TF pairs.

As illustrated in Table 7, the results of this analysis were consistent with these predictions for all measures of attention provided by mothers, with the exception of the CPRS–R Hyperactivity subscale (p = .054). When compared with children from concordant TF pairs (e.g., from pairs in which both twins had normal fluency), children who themselves had normal fluency but whose co-twin did not receive modestly but significantly elevated scores across maternally rated measures of attention. Put differently, our results suggest that having a co-twin who is fluency affected appears to place the fluency-typical member of the pair at an increased risk for receiving elevated Attention scores (but not Hyperactivity scores). These results also reduce the likelihood that rater bias or inflation can explain these associations, considering that the elevated Problem Behaviors scores in this analysis were received by children who had not been perceived by mothers as exhibiting a fluency problem.

In the second discordant co-twin-control analysis, we took a preliminary look at whether shared genes, the shared environment, or both might be primarily implicated in these cross-phenotype associations. To accomplish this, three groups of fluency-discordant pairs that varied in genetic relatedness were created. The first of these—the genetically unrelated (UR) group—was composed of "pseudo-pairs," formed so that one member was randomly selected (i.e., either the first- or the second-born twin within the pair) from all of the concordant fluency-affected pairs (UR-affected). The unaffected (i.e., TF) controls for these subjects were randomly selected from the larger group of concordant TF pairs. To account for gender differences—specifically, the higher proportion of boys in the UR-affected group—the cases and controls were made gender-proportional. The two remaining groups for this analysis were created by identifying all same-sex fluency-discordant pairs in the sample and distinguishing them by zygosity (i.e., into discordant MZ [MZd] and discordant DZ [DZd] pairs).

The UR pseudopairs share random (population base rate) numbers of segregating genes—the DZ twins sharing, on average, 50% of their segregating genes, and the MZ pairs sharing 100% of their segregating genes. An additional and important assumption of this design is

Table 7. Mean maternal *t* scores (and *SD*s) for attention measures for TF twins with either a fluency-affected (PS or HNF) or a TF co-twin.

Measure	*N*	TF twin with a PS or an HNF co-twin	*N*	TF index twin with a TF co-twin	*p*
DCB Attention Problems	821	51.85 (10.66)	7,191	49.80 (9.79)	< .01
CBCL Attention Problems	490	51.85 (11.01)	4,610	49.88 (9.93)	< .01
CPRS–R Attention Problems	231	52.23 (11.30)	2,186	49.51 (9.43)	< .01
CPRS–R ADHD	230	52.25 (9.70)	2,200	49.61 (9.70)	< .01
CPRS–R Hyperactivity	232	51.35 (10.45)	2,206	50.03 (9.84)	.054

that MZ and DZ pairs have a generally comparable shared environment; in other words, there are no differences in the common environment that MZ and DZ twins experience, on average. As before, the dependent variables of interest across these three groups were maternal scores on the attention measures at ages 5 and 7 years. Table 8 summarizes the attention scores obtained from subjects in these fluency-discordant groups (unrelated [URd], MZd, and DZd).

As can be seen in Table 8 and Figure 1, the results that we obtained do support, in part, the possibility that factors shared by biological twins may mediate the association between fluency and the attentional behaviors we examined. The first thing to note in Figure 1 is that, as expected, the fluency-affected (PS or HNF) members of the discordant pairs (the black bars) did tend to receive higher scores on the attention measures than did the TF members of the pair (the white bars). Also, consistent with our predictions, the most score-discrepant of the groups were the URd individuals. For this group, all attention scores between the fluency-affected and unaffected members of the pseudopairs were significantly different at $p < .01$, with the affected member of the pair receiving higher mean scores. Scores for individuals who were genetically related (the true twin pairs) were more similar to one another than were scores obtained from the pseudopairs. However, contrary to what we would expect to see if shared genes were the exclusive contributors to this association, there was no particular trend for the MZd subjects to be more alike in scores than the DZd subjects.

Discussion

The present study is the first to use a twin design to provide evidence for the presence of common mechanisms that may influence the expression of speech fluency and attentional regulation in children. To examine these relationships, maternal, paternal, and teacher ratings of attention were obtained at two time points (ages 5 and 7 years) from a large population sample of twin children who varied in reported fluency stability. These children were participants in a longitudinal twin study in the Netherlands; as such, they were not selected because of reported problems with either fluency or attention. In addition, by examining cross-twin attention data in fluency-discordant pairs, the present study has provided preliminary evidence that disruptions in speech fluency and attention may have shared etiological roots.

The primary results of the across-group analyses suggest that there is a relationship between speech fluency and attention. These findings affirm those of earlier studies that reported a positive association between the presence of attentional problems and/or impulsivity in clinical samples of persons who stutter (Alm & Risberg, 2007; Conture et al., 2006; Embrechts et al., 2000; Karrass et al., 2006; Monfrais-Pfauwadel & Lacombe, 2002; Riley & Riley, 2000). The present study expands upon this knowledge by suggesting that these associations are present even among children who are ascertained from the general population, most of whom (particularly in the HNF group) would not be viewed as having a fluency disorder by those who were concomitantly rating their attention.

More specifically, in the present study, children who were both PS and HNF were rated by mothers and fathers as displaying significantly more attentional problems at ages 5 and 7 years than were TF children on each of five attention-related rating scales we examined ($p < .002$ for all midparent t scores for both groups). Interestingly, there were no significant differences between the independent ratings of fathers and mothers on any measure and for any group, although there was a slight trend for maternal scores to be higher for some measures.

The magnitude of the differences between fluency-affected (PS and HNF) subjects and TF subjects in the present study was moderate for all measures except Hyperactivity, where the effect size was small. In practical terms, the fluency-affected children (PS and HNF) scored about one half of an *SD* above the TF mean across attention measures. These mean differences suggest that most children in the PS and HNF groups would probably not be perceived as having a clinically concerning attentional disorder. Nonetheless, from a theoretical perspective, the consistency of our findings across time points, raters, and measures suggests that this elevation in attention ratings, even if subclinical on average, may be highlighting an important pathogenic mechanism for fluency development. It is interesting to note that Finneran, Francis, and Leonard (2009) made a similar argument recently when discussing the theoretical role of subtle (subclinical) attentional difficulties in children with specific language impairment.

To further explore the magnitude of the group differences we obtained, the proportion of subjects in each of the three fluency groups who met our criterion as potentially "attention disordered" (t score of 65 or higher) was identified. These results were compared with two previous studies that used similar types of attention measures to identify CWS whose attention problems were judged to be severe enough to warrant a probable diagnosis of attention-deficit disorder (ADD) or ADHD. The prevalence of co-occurring stuttering and attention problems in these studies ranged from 4% reported by Arndt and Healey (2001) to 18% reported by Biederman et al. (1993) to 26% reported by Riley and Riley (2000).

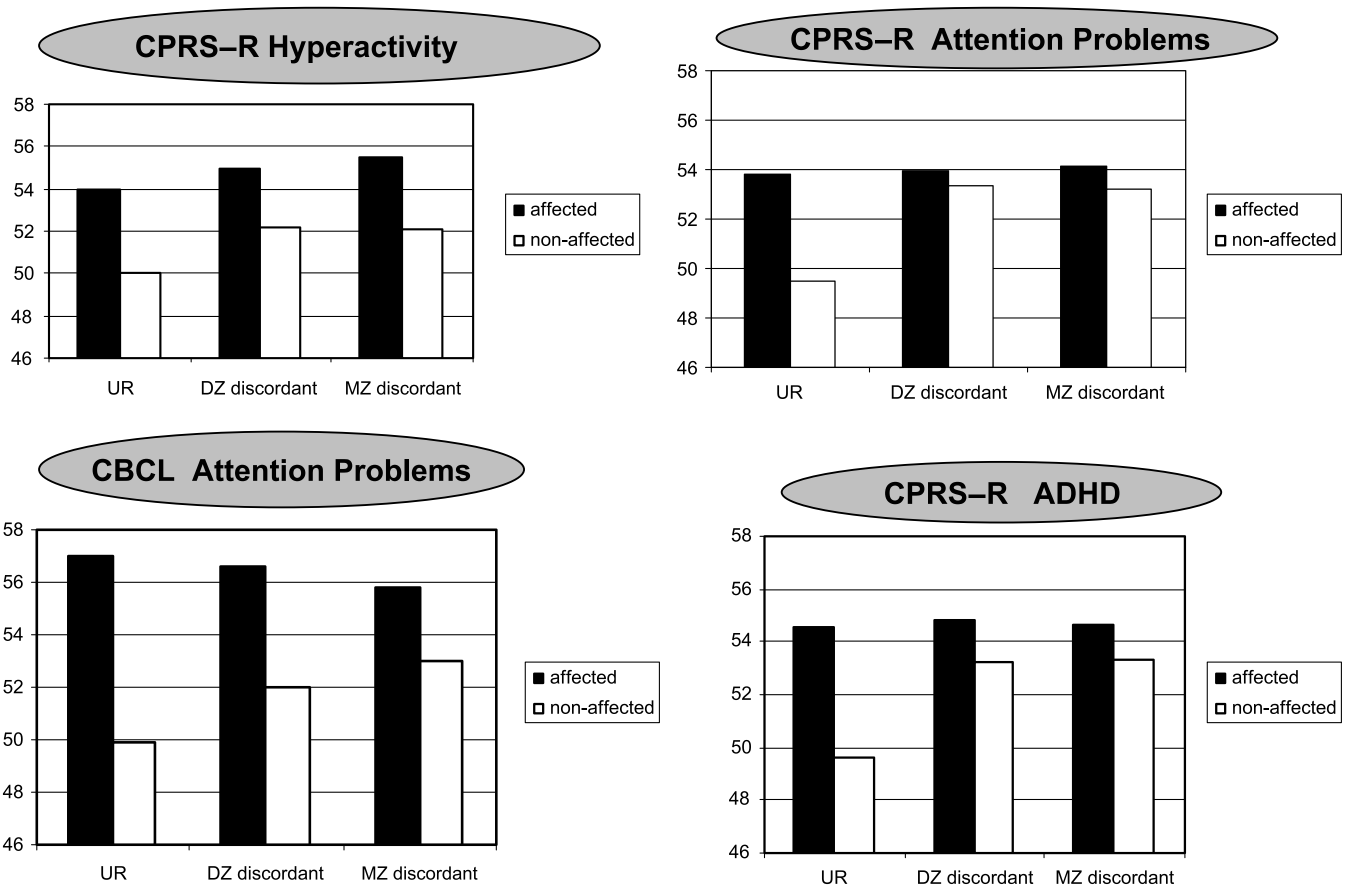
Table 8. Mean maternal attention *t* scores (and *SD*s) in fluency-discordant pairs as a function of genetic relatedness.

	Genetically unrelated groups (URd)					Dizygotic twin pairs (DZd)				Monozygotic twin pairs (MZd)			
	N	Fluency-affected child *t* *M (SD)*	*N*	Fluency-typical child *t* *M (SD)*	*p*	*N*	Fluency-affected twin *t* *M (SD)*	Fluency-typical co-twin *t* *M (SD)*	*p*	*N*	Fluency-affected twin *t* *M (SD)*	Fluency-typical co-twin *t* *M (SD)*	*p*
DCB Attention problems	571	54.19 (11.56)	7,191	49.80 (9.79)	< .01*	306	54.60 (11.78)	51.82 (11.18)	< .01*	211	54.64 (10.92)	51.98 (9.56)	< .01*
CBCL Attention problems	329	56.95 (11.56)	4,610	49.88 (9.93)	< .01*	185	56.62 (13.49)	52.04 (10.77)	< .01*	131	55.83 (11.83)	53.05 (10.07)	< .01*
CPRS-R Attention problems	144	53.81 (12.12)	2,186	49.51 (9.43)	< .01*	87	53.95 (10.72)	53.27 (10.87)	.72	70	54.13 (11.44)	53.22 (9.83)	.49
CPRS-R ADHD	147	54.57 (12.50)	2,200	49.61 (9.70)	< .01*	87	54.84 (11.07)	53.20 (11.98)	.32	69	54.66 (10.82)	53.36 (9.87)	.31
CPRS-R Hyperactivity	148	53.94 (13.39)	2,206	50.03 (9.84)	< .01*	88	54.93 (11.96)	52.17 (11.32)	.09	70	55.48 (12.09)	52.09 (10.16)	< .01*

Note. URd = genetically unrelated cases and controls; DZd = dizygotic twin pairs; MZd = monozygotic twin pairs.

*Intrapair scores differ at $p < .01$.

Figure 1. Maternal attention *t* scores at age 7 years for the three fluency-discordant groups. CPRS–R = Conners' Parent Rating Scale—Revised; UR = genetically unrelated pseudopairs; DZ = dizygotic; MZ = monozygotic; affected = probable stuttering or highly nonfluent; non-affected = typically fluent; CBCL = Child Behavior Checklist; ADHD = attention-deficit/hyperactivity disorder index.



Similar to Biederman and colleagues, we found that between 15% and 19% of the PS and HNF children exceeded our "ADD threshold," compared with only 6%–8% of the TF children. ORs for each attention scale were subsequently obtained to provide an index of relative risk for the expression of a "probable attentional disorder." Results of these analyses demonstrated that children in both the PS and HNF groups were approximately 2.5 times more likely than TF children to exceed our clinical threshold on the attention (but not hyperactivity) measures.

Children who participate in the Netherlands Twin Study are rated by teachers on a number of behavioral and achievement variables. For the present study, teacher ratings of attention and activity at age 7 years were selectively examined for all participating subjects. For the PS group, both of the attention measures completed by the teachers (Attention Problems subscale of the TRF and Attention Problems index of the CTRS–R) were significantly elevated relative to the TF group. For the HNF subjects, the CTRS–R subscale reached significance, whereas the TRF subscale did not. Interestingly, there were no significant group differences for either teacher scale measuring hyperactivity (ADHD and Hyperactivity indexes of the CTRS–R). These results provided interesting and independent corroborative support for the existence of perceived attentional deficits (but not hyperactivity) among the fluency-affected children, particularly for the PS group. Given the background of modest interjudge agreement between parents and teachers that has been reported for these subscales, the similar patterns that emerged between parents and teacher ratings in our study were encouraging.

How Are Fluency and Attention Related to One Another?

The results of this study lend support to emerging models that postulate that speech fluency (specifically, stuttering) may be part of a broader network of internal processes that interact with and influence one another and are sensitive to disruptive input from the environment

(Conture et al., 2006; DeNil, 1999; Smith & Kelly, 1997). At present, these models of stuttering etiology emphasize the importance of feedback loops involving the motor control, limbic, and/or auditory systems. In addition, as is implicit in these models, the feedback-driven systems that are implicated in fluency regulation require a central (executive) mechanism to function: After all, output must be detected before it can be modified. One of the contributions of the present findings, therefore, is to enhance the profile of executive attentional processes in models that describe the development and maintenance of speech fluency, both in speakers who stutter and in those who do not.

Our results suggest that children with unstable fluency are significantly more likely than children with typical fluency to have elevated scores on scales assessing self-regulation/inhibition, as measured by items such as "can't concentrate," "is inattentive or easily distracted," "daydreams," "fidgets with hands," and "has difficulty engaging in tasks." Overall, when compared with their TF peers, the fluency-affected children in our study were consistently rated as having more difficulty focusing upon or engaging in desirable activities and regulating their behavior to inhibit undesirable activities. If one is willing to consider speech nonfluencies (very loosely) as "undesirable activities" or "errors" that are not properly detected or inhibited at a cognitive level (Kolk & Postma, 1997), then a theoretical relationship between nonfluent speech and other self-regulatory problems is logically coherent.

Although the present study cannot unequivocally indicate whether unstable attention and unstable fluency arise from shared sources or are etiologically independent of one another, the results of both of our discordant cotwin analyses provide some provocative clues. First, our data indicate that the relationship between unstable attention and unstable fluency is complex and that both shared genes and shared environmental factors are involved in any etiological association that exists between these two traits. The absence of a clear MZ–DZ difference in attention scores within the fluency-discordant pairs suggests that although common genes may play a role in increasing the susceptibility of children to express a range of self-regulatory problem behaviors (i.e., unstable fluency and poor attention), shared environmental factors may also turn out to be of considerable importance for understanding these associations in young children. This finding is similar to that reported by Goldsmith et al. (1997), who found that "effortful control" (a precursor of self-regulation) was significantly mediated by both underlying additive genetic and shared environmental effects in their sample of twin toddlers.

Second, our results reveal that the complex underlying factors that create unstable fluency within a family also appear to increase the liability for disrupted attention in that family, even among individuals who themselves have typical fluency. Recall that, in our study, children who themselves were fluency typical but whose co-twin was fluency affected had significantly elevated maternal attention scores when compared with twins who were members of concordant fluency-typical pairs. Taken together, these preliminary results bring into question models of stuttering etiology which suggest that "high reactivity" and stuttering are necessarily independently acquired traits (e.g., Conture et al., 2006). An alternative explanation, supported by our data, is that the liability to express both high nonfluency and problems with self-regulation (attention regulation and perhaps other self-regulatory problems) may arise from a common set of mechanisms.

Clearly, it will be necessary to corroborate the descriptive associations between attention and fluency we obtained with more sophisticated behavioral and biological assessments. One model for such an approach has been provided by Comings (1994) and Comings and colleagues (1996), using Tourette syndrome (TS) as the index phenotype. Using sophisticated analytical procedures, these investigators obtained ratings for 20 behavioral problems believed to be related to TS (including stuttering, obssessive–compulsive disorder, oppositional defiant disorder, and ADHD) and then correlated these ratings with molecular genetic markers in TS and control families. Three specific polymorphisms (genetic variants) affecting known dopamanergic genes (DRD2, DβH, and DAT1) were selectively examined. Results of this study suggested that the degree of "loading" for markers of the three dopamanergic genes was associated with problem behavior scores in a surprisingly additive fashion; that is, having all three putative gene markers was associated with a poorer outcome on self-regulatory problem behavior measures than was having only two of the markers, and having only one marker resulted in an even more favorable outcome. These investigators proposed that the disorder phenotypes they examined, although distinct in many ways, might actually represent variants along a spectrum of "impulsive disabilities" (that includes stuttering) whose neurobiological roots could all be traced to deficits in the metabolism of dopamine. Although it remains to be seen whether the dopamanergic system will prove to be central to understanding these relationships in the long term—and, specifically, whether dopamine metabolism will be found to be relevant for stuttering (Wu et al., 1997), the methodological and theoretical approach used in these studies might be relevant for future investigations that seek to explore the concept of an "impulsivity spectrum" in families of persons with a fluency disorder.

Who Are the HNF Children?

Finally, one of the most novel findings of the present study was the degree of similarity observed between the probable stutterers and the highly nonfluent children on measures of attention. These findings suggest that these two groups of children, selected from a population sample entirely on the basis of maternal perceptions of highly nonfluent speech, display a very similar liability for expressing attentional difficulties at both sampling points and when assessed by multiple raters. Recent genetic modeling of these twin data demonstrated that high nonfluency in early childhood is itself a moderately heritable phenotype (van Beijsterveldt, Felsenfeld, & Boomsma, 2010). Taken together, these results suggest that children who are highly nonfluent (but not perceived as stuttering) at age 5 years are an interesting group to study in more detail. Although it is possible that high nonfluency is generally a benign and transient childhood behavior, it is also possible that high nonfluency may be a robust and measurable marker of functionally important self-regulatory problems in children and, potentially, in adults.

Research on the clinical or theoretical significance of high nonfluency (also called *normal disfluency*) in young children has not received widespread attention in the literature. When normal nonfluency in children has been the subject of study, the focus has typically been to either differentiate it from early stuttering (Ambrose & Yairi, 1999; Curlee, 1999; Yairi & Lewis, 1984) or to describe the frequency of various nonfluencies in samples of typically developing children of different ages (see Yairi, 1997, for a review of these studies). In an illustrative investigation of the latter type, Yairi (1981) analyzed the frequency and types of nonfluencies produced by 33 two-year-old children who were not considered by parents or the examiner to have any speech, language, or fluency difficulties. One of the most striking findings of this study was that even at this young age, the nonfluencies produced by these children were continuously, although not strictly normally, distributed. The frequency of nonfluencies per 100 words spoken in this group ranged from 0 to 25.6, with a clustering of scores in the middle range of 5–8 nonfluencies. Two of the subjects were noted to produce a high number of nonfluencies relative to their peers; both were considered fluency outliers, but neither were diagnosed with stuttering. Unfortunately, because nonspeech data about the subjects were not provided, we do not know whether these two HNF cases differed from the remaining subjects for any of the domains included in the present study.

Some investigators have suggested that HNF children perform less well on various measures of language structure and vocabulary than do TF children (Muma, 1971; Westby, 1979), although other studies do not confirm these findings (Haynes & Hood, 1977). In addition, there is a body of clinical work that supports the existence of a "trading off" relationship between expressive language demands and fluency stability in some children who do and do not stutter (Boscolo, Bernstein Ratner, & Rescorla, 2002; Lybolt, 1986; Merits-Patterson & Reed, 1981; Stoker & Usprich, 1976). This relationship appears to be fluid and may reflect how effectively a young speaker can "balance" stable fluency and complex language formulation at a given point in time. Given this background, it is reasonable to speculate that at least a subset of the HNF children in the present sample may have had weak or "overtaxed" language skills at the time their fluency was being rated by parents.

In one particularly informative study examining the language–fluency relationship, Westby (1979) assessed the language skills of three groups of 5- and 6-year-old subjects: (a) children who stuttered; (b) children who were highly nonfluent but nonstuttering; and (c) TF (control) children. Interestingly, Westby observed that both groups of nonfluent children scored significantly more poorly than the control children across the experimenter-administered language measures. When the two nonfluent groups were compared with one another, few significant between-group differences were found. These findings, obtained nearly 3 decades ago, provided early evidence that foreshadowed the present results by suggesting that HNF children may experience some of the same risks as CWS.

Limitations and Future Directions

In the present study, we did not directly assess the attention and fluency performance of our large number of participating children (more than 20,000), and we do not know how well the parental and teacher ratings of fluency that we used would agree with speech diagnoses provided by professionals. Also unavailable at present are data establishing the proportion of children in the study who were receiving services for either attention or fluency disorders. In addition, our study examined only one of the executive attention mechanisms described by Posner and Raichle in 1994. Future work should expand this focus by measuring how children across the fluency continuum perform on tasks such as error detection and correction and the resolution of cognitive conflicts. Finally, implicit in the present study is the assumption that speakers can be reliably identified as members of a fluency category (i.e., "highly fluent," "highly nonfluent," "typically fluent"). At present, data that directly test this assumption are lacking. Future studies should be designed to determine how well listeners agree on prototypical samples representing these fluency categories as produced by both children and adults.

One important challenge for future research will be to refine our understanding of the complex relationship between speech fluency and executive and other self-regulatory skills in children and adults whose speech spans the full fluency continuum, using direct and potentially novel procedures. In addition, future work should focus on developing a model of fluency control that includes the possibility that unstable fluency is part of a broader spectrum of regulatory problems. One starting point for this approach would be to obtain the prevalence of comorbid “impulsive disorders” in families with and without probands who stutter, following methods described by Comings (1994) for TS.

In terms of clinical implications, the present investigation may help to contribute to clinical assessment and treatment of both children and adults. For example, these findings should help to reinforce the need for clinicians to adapt their therapeutic strategies to support children with both stuttering and attentional disorders, considering this subgroup will not be uncommon (Healey & Reid, 2003). For adults, some therapy models have emphasized the important role that executive constructs such as “self-control,” “self-evaluation,” and “self-regulation” may play in facilitating the acquisition of fluent speech and in long-term maintenance of fluency gains (see Finn, 2008, for a recent review). New clinical research could certainly build upon this knowledge. For example, it might be possible to develop treatment modules that explicitly enhance executive attentional skills in older children, adolescents, and adults who stutter and then to measure their effectiveness by comparing outcomes to those of clients who are randomly assigned to receive otherwise comparable treatments that do not include executive skills training.

Finally, our results suggest that young CWS and those who are highly nonfluent may be at an increased risk for (typically mild) problems in attentional regulation. As such, it may be appropriate to monitor this area in young clients with poor fluency to ensure that emerging difficulties are not overlooked. Although standard assessment batteries for stuttering do not typically include parental ratings of attention or executive functioning, these measures might help to identify the subgroup of children who are experiencing these difficulties. In some cases, these findings might be helpful when counseling parents of children who stutter. Although it is still premature to suggest that a definitive link across the behaviors we have examined has been identified, it might be appropriate to suggest to parents that such links are becoming increasingly well supported in the literature. For some, it may be very reassuring to learn that the disparate problems in fluency, attention, and possibly other self-regulatory behaviors that they have been observing in their child might arise from common and coherent etiological roots.

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Appendix (p. 1 of 2). Individual items included on the Attention and Activity subscales used in the present study (both parent and teacher forms).

Subscale items: Parent report scales

DCB Attention Problems (age 5)	CBCL/4-18 Inattention Problems (age 7)	CPRS–R Attention Problems (age 7)	CPRS–R ADHD index (age 7)	CPRS–R Hyperactivity index (age 7)
Jumps from one activity to another	Acts too young for his/her age	Has difficulty doing homework	Is inattentive, easily distracted	Is always "on the go"
Does not attend to activity	Can't concentrate; can't pay attention for long	Fails to complete assignments	Has a short attention span	Hard to control in malls or while grocery shopping
Does not attend to adults	Can't sit still, is restless, or is hyperactive	Needs close supervision to get through tasks	Fidgets with hands or feet or squirms in seat	Runs about excessively in situations where it is inappropriate
Is distracted by others	Is confused or seems to be in a fog	Has difficulty engaging in tasks	Is messy or disorganized at home or school	Has difficulty waiting in line or awaiting his/her turn in games or groups
Does not finish an activity	Daydreams or gets lost in his/her thoughts	Has trouble concentrating in class	Only attends if it is something he/she is very interested in	Has difficulty playing or engaging in leisure activity quietly
	Impulsive or acts without thinking	Does not follow through on instructions and fails to finish schoolwork or chores	Avoids, expresses reluctance about, or has difficulties engaging in tasks that require sustained mental effort (such as homework)	Restless in the "squirmy" sense
	Is nervous, high-strung, or tense	Avoids, expresses reluctance about, or has difficulties engaging in tasks that require sustained mental effort (such as homework)		
	Nervous movements or twitching		Gets distracted when given instructions to do something	
	Poor schoolwork		Has trouble concentrating in class	
	Poorly coordinated or clumsy		Leaves seat in classroom where remaining seated is expected	
	Stares blankly		Does not follow through on instructions and fails to finish schoolwork or chores Is easily frustrated in efforts Distractibility or attention span is a problem	

Appendix (p. 2 of 2). Individual items included on the Attention and Activity subscales used in the present study (both parent and teacher forms).

Subscale items: Teacher report scales (collected from children at age 7)

TRF/4-18 Attention Problems	CTRS–R ADHD	CTRS–R Hyperactivity
Acts young	Is inattentive, easily distracted	Is restless
Is noisy	Disturbs other children	Is always "on the go"
Fails to finish assignments	Cannot remain still	Leaves seat in classroom where remaining seated is expected
Can't concentrate	Fidgets with hands	Has difficulty waiting his/her turn
Can't sit still	Has short attention span	Runs about excessively in situations where it is inappropriate
Is confused	Only attends if it is something he/she is really interested in	Has difficulty playing or engaging in leisure activity
Fidgets	Distractibility/attention span is a problem	Is excitable/impulsive
Daydreams	Interrupts or intrudes on others	
Can't follow directions	Fails to finish assignments	
Is impulsive	Does not follow through on instructions	
Has difficulty with learning	Is excitable/impulsive	
Is apathetic	Is restless, always up and on the go	
Is clumsy		
Is messy		
Is inattentive		
Stares blankly		
Underachieves		
Is nervous		
Has poor schoolwork		
Fails to carry out tasks		

Note. DCB = Devereux Child Behavior scale; CBCL/4-18 = Child Behavior Checklist/Ages 4–18; CPRS–R = Conners' Parent Rating Scale—Revised; TRF/4-18 = Teacher's Report Form/Ages 4–18; CTRS–R = Conners' Teacher Rating Scale—Revised; ADHD = attention-deficit/hyperactivity disorder.