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Visuomotor skill learning in young adults with Down syndrome

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ABSTRACT

Background: Individuals with Down syndrome (DS) have impaired general motor skills compared to typically developed (TD) individuals.

Aims: To gain knowledge on how young adults with DS learn and retain new motor skills.

Methods and Procedures: A DS-group (mean age = 23.9 ± 3 years, N = 11), and an age-matched TD-group (mean age 22.8 ± 1.8, N = 14) were recruited. The participants practiced a visuomotor accuracy tracking task (VATT) in seven blocks (10.6 min). Online and offline effects of practice were assessed based on tests of motor performance at baseline immediate and 7-day retention.

Outcomes and Results: The TD-group performed better than the DS-group on all blocks (all P < 0.001). Both groups improved VATT-performance online from baseline to immediate retention, (all P < 0.001) with no difference in online effect between groups. A significant between-group difference was observed in the offline effect (Δ TD - Δ DS, P = 0.04), as the DS-group's performance at 7-day retention was equal to their performance at immediate retention (Δ DS, P > 0.05), whereas an offline decrease in performance was found in the TD-group (Δ TD, P < 0.001).

Conclusions and Implications: Visuomotor pinch force accuracy is lower for adults with DS compared to TD. However, adults with DS display significant online improvements in performance with motor practice similar to changes observed for TD. Additionally, adults with DS demonstrate offline consolidation following motor learning leading to significant retention effects.

What this paper adds

Learning new motor skills is fundamental throughout our lifespan. Persons with Down syndrome have other prerequisites for learning new tasks, related to psychological, physiological, and anatomical factors imposed by the syndrome. This study is the first to investigate online and offline learning effects of a single motor skill training session in adults with DS. Our results show generally lower motor performance in DS individuals compared to the typically developed population, but with equal online learning effects. Both groups demonstrate retention, i.e., offline stabilisation but while TD demonstrate negative offline effects, this was not the case for DS. These results should be taken into consideration when planning training of motor and general life skills for adults with DS. This work lays the ground for further investigations of the trajectory of the early learning processes and the mechanisms involved when this target group acquires new skills.

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Data Availability

Data will be made available on request.

1. Introduction

The ability to learn and retain new motor skills is fundamental for everyday functioning, it allows us to perform movements, to meet the demands of our environment, and adapt to changes in it (Schmidt & Lee, 1999). Individuals born with Down syndrome (DS), are presented with a range of physical and psychological challenges including intellectual disability (Grieco et al., 2015) and impaired general motor skills (Vicari, 2006), which have been attributed to altered anatomical, physiological, and neurological development (Dierssen, 2012). Because of this, persons with DS have a higher need for support from health care and social services throughout their lives (Tsou et al., 2020). One physical deficit observed in children and adolescents with DS is manual dexterity (Camargo Oliveira & Cavalcante Neto, 2016; Padia et al., 2022). Manual dexterity is responsible for the flexible and diverse movements of the hands requiring coordination and precision of many segments, which is used continuously in everyday life, when picking up items, writing etc. (Sobinov & Bensmaia, 2021). Unfortunately, we do not know how this deficit develops into adulthood for the population with DS, nor how training of skills within this domain works within the population. Knowledge about the characteristics of the ability to learn, consolidate and retain new fine motor skills is valuable for the population with DS and professionals working with the population. It can lead to improved planning and structure of motor skill training in the everyday life of individuals with DS, e.g., through improvements in manual dexterity (Hall et al., 2011; Jain et al., 2022).

Previous studies have demonstrated that adults with DS can improve their motor performance during practice (Kerr & Blais, 1985; Reilly et al., 2017), and intervention studies have shown that the group can improve their gross motor skills over several training sessions. The latter has been observed as improvements in gait parameters after 10 weeks of Nordic walking (Skiba et al., 2019) and as improved dribbling skills after 16 weeks of soccer practice (Perić et al., 2021). These results confirm the ability of skill learning in people with DS, but the detailed dynamics of online learning (i.e., the change in performance during acquisition of a skill) and the subsequent offline consolidation and retention processes of motor skills (i.e., the changes in skilled performance after acquisition of a skill (Dayan & Cohen, 2011)) in adults with DS, has to our knowledge not yet been investigated.

Motor skill learning is a complex process, and it is influenced by a range of factors (Dayan & Cohen, 2011), e.g., the organization of task practice (Lage et al., 2015), the type and availability of (augmented) feedback (Oppici et al., 2021) and interindividual differences such as cognitive abilities (Seidler et al., 2013). Both implicit and explicit memory processes are involved in motor skill learning (Hikosaka et al., 2002), and age and baseline skill level seem to influence to which memory system dominate the learning process (Nemeth et al., 2013). Individuals with DS display impairments in explicit memory, and a relatively preserved implicit memory (Vicari et al., 2000), and this distinct memory profile could influence how motor skills are acquired and retained in the population. Motor skill learning is influenced by individual cognitive abilities such as executive functions (Schmidt et al., 2017) and spatial working memory (Seidler et al., 2013). Part of the intellectual disability profile of individuals with DS is a deficit in these specific functions (Dierssen, 2012; Lanfranchi et al., 2009), thus the groups' performance on tests probing these abilities could help explain part of the potential differences in motor skill learning.

On the basis on this knowledge, the present paper addresses the questions: What are the dynamics of online and offline motor skill learning for individuals with DS, and are the dynamics different from typically developed (TD) individuals? Furthermore, can the potential differences in the dynamics of motor skill learning be related to differences in cognitive abilities? To address these questions, a visuomotor accuracy tracking task (VATT), was used. The task involves a subdomain of manual dexterity: Control of dynamic pinch force in a precision grip, to track visual targets on a computer monitor. We evaluated the within-session improvement in motor performance (online effects) as well as the performance seven days after acquisition in a delayed retention test (offline effects). The inclusion of a delayed retention test makes it possible to distinguish online performance improvement and overall learning after an offline consolidation period (Kantak & Winstein, 2012). We hypothesized that the participants with DS would show improved motor performance during acquisition and display retention of the motor task seven days later. We expected the TD-group to perform better than the DS-group on the baseline assessment, and that the TD-group would display larger online improvements compared to the DS-group, possibly due to the use of explicit processes dominating early skill acquisition.

The VATT of this study was chosen as it requires control over a dynamic pinch-force output, a part of our manual dexterity, relevant for everyday life. The task was performed in 4 separate phases: Baseline, acquisition, immediate retention, and 7-day retention. It involved five different targets appearing on the screen in a repeated sequence. During acquisition, the sequence was colour-coded, i.e., a specific target colour always appeared in the same position on the screen. To investigate to what extent improved motor performance could be related to explicit sequence learning versus improved motor acuity, we implemented two different blocks at the baseline and retention tests (Shmuelof et al., 2014). One block with the same sequenced target order as in the acquisition phase, and the other with a random target order. Both blocks were non-colour coded. The presence of sequence learning was investigated by comparing the performance on the final, sequenced acquisition block (with colours), to the performance on the sequenced retention block (without colours), and by comparing performance on the sequenced and random retention blocks. We expected to observe sequence learning in the TD-group, due to employment of an explicit learning strategy, while we expected this to be absent in the DS-group. To investigate if potential differences in online and offline effects between groups could be related to specific motor or cognitive abilities, we administered a battery of cognitive tests and assessment of fine motor skills.

2. Methods

2.1. Participants

Twelve adults with Down syndrome (DS-group) and 14 age-matched typically developed adults (TD-group), were recruited. If a participant had been part of a similar research project previously and tried a task of a similar design, they were excluded from this study. One participant in the DS-group was excluded, since the person were not able to complete the training. Participants' characteristics is shown in Table 1. The study was approved by [NATIONAL ETICHS COMITEE] ([REGISTRATION NUMBER]) and adhered to the Helsinki Declaration II. All participants gave their informed consent before participation and only participants without guardianship were included.

2.2. Experimental design

Each participant visited the lab twice over the course of one week. On day 0, the participants completed the baseline, motor acquisition, and immediate retention blocks. Exactly one week after their first visit, the participants completed 7-day retention tests and additional motor and cognitive tests.

2.3. Visuomotor accuracy tracking task

A computer-based dynamic visuomotor accuracy tracking task (VATT) performed with the dominant hand was applied. The task has similarities to tasks used in previously published works from our lab (Beck et al., 2020; Christiansen et al., 2018). The participants were seated approximately 50 cm from a screen at an adjustable table, which was set to ensure a comfortable position of the dominant arm. The tables' height was recorded, to ensure replication on the second visit. The task required the participants to control a cursor up or down to track a series of rectangular target boxes on the monitor, by accurately applying force in a pinch grip to a spring-loaded lever through a scissor handle, i.e., pinching two levers between the index finger and thumb. When force was applied to the levers, the dot moved upwards, when force was released, the dot moved downwards (Fig. 1, see also (Beck, 2021)). This was accomplished as the force was relayed to a load-cell (UU2-K10, Dacell, South Korea). Then the force was low-pass filtered (10 Hz), amplified (x100) (AM-310, Dacell, South Korea), and then fed to the PC through a USB-connected data board, sampling at 90 Hz (NI USB-6008, National Instruments, Austin, Texas). An in-house developed Python application ran the VATT. Continuous, online feedback was presented to the participants, as the cursor changed colour from red to blue, when the targets were hit correctly. The targets were presented for 2 s, with 0.2 s between targets. The participants were given augmented feedback, as their time on target in percent were displayed for 2 s after each block.

Performance was assessed in two blocks of 59.2 s (27 targets) at baseline, immediately after, and seven days after the training session. One of the blocks had a random target order and the other block had five targets appearing in a repeated sequence. At baseline, the random block was performed before the sequenced block. This order was reversed at both the immediate and 7-day retention tests, i.e., the sequenced block preceded the random block. The participants practiced the sequenced task in 7 blocks of 92.2 s each (42 targets), with a 1-minute break between blocks. At baseline and retention tests, all targets were displayed in red (non-colour coded). During acquisition, the five-target sequence had different colours (augmented information on the sequence), i.e., a target of a given colour always appeared in the same position on the screen (colour-coded sequence). Before the first baseline block, the participants completed a sequenced familiarization block with red targets (33 s, 15 targets). The participants were not informed about the differences in block structure. Between the baseline and practice blocks, the participants were seated in rest for 20 min. Between practice and immediate retention, the participants had a 1-minute break.

2.4. Basic executive functions

A computerized modified Eriksen flanker task was used to measure basic executive functions (inhibitory control, processing speed, working memory and flexibility, (Eriksen & Eriksen, 1974)). The flanker task applied in this study was originally developed by Adele Diamonds' research group (Schonert-Reichl et al., 2015). In this task, the participants were seated in front of a laptop with an external

Table 1
Demographic data of the participants.

Variables	DS	TD
Number of participants	11	14
Sex (f/m)	3/8	6/8
Age (years \pm SD)	23.9 \pm 3.0	22.8 \pm 1.8
Living status (F/I/M)	3/6/2	-
Type of DS (ND/TL/M/UK)	7/1/0/4	-

The distribution on age and sex in both the DS- and TD-group. In the DS-group we obtained data on living status, where F = living with Family, I = Independent living, and M = A mix of independent living, and living with family. Type of DS were determined through interviewing the families. ND = Non-disjunction, TL= Translocation, M = Mosaic, UK = Unknown.

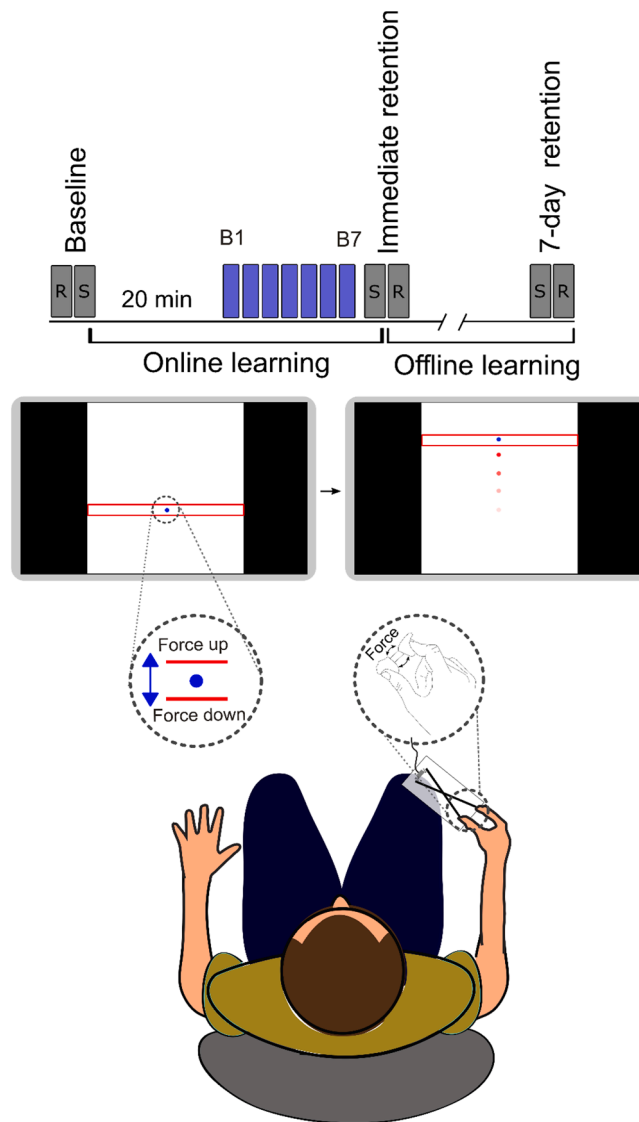


Fig. 1. Outline of the VATT and study protocol of the current study. The underarms are resting on an adjustable table, to ensure a comfortable position
Adapted from (Beck, 2021).

keyboard. Each trial consisted of a row of five fish turning left, right, or down (Fig. 2). The participants were instructed to ‘feed the correct fish as quickly as possible’, determined by which way the fish was facing. If the target fish were facing left, then the correct answer would be pressing the left control key with the left index finger. If the target fish were facing right the correct answer was pressing the numpad enter key with the right index finger. The buttons were marked with arrows showing the directions (left ctrl = ←, numpad enter = →). After a response, a new row of fish appeared. The task contained four trial types, determined by the distractor fish that were either facing the same direction (congruent), the opposite direction (incongruent), upwards (neutral), or without distractors. The four fish on the sides always turned in the same direction. The task consisted of three sub-tasks: A regular, a reversed, and a mixed flanker task. In the regular task the fish were blue, and the participants were instructed to feed the middle fish (target fish). In the reversed task the fish were pink, and the participants were told to feed the fish on the sides. The mixed task consisted of both blue and pink fish, and the participants were instructed to feed either the middle or the outside fish, depending on the colour of the fish. Before each sub-task, the participants completed practice rounds. In the regular and reversed tasks, the practice round consisted of 4 stimuli. Another practice round was required if 2 or less trials were correct. The mixed practice round consisted of 8 stimuli, and another practice round were performed if a participant got 5 or less correct answers. A maximum of three practice rounds were allowed for each sub-task. The test was terminated if a participant failed the third practice round before any the sub-tasks. The regular and reversed tasks consisted of 17 stimuli each (3 congruent, 10 incongruent, 2 neutral and 2 without distractors), and the mixed task consisted of 45 trials (12 congruent, 16 incongruent, 9 neutral and 8 without distractors). The average reaction times (RT) on all trials and the RTs on

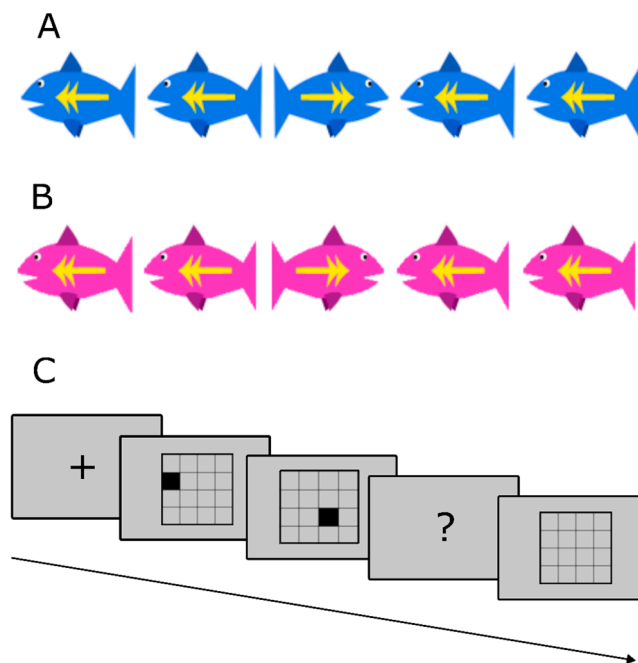


Fig. 2. Overview of the Flanker and Corsi block-tapping tasks. A: An example of a incongruent trial from the regular flanker task, where the distractors on the sides are pointing opposite of the middle fish. The correct response is to push the “right” button B: An example of a incongruent trial from the reversed flanker task, where the distractor in the middle is pointing in the opposite direction of the outside fish. The correct answer to this trial would be “left”. C: An overview of a 2-span trial from the Corsi block-tapping task. After a fixation cross, the grid appears, and one of the 16 squares changes colour to black followed by a second square. After the question mark, the empty grid appears, and the task is to repeat the sequence.

the congruent and incongruent trials were extracted as outcomes, as well as the accuracy rates on the congruent and incongruent trials (correct answers/total number of trials). The flanker effects were calculated as: incongruent – congruent, both in the RT and accuracy measures.

Flanker tasks have been applied to populations of individuals with DS previously (Ringenbach et al., 2021; Traverso et al., 2018). No validity or reliability studies have been conducted in the population with DS, but The Erisken flanker task have been found to an applicable measure of executive function in older adults with mild cognitive impairment (Guarino et al., 2020).

2.5. Visuospatial working memory

We applied a computerized forward span Corsi block-tapping task, developed by Aeschlimann and co-workers (Aeschlimann et al., 2017). The participants were placed in front of a laptop with a touchscreen with 16 white squares in a 4 by 4 grid. Following a fixation cross, one square at a time changed colours to black in a quasi-randomized pattern, followed by a question mark. Then the participants were instructed to tap the same boxes on the screen in the same order, with their index finger (Fig. 2). After an introduction and three practice rounds (two trials with 2-span tasks and one with a 3-span task), the task commenced at 2 spans. Four correct answers out of six trials were required to move on to the next level. If this was not accomplished, the task was terminated. The Corsi block-tapping task is the most used visuospatial task used in DS research and the test seem to be consistent with general ability levels (Yang et al., 2014), but the test has not been formally investigated in validity or reliability studies on the population.

2.6. Test of fine motor skills

The Purdue Pegboard test (Lafayette Instrument Company, Lafayette, Indiana, USA) were performed to measure the participants' manual dexterity (Tiffin & Asher, 1948). The test consisted of four sub-tasks: 1) The participants placed as many pins in the board as possible in 30 s with their dominant hand. 2) Similar to 1), but with the non-dominant hand. 3) The participants worked bimanually and had to place as many pairs of pins as possible in 30 s 4) The assembly task, where the subjects were instructed to build as many assemblies as possible using three different parts in 60 s. An assembly consisted of four parts, assembled in the following order: A pin, then a washer is placed over the pin, a collar, and finally a washer. The parts are placed with alternating hands, starting with the dominant hand placing the pin. Before each task, the experimenter instructed and demonstrated the task, and the participants practiced the task. Outcomes were the Sum of Scores (the sum of correctly placed pins in sub-task 1), 2), and 3)) and the Assembly score (the number of correctly placed parts in subtask 4)). The Purdue Pegboard test have been found to have a test-retest reliability of 0.86 and 0.92 (Chen & Ringenbach, 2015), and a predictive validity of 0.7 in adults with intellectual disability (Neeman, 1986).

2.7. Data analysis and statistics

All analyses were performed in R statistics (R Statistics 4.0.0, 2020). A linear mixed effects model was fitted to the data ("lme4" R-package (Bates et al., 2015)), to investigate the effect of time (i.e., the performance on the 13 blocks), group and type of block (sequence or random) on motor performance. Time on target in seconds, were translated to a score between 0 and 100 points, with each point being equal to 0.02 s. Group, time, and block type were included as fixed effects, together with sex and Corsi block-tapping span performance (the level at which the participants failed the test). Sex and Corsi block-tapping task performance were included to control for the variation these factors could introduce to the data. Since the participants were likely to display differences in baseline performance and our experiment is a repeated measures design, random intercepts were fitted for each participant.

(1): Time on target \sim Group \times Block \times Type + Sex + Corsi lock span number + (1|participant).

The equation above is presented in R-terminology where "(1|participant)" represents the individual intercepts and the \times 'es indicates the interactions between the fixed effects of Group, Block and Type. If significant effects of the interactions between Time on target and Group and Block (time), were observed, post hoc analyses of within- and between-group differences were performed. The post hoc analyses investigated differences in online and offline learning, and differences in performance between the random and sequence blocks, to investigate sequence learning. P-values from the post-hoc analyses were corrected for multiple comparisons with the single step method. Paired T-tests were used to investigate differences between the groups on the motor and cognitive tests. Unpaired T-tests were used to determine the presence of flanker effects within the groups. Alpha level was set at $P < 0.05$, for all analyses. Results from the linear mixed effects model are presented as model estimated means \pm standard error (SE). The results of the motor and cognitive test are presented as mean \pm SE. Correlation matrices in each group were computed for the motor and cognitive test and VATT performance on the sequenced task at baseline and online and offline effects, and for the absolute motor performance level on day 0 and offline effects.

3. Results

One participant from each of the groups did not complete the 7-day retention tests and the motor and cognitive tests, the TD-participant due to a hand injury, and the DS-participant were not able to come to the lab on the day of the retention tests. Two

Table 2
Outcomes of the motor and cognitive tests.

Variables	DS	TD	Sig. level
Flanker test			
Regular RT all (ms)	1400 \pm 160	464 \pm 17	***
Regular congruent RT (ms)	1376 \pm 176	469 \pm 22	***
Regular incongruent RT (ms)	1406 \pm 160	454 \pm 16	***
Regular congruent acc.	0.83 \pm 0.07	1 \pm 0	
Regular incongruent acc.	0.45 \pm 0.11	0.96 \pm 0	**
Regular flanker effect RT (ms)	26 \pm 123	-16 \pm 13	
Regular flanker effect acc.	-0.38 \pm 0.11	0.04 \pm 0	*
Reversed RT all (ms)	1299 \pm 123	439 \pm 132	***
Reversed congruent RT (ms)	1101 \pm 1587	471 \pm 26	**
Reversed incongruent RT (ms)	1338 \pm 126	446 \pm 19	***
Reversed congruent acc.	0.93 \pm 0.06	1 \pm 0	
Reversed incongruent acc.	0.82 \pm 0.04	0.98 \pm 0	**
Reversed, flanker effect RT (ms)	290 \pm 115	-25 \pm 9	*
Reversed, flanker effect, acc.	-0.11 \pm 0.06	-0.02 \pm 0	
Mixed, RT all (ms)	1085 \pm 77	730 \pm 35	**
Mixed congruent RT (ms)	934 \pm 577	683 \pm 35	**
Mixed incongruent RT (ms)	1198 \pm 125	799 \pm 38	*
Mixed, congruent acc.	0.92 \pm 0.05	0.97 \pm 0	
Mixed, incongruent acc.	0.47 \pm 0.06	0.92 \pm 0	***
Mixed flanker effect, RT (ms)	268 \pm 104	117 \pm 27	
Mixed flanker effect, acc.	-0.45 \pm 0.04	-0.05 \pm 0	***
Corsi block-tapping task			
Span number	2.8 \pm 0.4	6.4 \pm 0.4	***
Correct answers	6.9 \pm 1.8	26.5 \pm 1.7	***
Pegboard			
Sum of scores	18.5 \pm 1.7	39.2 \pm 1.0	***
Assembly	13.1 \pm 1.3	43.7 \pm 1.4	***

Sum of scores = The sum of pins placed with the right, left and both hands in three separate trails. Assembly score = The number of correctly placed parts in the assembly task. RT = Reaction time. Acc. = Accuracy ratio, calculated as number of correctly answered trials divided by the number of total trials. Flanker effect RT = Reaction time in the congruent trials subtracted from the reaction time in incongruent trials. Flanker effect acc. = Accuracy ratio in the congruent trials subtracted from the accuracy ratio in the incongruent trials. Corsi Span number = The number of spans at which the participants failed the task. Correct answers = The number of correctly performed trials. Data is presented as means \pm SE. Stars indicate the P-values of the paired T-tests between the groups. Significance codes: "*" denotes $p < 0.05$, "**" denotes $p < 0.01$, "***" denotes $p < 0.001$.

participants in the DS-group did not complete the sequence baseline block.

3.1. Fine motor skills and cognitive test performance

Paired T-tests showed that the TD-group performed better on the pegboard task and the Corsi block-tapping task compared to the DS group (all P-values < 0.001, Table 2). The results from the flanker tasks showed that the TD-group had lower reaction times (RT) on the three sub-tasks, both in the average RT for all trials and on the congruent and incongruent trials (Table 2): Regular flanker (all P-values < 0.001), reversed flanker (all P-values < 0.01) and mixed flanker (all P-values < 0.05). The TD-group was more accurate on the incongruent trials on all three tasks compared to the DS group (all P-values < 0.01), but no differences were observed on the congruent trials. No flanker effects were observed for RT in either of the groups on the regular flanker task (DS-group: $P = 0.923$, TD-group: $P = 0.192$). The DS-group displayed a flanker effect in accuracy on the regular flanker task ($P = 0.006$), while the TD-group did not ($P = 0.104$), this was seen as a significant difference between the groups in the paired T-test ($P < 0.05$). On the reversed flanker task, the DS-group presented a flanker effect on RT ($P = 0.025$), while the TD-group did not ($P = 0.083$). The paired t-test revealed a significant difference in the flanker effect on RT between the groups ($P = 0.019$). With regards to accuracy, both groups displayed flanker effects (DS-group: $P = 0.045$, TD-group: $P = 0.040$). On the mixed task, both groups presented flanker effects on RT (DS-group: $P = 0.016$, TD-group: $P = 0.001$), and accuracy (DS-group: $P < 0.001$, TD-group: $P = 0.012$). The paired T-tests between the groups showed no difference between the groups on the flanker effect on RT ($P = 0.105$), and a significant difference between the groups on the flanker effect on accuracy ($P < 0.001$).

3.2. Motor performance on the VATT

Main effects of both Block ($F = 29.6$, $P < 0.001$) and Group were observed ($F = 30.1$, $P < 0.001$), so post hoc analyses were performed. The post hoc analyses revealed that the TD-group performed ~ 30 points better than the DS group at all timepoints (all $P < 0.001$), equivalent of 0.6 s time on target on each target (Fig. 3). A significant general effect of sex was found ($F = 4.6$, $P = 0.04$) on VATT performance in favour of males in the study population, while no significant effect of Corsi block-tapping performance were observed ($P = 0.88$).

3.3. Online effects

Changes in visuomotor performance are presented as changes in time on target score. The learning curves for both groups are depicted in Fig. 3. The improvements in motor performance within session (online) and between sessions (offline) are shown on Fig. 4. Both the groups improved their online motor performance on the sequence task from baseline to immediate retention, (immediate retention – baseline, Δ_{online}) ($\Delta_{\text{online DS}} = 8.4 \pm 1.7$, $P < 0.001$; $\Delta_{\text{online TD}} = 11.9 \pm 1.5$, $P < 0.001$, Fig. 4 A and B). No difference in online learning was observed between the groups ($\Delta_{\text{online TD}} - \Delta_{\text{online DS}}$).

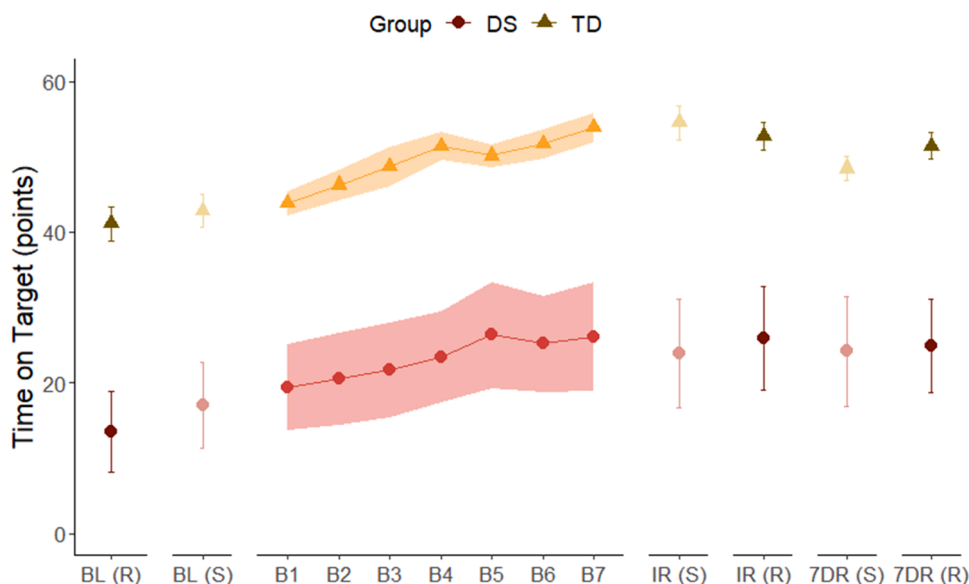
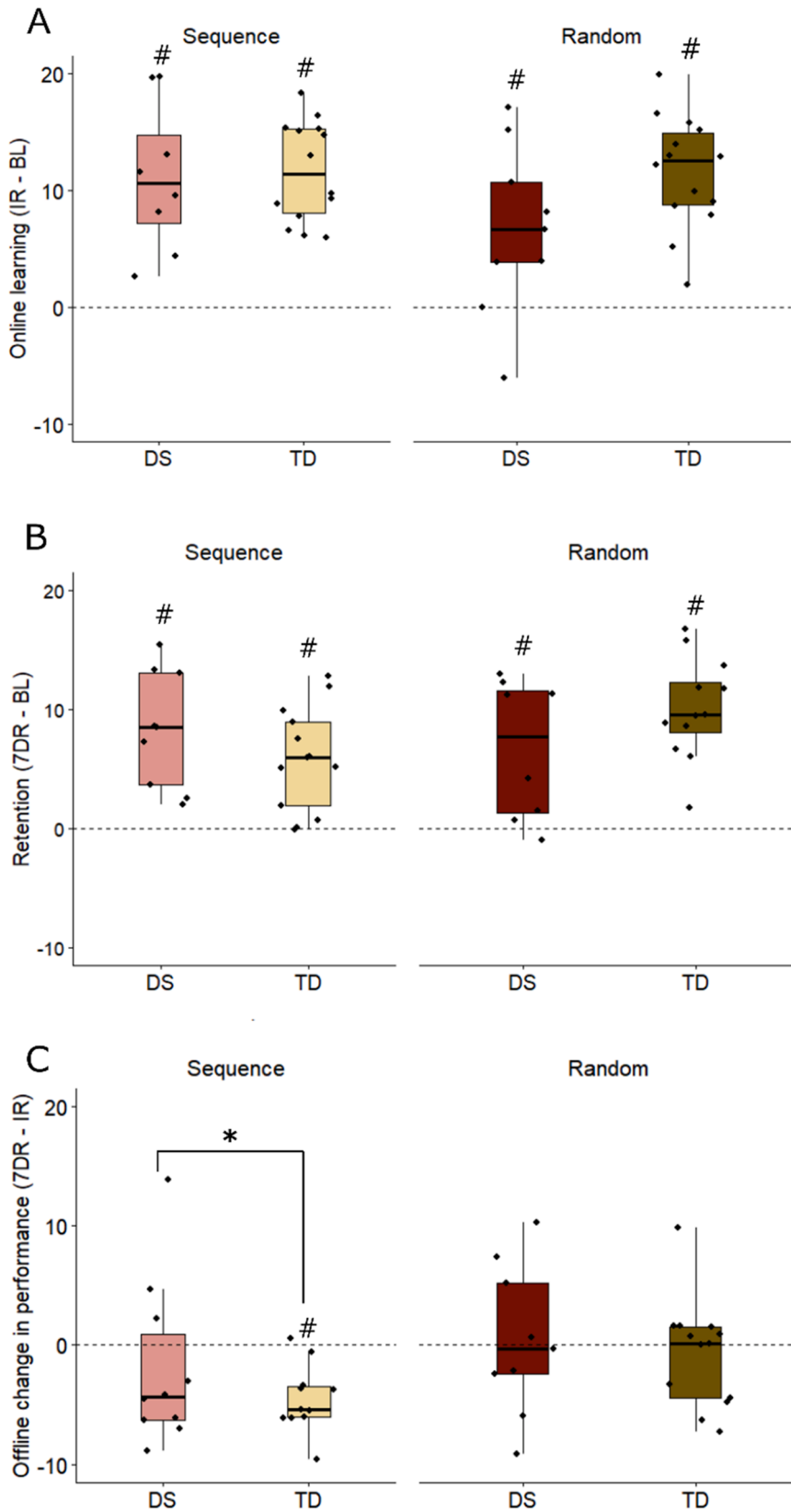


Fig. 3. Motor Performance at baseline, during acquisition and at retention tests. Motor performance as percentage points (pp) time on target on all blocks. Black and grey colours indicate random and sequence blocks at baseline, immediate and 7-day retention, respectively. Coloured blocks indicate acquisition blocks. Error bars and ribbons represent error as confidence intervals. Abbreviations: BL = Baseline; IR = Immediate retention; 7DR = 7-day retention, (S) = Sequence; (R) = Random.



(caption on next page)

Fig. 4. Online and offline effects. Box plots with individual values displaying changes in performance on both the sequenced and random tasks. (A) online performance change from baseline to immediate retention, (B) retention of VATT performance, measured as performance changes from baseline to 7-day retention, and (C) offline change in performance from immediate to 7-day retention. The dotted lines at 0 on the y-axes is equal to the groups' motor performance on the baseline blocks (A and B) and immediate retention blocks (C). BL = Baseline, IR = Immediate retention, 7DR= 7-day retention. # = Significant within-group difference. * = Significant between-group difference in the relative changes.

3.4. Retention and offline effects

Both groups showed retention of their performance on the VATT, as their performances on the 7-day retention tests were significantly better than their baseline performances on both the sequence (DS: 7.9 ± 1.7 , $P < 0.001$, TD: 5.8 ± 1.5 , $P < 0.001$, Fig. 4B) and random tasks (DS: 11.3 ± 6.7 , $P < 0.001$, TD: 10.9 ± 1.5 , $P < 0.001$, Fig. 4B). No difference between the groups was observed in retention (7-day retention – baseline). No difference in motor performance was observed in the DS-group from immediate to 7-day retention on the sequence task (7-day retention – immediate retention, Δ offline) (Δ offline DS: -0.5 ± 1.7 , $P = 0.995$, Fig. 4C). The TD-group had a significant offline decrease in performance on the sequence task (Δ offline TD: -6.1 ± 1.5 , $P < 0.001$, Fig. 4C). A between-group difference was observed in the offline effects in performance on the sequence task from immediate to 7-day retention (Δ offline TD – Δ offline DS: -5.6 ± 2.2 $P = 0.04$, Fig. 4C).

3.5. Implicit and explicit learning

The presence of explicit sequence learning vs. implicit learning of the task, was investigated within groups by analysing performance differences between the sequence and colour-coded practice block 7, and both the sequenced non-colour-coded and the random immediate retention blocks. This was also done between the retention blocks at day 7. No significant differences in performance within or between groups were observed in these analyses.

3.6. Correlations between online performance and offline effects

The correlation matrices between absolute motor performance on the immediate retention blocks and the offline performance changes from immediate to 7-day retention, showed significant negative correlations within the TD-group (Offline effects and immediate retention performance: Sequence, $R = -0.79$, $P = 0.001$, Random, $R = -0.69$, $P = 0.009$). These correlations were not observed in the DS-group.

3.7. Correlations between motor performance and motor and cognitive tests

The correlation matrices between motor performance on the VATT and the motor and cognitive tests in the two groups, showed a significant negative correlation between the number of correctly answered trials on the Corsi block-tapping task and offline effects on the random task in the TD-group ($R = -0.58$, $P = 0.04$). No other correlations between either baseline nor online and offline changes and the results of the motor and cognitive test were observed within the groups.

4. Discussion

This study is the first to investigate motor skill learning on a task that requires continuous dynamic control of pinch force in adults with DS, and the first to investigate both online and offline effects of motor practice.

4.1. Online effects and sequence learning

The participants in both the DS- and TD-group demonstrated positive online effects of motor practice i.e., improved their performance from baseline to immediate retention tests. This finding is in line with previous studies, which have shown that individuals with DS can improve their motor performance during practice (de Mello Monteiro et al., 2017; Kerr & Blais, 1985, 1987). Kerr and Blais used a pursuit tracking task performed with a steering wheel, which required the participants to accurately move the wheel to a given target location to investigate probability learning in youths with DS (Kerr & Blais, 1985, 1987). The pursuit tracking task somewhat resembles the visuomotor tracking element of the VATT in the present study. The researchers observed online performance improvements as a ~23% decrease in total trial time on 8 blocks with 800 trials (from ~2300 ms to ~2080 ms) (Kerr & Blais, 1985), while the present study shows a 40% increase in time on target on 7 blocks with 294 trials (from 17.03 to 23.88, Fig. 3). Direct comparisons in the magnitude of the improvements should be made with care, as the tasks are innately different. The aim of the pursuit tracking task was to investigate probability learning in persons with DS, not how the group acquired the motor skill of steering the wheel in a precise manner, while the aim with the VATT in the present study was to assess changes in the ability to control pinch force.

Despite differences in baseline motor performance, the DS- and TD-group showed equal improvements in online motor performance, demonstrating similar rates of learning in the two groups. No previous study has reported similar findings. In some previous studies, the age-matched control groups did not improve their performance on the tasks; they performed at maximum level at the baseline test leaving no further room for improvement i.e., a ceiling effect (de Mello Monteiro et al., 2017; Kerr & Blais, 1985, 1987).

This discrepancy in online effects between the present and earlier studies could be explained by differences in task designs, including difficulty levels. For instance, the study by Monteiro and colleagues applied a prediction reaction time model, which seem to leave very little room for improvements in the TD-group. The researchers also divided the DS-group into a high and low performing group based on their baseline scores. They observed that only the low performing group improved their scores. The requirements for precise pinch force control in the visuomotor tracking task of the present study, was chosen deliberately, as the aim was to investigate potential differences in motor skill learning between the groups, and that requires a task which challenges participants in both groups.

In addition to motor acuity, the task of the present study involves a prediction and reaction element as well; the faster the participants move the cursor to a target when it appears, the higher a score they can obtain. Thus, it could be speculated that part of the differences between the groups could be attributed to differences in RT and potentially to prediction on the sequence-based task. To our knowledge no previous study has applied such a model in individuals with DS. The flanker task used in the present study includes an RT-element. Here, the results showed significantly higher RTs on all three tasks in the DS-group compared to the TD-group, ranging from ~950 ms on the regular task to ~300 ms on the mixed task, which is in line with previous findings (Davis et al., 1991). This difference in reaction time could partly explain the difference in VATT-performance of the DS-group and the TD-group, however, further experiments and analyses are necessary to conclude on the role of differences in motor reaction time in the VATT.

The protocol of the present study was chosen based on pilot experiments, which indicated that protocols similar to previous studies (e.g., 6 blocks of 3 min) were exhausting for the participants with DS to complete (Beck et al., 2020; Thomas et al., 2016). We expected to observe sequence learning after exposure to the colored sequence during the seven practice blocks in the TD-group, as this has been observed previously (Krakauer et al., 2019; Shea et al., 2006). However, no differences in performance between Block 7 (sequenced and colour-enhanced) and the sequence block at immediate retention (sequenced, un-coloured), nor between the sequence and random block at immediate retention were observed in the TD-group. We did not observe sequence learning within the DS-group as expected. It could be the reduced amount of total practice time and less exposure to the sequence that is the cause of the absence of sequence learning in the TD-group. If the participants had been told to be aware of a sequence in the task, it is possible that sequence learning would have been observed.

The results for online effects demonstrate that individuals with DS improve performance with motor practice to the same extent as TD individuals. Since initial skill acquisition is influenced by involvement of cognitive processes and explicit aspects of learning (Krakauer et al., 2019) we have expected higher performance gains in the TD-group since this group is characterized by higher performance on the cognitive tests. This was however not the case. While the similar online effects between the two groups may be influenced by the higher baseline performance in the TD-group, it can nevertheless be concluded that DS-group demonstrate skill acquisition.

4.2. Offline effects, consolidation, and retention

To investigate offline effects and retention on motor learning, we employed delayed retention tests seven days after acquisition of VATT-performance. The present study is, to the authors knowledge, the first to apply such a design in adults with DS. Earlier studies have demonstrated that persons with DS improve both their gross (Almeida et al., 1994; Perić et al., 2021; Skiba et al., 2019) and fine motor performance (Latash et al., 2002) with several practice sessions practice. The previous studies did not include performance measures between sessions; thus, it is not possible to gauge the offline effects or retention of the motor practice between single sessions. The results of the present study show that adults with DS exhibit retention of VATT-performance, as significant increases in motor performance at 7-day retention compared to the baseline performance. The DS-group exhibited the same level of retention as the TD-group, indicating similar overall learning of the two groups (Kantak & Winstein, 2012).

A difference between the groups in the offline changes in motor performance on the sequence task was observed (Fig. 4B). The DS-group maintained their performance from immediate to 7-day retention, while performance decreased in the TD-group. This difference between the groups could be explained by the difference in absolute performance, which was significant on all blocks throughout the study (~0.6 s time on target per target). Indeed, we observed negative correlations between absolute motor performance on both the retention blocks on day 0 and the offline change in motor performance on the sequence task, indicating that a high absolute performance on day 0, correlated with a larger decrease in performance from immediate to 7-day retention. The TD-group might have reached a performance close to the ceiling of the task, thus making it less likely for the group to achieve the same level of performance seven days after the acquisition. An indication of this, is the negative correlations observed between performance on the immediate retention tasks and the offline effects in the TD-group. Reis and colleagues observed either a performance maintenance or decrease on a continuous pinch task, suggesting that this offline pattern might be the default on continuous pinch tasks, contrary to serial tapping tasks, where offline increases in performance have been observed (Reis et al., 2009; Siengsukon & Al-Sharman, 2011).

Additionally, the functional task difficulty, i.e., the difference in task difficulty imposed by differences in skill level of the participants is important to consider (Guadagnoli & Lee, 2004). We observe a significant difference in absolute skill level throughout the experiment, with the TD-group performing at a higher level. The difficulty level of a task during acquisition is related to performance level at retention in an inverted U-shaped manner, where both a too easy and too hard task is detrimental to retention of the task (Akizuki & Ohashi, 2015). It could be speculated that the task was closer an optimal challenge point for the DS-group, while it was too easy for the TD-group, and thus the negative offline effect was observed. To address the issue of functional difficulty level, future studies should include tasks, that are adjusted to ensure the same absolute performance level at baseline, and a control group that is matched to the same absolute online performance level as the DS-group. Motivation for task performance at the 7-day retention test could also have affected the results. If the participants in the TD-group were less motivated for performing the task, this could influence task performance negatively e.g., through lack of attention (Wulf & Lewthwaite, 2016). We can however only speculate on the

potential influence of motivation since no measures of motivation for task performance were obtained.

The differences in offline effects between the groups could point to differences in how the explicit and implicit memory systems are engaged during online motor learning (Robertson et al., 2004). Previous research has shown that individuals with DS have deficits in explicit memory and implicit memory abilities compared to typically developed peers (Vicari et al., 2000). It could be speculated that the DS-group employs a predominantly implicit learning strategy during skill acquisition, and that could be part of the reason for the maintained motor performance observed in the DS-group. The TD-groups' decrease in visuomotor performance at 7-day retention could indicate the engagement of explicit, cognitive processes that is prone to interference due to the presentation of competing knowledge during the offline period (Fletcher et al., 2005). However, we did not find indications of explicit sequence learning on day 0, indicating loss of explicit knowledge of the sequence is not the sole cause for the performance decrease in the TD-group.

4.3. Variability in performance and motor and cognitive tests

Several factors could contribute to the difference in general motor performance level between the groups in the present study. Recent studies have demonstrated increased accuracy and lower variability on the pinch force task with increased age during neurotypical development (Beck et al., 2021a, 2021b). Studies investigating the ability to modulate or hold a specific force output have shown greater variability and a generally lower force output in individuals with DS (Heffernan et al., 2009; Rao et al., 2017). In the present study, we observe greater interindividual variability in the DS-group, than in the TD-group, evident as larger confidence intervals in Time on Target score (Fig. 3). In addition, children and adolescents with DS have a different hand motor control development with specific grasping characteristics; they generally grasp objects with fewer fingers and the fingers not used for the actual grasping are often extended (Jover et al., 2010). These previous findings taken together with the results from the motor and cognitive tests of the present study, could explain the difference in visuomotor performance observed between the groups: Higher reaction times along with difficulties in controlling the pinch force will reduce time on target on the VATT. We observed a general effect of male sex, indicating that males across both groups performed better than females on the VATT. This difference between male and female performance have been observed in other studies with visuomotor tracking paradigms, and the difference could be related to a faster decision-making process in males (Mathew et al., 2020).

The results of the motor and cognitive tests showed that the TD-group performed better than the DS-group on almost all the outcomes, which is testament to some of the deficits presented to the population with DS. The area, where the DS-group were closest to the TD-group were the accuracy on the congruent trials in the flanker tasks, where no significant differences were observed between the groups on the three different tasks. The results of the present study do not indicate any connection between performance on the selected motor and cognitive tests and online motor performance or offline effects of VATT-performance. The only significant correlation observed was a negative correlation between the total number of correct answers on the Corsi block-tapping task and offline effects on the random task in the TD-group, indicating that the participants who scored high on the Corsi task had the highest reduction in motor performance in the offline period. The absence of any other correlations could be explained by a lack of power within the groups, and 2) the lack of correlation between VATT-performance and pegboard performance could be explained by differences in task demands. Whereas the VATT requires unimanual visuomotor accuracy, the Pegboard task requires speed, bimanual coordination and additional cognitive control, which may not have been captured in the measured cognitive parameters.

4.4. Clinical implications

The results of the present study shows how the DS-group acquires and retains a novel visuomotor skill. The online improvement in the DS-group was similar to that of the TD-group and the stability of the motor skill over the 7-day period observed in the DS-group, could implore clinicians and other professionals working with the target group, to focus on practicing skills involving manual dexterity. Our results and previous research indicate a deficit within this domain; however, we show a potential for improvement and overall learning with a relatively small amount of practice. It could be speculated that with a continuous increase in task difficulty over time, the individuals with DS would experience continuous improvements in performance, as seen in TD-individuals (Christiansen et al., 2018).

4.5. Limitations

An investigation of the presence of explicit knowledge about the motor sequence, e.g., as a questionnaire would have been advantageous, as it would have provided an indication of learning strategies. It is important to note that the observed difference between the groups in offline effects, needs to be investigated further. Indeed, the maintenance of motor performance in the DS-group seems to be driven by a large offline improvement in one participant and small improvements in three, while the remaining seven participants reduced their performance roughly by the same amount as the TD-group (Fig. 4C). A larger number of participants in both groups would have been advantageous to the study, in order to diminish the influence of outliers, and more in-depth analysis based on age and functioning level. However, our data analysis demonstrates a statistically significant difference between the groups, which we have addressed. The VATT used in the present study has been used in multiple other studies previously (Beck et al., 2020; Christiansen et al., 2018), and it has close resemblance to the Sequential Visual Isometric Pinch Task (SVIPT) task (Reis et al., 2009; Statton et al., 2015). The task is suited for investigating visuomotor pinch accuracy, for investigating visuomotor skill learning and for assessing retention effects i.e., both within and between-session learning effects. Since the present experiment represents a proof-of-concept study investigating skill learning in DS we chose this lab-based task since it has proven to be efficient in investigating dynamics of visuomotor

skill learning in previous studies. That being said, the employed learning paradigm naturally also has its limitations, which should be acknowledged. While the task and learning paradigm allows assessment of changes in pinch force control and accuracy with motor practice, it also has limitations relating to ecological validity. While visuomotor accuracy and fine motor control is required in many everyday skills, we also acknowledge that the results do not necessarily transfer to other everyday skills. Nevertheless, we believe that the findings of the present study have merit in particular since they shed light on the dynamics of visuomotor skill learning. Lastly, it would have been of interest to have information on the level of severity of the intellectual disability of the DS-group, as this could influence the performance of motor tasks (Gimenez et al., 2017). In that vein, future studies should also consider including a control group matched for mental age, as this would give further insights into the impact of cognitive level on motor learning in the population with DS.

5. Conclusion

Motor performance in the DS-group was lower compared to the TD-group throughout the experiment. The groups displayed similar improvements in online motor performance, i.e., similar learning rates on the visuomotor task. The inclusion of a delayed retention test in a motor learning scenario for young adults with DS is novel and allowed an investigation of how a new motor skill is retained in this population. Both groups demonstrated significant retention seven days after motor practice, indicating similar overall learning effects between the groups. While the TD-group displayed a decrease in mean performance at 7-day retention in the sequence task from the groups' mean immediate retention performance level, mean motor performance was maintained in the DS-group and a difference between the groups was observed. These findings demonstrate that individuals with DS can indeed acquire and retain motor skills with practice, and that some of the dynamics of motor learning are similar to what is observed in the typically developed population, i.e., online learning and overall learning. The cause for the difference in offline effects between the groups is unclear, but we suggest it is the result of the difference in absolute performance and functional difficulty level. The results did not indicate any sequence learning in the groups, as no differences in motor performance between the last sequence-coloured acquisition block and the sequenced non-coloured retention block or the random retention block were observed.

CRedit authorship contribution statement

All authors conceptualized the project together. LMH was responsible for the conducting the experiments and the formal analysis of the data. JW and JLJ assisted with supervision throughout the project. All authors contributed to writing the original draft and to the review and editing phase.

Declaration of Competing Interest

The authors have no conflicts of interest, neither economic nor scientific, in relation to this paper.

Data Availability

Data will be made available on request.

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