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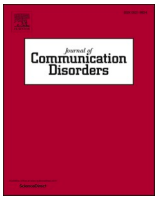

Cognitive flexibility in children with Developmental Language Disorder: Drawing of nonexistent objects

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ABSTRACT

Cognitive flexibility is the ability to adapt thoughts and behaviors to new environments. Previous studies investigating cognitive flexibility in children with Developmental Language Disorder (DLD) present contradictory findings. In the current study, cognitive flexibility was assessed in 5- and 6-year-old preschoolers with DLD ($n = 23$) and peers with typical development (TD; $n = 50$) using a nonexistent object drawing (NEOD) task. The children were asked to draw a nonexistent man and a nonexistent house. The children with DLD did not differ from their peers with TD on simple category changes, which were comprised of changes in the size or shape of parts of the object, change of the whole shape of the object, and deletion of parts of the object. Nevertheless, children with DLD made fewer more complex, high-level category changes, which included same-category insertions, position exchange of object's parts, and cross-category insertions. The difference between DLD and TD on high-level category changes was related to differences between the two groups in verbal short-term memory and inhibition. Furthermore, children with DLD made no changes to their original drawings of an existing man and house more often than their peers with TD. It is concluded that children with DLD aged 5–6 years show less flexibility on the NEOD task than age-matched children with TD. This difference in cognitive flexibility may be related to lower levels of verbal short-term memory and inhibition ability of children with DLD, or to different use of these cognitive skills on the NEOD task.

1. Introduction

Children often need to adapt their thoughts and behaviors to changing situations in their everyday life. In order to do this, they need cognitive flexibility. Cognitive flexibility comprises one of the executive functions (Miyake & Friedman, 2012; Miyake et al., 2000). Executive functions are a set of general-purpose control processes that regulate a person's thoughts and behaviors and include inhibition and updating of the working memory contents, in addition to cognitive flexibility. Deficits in cognitive flexibility have been associated with several neurodevelopmental disorders, including Developmental Language Disorder (e.g., Farrant, Maybery, &

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Fletcher, 2012; Kapa, Plante, & Doubleday, 2017; Roello, Ferretti, Colonnello, Levi, & 2015). However, other studies found that cognitive flexibility is spared in children with Developmental Language Disorder (e.g., Henry et al., 2011; Im-Bolter, Johnson, & Pascual-Leone, 2006). Given the inconsistent findings across studies, additional research is needed to gain a clearer picture of cognitive flexibility in children with DLD. The current study is the first to study cognitive flexibility in children with Developmental Language Disorder using a nonexistent object drawing task (Karmiloff-Smith, 1990).

2. Cognitive flexibility

Cognitive flexibility is the human ability to adapt the cognitive processing strategies, responses and representations to new and unexpected conditions in the environment (Cañas, Quesada, Antolí, & Fajardo, 2003; Legare, Dale, Kim, & Deák, 2018). Cognitive flexibility is used for switching between tasks, changing perspectives and thinking outside the box (Diamond, 2013). Other terms for cognitive flexibility include attentional flexibility or set-shifting/task-switching. The term attentional flexibility is typically used in studies that focus on the processes required for shifting attention. Studies that use notions such as set-shifting or task-switching define cognitive flexibility by the task used to measure it, i.e. a set-shifting or task-switching task (Dajani & Uddin, 2015). Cognitive flexibility is associated with better reading abilities in childhood (de Abreu et al., 2014), higher resilience (Genet & Siemer, 2011) and levels of creativity in adulthood (Chen et al., 2015), and better quality of life in the elderly (Davis, Marra, Najafzadeh, & Liu-Ambrose, 2010).

According to the well-known unity/diversity framework, cognitive flexibility is one of the executive functions (Miyake & Friedman, 2012; Miyake et al., 2000; note that Miyake and colleagues refer to the construct as 'shifting'). Research has shown that children as young as 3–4 years old can already successfully shift between two simple response sets, provided that rules are placed in a story context (Hughes, 1998) or demands on inhibition are reduced (Rennie, Bull, & Diamond, 2004). Various studies reported significant growth in cognitive flexibility between the ages of 3 and 6 years (Deák, 2000; Zelazo, Müller, Frye, & Marcovitch, 2003; Zelazo, Frye, & Rapus, 1996). Other research indicates a sharp increase between ages 7 and 9 years (Dick, 2014). The development of cognitive flexibility appears relatively gradual compared to inhibition, which has been found to undergo a strikingly strong increase in the preschool years and less growth at later ages (Best & Miller, 2010).

Most studies investigated children's cognitive flexibility using set-shifting tasks (Dajani & Uddin, 2015; Legare et al., 2018), which assess the ability to change previously learned behaviors if these are no longer relevant. Two tests are commonly used in research with kindergarteners, which is the population targeted in the current research: (1) Dimensional Change Card Sort (DCCS) in which cards are sorted using a simple rule, such as sorting cards based on color. After a number of items, the card sorting rule changes and children are asked to sort them based on shape. (2) Flexible Item Selection Task (FIST), which differs from the DCCS in that instead of telling children the rule explicitly, they need to generate it from a visual display. The DCCS and FIST are typically (computerized) tasks in which children are presented with a large number of items suited to their age, to which they respond as fast as possible. Outcomes are measured as accuracy. The DCCS also allows calculation of switch costs, which is the difference in response times between switch and non-switch trials.

Cognitive flexibility has also been tested in an entirely different way, using a nonexistent object drawing (NEOD) task (Low, Goddard, & Melser, 2009; Spensley & Taylor, 1999a,b; Ten Eycke & Müller, 2018). When drawing, children typically use schemata based on sequentially ordered and practiced movements. In NEOD tasks, they are asked to "draw X", and then to draw "a nonexistent X, " such as an X that they invent, that they have never seen before, a strange X, an X with something funny or odd about it (Karmiloff-Smith, 1990; Spensley & Taylor, 1999a,b). To solve this task, children need cognitive flexibility to modify and alter the procedurally encoded schemata. NEOD tasks are particularly interesting because, unlike set-shifting tasks, they are production tasks where children's output provides information on how they solve tasks in which something new and unexpected is asked of them. When solving a NEOD task, children can make size, shape or deletion changes, change the location of elements, and add or insert elements. These different types of modifications can be classified into inter-representational or intra-representational flexibility and complexity (Berti & Freeman, 1997; Spensley & Taylor, 1999a,b; Zhi, Thomas, & Robinson, 1997). Inter-representational flexibility refers to cross-category insertions, that is, combining components of different categories in one drawing, such as a house with wings. Intra-representational flexibility is observed when children make changes within the components of a category, such as a man with two heads.

Karmiloff-Smith (1990) concluded that different types of modifications are associated with different ages and development phases. She tested fifty-four children between the ages of 4 and 11 years, where each child produced six drawings. Children aged 4–6 years ($n = 22$) spontaneously made simple intra-representational changes: they modified size or shape, or deleted parts. Eight to 10-year-old children ($n = 32$) more often also made complex intra-representational changes: exchanging the position of elements, same-category insertions (such as a man with two heads), and inter-representational changes (cross-category insertions). The former three categories (changes of size, shape, deletion) were classified by Karmiloff-Smith as representing simpler low-level changes, while the latter three categories (element exchange, same-category insertion, cross-category insertion) represent more complex changes. In this study, Karmiloff-Smith also found that younger children drew their original schema again, and did not modify their drawing, more often than older children.

3. Cognitive flexibility and Developmental Language Disorder

For the purpose of the current study, children with Developmental Language Disorder (DLD) were given a NEOD task. DLD is a developmental disorder involving delayed language in the absence of any obvious cause such as a lack of environmental stimulation, hearing impairment, low intelligence, or neurological damage (Leonard, 2014). Although DLD primarily affects children's language

ability, there is increasing evidence that children with DLD have difficulties with domain-general executive functions (sustained attention: Ebert & Kohnert, 2011; visuospatial working memory: Vugs, Cuperus, Hendriks, & Verhoeven, 2013; inhibition and shifting: Pauls & Archibald, 2016; Vissers, Koolen, Hermans, Scheper, & Knoors, 2015). Executive function weaknesses are most prominent in verbally-loaded and more demanding tasks (Lukács et al., 2016; Montgomery, Magimairaj, & Finney, 2010; Noterdaeme et al., 2000; Spaulding, Plante, & Vance, 2008).

Findings regarding cognitive flexibility are not unequivocal. Some studies show that the performance of children with DLD is lower than the performance of peers with typical development (TD) on nonverbal inhibition and working memory, but is similar on set-shifting tasks (e.g., Im-Bolter et al., 2006; Henry et al., 2011). These studies investigated children who were 7–12 years old, with an average age of 10–11 years. Other studies that investigated younger preschool children found that children with TD outperformed children with DLD on a DCCS task (Farrant et al., 2012; Kapa et al., 2017) and on a FIST task (Roello et al., 2015). The discrepancy between these results may suggest that children with DLD have protracted cognitive flexibility development and/or later onset of cognitive flexibility development, which is reflected in lower scores at younger ages, but score similar to their peers at older ages. However, Dibbets, Bakker and Jolles (2006) investigated 6-year-old children and found no evidence for differences in task switching between children with DLD and TD. In addition, a meta-analysis of 22 studies (and 29 different samples) with children between ages 4 and 14 years did not point to age as a moderator of cognitive flexibility differences between DLD and TD (Pauls & Archibald, 2016). This meta-study revealed that children with DLD tend to perform lower than their peers with TD on tasks testing cognitive flexibility, but the size of the effect was small, while inhibition showed a moderate group effect. Pauls and Archibald suggested that the small group effect found for cognitive flexibility may, in fact, be driven by an inhibition deficit, as successful shifting from one task to another requires suppression of former tasks.

There is, to the best of our knowledge, no previous research that used a NEOD task to investigate cognitive flexibility abilities of children with DLD. Both NEOD and set-shifting tasks measure children's ability to adapt cognitive processing strategies, responses and representations to new and unexpected conditions. However, these tasks differ considerably. First, unlike DCCS and FIST tasks, the NEOD task is a production task. Second, set-shifting tasks are relatively fixed, i.e. the rules and representations that children shift between are more or less given, contrary to the NEOD task in which the representation to which a child switches is open, and the only requirement is that it is nonexistent. Furthermore, set-shifting tasks focus on the attentional processes required for cognitive flexibility more than the NEOD task, whereas the NEOD task taps into those aspects of cognitive flexibility that have to do with imagination (Ten Eycke & Müller, 2018). Comparing DLD and TD on the NEOD task can thus provide more insight into how DLD and TD differ with respect to the broad construct of cognitive flexibility.

4. The current study

As part of the current research, we aimed to determine whether differences in cognitive flexibility will be found in children with DLD compared to age-matched children with TD by using a NEOD task. This task has the potential to identify a difference in cognitive flexibility between the two groups, and enables determining whether children with DLD solve the task by using a strategy that is more typical of younger children with TD. We did not collect data from younger children with TD and were therefore unable to make direct comparisons with younger children with TD. However, comparisons with observations in previous research may enable us to cautiously interpret the drawings of the children with DLD developmentally, that is, whether they reflect an earlier developmental phase. The research question that guided our study was: Do children with DLD and children with TD differ in cognitive flexibility as measured by a NEOD task?

We expected indications of a protracted cognitive flexibility development for the children in our study, who were 5 to 6 years old, based on their performance on the NEOD task. Specifically, we hypothesized that:

(a) Children with DLD are more likely to make no modifications or simple intra-representational modifications (low-level change categories) compared to their peers with TD, because such changes are linked to younger ages (Karmiloff-Smith, 1990).
(b) Children with DLD are less likely to make complex intra-representational modifications (i.e., higher level within-category insertions) than their peers with TD.
(c) Children with DLD are less likely to make inter-representational modifications (cross-category insertions) than their peers with TD, because such changes appear to reflect more advanced cognitive flexibility (Adi-Japha et al., 2010).

Cognitive flexibility as measured using the NEOD task could be associated with other factors that are also likely to differ across DLD and TD, and that could create a confound. We considered four additional factors in our analyses in order to explore the robustness of the differences between TD and DLD on the NEOD task: basic drawing skills, nonverbal intelligence, verbal short-term memory (verbal STM) and inhibition. Children with DLD may score lower on basic drawing skills because this requires motor skills, and previous research has shown that these children have motor weaknesses (Diepeveen, van Dommelen, Oudesluys-Murphy, & Verkerk, 2018; Johnston & Weismer, 1983). To test for a possible motor mechanism related to the basic drawing level, we assessed visual-motor integration, and tested whether visual-motor integration contributed to group differences in basic drawing skills. We hypothesized that although visual motor skills contribute to group differences in basic drawing, they would not fully explain these differences, and that the basic drawing level (rather than visual motor skills) should thus be used as a motor predictor for explaining group differences in the NEOD. We therefore controlled for basic drawing skills when testing cognitive predictors for group differences in the NEOD change categories.

We considered three cognitive measures as predictors for group differences in the NEOD task: nonverbal intelligence, verbal STM,

and inhibition.

Nonverbal intelligence describes thinking skills and problem-solving abilities that do not fundamentally require verbal language production and comprehension. It involves manipulating or problem-solving about visual information (Kuschner, 2013). Children with DLD score within the normal range on nonverbal intelligence, although they tend to score somewhat lower than TD controls in between-group comparisons (Gallinat & Spaulding, 2015). Nonverbal intelligence has been found to be related to cognitive flexibility (Arffa, 2007).

Verbal STM is the ability to temporarily retain phonological information in mind and is typically impaired in children with DLD (Archibald & Gathercole, 2006; Weismer et al., 2000). Because of their verbal STM deficits, children with DLD may be less able to retain verbal instructions than their peers with TD, which may affect performance in nonverbal tasks with verbal instructions (Pauls & Archibald, 2016). For example, previous research showed that when verbal STM is controlled, differences in executive function tasks between children with TD and with DLD disappear (Lukács et al., 2016), and the same may be true for the NEOD task.

Inhibitory control has been suggested to involve three processes: inhibition of a prepotent response, resistance to distracters, and resistance to interference from prior knowledge (Friedman & Miyake, 2004). Adaptation to a new environment and responding to new conditions is central to cognitive flexibility, and requires that previously engaged responses be inhibited (Dajani & Uddin, 2015). For example, in the NEOD task, children must inhibit typical objects drawing schemata they have just used in order to produce the unusual drawing of the same object. It is thus expected that inhibition of a prepotent response is involved in the NEOD task, and may impact any differences between the performance of children with DLD and with TD on the NEOD task, because children with DLD tend to have lower inhibition than children with TD (Pauls & Archibald, 2016).

The following hypothesis was formulated regarding the cognitive predictors:

(a) For the assessed cognitive variables, we predicted that the DLD group would perform lower than the TD group on verbal STM and inhibition. Furthermore, nonverbal intelligence, verbal STM and inhibition were expected to be associated with NEOD modifications, and better verbal STM and inhibition abilities were expected to be associated with a higher probability that the child would actually make a high-level modification (complex intra-representational and inter-representational modifications) and with a higher frequency of use of such high-level modification, as these high-level modifications are more pronounced in older ages and reflect more advanced cognitive flexibility.

5. Methods

5.1. Study approval

The study was approved by the Israeli Ministry of Education (287/8918/2015). Consent was obtained from the parents of the participating children.

5.2. Participants

The sample included 73 kindergarten children aged 5–6 years ($M = 69.92$ months, SD = 3.49 months, see Table 1): 23 children with DLD and 50 children with TD. Participating children were from kindergartens in the same municipal area. The DLD sample included 12 boys and 11 girls. The TD sample consisted of 24 boys and 26 girls. In terms of gender, the groups were similarly composed ($\chi^2(1) = 0.11$, $p = .74$).

Children with DLD were recruited from language kindergartens. Children are admitted to a language kindergarten based on significant primary language impairment, normal nonverbal intelligence, and sound adaptive behavior skills. Children are referred to a language kindergarten by a placement committee. A referral to the placement committee is made following a recommendation by a child neurologist or a clinical or educational psychologist and a diagnosis of a speech therapist (Ministry of Education, 2020). All children referred to the placement committee are administered the Wechsler Preschool and Primary Scale of Intelligence (WPPSI-III;

Table 1
Descriptive statistics of the TD and DLD groups.

	TD $n = 50$		DLD $n = 23$		Wilcoxon Z	Effect size (r)
	M	SD	M	SD		
Age	70.32	3.25	69.04	3.90	0.98	0.01
Nonverbal intelligence	101.20	10.76	97.34	14.38	1.48	0.29
Language composite	0.79	0.97	-2.92	1.99	6.76***	0.79
Basic drawing level	2.69	0.44	2.46	0.79	3.93***	0.46
Verbal STM	12.10	3.42	6.52	3.37	5.09***	0.60
Inhibition	13.46	2.06	10.04	2.51	4.78***	0.55
Visual-motor skills	99.68	8.10	95.3	4.45	2.41*	0.28

Note. *$p < .05$, ***$p < .001$. Language composite is a Z-score based on the Goralnik test; Nonverbal intelligence was measured by Raven Standard Scores (M=100, SD=15); Basic drawing level was measured on a 1-4 scale; Verbal STM was measured with Kaufman ABC subtests (M=10, SD=3); Inhibition = number of accurate responses out of 16; Visual-motor skills were measured with Beery-VMI and represented by standard scores (M=100, SD=15). $r = Z/\sqrt{N}$.

Wechsler, 2002). Children are diagnosed as having a language disorder by a speech therapist based on a score of more than 1 SD below the mean in at least two language tests that are used in clinical practice in Israel and who have a normal performance IQ (Friedmann & Novogrodsky, 2004). The placement committee (comprised of the special education kindergarten district supervisor, municipal kindergarten psychologist, and special education therapists) tests eligibility based on standardized cognitive assessments, speech-language pathologist referral, and information from previous kindergarten teachers and caregivers that it receives.

Children with TD were recruited from regular kindergartens. Kindergarten teachers identify children at risk for a developmental delay in the first 3 months of the school year (Ministry of Education, 2019). Children identified by their teacher as "at risk" in the TD group and for developmental delays other than language in the DLD group were not included in the study.

All children participating in the current study were Hebrew-speaking monolinguals and scored within the normal range of the Raven test (Stand. Score ≥ 80, see Table 1). Language kindergarten children were identified as having DLD if they scored $Z \leq 1.25$ SDs below the norm on the Goralnik Screening Test for Hebrew (Goralnik, 1995). The Goralnik test assesses children's abilities in Hebrew and includes subtests for vocabulary, sentence repetition, comprehension, oral expression, pronunciation, and story-telling (see below). Two of 25 language kindergarten children we assessed scored above this range and did not participate in the study. Table 1 presents the participating children's overall Z scores for normative data, but raw scores were used in the analyses.

5.3. Measures

Cognitive flexibility (NEOD task). The children were first asked to draw a man using an HB pencil or felt-tip pen (one color). They were then asked to draw "a man that does not exist" (Karmiloff-Smith, 1990). Several phrasings were used to enable the children to understand the task: "A man you invent, one you have never seen before, a strange man, with something funny or odd, something make-believe, pretend." This phrasing, following Spensley and Taylor (1999a,b), is more elaborate than the original phrasing used by Karmiloff-Smith (1990), and includes an explicit request to add something to the man. All children heard the same instructions. After drawing the nonexistent man, the children were asked to verbalize why such a man does not exist. To give generality to the data, the children were also asked to draw a house and a house that does not exist (Kasirer et al., 2020; Adi-Japha et al., 2010).

Following the procedure developed by Karmiloff-Smith (1990), two independent raters scored the categories of changes compared with the original drawing as no change, deletion of elements, change in element shape or size, whole-shape changes, insertion of new (same-category) elements, position or orientation changes, and cross-category insertions. As in the original study, the categories were not mutually exclusive. Cohen's kappa coefficients for inter-rater agreement were greater than .90 ($p < .001$) in each change category. Disagreements were settled by discussion with an additional experienced rater.

In her study, Karmiloff-Smith (1990) found that changes made by older children (8–10 years old versus 4–6 years old) included more frequent insertions of new (same-category) elements, position or orientation changes, and cross-category insertions. These can thus be considered as reflecting a higher level of cognitive flexibility. For this reason, we grouped insertion of new (same-category) elements, position or orientation changes, and cross-category insertions into "high-level change categories". Deletion of elements, change in element shape or size, and whole-shape changes were combined into "low-level change categories". There were thus 6 possible change categories: 3 low-level change categories and 3 high-level change categories. The children could make multiple changes in one drawing and categories were not mutually exclusive.

Basic drawing level. The complexity level of the original man/house drawing was scored on a scale of 1–4: 1 = non-recognizable object; 2 = a recognizable figure composed of two line objects (e.g., in the house drawing of a rectangle and a triangle above); 3 = a recognizable figure composed of three line objects, of which two are integrated (e.g., in the man a body, head and eyes within the head); 4 = a recognizable figure that includes more complex graphic formulas (e.g., figures composed by four or more line objects of which three are integrated, as in a house drawing with a rectangular house and a triangular roof, with a cross within a window within the house, or a 3-dimensional drawing, see Adi-Japha et al., 2010, Kellogg, 1970). The two independent raters who scored the drawings for the change categories also scored the two drawings for complexity. Weighted Cohen's kappa coefficient for an ordinal scale were .92 ($p < .001$), on average, for the two drawings. Disagreements were settled by discussion with an additional experienced rater. The score of the two drawings was averaged.

Verbal STM. The forward number recall test for children consists of predefined sets of random strings of numbers of increasing length (children: K-ABC; Kaufman & Kaufman, 1983) and tests verbal STM. Participants repeat the string in the same order. Testing continues until the participant makes two consecutive errors in a same-length set. This version of the K-ABC was adapted for Hebrew and has been normed in Israel (M = 10 and SD = 3 for each subtest). On average, test-retest and internal consistency reliabilities of the K-ABC subtests were reported as 0.85 and 0.62, respectively (Phizer, Shimborsky, Walf, & Hazani, 1995).

Inhibition. The inhibition task resembled a go/no-go task. The stimuli were presented on a 16" laptop screen. The task involved 16 repeats of blocks of 3 stimuli (overall 48 stimuli) consisting of one of 6 aquarium animals (a yellow-blue striped fish, a green fish, a jellyfish, a starfish, a sea turtle, and an octopus) appearing in a random location on the screen for 3 s. The time between stimuli was 3–6 s, and the children had up to 3 s to respond. The children were asked to respond as rapidly and accurately as possible, using a computer key labeled in yellow. The children were instructed to tap the yellow key only when the yellow blue-striped fish appeared. They were told that other animals may appear as well, and that in that case they should not strike any key. The yellow-blue striped fish appeared with a probability of 2/3 and the other animals had equal probabilities of about 7% to appear. This task thus stresses inhibition of prepotent response. Each block included 2 repeats of the go stimuli and one no-go stimulus in a random order. This ensured that there were no more than two successive trials with a no-go stimulus (Howard & Okely, 2015). Prior to the task, the children practiced on 1 block. Accuracy (number of correctly responded blocks) was coded.

A block was scored as accurate if the child correctly pressed the button for the (two) go stimuli and rejected the (one) no-go

stimulus. We were unable to single the no-go stimulus, because the number of button presses to these stimuli was not recorded. However, as the prepotent response was a button press of the yellow button, it is likely that incorrect blocks were due to a button press of the yellow key to the (one) no-go stimulus in that block. It has been suggested that in go/no-go tasks the go trials index sustained attention while the no-go trials index actual inhibition processes (Ashley et al., 2019; Lewis et al., 2017; Willner et al., 2015). It may therefore be suggested that the current task is not a pure measure of inhibition, but rather a mix of inhibition and sustained attention.

Visual-motor skills. The Beery-VMI is a standardized test ($M = 100$, SD = 15) that evaluates visual-motor integration skills (often associated with copying, e.g., Ogawa, Nagai, & Inui, 2010) for children aged 2 years to adult. Participants copy progressively difficult geometric shapes. The test is stopped after subjects fail to correctly copy three consecutive shapes. The final score is the number of correct shapes copied. Overall test-retest and inter-rater reliabilities were reported, .84–.88 and .93–.98, respectively (Beery, Buktenica & Beery, 2006).

Language assessment. The Goralnik Screening Test for Hebrew (Goralnik, 1995) was administered in order to assess proficiency in Hebrew. The test includes subtests for vocabulary, sentence repetition, comprehension, oral expression, pronunciation, and story-telling. The Goralnik test was designed to screen monolingual Hebrew-speaking children aged 2;7 - 6;0 who are at risk for DLD. The scores are raw scores, with a total of 180 points. The Goralnik manual enables calculation of a standardized Z-score based on age-appropriate norms, used for identifying DLD (Goralnik, 1995; Altman, Armon-Lotem, Fichman, & Walters, 2016). Participating children identified with DLD scored $\leq$1.25 SD below the mean, while children with TD scored $\geq$0.9 SD below the mean. The Goralnik test provides norms in 6-month intervals. The last norm is for children aged 67–72 months. We used this norm for children aged 73–78 months, as these children share the same educational level (Kindergarten).

5.4. Procedures

The children were tested individually in a quiet room in the kindergarten by R.B or N.S. The tests were administered in a fixed order and the children were tested in two separate sessions as part of a larger study (Adi-Japha, Berke, Shaya, & Julius, 2019). Background variables were tested before the flexibility task. The first session involved the language, verbal STM and inhibition assessments. The second session included the assessment of the visual-motor skills and the cognitive flexibility task.

5.5. Data analysis

The NEOD task has an ordinal scale for the basic drawing level and a nominal scale for the change categories. Language scores were negatively skewed. Non-parametric statistics was therefore preferred. We first checked how the two groups scored on background measures: language, nonverbal intelligence, basic drawing level, verbal STM, inhibition, and visual-motor skills. We expected that the children with DLD would perform lower on language than the children with TD, but similarly on nonverbal intelligence, conforming to the DLD profile. We also expected the children with DLD to perform lower than the children with TD on visual-motor skills, basic drawing level, verbal STM and inhibition. The Wilcoxon Z-test was used in order to establish group differences. $r = Z/\sqrt{N}$ was used to estimate the magnitude of the effect (with r = .1, small effect; .3, medium effect, .5, large effect; Rosenthal, 1994, pp. 231–244; Pallant, 2007, p. 225).

The results of the drawing test were compared across TD and DLD in order to investigate whether and how DLD is associated with cognitive flexibility. A χ^2 test was applied to compare the number of children who made no changes across the two groups. We also indicated whether or not each child used a change category at least once across the two drawings (nonexistent man and house), resulting in a binary 0/1 variable per category. A χ^2 test was used to compare the number of children who used each category, and the number of children who used at least one of the low- and high-level change categories.

Spearman correlations and hierarchical (linear as well as logistic) regression analyses were conducted to determine whether any of the differences between the TD and DLD groups were affected by the inclusion of background measures on their own or in combination with others, i.e. basic drawing level, nonverbal intelligence, verbal STM, and inhibition. Three regression models were compared: Model 1 included basic drawing level as a predictor for determining whether the factor Group (TD, DLD) explained any variance beyond basic drawing level. Model 2 included basic drawing level and nonverbal intelligence as predictors in addition to Group. Model 3 included basic drawing level, nonverbal intelligence, and verbal STM or inhibition as predictors in addition to Group. Because verbal STM and inhibition were highly correlated ($r = 0.67$, $p < .001$), we tested them in two separate models (3A and 3B).

6. Results

The descriptive results in Table 1 show how the TD and DLD groups compare in terms of language, nonverbal intelligence, basic drawing level, verbal STM, inhibition, and visual-motor skills. The two groups did not differ in age and nonverbal intelligence.

However, in addition to language scores, verbal STM and inhibition, the groups differed in their visual-motor skills and basic drawing level. Basic drawing skills and visual-motor skills were significantly correlated across the full sample (TD and DLD, r_s (73) = 0.39, $p = .001$). Ordinal logistic regression applied to the sum of scores of the basic drawing level across the two drawings indicated that visual-motor skills significantly contributed to the variability in basic drawing level (B = 0.50, SD = 0.22, $W^2 = 5.08$, $p = .024$), but did not eliminate the contribution of the group variable (B = 1.58, SD = 0.60, $W^2 = 7.20$, $p = 0.007$).

Table 2 presents the frequency (per child) of a specific change category across the two drawings (continuous variable). Use of change categories was scored per drawing, and children could score 0–2 for each change category across the two drawings. Table 2 also

indicates the number of children who exhibited use of each category at least once across the two drawings (a binary 0/1 variable, for example 12 out of the 50 children with TD displayed same-category insertion at least once in their drawings). Because the frequency counts were low for several change categories, only the binary score (i.e., the number of children who exhibited that change category) was compared between groups.

The binary score was compared between the two groups in order to study the hypotheses that children with DLD are more likely to make no modifications or simple intra-representational modifications (low-level change categories) compared to their peers; that they are less likely to make complex intra-representational modifications (exchanging the position of elements and same-category insertions) as well as inter-representational modifications (cross-category insertions) than their peers with TD (Table 2). As mentioned, change categories in Karmiloff-Smith's (1990) developmental study were grouped into low- and high-level change categories, where the former is typical of 4–6 year-olds, and the latter is more common in older children. Table 2 also addresses group difference in low- and high-level change categories. χ^2 tests comparing the number of children who used at least one of the low- and high-level change categories did not indicate differences for the low-level change categories, but did indicate group differences in the high-level change categories. Specifically, more children in the TD group used same-category insertions (a type of complex intra-representational modification) and cross-category changes (inter-representational modification) than children in the DLD group.

Fig. 1 shows typical examples of drawings made by the children with DLD and their peers with TD in the NEOD task. Fig. 1A–C shows examples of drawings with same-category insertions, and Fig. 1E and F shows examples of drawings with cross-category insertions. These drawings were made by children with TD. Fig. 1G–I are examples of drawings with deletions made by children with DLD. NEOD categories are not mutually exclusive, for example, Fig. 1D involves deletion of windows.

The NEOD data in the current study further indicated that relatively more children in the DLD group (6/23 = 26.08%) made no change in their drawings of a man and a house than in the TD group (3/50 = 6% TD) (χ^2 (1) = 5.88, p = .015). It should be noted that the 6 children with DLD who made no change in their drawings did not differ from the other children in the DLD group in terms of language (Z = 0.11, p = .919) or nonverbal IQ scores (Z = 0.17, p = .878). Furthermore, the data indicated that in the TD group 13/50 children (26%) and in the DLD group 10/23 children (43.47%) made only low-level modifications (no significant difference, χ^2 (1) = 2.23, p = .135); in the TD group 29/50 children (58%) and in the DLD group 7/23 children (30.43%) made both low- and high-level changes (a higher proportion in the TD group, χ^2 (1) = 4.78, p = .029); and finally, in the TD group 5/50 children (10%) (and none of the children with DLD) made only high-level modifications to their drawings (χ^2 (1) = 5.33, p = .021).

Due to the low frequency of specific change categories, the analysis of background predictors to performance on the NEOD task related only to high- vs. low-level change categories. Table 3 presents Spearman correlations between background measures and use of all change categories (overall, how well children solved the task), low- or high-level change categories for children with DLD and their peers with TD. There were no significant group differences in the level of correlations between background variables and overall use of change categories (tested using the Fisher r-to-z transformation). No significant group differences emerged for the correlation between background variables and low- or high-level changes. There was, however, a non-significant trend toward a stronger association between the inhibition score and the frequency of low-level changes in children with DLD compared to their peers with TD (Z = 1.81, p = .070). It should be noted that performance in the NEOD did not correlate with the standardized language score (r_s (50) = .22, p = .127), r_s (23) = .25, p = .249).

The interpretation of the correlation of high-level changes in the DLD group is not clear due to the zero-clustered data with 16/23 children having the value 0 (Huson, 2007). To verify the association, we also conducted a Mann-Whitney test to compare the values of NEOD predictors between children with DLD who have (n = 7) and do not have high-level changes (n = 16). The results confirmed the pattern of significant associations in Table 3, with Z = 0.58, p = .624; Z = 1.52, p = .135; Z = 0.00, p = 1.00; and Z = 3.04, p = 0.010, for basic drawing level, non-verbal intelligence, verbal STM and inhibition, respectively.

Table 4 (left hand side) shows the results of regression models testing whether any difference between the TD and DLD group in the number of high-level changes drawn by children were affected by the inclusion of background measures (i.e., basic drawing level, nonverbal intelligence, verbal STM, and inhibition) on their own or in combination with others. Linear regression models suggest that nonverbal intelligence contributed to the explained variance, and the addition of verbal STM or inhibition made a further contribution and resulted in the disappearance of the difference between the TD and DLD groups. Logistic regression (right hand side of Table 4) pertaining to the difference between the number of children in the TD and DLD groups who had (i.e., a binary variable = had/did not

Table 2
Frequency of changes by group and category for the nonexistent object drawing task.

	TD n=50			DLD n=23				
	M	SD	n	M	SD	n	χ^2	P
Low-level change categories	1.40	0.98	42	1.69	1.36	17	1.03	.309
Size or shape of item	0.62	0.72	24	0.43	0.72	7	1.98	.158
Change of whole shape	0.20	0.45	9	0.39	0.72	6	0.63	.427
Deletion	0.58	0.70	23	0.87	0.76	15	2.33	.127
High-level change categories	1.26	1.04	34	0.30	0.47	7	9.03	**.003**
Same-category insertion	0.26	0.48	12	0.04	0.20	1	4.16	**.041**
Orientation	0.16	0.46	6	0.04	0.20	1	1.06	.302
Cross-category change	0.84	0.84	28	0.22	0.42	5	7.46	**.006**

Note. χ^2 tests compared the number of children who used each category.

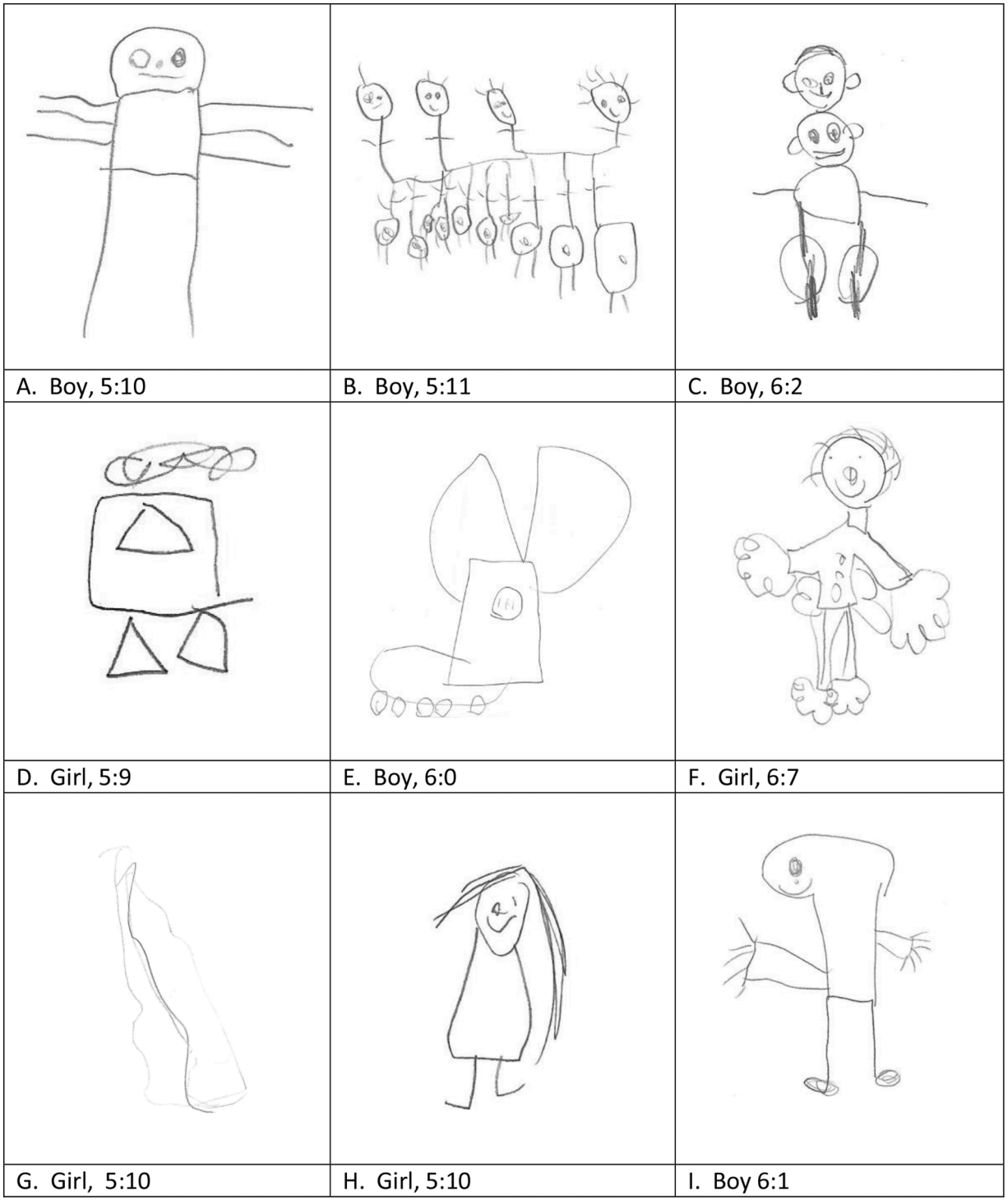

Fig. 1. Drawing examples. A-F, children with typical development (no language disorders); G-I children with a developmental language disorder. A. "He has 3 hands, actually 6, because 3 and 3 are 6". B. "A man with 4 heads, 4 necks, many hands and belly buttons". C. "A man with huge knees (laughs), a belly and hands, 2 heads and 4 eyes". D. "The house has hair, there is a triangle inside this house, and it has legs". E. "It has wheels and ears, and there are people in the house (points to the window)". F. "The man flies, he has wings". G. "A house with a 'delete' line". H. "She has no hands". I "The eye is one".

Table 3
Correlations between background measures and dependent variables.

Spearman r	Basic drawing level		Nonverbal intelligence		Verbal STM		Inhibition	
	TD	DLD	TD	DLD	TD	DLD	TD	DLD
	$n = 50$	$n = 23$	$n = 50$	$n = 23$	$n = 50$	$n = 23$	$n = 50$	$n = 23$
All change categories	.06	.24	.27[a]	.27	.32*	.17	.31*	.58**
Low-level change categories	.05	.24	.07	.22	.16	.20	.04	.48*
High-level change categories	.05	.11	.33*	.32	.32*	.00	.36*	.65**

Note. $p = .056$ *$p < .05$, **$p < .01$, ***$p < .001$.

Table 4
Regression analyses explaining the high-level changes across the TD and DLD groups.

	High-level changes by number of changes (averaged across two drawings)				High-level changes by child			
Model 1	B	SE	t	p	B	SE	OR	p
Basic drawing level	.08	.09	0.87	0.386	.19	.63	0.67	.413
Group	.42	.13	3.13	**.003**	.66	.31	4.57	**.032**
	$R^2 = .206$, $p < 0.001$.				$\chi^2 = 9.83$, $p = .007$; Cox & Snell $R^2 = .126$			
Model 2								
Basic drawing level	.06	.09	0.70	.487	.16	.24	0.46	.496
Nonverbal intelligence	.07	.03	2.51	**.015**	.22	.08	7.09	**.008**
Group	.39	.13	2.99	**.004**	.66	.33	3.81	**.051**
	$\Delta R^2 = .066$, $p = 0.015$; $R^2 = .272$				$\Delta\chi^2 = 8.17$, $p = .004$; $\chi^2 = 18.00$ Cox & Snell $R^2 = .218$			
Model 3A								
Basic drawing level	.05	.09	0.52	.625	.15	.35	.34	.561
Nonverbal intelligence	.07	.03	2.22	**.029**	.22	.09	6.30	**.012**
Verbal STM	.09	.04	2.18	**.033**	.26	.11	5.15	**.023**
Group	.23	.15	1.55	.125	.18	.40	.20	.654
	$\Delta R^2 = .047$, $p = 0.033$; $R^2 = .320$				$\Delta\chi^2 = 5.56$, $p = .018$; $\chi^2 = 23.56$ Cox & Snell $R^2 = .276$			
Model 3B								
Basic drawing level	-.01	.09	-.14	.885	-.08	.28	.09	.768
Nonverbal intelligence	.05	.02	1.92	**.058**	.20	.09	4.45	**.035**
Inhibition	.15	.05	3.12	**.003**	.55	.16	11.68	**.001**
Group	.20	.13	1.48	.141	.12	.39	.09	.755
	$\Delta R^2 = .092$, $p = 0.003$; $R^2 = .364$				$\Delta\chi^2 = 14.88$, $p < .001$; $\chi^2 = 32.88$ Cox & Snell $R^2 = .370$			

have) high-level change categories in their drawings was used to account for the non-linear distribution of the number of high-level changes drawn by children. Nevertheless, the same findings were found in the two analyses (Ghanamah, Eghbaria-Ghanamah, Karni, & Adi-Japha, 2020). It should be noted that the linear regression suggests a model for predicting the frequency of use of high-level change categories, where the R^2 improved from .206 with just Group as predictor to .320 in Model 3A (.364 in model 3B, Table 4). The logistic regression, however, suggests a model for predicting whether a child would use a high-level change category, with an improvement in model prediction represented by Cox and Snell R^2 from .126 with just Group as predictor to .276 in Model 3A (.370 in model 3B, Table 4). Chi square change statistics represent the significance of the step.

7. Discussion

Cognitive flexibility has been studied in the context of communication disorders. The goal of our study was to investigate cognitive flexibility in 5- to 6-year-old preschoolers with DLD using the NEOD task (Karmiloff-Smith, 1990; Low et al., 2009; Spensley & Taylor, 1999a,b; Ten Eycke & Müller, 2018), which is a drawing task that has not been used with children with DLD to date. In the NEOD task, the children were first asked to draw a man. After that, they were asked to draw a nonexistent man. The same procedure was followed for the object 'house'. The children could make no changes, or make modifications which could be low-level changes (change to the size/shape of the item, change of the whole shape, deletion of parts of the item) or high-level changes (same-category insertion, change to the location/orientation of the item, cross-category changes).

Three main findings emerged from the study: (1) Children with DLD made no modifications more often than children with TD. (2) Children with DLD were less likely to make high-level changes. In particular, they made fewer inter-representational cross-category insertions and, to a lesser degree, fewer intra-representational same-category insertions. (3) This difference in high-level category changes between children with DLD and with TD disappeared when verbal STM or inhibition were added to the regression models.

7.1. Not an isolated language impairment

The weaker cognitive flexibility of children with DLD, signaled by a higher likelihood to make no changes and a lower likelihood to

make high-level changes, is in line with the findings reported by Farrant et al. (2012) who found that 5-year-old children with DLD performed lower than age-matched peers with TD on a DCCS task, which also tests cognitive flexibility. The results of our study supplement this research by showing that cognitive flexibility weaknesses in kindergarten children with DLD are not only found in a set-shifting task, but also in a productive NEOD task. Studies with older children (aged 7–12 years, and 10-11 years on average) reported equal performance on set-shifting tasks across children with DLD and TD (Im-Bolter et al., 2006; Henry et al., 2012), suggesting that age impacts cognitive flexibility differences between children with DLD and TD. The pattern that emerged from our study is that the children with DLD nearly always made age-appropriate low-level changes and hardly ever high-level changes. They were as likely as their peers to make low-level changes, but less likely to make higher level changes. It is interesting to note that task success of the DLD group (74%), that is, the percentage of children who made changes, was lower than that of children with TD who were 5-months younger (91%) presented in the original study (Karmiloff-Smith, 1990; success of the children with TD who were 5-months older in the current study was 94%). It should be further noted that the phrasing used in the current study included an explicit request to add "something funny or odd," which may have somewhat changed the type of response (Low et al., 2009), where children with TD responded with a higher rate of insertions (same- as well as cross-category insertions) than in Karmiloff-Smith's (1990) study.

That children with DLD show cognitive flexibility limitations is compatible with a growing body of research on domain-general executive function impairments in DLD (e.g., Ebert & Kohnert, 2011; Vugs et al., 2013; Pauls & Archibald, 2016; Vissers et al., 2015). In line with these studies as well as our predictions, the children with DLD performed lower than TD controls on inhibition, in addition to lower performance on verbal STM. The children with DLD in our study had moreover lower visual-motor skills and a lower basic drawing level than the children with TD, indicating weaknesses that extend to motor development (Diepeveen et al., 2018; Johnston & Weismer, 1983). These results demonstrate that although the language impairments of children with DLD are primary, these impairments are typically not isolated, and many children with DLD also have significant impairments outside the domain of language.

7.2. The role of verbal STM and inhibition

We investigated the effect of basic drawing level, nonverbal intelligence, verbal STM, and inhibition on the relation between cognitive flexibility and the presence of DLD in order to better understand the observed differences between DLD and TD. The effect of DLD on the use and likelihood of high-level category changes remained significant in the models that included basic drawing skills and nonverbal intelligence. Basic drawing level was not associated with high-level category changes, whereas nonverbal intelligence did show a significant association, confirming that high-level changes in the NEOD task are related to higher order cognition. The difference between the DLD and TD groups disappeared when verbal STM or inhibition were included in the regression model.

A similar effect of verbal STM was found by Lukács et al. (2016), but only for verbal executive function tasks (in their study, children with TD and DLD performed equally on nonverbal executive function tasks). The impact of verbal STM suggests that verbal abilities impact children's performance in a NEOD task, and specifically children's ability to make high-level category changes. According to Vygotsky (1978), thought is mediated by language. In line with this view, children use private or inner speech to solve problems (Neuman, Leibowitz, & Schwarz, 2000; Damianova, Lucas, & Sullivan, 2012; Welsh, 1987). Deák (2003) described how language enhances and enables the expression of flexible cognition, and provides the potential for innovative conceptualization. As such, verbal mediation, and by implication verbal STM, may help children to arrive at more complex solutions of the NEOD task, and in particular to come up with cross-category insertions. In line with this suggestion, the association of verbal STM with high-level changes was significant only in the TD group, while for children with DLD, verbal STM and occurrences of high-level changes were not associated ($r_s = .00$). Although this group difference in correlation level was not significant, the finding supports the role of verbal STM in the more complex solutions to the NEOD, and their lower frequency in children with DLD. The effect of verbal STM also fits within a general theory of executive functioning development, such as the integrative framework proposed by Garon and colleagues (2008; see Kapa et al., 2017, for an application to DLD). This framework holds that attention underlies executive function abilities and that developmental hierarchies could result in cascading effects. Verbal STM, like attention, is a basic and early available ability (Gathercole and Adams, 1993; Gathercole, Pickering, Ambridge, & Wearing, 2004) that comprises a foundation for later developing executive functions.

Inhibition is related to the expression of cognitive flexibility in young children (Davidson et al., 2006). When inhibition was included in the regression models that explained the use of high-level changes, group differences were no longer significant. This suggests that group differences in the level of inhibitory control may explain differences in task performance and is in line with Pauls and Archibald's (2016) suggestion that an inhibitory control deficit may disable children with DLD to sufficiently clear former tasks (in our study: schemata) from the current focus of attention. This, in turn, may prevent them from focusing on the new task or coming up with a novel and more uncommon solution. It is possible that children with DLD and their peers with TD used somewhat different mechanisms when solving the NEOD. A strong association for inhibition with the use of change categories emerged for children with DLD, whereas in children with TD, a similar correlation level was found for nonverbal intelligence, verbal STM and inhibition with the use of change categories. In particular, the association of inhibition with low-level changes was stronger in children with DLD than in their peers with TD. Children with DLD may have relied on inhibition for solving the NEOD, forming mainly deletions to object parts. Use of inhibitory control did not suffice, however, for performing high-level changes, possibly because of its overall lower level in this group. A pattern of different mechanisms used to solve the NEOD task was also found when task performance was compared between high-functioning children with ASD and peers: the former relied mainly on executive functions for solving the task, whereas children with TD used additional cognitive processes (Ten Eycke & Müller, 2018).

7.3. Limitations and future research

The study has several limitations. First, the DLD sample size is relatively small. Moreover, hearing was not screened and DLD diagnoses were verified based on a screening language test. Although this test has been used in the literature (e.g., Altman et al., 2016), other language assessments may have yielded different findings. In particular, it may be suggested that children with DLD understood the task less well due to their poorer language skills. However, the findings of the current study do not support such an interpretation, because the children with DLD who did not modify their drawings had language scores similar to their peers, and the language composite score did not correlate with NEOD performance.

Second, we did not test the children with other commonly-used measures of cognitive flexibility, such as set-shifting tasks. Although our findings are compatible with research that used the DCCS task with 5-year-old children with DLD (Farrant et al., 2012), it remains to be seen whether a NEOD task and a DCCS task measure the same underlying construct. Low correlations between different cognitive flexibility tasks suggest that the construct of cognitive flexibility is fractioned (Legare et al., 2018), and different tasks may tap into different subcomponents of cognitive flexibility.

Finally, our measure of inhibition also measured sustained attention, as we were unable to single out the no-go stimulus, and reliability estimates for our task, such as the split-half test (Green et al., 2016), could not be calculated. Sustained attention is the ability to maintain focus on a task despite the absence of task events that are intrinsically arousing (Robertson, Manly, Andrade, Baddeley, & Yiend, 1997). It is a core and crucial ability that underlies children's performance in more formal structured experimenter-demand tasks, including tasks which test cognitive flexibility (Garon, Bryson, & Smith, 2008), and is often impaired in children with DLD (Ebert & Kohnert, 2011). Consequently, between-group differences in sustained attention could also have contributed to explaining the between-group differences on the NEOD task. It should be noted that high shared variance between the digit span task measuring verbal STM and go/no-go task measuring inhibition could reflect a basic contribution of sustained attention to both tasks.

Our findings open venues for future research. Future research could aim to tease apart effects of inhibitory control and sustained attention in order to determine which ability potentially underlies a cognitive flexibility deficit of children with DLD. Other research could focus on investigating relationships between cognitive flexibility and language development. We did not observe significant correlations between children's performance on the NEOD task and the language composite scores, but relations may exist for specific aspects of language. Whether or not impairments in the different developmental domains (e.g., verbal, nonverbal cognition, motor) are related, either directly or indirectly, is an issue that requires further research.

8. Conclusions

Preschoolers with DLD are less cognitively flexible than their peers with TD. The results of a drawing task in which children were asked to draw nonexistent objects demonstrated that 5- to 6-year-old children with DLD made no changes to their initial drawings more often, and were less likely to make more complex changes than aged-matched children with TD, pointing to lower cognitive flexibility. Nevertheless, the performance of these children was within age expectations as compared to the 4-to-6 year olds studied in the original Karmiloff-Smith (1990) study. The difference between the two groups disappeared when verbal STM or inhibition were statistically controlled. These findings suggest that the lower cognitive flexibility of children with DLD is, at least in part, explained by their lower verbal STM and inhibition ability.

Author statement

EB was involved in conceptualization, and writing (original draft, review & editing); RB and NS were involved in investigation, and validation; EAJ was involved in methodology, formal analysis and validation, writing (original draft, review & editing), visualization, resources, supervision, and project management.

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