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RESEARCH ARTICLE

Why do individuals with Williams syndrome or Down syndrome fail the Weather Prediction Task?

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Abstract

Williams syndrome (WS) and Down syndrome (DS) are two neurodevelopmental disorders with distinct genetic origins characterized by mild to moderate intellectual disability. Individuals with WS or DS exhibit impaired hippocampus-dependent place learning and enhanced striatum-dependent spatial response learning. Here, we used the Weather Prediction Task (WPT), which can be solved using hippocampus- or striatum-dependent learning strategies, to determine whether individuals with WS or DS exhibit similar profiles outside the spatial domain. Only 10% of individuals with WS or DS solved the WPT. We further assessed whether a concurrent memory task could promote reliance on procedural learning to solve the WPT in individuals with WS but found that the concurrent task did not improve performance. To understand how the probabilistic cue–outcome associations influences WPT performance, and whether individuals with WS or DS can ignore distractors, we assessed performance using a visual learning task with differing reward contingencies, and a modified WPT with unpredictive cues. Both probabilistic feedback and distractors negatively impacted the performance of individuals with WS or DS. These findings are consistent with deficits in hippocampus-dependent learning and executive functions, and reveal the importance of congruent feedback and the minimization of distractors to optimize learning in these two populations.

KEYWORDS
explicit, feedback-based learning, genetic disorders, multiple memory systems, multiple-cue learning, probabilistic classification learning, Trisomy 21

1 | INTRODUCTION

Williams syndrome (WS) and Down syndrome (DS; Trisomy 21) are two neurodevelopmental disorders with distinct genetic origins characterized by mild to moderate intellectual disability (de Graaf et al., 2017; Ewart et al., 1993; Korenberg et al., 2000; Lejeune et al., 1959; Martens et al., 2008). Even though individuals with these syndromes have relatively similar mental ages and IQs (WS mental age range: 4.1–10.7 years of age, IQ: mean 55, range 40–70; DS mental age range: 4.3–9 years of age, IQ: mean 50, range 30–70 (Banta Lavenex et al., 2015; Bostelmann et al., 2017; Bostelmann et al., 2018; Bostelmann et al., 2020; Martens et al., 2008; Mégarbané et al., 2013; Vicari et al., 2005, 2006; Vicari et al., 2007; current study)), they exhibit distinct cognitive profiles with different strengths and weaknesses (Jarrold et al., 1999; Karmiloff-Smith, 2013; Porter & Coltheart, 2006; Vicari, 2001; Vicari et al., 2005; Vicari et al., 2016; Wang & Bellugi, 1993, 1994). Indeed, some abilities considered to be relatively preserved in individuals with DS are often more impaired in individuals with WS, and vice versa. For example, individuals with DS exhibit poor language capacities, and impaired verbal and visuo-perceptual memory, but relatively spared nonverbal capacities (e.g., graphic, constructive, and visuo-spatial abilities) (Bihrle et al., 1989; Jarrold et al., 1999; Jarrold et al., 2007; Porter & Coltheart, 2006; Ricketts, 2011; Vicari et al., 2016; Wang & Bellugi, 1994). In contrast, individuals with WS exhibit relative strengths in language production and face recognition, but severe deficits in visuo-spatial processing, including the ability to decipher, judge, recall, and reconstruct the relationship between visually presented forms and objects (Atkinson et al., 2003; Bellugi et al., 2000; Martens et al., 2008; Mervis et al., 2000; Porter & Coltheart, 2006; Vicari et al., 2016). Since individuals with these syndromes have overlapping mental ages but often exhibit opposing cognitive strengths and weaknesses, there is a long history of comparing the performance of individuals with these syndromes on a variety of different tasks. Findings from these comparative studies serve to further characterize the cognitive profiles associated with each of these syndromes, which is important for the families and caregivers of individuals with these syndromes. However, comparative studies also provide critical empirical evidence for understanding brain structure–function relations in both typical and atypical development, as syndrome-specific brain abnormalities enable researchers to make and test syndrome-specific hypotheses about performance.

For example, individuals with WS and individuals with DS exhibit different profiles in their real-world spatial learning abilities (Banta Lavenex & Lavenex, 2021). Indeed, they both show impaired hippocampus-dependent place learning and enhanced striatum-dependent response learning, but to different extents (Bostelmann et al., 2017; Bostelmann et al., 2018). Consistent with observed hippocampal abnormalities (Meyer-Lindenberg et al., 2005), individuals with WS are severely impaired at low-resolution hippocampus-dependent place learning (NB: since high-resolution abilities require low-resolution abilities, high-resolution abilities were not tested in WS), but exhibit strongly facilitated striatum-dependent response learning (Bostelmann et al., 2017). In contrast, individuals with DS demonstrate relatively preserved low-resolution place learning which is dependent on the CA1 region of the hippocampus, but impaired high-resolution place learning which is dependent on pattern separation and the dentate gyrus, and weakly facilitated striatum-dependent response learning (Banta Lavenex et al., 2015; Bostelmann et al., 2018). The fact that high-resolution place learning is particularly impacted in DS is consistent with the finding that DS has a particularly important impact on the dentate gyrus within the hippocampal formation (Contestabile et al., 2010; Koenig et al., 2021; Menghini et al., 2011; Pennington et al., 2003). Thus, as predicted by the competitive interaction hypothesis (Chang & Gold, 2003; Jacobson et al., 2012; Packard & Goodman, 2013; Schroeder et al., 2002; White & McDonald, 2002), individuals with WS exhibit strongly facilitated striatum-dependent response learning in the presence of severe impairment in hippocampus-dependent place learning, whereas individuals with DS exhibit weakly facilitated striatum-dependent response learning in the presence of impairments in only some aspects of hippocampus-dependent place learning as compared to typically developing (TD) children in the same mental age range (Banta Lavenex et al., 2015; Bostelmann et al., 2017; Bostelmann et al., 2018). Here, we used the Weather Prediction Task (WPT), a commonly used clinical neuropsychological task, which can be solved using different learning strategies dependent on either the hippocampus or the striatum, to determine whether cognitive profiles similar to those observed in the spatial domain in individuals with WS or DS can be observed outside the spatial domain.

The WPT was originally thought to assess striatum-dependent procedural or "implicit" learning (Knowlton et al., 1994) and is still commonly used to assess striatum-dependent procedural learning and memory in typical and atypical development and in neurological disorders (e.g., Fernandez et al., 2021; Gabay & Goldfarb, 2017; Karcher et al., 2019; Labouliere et al., 2016; Mayor-Dubois et al., 2016). In this task, participants must predict the weather outcome (sunshine or rain) based on different combinations of four tarot cards (cues). However, just like the weather, cue–outcome associations are probabilistic and thus not perfectly correlated. Whereas two highly predictive cues are associated with rain or sunshine on 75% of trials and with the opposite outcome on the remaining trials, two less predictive cues are associated with rain or sunshine on 57% of trials and with the opposite outcome on the remaining trials. Importantly, participants are not explicitly informed about the probabilistic nature of the task. It was therefore initially assumed that participants gradually learn the cue–outcome associations by trial-and-error feedback using a multi-cue strategy, which was described as a procedural learning strategy enabling the prediction of the correct outcome based on the combination of all cue–outcome probabilities (Knowlton et al., 1994). However, some studies have suggested that participants can use different learning strategies to solve the WPT (e.g., Bochud-Fragnière et al., 2022; Foerde et al., 2006; Hopkins et al., 2004; Li et al., 2016; Price, 2009). For example, Foerde and colleagues showed that both declarative and habit (procedural) learning can support similar above chance performance on the WPT (Foerde et al., 2006; Foerde et al., 2007). Indeed, in an fMRI study, they showed that medial temporal lobe structures are normally activated when young healthy adults solve the WPT. However, medial temporal lobe structures do not seem to be involved when the same participants solve the WPT in a dual-task condition including a concurrent working memory task. Moreover, in the single-task condition, significant declarative knowledge was gained and the activity of the hippocampus correlated with task performance, suggesting that the hippocampus was engaged in solving the WPT. In contrast, in the dual-task condition when the hippocampus was solicited with a concurrent working memory task (i.e., high tones counting during the WPT), participants still solved the WPT but acquired less declarative knowledge and the activity of the putamen correlated with WPT performance, suggesting that participants were pushed toward using a striatum-dependent procedural learning strategy to solve the WPT. This study suggests that, although the healthy adults preferentially used the hippocampus-dependent learning and memory system to solve the WPT in classical single-task condition, they can also solve the task using the striatum-dependent memory system. Thus, patients with developmental or acquired hippocampal deficiency might also have the possibility to solve the WPT using at least the striatum-dependent memory system as already shown with some amnesic patients (Hopkins et al., 2004; Knowlton et al., 1994).

In agreement with these findings, a new strategy analysis has shown that most healthy individuals use a learning strategy likely dependent on the hippocampus to solve the WPT under single-task conditions. Indeed, Bochud-Fragnière et al. (2022) demonstrated that 20- to 30-year-old adults solve a shortened version of the WPT (i.e., 100 trials) using a flexible cognitive strategy that is likely dependent on the hippocampus. In their study, they introduced new “pattern analyses” as a more appropriate manner of characterizing the learning strategies used to solve the WPT. First, they computed the performance of every single participant on each of the 14 individual patterns of cues. Second, they compared the overall performance of young adults for each individual pattern to the performance that would be expected if participants were using a particular learning strategy. Finally, they compared the different levels of performance between the 14 patterns to characterize how each individual cue influenced the participants' choices when presented alone or in combination with other cues. They found that most young adults do not use a multicue strategy to solve the WPT (i.e., predicting the correct outcome based on their implicit knowledge about the probabilities of cue–outcome associations acquired across successive trials), as initially proposed (Knowlton et al., 1994). Instead, their analyses demonstrated that the responses of young adults matched the use of a hierarchical strategy, in which participants consider the highly predictive cues primarily and the less predictive cues secondarily, but only when the two incongruent highly predictive cues were not present simultaneously. This strategy, by which the participant determines the correct outcome following a series of hierarchical comparisons of the cues on each trial, is akin to a context-dependent or conditional learning strategy that has been shown to be dependent on the hippocampus (Andersen et al., 2007; Greene et al., 2006; Ridley et al., 1995).

Bochud-Fragnière et al. (2023) then investigated when and how young healthy TD children from 3 years of age solve the WPT, which had been previously evaluated only with children from 8 years of age (Mayor-Dubois et al., 2016) and with a strategy analysis that was later shown to be inadequate (for a more complete discussion see Bochud-Fragnière et al. (2022)). Using the same methodological design and analyses as Bochud-Fragnière et al. (2022), they showed that whereas some TD children demonstrated the capacity to solve the WPT from 5.5 years of age using the two most predictive cues and neglecting the two less predictive cues, most TD children from 7.5 years of age used a hierarchical learning strategy as 20- to 30-year-old adults did (Bochud-Fragnière et al., 2022; Bochud-Fragnière et al., 2023). Bochud-Fragnière et al. (2023) suggested that the evolution of the learning strategies used by TD children from middle to late childhood, progressing from the use of the two most predictive cues strategy to the hierarchical strategy, may reflect their increasing ability to use conditional information concomitant with the development of the hippocampus-dependent learning and memory system (Lavenex & Banta Lavenex, 2013).

The aim of the current descriptive study was to determine whether individuals with WS or DS can solve the WPT, a task that can be solved by TD children from 5.5 years of age using the two most predictive cues strategy, and by TD children from 7.5 years of age using a conditional, hierarchical strategy. Depending on whether individuals with WS or DS are able to solve the WPT or not, we further aimed to characterize the learning strategy they employed, or to identify intrinsic factors of the WPT, which may negatively impact their performance, respectively.

2 | EXPERIMENT 1: STANDARD WPT

In Experiment 1, we sought to determine whether individuals with WS or DS can solve the standard WPT, in which participants must predict the weather based on probabilistic cue–outcome associations.

2.1 | Materials and methods

2.1.1 | Participants

We recruited 40 individuals with WS (22 females), 17 individuals with DS (8 females), 22 young healthy adults (11 females) and 71 TD children (31 girls) with a chronological age similar to the mental age of individuals with WS or DS. Two of the 71 children (one 3.59-year-old boy and one 3.70-year-old boy) were excluded because they did not finish the task, leaving a total of 69 TD participants (Table 1). Participants with WS or DS were recruited in Switzerland (WS: $n = 9$; DS: $n = 16$) and Italy (WS: $n = 29$; DS: $n = 1$). Exclusion criteria for individuals with WS or DS were severe motor, comprehension or attentional difficulties which precluded completing testing. Thus, we excluded one 7.13-year-old boy with WS and one 26.14-year-old man with WS, leaving a total of 38 participants with WS (Table 1). We did not exclude any individuals with DS (Table 1). No behavioral or performance differences were observed between the Swiss and the Italian participants with WS or DS (data not shown), and thus the data for these populations were grouped

TABLE 1 Chronological and mental ages of participants retained in Experiment 1.

		Chronological age (years)		Mental age (years)	
Participants	*n*	Mean	Range	Mean	Range
WS	38	25.9	9.4–47.9	6.5[a]	4.1–10.7[a]
DS	17	18.0	6.8–30.8	5.6	4.3–7.2
TD children	69	6.6	3.0–11.9	N/A	N/A
TD adults	22	26.3	21.7–30.8	N/A	N/A

N/A, not assessed.
[a]One 23-year-old individual with WS did not perform the Leiter-R Brief IQ.

for analysis and presentation. According to parents or caregivers, no participant exhibited signs of age-related dementia. The mental age of all participants with DS and all but one participants with WS was determined using the Leiter International Performance Scale-Revised (Table 1; Subtests included in the Brief IQ from which mental age is calculated are: Figure Ground, Form Completion, Sequential Order, and Repeated Patterns) (Roid & Miller, 1997). All TD children and young healthy adults were recruited in Switzerland. Exclusion criteria were atypical development (e.g., developmental delay or high potential), neurological disorders (e.g., epilepsy) or chronic substance use (drugs or medications). Although the mental age of TD children and adults was not assessed, we assumed that it matched their chronological age. The results of the TD children and adults have been published previously (Bochud-Fragnière et al., 2022; Bochud-Fragnière et al., 2023).

Testing took place between 8:00 AM and 6:30 PM, in a quiet room with a table and two chairs. All participants or their parents gave their written informed consent. Human subjects research was approved by the Cantonal Ethics Commission for Human Research (Vaud, Switzerland; project no PB_2017-00074 (60/14)) and was in accordance with the code of ethics of the World Medical Association (Declaration of Helsinki) for experiments involving human subjects in research.

2.1.2 | General procedure

The task, already described in Bochud-Fragnière et al. (2022) and Bochud-Fragnière et al. (2023), was modified from the original WPT (Knowlton et al., 1994) and from the Ice Cream Task (Shohamy et al., 2004) to test young children and individuals with learning disabilities. First, cartoonish sea animals were used as cues instead of tarot cards, and two different smiley faces served as outcomes (Figure 1). Second, to give participants with low mental ages a better chance to complete the task we used only 100 trials and associative outcome probabilities (i.e., .20, .40, .60, .80) used previously (Gluck et al., 2002; Mayor-Dubois et al., 2016). The number of presentations of each pattern was adapted to respect these probabilities (Table 2). The task was programmed with E-Prime® 3.0 software (Psychology Software Tools, Sharpsburg, PA, USA) and was presented on a computer tablet with a tactile screen (HP Spectre x360 Convertible 13-w0XX running with Microsoft© Windows® 10 64-bit; Processor Intel Core i7; 8 GB RAM; 13.3-in (1920 × 1080); landscape orientation). The participant's choice was recorded automatically by E-prime when they touched one of the two outcome stimuli. During the whole experiment, the experimenter sat next to the participant and wore sunglasses to preclude cueing participants with respect to the correct choice with their eye gaze.

2.1.3 | Stimuli and detailed procedure

We used two sets of four cues (cartoonish pictures of sea animals) and the two outcomes (a smiley face with sunglasses enjoying the sun, hereafter referred to as "sun"; a smiley face wearing a knit hat enjoying the snow, hereafter referred to as "snow"; Figure 1) created by Bochud-Fragnière et al. (2022). One set of four cues was pseudo-randomly attributed to each participant. The WPT was divided into two types of trials: learning trials and test trials. For both types, each trial was preceded by a fixation cross displayed for 1 s at the center of the screen. Subsequently, the stimuli appeared. During the learning trials, participants were required to choose which of two outcomes (sun or snow) was predicted by the combination of one to three cues, chosen from amongst the four sea animals that combined to produce 14 unique patterns (Table 2). The instructions were the same for all participants: "You will see some fish, and you will have to discover if they prefer to play in the sun or in the snow." Then, the experimenter or the participant touched the screen to begin the task. The order of the trials and the position of the cues (each cue could appear at one of four possible positions on the screen: left, center left, center right, right) were randomly determined with E-prime. The position of the two outcome images, with snow on the left and sun on the right, was fixed for all trials and all participants (Figure 1).

Participants thus had to learn, using trial-by-trial feedback, which cues were associated with which outcome. Once participants had indicated their choice by touching the screen, a congruent or incongruent outcome image appeared depending on the predetermined probability associated with the cues, while the cues remained visible. The experimenter provided verbal feedback consistent with the visual feedback until participants demonstrated the ability to understand the visual feedback, as evidenced when participants nodded their head or gave themselves verbal feedback: "yes," "right," when the visual feedback fit the answer of the participant; "no," "not this one," "it's no big deal," "oops, no," when the visual feedback did not fit her or his answer. Note that the two last feedback phrases were used to lessen the impact of a

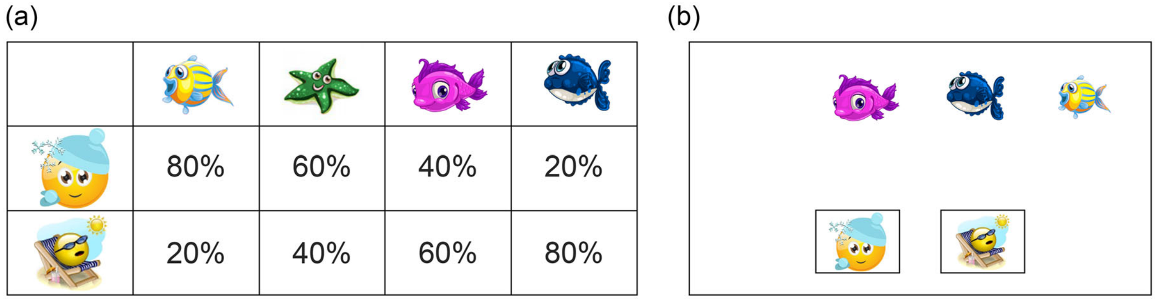

FIGURE 1 (a) Example of one set of four cues (sea animals), which were associated with each outcome (sun or snow) with a fixed level of probability across all patterns in the WPT of Experiment 1. Since there was no difference in performance between sets (data not shown), data obtained with different sets were combined for presentation. All sets of cues were already described (Bochud-Fragnière et al., 2022; Bochud-Fragnière et al., 2023). (b) Representation of the computer screen for one trial when three animals were displayed, and the participant had to choose between snow and sun. Note that one, two, or three animals could be displayed on any given trial; all four animals were never displayed simultaneously.

TABLE 2 Number of occurrences of each pattern and their association with each outcome in the WPT of Experiment 1.

	Cues				Outcome A	Outcome B	
Pattern	1	2	3	4	Snow	Sun	Total number of occurrences
1	0	0	0	1	8	1	9
2	0	0	1	0	4	1	5
3	0	0	1	1	12	1	13
4	0	1	0	0	1	3	4
5	0	1	0	1	5	1	6
6[a]	0	1	1	0	1	2	3
7	0	1	1	1	9	1	10
8	1	0	0	0	1	8	9
9[a]	1	0	0	1	2	2	4
10	1	0	1	0	1	6	7
11	1	0	1	1	2	1	3
12	1	1	0	0	1	12	13
13	1	1	0	1	2	3	5
14	1	1	1	0	1	8	9
Total	7	7	7	7	50	50	100

Note. The probability that each individual cue was associated with each outcome was calculated across all patterns. See main text for details.
[a]Patterns 6 and 9 do not have a correct answer, because they combine either two cues that are highly predictive for sun and snow (pattern 9) or two cues that are less predictive for sun and for snow (pattern 6). They were not included in the analyses, which thus comprised 93 trials instead of 100.

series of negative feedbacks and to maintain participants' motivation. Participants then pressed a red button at the bottom of the screen to advance to the next trial. Note that a participant can make an optimal (correct) prediction but receive incongruent feedback. For example, even though fish X was correlated 80% of the time with sun, 20% of the time when fish X was shown, the snow outcome was displayed on the screen. Therefore, incongruent trials could not be differentiated from congruent trials based on feedback, but rather only by considering the feedback given for each cue cumulatively across trials. The choice was scored as correct if it corresponded to the most probable outcome for that combination of cue(s), regardless of the displayed outcome image. For example, for pattern 0001, the appropriate prediction scored as correct is always outcome A (snow), even if the feedback will be outcome B (sun) on some of the trials (Table 2).

After 100 training trials, four test trials without feedback were presented during which each cue was displayed individually (patterns 0001, 0010, 0100, 1000) in the presence of the two possible outcomes, and participants were asked to choose which outcome was associated with each individual cue. Once participants indicated their choice by touching the outcome image on the screen, the next trial began with the fixation cross. The instructions for the test trials were the same for all participants: "You will see a fish on the computer screen and you will have to recall if it preferred to play in the snow or in the sun." Then, the experimenter or the participant touched the screen to begin the test trials.

For both learning and test trials, the computer recorded the choice of the participant and the reaction time. Since reaction time depends on self-confidence and because limiting the time available to respond could induce impulsivity, the stimuli were presented for as long as needed for the participant to choose one outcome by touching the corresponding image. Although we did not impose a response time inferior to 5 s as was originally done (Knowlton, Mangels, et al., 1996; Knowlton et al., 1994; Knowlton, Squire, et al., 1996), our participants' average response times (WS: $M = 3.77$ s; $SD = 2.52$; DS: $M = 2.61$ s; $SD = 0.82$; Children: $M = 2.59$ s; $SD = 1.15$; Adults: $M = 2.62$ s; $SD = 0.55$) were close to those observed in previous studies. These reaction times showed that our participants were concentrated on the task and that they were not responding impulsively, nor were they taking an unusually long time to respond. Nevertheless, there were differences in the response times of the different groups of participants ($F_{(3,142)} = 5.296$, $p = .002$, $\eta^2_p = .101$). The average response time of individuals with WS was higher than that of young adults ($t_{(42.747)} = 2.696$, $p = .010$, $d_s = 0.562$), TD children ($t_{(45.650)} = 2.722$, $p = .009$, $d_s = 0.668$), and individuals with DS ($t_{(50.136)} = 2.547$, $p = .014$, $d_s = 0.538$).

2.1.4 | Data analysis

We used two-tailed paired-sample t-tests to determine whether participants exhibited a bias toward choosing one of the two outcomes, sun or snow, irrespective of the cues presented. We normalized the number of times outcome A (snow) and outcome B (sun) were chosen by dividing these numbers by the number of times they were presented (i.e., 46 and 47, respectively; the difference is due to the overall distribution of cue and outcome combinations; Table 2). As a group, 20- to 30-year-old adults did not choose "sun" more than "snow" during training trials ($p = .123$) or test trials ($p = .748$; see Supplemental Material S1 for full statistical results) (Bochud-Fragnière et al., 2022). Although, TD children did not choose "sun" more than "snow" during training trials ($p = .374$), there was a tendency to choose "sun" more than "snow" during test trials ($p = .056$). A detailed analysis revealed that only children who failed the task exhibited a slight bias toward choosing "sun" during training ($p = .064$) and test ($p = .020$) trials. In contrast, children who performed above chance level did not exhibit any bias toward choosing one of the two different outcomes during training ($p = .110$) or test ($p = .788$) trials (Bochud-Fragnière et al., 2023). As a group, individuals with DS did not choose "sun" more than "snow" during training ($p = .773$) or test ($p = .817$) trials. In contrast, as a group, individuals with WS chose "sun" more than "snow" during training ($p = .027$) and test ($p = .016$) trials.

Since there was no consistent effect of gender in any of the analyses performed (data not shown), data from males and females were combined for analysis and presentation. To determine whether a group of participants performed above chance during training and test trials, we performed one-sample t-tests comparing the average performance of the group of participants to the theoretical mean. As was done in previous studies (Foerde et al., 2006; Gluck et al., 2002; Knowlton et al., 1994), we defined a passing criterion based on performance during learning trials. Individual above chance performance during training was determined statistically using a chi-square test as 56 correct choices out of 93 ($\chi^2_{(1)} = 3.882$, $p = .049$). Test trials were not used to determine whether a participant solved the WPT because: (1) there were only four trials, and thus insufficient statistical power at the individual level, and (2) they only give information about the pattern containing only one single cue. In contrast, test trials do give information about possible "declarative" information learned by each group of participants regarding single cue–outcome associations. Because TD children's performance varied with age, we used the same age groups as in Bochud-Fragnière et al. (2023) (3–5.5 years ($n = 25$), 5.5–7.5 years ($n = 25$), and 7.5–12 years ($n = 19$)). We separated individuals with WS or DS into similar age groups based on their mental age (WS: 3–5.5 years ($n = 10$), 5.5–7.5 years ($n = 20$) and 7.5–12 years ($n = 7$); NB: the 3- to 5.5-year-old group included three WS participants with a mental age of 5.5 years and one 23-year-old woman with WS did not perform the Leiter-R Brief IQ, she was therefore not included in the analyses that considered the mental age of the individuals; DS: 3–5.5 years ($n = 7$), 5.5–7.5 years ($n = 10$) and 7.5–12 years ($n = 0$)). We performed General Linear Model analyses to compare the number of correct choices made during training trials by participants of each group (i.e., 20- to 30-year-old adults, 3- to 5.5-year-old TD children, 5.5- to 7.5-year-old TD children, 7.5- to 12-year-old TD children, 3- to 5.5-MA DS, 5.5- to 7.5-MA DS, 3- to 5.5-MA WS, 5.5- to 7.5-MA WS, and 7.5- to 12-MA WS). Post hoc analyses were performed with paired-sample t-tests when F ratios were significant, thus controlling for Type I error rate (Carmer & Swanson, 1973). We did not use corrections for multiple comparisons for individual t-tests because in our study the risk of reporting a difference that may not exist (type I error) is not worse than the risk of missing a difference that may exist (type II error). Accordingly, we followed the recommendations of Rothman (1990), who argues that *"not making adjustments for multiple comparisons is preferable because it will lead to fewer errors of interpretation when the data under evaluation are not random numbers but actual observations on nature,"* and Saville (1990), who also argues that a procedure without correction is preferable because it provides greater consistency to compare results between studies. We used chi-square tests, performed on the raw number of participants, to determine whether the number of participants who performed above chance level differed between groups.

All statistical analyses were performed with IBM SPSS statistics (version 27.0). Significance level was set at $p < .05$ for all analyses. We reported effect size with partial eta squared (η^2_p) for ANOVAs, Cohen's d_s for independent samples t-tests, and Cohen's d_z for paired or one-sample t-tests.

2.2 | Results

2.2.1 | Group performance

During training trials, as a group, 20- to 30-year-old adults ($M = 68.95$, $SD = 9.40$, $t_{(21)} = 11.200$, $p < .001$, $d_z = 2.388$), 7.5- to 12-year-old TD children ($M = 64.32$, $SD = 11.84$, $t_{(18)} = 6.558$, $p < .001$, $d_z = 1.504$), and 5.5- to 7.5-year-old TD children ($M = 54.36$, $SD = 13.07$, $t_{(24)} = 3.007$, $p = .006$, $d_z = 0.601$) made more correct choices than if they had chosen randomly. In contrast, 3- to 5.5-year-old TD children ($M = 45.96$, $SD = 3.75$, $t_{(24)} = 0.721$, $p = .478$, $d_z = 0.144$) did not make more correct choices than if they had chosen randomly. Similarly, none of the groups of individuals with WS or DS made more correct choices than if they had chosen randomly (all $p > .170$; Supplemental Material S1). Accordingly, there were differences in performance between groups during training trials ($F_{(8,136)} = 15.210$, $p < .001$, $\eta^2_p = .472$; see Supplemental Material S2, Table S1 for post hoc comparisons).

During tests trials, as a group, 20- to 30-year-old adults ($M = 3.19$, $SD = 0.75$, $t_{(20)} = 7.278$, $p < .001$, $d_z = 1.588$, one participant did not perform the test trials), 7.5- to 12-year-old TD children ($M = 2.89$, $SD = 0.99$, $t_{(18)} = 3.923$, $p < .001$, $d_z = 0.900$), and 5.5- to 7.5-MA individuals with DS ($M = 2.40$, $SD = 0.52$, $t_{(9)} = 2.449$, $p = .037$, $d_z = 0.775$) made more correct choices than if they had chosen randomly. In contrast, no other group made more correct choices than if they had chosen randomly (all $p > .106$; Supplemental Material S1). Accordingly, there were differences in performance between groups during test trials ($F_{(8,135)} = 2.229$, $p = .029$, $\eta^2_p = .117$; see Supplemental Material S2, Table S2 for post hoc comparisons).

In sum, during training trials, as a group, only 20- to 30-year-old adults, 5.5- to 7.5-year-old and 7.5- to 12-year-old TD children performed above chance level. No group of individuals with WS or DS performed above chance level, not even those with a MA greater than 5.5 years of age who might be expected to pass based on their mental age. During test trials, 20- to 30-year-old adults and 7.5- to 12-year-old TD children performed above chance level, whereas 5.5- to 7.5-year-old TD children did not. Surprisingly, 5.5- to 7.5-MA individuals with DS performed slightly above chance level during test trials. However, despite the existence of group differences, interindividual variability within each group warranted a more detailed analysis of performance at the individual level.

2.2.2 | Individual performance

The number of participants who performed the task above chance level (≥ 56 correct choices; $\chi^2_{(1)} \geq 3.882$, $p \leq .049$) differed between groups (Figure 2; $\chi^2_{(8)} = 81.498$, $p < .001$, see Supplemental Material S2, Table S3 for pairwise comparisons). Twenty of the twenty-two 20- to 30-year-old adults (91%) performed the WPT above chance (Supplemental Material S2, Table S4); none of the twenty-five 3- to 5.5-year-old TD children (0%), nine of the twenty-five 5.5- to 7.5-year-old TD children (36%), and fifteen of the nineteen 7.5- to 12-year-old TD children (79%) performed the WPT above chance level (Figure 2; Supplemental Material S2, Table S5). One of the 7 individuals with DS with a mental age between 3 and 5.5 years (14%) and 1 of the 10 individuals with DS with a mental age between 5.5 and 7.5 years (10%) performed the WPT above chance level (Figure 2; Supplemental Material S2, Table S6). None of the 10 individuals with WS with a mental age between 3 and 5.5 years (0%), none of the 20 individuals with WS with a mental age between 5.5 and 7.5 years (0%), and 3 of the 7 individuals with WS with a mental age between 7.5 and 12 years (43%) performed the WPT above chance level (Figure 2; Supplemental Material S2, Table S7).

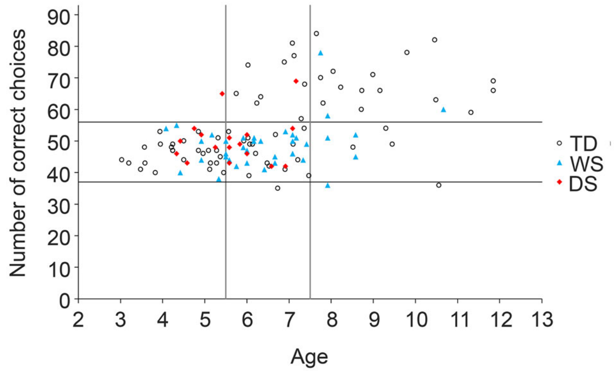

FIGURE 2 Individual performance of 3- to 12-year-old TD children, individuals with WS, and individuals with DS on the training trials of the WPT in Experiment 1. The upper black line represents the number of correct choices (56/93) corresponding to above chance performance at the individual level, whereas the lower line represents the number of correct choices (37/93) defined as below chance performance at the individual level (both $\chi^2_{(1)} = 3.882$, $p = .049$). The x-axis represents chronological age (years) for TD children and mental age (years) for individuals with WS or DS. The two vertical lines separate three different age groups (3–5.5 years, 5.5–7.5 years, and 7.5–12 years). Note that the individual with WS whose mental age was not measured does not appear in the figure but appears in Supplemental Material S2, Table S6.

In sum, 61% of the TD children and 12% of the individuals with DS exhibited the ability to solve the WPT from a mental age of about 5.5 years, whereas three individuals with WS exhibited this ability from a mental age of about 7.5 years (11% of all individuals > 5.5 years of age, or 43% of all individuals > 7.5 years of age). However, it is important to note that not all older TD children or individuals with WS or DS with higher mental ages were able to solve the task.

2.2.3 | Discussion: Experiment 1

Experiment 1 was designed to determine whether individuals with WS or DS can solve the standard WPT. We found that only about 10% of individuals with WS or DS were able to perform above chance level. Most individuals with WS or DS were thus unable to implement any of the learning strategies that can be used to solve the WPT (Bochud-Fragnière et al., 2022; Bochud-Fragnière et al., 2023). These results

TABLE 3 Chronological and mental ages of participants with WS retained in Experiment 2.

		Chronological age (years)		Mental age (years)	
Participants	*n*	Mean	Range	Mean	Range
WS	12	24.9	10.9–44.8	7.2	5.9–10.7

were unexpected given the facilitated striatum-dependent spatial response learning capacities of individuals with WS or DS (Bostelmann et al., 2017; Bostelmann et al., 2018), and the fact that the WPT can be solved using a striatum-dependent procedural learning strategy (Bochud-Fragnière et al., 2022; Gluck et al., 2002; Knowlton et al., 1994). Instead, the results are consistent with the fact that most young healthy individuals use a hippocampus-dependent learning strategy to solve the WPT (Bochud-Fragnière et al., 2022; Foerde et al., 2006), and that several types of hippocampus-dependent learning are impaired in individuals with WS or DS (Banta Lavenex et al., 2015; Bostelmann et al., 2017; Bostelmann et al., 2018; Costanzo, Vicari, et al., 2013; Edgin et al., 2014; Meyer-Lindenberg et al., 2005; Pennington et al., 2003; Vicari et al., 2000). Nevertheless, to ensure that individuals with WS were not capable of employing a striatum-dependent procedural strategy to solve the WPT, we wanted to test participants under conditions that promote the use of such learning strategy.

3 | EXPERIMENT 2: WPT + CONCURRENT MEMORY TASK

Since in Experiment 1 individuals with WS were unable to solve the WPT, we hypothesized that if they were recruiting their hippocampus to solve the task (similar to older TD children and adults), impaired hippocampal function would preclude above chance performance. Accordingly, in Experiment 2, we evaluated whether the recruitment of the hippocampus with a concurrent working memory task (Banta Lavenex et al., 2014; Yonelinas, 2013) could lead to the preferential use of a striatum-dependent learning strategy to solve the WPT, as was previously reported in TD adults (Foerde et al., 2006; Foerde et al., 2007). Since individuals with WS demonstrate greater facilitation of striatum-dependent response learning than individuals with DS in the spatial domain, we tested a subset of participants with WS in a dual-task condition following a procedure designed to promote reliance on a striatum-dependent learning strategy for solving the WPT.

3.1 | Materials and methods

3.1.1 | Participants

A subgroup of 12 individuals with WS (7 females; Table 3) who performed Experiment 1 participated in Experiment 2, including the 3 individuals with WS who solved the WPT in Experiment 1.

3.1.2 | Detailed procedure and data analysis

In Experiment 2, participants had to solve the WPT while performing a concurrent memory task. Testing material and conditions were the same as described for Experiment 1, except that participants tested with stimuli set A in Experiment 1 were tested with stimuli set C in Experiment 2, and vice versa. Since most participants with WS have difficulties with counting, we did not use tone counting as the secondary memory task, as was done previously (Foerde et al., 2006; Foerde et al., 2007). Instead, participants heard pairs of words (one word per second) that contained one word corresponding to a food item and one word corresponding to an object. Participants had 1 s after hearing the two words to repeat the word corresponding to a food item. The order of the words in the pair, food item versus object, was pseudorandom and the pairs of words were fixed for all participants. We did not perform an analysis of the responses to the word memory task, but a second experimenter verified that the participant answered correctly and was thus actually performing the secondary task. Data analysis of the WPT was performed as described in Experiment 1. Individual above chance performance during training was determined statistically using a chi-square test as 56 correct choices out of 93 ($\chi^2{}_{(1)} = 3.882$, $p = .049$).

3.2 | Results

As a group, participants with WS did not make more correct choices than if they had chosen randomly across training trials (M = 47.25, $SD = 2.90$, $t_{(11)} = 0.897$, $p = .389$, $d_z = 0.259$) or test trials ($M = 1.67$, $SD = 0.89$, $t_{(11)} = 1.301$, $p = .220$, $d_z = 0.376$). More importantly, none of the 12 participants with WS performed above chance level ($\geq$ 56 correct choices; all $\chi^2{}_{(1)} \geq 3.882$, $p \leq 0.049$; Supplemental Material S2, Table S8). As a group, participants with WS chose "sun" more often than "snow" during training trials ("sun": $M = 1.31$, $SD = 0.41$; "snow": $M = 0.68$, $SD = 0.42$; $t_{(11)} = 2.633$, $p = .023$, $d_z = 0.760$), but not during test trials ("sun": $M = 2.33$, $SD = 0.65$; "snow": $M = 1.67$, $SD = 0.65$; $t_{(11)} = 1.773$, $p = .104$, $d_z = 0.512$).

3.3 | Discussion: Experiment 2

Although the WPT can be solved using different learning strategies dependent on dissociable memory systems (e.g., Bochud-Fragnière et al., 2022; Foerde et al., 2006; Gluck et al., 2002), Experiment 1 showed that very few individuals with WS were able to solve the task and that most individuals with WS could not employ any successful learning strategy to solve the WPT under normal conditions. We

TABLE 4 Chronological and mental ages of participants retained in Experiment 3.

		Chronological age (years)		Mental age (years)	
Participants	*n*	Mean	Range	Mean	Range
WS	25	27.5	12.7–44.8	7.1	5.5–10.7
DS	14	18.8	7.5–31.9	5.7	4.3–7.2

thus hypothesized that a concurrent hippocampus-dependent memory task may help individuals with WS to solve the WPT using a striatum-dependent learning strategy, as was shown in healthy adults (Foerde et al., 2006; Foerde et al., 2007). Experiment 2 showed that none of the 12 participants with WS tested was able to solve the WPT in the dual-task condition, thus refuting our hypothesis. Note that our subgroup of participants included the three participants who solved the WPT in Experiment 1 and nine participants who did not. Thus, instead of helping individuals with WS to solve the WPT, the concurrent verbal memory task had a detrimental effect on the performance of the three individuals who succeeded in the single-task condition.

Although the results of both experiments are consistent if we consider that the WPT is primarily assessing hippocampus-dependent learning abilities, it is important to determine which aspects of the WPT present difficulties for individuals with WS or DS. Specifically, the probabilistic nature of the WPT and the lack of absolute congruent feedback may negatively impact learning in participants who doubt their understanding of the cue–outcome associations or lack the ability to learn with weak reward contingency ratios. Alternatively, some participants might be unable to distinguish highly predictive and less predictive cues, and thus treat all cues as if they have the same weight. Experiments 3 and 4 aimed to determine whether these factors have a detrimental impact on the performance of individuals with WS or DS on the WPT.

4 | EXPERIMENT 3: VISUAL LEARNING WITH DETERMINISTIC OR PROBABILISTIC FEEDBACK

In Experiment 3, we assessed the influence of probabilistic feedback on the learning abilities of individuals with WS or DS. We tested participants with four variants of a visual learning (VL) task: two deterministic tasks in which the stimuli were either always rewarded or never rewarded (i.e., 100% vs. 0%) and two probabilistic tasks in which the feedback contingencies fit the cue–outcome associations of the WPT (i.e., 80% vs. 20% and 60% vs. 40%).

4.1 | Materials and methods

4.1.1 | Participants

Participants were 25 individuals with WS (15 females) and 14 individuals with DS (6 females), who had participated in Experiment 1 (Table 4). Five additional participants with WS were not included in the present analyses because either the experimenter made a mistake when giving the instructions (*n* = 3; one 22.20-year-old woman, one 23.11-year-old woman, and one 20.58-year-old man), or the participant did not follow the instructions (*n* = 2; one 47.86-year-old woman and one 23.28-year-old woman). All participants with DS were included in the analyses.

4.1.2 | General procedure

A visual learning task was created to test under which reward contingencies participants can learn the nonconditional relationships between two stimuli presented simultaneously. Each pair of stimuli comprised two cartoonish sea animals (Figure 3), and a smiley face served as positive feedback for choosing the correct stimulus. The task included four variants performed successively in which the rewarding probabilities for the stimuli of each pair were: (1) 100% and 0%; (2) 80% and 20%; (3) 60% and 40%; (4) 100% and 0% (Figure 3). All testing material and conditions not specifically mentioned below were the same as described for Experiment 1.

4.1.3 | Stimuli and detailed procedure

We created nine unique pairs of stimuli (Figure 3) and used a smiley face for positive feedback signaling a correct choice and a blank screen for negative feedback signaling an incorrect choice. All participants were tested with the same stimuli. For each variant of the task, each trial was preceded by a fixation cross displayed for 1 s at the center of the screen. Subsequently, a pair of stimuli appeared (either Pair 1 or Pair 2). Each pair was presented for as long as needed for the participant to choose one of the two images by simply touching the image. Each pair was presented 20 times during the task, for a total of 40 trials. The presentation order of the pairs and the positions of the stimulus within each pair (left or right side of the screen) were pseudo-randomly predetermined and fixed for all participants. The instructions were the same for all participants: "You will see two fish, and you will have to discover which one is the winner/stronger." These instructions were given before the first pretraining trial and before the first trial of the first task (VL100%). Between the different tasks, the instructions were also the same for all participants: «Now we will play the same game with another set of fish». Then, the experimenter or the participant touched the screen to begin the task.

Experiment 3 began with two presentations of a pretraining pair of stimuli: The participants saw a pair of fish and they had to discover

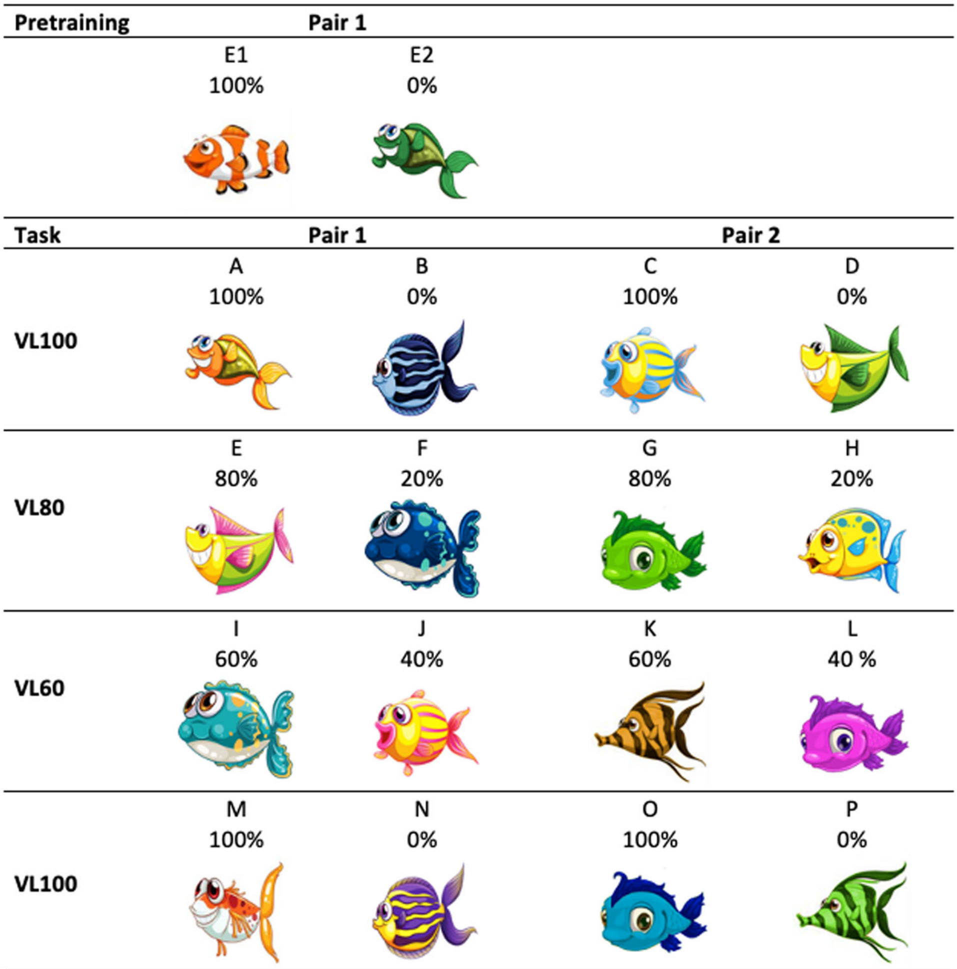

FIGURE 3 Pairs of stimuli used in the pretraining trial or the different variants of the task in Experiment 3. "VL100%" is a classical visual learning task, in which one stimulus of each pair was always associated with a smiley face and considered as the correct stimulus. "VL80%" and "VL60%" are probabilistic visual learning tasks, in which one stimulus of each pair was associated in a probabilistic manner, either 80% or 60% of the trials, with a smiley face and considered as the correct stimulus. No letters or probabilities were shown on the screen. The order of presentation of the pairs and the positions (left-right) of the stimulus within each pair were pseudo-randomly predetermined and fixed for all participants. The correct stimulus for each pair was fixed for all participants.

which one was reinforced. If participants touched the correct stimulus, the pair disappeared, and a smiley face appeared. If participants touched the wrong stimulus, the pair remained visible until they chose the correct stimulus, after which the pair disappeared and a smiley face appeared. The participants had to press a red button at the bottom of the screen to start the next trial. After two correct answers on the pretraining trial, the experimenter repeated the instructions, and she or the participant touched the screen to begin the first task.

During the two deterministic visual learning tasks (VL100% tasks, which were the first and the last tasks in the series), if participants chose the correct stimulus the pair of stimuli disappeared, and a smiley face appeared. If they chose the wrong stimulus, the screen turned blank. Thus, the feedback was always 100% congruent or deterministic (Ashby & Maddox, 2005; Atallah et al., 2008). The experimenter provided verbal feedback consistent with the visual feedback until participants demonstrated the ability to understand the visual feedback (when participants nodded their head or gave verbal feedback themselves). The participants then pressed a red button at the bottom of the screen to advance to the next trial. Verbal feedback was used to ensure that participants understood the goal of the task and understood that the same fish was always the winner. Furthermore, additional guidance was given before and during the first 20 trials of the first VL100% task only: Before the task began, the experimenter said, "I will tell you a secret. When you see these two fish, this fish (experimenter points to fish) always wins. Always." During the first 20 trials of the first VL100% task (i.e., the first 10 presentations of each pair), the participant was instructed to first show which fish they wanted to touch and were instructed to wait for the approval of the experimenter before touching it. If the participant showed the correct stimulus, the experimenter said, "yes" or "go." If the participant showed the incorrect stimulus, the experimenter said, "No, never this one" and then said, "Always this one," while pointing to the correct fish. After the first 20 trials, the experimenter told the participant that they could do it alone, without waiting for approval to make a choice. Therefore, from the 21st trial on, the experimenter only gave the verbal feedback corresponding to the visual feedback when it appeared. This method of instruction and training was adopted since we had observed in a previous experiment employing the VL100% task that some young children and individuals with WS or DS interpreted each trial as a new and unique trial in which the "winner" could not be determined based on the results of previous trials. However, this did not mean that the participant did not or could not remember which fish was the winner on the previous trial, only that they considered the different trials as being unrelated. By implementing this instructional and training procedure at the beginning of the task we could ensure that participants understood, and our results showed that these individuals were indeed capable of learning and remembering the winner under this specific reward contingency.

During the two probabilistic visual learning tasks (VL80% and VL60%), participants received visual and verbal feedback that fit the reinforcement contingencies of the stimuli. During the VL80% task, stimuli E and G were reinforced 80% of the time, whereas stimuli F and H were reinforced 20% of the time. During the VL60% task, stimuli I and K were reinforced 60% of the time, whereas stimuli J and L were reinforced 40% of the time. A choice was scored as correct if it corresponded to the stimulus with the highest reinforcement contingency of the pair (i.e., stimuli E, G, I, and K), regardless of the feedback (Figure 3). Thus, similar to the WPT in Experiments 1 and 2, participants can make an optimal (correct) choice but receive incongruent feedback.

4.1.4 | Data analysis

To determine whether a group of participants performed above chance level during each task, we used one-sample *t*-tests to compare the average number of correct choices in the last 20 trials of each task with the theoretical mean. We did not perform these analyses including all trials, since our instructions precluded error during the first 20 trials of the first VL100% task. Individual above chance performance during training was defined as 15 correct choices out of 20 trials ($\chi^2_{(1)} \geq 5.00$, $p \leq .025$). Note that a participant could perform above chance on a task by performing above chance on only one of the two pairs. We also used chi-square tests to determine whether the number of participants who performed above chance level differed between WS and DS. Since there was no effect of gender, data from males and females were combined for analysis and presentation. All statistical analyses were performed with IBM SPSS statistics (version 27.0). Significance level was set at $p < .05$ for all analyses. Chi-square tests were performed on the raw number of participants. We reported effect size with Cohen's d_z for one-sample *t*-tests.

4.2 | Results

4.2.1 | Group and individual performance

The percentages of individuals with WS or DS who performed above chance, as a group and by age group and reward contingency, are shown in Table 5. As a group, participants with WS made more correct choices than if they had chosen randomly in the last 20 trials of the first VL100% task ($M = 19.08$; $SD = 2.14$; $t_{(24)} = 21.222$, $p < .001$, $d_z = 4.244$), the VL80% task ($M = 13.16$; $SD = 4.92$; $t_{(24)} = 3.210$, $p = .004$, $d_z = 0.642$) and the last VL100% task ($M = 15.60$; $SD = 5.01$; $t_{(24)} = 5.591$, $p < .001$, $d_z = 1.118$), but not in the last 20 trials of the VL60% task ($M = 9.28$; $SD = 3.64$; $t_{(24)} = 0.990$, $p = .332$, $d_z = 0.198$). Similarly, as a group, participants with DS made more correct choices than if they had chosen randomly in the last 20 trials of the first VL100% task ($M = 19.79$; $SD = 0.58$; $t_{(13)} = 63.245$, $p < .001$, $d_z = 16.903$), the VL80% task ($M = 14.93$; $SD = 3.67$; $t_{(13)} = 5.027$, $p < .001$, $d_z = 1.344$) and the last VL100% task ($M = 17.79$; $SD = 2.86$; $t_{(13)} = 10.185$, $p < .001$, $d_z = 2.722$), but not in the last 20 trials of the VL60% task ($M = 9.43$; $SD = 3.57$; $t_{(13)} = 0.599$, $p = .559$, $d_z = 0.160$). The number of participants who performed the different tasks above chance did not differ between the two groups (see detailed statistics in Supplemental Material S1 and individual results in Supplemental Material S2, Tables S9 and S10).

We further compared the number of participants who passed the different tasks within each group. There were differences in the number of participants with WS (irrespective of mental age) who performed the different tasks above chance level ($\chi^2_{(3)} = 38.215$, $p < .001$). More participants with WS performed above chance the first VL100% task than the VL80% task ($\chi^2_{(1)} = 15.062$, $p < .001$), the VL60% task ($\chi^2_{(1)} = 35.280$, $p < .001$), and the last VL100% task ($\chi^2_{(1)} = 5.711$, $p = .017$). More participants with WS performed above chance the VL80% task than the VL60% task ($\chi^2_{(1)} = 7.018$, $p = .008$). More participants with WS performed above chance the last VL100% than the VL60% task ($\chi^2_{(1)} = 17.014$, $p < .001$), but not the VL80% task ($\chi^2_{(1)} = 2.885$, $p = .089$).

There were also differences in the number of participants with DS who performed the different tasks above chance level ($\chi^2_{(3)} = 27.963$, $p < .001$). More participants with DS performed above chance the first VL100% task than the VL80% task ($\chi^2_{(1)} = 9.333$, $p = .002$), the VL60% task ($\chi^2_{(1)} = 24.267$, $p < .001$), but not the last VL100% task ($\chi^2_{(1)} = 3.360$, $p = .067$). More participants with DS performed above chance the VL80% task than the VL60% task ($\chi^2_{(1)} = 6.300$, $p = .012$). More participants with DS performed above chance the last VL100% task than the VL60% task ($\chi^2_{(1)} = 14.583$, $p < .001$), but not the VL80% task ($\chi^2_{(1)} = 2.489$, $p = .115$).

We also evaluated whether the number of participants with WS who performed above chance level was different between the 5.5–7.5 years and 7.5–12 years mental age groups. There was no difference between mental age groups in the number of participants with WS who passed the first VL100%, the VL60% and the last VL100% ($\chi^2_{(1)} = 0.845$, $p = .358$, $\chi^2_{(1)} = 0.521$, $p = .470$ and $\chi^2_{(1)} = 1.990$, $p = .158$, respectively). However, there was a difference between mental age groups in the number of participants with WS who performed above chance level in the VL80% ($\chi^2_{(1)} = 4.001$, $p = .045$).

There was no difference between the number of participants with DS who performed above chance level between the 3.5–5.5 years and 5.5–7.5 years mental age groups in the first VL100%. There was no difference between mental age groups in the number of participants

TABLE 5 Percentage of participants performing each task above chance in Experiment 3.

		Tasks			
	N	VL100%	VL80%	VL60%	VL100%
Participants with DS	14	100%	50%	7%[a]	79%
MA: 3–5.5	5	100%	40%	20%[a]	40%
MA: 5.5–7.5	9	100%	56%	0%	100%
Participants with WS	25	92%	40%	8%[a]	64%
MA: 5.5–7.5	18	89%	28%	6%[a]	56%
MA: 7.5–12	7	100%	71%	14%	86%

[a]Only one participant in each (sub)group performed the VL60% task above chance level.

with DS who passed the VL80% and the VL60% ($\chi^2_{(1)} = 0.311$, $p = .577$ and $\chi^2_{(1)} = 1.938$, $p = .164$, respectively). However, there was a difference between mental age groups in the number of participants with DS who performed above chance level in the last VL100% ($\chi^2_{(1)} = 6.873$, $p = .009$).

In sum, nearly all participants with WS or DS were able to solve a visual learning task when the feedback was deterministic (100% congruence). In contrast, only about half of these participants were able to solve the visual learning task when the feedback was probabilistic with high congruence (80%), and most participants with WS or DS failed to solve the visual learning task when the feedback was probabilistic with low congruence (60%). Importantly, a majority of participants with WS or DS, but not all, were again able to solve the visual learning task when the feedback returned to deterministic (100% congruence), after having been tested in two conditions with probabilistic feedback. Finally, the proportion of individuals that passed or failed was influenced by mental age and syndrome, with lower mental age participants performing worse than higher mental age participants, and lower mental age participants with DS performing equivalently to higher mental age participants with WS.

4.3 | Discussion: Experiment 3

Experiment 3 was designed to determine whether probabilistic feedback, a predominant feature of the WPT, may impact the learning abilities of individuals with WS or DS. We assessed whether individuals with WS or DS were able to solve deterministic or probabilistic variants of a visual learning task. As expected, the reward contingencies impacted the performance of most participants with WS or DS. Most participants with WS or DS were able to learn the deterministic relationships between two stimuli presented simultaneously, especially when they were guided in their initial learning. In contrast, about 50% of participants with WS or DS were not able to learn the information with incongruent feedback, even when it was highly predictive (i.e., 80% congruence). This capacity was more severely impaired when the feedback was less predictive (i.e., 60% congruence). Our results highlight the importance of congruent feedback to facilitate learning in individuals with WS or DS. They further suggest that only half of the participants with WS or DS may be able to use the information provided by the highly predictive cues to solve the WPT, since they have a similar 80% reward contingency. In contrast, few individuals with these syndromes may be able to use the information provided by less predictive cues to solve the WPT. Therefore, individuals with these syndromes may not be able to rely on the procedural multicue strategy or any strategy, which requires consideration of the less predictive cues to solve the WPT. However, several strategies that may be used to perform the task above chance level do not require participants to consider all the cues (Bochud-Fragnière et al., 2022; Bochud-Fragnière et al., 2023; Gluck et al., 2002; Meeter et al., 2006). For example, it is possible to perform the WPT above chance by associating only the two highly predictive cues with their respective outcomes and responding uniquely on this basis. Indeed, 5.5- to 7.5-year-old TD children, in contrast to 7.5- to 12-year-olds and 20- to 30-year-old adults, exhibit above chance performance but only consider the two highly predictive cues and neglect the two less predictive cues (Bochud-Fragnière et al., 2023). Since, as a group, individuals with WS or DS do not solve the WPT using the two highly predictive cues, we hypothesized that it was perhaps because they are not capable of learning that some cues are highly predictive.

5 | EXPERIMENT 4: DETERMINISTIC WPT WITH UNPREDICTIVE CUES

In Experiment 4, we assessed whether individuals with WS or DS could solve a modified WPT-like task in which two cues were deterministic (100% congruence) and two cues were unpredictive (50% chance of being associated with either outcome). We hypothesized that the unpredictive cues would serve as distractors, preventing some individuals with WS and DS from recognizing the predictive nature of the two predictive cues.

5.1 | Materials and methods

5.1.1 | Participants

Participants were 27 individuals with WS (16 females) and 7 individuals with DS (1 female), who participated in Experiment 1 (Table 6).

TABLE 6 Chronological and mental ages of participants retained in Experiment 4.

		Chronological age (years)		Mental age (years)	
Participants	*n*	Mean	Range	Mean	Range
WS	27	28.2	12.8–47.9	6.8	4.3–10.7
DS	7	22.3	9.9–32.6	5.8	4.4–7.2

TABLE 7 Number of occurrences of each pattern and their association with each outcome in the deterministic classification learning task of Experiment 4.

	Cues				Outcome A	Outcome B	
Pattern	1	2	3	4	Snow	Sun	Total number of occurrences
1	0	0	0	1	12	0	12
2	0	0	1	1	12	0	12
3	0	1	0	1	12	0	12
4	0	1	1	1	12	0	12
5	1	0	0	0	0	12	12
6	1	0	1	0	0	12	12
7	1	1	0	0	0	12	12
8	1	1	1	0	0	12	12
Total	4	4	4	4	48	48	96

The COVID-19 pandemic prevented us from testing more participants with DS in a timely manner, and we decided not to resume testing after a 2-year hiatus. All testing material and conditions not specifically mentioned below were the same as described for Experiment 1.

5.1.2 | Procedure

Participants were tested with a deterministic classification learning task in which two cues predicted the outcome in a deterministic manner (100% congruence) and two other cues did not predict the outcome (50% chance of being associated with either outcome; Supplemental Material S2, Figure S1). The number of presentations of each pattern was adapted to respect these association probabilities and only the patterns that were associated coherently with one particular outcome were presented (Table 7). The rest of the procedure was the same as described for the standard WPT in Experiment 1.

5.1.3 | Data analysis

Individual above chance performance during training was defined as 58 correct choices out of 96 trials ($\chi^2_{(1)} = 4.167, p = .041$). Only individuals with WS that did not perform above chance level exhibited a slight bias for "sun" as compared to "snow" in the training trials ("sun": $M = 56.19$, $SD = 18.49$; "snow": $M = 39.81$, $SD = 18.49$; $t_{(15)} = 1.771$, $p = .097$, $d_z = 0.443$; see Supplemental Material S1 for all other analyses). Since there was no effect of gender, data from males and females were combined for analysis and presentation. Because the performance of participants with WS or DS varied with mental age in the standard WPT in Experiment 1, we separated participants into three mental age groups (WS: 3–5.5 years (n = 5), 5.5–7.5 years (n = 16), and 7.5–12 years (n = 6); *NB*: the 3–5.5 years group includes two WS participants with a mental age of 5.5 years; DS: 3–5.5 years (n = 3), 5.5–7.5 years (n = 4) and 7.5–12 years (n = 0)). All statistical analyses were performed as described for the previous experiments.

5.2 | Results

As a group, 3- to 5.5-MA participants with WS did not make more correct choices than if they had chosen randomly during training trials ($M = 54.00$; $SD = 10.32$; $t_{(4)} = 1.300$, $p = .263$, $d_z = 0.581$; Figure 4 and Supplemental Material S2, Table S11) or test trials ($M = 1.20$; $SD = 0.84$; $t_{(4)} = 0.535$, $p = .621$, $d_z = 0.239$). Indeed, no individual with WS in this MA group exhibited above chance performance during training trials. In contrast, 5.5- to 7.5-MA participants with WS made more correct choices than if they had chosen randomly during training trials ($M = 57.44$; $SD = 13.46$; $t_{(15)} = 2.805$, $p = .013$, $d_z = 0.701$) but not test trials ($M = 1.31$; $SD = 0.79$; $t_{(15)} = 1.576$, $p = .136$, $d_z = 0.394$), and 7.5- to 12-MA participants with WS made more correct choices than if they had chosen randomly during training trials ($M = 83.50$; $SD = 19.90$; $t_{(5)} = 4.370$, $p = .007$, $d_z = 1.784$) and all six participants with WS had the maximum of two correct choices during test trials ($M = 2.00$; $SD = 0.00$).

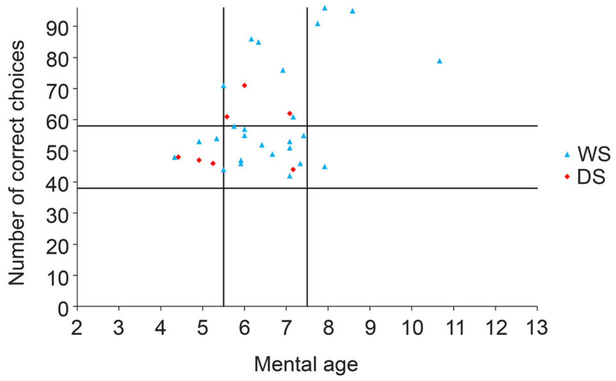

FIGURE 4 Individual performance of participants with WS and DS during training trials of the WPT with 100% congruence and unpredictive cues in Experiment 4. The upper black line represents the number of correct choices defined as individual above chance performance (58/96; $\chi^2_{(1)} = 4.167$, $p = .041$). The lower line represents the number of correct choices defined as individual below chance performance (38/96; $\chi^2_{(1)} = 4.167$, $p = .041$). Vertical lines separate mental age groups (3–5.5 years, 5.5–7.5 years, and 7.5–12 years). Note that two participants with WS with the same mental age (i.e., 8.58 years) performed similarly with a score at 95 and their data are thus overlapping on the graph (Supplemental Material S2, Table S11).

As a group, 3- to 5.5-MA participants with DS did not make more correct choices than if they had chosen randomly during training trials ($M = 47.00$; $SD = 1.00$; $t_{(2)} = 1.732$, $p = .225$, $d_z = 1.000$; Figure 4 and Supplemental Material S2, Table S12) or test trials ($M = 1.67$; $SD = 0.58$; $t_{(2)}$ 2.000, $p = .184$, $d_z = 1.155$). Although 5.5- to 7.5-MA participants with DS did not make more correct choices than if they had chosen randomly during training trials ($M = 59.50$; $SD = 11.27$; $t_{(3)} = 2.041$, $p = .134$, $d_z = 1.020$), all four participants with DS had the maximum of two correct choices during test trials ($M = 2.00$; $SD = 0.00$).

Finally, we compared the number of individuals with WS or DS that performed above chance in this deterministic WPT with unpredictive cues with those that performed above chance in the standard WPT. More individuals with WS with a MA above 5.5 years performed above chance in the current task (11 of 24) than in the standard WPT (3 of 31; $\chi^2_{(1)} = 9.319$, $p = .002$). Similarly, more individuals with DS with a MA above 5.5 years performed above chance in the current task (3 of 4) than in the standard WPT (1 of 10; $\chi^2_{(1)} = 5.915$, $p = .015$).

5.3 | Discussion: Experiment 4

Experiment 4 was designed to determine whether the presence of unpredictive cues impacts the learning abilities of individuals with WS or DS. We assessed whether individuals with WS or DS were able to solve a modified version of the WPT in which the cue–outcome associations were either deterministic (100% congruence) or unpredictive. We found that the presence of unpredictive cues is particularly detrimental for participants with lower mental ages (i.e., 3- to 5.5-MA individuals with WS or DS) and for 50% of 5.5- to 7.5-MA individuals with WS, who may not be able to either recognize the predictive cues or ignore unpredictive or meaningless cues. In contrast, more than a third of WS with a mental age above 5.5 years benefited from the deterministic nature of the task, and more individuals with WS or DS passed this modified WPT than the standard WPT, suggesting that the probabilistic nature of the reward contingencies hindered their performance in the standard WPT.

6 | GENERAL DISCUSSION

The current descriptive study was designed first and foremost to determine whether and how individuals with WS or DS solve the WPT, a probabilistic classification learning task that can be solved using different strategies that depend on distinct learning and memory systems. Based on our findings, we then assessed whether a concurrent hippocampus-dependent memory task could promote reliance on a striatum-dependent learning strategy to solve the WPT in a subgroup of individuals with WS. To elucidate factors that may contribute to the inability of individuals with WS or DS to solve the WPT, we further assessed the impact of probabilistic reward contingencies and unpredictive cues (see summaries of individual performance in Supplemental Material S2, Tables S13 and S14).

In Experiment 1, only about 10% of individuals with WS or DS were able to solve the standard WPT. TD children exhibited this capacity from 5.5 years of age, while only two individuals with DS with a mental age above 5.5 years and three individuals with WS with a mental age above 7.5 years exhibited this capacity. As a group, 5.5- to 7.5-MA individuals with WS were severely impaired, whereas 7.5- to 12-MA individuals with WS and 5.5- to 7.5-MA individuals with DS were somewhat less impaired, as compared to TD children in the same mental age range.

In Experiment 2, a concurrent memory task failed to drive individuals with WS to use a procedural learning strategy to solve the WPT. None of the 12 individuals with WS was able to solve the WPT in the dual-task condition. Instead, the concurrent verbal memory task had a detrimental effect on the performance of the three individuals with WS who succeeded in the single-task condition in Experiment 1.

In Experiment 3, probabilistic feedback had a negative impact on the performance of individuals with WS or DS in a simple visual learning task. More than 90% of individuals with WS or DS were able to learn the deterministic relationships between two stimuli presented simultaneously, especially when they were guided in their initial learning. In contrast, only about 50% of the individuals with WS or DS were able to learn which stimuli were rewarded when incongruent feedback was provided, even if it was highly predictive (i.e., 80% congruence). Less than 10% of individuals exhibited some evidence of learning when the feedback was less predictive (i.e., 60% congruence).

In Experiment 4, unpredictive cues were also found to impact the ability of individuals with WS or DS to select and act upon predictive information in a modified WPT-like task. Interestingly, however, more individuals with WS or DS were able to solve this task than the standard WPT, suggesting that these individuals were primarily hindered by probabilistic reward contingencies. Although some individuals from both groups with a mental age of about 5.5 years were able to solve the task, not all participants with this mental age or higher demonstrated this ability. Nevertheless, individuals with lower mental ages were more impacted by the presence of unpredictive cues than individuals with higher mental ages.

In sum, most individuals with WS or DS were unable to use any of the learning strategies that can lead to above chance performance in the WPT (e.g., Bochud-Fragnière et al., 2022). Moreover, the findings of all four experiments are consistent with learning impairments arising from deficits in hippocampus-dependent and executive functions in WS and DS, and reveal the importance of congruent feedback, explicit instructions, and the minimization of distractors to optimize learning in individuals with WS or DS.

6.1 | Comparison with previous studies

The current study revealed that most individuals with WS or DS are unable to solve the standard WPT and identified different factors that may impact performance on this task. Previous studies have investigated the ability of TD children to solve the WPT (Bochud-Fragnière et al., 2023; Mayor-Dubois et al., 2016). Whereas Mayor-Dubois et al. (2016) only tested children from 8 years of age, Bochud-Fragnière et al. (2023) tested children from 3 years of age. Our results are consistent with the findings from this latter study and show that atypically developed individuals with a mental age inferior to 5.5 years also fail to solve the WPT. Our findings that the ability to solve the WPT evolves with mental age in individuals with WS or DS is also consistent with the study by Bochud-Fragnière et al. (2023), which found that the one third of 5.5- to 7.5-year-old TD children who performed above chance relied on the two most predictive cues and neglected the two less predictive cues. In contrast, about 80% of 7.5- to 12-year-old TD children could solve the WPT, and similar to 20- to 30-year-old adults, they relied on a hierarchical strategy: They considered the highly predictive cues primarily and the less predictive cues secondarily (Bochud-Fragnière et al., 2022).

In the current study, we were unable to perform the pattern analyses proposed by Bochud-Fragnière et al. (2022) to determine which strategy was used by the different groups of participants because too few individuals with WS or DS performed above chance level. Nor did we perform the strategy analyses developed by Gluck and colleagues (Gluck et al., 2002; Meeter et al., 2006), because the goodness of fit of different strategies cannot be distinguished statistically to infer the specific learning strategies used to solve the WPT (Bochud-Fragnière et al., 2022; Bochud-Fragnière et al., 2023). Nevertheless, our findings that neither individuals with WS nor individuals with DS, even those with higher mental ages, can solve the probabilistic visual learning task with low congruence (i.e., 60%) are consistent with the fact that 5.5- to 7.5-year-old TD children do not rely on the less predictive cues (that are also only predictive 60% of the time) to solve the WPT (Bochud-Fragnière et al., 2023). Moreover, the fact that most individuals with DS with a mental age of 5.5 years or higher are able to select meaningful information and ignore distractors in the deterministic WPT with unpredictive cues (Experiment 4) is consistent with the two predictive cues strategy used by 5.5- to 7.5-year-old TD children. Finally, the fact that the mental age at which individuals with WS exhibited this capacity is higher than for individuals with DS (i.e., 7.5–12 years vs. 5.5–7.5 years, respectively) further demonstrates differences in the cognitive profiles of WS and DS. Although we acknowledge that the tasks described in Experiments 3 and 4 could also be used to determine why most 3- to 7.5-year-old and some older TD children fail the standard WPT, this would go beyond the aim of the current study.

The current study was designed in the context of previous work, which demonstrated impaired hippocampus-dependent place learning and enhanced striatum-dependent spatial response learning in WS and DS (Bostelmann et al., 2017; Bostelmann et al., 2018), and aimed to determine whether individuals with these syndromes exhibit similar cognitive profiles outside the spatial domain. In a series of studies, Bostelmann and colleagues showed that the performance of individuals with WS in a low-resolution place learning task with vision and a low-resolution cognitive mapping task without vision was severely impaired (Bostelmann et al., 2017; Bostelmann et al., 2018; Bostelmann et al., 2020). In contrast, individuals with WS exhibited facilitated striatum-dependent spatial response learning abilities as compared to TD children. Similarly, although individuals with DS exhibited relatively preserved low-resolution place learning and cognitive mapping, they exhibited impaired hippocampus-dependent spatial pattern separation and facilitated striatum-dependent spatial response learning, as compared to TD children. Because the WPT can be solved using a procedural learning strategy thought to depend on the striatum (Bochud-Fragnière et al., 2022; Gluck et al., 2002; Knowlton et al., 1994), the inability of most individuals with WS and DS to perform the WPT was unexpected.

Previous experiments had shown that a concurrent working memory task can promote the use of a striatum-dependent habit learning strategy in healthy young adults (Foerde et al., 2006; Foerde et al., 2007), but see (Newell et al., 2007). Moreover, there was a correlation between medial temporal lobe activity and explicit knowledge in the single-task condition, whereas there was a correlation between striatal activity and overall performance in the dual-task condition (Foerde et al., 2006). Considering these results, we tested whether a secondary working memory task might promote the use of a striatum-dependent learning strategy and improve the performance of individuals with WS on the WPT. Instead, we found that none of the 12 participants with WS tested in the dual-task condition was able to solve the WPT. Moreover, rather than improving the performance of individuals with WS via the facilitation of striatum-dependent learning, the concurrent verbal memory task had a detrimental effect on the performance of the three participants who succeeded in the single-task condition. Thus, in agreement with previous findings (Bochud-Fragnière et al., 2022; Foerde et al., 2006; Foerde et al., 2007; Hopkins et al., 2004; Lagnado et al., 2006; Newell et al., 2007; Price, 2009), our results are consistent with the perspective that the standard WPT engages primarily the hippocampus-dependent declarative memory system. Accordingly, based on their spatial memory profile (Banta Lavenex et al., 2015; Bostelmann et al., 2017; Bostelmann et al., 2018; Edgin et al., 2014;

Pennington et al., 2003; Vicari et al., 2000), performance on the standard WPT should be impaired in individuals with DS and even more so in individuals with WS. Indeed, we found that individuals with WS were more impaired than individuals with DS in the same mental age range: the youngest individual with DS who performed above chance level was about 5.5 years of mental age, whereas no individual with WS under 7.5 years of mental age performed the task above chance level. It is interesting to note that individuals with WS were more impaired than individuals with DS despite testing conditions that may potentially facilitate the performance of individuals with WS, as compared to individuals with DS, considering the "hyper social" phenotype of individuals with WS (Jones et al., 2000; Kozel et al., 2021; Martens et al., 2008) and the motivational effect of social interaction on individuals with WS (Kleberg et al., 2023), such as the verbal questions of the experimenter, the verbal feedback and encouragement of the experimenter, and the smiley face as a visual feedback. Moreover, if both the WPT and the concurrent memory task recruit the hippocampal memory system (Newell et al., 2007), it is not surprising that participants with WS demonstrated even greater impairment under dual-task conditions (Banta Lavenex et al., 2014; Olton et al., 1979). Altogether, our findings suggest that impaired hippocampal function may underlie, at least in part, the inability of individuals with WS or DS to solve the WPT, a task that normally depends on proper hippocampal function. Nevertheless, we further investigated specific elements of the WPT, which could impact the performance of individuals with WS or DS.

6.2 | Factors impacting WPT performance in individuals with WS or DS

Several factors may have contributed to individuals with WS or DS failing the WPT. First, we found that one individual with WS alternated systematically between the two outcomes (sun and snow; data not shown). Second, we found that individuals with WS that did not solve the task tended to choose the "sun" outcome irrespective of the cues present on the screen. Whereas this may reveal a bias for these individuals for the "sun" outcome, it may also be because the "sun" outcome was always presented on the right side of the screen and 82% of the individuals with WS used their right hand to perform the task. Although we could have counterbalanced the positions of the outcomes to control for such a potential bias, this might have increased errors due to inattention when choosing the outcome. Nonetheless, 76% of the individuals with DS also used their right hand to perform the task, but as a group, they did not exhibit a bias for choosing the "sun" outcome. Third, we identified two intrinsic components of the WPT, namely, probabilistic feedback and the presence of less predictive cues, which likely impacted the performance of individuals with WS and DS. We designed two experiments to test the influence of these factors on individuals with WS or DS.

6.3 | Probabilistic feedback

Our results indicated that individuals with WS or DS who passed the WPT did not rely on the procedural multicue strategy, nor any of the other strategies that require participants to take into consideration the reward contingencies of both the highly and the less predictive cues. In Experiment 3, we tested whether participants with WS or DS could perform a visual learning (VL) task in which they had to learn which stimulus of a pair was rewarded when the reward contingencies varied sequentially from 100%, to 80%, to 60% and then back to 100%. About half of our participants with WS or DS performed the VL80% task above chance level, indicating that they may be able to rely on the highly predictive cues to solve the standard WPT. In contrast, less than 10% of our participants with WS or DS were able to perform the VL60% task above chance level. Although this may be taken to indicate that these participants can rely on the less predictive cues to solve the WPT, if we consider their results in more detail, we find that the two participants with WS and the one participant with DS who performed the VL60% task above chance level did not perform the standard WPT above chance. Moreover, the facts that the participant with DS who performed the VL60% task above chance level did not perform above chance for either pair of stimuli analyzed separately and that he did not perform the VL80% task above chance together suggest that this is a spurious finding that does not refute our interpretation that individuals with DS are unable to rely on the less predictive cues to solve the standard WPT. Although we were not able to perform the pattern analyses proposed by Bochud-Fragnière et al. (2022) due to the low number of successful participants, our visual learning task with varying reward contingencies (Experiment 3) was nonetheless informative about how individuals with WS or DS could solve the standard WPT. Our data suggest that the few participants with WS or DS who were successful at the WPT relied primarily on the highly predictive cue(s).

According to Bochud-Fragnière et al. (2022), strategies that are based on relying on one or two of the highly predictive cue(s) can enable above chance performance. Moreover, they found that the majority of successful 5.5- to 7.5-year-old TD children use the two most predictive cues strategy to solve the WPT (Bochud-Fragnière et al., 2023). The use of this strategy by individuals with WS or DS would be consistent with the behavior of the younger TD children who solved the WPT. Anecdotally, the three participants with WS who passed the standard WPT above chance level also performed the probabilistic VL80% task above chance level; the one participant with DS who performed the WPT above chance level did not. Indeed, the three successful participants with WS all exhibited the same performance profile, succeeding on the standard WPT, the VL100% task, the VL80% task and the deterministic WPT with unpredictive cues. In contrast, not all participants with WS or DS who solved the probabilistic visual learning task solved the standard WPT, indicating that we cannot predict an individual's ability to perform the WPT based on their ability to perform its separate components.

6.4 | Unpredictive cues

The use of the two most predictive cues strategy requires the ability to learn the valence of one or both of the highly predictive cues, while at the same time learning that the less predictive cues are unreliable and subsequently ignoring them. A previous study showed that 5.5- to 7.5-year-old TD children used the two most predictive cues strategy and neglected the two less predictive cues to solve the standard WPT (Bochud-Fragnière et al., 2023). In fact, there are several strategies that do not require taking all the cues into consideration to perform above chance on the standard WPT (e.g., the two most predictive cue or one-cue strategies). However, individuals who use such strategies must be able to ignore one or more other cues.

Our current results show that the performance of many individuals with WS or DS is impacted by the presence of unpredictive cues, especially individuals with lower mental ages. None of the individuals with WS or DS with a mental age lower than 5.5 years was able to solve the deterministic WPT where two cues predicted the outcome 100% of the time, and two other cues were unpredictive. Accordingly, the design of the WPT, which includes four cues, may require cognitive capacities that are typically not available to individuals with WS or DS with lower mental ages. In contrast, for several individuals with WS or DS and a mental age higher than 5.5 years who did not solve the standard WPT, rendering two of the cues deterministic (100% congruence) enabled them to solve the deterministic WPT. Thus, for these individuals with WS and DS, the major impediment to solving the WPT appears to be its probabilistic nature.

In sum, although we cannot identify all the factors that led individuals with WS or DS to fail the WPT, what we can conclude is that they were not able to identify and implement any of the learning strategies that can lead to successful WPT performance. Moreover, the combination of probabilistic feedback and less predictive cues in the WPT likely makes it impossible for most individuals with WS or DS to solve this task. In the real world, predicting the actual weather or the numerous possible outcomes of a single event, or understanding that the same word can have different meanings, or that some behaviors are okay in one context but not another, are likely very difficult for these individuals. Our current experiments demonstrate the importance of congruent feedback, explicit instructions, and the minimization of distractors to optimize learning in individuals with WS or DS.

6.5 | Cognitive processes implicated in solving the WPT

The WPT is a complex task that likely implicates several cognitive processes simultaneously. To solve the WPT, participants must use trial-by-trial feedback to learn which cues are predictive of the outcome and then commit this information to memory for use in future trials. Participants must also be able to ignore incongruent trials, when a cue that is usually highly predictive for one outcome is followed by the opposite outcome.

To use trial-by-trial feedback, participants must solicit executive functions supported by the prefrontal cortex (Diamond, 2013; Miller & Wallis, 2009; Milner, 1963; Stuss & Benson, 1984) such as sustained and selective attention to focus on the different cues on each trial, as well as hippocampus-dependent working memory (Banta Lavenex et al., 2014; Yonelinas, 2013). Participants also need to solicit planning and cognitive flexibility to test hypotheses about the outcome in prospective trials and to continue learning the rules and applying previously learned rules across the trials. Finally, participants must also be able to inhibit responding to the presence of a highly predictive cue before they have assessed all the cues present on that trial. Previous research has shown that individuals with WS or DS exhibit a variety of executive function impairments. Individuals with WS have particular impairments in response inhibition (Carney et al., 2013; Foti et al., 2015; Greer et al., 2013), selective attention (Breckenridge et al., 2013), attentional control (Breckenridge et al., 2013; Greer et al., 2013), and planning (Costanzo, Varuzza, et al., 2013), whereas individuals with DS have particular impairments in response inhibition (Carney et al., 2013; Costanzo, Varuzza, et al., 2013; Traverso et al., 2018), interference suppression (Traverso et al., 2018), selective attention (Breckenridge et al., 2013), and attentional control Syndrome(Breckenridge et al., 2013). Accordingly, we found that a majority of individuals with WS or DS, in particular those with lower mental ages, were impacted by the presence of unpredictive cues in the deterministic WPT task. Thus, less predictive cues in the standard WPT likely served as distractors and interfered with the ability of these individuals to focus on the highly predictive cues. Impairments in response inhibition in both groups might have also led to impulsive responding, which would not only engender errors but also interfere with their ability to assess cue–outcome associations since the entire array of cues may not be attended to. Thus, in addition to impairments in hippocampus-dependent learning, impairments in executive functions may also play a role in the inability of most individuals with WS or DS to solve the WPT.

To learn and remember the cue–outcome associations, participants must solicit memory functions. Since individuals with WS and individuals with DS exhibit preserved spatial response learning, a strategy that relies on the striatum (Banta Lavenex & Lavenex, 2021; Bostelmann et al., 2017; Bostelmann et al., 2018; Chang & Gold, 2003), and since previous studies showed that the striatum can be recruited when solving the WPT under very specific conditions (Foerde et al., 2006; Foerde et al., 2007), we hypothesized that most individuals with WS or DS would be able to solve the WPT. Our hypothesis was not confirmed, since more than 90% of individuals with WS or DS were unable to implement any strategy, whether striatum- or hippocampus-dependent to solve the task. However, the results from the present study are consistent with previous results in TD adults and children: the WPT is likely solved using hippocampus-dependent memory processes (Bochud-Fragnière et al., 2022; Foerde et al., 2006; Foerde et al., 2007) and does not elicit the striatum except under very specific conditions (Foerde et al., 2006; Foerde et al., 2007). When considering the probabilistic nature of the WPT, the striatum may not be the brain structure of choice to solve the task. Indeed, the striatum is particularly implicated in behaviors for which there is a one-to-one relation between the stimulus and the response, and when reward contingencies are deterministic (Shohamy, 2011). In contrast, the hippocampus is particularly implicated in behaviors that require the simultaneous comparison of numerous pieces of information, and for which cognitive flexibility is required (Rubin et al., 2014), as is the case for the WPT.

In sum, although the standard WPT is a complex cognitive task which may theoretically be solved using different strategies that depend on distinct learning and memory systems, healthy young adults preferentially use the hippocampus-dependent memory system to solve the WPT (Bochud-Fragnière et al., 2022; Foerde et al., 2006; Foerde et al., 2007). We propose that impairments in both hippocampal function (Banta Lavenex et al., 2015; Bostelmann et al., 2017; Bostelmann et al., 2018; Meyer-Lindenberg et al., 2005), which impacts probabilistic learning and cognitive flexibility, and deficits in executive functions (Breckenridge et al., 2013; Costanzo, Varuzza, et al., 2013), which impact attention and inhibition processes, may both contribute to the inability of most individuals with WS or DS to solve the WPT.

7 | CONCLUSION

Our study provided evidence that most individuals with WS or DS are impaired at the WPT, and further identified learning deficits due to partial reward contingencies and the inability to ignore distractors as potential factors underlying poor performance on the WPT. Altogether, our findings are consistent with learning impairments arising from hippocampus and executive function deficits in WS and DS, and highlight the importance of congruent feedback, explicit unambiguous instructions and materials, and the minimization of distractors to facilitate learning in individuals with WS or DS.

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CONFLICT OF INTEREST STATEMENT

The authors report there are no competing interests to declare.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article are available at https://doi.org/10.5281/zenodo.11234423. ~~will be made available by the authors, without undue reservation.~~

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