Executive Functioning, Parenting Style, and Risk-Taking Behaviors among the Offspring of Parents with Huntington's Disease

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Abstract

Patients of Huntington's Disease experience decreased capacity for executive functioning. Moreover, HD patients also experience increased levels of impulsivity and risk-taking behaviors. Consequences of these behaviors include disrupted relationships, social isolation, and both legal and financial consequences, all leading to increased stress. As a result, children of parents with HD may have less warmth and structure in their relationships. This may lead to deficits in executive functioning and increases in risky behaviors in their children. Understanding how parenting style moderates the relationship between children's executive functioning and their risk-taking behaviors may allow future research to develop intervention strategies to create a protective effect against these negative behaviors for children of HD parents.

Introduction

Huntington's disease (HD) is a neurodegenerative disorder with onset typically in the fourth or fifth decade of life (Bates et al. 2015). Three of the components of HD are progressive deficits in motor, cognitive, and emotional functions. Impairment of motor skills starts presenting early with small, uncontrollable twitches in the hands and feet, a symptom known as chorea. Ultimately, this impairment affects the entire body, resulting in slurred speech, inability to walk, and even function completely, which results in death. Cognitively, HD leads to a loss of executive functioning, including deficits in working memory and attentional and impulse control. Emotional symptoms of HD involve apathy, irritability, and depression.

HD is caused by mutations in the HTT gene, which is responsible for synthesizing a protein called huntingtin. Huntingtin protein is a normal part of our brains and plays a role in neuron development. However, mutations of the HTT gene in patients with HD cause the production of a mutant form of huntingtin, resulting in neurodegeneration. Additionally, this condition is an autosomal dominant and fully penetrant disorder, which means that there is a 50% chance a child will have HD if their parent does. (Bates 2003).

With regard to cognitive function, executive functioning (EF) is a set of general-purpose psychological processes and includes things like, planning, concentration, working memory, and self-regulation. Before considering EF in patients with HD, it is important to first understand the foundations and components of EF. In a structural model proposed by Miyake and Friedman (2012), standardized laboratory tasks have shown both unity and diversity in executive functioning. In other words, different executive functioning skills are correlated with one another, giving rise to an underlying general ability for executive functioning (unity). However, these executive functions can also be separated and tested individually as well (diversity).

Specifically, updating, shifting, and inhibition are the three components that fall under the unity umbrella of executive functioning, coined as “common EF”. Updating is the ability to constantly monitor and reappraise the contents of our working memory, shifting is the ability to swap between tasks, and inhibition is the ability to control attention or focus. In general, all three of these measures simultaneously relate to one another, while also relating differently to other measures, such as IQ and examinations of frontal lobe functioning.

Specifically, inhibitory control is our ability to meaningfully control our attention, thoughts, behaviors, and emotions in order to “cancel” our inherent predispositions to react a certain way (Diamond 2013). These scenarios can range from being able to control an outburst anger at an inappropriate time to resisting the temptation of vices like using illegal substances. Since mutant huntingtin causes neurodegeneration within the prefrontal cortex, this posits patients of HD to have issues with their inhibitory control, which can result in harmful risk-taking behaviors due to increased impulsivity.

Impulsivity involves the tendency to make up one’s mind quickly, taking action with less thought than others would (Barratt 1994). Furthermore, three dimensions of impulse have also been proposed. First, motor impulse involves taking physical actions without thinking. Cognitive impulse involves making decisions significantly faster than others. And most importantly, the non-planning dimension of impulse involves a tendency to think only in the short-term, without any consideration of future consequences. Ultimately, impulsivity limits our ability to reliably consider outcomes for both ourselves and others, which can potentially have detrimental effects on our relationships.

To elaborate on risk-taking behaviors, patients with HD have been characterized by risky behavior in several different facets of life. For example, Kalkhoven et al (2014) found that HD patients were less likely to consider long-term rewards than typical populations, and that HD patients were more susceptible to risky gambling decisions. Furthermore, Schultz et al (2017) found that HD patients were more likely to use substances like tobacco, alcohol, and hard drugs, than typical populations, and that this substance abuse led to the worsening of their deficits in motor function. Overall, HD patients tend to be more at risk for dangerous risk-taking behavior, which can ultimately lead to issues with relationships and even isolation. Despite the importance of examining risky behaviors in HD patients, there have not been many ways to reliably assess these behaviors empirically. In fact, the only screening tool to assess these behaviors was designed by McDonnel et al (2020), known as the Risk Behavior Questionnaire, or the RBQ-HD. This study found that the RBQ-HD was reliable for assessing risky behaviors when using both patients and caregivers as informants.

While it may seem that executive functioning is only involved in specific processes, like inhibitory control, in reality, several studies have shown that poor executive functioning as a whole is correlated with neuropsychiatric conditions. For example, Moran (2016), showed that individuals suffering from anxiety were more likely to perform worse on measures of EF. Moreover, Snyder (2013) showed that patients with major depressive disorder had slower processing speeds and deficits in measures of EF. While not neurodegenerative, these studies show that poor executive functioning is a marker of poor mental health. If poor executive functioning is correlated with neuropsychiatric symptoms, then the question is raised if improving executive function can be used as an intervention to reduce these neuropsychiatric symptoms.

On another note, previous research has shown that parenting style is an effective predictor of children’s behaviors. For example, positive parenting styles like warmth have been shown to be associated with a lower degree of the child experiencing externalizing behaviors, of which many risk-taking and impulsive behaviors may fall. (McKee et al. 2007). On the other hand, more negative parenting styles have been shown to be positively associated with familial conflict and internalizing behaviors (Warmuth et al. 2020). With regards to how parenting style may affect children's executive functioning ability, a study done by Compas et al. (2017) posits that positive parenting styles were associated with increased use of children's secondary control coping strategies, which require the use of strong executive functioning skills. Finally, positive parenting styles have been shown to be associated with better attentional and impulse control among children with ADHD (Healey et al 2011). Understanding that parenting style can relieve some of the burden placed on these neurodivergent children, the same concept can be applied to at-risk HD children.

With regard to HD specifically, several studies have already examined links between coping, HD, and executive functioning. For example, since the children of parents with diagnosed HD experience great deals of psychological distress, Ciriegio et al. (2020) examined working memory, secondary control coping, and anxiety and depression symptoms in the offspring of parents with HD. Interestingly, these offspring performed significantly worse on tests of working memory and had elevated levels of anxiety and depression symptoms. Moreover, working memory, secondary control coping, and neuropsychiatric symptoms were not just correlated, but also revealed that working memory explained the increased anxiety and depressive symptoms through a lack of secondary control coping. In a second study, Ciriegio et al. (2022) examined similar measures in adults with HD as well. Here, better inhibitory skills were associated with greater use of secondary control coping, which in turn was associated with less anxiety and depression symptoms. Overall, both these studies imply that HD-affected populations would greatly benefit from interventions that improve their executive functioning. In turn, this may improve their secondary control coping skills, which can improve their mental health and reduce symptoms of anxiety and depression.

Currently, HD can be diagnosed by genetic test for only individuals above the age of 18. However, a genetic test only confirms or denies the presence of the mutated HTT gene and says very little about onset and progression of neurodegeneration. Instead, this is usually measured by a CAP score. One component of the CAP score is the number of CAG repeats on the HTT gene. Typically, the more CAG repeats on the gene, the more severe the progression of HD is. A CAP score is generated from the number of repeats multiplied by the patient’s age, CAP standing for CAG Age Product (Pfalzer et al 2022).

However, deficits in executive function may be an early marker of HD before any physical signs of chorea are even present. To elaborate, a study by You et al. (2014) examined executive function abilities, including inhibitory control in premanifest HD patients for correlations with the disease markers of disease burden and striatal volume. Interestingly, disease burden was found to be higher for patients with lower working memory scores. Although this study only involved 15 participants, it does pave the way for future research considering executive function as a measure of determining onset and progression of HD.

Overall, among at-risk offspring, it is hypothesized a) that there will be an association between worsened levels of executive functioning and greater risk-taking behaviors. Furthermore, it is hypothesized that b) parenting style will function as a moderator between executive functioning and risk-taking behaviors.

Method

Participants

Participants include 56 at-risk offspring who have an HD-affected parent. Participants ranged from 8-38 and had a mean age of 19.

Procedure

HD patients and their offspring were recruited through the Huntington's Disease Multidisciplinary Clinic at Vanderbilt University Medical Center. Inclusion criteria for the HD sample are: participants must be fluent in English; parents with HD must be part of the Huntington's Disease Multidisciplinary Clinic at Vanderbilt University Medical Center; parents with HD can range in disease severity (premanifest, prodromal, motor manifest HD); Exclusion criteria for HD sample includes a diagnosis of autism spectrum disorder or psychosis in the HD parent or their offspring.

Exclusion criteria for the healthy control sample are a diagnosis of any neurological or neurodegenerative disease, autism spectrum disorder, or psychosis in the parent or offspring.

Measures

Data from the NIH Toolbox Cognition battery was used. The Toolbox Cognition Battery is designed for individuals aged 7 through adulthood. This computer-based assessment measures executive function including episodic memory, language, working memory, and attention. More specifically, participants were administered the following subtests: (a) Dimensional Card Sort Task, an executive function task of capacity to plan, organize, and monitor in a goal-oriented manner; (b) Flanker Task, an attention task that requires focus on a given stimulus while inhibiting attention to surrounding stimuli; (c) List Sorting Task, a working memory task requiring immediate recall and sequencing of various orally and visually presented stimuli; and (d) Picture Sequence Memory Task, an assessment of episodic memory by recalling an increasingly lengthy series of activities presented one at a time.

The quality of the parent-offspring relationship was assessed through behavioral observations conducted and recorded in private offices. Parent-offspring dyads were asked to talk together during two interactions that will be video recorded. Parents and their offspring were first asked to discuss a positive experience they have shared together recently for 10 minutes. Then, parents and their offspring were asked to discuss a recent source of stress in their family for 10 minutes. Trained members of the research staff coded all videotapes using the standardized coding system: Iowa Family Interaction Rating Scales (IFIRS).

Adult offspring completed the Adult Self-Report (ASR) which is a reliable and valid standardized self-report tool to assess emotional and behavioral functioning. The ASR contains 126 items on problem behaviors that have occurred in the past 6 months. For each item, the respondent indicates on a three-point scale how true the item is from 0 “not true” to 2 “very true or often true”. Adolescents completed the Youth Self-Report (YSR),122 which complements the ASR but is completed by adolescents. The ASR and YSR provides scores on three broadband scales (Internalizing, Externalizing, Total Problems) and eight syndrome scales (e.g., Anxious/Depressed). Parents also filled out the Adult Behavior Checklist (ABCL) or Child Behavior Checklist (CBCL) for their offspring, which serves as a parental-reported complement to the ASR/YSR.

Results

Descriptive statistics were provided for variables of interest. The total sample consisted of 56 participants with a mean age of 19.09, ranging from 8 to 38 years old. However, not all participants completed all measures, so the sample size varies from N=26 for RBQ Sum to N=56 for the working memory test.

Table 1

Descriptive Statistics for Key Variables

	N	Minimum	Maximum	Mean	Std. Deviation
Offspring Age	56	8	38	19.09	7.96
Offspring Flanker Inhibitory Control T-Score	54	1.00	56.00	32.7	13.31
Offspring Dimensional Change Card Sort T-score	54	10.00	99.00	50.00	17.27
Offspring Working Memory T-score	56	8.00	99.00	51.17	16.6
Offspring Processing Speed T-score	53	1.00	555.00	56.2	72.9
Parent Warmth	42	1.00	9.00	4.78	1.91
Parent Child-Centeredness	42	2.00	8.00	5.8	1.59
Parent Child-Monitoring	42	2.00	7.00	5.09	1.44
Offspring Self-Report Attention Problems	51	50.00	78.00	59.2	9.63

Offspring Self-Report Rule Breaking Behaviors	51	50.00	77.00	53.9	6.06
Offspring Self-Report ADHD Problems	51	50.00	77.00	59.3	8.52
Parent-Report on Offspring Attention Problems	39	40.00	88.00	55.07	8.85
Parent-Report on Offspring Rule Breaking Problems	39	35.00	74.00	53.17	7.04
Parent-Report on Offspring ADHD Problems	39	50.00	75.00	54.92	6.92
RBQ Sum	26	.00	24.00	6.15	5.86

Bivariate Correlational Analysis

Table 2 presents correlations between the key variables, examining for possible associations between measures of EF and RBQ, as well as parenting style. It was found that the RBQ Sum was not significantly correlated with any measures of EF, possibly due to small sample size (n=26). As a result, measures from the ASEBA self-report and parent-report that reflected risky behaviors and impulsivity, including ADHD symptoms, attentional problems, and rule-breaking behaviors. One measure of EF to note is working memory (WM). WM was significantly negatively correlated with parent-reported symptoms of ADHD (r = -0.39, p = .014), parent-reported attentional problems (r = -0.50, p = .001), and parent-reported rule-breaking behaviors (r = -.54, p < .001). This is consistent with the hypothesis that worsened levels of executive functioning predict greater levels of risky behaviors and impulsivity. However, it should be noted that self-reported measures of attention problems, rule-breaking behavior, and ADHD symptoms were not significantly correlated with EF, nor was the self-reported measure of the RBQ. Additionally, the Flanker Inhibitory Control and Dimensional Card Sort measures were not significantly correlated with any measures of risky behavior either. Regarding parenting style, it was found that significant negative correlations exist between working memory and parental warmth (r = -0.33, p = .03). This is opposite to the hypothesis and suggests that worse working memory ability is predicted by higher degrees of parental warmth. Regarding age, no significant moderating or interaction effects were found.

Table 2 *Bivariate correlations between key variables*

Variables	1	2	3	4	5	6	7	8	9	10	11
1. Offspring Age	—										
2. Working Memory T-Score	-.106	—									
3. Parental Warmth	-.206	-.335*	—								
4. Parental Child-Centeredness	-0.206	-.195	.758*	—							
5. Parental Child-Monitoring	-.297	-.192	.658**	812**	—						
6. Child Attention Problems (C)	-.354*	-.024	.172	.260	.339*	—					
7. Child Rule Breaking Behaviors (C)	-.029	-.134	.311	.315	.307	.208	—				
8. Child ADHD Symptoms (C)	-.294*	.007	.006	.103	.259	.892**	.20	—			
9. Child Attention Problems (P)	-.269	-.502**	-.22	.216	.39*	.46**	.269	.463**	—		
10. Child Rule Breaking Behaviors (P)	-.157	-.541**	.31	.101	.167	.273	.484**	.21	.589**	—	
11. Child ADHD Symptoms (P)	-.30	.39*	-.025	-.127	.258	.423**	.249	.513**	.658**	.58**	—

Note. (C) Child Self-Report, *(P)* Parent Report on Child. * p<.05. ** p<.01

Regression Analysis

Based on the significant correlations between parent-reported behaviors and working memory, analyses of linear regression were run to test for moderating effects of a warm parenting and age on this relationship. A main effect was observed for parental warmth on parent-reported attention problems ($\beta = .56$, $p = .004$), on parent-reported rule-breaking behaviors ($\beta=0.647$, $p<0.001$), and on parent-reported ADHD symptoms ($\beta = .504$, $p = .013$). Ultimately, it was found that the only moderating effect that existed was that warmth moderated the relationship between working memory and parent-reported rule-breaking problems ($\beta = -.40$, $p = .023$). Higher degrees of parental warmth reduced rule-breaking behaviors among low working memory-performing offspring.

Table 3

Regression Models using Warmth as a Moderator

		Unstandardized Coefficients		Standardized Coefficients			Correlations			Collinearity Statistics	
Model		B	Std. Error	Beta	t	Sig.	Zero-order	Partial	Part	Tolerance	VIF
1	(Constant)	54.103	1.715		31.542	<.001					
	Centered WM	.471	.151	.564	3.130	.004*	.509	.531	.515	.834	1.199
	Centered Warmth	.280	.947	.058	.295	.770	-.222	.059	.049	.707	1.415
	WM x Warmth Interaction	-.157	.107	-.277	-1.469	.154	-.177	-.282	-.242	.759	1.317

Dependent Variable: Parent-Report on Offspring Attention Problems

Model		Unstandardized Coefficients B	Unstandardized Coefficients Std. Error	Standardized Coefficients Beta	t	Sig.	Correlations Zero-order	Correlations Partial	Correlations Part	Collinearity Statistics Tolerance	Collinearity Statistics VIF
2	(Constant)	51.348	1.242		41.337	<.001					
	Centered WM	.447	.109	.647	4.096	<.001**	.580	.634	.591	.834	1.199
	Centered Warmth	.177	.686	.044	.259	.798	-.310	.052	.037	.707	1.415
	WM x Warmth Interaction	-.187	.077	-.401	-2.421	.023*	-.294	-.436	-.349	.759	1.317

Dependent Variable: Parent-Report on Offspring Rule Breaking Problems COMBINED CBCL/ABCL

Model		Unstandardized Coefficients B	Unstandardized Coefficients Std. Error	Standardized Coefficients Beta	t	Sig.	Correlations Zero-order	Correlations Partial	Correlations Part	Collinearity Statistics Tolerance	Collinearity Statistics VIF
3	(Constant)	53.788	1.255		42.851	<.001					
	Centered WM	.293	.110	.504	2.660	.013*	.357	.470	.460	.834	1.199
	Centered Warmth	1.099	.693	.326	1.585	.125	.025	.302	.274	.707	1.415
	WM Warmth Interaction	-.147	.078	-.374	-1.882	.072	-.171	-.352	-.326	.759	1.317

Dependent Variable: Parent-Report on Offspring ADHD Problems COMBINED CBCL/ABCL

Table 4

Regression Models using Age as a Moderator

Model	Unstandardized Coefficients B	Unstandardized Coefficients Std. Error	Standardized Coefficients Beta	t	Sig.	Correlations Zero-order	Correlations Partial	Correlations Part	Collinearity Statistics Tolerance	Collinearity Statistics VIF

4	(Constant)	53.886	1.285		41.936	<.001					
	Centered WM	.379	.110	.484	3.433	.002*	.502	.502	.483	.993	1.007
	Centered Age	.229	.162	.210	1.415	.166	.269	.233	.199	.899	1.113
	WM x Age Interaction	.008	.019	.065	.439	.663	.136	.074	.062	.904	1.106

Dependent Variable: Parent-Report on Offspring Attention Problems COMBINED CBCL/ABCL

		Unstandardized Coefficients		Standardized Coefficients			Correlations			Collinearity Statistics	
Model		B	Std. Error	Beta	t	Sig.	Zero-order	Partial	Part	Tolerance	VIF
5	(Constant)	52.096	1.019		51.139	<.001					
	Centered WM	.332	.087	.533	3.794	<.001*	.541	.540	.531	.993	1.007
	Centered Age	.074	.129	.085	.577	.568	.157	.097	.081	.899	1.113
	WM x Age Interaction	.009	.015	.092	.628	.534	.125	.105	.088	.904	1.106

Dependent Variable: Parent-Report on Offspring Rule Breaking Problems COMBINED CBCL/ABCL

		Unstandardized Coefficients		Standardized Coefficients			Correlations			Collinearity Statistics	
Model		B	Std. Error	Beta	t	Sig.	Zero-order	Partial	Part	Tolerance	VIF
6	(Constant)	54.338	1.057		51.402	<.001					
	Centered WM	.224	.091	.367	2.471	.018*	.390	.385	.366	.993	1.007
	Centered Age	.257	.133	.301	1.928	.062	.300	.310	.285	.899	1.113
	WM x Age Interaction	-.010	.015	-.099	-.638	.528	-.002	-.107	-.094	.904	1.106

Dependent Variable: Parent-Report on Offspring ADHD Problems COMBINED CBCL/ABCL

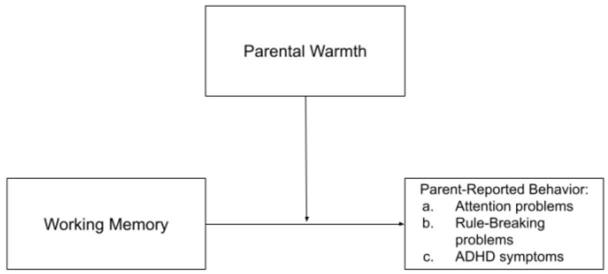

Figure 1. Proposed model of parental worth moderation on the relationship between working memory and parent-reported behaviors.

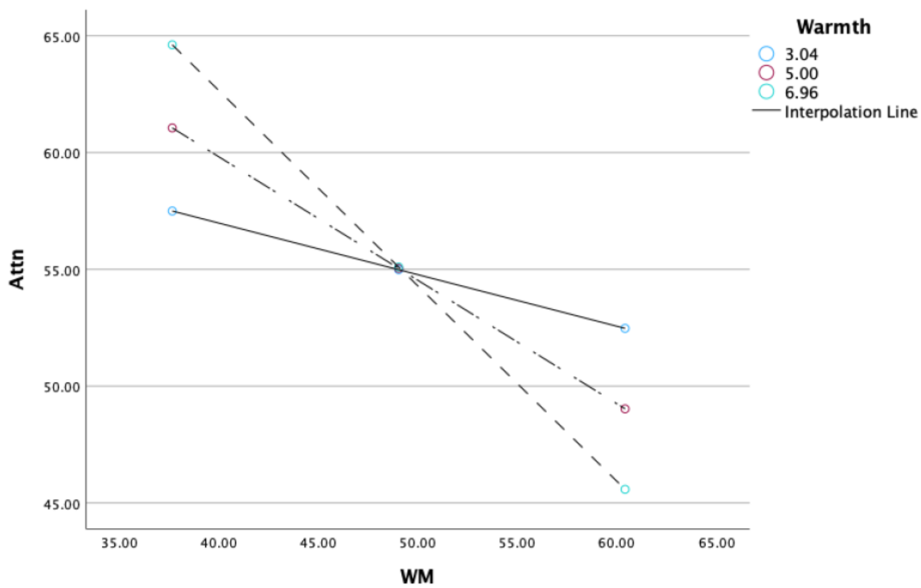

Figure 2. Working Memory and Attentional Problems Moderated by Warmth

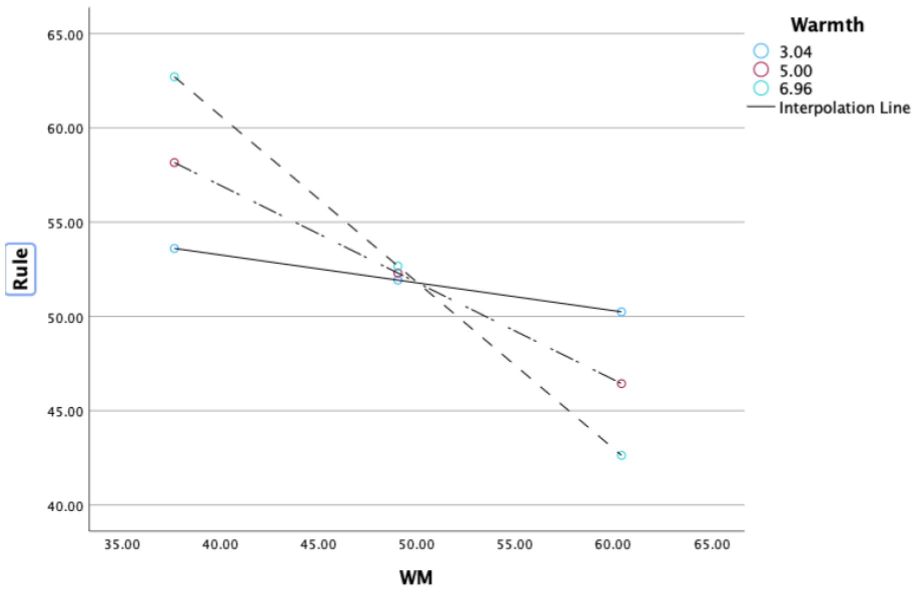

Figure 3. Working Memory and Rule Breaking Behaviors Moderated by Warmth

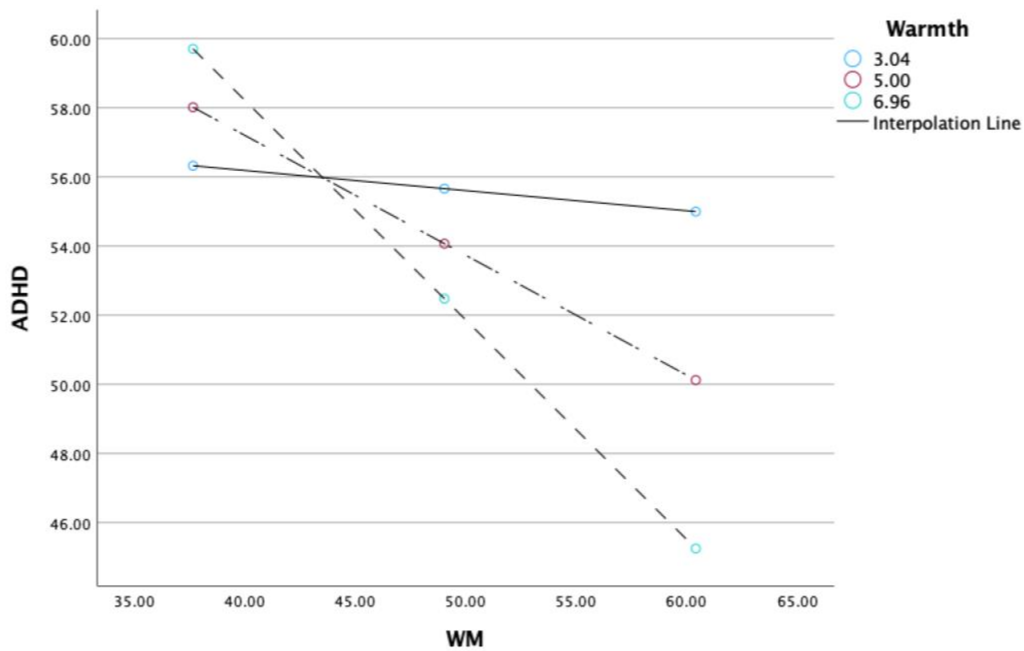

Figure 4. Working Memory and ADHD Symptoms Moderated by Warmth

Discussion

Ultimately, it was found that none of the measures of executive functioning or parenting were correlated with the total score on the Risky-Behavior Questionnaire. However, this is most likely due to the relatively smaller sample size of participants who completed the RBQ. Because of this, three measures from the ASEBA self-report and behavior checklist were used to represent risky and impulsive behavior.

Attentional problems and ADHD symptoms were chosen to represent impulsivity associated with risk-taking behaviors, while rule-breaking behavior problems were chosen to represent the conduct issues associated with many risk-taking behaviors. Of the different measures of executive functioning, significant negative correlations were found between working memory and these measures of risky behavior, indicating that poor working memory ability is associated with greater attentional problems, ADHD symptoms, and rule-breaking behaviors.

However, it should be noted that these significant correlations exist only for parent-reported surveys, while self-reported surveys for these behaviors were non-significant. This is important to understand since these offspring may not realize they are even taking these risky behaviors. This is consistent with the hypothesis about deficits in executive functioning and specifically working memory. At-risk offspring suffering from these cognitive deficits may have a difficult time using abstract thinking to fully understand the consequences of dangerous behaviors, leading to impulsivity. As a result, there may be a self-report bias, explaining the discrepancy between significance between parent and self-reported behaviors.

Regarding parenting style, an effect opposite to the hypothesis was actually found, and executive functioning had a negative correlation with parental warmth. To elaborate, lower offspring working memory scores was associated with higher degrees of parental warmth. While it was originally hypothesized that warm and structured parenting would lead to cognitively stronger offspring, it is important to remember that this model did not test for directionality between these variables. In fact, this negative correlation may be explained by the fact that parents feel the need to compensate and support their children who struggle more cognitively, suggesting that parents might increase positive behaviors if they notice their children are predisposed to cognitive deficits.

Results from the linear regression analysis did not confirm the second hypothesis that parenting style functioned as a moderator between EF and risky behaviors. Significant main effects were found for all models between working memory and parent-reported risky and impulsive behaviors. However, only one significant interaction effect was found. The interaction effect between working memory and parental warmth showed significance, suggesting that the two values depend on one another in predicting risky behaviors. Furthermore, this value was negative, indicating that an increase in warm parenting may reduce the amount of risky behavior as predicted by working memory.

There are several limitations that affect this study. First, the sample size is relatively small and differed between measures. For example, the RBQ only had 26 participants, and only 18 of these participants completed the interaction task to assess parenting style. This small sample size may explain the lack of significance when using the RBQ. For reference, correlations using the CBCL and WM were significant and had a sample size of 37. Additionally, the data used in this study was cross-sectional, and as a result, we cannot test for the directionality of any of these correlations. Furthermore, due to a small sample size, the age group was expanded to include all offspring rather than just those under 18. Parenting effects are most likely stronger for children under 18 rather than offspring above 30 who have been living away from home, yet this study was unable to fully capture this.

Despite these limitations there are still several strengths to this study as well. For example, this study uses a multi-informant survey. Ultimately, surveying both offspring and their parents about offspring risky behavior revealed an important discrepancy in ADHD symptoms, attentional problems, and rule-breaking problems that would not have been captured otherwise. Various measures of specific EF processes also reveal more detailed framework in which components of EF contribute to these risky and impulsive behaviors.

For future studies, a larger sample size would help elaborate on some of these correlations and interactions within this study. Furthermore, adding a longitudinal design to the study would help establish directionality as well. Furthermore, it would be beneficial to specifically study offspring under 18 in a separate group than those who are over 18. Doing so may reveal a stronger interaction effect of parenting style. Additionally, an intervention study may wish to focus on working memory and measure risk-taking behaviors as an outcome. Ultimately, these findings present valuable new information on the relationship between executive functioning and risk-taking behaviors among at-risk offspring, suggesting a possible area of intervention in working memory specifically.

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