#### Visuomotor skill learning in young adults with Down syndrome

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# 17 Abstract

- 18 Background: Individuals with Down syndrome (DS) have impaired general motor skills
- 19 compared to typically developed (TD) individuals.
- 20 Aims: To gain knowledge on how young adults with DS learn and retain new motor skills.
- 21 Methods and Procedures: A DS-group (mean age =  $23.9 \pm 3$  years, N = 11), and an age-
- 22 matched TD-group (mean age  $22.8 \pm 1.8$ , N= 14) were recruited. The participants practiced a
- 23 sequence visuomotor accuracy tracking task (VATT). Online and offline effects of practice
- 24 were assessed in immediate and 7-day retention tests. Participants practiced the task in seven
- 25 blocks (10.6 minutes).
- 26 Outcomes and Results: The TD-group performed better than the DS-group in all blocks (all P
- 27 < 0.001). Both groups improved VATT-performance online from baseline to immediate
- 28 retention (all P < 0.001). The DS-groups' performance at 7-day retention was at the same
- 29 level as the immediate retention tests ( $\Delta DS$ ). An offline decrease in performance was found
- 30 in the TD-group ( $\Delta$ TD, P < 0.001). A between-group difference was observed in the offline
- 31 effect on the sequence task ( $\Delta$ TD  $\Delta$ DS, P = 0.04).
- 32 Conclusions and Implications: The motor performance of adults with DS is lower compared
- to their TD peers. However, adults with DS display significant online performance
- 34 improvement during training, and offline consolidation following motor learning.

#### 36 What this paper adds

37 Learning new motor skills is fundamental throughout our lifespan. Persons with Down 38 syndrome have other prerequisites for learning new tasks, related to psychological, 39 physiological, and anatomical factors imposed by the syndrome. This study is the first to 40 investigate online and offline learning effects of a single motor skill training session in adults 41 with DS. Our results show generally lower motor performance in DS individuals compared to 42 the typically developed population, but with equal online learning effects. Both groups 43 demonstrate retention, i.e., offline stabilisation but while TD demonstrate negative offline 44 effects, this was not the case for DS. These results should be taken into consideration when 45 planning training of motor and general life skills for adults with DS. This work lays the 46 ground for further investigations of the trajectory of the early learning processes and the 47 mechanisms involved when this target group acquires new skills.

#### 48 **1. Introduction**

49 The ability to learn and retain new motor skills is fundamental for everyday functioning, it 50 allows us to perform movements, to meet the demands of our environment, and adapt to 51 changes in it (R. A. Schmidt & Lee, 1999). Individuals born with Down syndrome (DS), are 52 presented with a range of physical and psychological challenges including intellectual 53 disability (Grieco et al., 2015) and impaired general motor skills (Vicari, 2006), which have 54 been attributed to altered anatomical, physiological, and neurological development (Dierssen, 55 2012). Because of this, persons with DS have a higher need for support from health care and 56 social services throughout their lives (Tsou et al., 2020). Knowledge about the characteristics 57 of the ability to learn, consolidate and retain new motor skills is valuable for the population 58 with DS and professionals working with the population. It can lead to improved planning and 59 structure of motor skill training in the everyday life of individuals with DS (Hall et al., 2011; 60 Jain et al., 2022).

61 Previous studies have demonstrated that adults with DS can improve their motor 62 performance during practice (Kerr & Blais, 1985; Reilly et al., 2017), and intervention 63 studies have shown that the group can improve their gross motor skills over several training 64 sessions. The latter has been observed as improvements in gait parameters after 10 weeks of 65 Nordic walking (Skiba et al., 2019) and as improved dribbling skills after 16 weeks of soccer 66 practice (Perić et al., 2021). These results confirm the ability of skill learning in people with 67 DS, but the detailed dynamics of online learning (i.e., the change in performance during 68 practice) and the subsequent offline consolidation and retention of motor skills in adults with 69 DS, has to our knowledge not yet been investigated.

70 Motor skill learning is a complex process, and it is influenced by a range of factors (Dayan & 71 Cohen, 2011), e.g., the organization of task practice (Lage et al., 2015), the type and 72 availability of (augmented) feedback (Oppici et al., 2021) and interindividual differences 73 such as cognitive abilities (Seidler et al., 2013). Both implicit and explicit memory processes 74 are involved in motor skill learning (Hikosaka et al., 2002), and age and baseline skill level 75 seem to influence to which memory system dominate the learning process (Nemeth et al., 76 2013). Individuals with DS display impairments in explicit memory, and a relatively 77 preserved implicit memory (Vicari et al., 2000), and this distinct memory profile could 78 influence how motor skills are acquired and retained in the population. Motor skill learning is 79 influenced by individual cognitive abilities such as executive functions (M. Schmidt et al., 80 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.

84 On the basis on this knowledge, the present paper addresses the questions: What are the 85 dynamics of online and offline motor skill learning for individuals with DS, and are the 86 dynamics different from TD individuals? Furthermore, can the potential differences in the 87 dynamics of motor skill learning be related to differences in cognitive abilities? To address 88 these questions, a visuomotor accuracy tracking task (VATT), was used. The task involves 89 control of dynamic pinch force in a precision grip, to track visual targets. We evaluated the 90 within-session improvement in performance (online effects) as well as the retention seven 91 days after motor practice (offline effects). We hypothesized that the participants with DS 92 would show improved motor performance during acquisition and display retention seven 93 days later. We expected the typically developed (TD) adults to perform better than the DS-94 group in the baseline assessment, and that they would display larger online improvements 95 compared to the DS-group, possibly due to the use of explicit processes dominating early 96 skill acquisition.

97 The task involved five different targets appearing on the screen in a repeated sequence. 98 During motor practice, the sequence was colour-coded, i.e., a specific target colour always 99 appeared in the same position. By implementing two different tests at baseline and retention, 100 one with the sequenced target order, but without colour-coding and one with a random target 101 order, we aimed at investigating to what extent improved motor performance could be related 102 to explicit sequence learning versus improved motor acuity (Shmuelof et al., 2014). The 103 presence of sequence learning was investigated by comparing the performance in the final 104 sequenced practice block (with colours), to the performance in the sequenced, non-colour 105 coded retention block, and by comparing performance in the sequenced and random retention 106 blocks. We expected to observe sequence learning in the TD-group, due to employment of an 107 explicit learning strategy, while we expected this to be absent in the DS-group. To investigate 108 if potential differences in online and offline effects between groups could be related to 109 specific motor or cognitive abilities, we administered a battery of motor and cognitive tests 110 and assessment of fine motor skills.

111

#### 112 **2. Methods**

#### 113 2.1 Participants

Twelve adults with Down syndrome (DS-group) and 14 age-matched typically developed adults (TD-group), were recruited. One participant in the DS-group were excluded, since the person were not able to complete the training. Participants' characteristics is shown in Table 1. The study was approved by The Danish National Committee on Health and Research Ethics (H-19017828) and adhered to the Helsinki Declaration II. All participants gave their informed consent before participation and only participants without guardianship were included.

121 [Table 1]

122

#### 123 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 practice and immediate retention blocks. Exactly one week after their first visit, the participants completed 7-day retention tests and additional motor and cognitive tests.

128

### 129 *2.3 Visuomotor accuracy tracking task*

130 A computer-based dynamic visuomotor accuracy tracking task (VATT) performed with the 131 dominant hand was applied. The task has similarities to tasks used in previously published 132 works from our lab (Beck et al., 2020; Christiansen et al., 2018). The participants were seated 133 approximately 50 cm from a screen at an adjustable table, which was set to ensure a 134 comfortable position of the dominant arm. The tables' hight was recorded, to ensure 135 replication on the second visit. The task required the participants to control a cursor to track a 136 series of target boxes on the monitor, by accurately applying pinch force to a spring-loaded 137 lever, relaying the force to a load-cell (UU2-K10, Dacell, South Korea) (Figure 1, see also 138 Larsen et al., 2016). Then the force was low-pass filtered (10 Hz), amplified (x100) (AM-139 310, Dacell, South Korea), and then fed the PC through a USB-connected data board, 140 sampling at 90 Hz (NI USB-6008, National Instruments, Austin, Texas). An in-house 141 developed Python application ran the VATT. Continuous, online feedback was presented to 142 the participants, as the cursor changed colour from red to blue, when the targets were hit 143 correctly. The targets were presented for 2 s, with 0.2 s between targets. The participants

144 were given augmented feedback, as their time on target in percent were displayed for 2 s after 145 each block. Performance was assessed in two blocks of 59.2 s (27 targets) at baseline, 146 immediately, and seven days after the training session; a block with a random target order 147 and a block with five targets appearing in a repeated sequence. At baseline the random block 148 was performed before the sequence block. The order was reversed at both retention tests. The 149 participants practiced the sequenced task in 7 blocks of 92.2 s each (42 targets), with a 1-150 minute break between blocks. At baseline and retention, all targets were displayed in red. 151 During practice, the five-target sequence had different colours (augmented information on the 152 sequence), i.e., the same colour always appeared in the same position. Before the first 153 baseline block, the participants completed a sequenced familiarization block with red targets 154 (33 s, 15 targets). The participants were not informed about the differences in block structure. 155 Between the baseline and practice blocks, the participants were seated in rest for 20 minutes. 156 Between practice and immediate retention, the participants had a 1-minute break.

157

158 [Figure 1]

### 159 2.4 Visuospatial working memory

160 We applied a computerized forward span corsiblock tapping task, developed by Aeschlimann 161 and co-workers (Aeschlimann et al., 2017). The participants were placed in front of a laptop 162 with a touchscreen with 16 white squares in a 4 by 4 grid. Following a fixation cross, one 163 square at a time changed colours to black in a quasi-randomized pattern, followed by a 164 question mark. Then the participants were instructed to tap the same boxes on the screen in 165 the same order, with their index finger. After an introduction and three practice rounds (two 166 trials with 2-span tasks and one with a 3-span task), the task commenced at 2 spans. Four 167 correct answers out of six trials were required to move on to the next level. If this was not 168 accomplished, the task was terminated.

169

170 2.5 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). The participants were seated in front of a

175 laptop with an external keyboard. In the task, images of rows of five fish turning left or right 176 was displayed, and the participants were instructed to 'feed the correct fish as quickly as 177 possible', either by pressing the left control key with their left index finger or the numpad 178 enter key with their right index finger, determined by the way the fish was facing. The 179 buttons were marked with arrows showing the directions (left ctrl =  $\leftarrow$ , numpad enter =  $\rightarrow$ ). 180 The task contained four trial types, determined by the distractors that were either facing the 181 same direction (congruent), the opposite direction (incongruent), upwards (neutral), or 182 without distractors. The task consisted of three sub-tasks: A regular flanker task, a reversed, 183 and a mixed task. In the regular task the fish were blue, and the participants were instructed 184 to feed the middle fish. In the reversed task the fish were pink, and the participants were told 185 to feed the fish on the sides. The mixed task consisted of both blue and pink fish, and the 186 participants were instructed to feed either the middle or the outside fish, depending on the 187 colour of the fish. Before each sub-task, the participants completed practice rounds. In the 188 regular and reversed tasks, the practice round consisted of 4 stimuli. Another practice round 189 was required if 2 or less trials were correct. The mixed practice round consisted of 8 stimuli, 190 and another round were performed if a participant got 5 or less correct answers. A maximum 191 of three practice rounds were allowed for each sub-task. The test was terminated if a 192 participant failed the third practice round before any the sub-tasks. The regular and reversed 193 tasks consisted of 17 stimuli each (3 congruent, 10 incongruent, 2 neutral and 2 without 194 distractors), and the mixed task consisted of 45 trials (12 congruent, 16 incongruent, 9 neutral 195 and 8 without distractors). The average reaction times (RT) on all trials and the RTs on the 196 congruent and incongruent trials were extracted as outcomes, as well as the accuracy rates on 197 the congruent and incongruent trials (correct answers/total number of trials). The flanker 198 effects were calculated as: incongruent – congruent, both in the RT and accuracy measures.

199

#### 200 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 seconds 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 seconds. 4) The assembly task, where the subjects were instructed to build as many

207 assemblies as possible using three different parts in 60 seconds. Before each task, the 208 experimenter instructed and demonstrated the task, and the participants practiced the task.

209 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)).

211

### 212 2.7 Data analysis and statistics

213 All analyses were performed in R statistics (R Statistics 4.0.0, 2020). A linear mixed effects 214 model was fitted to the data ("Ime4" R-package (Bates et al., 2015)), to investigate the effect 215 of time (i.e., the performance on the 13 blocks), group and type of block (sequence or 216 random) on motor performance. Time on target in seconds, were translated to a score 217 between 0 and 100 points, with each point being equal to 0.02 s. Group, time, and block type 218 were included as fixed effects, together with sex and corsiblock span performance (the level 219 at which the participants failed the test). Sex and corsiblock performance were included to 220 control for the variation these factors could introduce to the data. Since the participants were 221 likely to display differences in baseline performance and our experiment is a repeated 222 measures design, random intercepts were fitted for each participant.

(1): Time on target ~ Group x Block x Type + Sex + Corsiblock span number +
(1|participant)

225 The equation above is presented in R-terminology where "(1|participant)" represents the 226 individual intercepts and the x'es indicates the interactions between the fixed effects of 227 Group, Block and Type. If significant effects of the interactions between Time on target and 228 Group and Block (time), were observed, post hoc analyses of within- and between-group 229 differences were performed. The post hoc analyses investigated differences in online and 230 offline leaning, and differences in performance between the random and sequence blocks, to 231 investigate sequence learning. P-values from the post-hoc analyses were corrected for 232 multiple comparisons with the single step method. Paired T-tests were used to investigate 233 differences between the groups in the motor and cognitive tests. Unpaired T-tests were used 234 to determine the presence of flanker effects within the groups. Alpha level was set at P < P235 0.05, for all analyses. Results from the linear mixed effects model are presented as model 236 estimated means  $\pm$  standard error (SE). The results of the motor and cognitive test are 237 presented as mean  $\pm$  SE. Correlation matrixes were computed for each motor and cognitive

- tests and VATT performance on the sequenced task at baseline and online and offline effects,
- and for the absolute motor performance level on day 1 and offline effects.
- 240

#### **3. Results**

### 242 *3.1 Fine motor skills and cognitive test performance*

243 Paired T-tests showed that the TD-group performed better in the pegboard task and the 244 corsiblock tapping task compared to the DS group (all P-values < 0.001, Table 2). The results 245 from the flanker tasks showed that the TD-group had lower reaction times (RT) on the three 246 sub-tasks, both in the average RT for all trials and on the congruent and incongruent trials 247 (table 2): Regular flanker (all P-values < 0.001), reversed flanker (all P-values < 0.01) and 248 mixed flanker (all P-values < 0.05). The TD-group was more accurate on the incongruent 249 trials in all three tasks compared to the DS group (all P-values < 0.01), but no differences 250 were observed on the congruent trials. No flanker effects were observed for RT in either of 251 the groups in the regular flanker (DS-group: P = 0.923, TD-group: P = 0.192). The DS-group 252 displayed a flanker effect in accuracy in the regular flanker task (P = 0.006), while the TD-253 group did not (P = 0.104), this was seen as a significant difference between the groups in the 254 paired T-test (P < 0.05). In the reversed flanker task, the DS-group presented a flanker effect 255 on RT (P = 0.025), while the TD-group did not (P = 0.083). The paired t-test revealed a 256 significant difference in the flanker effect on RT between the groups (P = 0.019). With 257 regards to accuracy, both groups displayed flanker effects (DS-group: P = 0.045, TD-group: 258 P = 0.040). In the mixed task, both groups presented flanker effects on RT (DS-group: P =259 0.016, TD-group: P = 0.001), and accuracy (DS-group: P < 0.001, TD-group: P = 0.012). The 260 paired T-tests between the groups showed no difference between the groups in the flanker 261 effect in RT (P = 0.105), and a significant difference between the groups in the flanker effect 262 on accuracy (P < 0.001).

263

264 [Table 2]

265

266 *3.2 Motor performance in the VATT* 

267 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-

269 group performed ~30 points better than the DS group at all timepoints (all P < 0.001),

equivalent of 0.6 seconds time on target on each target (Figure 2). A significant effect of sex was found (F = 4.6, P = 0.04) in the VATT, while no significant effect of corsiblock performance were observed (P = 0.88).

273

#### *3.3 Online effects*

275 Changes in visuomotor performance are presented as changes in time on target score. The 276 learning curves for both groups are depicted in Figure 2. The improvements in motor 277 performance within session (online) and between sessions (offline) are shown on Figure 3. 278 Both the groups improved their online motor performance in the sequence task from baseline 279 to immediate retention, (immediate retention – baseline,  $\Delta$ online) ( $\Delta$ online DS = 8.4 ± 1.7, P 280 <0.001;  $\Delta$ online TD = 11.9 ± 1.5, P < 0.001, Figure 3 A and B). No difference in online 281 learning was observed between the groups ( $\Delta$ online TD –  $\Delta$ online DS).

282

### 283 *3.4 Retention and Offline effects*

284 Both groups showed retention of the task, as their performances on the 7-day retention tests 285 were significantly better than their baseline performances on both the sequence (DS:  $7.9 \pm$ 286 1.7, P < 0.001, TD: 5.8  $\pm$  1.5, P < 0.001, Figure 3B) and random tasks (DS: 11.3  $\pm$  6.7, P < 287 0.001, TD:  $10.9 \pm 1.5$ , P < 0.001, Figure 3B). No difference between the groups was observed 288 in retention. No difference in motor performance was observed in the DS-group from 289 immediate to 7-day retention in the sequence task (7-day retention – immediate retention, 290  $\Delta$ offline) ( $\Delta$ offline DS: -0.5 ± 1.7, P = 0.995, Figure 3C). The TD-group had a significant 291 offline decrease in performance in the sequence task ( $\Delta$ offline TD: -6.1 ± 1.5, P < 0.001, 292 Figure 3C). A between-group difference was observed in the offline effects in performance in 293 the sequence task from immediate to 7-day retention ( $\Delta$ offline TD –  $\Delta$ offline DS: -5.6 ± 2.2 P 294 = 0.04, Figure 3C).

295

296 [Figure 3]

297

298 3.5 Implicit and explicit learning

The amount 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

302 retention blocks. This was also done between the retention blocks at day 7. No significant

303 differences in performance within or between groups were observed in these analyses.

304

## 305 3.6 Correlations between online performance and offline effects

A correlation matrix between motor performance on the immediate retention blocks and the performance changes from immediate and 7-day retention, showed significant negative correlations between the offline change in performance on the sequence task and absolute motor performance in both the random (R = -0.48, P = 0.020) and the sequence immediate retention block (R = -0.57, P = 0.005).

311

### 312 3.7 Correlations between motor performance and motor and cognitive tests

313 The correlation matrix for the flanker task showed a significant negative correlation between 314 baseline VATT performance and reaction time on the congruent trials in the regular flanker 315 task (P = 0.005; Sequence: R = -0.75, P < 0.001), and a significant positive correlation 316 between accuracy on the incongruent trials and baseline VATT performance (R = 0.68, P < 0.68, P317 (0.001). In the flanker effects, we observed significant positive correlations between the 318 baseline VATT performance and the flanker effects on accuracy on the regular (R = 0.46, P 319 =0.031) and mixed flanker task (R = 0.75, P < 0.001). We did not observe any correlations 320 between baseline VATT performance and the flanker effects on reaction time. In the 321 Pegboard task, significant positive correlations were observed between baseline VATT 322 performance and both Sum of Scores (R = 0.78, P < 0.001) and Assembly (R = 0.85, P < 0.001) 323 0.001). Positive significant correlations were observed between baseline VATT performance 324 and corsiblock performance (span number: R = 0.75, P < 0.001, correctly answered trials: R =325 0.78, P < 0.001). The correlation matrixes also included the online and offline effects. No 326 correlations were observed between online or offline effects and outcomes from the flanker, 327 Pegboard or corsiblock tasks (P-values > 0.05).

328

#### 329 **4. Discussion**

This study is the first to investigate motor skill learning in 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.

#### 334 *4.1 Online effects and sequence learning*

335 The participants in both the DS- and TD-group demonstrated positive online effects of motor 336 practice i.e., improved their performance from baseline to immediate retention tests. This 337 finding is in line with previous studies, which have shown that individuals with DS can 338 improve their motor performance during practice (de Mello Monteiro et al., 2017; Kerr & 339 Blais, 1985, 1987). Kerr and Blais used a pursuit tracking task performed with a steering 340 wheel, which required the participants to accurately move the wheel to a given target location 341 to investigate probability learning in youths with DS (Kerr & Blais, 1985, 1987). The pursuit 342 tracking task somewhat resembles the visuomotor tracking element of the VATT in the 343 present study. The researchers observed online performance improvements as a ~23% 344 decrease in total trial time in 8 blocks and 800 trials (from ~2300 ms to ~2080 ms) (Kerr & 345 Blais, 1985), while the present study shows a 40% increase in time on target in 7 blocks with 346 294 trials (from 17.03 to 23.88, Figure 2). Direct comparisons in the magnitude of the 347 improvements should be made with care, as the tasks are innately different. The aim of the 348 pursuit tracking task was to investigate probability learning in persons with DS, not how the 349 group acquired the motor skill of steering the wheel in a precise manner, while the aim with 350 the VATT in the present study was to assess changes in the ability to control pinch force.

351 Despite differences in baseline motor performance, the DS- and TD-group showed equal 352 improvements in online motor performance. No previous study has reported similar findings. 353 In some previous studies, the age-matched control groups did not improve their performance 354 in the tasks; they performed at maximum level at the baseline test leaving no further room for 355 improvement i.e., a ceiling effect (de Mello Monteiro et al., 2017; Kerr & Blais, 1985, 1987). 356 This discrepancy in online effects between the present and earlier studies could be explained 357 by differences in task designs, including difficulty levels. For instance, the study by Monteiro 358 and colleagues applied a prediction reaction time model, which seem to leave very little room 359 for improvements in the TD-group. The researchers also divided the DS-group into a high 360 and low performing group based on their baseline scores. They observed that only the low 361 performing group improved their scores. The requirements for precise pinch force control in 362 the visuomotor tracking task of the present study, was chosen deliberately, as the aim was to 363 investigate potential differences in motor skill learning between the groups, and that requires 364 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

367 higher a score they can obtain. Thus, it could be speculated that part of the differences 368 between the groups could be attributed to differences in RT and potentially to prediction in 369 the sequence-based task. To our knowledge no previous study has applied such a model in 370 individuals with DS. The flanker task used in the present study includes an RT-element. 371 Here, we find significantly higher RTs on all three tasks in the DS-group compared to the 372 TD- group, ranging from  $\sim$ 950 ms in the regular task to  $\sim$ 300 ms in the mixed task, which is 373 in line with previous findings (Davis et al., 1991). This difference in reaction time could 374 partly explain the difference in VATT-performance of the DS-group and the TD-group, 375 however, further experiments and analyses are necessary to conclude on the role of 376 differences in motor reaction time in the VATT.

377 The protocol of the present study was chosen based on pilot experiments, which indicated 378 that protocols similar to previous studies (e.g., 6 blocks of 3 minutes) was exhausting for the 379 participants with DS to complete (Beck et al., 2020; Thomas et al., 2016). We expected to 380 observe sequence learning after exposure to the colored sequence during the seven practice 381 blocks in the TD-group, as this has been observed previously (Krakauer et al., 2019; Shea et 382 al., 2006). However, no differences in performance between Block 7 (sequenced and colour-383 enhanced) and the sequence block at immediate retention (sequenced, un-coloured), nor 384 between the sequence and random block at immediate retention was observed in the TD-385 group. We did not observe sequence learning within the DS-group as expected. It could be 386 the reduced amount of total practice time and less exposure to the sequence that is the cause 387 of the absence of sequence learning in the TD-group. If the participants had been told to be 388 aware of a sequence in the task, it is possible that sequence learning would have been 389 observed.

390 The results for online effects demonstrate that individuals with DS improve performance with 391 motor practice to the same extent as TD individuals. Since initial skill acquisition is 392 influenced by involvement of cognitive processes and explicit aspects of learning (Krakauer 393 et al., 2019) we could have expected higher performance gains in the TD-group since this 394 group is characterized by higher performance in the cognitive tests. This was however not the 395 case. While the similar online effects between the two groups may be influenced by the 396 higher baseline performance in the TD-group, it can nevertheless be concluded that DS-group 397 demonstrate skill acquisition.

400 To investigate offline effects and retention on motor learning, we employed delayed retention 401 tests seven days after acquisition of the VATT. The present study is, to the authors 402 knowledge, the first to apply such a design in adults with DS. Earlier studies have 403 demonstrated that persons with DS improve both their gross (Almeida et al., 1994; Perić et 404 al., 2021; Skiba et al., 2019) and fine motor performance (Latash et al., 2002) with several 405 practice sessions practice. The previous studies did not include performance measures 406 between sessions; thus, it is not possible to gauge the offline effects or retention of the motor 407 practice between single sessions. The results of the present study show that adults with DS 408 exhibit retention on the VATT, as significant increases in motor performance at 7-day 409 retention compared to the baseline performance. The DS-group exhibited the same level of 410 retention as the TD-group.

411 A difference between the groups in the offline changes in motor performance on the sequence 412 task was observed (Figure 3B). The DS-group maintained their performance from immediate 413 to 7-day retention, while performance decreased in the TD-group. This difference between 414 the groups could be explained by the difference in absolute performance, which was 415 significant on all blocks throughout the study ( $\sim 0.6$  s time on target per target). Indeed, we 416 observed negative correlations between absolute motor performance on both the retention 417 blocks on day 1 and the offline change in motor performance in the sequence task, indicating 418 that a high absolute performance on day 1, correlated with a larger decrease in performance 419 from immediate to 7-day retention. The TD-group might have reached a performance close to 420 the ceiling of the task, thus making it less likely for the group to achieve the same level of 421 performance seven days after the acquisition. Reis and colleagues observed either a 422 performance maintenance or decrease on a continuous pinch task, suggesting that this offline 423 pattern might be the default on continuous pinch tasks, contrary to serial tapping tasks, where 424 offline increases in performance have been observed (Reis et al., 2009; Siengsukon & Al-425 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 a in 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 433 the DS-group, while it was too easy for the TD-group, and thus the negative offline effect 434 was observed. To address the issue of functional difficulty level, future studies should include 435 tasks, that are adjusted to ensure the same absolute performance level at baseline, and a 436 control group that is matched to the same absolute online performance level as the DS-group. 437 The differences in offline effects between the groups could point to differences in how the 438 explicit and implicit memory systems are engaged during online motor learning (Robertson et 439 al., 2004). Previous research has shown that individuals with DS have deficits in explicit 440 memory and implicit memory abilities comparable to typically developed peers (Vicari et al., 441 2000). It could be speculated that the DS-group employs a predominantly implicit learning 442 strategy during skill acquisition, and that could be part of the reason for the maintained motor 443 performance observed in the DS-group. The TD-groups' decrease in visuomotor performance 444 at 7-day retention could indicate the engagement of explicit, cognitive processes that is prone 445 to interference due to the presentation of competing knowledge during the offline period 446 (Fletcher et al., 2005). However, we did not find indications of explicit sequence learning on 447 day 1, which contradicts the notion that it is a loss of explicit knowledge of the sequence that 448 is the cause of the performance decrease in the TD-group.

449

### 450 4.3 Variability in performance and motor and cognitive tests

451 Several factors could contribute to the difference in general motor performance level between 452 the groups in the present study. Recent studies have demonstrated increased accuracy and 453 lower variability in the pinch force task with increased age during neurotypical development 454 (Beck, Spedden, & Lundbye-Jensen, 2021; Beck, Spedden, Dietz, et al., 2021). Studies 455 investigating the ability to modulate or hold a specific force output have shown greater 456 variability and a generally lower force output in individuals with DS (Heffernan et al., 2009; 457 Rao et al., 2017). In the present study, we observe greater interindividual variability in the 458 DS-group, than in the TD-group, evident as larger confidence intervals in Time on Target 459 score (Figure 2). In addition, children and adolescents with DS have a different hand motor 460 control development with specific grasping characteristics; they generally grasp objects with 461 fewer fingers and the fingers not used for the actual grasping are often extended (Jover et al., 462 2010). These previous findings taken together with the results from the motor and cognitive 463 tests of the present study, could explain the difference in visuomotor performance observed 464 between the groups: Higher reaction times along with difficulties in controlling the pinch 465 force will reduce time on target in the VATT. We observed significant correlations between

baseline VATT performance, and performance in the pegboard, corsiblock, and flanker tasks.
This indicates a relation between performance on the VATT, and the skills measured by these
tests, while they also are an expression of the general cognitive and motor challenges related
to DS.

470

#### 471 *4.4 Limitations*

472 An investigation of the presence of explicit knowledge about the motor sequence, e.g., as a 473 questionnaire would have been advantageous, as it would have provided an indication of 474 learning strategies. It is important to note that the observed difference between the groups in 475 offline effects, needs to be investigated further. Indeed, the maintenance of motor 476 performance in the DS-group seems to be driven by a large offline improvement in one 477 participant and small improvements in three, while the remaining seven participants reduce 478 their performance roughly by the same amount as the TD-group (Figure 3C). Despite this, our 479 data analysis demonstrates a statistically significant difference between the groups. Lastly, it 480 would have been of interest to have information on the level of severity of the intellectual 481 disability of the DS-group, as this could to influence the performance of motor tasks 482 (Gimenez et al., 2017).

483

### 484 **5.** Conclusion

485 Motor performance in the DS-group was lower compared to the TD-group throughout the 486 experiment. The groups displayed similar improvements in online motor performance. The 487 inclusion of a delayed retention test in a motor learning scenario for young adults with DS is 488 novel and allowed an investigation of how a new motor skill is retained in this population. 489 Both groups demonstrated significant retention seven days after motor practice. While the 490 TD-group displayed a decrease in performance at 7-day retention in the sequence task from 491 the groups' immediate retention performance level, motor performance was maintained in the 492 DS-group and a difference between the groups was observed. These findings demonstrate 493 that individuals with DS can indeed acquire and retain motor skills with practice. The cause 494 for the difference in offline effects between the groups is unclear, but we suggest it is the 495 result of the difference in absolute performance and functional difficulty level. The results did 496 not indicate any sequence learning in the groups, as no differences in motor performance

497 between the last sequence-coloured acquisition block and the sequenced non-coloured

498 retention block or the random retention block were observed.

499

### 500 **Conflicts of interests**

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

- 502 paper.
- 503

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673	

## 675 **Tables and figures**

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### Table 1: Demographic data of the participants

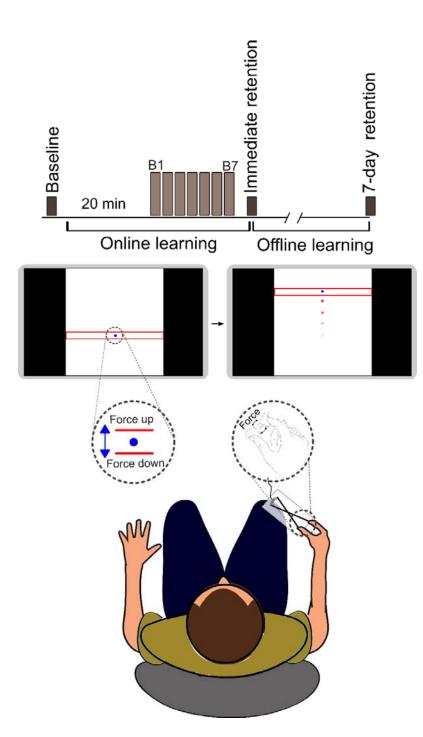
Variables	DS	TD
Number of participants	11	14
Sex (f/m)	3/8	6/8
Age (years $\pm$ SD)	$23.9\pm3.0$	$22.8 \pm 1.8$

Variables	DS	DS		TD	
Pegboard					
Sum of scores	$18.5 \pm$	1.7	$39.2 \pm$	1.0	***
Assembly	13.1 ±	1.3	$43.7 \hspace{0.2cm} \pm \hspace{0.2cm}$	1.4	***
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 ±	13	
Regular flanker effect acc.	-0.38 $\pm$	0.11	$0.04$ $\pm$	0	*
Reversed RT all (ms)	1299 ±	123	$439 \hspace{0.2cm} \pm \hspace{0.2cm}$	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 ±	9	*
Reversed, flanker effect, acc.	-0.11 ±	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	***
Corsiblock 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	***

#### Table 2: Outcomes of the motor and cognitive tests

678 Sum of scores = The sum of pins placed with the right, left and both hands in three separate 679 trails. Assembly score = The number of correctly placed parts in the assembly task. RT =680 Reaction time. Acc. = Accuracy ratio, calculated as number of correctly answered trials dived 681 by the number of total trials. Flanker effect RT = Reaction time in the congruent trials 682 subtracted from the reaction time in incongruent trials. Flanker effect acc. = Accuracy ratio in 683 the congruent trials subtracted from the accuracy ratio in the incongruent trials. Corsiblock 684 Span number = The number of spans at which the participants failed the task. Correct 685 answers = The number of correctly performed trials. Data is presented as means  $\pm$  SE. Stars 686 indicate the P-values of the paired T-tests between the groups. Significance codes: "\*" 687 denotes p < 0.05, "\*\*" denotes p < 0.01, "\*\*\*" denotes p < 0.001.

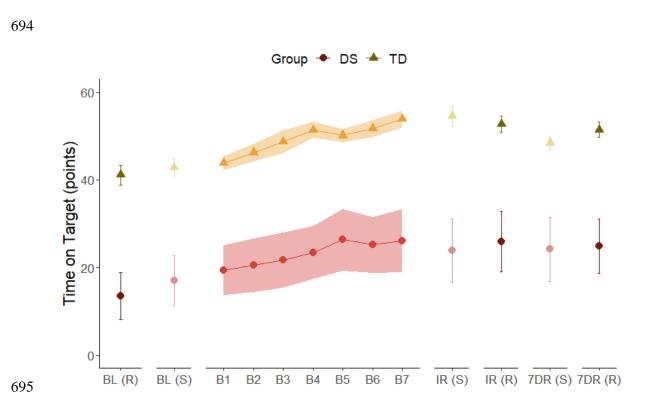


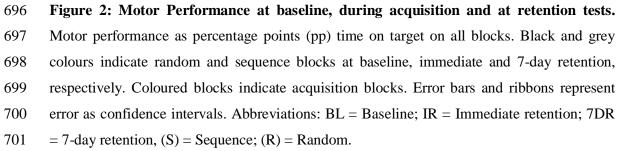


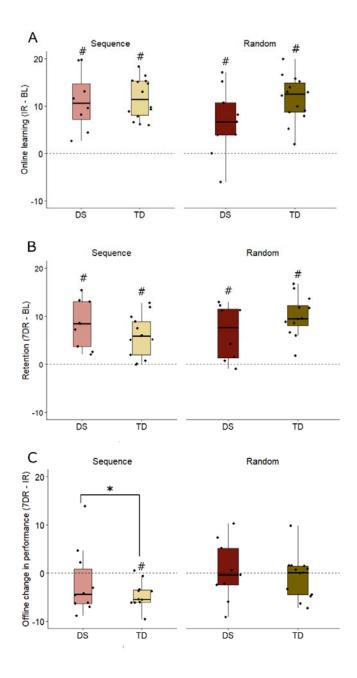
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- 690 **Figure 1**: Outline of the VATT and study protocol of the current study. The underarms are
- 691 resting on an adjustable table, to ensure a comfortable position.

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702

703 Figure 3: Online and offline effects. Box plots with individual values displaying changes in 704 performance on both the sequenced and random tasks. (A) online performance change from 705 baseline to immediate retention, (B) retention of the VATT, measured as performance 706 changes from baseline to 7-day retention, and (C) offline change in performance from 707 immediate to 7-day retention. The dotted lines at 0 on the y-axes is equal to the groups' motor 708 performance on the baseline blocks (A and B) and immediate retention blocks (C). BL =709 Baseline, IR = Immediate retention, 7DR = 7-day retention. # = Significant within-group 710 difference. \* = Significant between-group difference in the relative changes.