Cognitive Flexibility Among Individuals With Down Syndrome: Assessing the Influence of Verbal and Nonverbal Abilities

Colin Campbell, Oriane Landry, Natalie Russo, Heidi Flores, Sophie Jacques, and Jacob A. Burack

Abstract
The influences of verbal mental age (VMA) and performance mental age (PMA) on cognitive flexibility were examined among a group of participants with Down syndrome (DS), in order to disentangle the relative contributions of each. The impaired cognitive flexibility typically observed among individuals with DS in combination with uneven VMA and PMA development suggests an opportunity to further understand the developmental relationship between VMA, PMA, and cognitive flexibility. We examined the performance of 22 participants with DS on the Flexible Item Selection Task (FIST), used for measuring cognitive flexibility among preschoolers. Partial correlations revealed that only VMA was related to the FIST after controlling for PMA, highlighting the role of verbal abilities in the development of cognitive flexibility.

Key Words: Down syndrome; cognitive flexibility; executive function
Card Sorting Test (WCST; Berg, 1948), in which participants are required to match items based on changing dimensional criteria. On matching item tasks designed for children, such as the Dimensional Change Card Sort (DCCS; e.g., Frye et al., 1995; Zelazo et al., 1996) and the Flexible Item Selection Task (FIST; Jacques & Zelazo, 2001, 2005) tests, essential advances in cognitive flexibility are observed between the ages of 3–5 years. Although possible links to nonverbal visual-spatial skills have been reported (Blair & Razza, 2007; Kharitonova & Munakata, 2011), the developmental changes within cognitive flexibility are primarily associated with concurrent development in language abilities (Frye et al., 1995; Hongwanishkul et al., 2005; Zelazo et al., 1996), as tasks that are used to assess cognitive flexibility such as the DCCS and the Self-Ordered Pointing task (SOP) (Petrides & Milner, 1982) have been linked to language ability (Kirkham et al., 2003; Hongwanishkul et al., 2005; Jacques & Zelazo 2005). As the deficits in language are over and above the cognitive delay associated with DS (Chapman, 1998; Chapman & Kay-Raining Bird, 2012), they may also limit success on cognitive flexibility tasks which are dependent on strong language abilities (Jacques & Zelazo, 2005). This notion is supported by Landry et al.’s (2012) findings that performance on the DCCS and SOP was uniquely correlated with verbal development and not to nonverbal intelligence.

As in the study of all aspects of typical cognitive functioning, the attempt to delineate the factors that contribute to the development of cognitive flexibility is limited by the inability to tease apart patterns of developmental relations among factors that are inherently linked and those that co-occur by happenstance. Thus, the developmental pattern of persons with DS, which is characterized by a relative weakness in verbal as compared to nonverbal visual-spatial abilities (Chapman, 2006; Rowe et al., 2006; Thase et al., 1984; Wang, 1996), provides an opportunity to examine the development of cognitive flexibility when development across domains is not “lock-in-step” as it is among typically developing children.

The potential utility of this study is further enhanced by the initial evidence that persons with DS are less cognitively flexible than would be expected based on developmental level as indicated by mental age. In a study in which the participants with DS were less able to switch between two sets of sorting rules of the DCCS as compared to typically developing preschool participants. The participants with DS, who were adults, tended to use a single set of rules throughout the task regardless of any change in instruction, although there were some participants who were able to switch flexibly. Thus, in order to extend this preliminary evidence of impairment in cognitive flexibility among persons with DS to another paradigm and to examine the factors that contribute to the development of this area of functioning, we assessed the relative influences of the developmental factors of chronological age (CA), performance mental age (PMA), and verbal mental age (VMA) on performance on the FIST (Jacques & Zelazo, 2001) among children and adolescents with DS. The FIST, which is an item-matching task, was selected as the measure of cognitive flexibility for this study because it was originally designed for use with preschoolers between the ages of 3–5 years, which makes the FIST useful for populations with delayed cognitive development. The combination of the discrepancy between CA and mental age that is inherent to all groups with intellectual disability and the typical disparity that exists between abilities in visual-spatial processing, a relative strength among individuals with DS, and verbal abilities, an area of relative weakness, allowed us a unique opportunity to assess specific contributions of primary developmental factors to performance on cognitive flexibility.

Method

Participants
Twenty-six English-speaking children, adolescents, and young adults with DS were originally recruited from the Down Syndrome Research Foundation in Vancouver, British Columbia, Canada; Summit School in Montreal, Quebec, Canada; and the Down Syndrome Research Institute in London, Ontario, Canada. None of the participants had any other comorbid neurodevelopmental disorders, including autism spectrum disorder. Four of the recruited children were excluded from the study, three for not being able to understand the task instruction and one for having a VMA less than 34 months. Thus, 22 children, adolescents and young adults (21 female) with DS, ranging in CA from 9 years,
4 months to 23 years, 0 months with a mean age of 15 years, 3 months (SD = 51.8 months), participated in the study. Performance mental age was assessed with the Leiter International Performance Scale, Revised (Leiter-R) and ranged from 48 to 84 months (M = 62.66 months; SD = 9.33 months), while VMA was assessed with the Peabody Picture Vocabulary Test-Third Edition (PPVT-III) and ranged from 38–98 months (M = 64.05 months, SD = 20.26 months). The CA, VMA, and PMA of each of the participants are provided in Table 1. The difference in mean scores between the VMA and PMA was not significant, whereas the correlation between them was significant, r (20) = .77, p < .001. VMA was significantly correlated with CA, r (20) = .44, p < .05, while the correlation between PMA and CA approached significance, r (20) = .33, p = .07.

Table 1
Chronological Age (CA) in Months, and Verbal Mental Age (VMA) and Performance Mental Age (PMA) Estimates in Months by Participant

<table>
<thead>
<tr>
<th>Participant</th>
<th>Gender</th>
<th>CA</th>
<th>VMA</th>
<th>PMA</th>
</tr>
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<td>48</td>
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<td>Female</td>
<td>231</td>
<td>38</td>
<td>48</td>
</tr>
<tr>
<td>5</td>
<td>Female</td>
<td>157</td>
<td>59</td>
<td>55</td>
</tr>
<tr>
<td>6</td>
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<td>67</td>
</tr>
<tr>
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<td>Male</td>
<td>233</td>
<td>86</td>
<td>65</td>
</tr>
<tr>
<td>8</td>
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<td>212</td>
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<td>71</td>
</tr>
<tr>
<td>9</td>
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<td>173</td>
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<td>55</td>
</tr>
<tr>
<td>10</td>
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<td>214</td>
<td>87</td>
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</tr>
<tr>
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<td>Female</td>
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<td>98</td>
<td>84</td>
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<td>55</td>
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<td>58</td>
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<tr>
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</tr>
<tr>
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<td>66</td>
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<td>61</td>
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<td>276</td>
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<tr>
<td>22</td>
<td>Female</td>
<td>122</td>
<td>64</td>
<td>64</td>
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</table>

Mean: 183.95, SD: 51.76

Institutional review board approval was obtained from the institution of the senior author (McGill University, Canada). Prior to the administration, consent was obtained from a parent or caregiver. At the beginning of the administration, the participants were told the instructions of the task and verbally asked whether they would like to participate. The participants were told that they could ask for help if they did not understand something and that they could stop at any time. They were also told that their performance in the study would not be reported to anyone, including their parents or teachers.

Measures

Flexible Item Selection Task. The FIST is a task designed to assess cognitive flexibility by having children match items on two different dimensions and is appropriate for use with preschoolers (Jacques & Zelazo, 2001). On each of 18 trials, the participants were shown three items that varied on shape, color, and size (e.g., a small blue boat, a small blue teapot, and a small yellow boat) and asked to select two items that matched on a particular dimension as shown in Figure 1. The trials each consisted of sets of three items that were identical on one dimension (e.g., size) but differed on the other two relevant dimensions (e.g., shape and color). The participants were asked by the examiner to “Show me two pictures that go together in one way.” After completing the first match (e.g., the two boats), the participants were asked to “Show me two pictures that go together but in another way.” Once they selected a second pair (e.g., the two blue items), they moved on to a different trial. At the beginning of the task, one demonstration trial was completed by the examiner in order to demonstrate the purpose of the task. In order to determine whether the participants understood the basic task instructions, they each completed two practice trials in which they obtained feedback. All of the participants completed the same 15 test trials presented in a counterbalanced order.

Scoring. On each trial, the participants who correctly identified a match on their first attempt received a score of one for the first match. They also received a score of one on the second match if they correctly identified the other possible pair of items, but only if their first match had been correct. This scoring system was adopted because
an incorrect match on the first attempt meant that the other two remaining matches were inevitably correct. Scoring a second match as correct while the first match was incorrect would have inflated the scores on second matches. The raw scores from the first and second matches were analyzed separately, with scores from the first matches assessing abstraction abilities, and scores from the second matches representing cognitive flexibility.

Peabody Picture Vocabulary Test–Third Ed. (PPVT-III; Dunn & Dunn, 1997). The PPVT-III was used to assess the receptive vocabulary of individuals, and for the current study provided an estimate of VMA. In order to complete the task, the participants had to select the correct image out of four that matched the word expressed by the examiner until they reached a performance ceiling.

Leiter International Performance Scale–Revised (LEITER-R). Leiter-R Brief IQ Screener was used to assess PMA. The Brief IQ index is based on four subtests that assess visualization (figure ground and form completion) and fluid reasoning (sequential order and repeated patterns). The Leiter-R is administered with minimal verbal instruction by the examiner so as to minimize the influence of verbal abilities (Roid & Miller, 1997). For the current study, the Brief IQ composite score was obtained in order to determine the PMA in months.

Procedure

The measures for the study were administered individually in one session at either of the two participating centers by examiners who were blind to the study’s hypotheses. In addition to the FIST, PPVT-III and Leiter-R, other tasks not related to the current study were also completed by the participants.

Results

The mean number of correct responses on the FIST was 13.14 out of 15 for their first match (SD = 1.75) and 8.55 out of 15 for their second match (SD = 3.00). In order to assess the relation between task performance on each match and CA, VMA and PMA (all scored in months), Pearson correlations were calculated. The difference in mean scores between the VMA and PMA was not significant. However, as shown in Table 2, the correlation between them was significant, r (20) = .77, p < .001, using a Bonferroni.1 VMA was not significant correlated with CA, r (20) = .44, p < .05 and neither was the correlation between PMA and CA, r (20) = .33, p = .07. As also shown in Table 2, both the PMAs and the VMAs were positively correlated with FIST first match (PMA, r [20] = .63, p < .002; VMA, r [20] = .58, p < .005) and second match (PMA, r [20] = .61, p < .003; VMA, r [20] = .70, p < .001). Chronological age was not significantly correlated with performance on the first, r (20) = .41, p = .058, or second match, r (20) = .20, p = .367. The correlations between second match accuracy on the FIST and PMA are presented in Figure 2, and the correlations between second match accuracy and VMA are presented in Figure 3.

To disentangle the unique contribution of PMA and VMA to performance on both the first and second matches on the FIST, partial correlations were used as follow up analyses.2 When controlling for PMA no significant association remained between correct first pair matches on the FIST and VMA, r (20) = .12, p = .42. Similar results were found when controlling for VMA with PMA and first pair matches on the FIST, r

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1 Because of the number of simple correlations calculated (n = 10), a Bonferroni correction was used to protect against inflated experiment-wise errors. Thus, for 10 correlations, we used $p = .005$ for experiment-wise Type I error rate of $p = .05$.

2 Note that Bonferroni corrections were not applied to these followup analyses. Unlike the overall simple correlations, the purpose of the followup analyses was to try to identify patterns of unique correlations between specific measures of mental age and performance on different aspects of the FIST once other significant measures had been controlled. Given the small number of participants and the more conservative nature of partial correlations in relation to simple correlations (by controlling for the contribution of other variables), we felt that applying a correction in this context would lead to an unacceptably high Type II error rate.
(20) = .35, \( p = .12 \). In contrast, when controlling for PMA, VMA and correct second pair matches on the FIST were correlated, \( r (20) = .45, p = .04 \). PMA and correct second pair matches on the FIST were not significantly correlated with PMA when VMA was controlled, \( r (20) = .15, p = .51 \). For a more detailed presentation of the partial correlation results, please see Table 3.

**Discussion**

The findings of this study on the relative contributions of CA, PMA, and VMA to performance on a task of cognitive flexibility task among 22 children, adolescents, and young adults with DS provide a more precise understanding of the contributors to the development of cognitive flexibility. The participants’ VMA was most strongly associated with performance on the second match, which is considered the measure of cognitive flexibility on this task. The finding that VMA was the variable that best accounted for performance on the cognitive flexibility match on the FIST using partial correlations suggests that verbal abilities might be primarily responsible for the development of cognitive flexibility at least among persons with Down syndrome. In contrast, the correlation between PMA and the FIST second match may be more reflective of variance that is shared with VMA, as the relationship between FIST second match and PMA was not significant when VMA was controlled. One indication of the extent of the deficits noted among the participants on the FIST related to the finding that even the participants with PMAs or VMAs of 60 months or greater were unable to reach ceiling levels of performance, as would be expected from reports of the performance of typically developing 5-year-old children (Jacques & Zelazo, 2001). This finding is consistent with evidence of a specific deficit in cognitive flexibility above and beyond intellectual abilities among persons with DS (Zelazo et al., 1996).

Despite the nuanced findings, this study must be considered preliminary, as it is limited in certain aspects. One, 21 of the 22 participants were female and gender differences might have affected the findings, although sex-related differences have yet to be reported for this task. Two, despite considerable evidence that visual-spatial abilities are typically stronger than verbal abilities among persons with DS (Chapman, 2006; Cleland et al., 2010; Silverman, 2007), no differences were found between the VMA and PMA scores. This discrepancy between the current findings and the literature may be due to the preponderance of female participants or, more likely, to our use of receptive vocabulary as the measure of verbal intelligence. Among individuals with DS, scores on measures of receptive vocabulary may be as high as on those of nonverbal processing (Chapman, 2006). Thus, a more comprehensive measure of verbal intelligence may be helpful. Three, the high correlations between scores on the Leiter-R and PPVT-III may call into question the

![Figure 2. Total correct FIST second matches by performance mental age in months.](image)

<table>
<thead>
<tr>
<th>Table 2</th>
<th>Correlations between CA, PMA, VMA, and FIST Performance</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>CA</td>
</tr>
<tr>
<td>CA</td>
<td>1.0</td>
</tr>
<tr>
<td>PMA</td>
<td></td>
</tr>
<tr>
<td>VMA</td>
<td></td>
</tr>
<tr>
<td>FIST first match</td>
<td>1.0</td>
</tr>
<tr>
<td>FIST second match</td>
<td></td>
</tr>
</tbody>
</table>

*Correlation is significant at the 0.005 level (using a Bonferroni correction).
extent to which the two measures assess separate abilities or domains of functioning. Thus, despite the fact that the Leiter-R and PPVT-III were both designed to assess different constructs (nonverbal intelligence and receptive vocabulary respectively), these two measures are often highly correlated. For example, in a longitudinal study, Roberts et al. (2007) found that the scores of 39 boys with DS on the PPVT-III and Leiter-R were correlated at a level, $r(39) = .87$, $p < .05$, similar both to typically developing boys, $r(41) = .76$, $p < .05$, and to the findings in the current study. Four, the participants’ CA varied considerably, but this does not seem that important since CA was not related to performance and because the more important scores of MA did not vary as much.

In summary, the current study found that VMA was the only age-related variable that was continued to relate to second-match performance on the FIST when PMA was controlled among a group of participants with DS with mean mental ages just higher than five years. This finding is particularly noteworthy in light of the fact that VMA and PMA were highly correlated. This finding highlights the potential role of verbal abilities as a contributing factor to the development of cognitive flexibility. The extent to which this pattern of results is also found among typically developing children or among persons with other syndromes remains to be determined. Unique relationships between different indices of developmental level and cognitive abilities among persons with DS as well as other syndromes associated with intellectual disability, such as fragile X syndrome (Cornish et al., 2001), phenylketonuria (Antshel & Waisbren, 2003) and Prader-Willi Syndrome (Walley & Donaldson, 2005) have the potential to be informative for understanding both cognitive development in these individual groups and the development of cognition more generally.

### References
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Résumés en Français

L’entraînement de la mémoire par ordinateur mène à des améliorations soutenues des habiletés visuo-spatiales de la mémoire à court terme chez les enfants avec un syndrome de Down

Stephanie J. Bennett, Joni Holmes et Sue Buckley


La flexibilité mentale chez des personnes avec un syndrome de Down : évaluation de l’influence des habiletés verbales et non verbales

Colin Campbell, Oriane Landry, Natalie Russo, Heidi Flores, Sophie Jacques et Jacob A. Burack

Les influences de l’âge mental verbal (AMV) et de l’âge mental de performance (AMP) sur la flexibilité mentale ont été évaluées auprès d’un groupe de participants avec un syndrome de Down (SD) pour départager l’apport relatif de chacun. La flexibilité mentale affaiblie typiquement observée chez les personnes avec un DS combinée à des développements inégaux de l’AMV et de l’AMP suggère une occasion de mieux comprendre la relation entre l’AMV, l’AMP et la flexibilité mentale. Nous avons examiné la performance au Flexible Item Selection Task (FIST), un test mesurant la flexibilité mentale d’enfants d’âge préscolaire, chez 22 participants ayant un SD. Des corrélations partielles révèlent que seule l’AMV était reliée au FIST en contrôlant pour l’AMP, soulignant le rôle des habiletés verbales dans le développement de la flexibilité mentale.

Profil du cortisol diurne chez des adultes présentant le syndrome de Williams en contexte nouveau et familier

Miriam Diane Lense, Andrew J. Tomarken et Elisabeth M. Dykens

Le syndrome de Williams (SW) est une maladie génétique associée à des taux élevés d’anxiété et des difficultés sociales. Nous avons examiné le cortisol diurne, un biomarqueur de la réponse au stress, chez des adultes présentant le SW en contexte nouveau et familier et nous avons comparé ces profils à ceux d’adultes ayant un développement typique (DT). Les participants SW et DT avaient des profils similaires en contexte familier. En contexte nouveau, les participants ayant un SW avaient un taux de cortisol plus élevé en fin de journée et lorsqu’il y avait davantage de demandes sociales. Le déclenchement de la réponse du cortisol chez les participants SW a été associé avec les plaintes somatiques et les difficultés sociales rapportées par les parents. Les résultats suggèrent que les adultes ayant un SW ont un profil de cortisol diurne typique qui peut être sensible aux transitions sociales et occupationnelles durant la journée.

Les effets de la reconnaissance des symptômes et des étiquettes diagnostiques sur les croyances du public, leurs réactions emotives et sur le stigma associé à la déficience intellectuelle

Katrina Scior, Theresa Connolly et Janice Williams

Les étiquettes sont fermement rejetées par le mouvement de la défense des droits des personnes handicapées et pourtant les effets complexes de l’utilisation d’étiquettes sur les croyances populaires sont peu compris. Cette étude examine les effets des étiquettes diagnostiques sur les réactions du public à l’égard des personnes ayant une déficience intellectuelle. Une
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