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Profiles of Everyday Executive Functioning in Young Children With Down Syndrome

Lisa A. Daunhauer,

Colorado State University, Room 447, Behavior Sciences Building, Fort Collins, CO 80523-1570

Deborah J. Fidler,

Colorado State University

Laura Hahn,

Kansas University

Elizabeth Will,

Colorado State University

Nancy Raitano Lee, and

National Institute of Mental Health

Susan Hepburn

University of Colorado, School of Medicine

Abstract

We investigated executive functioning (EF) in children with Down syndrome (DS; $n = 25$) and typically developing (TD) children matched for mental age (MA; $n = 23$) using the Behavior Rating Inventory of Executive Function-Preschool. We sought to (1) compare children with DS to a developmentally matched control group, and (2) to characterize the EF profile of children with DS. Across teacher and parent reports, significant deficits in working memory and planning were observed in the DS group. Parents, but not teachers, of children with DS also reported difficulties in inhibitory control relative to the comparison group. Results extend earlier findings regarding EF impairments in children with DS. The complementary role inhibitory control may play in this profile is discussed.

Keywords

executive function; cognition; Down syndrome; trisomy 21

From childhood, individuals with Down syndrome (DS) have a high probability of demonstrating a distinct cognitive profile including deficits in verbal processing, working memory, and goal-directed behavior (for a review see Daunhauer & Fidler, 2011). DS is the most common neurogenetic syndrome associated with intellectual disability (ID), affecting roughly 1:732 live births (Canfield et al., 2006). Researchers have found that the incidence

of DS increased by 31.1% from 1979 to 2002 (Shin, Besser, Kucik, Lu, Siffel, & Correa, 2009), a trend paralleling increased births by women of advanced maternal ages. However, critical gaps persist in our understanding of the DS cognitive phenotype. Recent evidence suggests that individuals with DS demonstrate deficits in adaptive, goal-directed behaviors known as executive function (EF) skills (Kogan et al., 2009; Lee et al., 2011; Rowe, Lavender, & Turk, 2006). Furthermore, in a review of the DS neuroanatomical phenotype, Nadel cites specific reductions in the size of the frontal lobes (Nadel, 2003), an area of the brain associated with EF (Tau & Peterson, 2010). Yet, despite converging neuroanatomical and behavioral evidence, a paucity of comprehensive work has been conducted on the development of EF in young children with DS.

EF is a molar term referring to the interactive cognitive and emotional processes integral to adaptive, goal-directed actions including working memory, inhibition, emotional control, shifting, and planning (Carlson, 2005; Hughes 2011; Pennington & Ozonoff, 1996; Zelazo, Carter, Reznick, & Frye, 1997; Zelazo & Muller, 2011). Aspects of EF have been measured in infancy (Diamond, 1985; 1990) and continue to develop through childhood (e.g. Carlson, 2005). Greater understanding of EF has come from the concepts of hot and cool aspects of EF (Hongwanishkul, Happaney, Lee, & Zelazo, 2005). Hot EF incorporates affect and motivation and is associated with the ventromedial regions of the prefrontal cortex (e.g., gift delay; Hongwanishkul et al., 2005; Zelazo, Qu, & Muller, 2005). Cool EF elicits primarily cognitive demands and is associated with the dorsolateral prefrontal cortex (e.g., the Dimensional Change Card Sorting Task; Hongwanishkul et al., 2005).

There are several issues complicating the measurement and understanding of EF in early development. First, it is not fully known how task reliability and validity is affected when modifying EF tasks for developmental appropriateness (Garon, Bryson, & Smith, 2008; Hughes, 2011; Lehto, Juujarvi, Kooistra, & Pulkkinen, 2003). Currently there is no one universally accepted battery for assessing EF in early development. Second, the role of context in EF performance is complex. For example, Gioia and colleagues (2003) found only small to medium correlations between teachers' and parent's reports of EF observed at school and home in preschool-aged children (r 's ranging from .06–.28). This suggests that EF demands and performance may vary across school and home settings or perhaps be interpreted differently by the reporters. Additionally, with regard to the role of context, it is not fully known how lab-based performance tasks typically performed in a quiet area, one task at a time, administered by one examiner, compare to a person's performance of multifaceted EF demands in real life contexts (Bakar et al., 2011; Gioia, Isquith, Guy, & Kenworthy, 2000; Gioia, Kenworthy, & Isquith 2010).

As a final point, researchers examining EF in early development are still considering whether EF is a unitary construct or one made up of dissociable components. Existing evidence suggests that components of EF such as working memory and inhibition may have differing developmental trajectories (for a review see Garon et al., 2008; also see Diamond, 2001; Diamond, 2002; Miyake, Friedman, Emerson, Witzki, & Howerter, 2000; Murray & Kochanska, 2002). In examining EF in early development, Wiebe, Espy, and Charak (2008) found evidence for a unitary construct of EF in typically developing children 2 to 6 years of age. They found that models including multiple factors did not account for additional

variance beyond the unitary model. Additional insight comes from other researchers indicating that in typically developing children, as performance on EF tasks improves with age, intercorrelations among them decreases (see Tsujimoto, 2008, for a review).

Although questions regarding the dissociability of EF skills in typically developing children persist (Wiebe et al., 2008), the extant research on EF profiles in children with developmental disabilities supports a dissociable model. Researchers examining the validity of a parent-report measure of performance in everyday activities for school-aged children found evidence supporting this concept of a specific, multicomponent model of EF in a heterogeneous clinical sample including children with attention deficit hyperactivity disorder (ADHD), autism spectrum disorder (ASD), learning disabilities and other neurodevelopmental disorders (Gioia, Isquith, Retzlaff, & Espy, 2002). Furthermore, a body of research using laboratory tasks indicates that components of EF can be more impaired relative to overall mental age (MA) in specific neurodevelopmental disorders, whereas other areas are less impaired and MA appropriate. Ozonoff and Jensen (1999) hypothesized that when thoroughly assessed, different neurocognitive disorders may be associated with specific profiles of strengths and challenges in EF, an “executive fingerprint” (p. 175). Characterizing the EF profile in individuals with DS will be critical in that EF skills are associated with academic achievement (Blair & Razza, 2007, McClelland et al. 2007), social skills (Diamond, Barnett, Thomas, & Munro, 2007), and health outcomes (Riggs, Greenberg, & Rhoades, 2010) in typically developing children, as well as aspects of school participation and behavior related to academic achievement in children with developmental disabilities (DD; Zingerevich & LaVesser, 2009). Therefore, a better understanding of the EF profile in young children with DS will help establish an innovative framework for early intervention to improve developmental outcomes for this population.

At present, most existing studies examining EF in DS have focused on older children and adults using selected performance-based, laboratory tests in contrast to a comprehensive examination. Nonetheless, there is mounting evidence that individuals with DS demonstrate deficits in EF skills including deficits in working memory and perhaps planning, along with mixed evidence for deficits in both inhibition and shifting (see our broad review of the extant literature in Lee et al., 2011). Specifically, evidence suggests working memory impairments beyond those expected for overall MA (e.g., Baddeley & Jarrold, 2007), and mixed evidence exists regarding performance in other component areas of EF. Our team’s previously published findings highlight a potentially distinct EF profile in school-aged children with DS that includes significant levels of weaknesses beyond those anticipated for overall MA in the domains of both working memory and planning/organization as reported by parents (Lee et al., 2011). These findings suggest that even at a young MA (2–4 years), children with DS were reported to have a specific EF profile that included impairments in “cool” executive functions beyond those expected for their overall (delayed) developmental level. However, the generalizability of this work was limited in that previous work compared a DS group to published norms only. Therefore, a primary goal of this study was to replicate the findings of Lee et al. (2011) in a new sample of children with DS with two important additions: (1) inclusion of teacher-reported observations, and (2) inclusion of a contemporary sample of MA-matched TD children. This is a critical next step in

understanding the EF profile in children with DS. No children who participated in the Lee et al. (2011) study were included in the present study.

Therefore, for this new study we asked:

1. Do young children with DS present with significant levels of deficits in everyday EF compared to MA- matched comparison group of TD children as reported by (a) their teachers and (b) their parents?
2. Do teachers and parents concur in their reports of everyday EF function in children with DS?
3. What is the relative risk for school-age children with DS for presenting with executive dysfunction as reported by (a) their teachers and (b) their parents?

Method

Participants

Participants were 25 children with DS and 23 MA-matched children with TD, their teachers, and parents. Groups were equated for nonverbal MA using the Leiter-R Brief IQ subscales (Leiter-R; Roid & Miller, 1995; 1997, see Table 1 for participant characteristics).

Inclusion criteria for all participants included the following: (a) They had a complete Behavior Rating Index of Executive Function-Preschool version (BRIEF-P) questionnaire filled out by their teacher and/or primary caregiver; (b) they completed the Leiter-R Brief IQ to obtain an estimate of nonverbal MA; (c) their MA, as estimated by a direct developmental assessment (see the following section for details), fell within the normative sample age range of the BRIEF-P which is 2:0 to 5:11; (d) they had no history of traumatic events such as a head injury and no medical/genetic conditions beyond those associated with DS. Additional inclusion criteria for children with DS included (e) that children were reported to be free of an autism spectrum disorder clinical diagnosis as reported by their parents. One child with DS was unable to complete the entire Leiter-Brief IQ inclusion criteria and thus was not included in this report.

Parents who consented to invite their child's teacher into the study were asked to provide contact information for the teacher with whom the child spent the most time. Therefore, 100% of the TD comparison group's teachers were general education teachers; whereas 52.50% of the DS group's teachers were general education teachers and 47.50% of the teachers were special education teachers. It should be noted that one child in the DS study group had a report from both a general education and a special education teacher. For this child, the average of the teachers' ratings was used for this study.

Procedures

Written consent was obtained from the parents of child participants prior to completing any measures. Additionally, consent was obtained from the parents to contact their child's teacher and consent was also obtained from the teacher. Both teachers and parents completed one BRIEF-P questionnaire for each participant. Children were evaluated by

experienced researchers for nonverbal MA assessment. Assent was also obtained from the children to participate in the developmental measures. We obtained approval from the university institutional review board at Colorado State University.

Participants were recruited from the Rocky Mountain Down Syndrome Association, the Poudre School District in Fort Collins, CO, and JFK Partners, a University Center of Excellence in Developmental Disabilities at the University of Colorado School of Medicine, Anschutz Medical Campus, Aurora, CO.

Measures

Nonverbal MA—Children completed the subtests for the Leiter-R Brief IQ composite (Roid & Miller, 1997) to determine nonverbal MA. The Brief IQ Composite of the Leiter-R is derived from the following four visual reasoning subtests: figure ground, form completion, sequential order, and repeated patterns to measure nonverbal MA. This assessment has been standardized on a national sample of almost 2,000 individuals from 2.0 to 20.11 years old. The authors reported that Leiter-R Brief IQ Composite demonstrated adequate concurrent validity correlating with the WISC-III Full Scale and Performance IQs (.85) and high test-retest reliability (upper .80s–.90s). Mental age scores were obtained for each participant for the BRIEF-IQ composite in order to generate an estimate of overall MA. This MA score was used to determine if a participant would be included in the current study (i.e., if the MA fell in the equivalent 2:0 to 5:11 CA range used to standardize the BRIEF-P). These matching procedures were critical in that overall developmental level was already accounted for when conducting analyses to identify specific areas of impairment in the DS group. Using MA-appropriate norms and a comparison group of MA-matched TD children made it possible to identify EF skills that were impaired beyond expectation for a child's overall level of functioning. In other words, with this study, it was possible to ask whether an 8-year-old child with DS who had an MA of 4 years demonstrated clinical levels of impairments when compared to 4-year-old children (via the TD comparison group), not other 8-year-old children.

Everyday executive function skills assessment—We used the BRIEF-P (Gioia Espy, & Isquith, 2003) to examine EF skills for all participants. We used the BRIEF-P version exclusively in this study because the questionnaire was more developmentally appropriate for our participants than a version that matched their chronological age (see Lee et al. 2011 for a further discussion on this topic). The 63-item BRIEF-P obtains reports from teachers or caregivers to describe a student's or child's behavior using a 3-point Likert scale indicating how frequently a child engages in a given behavior (Never, Sometimes, Often). The BRIEF-P was normed on 460 children ages 2:0 to 5:11 representative of the U.S. population. It consists of five empirically-derived clinical scales, three Indexes, and a total score, called the Global Executive Composite (see Table 2 for a description of each domain). The BRIEF-P has adequate internal consistency (Cronbach's alpha from .80–.97 for the scales). Test-retest reliability correlations for the Indexes and Clinical scales ranged from .78 to .90 for parent reports and .65 to .94 for teacher reports (Gioia et al., 2003). Convergent and discriminant validity was established through an examination of

correlations between the BRIEF-P Indexes/Clinical Scales and other ratings scales thought to assess similar or dissimilar domains.

Finally, the BRIEF-P has been found to detect differences in typically developing children and two heterogeneous clinical samples that included children with ADHD, ASD, or a language disorder (Gioia et al., 2003). Furthermore, the BRIEF-P's validity data suggested that different clinical groups (e.g., an entire ASD group, ADHD group, and a language disorders group) presented with specific profiles. For example, the ASD group exhibited more problems with shifting than any other group.

Data Analyses

To obtain scores on the BRIEF-P, raw scores from each of the scales/indexes were converted to age- and gender-referenced normative T-scores. T-scores were used in all between-group analyses for this project. It is important to emphasize that, for the DS group, T-scores were calculated using the child's *nonverbal MA* (not chronological age), as measured by the Leiter-R Brief IQ subtests (as per Lee et al., 2011). Raw scores were converted into age equivalents using procedures outlined in the Leiter-R manual (Roid & Miller, 1997). This made it possible to produce T-scores that accounted for the child's overall level of developmental delay, and that facilitated the identification of impairments in EF beyond those that would be expected when compared to TD children at similar developmental levels. For example, the T-scores of an 8-year-old child with DS who had an MA of 4 years (as per the Leiter-R) was calculated using the BRIEF-P standardization norms for TD 4-year-old children. T-scores for the TD comparison group were calculated using the child's chronological age. Higher T-scores denote greater levels of EF impairments. On the BRIEF-P, T-scores at or above a 65 can be suggestive of clinical significance (Gioia et al., 2003). All variables were examined and determined to meet the approximate assumptions for parametric analyses.

Results

Comparison of EF in DS With an MA-Matched TD Group as Reported by Teachers and Parents

We conducted a mixed MANOVA to assess if there was a significant difference in everyday EF by group (DS or TD) and reporter (teacher or parent) on the BRIEF-P total (Global Executive Composite) and Index scores (Inhibitory Self Control, Flexibility, and Emergent Metacognition). See Table 3 for means and standard deviations of all teacher and parent BRIEF-P reports. Significant main effects were found for both group, $F(4, 31) = 8.86, p < .001, \eta^2 = .99$, and reporter (teacher vs. parent, $F(4, 31) = 4.04, p = .009, \eta^2 = .34$). There was no significant interaction effect for group x reporter, $F(4, 31) = 1.19, p = .33, \eta^2 = .13$. Figures 1 and 2 report the percentage of each group exhibiting T scores ≥ 65 , the level indicative of clinical levels of impairments as per the BRIEF-P manual (Gioia et al., 2003)

Teacher reports: Follow-up ANOVAs for significant main effect of group—

Analyses of the teacher-reported total and index scores with ANOVAs indicated that in comparison to the MA-matched TD group, students with DS were reported to have

significantly greater deficits in the Global Executive Composite and the Emergent Metacognition Index, $F(1, 34) = 11.78, p = .002, \eta^2 = .26$; and $F(1, 34) = 18.21, p = .001, \eta^2 = .35$ respectively, using a Bonferroni adjustment ($.05/4 = .0125$). No significant between-group differences were observed for the Flexibility and the Inhibitory Self-Control Indices, $F(1, 34) = 5.05, p = .03, \eta^2 = .13$ and $F(1, 34) = 3.50, p = .07, \eta^2 = .09$ respectively. Further examination of the clinical scales comprising the Emergent Metacognition Index indicated that students with DS were reported to have significantly more problems compared to their TD peers at equivalent MA levels with both Working Memory and Plan/Organize, $F(1, 34) = 25.90, p = .001, \eta^2 = .43$; and $F(1, 34) = 7.50, p = .01, \eta^2 = .18$ respectively, using a Bonferroni adjustment ($.05/2 = .025$). In summary, teachers reported that their students with DS demonstrated significant challenges in both Working Memory and Plan/Organize.

Parent reports: Follow-up ANOVAs for significant main effect of group—

Follow-up ANOVAs with the parent-reported total and index scores indicated that in comparison to the MA-matched TD group, the DS group was reported to have significantly more problems on the total Global Executive Composite and index scores of Emergent Metacognition and Inhibitory Self Control, $F(1, 46) = 19.36, p = .001, \eta^2 = .30$; $F(1, 46) = 35.60, p = .001, \eta^2 = .44$; and $F(1, 46) = 9.22, p = .004, \eta^2 = .17$ respectively, using a Bonferroni adjustment ($.05/4 = .0125$). Mean differences for the Flexibility index were not statistically significant with this Bonferroni adjustment, $F(1, 46) = 5.81, p = .02, \eta^2 = .11$. Given that both Emergent Metacognition and Inhibitory Self Control scores were significantly poorer in the DS group, we examined group differences in their respective clinical scales. The DS group had more pronounced deficits in Working Memory, Plan/Organize, and Inhibit than their MA-matched, TD counterparts, $F(1, 46) = 22.65, p = .001, \eta^2 = .33$; $F(1, 46) = 17.27, p = .001, \eta^2 = .27$; and $F(1, 46) = 8.06, p = .001, \eta^2 = .15$ respectively, using a Bonferroni adjustment ($.05/4 = .0125$). Neither the clinical scale Shift, nor Emotional Control were significantly lower in the DS group, $F(1, 46) = 4.35, p = .042, \eta^2 = .09$ and $F(1, 46) = 2.555, p = .117, \eta^2 = .05$ respectively. In summary, parents reported that their children with DS demonstrated significant challenges in Working Memory, Plan/Organize, and Inhibit in comparison to parent reports of typically developing children.

Congruence of teacher and parent reports by group: Follow-up for significant main effect for reporter—As described previously, a main effect for reporter (teacher or parent, $F(4, 31) = 4.04, p = .009, \eta^2 = .34$) was observed. Therefore, we examined the relationship of teacher to parent reports of EF by domain and group. Overall, the magnitude of the relationships between ratings from both teacher and parent report data in the DS group were large (all but one $r = .70$ s). The magnitude of the relationship between teacher and parent reports for in the TD group ranged from $r = .10$ s-. $.40$ s (see Table 4 for all coefficients). Finally, post-hoc Spearman correlations were conducted to examine the association between MA and the Global Executive Composites for both the teacher and parent reports. Findings indicate that the magnitude of the relationship between MA and EF is small to medium for both groups (see Table 5).

The Risk of Clinical-Level EF Problems in Children With Down Syndrome

Relative risks coefficients were calculated for all BRIEF-P clinical scales in which the DS group demonstrated significant impairments in comparison to the expected rate of such elevations based on the published norms (~7%; Gioia et al., 2003). The relative risk calculations were completed using the frequencies derived from the number of children having a T-score of 65 or greater on each clinical scale, denoting clinical-level impairment for that MA in both the DS group and the standardization population (Gioia et al., 2003). According to the teachers' reports, the relative risk of students with DS demonstrating clinical-levels of impairments was 9.77 times higher for Working Memory (95% CI [4.49, 21.26]) and 6.77 times higher for Plan/Organize (95% CI [2.87, 15.9]) than the standardization population. Based on parent-reported data, the risk of children demonstrating clinical-levels of impairments was 9.14 times in Working Memory (95% CI [4.22, 19.80]), 5.71 times in Plan/Organize (95% CI [2.42, 13.51]), and 4.57 times in Inhibit (95% CI [1.83, 11.41]) compared to the standardization population. The confidence intervals for these relative risk analyses do not include 1 in the interval which indicates that they are significant (Portney & Watkins, 2009).

Discussion

This study characterized the EF profiles of a group of children with DS (without co-occurring ASD) and a MA-matched, typically developing comparison group. The BRIEF-P, an assessment of everyday executive function, was used to examine reports by teachers and parents. Consistent with parent-reported data from Lee et al. (2011), significant deficits were observed in the DS group, as reported by both teachers and parents, in the areas of working memory and planning/organizing when compared to the MA-matched TD comparison group. Additionally, parent-reported data also indicated that the DS group had significantly greater impairments on the clinical scale Inhibit (inhibitory control) than the comparison TD group. However, in the teacher-reported data, inhibitory control was not reported to significantly differ between groups.

Characterizing EF Deficits in DS

In contrast to the MA-matched TD group, the DS group's profile included clinical-levels of impairments in the areas of working memory (68.4% of participants) and planning (47.4%) as reported by teachers. As reported by parents, significantly more children in the DS group also demonstrated clinical levels of impairments in working memory (64.0% of participants) and planning (40.0%), as well as inhibitory control (32.0%) when compared to the TD comparison group. However, the DS group was not found to demonstrate significantly greater difficulties with inhibitory control than the TD comparison group based on teacher report. The EF profile observed in the present DS study group maps roughly onto the Hongwanishkul et al.'s (2005) concept of cool EF functions with a possible subset of children with DS also experiencing deficits in inhibitory control. This profile of EF deficits in DS lends support to Ozonoff and Jensen's (1999) hypothesis for specific EF profiles in individuals with neurodevelopmental disorders.

Working memory deficits—Deficits in the working memory component of this profile have been widely reported in the literature in school-aged and older individuals with DS using laboratory-based performance measures (for a review see Baddeley & Jarrold, 2007; see also Conners, 2003; Edgin, Pennington, & Mervis, 2010; Pennington et al., 2003). Furthermore, the extant literature suggests that individuals with DS perform more poorly on working memory tasks with an auditory component and that this impairment is related to language processing (see Conners 2003 for a review), particularly in the phonological domain (Lee, Pennington, & Keenan, 2010). Over 60% of the DS study group showed clinical levels of impairment in this area by both teachers and parents, when compared to a MA- matched comparison group. Children with DS were nine times more likely to experience clinical levels of problems in working memory at either school or home compared to developmentally equivalent TD children. Furthermore, when examining the magnitude of this working memory deficit utilizing Cohen's *d* (Cohen, 1988), a measure of effect size, we find Cohen's *d* to be 1.8 for teachers and 1.4 for parents. As a direct comparison to the lab-based working/short-term memory literature focused on the verbal domain, we refer to Lee et al. (2010) who calculated the mean effect size (and 95% CI) of this impairment for eight published studies that compared DS performance to that of mental-aged matched TD controls. They reported a mean effect size of 1.97 with a 95% confidence interval of 1.18 to 2.75. Both teacher and parent effect sizes reported in the current study fall within that 95% CI, providing support for the convergence of questionnaire and lab-based measures of working memory and the importance of this deficit to the DS neurocognitive phenotype. It is possible that teachers and parents of children with DS may be familiar with literature describing working memory deficits in this population and thus rated the children accordingly. However, the BRIEF-P does not label domains and intermixes rating items for each domain, therefore, minimizing report bias.

The long-term implications of these working memory deficits are poorly understood. Researchers have found that typically developing kinder-gartners' cool EF tasks performance (e.g., working memory) at the beginning of the school year differentially predicted both math achievement and learning-related behavior in the following spring (Brock, Rimm-Kaufman, Nathanson, & Grimm, 2009). However, no evidence to date examines how early deficits in working memory moderate outcomes for individuals with DS. Promising evidence exists for working memory training in children with DS (Conners, Rosenquist, Arnett, Moore, & Hume, 2008), but see also Hulme & Mackenzie, (1992) who failed to find support for training, as well as other groups (Morrison & Chein, 2011). There is much to be learned regarding working memory and children with DS, including the most effective mechanism of change to improve working memory deficits and understanding functional outcomes that can effectively be improved by intervention.

Planning deficits—Less evidence is available on planning and problem-solving skills in individuals with DS. Extant research examining specific planning and problem-solving performance tasks indicates deficits in this area as well (e.g., Fidler, Hepburn, Mankin, & Rogers, 2005; Kasari & Freeman, 2001; Rowe et al., 2006). Based on the parent report, about 37% of the children with DS in the present study were reported to have clinical levels of impairment in planning skills beyond those expected for their overall level of

development. This level of impairment is in line with the findings by Lee et al. (2011). In the present sample, students with DS were almost seven times more likely than their MA-matched peers to present with planning impairments as reported by their teachers. Based on parent report, children are almost six times more likely to have planning deficits. It is possible that school tasks demand more planning challenges (e.g., sequencing steps to complete a learning activity) in perhaps a more dynamic and demanding environment than the family/home context reported by the parents. More data are needed to understand how deficits in planning abilities affect participation and performance in the educational context.

Inhibitory control deficits—In this study, we found significant group differences in parent-reported inhibitory control. The existing literature on inhibition in individuals with DS provides mixed evidence (e.g., Kopp, Krakow, & Johnson, 1983, reported evidence for a deficit and Pennington et al., 2003, reported no evidence for a deficit). Researchers have acknowledged that the concept of inhibition in EF is complex because inhibition is operationalized to include several types of functions such as behavioral inhibition and disregarding interference. Additionally, inhibition is often integrated in many other tasks involving executive cognitive functions (for a brief review see Zelazo & Müller, 2011). This issue regarding the operationalization of inhibitory control may contribute to the mixed evidence in this population. On the BRIEF-P, items measuring inhibitory control focus on behavioral inhibition, or the ability of children to have control over their behavior and stop a behavior as appropriate (Gioia et al., 2003; see also Table 2). Lee et al.'s (2011) findings indicated that parents reported clinical levels of impairment on the inhibit scale for their children with DS (31% of the sample). This is almost identical to the parent-reported data for the DS group from this study (32%).

Emotional control and shifting—Neither emotional control nor shifting were significant areas of impairment for the DS study group in contrast to the MA-matched comparison group as reported by both teachers and parents. This result parallels the findings of no significant impairments in these domains by Lee et al. (2011). It is important to remember that the present findings do not indicate that emotional control or shifting skills are necessarily at chronological age expectations for the DS group, but suggest that they are not excessively delayed in regards to the group's cognitive abilities. The results in regard to emotional control parallel other research. In the literature, social cognition issues with emotional recognition and theory of the mind have been established in individuals with DS (see Cebula, Moore, & Wishart, 2010), but fewer mood disorders and psychiatric comorbidities have been found in individuals with DS compared to MA-matched individuals with other ID (Dykens, 2007). The findings regarding shifting are an important addition to the body of inquiry on EF skills in individuals with DS. Currently there is mixed evidence in the literature for problems in shifting in adolescents with DS). One study reported that 5-year-olds with DS were able to perform a delayed non-matching to object task significantly better than their developmentally-matched peers with autism (Dawson, Meltzoff, Osterling, & Rinaldi, 1998). Conversely, in another study, almost 41% of a group of school-aged children with DS could not shift on the dimensional card sorting task (Edgin, 2003). On the BRIEF-P, items measuring shifting focus on children's ability to flexibly solve problems or switch attention (see Table 2 for examples). It is possible that these items assessing this skill

in everyday contexts could diverge from laboratory tasks of shifting. We discuss further issues regarding performance on the BRIEF-P and performance on laboratory tasks of EF in the following sections.

Further interpretation of EF profile in DS—It should be noted that the BRIEF-P was not specifically constructed to assess hot and cool EF; however, the empirically derived Indexes reflect these conceptual distinctions. As such the BRIEF-P can be used to cautiously contribute to the academic discussion of the DS profile when acknowledging the need for conservatism as items have not been tested for consistency of “coolness” or “hotness” within each domain. With this in mind, the Index Emergent Metacognition, which contains the scales Working Memory and Plan/ Organize, corresponds to what is often described in the literature as cool EF functions. The Indexes Flexibility (made up of the Inhibition and Shifting scales) and Inhibitory Self-Control (made up of Inhibition and Emotional Control) are in many ways congruent with hot EF domains. As emphasized by Riggs, Greenberg, and Rhoades (2011), no matter which EF framework one chooses to adopt (e.g., multidimensional), inhibitory control functions as both impulse control *and* as a system that allows a child to inhibit extraneous or competing stimuli. Therefore, typically cool tasks such as monitoring one’s progress during an activity also requires inhibitory control.

If the DS cognitive profile features a profile of primary deficits in working memory and planning, with a smaller number of children also demonstrating impairments in inhibitory control, what could be the neurobiological basis of these impairments? Prior research identifies the dorsolateral prefrontal cortex as traditionally being associated with cool EF functions, such as working memory and planning deficits, whereas the ventromedial prefrontal cortex has been associated with hot EF performance, such as inhibitory control (Zelazo & Muller, 2011). Thus, these should certainly be regions of future investigation for neuroimaging studies of DS. Existing studies of the DS neuroanatomical phenotype utilizing magnetic resonance imaging (MRI) report volumetric reductions in the frontal lobes (e.g., Jernigan & Bellugi, 1990; White Alkire, & Haier, 2003) but they traditionally have not focused on these frontal subregions.

Given that neuroanatomical and cellular dysfunction is widespread in the DS brain (e.g., in synapses and dendritic spines, Haydar & Reeves, 2011), the mechanisms underlying cognitive executive dysfunction in this population are not fully understood. Moreover, the developing brain in neurocognitive disorders such as DS is likely to be quite different from adult models of brain injuries that strongly inform our understanding of executive dysfunction (Thomas & Karmiloff-Smith, 2002). Thus, these models are helpful starting points but are likely to have limited utility as a means to fully describe the neuroanatomical underpinnings of the executive deficits in DS. Finally, as highlighted by Zelazo and Müller (2011) and Diamond (2000), various neuroanatomical components work together with other neural structures as a larger system to adaptively obtain goals. Thus, the nature of the neural underpinnings of executive dysfunction is likely complex. Consequently, it will be critical to characterize the EF performance, interrelatedness of domains of EF in the DS profile along with a better understanding of the potentially complex neurological foundations from early development in order to develop effective interventions. Taken as a whole, our study findings and cited arguments regarding the potential contributions of inhibitory control’s

role in successful completion of “cool” EF tasks highlight the need for a deeper understanding of the conceptualization of EF in DS and its neurobiological foundations.

Agreement Between Teacher and Parent BRIEF-P Reports

This study is among the first to use multiple informants to examine every day EF in DS in both the school and home context. Interestingly, overall the teacher and parent reports of the DS group (all but one in the $r = .70$ s) are highly correlated with each other. The group patterns of these correlations are on par with the BRIEF-P psychometric data describing congruence of parent and teacher reports for clinical and typically developing samples (Gioia et al., 2003). This may suggest that the demands across context and/or performance across context may vary. Although the BRIEF-P examines behaviors across contexts, it provides little information regarding functional performance. For example, the BRIEF-P is unable to highlight when a student has few limitations in some classroom activities and more in others. A deeper understanding of the EF demands at school and home will be critical in developing interventions that target relevant outcome variables in these contexts.

Limitations

The teacher and parent report methodology was both a strength and a limitation of this study. Teacher and parent reports may be useful during early development when it may be challenging to assess EF performance. However, one must conservatively interpret the current study's BRIEF-P findings in relationship to EF domains. Although existing studies comparing the school-aged BRIEF to laboratory tasks and assessments have found various amounts of convergence and divergence across studies (McAuley, Chen, Goos, Schachar, & Crosbie, 2010), comparisons between the BRIEF-P and laboratory-based performance tasks are scant. Mahone and Hoffman (2007) found only a small correlation between performance on the BRIEF-P and performance-based EF tasks in a sample of preschoolers with ADHD. However, the BRIEF-P scores for the ADHD group differed significantly from the comparison group suggesting the instrument may be sensitive to symptoms.

As highlighted by Mahone and Hoffman (2007), the BRIEF-P was not highly correlated with IQ in their study, indicating that perhaps this is an important instrument in understanding the behaviors of children with IQ scores on both the low and high the ends of the spectrum. Correlational analyses from the current study support this observation between the Leiter Brief IQ and the BRIEF-P Global Executive Composite; however, due to the small group sizes more information is needed. If, with more data, it becomes clear that the relation between everyday EF behavior and IQ in children with DS is not strong, this may have tremendous implications for educational, functional, and perhaps even pharmaceutical intervention.

Although the reports from teachers and parents may be important in determining performance in school and home life, it is possible that differences reported by teachers and parents may vary due to factors other than context. That is, teachers and parents may consciously or unconsciously use different comparison children when considering a target child's performance. For example a teacher may compare a child to other peers with disabilities whereas a parent may lack a point of comparison or use a sibling. Further

information is needed regarding EF performance across reporters and contexts to understand this issue. Additionally, the current study compared the DS group to a MA-matched comparison group. Therefore, the DS group was MA-appropriate for the BRIEF-P, but chronologically older than children used in the BRIEF-P normative sample. Consequently, life experience may also confound these findings. Overall, more data are needed to better understand the relationship of BRIEF-P ratings, EF laboratory task performance, and IQ to better inform intervention.

Another limitation of this study is the modest sample size and the use of an MA- matched, TD comparison group as opposed to a group of children with neurodevelopmental disabilities matched for both MA and chronological age (CA). As such, this study is unable to address any questions regarding the specificity of the EF profile observed in DS and whether this pattern is somewhat unique to this population, or whether it is a pattern observed more generally in children with other neurogenetic disorders or DDs in general. We argue that having a concurrent MA-matched comparison group is critical for understanding how EF skills differ from the normative expectations when accounting for overall developmental status. However, in the future, a group of MA- and CA- matched children with other DDs will be essential to control for confounds. Because the DS group was chronologically older than the MA-matched comparison group in this study; we used the BRIEF-P in a way that perhaps it was not intended. The BRIEF-P manual did report findings on clinical populations; however, the participants' MA was not reported (Gioia et al., 2003). Therefore, the greater amount of life experience in the DS group may be a confound of this study. In summary, a comparison group with developmental disabilities matched for both MA and CA would help better address questions regarding the specificity of the EF profile to DS (Seltzer et al., 2004).

Future Directions

Researchers studying typically developing children have found that EF may be a unitary construct in early development for typically developing children (Wiebe et al., 2008). However, this study further characterizes the everyday EF profile in DS that is observable in school-aged children. It will be important to understand how this profile may predict or moderate trajectories of adaptive behavior and academic achievement. Additionally, examining how children with DS perform on laboratory tasks of EF and how this performance corresponds to achievement in both academic and life skills will be a critical next step in characterizing the cognitive phenotypic profile in DS and identifying important targets for intervention. In the absence of a better understanding of EF in young children with DS, the development of effective early interventions, including pharmaceutical interventions targeting cognitive function in individuals with DS, will continue to remain challenging.

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Figure 1.

Teacher reports: Percentage of clinically elevated scores on BRIEF-P ($T \geq 65$) by group.

Note: I = Inhibit; EC = Emotional Control; S = Shift; WM = Working Memory; P/O = Plan/Organize.

Figure 2.

Parent reports: Percentage of clinically elevated scores on BRIEF-P ($T \geq 65$) by group.

Note: I = Inhibit; EC = Emotional Control; S = Shift; WM = Working Memory; P/O = Plan/Organize.

Table 1

Participant Characteristics

Characteristic	Group						<i>t</i> / <i>X</i> ²	<i>p</i>
	Down syndrome <i>n</i> = 25			Typically developing <i>n</i> = 23				
	<i>M</i>	<i>SD</i>	<i>Range</i>	<i>M</i>	<i>SD</i>	<i>Range</i>		
Chronological age (months)	96.56	17.31	61–133	39.78	5.00	30–46	15.71	.001
Mental age (months) ^a	50.12	8.32	40–67	49.96	5.32	35–57	.22	<i>p</i> = .83
	<i>f</i>	<i>n</i>	%	<i>f</i>	<i>n</i>	%		
Child's gender female	7	25	28.00	9	23	39.10	0.19	<i>p</i> = .89
Child White, Non-Hispanic	22	24	91.70	19	23	82.60		
Mother ed, college ^b	14	25	68.00	21	23	91.30		
Mother White, Non-Hispanic	23	24	96.00	19	23	82.6		
Father ed, college ^b	17	24	71.00	21	23	91.30		
Father White, Non-Hispanic	23	23	100	21	23	91.30		

^aLetter-R Brief IQ raw score used for statistical comparison.^bMother and Father's Education - Number/Percent who completed college.

Table 2

Description of BRIEF-P Clinical Scales and Indexes

Domain	Description, examples
Total Score	
Global Executive Composite (GEC)	Total score comprised of the five clinical scales Inhibit + Shift + Emotional Control + Working Memory + Plan/Organize
Indexes	
Inhibitory Self-Control Index (ISCI)	Comprised of the clinical scales Inhibit + Emotional Control
Flexibility Index (FI)	Comprised of the clinical scales Shift + Emotional Control
Emergent Metacognition Index (EMI)	Comprised of the clinical scales Working Memory + Plan/Organize
Clinical Scales	
Inhibit (I)	Behavioral regulation or inhibiting responses to avoid engaging in impulsive or inappropriate behavior <i>Examples</i> “Acts wilder or sillier than others in groups...” “Is impulsive”
Emotional Control (EC)	Emotion modulation and corresponding behavioral responses <i>Examples</i> “Has explosive, angry outbursts” “Becomes upset too easily”
Shift (S)	Flexibly moving between tasks or situations and alternating attentional focus when completing tasks <i>Examples</i> “Becomes upset with new situations” “Is upset by a change in plans or routine”
Working Memory (WM)	Maintaining information in the focus of one’s attention in order to complete a task or provide the appropriate response <i>Examples</i> “When given two things to do, remembers only the first or the last” “Has trouble finishing tasks”
Plan/Organize (PO)	Anticipating and preparing for future activities or tasks, handle current demands, and to impose order on information to accomplish a goal <i>Examples</i> “When instructed to clean up, puts things away in a disorganized, random way.” “Needs to be told to begin a task even when willing to do it”

Table 3

T-Scores on BRIEF-P by Group and Reporter

BRIEF-P Domains	Down syndrome group (<i>M, SD</i>)	Typically developing Group (<i>M, SD</i>)	Cohen's <i>d</i>	
<i>Teacher Reported^a</i>				
Global Executive Composite	66.21	54.29	6.00	1.18
Indexes:				
Inhibitory Self-Control	61.31	54.29	7.43	.64
Flexibility	55.89	49.24	4.60	.77
Emergent Metacognition	66.79	55.35	6.69	1.16
Clinical Scales:				
Inhibit	63.89	56.82	9.84	.57
Emotional Control	55.16	50.00	7.06	.51
Shift	55.47	48.76	6.72	.80
Working Memory	71.11	54.29	6.62	1.75
Plan/Organize	65.84	56.00	8.38	.90
<i>Parent Reported^b</i>				
Global Executive Composite	62.32	48.13	10.12	1.30
Indexes:				
Inhibitory Self-Control	56.92	47.35	9.30	.90
Flexibility	53.28	46.43	7.79	.71
Emergent Metacognition	67.08	49.43	10.38	1.77
Clinical Scales:				
Inhibit	59.76	49.65	10.28	.84
Emotional Control	49.88	45.26	7.26	.47
Shift	54.72	48.39	7.60	.62
Working Memory	67.08	51.21	10.07	1.41
Plan/Organize	60.76	46.74	10.22	1.23

^a DS *n* = 19 TD *n* = 17.^b DS *n* = 25, TD *n* = 23.

Table 4

Agreement of Parent and Teacher Reports, on the BRIEF-P by Group (Pearson r)

BRIEF-P Domain (T-scores)	Down syndrome	Typically developing
Global Executive Composite	.76**	.25
Emergent Metacognition	.56*	.33
Flexibility	.72**	.14
Inhibitory Self-Control	.72**	.48

*
 $p = .01$.**
 $p = .001$.

Table 5Correlation (Spearman's rho) Between Nonverbal MA^a and BRIEF-P

BRIEF-P Global executive composite (T- score)	Down syndrome	Typically developing
Parent Report Teacher	.37	.03
Report	.23	.42

^a Nonverbal MA = Brief IQ composite from the Leiter-R.