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Inhibitory Control in 6- to 8-year-olds with Williams Syndrome: Relations with Intellectual Ability and Parent Report Measure of Executive Function

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INTRODUCTION

- Executive function refers to the interrelated top-down cognitive processes that are integral in the management of actions, thoughts, and emotions (Diamond 2013).
- Williams syndrome (WS) is a genetic disorder caused by a hemideletion of 26-28 genes on chromosome 7q11.23.
- Individuals with WS often have mild to moderate intellectual disability, as well as impairments in executive function. The greatest executive function difficulty for individuals with WS is inhibitory control (Mervis & Greiner de Magalhães, in press).
- We directly evaluated inhibitory control using a laboratory measure of delay of gratification in 6 – 8-year-olds with WS. The purpose of the current study was to describe and characterize the performance of children with WS on a delay of gratification task. Furthermore, we evaluated the relations among child performance on the inhibitory control measure, child intellectual ability, and parent report of executive function.

METHODS

Participants:

- 33 children (16 boys, 17 girls) aged 6.01 – 8.05 years ($M = 6.91$ years, $SD = 0.70$) with genetically-confirmed classic WS deletions

Measures:

- Gift Wrap Task: Experimental behavioral measure which assesses inhibitory control (adapted from Kochanska et al., 1996)
 - The children were told that the examiner had a present for them and that it would be a "big surprise." The children were seated facing away from the gift and instructed to sit, wait, and not peek while the gift was being wrapped. The examiner then walked across the room and noisily wrapped the gift for approximately 60s.
 - The child's objective was to remain seated and inhibit the desire to look while the examiner was wrapping the gift.
 - A behavioral coding scheme was developed to characterize children's performance on the task. Coding was conducted using Behavioral Observation Research Interactive Software (BORIS, 2020) to evaluate the following:
 - Looking proportion = total time the child looked at gift divided by total duration of task
 - Standing proportion = total time the child was standing divided by total duration of task
 - Levels of inhibitory control:
 - 0 = never looked, remained seated
 - 1 = peeked/looked over shoulder at gift, remained seated
 - 2 = turned fully around to look at gift, remained seated
 - 3 = turned fully around to look at gift, stood up
 - The images below depict a child turned fully around to look at the gift while seated (level of inhibitory control = 2).



- Behavior Rating Inventory of Executive Function-2 (BRIEF-2; Gioia et al., 2015): Parent report measure which is used to assess behavioral, cognitive, and emotional aspects of executive function in children.
 - 3 indices: Emotional Regulation Index (ERI), Behavioral Regulation Index (BRI), and Cognitive Regulation Index (CRI)
 - Higher T-scores indicate greater difficulty in executive functioning (general population $M = 50$, $SD = 10$).
- Differential Ability Scales-II (DAS-II; Elliott, 2007): Standardized assessment used to measure intellectual ability. The General Conceptual Ability (GCA) standard score (SS; similar to IQ) is a composite score focused on verbal, reasoning, and conceptual abilities.
 - Higher scores indicate greater intellectual ability (general population $M = 100$, $SD = 15$).

RESULTS

Table 1. Descriptive Statistics: Gift Wrap Task

Measures	Mean (SD)	Median	Range
Looking Proportion	.34 (.36)	.30	0 – .99
Standing Proportion	.04 (.12)	0	0 – .59

- On average, children spent 34% of the time looking at the gift and 4% of the time standing up.

Table 2. Pass/Fail Statistics

	% Passed	% Failed
Boys	30.25	68.75
Girls	35.29	64.71
Total	30.30	69.70

- The proportion of children who passed (never looked, remained seated) or failed (looked at gift) did not differ as a function of sex ($\chi^2(1) = 0.41$, $p = .520$).

Table 3. Spearman Correlations: DAS-II, BRIEF-2

	Age	GCA	ERI	BRI	CRI
Looking Proportion	-.34	-.49**	.47**	.36*	.24

Note. * $p < .05$, ** $p < .01$

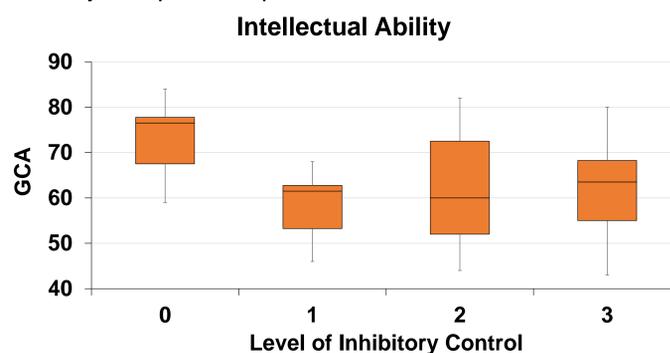
- Spearman correlations were conducted to assess the relation between looking proportion and age, intellectual ability, and parent report measure of executive function
- As intellectual ability increased, time spent looking at the gift decreased. Children with higher intellectual ability spent significantly less time looking at the gift.
- As ERI T-scores increased, time spent looking at the gift increased. Children with greater emotional regulation difficulties spent significantly more time looking at the gift.

Table 4. Descriptive Statistics: DAS-II, BRIEF-2 as a Function of Level of Inhibitory Control

Measures	Level of Inhibitory Control											
	Level 0 ($n = 10$)			Level 1 ($n = 6$)			Level 2 ($n = 11$)			Level 3 ($n = 6$)		
	Mean (SD)	Mdn	Range	Mean (SD)	Mdn	Range	Mean (SD)	Mdn	Range	Mean (SD)	Mdn	Range
GCA	73.20 (7.81)	76.5	59 – 84	60.00 (9.94)	61.5	46 – 72	61.73 (12.75)	60.0	44 – 82	62.00 (12.90)	63.5	43 – 80
BRI T-score	60.10 (10.02)	61.5	41 – 72	64.83 (10.91)	64.5	47 – 78	69.82 (9.03)	72.0	49 – 82	67.00 (6.00)	66.0	59 – 76
ERI T-score	58.00 (9.15)	55.0	48 – 74	57.17 (7.49)	57.0	47 – 66	66.27 (8.81)	69.0	51 – 79	68.83 (3.20)	68.0	65 – 74
CRI T-score	66.00 (5.25)	66.0	57 – 73	67.00 (12.70)	66.5	45 – 80	71.36 (7.07)	73.0	55 – 81	67.33 (3.88)	67.0	62 – 74

Effect of GCA and BRIEF-2 Indices on Level of Inhibitory Control

Separate Jonckheere-Terpstra tests for ordered alternatives were conducted to evaluate the effects of child intellectual ability and parent report of executive function on the level of inhibitory control:



- GCA had a significant effect on level of inhibitory control ($T_{JT} = 137.00$, $Z = -2.01$, $p = .045$).
- Results of post-hoc stepwise comparisons were: $0 > (1 = 2 = 3)$. Children who never looked and remained seated had significantly higher median GCA SSs than children who looked, turned fully around, or turned and stood up ($p < .05$). No other significant differences were detected.



- ERI T-scores had a significant effect on level of inhibitory control ($T_{JT} = 286.50$, $Z = 2.84$, $p = .005$).
- Results of post-hoc stepwise comparisons were: $0 < (2 = 3)$; $1 < 3$. Children who turned around or turned and stood up had higher median ERI T-scores than children who never looked. Children who turned around and stood up had higher median ERI T-scores than children who only peeked/looked over their shoulder ($ps < .05$).

- No statistically significant effect of BRI ($p = .077$) or CRI ($p = .347$) T-scores on level of inhibitory control was found.

DISCUSSION

- Deficits in inhibitory control for children with WS were evident. The majority of the children (70%) were unable to delay gratification, which is evidenced by the fact that children could not inhibit the urge to peek or look at the gift while it was being wrapped.
- Intellectual abilities was significantly related to children's performance on the laboratory measure of inhibitory control. Children who never looked and remained seated had significantly higher intellectual abilities than children who did not follow task instructions.
- Parent report of emotion regulation was significantly related to children's performance on the laboratory measure of inhibitory control. Children with better emotion regulation exhibited higher levels of inhibitory control.
- Inhibitory control underlies many facets of daily life, such as school achievement, mastery motivation, and adaptive skills (Mervis & Greiner de Magalhães, in press). These results highlight the need for research-based interventions to ameliorate deficits in executive function for children with WS.