



Skill-related physical fitness versus aerobic fitness as a predictor of executive functioning in children with intellectual disabilities or borderline intellectual functioning



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ABSTRACT

Children with intellectual disabilities (ID) or borderline intellectual disabilities (BIF) often demonstrate impairments in executive functioning (EF). Studies in typically developing children show that aerobic fitness (AF) is positively related with EF. Skill-related physical fitness (SF) might, however, be a stronger predictor of EF than AF, as cognitive challenges are inherent in application of these skills. In this study, AF and SF were examined *simultaneously* in relationship with domains of EF in children with ID or BIF. Seventy-three children (age range 8–11; 51 boys) with ID (IQ range 56–79) or BIF (IQ range 71–79) were measured annually over a period of 4 years on AF (20-m endurance shuttle run test) and SF (plate tapping and 10 × 5 m run). EF was measured with the Stroop Color-Word test (inhibition), Trailmaking and Fluency test (cognitive flexibility), Self-ordered pointing task (working memory) and the Tower of London (planning). Multilevel models showed that SF was significantly associated with inhibition and both measures of cognitive flexibility, but in the same models no significant associations between AF and EF were found. In addition, age was significantly related to working memory and cognitive flexibility, favouring the older children. In children with ID or BIF, SF is of greater importance than AF in relationship with core domains of EF.

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What this paper adds?

For the first time aerobic fitness and skill-related physical fitness were examined *simultaneously* in relationship with a wide range of EF domains (inhibition, cognitive flexibility, working memory and planning) in 8–11- year old children with ID or BIF. In this longitudinal study, the children were measured annually over a period of 4 years. Multilevel models were used in order to give insight into possible developmental changes per EF measure with increasing age. Next, multilevel modelling was used to examine the unique contributions of aerobic fitness and skill-related physical fitness to EF. The results showed a unique relationship between skill-related physical fitness and inhibition and cognitive flexibility. No significant associations between aerobic fitness and EF were found. Age was significantly related to working memory and cognitive

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flexibility, favouring the older children. In children with ID or BIF skill-related physical fitness is of greater importance in relationship with domains of EF than aerobic fitness.

Skill-related physical fitness versus aerobic fitness as predictor of executive functioning in children with intellectual disabilities or borderline intellectual functioning

1. Introduction

Children with intellectual disabilities (ID) demonstrate impairments in executive functioning (EF; Hartman, Houwen, Scherder, & Visscher, 2010; Kirk, Gray, Riby, & Cornish, 2015; Sgaramella, Carrieri, & Barone, 2012), which incorporates a collection of inter-related higher-cognitive processes responsible for purposeful, goal-directed behavior (Anderson, 2002). EF is an important predictor of academic achievement (Bull, Andrews, Espy, & Wiebe, 2008; Van der Niet, Hartman, Smith, & Visscher, 2014) and social functioning (Diamond, 2013). Core executive functions are cognitive flexibility, inhibition, and working memory which can be seen as relatively lower-level executive functions, compared to more complex executive functions like planning (Miyake et al., 2000; Diamond, 2013). In general, development of EF is characterized by an accelerated development between 5 and 10 years with a continued development into adolescence (Anderson, Anderson, Northam, Jacobs, & Catroppa, 2001; Anderson, 2002). However, the developmental trajectories of the specific EF processes reflect the differences in complexity: inhibition shows the largest improvements in the preschool years, and less change later on. Working memory and cognitive flexibility improve mostly after preschool years, whereas in planning the largest improvements can be seen in late childhood and adolescence (Best, Miller, & Jones, 2009).

An interesting question is whether or not decreased levels of physical fitness are related to EF in children with intellectual disabilities. Children with ID have demonstrated lower aerobic fitness levels than typically developing children (Hartman, Smith, Westendorp, & Visscher, 2015). Studies in typically developing children have shown that aerobic fitness was positively related to aspects of EF, such as inhibition, cognitive flexibility, and planning (Buck, Hillman, & Castelli, 2008; Hillman, Buck, Themanson, Pontifex, & Castelli, 2009; Van der Niet et al., 2014). Fewer studies in individuals with cognitive impairments have been conducted. A positive relationship between physical exercise and inhibitory control has been found in individuals with Down Syndrome (Chen & Ringenbach, 2016). A possible theory explaining the link between aerobic fitness and EF is the cardiovascular fitness hypothesis, which states that aerobic exercise might induce short and long term changes in brain regions critical to learning and memory, as a result of increased cerebral blood flow (Etnier et al., 1997). In addition, aerobic exercise resulted in increased levels of neurotransmitters like brain-derived neurotrophic factor (BDNF) and other growth factors, that promote neurogenesis and synaptic plasticity (Hötting & Röder, 2013). Recently, it has been shown that aerobic fitness was positively associated with differences in regional brain function and brain structure in children (Chaddock, Pontifex, Hillman, & Kramer, 2011). Furthermore aerobic fitness was related to increased white matter integrity in children, which may result in faster neural conduction between brain regions important for cognitive control (Chaddock-Heyman et al., 2014). Although in typically developing children, positive relationships have been found between aerobic fitness and executive functions, in children with ID no clear evidence is available for a wide range of executive functions.

Besides the links that have been found between aerobic fitness and EF, skill-related physical fitness might be a predictor of EF, and an even stronger predictor than aerobic fitness. It has been hypothesized that, besides the aerobic mechanisms, learning and developmental mechanisms play an important role, as skill-related movements provide learning experiences that aid cognitive development (Sibley & Etnier, 2003). Skill-related physical fitness consists of those components of physical fitness that have a relationship with enhanced performance in sports and motor skills, and important aspects are coordination and agility (Corbin, Pangrazi, & Franks, 2000). Coordination is the ability to use the senses, such as sight and hearing, together with body parts in performing motor tasks smoothly and accurately. Agility is the ability to rapidly change the position of the entire body in space with speed and accuracy (Corbin et al., 2000). Several brain structures play a pivotal role in both skill-related movements and EF. There are important striatal pathways between the cerebellum and dorso-lateral prefrontal cortex, brain regions that are critical for complex movements as well as complex cognitive performance (e.g. EF) (Diamond, 2000; Koziol et al., 2014). Pesce (2012) argued that qualitative demands inherent in movement tasks could be important in the relationship with cognition (e.g. EF), but these aspects are largely under-investigated. Many forms of exercise include cognitively demanding physical activities such as employing competitive strategies, anticipating the behavior of teammates or opponents, and dealing with changing task demands (Best, 2010). In typically developing children a review showed the strongest relationships between complex motor skills and higher order cognitive skills (Van der Fels et al., 2015). In individuals with cognitive impairments, i.e. adolescents with Down Syndrome, a positive relationship has been found between manual dexterity and planning ability (Holzapfel et al., 2015).

In children with ID not only are levels of skill-related physical fitness lower than in typically developing children (Hartman et al., 2015), but also performance of gross motor skills (i.e. locomotor and object control skills) is impaired (Hartman et al., 2010; Vuijk, Hartman, Scherder, & Visscher, 2010; Westendorp, Houwen, Hartman, & Visscher, 2011). A positive relationship has already been found between gross motor skills and planning in children with ID (Hartman et al., 2010) and between gross motor skills and cognitive flexibility in children with Down Syndrome (Schott & Holfelder, 2015). We hypothesize that skill-related physical fitness components, resulting in smoothly and accurately performed motor tasks, are also positively related to EF in children with ID.

As children with ID have low levels of aerobic and skill-related physical fitness, as well as EF, we need to investigate whether skill-related physical fitness is related to EF in a different way compared to aerobic fitness. We need to fully

understand what the unique contribution of these factors is on the level of EF in this vulnerable population. This will also shed light on the existing scientific discussion regarding the role of cardiovascular versus the learning and developmental mechanisms in the relationship with cognition in children (Pesce, 2012; Sibley & Etnier, 2003). In the present study, it was hypothesized that the relationship between skill-related physical fitness and EF is stronger than between aerobic fitness and EF in children with ID. Accordingly, the aim of the present study was to examine the possible relationships between aerobic fitness and skill-related physical fitness with core domains of EF (inhibition, cognitive flexibility, working memory) and planning in children with ID. For the first time aerobic fitness and skill-related physical fitness were examined *simultaneously* in relationship with a wide range of EF measures in children with ID in a 4-year longitudinal study. Age and severity of ID were also considered.

2. Method

2.1. Participants

Seventy-three children (IQ $M = 70.8$, range 56–79; 51 boys) from a primary special-needs school located in the northern regions of the Netherlands participated in a longitudinal study. In the year of enrolment, the age range of the children was 8–11 years ($M = 9.26$; $SD = 1.04$). Twenty-eight children with mild intellectual functioning (ID; 20 boys and 8 girls; IQ range 56–70) and forty-five children with borderline intellectual functioning (BIF; 31 boys and 14 girls; IQ range 71–79) were identified. IQ of children with BIF varies between 70 and 85, but it has no clear diagnostic code in either the DSM-5 or IDC-10 (American Psychiatric Association, 2013; World Health Organization, 1992). The children were included if they were healthy and were not diagnosed with Attention Deficit Hyperactivity Disorder or Autism Spectrum Disorders. Information about learning lags of the children were retrieved from the Child Academic Monitoring System (CAMS), which is a record that the majority of the schools in the Netherlands keep. It provides an overview of a child's progress in academic skills. A learning lag explains the amount of material not mastered per academic domain and is an indication of failures in achievement. For example, a child with a learning lag of 0.35 on reading has not mastered 35% of the reading level he/she should normally have achieved (See for a detailed description Westendorp, Hartman, Houwen, Smith & Visscher, 2011). The average learning lags of the children with BIF were 0.39 ($SD = 0.19$) for mathematics, 0.38 ($SD = 0.29$) for spelling, and 0.01 ($SD = 0.51$) for reading. The average learning lags of the children with ID were 0.62 ($SD = 0.15$) for mathematics, 0.65 ($SD = 0.22$) for spelling and 0.37 ($SD = 0.31$) for reading.

Over a period of 4 years, the children's aerobic and skill-related physical fitness, as well as EF was measured annually. Not all children participated in all measurements, as in a specific year children enrolled in the school during the 4-year period, children were absent during the measurements, and others left school. Informed consent was obtained for all children and all procedures were approved by the institutional Ethics Committee of the Center of Human Movement Sciences, University Medical Center Groningen, University of Groningen.

2.2. Materials

2.2.1. Assessment of aerobic and skill-related physical fitness

In the present study, one item of the Eurofit test battery was used to assess aerobic fitness and two items of the Eurofit test battery were used to assess skill-related physical fitness. The reliability and validity of the Eurofit for children is adequate (Adam, Klissouras, Ravazzolo, Renson, & Tuxworth, 1988) and the test has been widely used in Europe. In males with ID, adequate reliability was obtained (ICC varied from 0.94–0.99; Mac Donncha, Watson, McSweeney, & O'Donovan, 1999). The test battery has been used in adolescents with ID (Salaun & Berthouze-Aranda, 2012) as well as in children with ID (Hartman et al., 2015). Moderate to good test-retest reliability of the items that were used in the present study was obtained in young children (5–7-year olds) (Fjørtoft, 2000).

2.2.1.1. Aerobic fitness. For measuring aerobic fitness the 20-m endurance shuttle run (20MSR) was used. The 20MSR is a health-related item of the test battery. In the 20MSR test children run back and forth between two lines 20 m apart, pacing their run to audio signals that progressively increase in difficulty. The test ends when a child fails to reach the end lines prior to the beep on two successive occasions or when a child stops because of exhaustion. The performance is expressed as the number of stages completed. The protocol started at 8.0 km/hr and the velocity increased by 0.5 km/hr for each one minute stage. From the last stage that was completed, the maximal oxygen uptake (VO_{2max} in $mL\ kg^{-1}\ min^{-1}$) was estimated by using the following formula: $[31.025 + (3.238 \times velocity) - (3.248 \times age) + (0.1536 \times age \times velocity)]$ (Léger, Mercier, Gadoury, & Lambert, 1988).

2.2.1.2. Skill-related physical fitness. For measuring skill-related physical fitness, measures for upper and lower limbs were obtained. The plate tapping task (PT) was used which measures upper limb coordination, and the 10×5 m run test (10×5 m run) was used for lower limb coordination and agility. In the PT task, the children move their preferred hand between two discs when holding their other hand on a rectangle midway between the discs. The children tap the two discs alternately until 25 cycles are completed. The performance is expressed as the time needed to complete 25 cycles. In the 10×5 m run

test children are asked to run 5 m, make a turn, and run back for 10 times, covering a distance of 50 m in total. For both tests, the performance is expressed as the time in seconds, and the best score of two trials is recorded.

2.2.2. Assessment of executive functioning

2.2.2.1. Response inhibition (Stroop Color-Word test; Stroop). The Stroop Color-Word test measures inhibition of prepotent behavior (Stroop, 1935). This refers to three interrelated processes: (i) inhibition of a prepotent response, (ii) stopping of an ongoing response and (iii) interference control (Barkley, 1997). Interference control is the inhibition of a habitual response in favor of a less familiar one. The Stroop Color-Word test consists of 3 cards: a Word Card with color words (red, green, yellow and blue) printed in black ink, a Color Card with colored rectangles (red, green, yellow and blue) and a Color-Word Card on which the names of the colors are printed in an incongruent color of ink. Each card consists of 100 stimuli. During the word reading condition (i.e., Word Card), children have to read aloud as quickly as possible the names of the colors printed in black ink. During the color naming condition (i.e., Color Card), children have to mention the colors of the rectangles as quickly as possible. Finally, during the color-word condition (i.e., Color-Word Card), children have to name the color of the ink as quickly as possible and not to read the word itself. The total time needed for each card was registered. An interference measure was calculated by subtracting the average time needed to complete the Word Card and the Color Card from the time needed to complete the Color-Word Card (Valentijn et al., 2005). The Stroop Color-Word test has been used in children with developmental disorders (Qian, Shuai, Cao, Chan, & Wang, 2010). In general, the test-retest reliability coefficients of the three separate cards are high ($r > 0.80$) (Neyens & Aldenkamp, 1997). In contrast to the other items of EF, the Stroop test was administered in the last two years of the 4-year period.

2.2.2.2. Cognitive flexibility [Trailmaking test (TMT) and Fluency]. The TMT is a measure of cognitive flexibility (Reitan, 1971). The TMT is a paper- and-pencil task which consists of two parts: (i) Part A requires the child to draw a line to connect encircled numbers (1–25) randomly arranged on a page in ascending order (1-2-3-4, etc.), and provides an estimate of attention and psychomotor speed, (ii) Part B requires participants to draw a line to connect encircled numbers (from 1 to 13) and encircled letters (from A to L) in alternating order (1-A-2-B-3-C, etc.). The score of both parts of the test consists of the total execution time (in seconds). To obtain an accurate measure of cognitive flexibility, the time to complete trail A is subtracted from the time to complete trail B (Strauss, Sherman, & Spreen, 2006). The TMT has been used and validated in children from age 7 (Anderson, 1998; Strauss et al., 2006). The test has also been used in children with developmental disorders (Qian et al., 2010).

Fluency taps cognitive flexibility (Korkman, Kirk, & Kemp, 1998). Children are instructed to generate as many words as possible within a certain category: a semantic category or a phonemic category. In the semantic category words belong to a certain category, such as food or animals. In the phonemic category words begin with a certain letter, such as a 'k' or 'm'. The total number of correct responses is the outcome measure, and duplicate items are scored as incorrect. In the present study, the total score was the total number of correct responses within 60 s on 'food', 'animals', 'k', and 'm'. Fluency tests have been used in typically developing children (Lehto, Juujärvi, Kooistra, & Pulkkinen, 2003; Van der Elst, Hurk, Wassenberg, Meijs, & Jolles, 2011) and children with intellectual disorders (Danielsson, Henry, Messer, & Rönnerberg, 2012). Test-retest reliability is adequate (0.74) (Korkman et al., 1998; Korkman, Kirk, & Kemp, 2001).

2.2.2.3. Self-ordered pointing task (SOPT; Working memory). The SOPT assesses non-verbal working memory (Petrides & Milner, 1982). The SOPT consisted of 4 series of cards with abstract designs. The series of cards contains 6, 8, 10, and 12 abstract designs respectively. For a specific series, the same set of abstract designs was used, but they were arranged differently on each card. In a specific series, there were as many different cards as there were designs. For example, in the series with 6 abstract designs, there were 6 cards with the same 6 abstract designs printed, but the position of the abstract designs differ on each card. Children were instructed to point to a different design on each card without repeating a design already pointed to and after all cards of a series the child has pointed to all the different designs of that series. The test administrator turned around the cards; children were asked only to point to a design on the card. Children were instructed to point to the designs in any order they wished, but that they were not allowed to use a standard order (i.e. point to the same location on two consecutive cards) or to point to a specific design more than once. Prior to the official test, children were given a practice trial with a series of 3 designs. The difficulty of the test and the appeal on working memory increases with the increases of the amount of cards per series, respectively 6, 8, 10, and 12. The test score was the total number of errors in the 4 series. The test has been used in typically developing children (Cragg & Nation, 2007) as well as children with developmental disorders (Karama et al., 2008; Temple, Carney, & Mullarkey, 1996). The test has been shown to be an appropriate task for children, and the reliability of the task is acceptable (Cragg & Nation, 2007).

2.2.2.4. Tower of London (TOL; planning and problem-solving). The widely used Tower of London task was used to measure planning ability (strategic decision making) and problem solving (Shallice, 1982). The task is brief and easy to administer and readily comprehended by young children (Anderson, Anderson, & Lajoie, 1996). Using a board with three pegs of varying lengths and three differently colored beads with holes (red, yellow, and blue), children have to move the beads into a depicted goal state in a minimum number of moves (as indicated by the researcher). In this way, 12 problems varying in difficulty with the goal state having to be reached in two, three, four or five moves. The TOL is rated by assigning 3, 2 or 1 point(s) per problem depending on the number of trials required to reach the goal state, with 3 reflecting one trial, 2 two trials, and

Table 1

Descriptive results for aerobic fitness, skill related physical fitness and executive functions in the year of enrolment.

	Total groupM (SD, n = 69)	Children with IDM (SD)	Children with borderline intellectual functioning (BIF) M (SD)
Aerobic fitness			
20-m endurance shuttle run (ml·kg ⁻¹ ·min ⁻¹ , n = 69)	44.36 (3.46)	43.64 (2.40)	44.75 (3.88)
Skill-related physical fitness			
Plate tapping (s, n = 71)	22.67 (4.15)	23.32 (4.35)	22.28 (4.02)
10 × 5 m run test (s, n = 71)	27.15 (3.08)	27.16 (3.60)	27.14 (2.77)
Executive functions			
Stroop Color-Word test (s, n = 59)	80.19 (43.71)	87.21 (46.16)	75.06 (41.91)
Trailmaking test (s, n = 72)	158.67 (96.57)	166.37 (104.39)	141.97 (77.00)
Fluency (number, n = 73)	33.40 (8.90)	30.78 (8.32)	34.93 (8.95)
Self-ordered pointing task (number of errors, n = 71)	9.36 (2.79)	10.04 (2.71)	8.95 (2.79)
Tower of London (points, n = 61)	26.49 (3.77)	26.04 (2.71)	26.73 (4.23)

1 three trials. The TOL total score is the sum of the scores for all 12 problems, with a maximum of 36. The test has been used before in typically developing children (Van der Niet et al., 2014) and in children with intellectual disabilities (Hartman et al., 2010). The TOL has been tested and validated for use with children from age 7 (Anderson et al., 1996).

2.3. Data analysis

Statistical analysis was conducted using SPSS for Windows, version 22.0. First, descriptives were calculated for the aerobic fitness, skill-related physical fitness, and EF measures in the year of enrolment of the children. Second, multilevel modelling (MLwin 2.23) was used in order to give insight into possible developmental changes per EF measure with increasing age. In multilevel models longitudinal data, which are not independent, can be analyzed. The number of measurements may vary per child, assuming that the missing data are at random (Snijders & Bosker, 2011). Five multilevel models were constructed with age (in categories: 8, 9, 10, 11, and 12 years), age2, and severity of ID (ID or BIF) as possible predictors. The EF measures were used as dependent variables. Both age and age2 were entered into the model in order to examine if the best model fit was a linear or quadratic curve. It was tested whether or not the EF measures changed statistically significantly from the reference age (8 years) to older ages (9, 10, 11, and 12 years) after calculation of Z-scores per age group. For inhibition, the 10 year olds served as a reference category. In case of a significant contribution of severity of ID to the model, two separate curves (one for ID and one for BIF) were obtained. Interaction effects between severity of ID and age were also considered.

In order to examine the relationships between aerobic fitness, skill-related physical fitness and EF, first a factor analysis (Principal Component Analysis with Varimax rotation) was performed with the fitness measures. The results of the factor analysis demonstrated whether or not in our study sample, the raw scores of the skill-related items (PT and 10 × 5 m run) loaded on one factor and aerobic fitness on another factor. In case both PT and 10 × 5 m run loaded high on the intended factor (rotated factor loadings above 0.300), the raw scores were transformed into Z-scores, and a composite score was calculated for further analysis. Next, five multilevel models (one per EF item) were constructed with the measures of aerobic and skill-related physical fitness, and year of measurement as possible predictors. The measures of EF were treated as dependent variables. Age, gender, and severity of ID were added as covariates. For all analyses, statistical significance was adopted when $p < 0.05$.

3. Results

3.1. Descriptives and developmental changes per EF measure

In Table 1, the descriptives of aerobic physical fitness, skill-related physical fitness, and EF of the children in the year of enrolment are shown.

In Figs. 1 and 2, the developmental changes per EF measure are presented. The models were not influenced by age2, so this variable was not included in the final models. The inhibition model showed a statistically significant decrease over time (i.e. better performance) of the 12-year-old children compared to the 10-year-old children. The model was not influenced by severity of ID. The cognitive flexibility model (as measured with the TMT) showed a statistically significant decrease over time to perform the trail in the 9, 10, 11, and 12-year-old children compared to the 8-year-old children. The model was not influenced by severity of ID. The cognitive flexibility model (as measured with the fluency test) showed a statistically significant increase with increasing age. The severity of ID significantly influenced the model: Children with BIF had higher scores than children with ID. The working memory model demonstrated a statistically significant decrease of the number of errors with increasing age. Severity of ID contributed significantly to the model: the average number of errors was lower

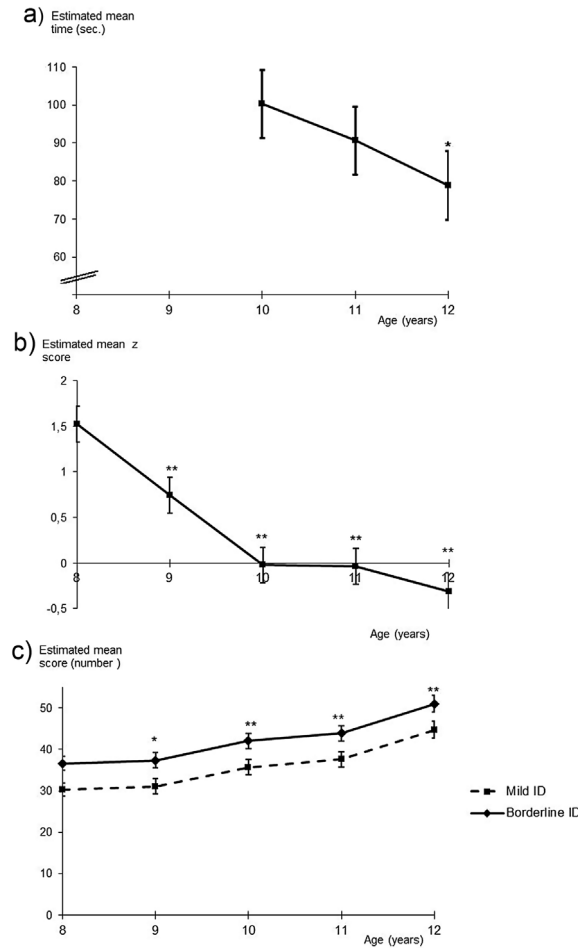


Fig. 1. a, b, c: Multilevel models for inhibition (a), cognitive flexibility (Trailmaking test, b), and cognitive flexibility (Fluency, c). Note: * = $p < 0.05$, ** = $p < 0.01$. Tested against the youngest age category.

in children with BIF compared with children with ID. The planning model showed that the 10, 11, and 12-year-old children had higher scores than the 8-year-old-children. Again, children with BIF had higher scores than the children with ID.

3.2. Aerobic fitness and skill-related physical fitness as predictors of EF

The factor analysis showed that both items of skill-related physical fitness loaded high on factor 1 (Eigenvalue = 1.550; Cumulative Explained Variance = 52%). Rotated factor loadings were 0.876 for PT and 0.884 for 10 × 5 m run. Aerobic fitness loaded high on factor 2 (Eigenvalue = 1.075; Cumulative Explained Variance = 88%) with a rotated factor loading of 0.979. The results demonstrated that it was appropriate to use a composite score of PT and 10 × 5 m run for further analysis.

In Tables 2a and 2b the results of the multilevel models are shown. In the models of inhibition and cognitive flexibility (measured with both the TMT and Fluency), skill-related physical fitness was a significant predictor (inhibition: $p = 0.02$; cognitive flexibility TMT: $p = 0.01$; cognitive flexibility Fluency: $p = 0.02$). Aerobic fitness did not contribute significantly to the outcome measures of inhibition and cognitive flexibility. The covariate gender was not significant in these models. The covariate age was not significant in the model of inhibition, but it contributed significantly to both models of cognitive flexibility. Severity of ID was a significant covariate in one of the cognitive flexibility models (Fluency).

In the models of working memory and planning, neither skill-related physical fitness nor aerobic fitness were significant predictors (see Table 2b). The covariate gender was not significant in either of these models. Age contributed significantly to the working memory model, and in the planning model a trend ($p < 0.1$) was found. Severity of ID was a significant covariate in the working memory model as well as the planning model.

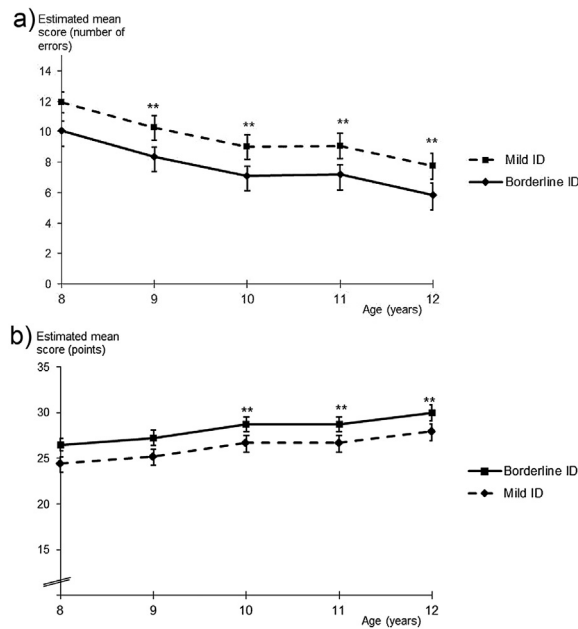


Fig. 2. a, b: Multilevel models for working memory (a) and planning (b). Note: * = $p < 0.05$, ** = $p < 0.01$. Tested against the youngest age category.

Table 2a

Multilevel models for aerobic fitness and skill-related physical fitness as predictors for inhibition (Stroop) and cognitive flexibility (Trailmaking test; TMT and Fluency).

	Beta	Inhibition (Stroop)		Cognitive flexibility (TMT) ^c			Fluency		
		SE	P	Beta	SE	P	Beta	SE	P
Fixed effects									
Intercept (constant)	85.78	64.79		3.15	1.15		22.41	11.79	
Age	-1.95	4.64	0.68	-0.33	0.08	0.003	2.31	0.86	0.03
Gender ^a	-3.85	8.32	0.66	0	0.14	1	-1.41	1.74	0.44
Severity of ID ^b	-8	8.64	0.38	-0.01	0.15	0.7	5.95	1.72	0.01
Year 2 ^c	-	-	-	0.04	0.17	0.82	-0.08	1.51	0.96
Year 3 ^c	reference			0.1	0.25	0.7	-0.55	2.32	0.81
Year 4 ^c	-18.14	5.39	0.01	0.08	0.26	0.76	-4.65	2.46	0.1
Aerobic fitness	0.65	0.77	0.43	0.01	0.02	0.63	-0.2	0.15	0.22
Skill-related physical fitness	8.66	3.09	0.02	0.19	0.05	0.01	-1.38	0.48	0.02
Random effects									
Variance between students	558.51	180.71		0.1	0.06		29.7	7.42	
Variance within students	568.14	123.93		0.55	0.07		34.47	4.28	
Deviance	1004.52			457.8			1353.52		
Deviance empty model	1187.37			632.47			1705.98		

^{a,b}Respectively 'boy' and 'ID' were the reference categories.

^cYear 1 was the reference category in the cognitive flexibility models. Year 3 was the reference category in the inhibition model.

4. Discussion

Relationships between aerobic physical fitness with EF have been studied frequently in primary school children, and the possible association between skill-related physical fitness and EF has received more attention in the past few years. For the first time aerobic fitness and skill-related physical fitness were examined *simultaneously* in relationship with EF in children with ID or BIF, which provides more insight into the unique contribution of these factors on the level of EF.

The current study showed that skill-related physical fitness was significantly associated with inhibition and both measures of cognitive flexibility in 8–11 year old children with ID or BIF, but no significant association between aerobic fitness and EF was found. The results support the learning and developmental hypothesis, which assumes that movements provide learning experiences that aid cognitive development, and the type of movement involved in physical activity might be more important than the actual physical exertion.

Table 2b

Multilevel models for aerobic fitness and skill-related physical fitness as predictors for working memory (Self-ordered pointing task; SOPT) and planning.

	Working memory (SOPT)			Planning (TOL)		
	Beta	SE	P	Beta	SE	P
Fixed effects						
Intercept (constant)	18.31	4.58		17.05	5.59	
Age	−0.71	0.34	0.02	0.87	0.44	0.08
Gender ^a	0.02	0.58	0.33	−0.28	0.77	0.73
Severity of ID ^b	−2.08	0.59	0.01	1.88	0.79	0.04
Year 2 ^c	−0.31	0.54	0.58	−0.36	0.71	0.62
Year 3 ^c	−0.22	0.91	0.81	−0.78	1.24	0.55
Year 4 ^c	−1.25	1.24	0.34	−0.71	1.3	0.6
Aerobic fitness	−0.04	0.06	0.53	0.03	0.07	0.68
Skill-related physical fitness	0.13	0.19	0.52	−0.19	0.24	0.45
Random effects						
Variance between students	2.92	0.88		5.23	1.44	
Variance within students	4.07	0.63		7.18	0.92	
Deviance	708.37			954.49		
Deviance empty model	862.08			1115.94		

^{a, b, c}Respectively 'boy', 'ID', and 'year 1' were the reference categories.

The present study extends the literature on children with intellectual disabilities, by showing that the relationships between skill-related physical fitness and EF persisted when aerobic fitness was examined simultaneously. These findings support our hypothesis that the skill-related tasks in our study were more complex than the aerobic fitness test. It is plausible that especially the selected items of skill-related physical fitness consist of those components that have a relationship with enhanced performance during more complex skills. Our results are in line with two other studies in typically developing children (5–6 year olds), showing a positive relationship between upper body coordination with inhibition (Livesey, Keen, Rouse, & White, 2006), and with a composite score of inhibition, cognitive flexibility, and working memory (Roebbers et al., 2014), but these studies did not take the effect of aerobic fitness into account. Our results contradict those of other studies showing positive relationships between aerobic fitness and cognitive control (with inhibition and cognitive flexibility as important underlying domains) in typically developing children (Chaddock et al., 2012; Hillman et al., 2009; Pontifex et al., 2011). In addition, the results conflict with a study that showed a positive relationship between physical fitness and EF (a composite score of cognitive flexibility and planning) in typically developing children (Van der Niet et al., 2014). However, it should be noted that in the study of Van der Niet et al. (2014), physical fitness was a composite score of aerobic fitness, skill-related physical fitness and strength (in total four items) which may have explained the different findings. A striking difference was that in our study sample the factor analysis resulted in two factors (one for aerobic fitness and one for skill-related fitness), whereas only one factor was obtained in typically developing children (Van der Niet et al., 2014). This could indicate that the cognitive demands inherent to the skill-related physical fitness items are more pronounced in children with ID or BIF than in typically developing children. This is also illustrated by a significant difference in skill-related physical fitness between children with ID or BIF favouring the latter group (Hartman et al., 2015). Furthermore, the children with ID or BIF scored significantly lower on physical fitness than typically developing children, and these differences persisted when the children grew older. Differences in skill-related physical fitness were even more pronounced (large Effect Size) than differences in aerobic physical fitness (medium Effect Size) (Hartman et al., 2015). Future studies in children with ID or BIF should take account of possible cognitive demands of physical fitness tasks, especially as these tasks are skill-related. In particular, the skill-related tasks could be appropriate for stimulating cognitive performance in these children, but this should be investigated in future intervention studies.

The current study showed specific relationships between skill-related physical fitness and the domains of EF: positive significant associations were demonstrated with inhibition and cognitive flexibility, but no significant associations were found with working memory and planning. In other studies, it has been shown that EF, in particular inhibition (Grandjean et al., 2012), cognitive flexibility (Lie, Specht, Marshall, & Fink, 2006), visual working memory (Postle, Druzgal, & D'Esposito, 2003) and problem solving (Rasmussen et al., 2006) have been attributed to the prefrontal cortex. During complex motor and cognitive tasks, there is a close link between the prefrontal cortex and the cerebellum (Diamond, 2000). Studies have shown that the fronto-cerebellar network is involved in cognitive tasks that require inhibition (Rubia, Smith, Taylor, & Brammer, 2007) and cognitive flexibility (Specht, Lie, Shah, & Fink, 2009). We assume that in our study, the skill-related physical fitness items required mainly inhibition and cognitive flexibility, and less working memory and problem solving. For both tasks a fast response is necessary for successful performance, which requires adequate inhibition skills and cognitive flexibility. Future studies should aim to gain insight into the role of the fronto-cerebellar network during specific EF tasks and skill-related physical fitness tasks in typically developing children and children with ID.

Age was an important factor in the tasks that measured cognitive flexibility and working memory. Both the developmental curves as well as the multilevel models in which age served as a covariate showed that the performance of the children

improved significantly with increasing age. Although for inhibition and planning, a significant improvement with age was demonstrated in the developmental curves, age effects disappeared in the multilevel models with all relevant covariates. It is remarkable that on the tasks that measured inhibition and planning, no significant improvements were observed in this study population of children with ID or BIF. In general, inhibition shows prominent improvement during the preschool years and continued improvement during childhood and adolescence (Best et al., 2009; Huizinga, Dolan, & Van der Molen, 2006). Working-memory and cognitive flexibility improve mostly after the preschool years, and planning ability seems to make the largest gains in late childhood and adolescence (Best et al., 2009). A possible explanation for the lack of age-effects in inhibition and planning might be the EF deficits that children with ID or BIF demonstrate (Hartman et al., 2010; Kirk et al., 2015). In addition, the current study population showed EF deficits as well, when comparing the scores with samples of typically developing children from previous studies (Hartman et al., 2010; Van der Niet et al., 2016). The lack of age-effects in children with ID or BIF together with lower EF performance compared to typically developing children might indicate that EF deficits increase when the children grow older. However, it is possible that larger improvements on inhibition and planning might be seen after primary school age. Further research is needed on the development of EF in children with ID or BIF, in order to determine which age range is most appropriate for stimulating the different domains of EF by training.

Strengths of the present study are the longitudinal design, the long duration of the study period (four years) and the relative large sample size of children with ID or BIF. A limitation is the possible association of additional variables with EF, i.e. socio-economical status, which has not been incorporated in the models presented here. Designing complete explanatory models for the variance in EF was outside the scope of the present study and the present models should be interpreted with this in mind. In addition, the longitudinal design did not allow us to draw firm conclusions about the causality of the relationships we found.

5. Conclusions

A unique relationship exists between skill-related physical fitness and inhibition and cognitive flexibility in 8- to 11-year old children with ID or BIF. The data suggest that in these children skill-related physical fitness is of greater importance in relationship with core domains of EF than aerobic fitness. The results support the learning and developmental hypothesis. The results are important as children with ID or BIF are a vulnerable population regarding both physical fitness as well as EF. In order to stimulate EF in children with ID or BIF, training of skill-related physical fitness should be considered as a possible effective intervention.

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