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### Sibling Mediation in an Executive Function Intervention for Children with Autism Spectrum Disorder

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SIBLING MEDIATION IN AN EXECUTIVE FUNCTION INTERVENTION FOR  
CHILDREN WITH AUTISM SPECTRUM DISORDER

A Thesis Presented to  
The Faculty of the School of Medicine  
Yale University

In Candidacy for the Degree of  
Master of Medical Science

July 2022

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## **ABSTRACT**

Autism spectrum disorder is a developmental disorder characterized by social communication deficits and restricted, repetitive behaviors. It is associated with deficits in cognitive flexibility, which is the ability to switch between tasks, shift attention, and adapt learned responses. Various interventions, such as the Unstuck and On Target curriculum, target cognitive flexibility. In addition, prior studies have supported including sibling mediation in interventions; however, the inclusion of siblings as mediators of the Unstuck and On Target curriculum has not yet been explored. **We propose to determine whether including siblings as mediators to a modified Unstuck and On Target curriculum improves cognitive flexibility outcomes, specifically performance on the Wisconsin Card Sorting Task, as well as the quality of the sibling relationship.** This randomized controlled trial will provide implications on the integration of typically developing sibling mediators in other contexts to help children with autism spectrum disorder.

## **Chapter 1 – Introduction**

### ***1.1 Autism Spectrum Disorder***

Autism spectrum disorder (ASD) is a lifelong developmental disability characterized by persistent deficits in social communication and restricted, repetitive behaviors, interests, and activities.<sup>1</sup> These symptoms must be present in the early developmental period, with the onset of symptoms typically seen by age 3, and cause clinically significant impairment in social areas of functioning.<sup>2</sup> As a spectrum disorder, the presentation and severity of ASD symptomatology varies considerably between individuals,<sup>3</sup> and severely affected children are more likely to be reliably identified and diagnosed at younger ages.<sup>4</sup> The symptoms must also not be better explained by an intellectual disability or global developmental delay.<sup>2</sup> The full diagnostic criteria as outlined by the DSM-V can be found in Table 1.

**Table 1. DSM-V Diagnostic Criteria for Autism Spectrum Disorder**

|  |
|--|
| <p><b>To meet diagnostic criteria for ASD according to DSM-5, a child must have persistent deficits in each of three areas of social communication and interaction (see A.1. through A.3. below) plus at least two of four types of restricted, repetitive behaviors (see B.1. through B.4. below).</b></p>  |
| <p>A. Persistent deficits in social communication and social interaction across multiple contexts, as manifested by the following, currently or by history (examples are illustrative, not exhaustive; see text):</p> <ol style="list-style-type: none"><li>1. Deficits in social-emotional reciprocity, ranging, for example, from abnormal social approach and failure of normal back-and-forth conversation; to reduced sharing of interests, emotions, or affect; to failure to initiate or respond to social interactions.</li><li>2. Deficits in nonverbal communicative behaviors used for social interaction, ranging, for example, from poorly integrated verbal and nonverbal communication; to abnormalities in eye contact and body language or deficits in understanding and use of gestures; to a total lack of facial expressions and nonverbal communication.</li><li>3. Deficits in developing, maintaining, and understand relationships, ranging, for example, from difficulties adjusting behavior to suit various social contexts; to difficulties in sharing imaginative play or in making friends; to absence of interest in peers.</li></ol> |

|   |
|---|
| <p>B. Restricted, repetitive patterns of behavior, interests, or activities, as manifested by at least two of the following, currently or by history (examples are illustrative, not exhaustive):</p> <ol style="list-style-type: none"> <li>1. Stereotyped or repetitive motor movements, use of objects, or speech (e.g., simple motor stereotypes, lining up toys or flipping objects, echolalia, idiosyncratic phrases).</li> <li>2. Insistence on sameness, inflexible adherence to routines, or ritualized patterns of verbal or nonverbal behavior (e.g., extreme distress at small changes, difficulties with transitions, rigid thinking patterns, greeting rituals, need to take same route or eat same food every day).</li> <li>3. Highly restricted, fixated interests that are abnormal in intensity or focus (e.g., strong attachment to or preoccupation with unusual objects, excessively circumscribed or perseverative interests).</li> <li>4. Hyper- or hypo-reactivity to sensory input or unusual interest in sensory aspects of the environment (e.g. apparent indifference to pain/temperature, adverse response to specific sounds or textures, excessive smelling or touching of objects, visual fascination with lights or movement).</li> </ol> |
| <p>C. Symptoms must be present in the early developmental period (but may not become fully manifest until social demands exceed limited capacities, or may be masked by learned strategies in later life).</p>  |
| <p>D. Symptoms cause clinically significant impairment in social, occupational, or other important areas of current functioning.</p>  |
| <p>E. These disturbances are not better explained by intellectual disability (intellectual developmental disorder) or global developmental delay. Intellectual disability and autism spectrum disorder frequently co-occur; to make comorbid diagnoses of autism spectrum disorder and intellectual disability, social communication should be below that expected for general developmental level.</p>   |

*Source: Diagnostic and Statistical Manual of Mental Disorders, 5th Edition, 2013, American Psychiatric Association.*

The prevalence of diagnosis has increased in the United States within the last 30 to 40 years,<sup>5</sup> with the latest available data on ASD demonstrating a prevalence of one in 44 children aged 8 years in the United States.<sup>6</sup> The reason for this accelerated increase is multifactorial given the broadening of diagnostic criteria, increased provider ascertainment at earlier ages, and improved parental awareness.<sup>5</sup> Additionally, there are



large research programs with the goal of improving early ASD detection in the community for faster connection of affected families to services for children with ASD.<sup>6</sup> Autism has also been found to be more prevalent in males than females.<sup>6,7</sup>

While there is no cure for ASD or its core symptoms, there are a variety of interventions available that aim to teach skills for daily life and reduce undesirable or inappropriate behaviors.<sup>8</sup> While research on this clinical population shows limited or ambiguous efficacy regarding pharmacotherapy, most of the literature supports the use of behavioral and psychological interventions for core and associated symptoms for ASD.<sup>9</sup> These treatment programs commonly incorporate applied behavior analysis, a modality which focuses on behavior change and skill building.<sup>5,8</sup> Since ASD affects and manifests in each person differently, interventions are usually catered toward the individual and their unique needs. Timely evaluation and diagnosis of ASD among young children are also important as early treatments are associated with improved outcomes.

### ***1.2 Challenges for Family***

ASD can cause significant challenges for affected children as well as place substantial burden on family and other caregivers.<sup>10</sup> The hallmark repetitive and restricted behaviors of ASD have been associated with increased family stress,<sup>11</sup> with parents reporting higher levels of stress and affective symptoms compared to parents of typically developing children and parents of children with other disabilities.<sup>1,12</sup> Delays and impairments in social relatedness in children with ASD have been associated with strained familial relationships, which further adversely impacts child functioning and development.<sup>13-15</sup>

With the sibling relationship likely the longest lasting relationship in a person's life, siblings of children with ASD face unique stressors compared to siblings of typically developing children.<sup>16</sup> Using tailored self-report questionnaires to measure the perceptions of the relationship quality, such as the Sibling Relationship Questionnaire (SRQ), researchers have identified altered patterns of hostility, closeness, and warmth among sibling relationships when one child has ASD.<sup>16-18</sup> For example, aggression and hostility from children with ASD has been shown to be a significant stressor for their typically developing (TD) siblings.<sup>19</sup> Siblings have several additional roles such as play companion, nurturer, conversation partner, and teacher,<sup>20</sup> and in 73% of cases, become the primary caregiver of their sibling with ASD after the death of the parents.<sup>16</sup> These alterations of a child's role in the family may give rise to negativity such as resentment and rivalry toward their sibling with special needs.<sup>18</sup> Fostering a healthy sibling relationship is especially critical as it relates to both children's well-being as well as the TD sibling involvement in lifelong care, friendship, and advocacy.<sup>21</sup>

### ***1.3 Executive Function***

Executive function is an umbrella term used to refer to the cognitive processes underlying goal directed and adaptive behavior.<sup>22</sup> The core domains of executive function include planning, working memory, cognitive flexibility, and inhibition,<sup>23</sup> which are theorized to be typically impaired in ASD as well as other neurodevelopmental disorders involving frontal lobe deficits, such as attention-deficit hyperactivity disorder and obsessive-compulsive disorder.<sup>24</sup> These deficits in the components of executive function are believed to contribute to problem behaviors; for example, cognitive inflexibility in regards to switching conversation topics can result in social interaction impairments.<sup>25</sup>

For an individual with ASD, what appears as opposition and stubbornness can actually be an innate protective effort to avoid being overwhelmed and overstimulated.<sup>26</sup> Executive function deficits not only often predict autistic behaviors, skills, and outwardly visible symptomology,<sup>27</sup> but also have been associated with decreased independence and poor outcomes extending into adulthood.<sup>11</sup> Progress and improvement in each executive function is essential to preparing an individual with ASD to meet the expectations of the adult world.<sup>26</sup> The effect of executive functions on an individual's social competence and functioning make them important targets of interventions for this population.<sup>28</sup>

Cognitive flexibility encompasses a range of interrelated characteristics that includes easily switching between tasks, shifting attention to different features, and adapting responses based on reward and punishment.<sup>29</sup> It is a key constituent of executive function that orients people to relevant environmental information and allows them to adapt to changes, switch perspectives, and adjust behaviors.<sup>30</sup> Cognitive flexibility difficulties in autism have been well-studied and found to predict adaptive functioning in youth with ASD.<sup>31</sup> Adaptive functioning, which is the ability to successfully meet age-appropriate demands in everyday life, is particularly impaired in ASD but may be malleable with intervention and support.<sup>20</sup> Cognitive flexibility skills in autism are necessary given the intrinsic difficulty detaching from specific objects or activities, and adapting to changing environments.<sup>30</sup>

One widely used neuropsychological test that assesses cognitive flexibility is the Wisconsin Card Sort Task (WCST), with research showing consistently impaired performance in the ASD population.<sup>32</sup> The cards differ from one another in three stimulus dimensions, and the task involves sorting the cards according to a rule based on one of

the dimensions.<sup>33</sup> These dimensions are color, shape, and number. The rule is changed after a set, or 10 consecutive correctly sorted cards, unbeknownst to the participant, who must figure out each new rule until the stack of cards is complete.<sup>32</sup> There are several measures of performance on the WCST as described in its manual, with most frequently used being the number of sets completed and various error measurements. Errors can be classified as perseverative, where the participant continues with the previous correct rule after the rule change despite negative feedback, or as failure to maintain set (FMS), where the participant commits an error after several correct responses despite positive feedback.<sup>34</sup> This task essentially measures the ability to shift mindsets flexibly depending on changing context.<sup>30</sup> In studies of WCST performance in autism, the greatest consistency is the impairment characterized by increased perseveration.<sup>32</sup> Cognitive flexibility is commonly measured in terms of perseverative errors and conceptual responses.<sup>30</sup>

#### ***1.4 Interventions***

Much of the treatment and support for those with ASD are geared toward improving basic characteristics, such as communication, and behavior characteristics, such as social skills.<sup>35</sup> Interventions starting as early as the child's second year of life have been linked with greater developmental gains and improvement in ASD symptoms compared to interventions given later in life.<sup>1</sup> No pharmacological treatments for core ASD symptoms, in the absence of co-occurring mental health and behavioral disorders, have yielded sufficient evidence for regulatory approval for use in clinical practice.<sup>9</sup>

There is much research supporting the use of peer-mediated interventions to target the core deficits of ASD.<sup>36</sup> With social difficulties limiting their opportunities to interact

with peers, children with ASD often have their family members being the most available people for communication.<sup>3</sup> Involving family members in delivering interventions has potential for improved generalization outcomes, learning opportunities, and development.<sup>1</sup> Siblings in particular have a special role as they function as a constant, daily source of social modeling that can significantly remediate the social deficits hallmark of ASD.<sup>19</sup> Older siblings often take on a teaching role, and these interactions can promote linguistic, cognitive, and emotional development.<sup>19</sup> Siblings provide children with ASD more opportunities to practice targeted skills in the home and other natural settings, which can improve long-term maintenance and retention of these skills learned from intervention.<sup>37</sup> Inclusion of siblings has been shown to not only enhance intervention effectiveness, but also promote relationship satisfaction within the pair.<sup>36</sup> Educating the typically developing sibling in mediation skills while fostering a positive sibling dynamic could have long term benefits for the whole family, especially in the early developmental period.<sup>19</sup>

One intervention that focuses on executive function impairments is the *Unstuck and On Target!* (UOT) curriculum, which is a cognitive behavioral intervention originally designed to be delivered through mainstream educational settings for children with ASD ages 8-11.<sup>11</sup> Being a verbally driven intervention, it is designed to serve those who have the cognitive verbal skills to benefit from higher order cognitive and social interventions.<sup>26</sup> Major components of the curriculum include teaching flexibility, its utility and importance, and associated skills through routines and scripts that are continuously practiced and reinforced until they are second-nature and automatic.<sup>26</sup> Some of the scripts included in these lessons were first developed for children with executive

function deficits resulting from traumatic brain injuries.<sup>11</sup> These lessons are first presented by a didactic class and then followed by activities such as role-playing or games.<sup>28</sup> The UOT curriculum has also been modified and demonstrated as acceptable in children with diagnoses other than ASD but with similar executive deficits in a community-clinic setting with co-occurring child and parent groups, despite not having enough power to detect statistically significant effects.<sup>38</sup> It has also been found as a useful tool to incorporate at home.<sup>26</sup>

### ***1.5 Statement of the Problem***

ASD is a lifelong developmental disability associated with impairments in executive function, including cognitive flexibility. These deficits contribute to maladaptive behaviors, as well as strained family functioning and sibling relationships. There is expanding evidence in the literature supporting the effectiveness of cognitive behavioral approaches geared towards behavior change and skill building. Although there are many studies encouraging the involvement of siblings in interventions for ASD, this has yet to be studied with the UOT curriculum, a promising intervention that targets executive function. Additionally, siblings offer potentially greater generalizability and longevity of intervention effects, an advantage that is missing from outside mediators. It is worthwhile to explore the inclusion of siblings as mediators in UOT and its effectiveness on cognitive flexibility skills as well as the quality of the sibling relationship.

### ***1.6 Goals and Objectives***

We propose this randomized control trial study in order to investigate the multidimensional effects of introducing typically developing siblings as mediators in a

modified UOT intervention. The primary aim is to determine if sibling inclusion in an established executive function intervention significantly improves cognitive flexibility outcomes in children with ASD. Outcomes will be measured through change in a composite WCST performance at baseline and follow-up by the children with ASD. The secondary aim of this is to investigate potential change in the quality of the sibling relationship using the self-report Sibling Relationship Questionnaire (SRQ) at baseline and follow-up by both participants in the sibling dyad.

### ***1.7 Hypothesis***

Among children aged 6 to 12 diagnosed with ASD, we hypothesize that participants randomized to the sibling-mediated UOT group will show significantly greater improvement from pre- to post-intervention in cognitive flexibility, as measured by the WCST, and sibling relationship quality, as measured by the SRQ, in comparison to the group receiving the UOT intervention without sibling mediation.

### ***1.8 Definitions***

*ASD*: Autism Spectrum Disorder.

*WCST*: Wisconsin Card Sorting Task.<sup>34</sup>

*UOT*: Unstuck and On Target!<sup>26</sup>

*SRQ*: Sibling Relationship Questionnaire.<sup>17</sup>

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## **Chapter 2 – Literature Review**

### ***2.1 Introduction***

A comprehensive literature search was conducted between December 2021 and July 2022 using PubMed, Ovid (Medline, APAPsycINFO), Cochrane Review, and Scopus. The studies, articles, and meta-analyses included in this literature review were analyzed for relevancy to the proposed research study based on their titles and abstracts. Key search terms used in each database to find literature pertinent to our study population include *autism spectrum disorder, ASD, autistic*, as well as *adolescents, school age*, and *children*. Search terms used to find studies on executive function interventions include *cognitive flexibility intervention, executive function intervention, EF intervention, Unstuck and On Target*, and *UOT*. The search terms used to find studies utilizing typically developing siblings as treatment mediators for this population include *sibling mediated intervention, SMI, sibling intervention, sibling mediator*, and *sibling support*. Several additional terms used for measurements of our proposed study's outcomes include *Wisconsin Card Sorting Task, WCST, Sibling Relationship Questionnaire*, and *SRQ*. These categories of our search terms were used in various combinations, utilizing the *and/or* functionalities of the databases stated above.

### ***2.2 Review of Empirical Studies***

#### ***2.2.1 Cognitive Flexibility Impairments in ASD***

Multiple studies have demonstrated the prevalence of cognitive flexibility impairments in the ASD population. One study investigating this executive function, as well as working memory, conducted cognitive assessments on 22 individuals with ASD, 14 typically developing siblings, and 15 age-matched control participants, with male to

female ratio not significantly different among the groups (chi-squared = 4.01).<sup>1</sup> Those in the ASD group met DSM-IV criteria for ASD, Asperger disorder, or pervasive developmental disorder not otherwise specified (PDD-NOS). Tests used to measure cognitive flexibility and working memory were the Wisconsin Card Sorting Task (WCST) and Verbal Learning Task (VLT), respectively.<sup>1</sup> The three variables analyzed for measuring WCST performance included number of categories achieved (CA), percentage of perseverative errors (%PEM), and reaction time (RT). Using multivariate analysis of variance (MANOVA) to examine the demographic variables and the aforementioned three performance variables, the study found that the ASD group had significantly worse scores on both the WCST and VLT in comparison to the other two groups. Analysis of demographic variables suggested that, since IQ levels did not significantly differ between groups, impairments in executive functions may be intrinsic to ASD, regardless of symptom severity.<sup>1</sup>

Another study examining executive function in individuals with ASD also investigated possible gender differences, given previous research demonstrating that women are less likely to be identified with ASD even if symptoms are equally as severe in their male counterparts.<sup>2</sup> This study, which included 99 men and 40 women with ASD compared to 35 neurotypical men and 25 neurotypical women, matched participants based on age, overall IQ, and verbal ability. Several instruments were used to measure executive functioning, including the WCST for cognitive flexibility. Using the number of perseverative errors, non-perseverative errors, and completed strategies as measurements of WCST performance, the results of MANOVA indicated that men and women with ASD performed worse in all measures than their neurotypical counterparts. Men with

ASD were also found to have significantly fewer perseverative errors than women with ASD.<sup>2</sup> Limitations of this study include that the participants had a higher end of average or above average IQ level, and were not matched on ASD symptom severity, both of which may influence assessment performance.

A different study that also focused on cognitive flexibility investigated for potential links to sociodemographic correlates, including age, gender, and education profile.<sup>3</sup> In a total sample size of 123 children aged 7 to 14 years and diagnosed with ASD, 94 boys and 29 girls were evaluated for cognitive flexibility patterns on the WCST. Participants were excluded if their IQ was less than 70. This study found that improved WCST performance was positively associated with child education level ( $p < 0.001$ ) and parental education ( $p < 0.001$ ).<sup>3</sup> A multivariate analysis of covariance (MANCOVA) was performed to investigate age and gender patterns of cognitive flexibility deficits in the participants. Strong evidence of gender differences was found ( $p = 0.009$ ) demonstrating that girls made significantly more perseverative errors ( $p = 0.012$ ) with fewer completed categories ( $p = 0.002$ ) compared to their male counterparts. There was no significant difference found between age groups for perseverative errors made ( $p = 0.07$ ).

One recent study that explored cognitive flexibility through computerized WCST performance included 14 adolescents with ASD and 22 adolescents without ASD or any other medical or genetic conditions, all of whom had an IQ between 50 and 85.<sup>4</sup> Although having a relatively small sample size, this study addressed the previously existing gap in literature regarding assessment of cognitive flexibility in autistic individuals with lower than average intelligence. Using one way analysis of variance (ANOVA) to compare WCST performance between the ASD and non-ASD groups, this

exploratory study found that there were no statistically significant differences between groups in the percentage of perseverative errors ( $p = 0.165$ ) and percentage of conceptual responses ( $p = 0.134$ ).<sup>4</sup> These results suggest there is a potential floor effect in terms of IQ for individuals with ASD that can affect measurements of cognitive flexibility, which should be considered for future studies investigating this executive function.

### ***2.2.2 Executive Function Interventions***

Many studies have found that, despite not being a hallmark deficit of ASD, executive functions (EF) are generally impaired at the group level in children with ASD, particularly in comparison with their typically developing (TD) peers.<sup>5</sup> Given their strong relationship with behavioral regulation in the context of adapting to new environmental stimuli, executive functions are a relevant and increasingly studied target for treatment.<sup>6</sup> Although there is no gold standard intervention for ASD symptoms, there is expanding evidence for cognitive training interventions to target EF impairments in this population.<sup>7,8</sup>

A systematic review of 19 studies, all either of a randomized control trial (RCT) or quasi-experimental design, evaluated the effectiveness of cognitive training programs on executive function and ASD core symptomatology.<sup>6</sup> A variety of clinical approaches were used within the studies, including the cognitive behavioral model, restorative techniques of cognitive remediation therapy, mindfulness practice, and cognitive enhancement programs.<sup>7,9</sup> This review found that in general, most EF interventions were beneficial in improving children's executive skills as well as quality of life in comparison to control activities or treatment as usual. One notable limitation in most of the reviewed studies is the focus of short-term training effects, as most studies took place over a period

of 2 to 8 weeks and assessed participants 1 week to 1 month after the end of training. This demonstrates a need for future research to include both immediate as well as follow-up measures to inform about the maintenance of treatment gains over a longer period of time. Another limitation was the heterogeneity of outcome measures that make it difficult to draw firm conclusions on training efficacy. Future studies should consistently use more standardized EF outcome measures and clinically relevant assessment tools. This systematic review also found that training programs have maximized efficacy when administered in therapeutic settings as well as daily life contexts.<sup>6</sup>

A quasi-experimental study assessed changes in WCST performance at baseline and after an intervention program consisting of 14 to 21 weekly language therapy sessions designed to target specific abilities related to cognitive flexibility.<sup>10</sup> Participants included ten children and adolescents with an age range of 5 to 13 years, diagnosed with ASD according to DSM-IV criteria. This longitudinal study lasted 3 years, with participants tested at baseline in 2012 to 2013, intervention phase taking place in 2014, and reassessment of the same measures post-intervention in 2015.<sup>10</sup> Statistical analysis was done with the non-parametric Wilcoxon matched signed-rank test in order to verify the change in WCST performance measures, and only three test items showed significant differences pre- and post-intervention: the number of perseverative errors (T1 M = 46.3; T2 M = 26.1;  $p = 0.028$ ), perseverative responses (T1 M = 58.6; T2 M = 31.9;  $p = 0.028$ ), and categories completed (T1 M = 2.1; T2 M = 3.8;  $p = 0.049$ ).<sup>10</sup> Although the results of this longitudinal study are significant and support internal validity, the major limitation of this study stems from its quasi-experimental, single-group design, which makes it difficult to draw causal conclusions regarding efficacy and external validity. A bigger

sample and experimental design would be more appropriate to generalize findings to the larger ASD population.

One randomized control study investigating a 12-week physical activity intervention, which comprised motor skill training and executive function training, also utilized change in WCST performance as a measure of cognitive flexibility.<sup>11</sup> There were 22 participants aged between 6 and 12 with a diagnosis of ASD according to DSM-IV criteria who were randomly split to an intervention and control group. ANCOVA results revealed that the intervention group performed significantly better compared to the control group on three WCST indices: perseverative responses ( $p < 0.01$ ), total correct ( $p < 0.01$ ), and conceptual-level response ( $p < 0.01$ ).<sup>11</sup> This randomized study was a significant addition to the literature of therapeutic options for children with ASD, however, an important limitation was the small sample size.

### ***2.2.3 Unstuck and On Target! Intervention***

The *Unstuck and On Target!* curriculum (UOT) was developed to improve executive function abilities, such as cognitive flexibility and goal directed behavior, and was designed for 8 to 11 year old students with ASD.<sup>12</sup> As this intervention was being designed, a preliminary study consisting of a small sample of 8 children was conducted, resulting in improvements in flexibility and collaborative behavior post intervention.<sup>12</sup>

A randomized control trial investigated the effectiveness of UOT compared to an established social skills (SS) curriculum in improving executive function in 3<sup>rd</sup> to 5<sup>th</sup> graders with ASD.<sup>7</sup> Participants were excluded if they did not meet the threshold of an IQ greater than 70 and verbal mental age of greater than or equal to 8, which were measured with the *Wechsler Abbreviated Scale of Intelligence* (WASI). Participants were also



excluded if they did not meet the diagnostic cutoff on the *Autism Diagnostic Observation Schedule* (ADOS). After evaluation for inclusion and eligibility, randomization occurred at the level of the school and not the individual participant to prevent cross-contamination of the two treatments. Participants were matched at the start of the intervention for autism symptomatology, age, parents' education, IQ, minority status, and medication status. A total of 10 schools with 47 students were assigned to the UOT intervention, and 4 schools with 20 students were assigned to the SS intervention. Both interventions were carried out in 28 sessions, each lasting 30 to 40 minutes, during a single school year and delivered by school staff within a mainstream educational setting. Teachers and parents had training sessions to reinforce the lessons specific to the intervention. Study evaluators were blinded to outcome data, which included a collection of direct child measures pre- and post-intervention. These measurements included the *WASI Block Design* task to assess problem solving, and the *Challenge Task* to assess flexible and planful behavior. Parent and teacher report measures were unblinded and included the Behavior Rating Inventory on Executive Function and the Social Responsiveness Scale. Results revealed that while both intervention groups improved, there were significantly greater improvements in measures of problem solving, flexibility, planning, and organizing from the UOT intervention compared to the SS intervention ( $p < 0.05$ ), with a medium-large effect size. When compared to the SS participants, there was more improvement observed in the UOT participants' ability to follow directions ( $p < 0.001$ ; UOT = 65.2% improved), transition smoothly ( $p < 0.001$ ; UOT = 63% improved), and avoid getting stuck ( $p < 0.05$ , UOT = 47.8% improved). The participants in the UOT group also generalized greater improvements in classroom behavior observations compared to SS participants, including

a reduction of negativity in the classroom ( $p = 0.05$ ; UOT = 39.1% improved). This study provided evidence that UOT is an effective EF intervention for children with ASD. Limitations of this study include a relatively small sample size, lack of longitudinal follow up to investigate changes preserved over time, and no isolation of specific modules of UOT.<sup>7</sup> Another notable limitation was a lack of test-retest reliability, validity, and normative data on the *Challenge Task*, which at the time of the study was an unpublished and un-normed assessment designed by the authors in order to measure cognitive flexibility skills.

Another study evaluated the feasibility and preliminary efficacy of a UOT intervention protocol modified for an outpatient clinic setting with a diagnostically diverse group of participants. Eligibility criteria included an IQ greater than 80 on WASI-II, verbal fluency, and parent-reported difficulties with EF, with no formal diagnosis required for participation. In the sample of 6 children aged between 8 and 11 who completed the full intervention, there were 3 children with ASD, 1 child with attention deficit hyperactivity disorder (ADHD), 1 child with post-traumatic stress disorder (PTSD), and 1 child with history of traumatic brain injury (TBI). In a single subject case design where each participant served as their own control, the modified UOT intervention was delivered in 10 weekly group therapy sessions lasting 90 minutes each, with concurrent child and parent groups. Each session represented one thematic topic of the published UOT protocol, with praise and tangible behavioral reinforcers, such as play money and prizes, added to the child protocol. The concurrent parent sessions provided psychoeducation to assist parents in understanding and reinforcing skills learned in the child group, in order to maximize intervention effectiveness and promote its

generalization to the home setting. Parent report measures included the child behavior check list (CBCL), a 118-item assessment which determines the range of behavioral and emotional problems rated on a 3-point Likert scale and has been utilized with children of both typical and atypical developmental trajectories. Clinician administered performance tasks of the Delis-Kaplan Executive Function System (DKEFS) included trail-making to assess flexibility, color-word interference test to assess inhibition and switching, and card sorting task to assess flexible thinking and behavior. This study found the UOT intervention, which was modified to fit a transdiagnostic patient population administered in an outpatient setting, to be feasible, defined through treatment fidelity, participant compliance, and participant satisfaction. The study did not have adequate power to detect significant effects despite demonstrated feasibility.<sup>13</sup> Preliminary efficacy data showed reliable increase in EF for three participants across parent-report measures. Group differences on the CBCL indicated reductions in overall behavioral and emotional problems post-treatment. The three DKEFS tasks demonstrated no significant changes on the group level. The transdiagnostic participant sample served as a strength as the diversity maximized the ecological validity of the study, but also served as a weakness as it limited the ability to ascertain clinically significant change that resulted from the intervention itself. Other limitations of this study include the small and heterogeneous sample, lack of a comparison group or control, lack of treatment-naïve clinical improvement and severity ratings, and mixed results.<sup>13</sup>

Despite only having two published studies in the available literature, UOT is an effective and promising intervention for youth with ASD. Future studies can address the current gap by using larger sample sizes, established and norm-referenced assessments as

outcome measures for executive function skills, and follow-up probes to investigate maintenance of the intervention results.

#### ***2.2.4 Sibling Relationship***

For many individuals, the bond with a sibling is the most enduring one a person can have, and the longest lasting of all immediate familial relationships.<sup>14,15</sup> For a person with a disability, this relationship is especially unique as their sibling takes on several roles including a playmate, friend, and teacher in childhood, and eventually an advocate and caretaker in adulthood.<sup>15</sup> These extra responsibilities assumed by the child coupled with increased demands that cut into parental availability for that neurotypical sibling, however, may give rise to problems or strains in family functioning.<sup>16,17</sup> In an early study investigating the sibling relationships with disabled versus nondisabled siblings, it was found that children with disabled siblings take on more household and sibling caregiving responsibilities ( $p < 0.01$ ), and recalled more negative behavior from their mothers ( $p < 0.01$ ) compared to children with non-disabled siblings.<sup>16</sup> In this study, typically developing (TD) children also self-evaluated their satisfaction with the sibling relationship in a 5-item questionnaire. Only one significant group effect was found, where children with disabled siblings reported they were happier with how they got along with their siblings, compared to children with non-disabled siblings ( $p < 0.01$ ).<sup>16</sup> None of the disabled children in this study, however, had a formal diagnosis of ASD, which is our proposed study's population of interest.

In a study investigating sibling perception of their relationships, the total sample size of 90 TD participants between the ages of 8 to 18 were split into three even groups of 30: those with a sibling diagnosed with ASD, those with a sibling diagnosed with Down

syndrome, and those with a sibling with no known disability.<sup>18</sup> This study utilized the brief version of the *Sibling Relationship Questionnaire* (SRQ), a 39-item assessment that evaluates four key factors of closeness/intimacy, power, conflict, and rivalry.<sup>19</sup> SRQ factor scores were analyzed in relation to the participant's group and gender using MANOVA, with a significant overall effect found for both group ( $p < 0.001$ ) and gender ( $p < 0.05$ ).<sup>18</sup> Results found that participants with a sibling who has ASD report less intimacy ( $p < 0.01$ ), less nurturance ( $p < 0.01$ ), and less prosocial behavior ( $p < 0.05$ ) compared to those who have siblings with Down syndrome or without a disability. The variety of social and communication deficits, which are characteristic of autism, as well as limitations in cognitive functioning, are strongly believed contribute to these findings. For sibling gender, there was only a significant trend found on the conflict factor of the SRQ, with males reporting more conflict with their siblings than females among all groups.

One recent RCT evaluated the effect of a sibling support group on the quality of the sibling relationship, in comparison to an attention-only control group.<sup>20</sup> TD siblings attended either program for 10 weeks, and data was collected in six different cohorts over the course of 3 years. Out of the 44 participating families, which included 54 TD siblings and their 44 siblings with ASD, there were 24 TD siblings randomized to the support group and 30 TD siblings randomized to the control group. The intervention sibling support group focused on providing TD siblings psychoeducation about ASD, problem-solving skills, and coping strategies. Sibling relationship quality included a TD sibling self-report of a modified 33-item version of the SRQ for siblings (SIB-S), with higher scores indicating a more positive perception of the relationship.<sup>19</sup> Results of the mixed

ANOVA found that the SIB-S was significantly higher post-intervention ( $M = 84.27$ ) than pre-intervention ( $M = 73.62$ ) for the support group ( $p < 0.001$ ), compared to the control group (T1  $M = 79.24$ ; T2  $M = 75.83$ ).<sup>20</sup> These self-reported improvements in relationship quality were confirmed with observational measures in the context of sibling play. These findings strongly suggest that sibling relationship quality is related to TD sibling knowledge not only about ASD, but also on how to implement problem-solving and coping strategies.<sup>20</sup> Because improving the sibling relationship early in development can lead to positive health and social outcomes for each sibling and overall family functioning, the inclusion of TD siblings in interventions for children with ASD holds promise as an effective treatment option with multidimensional benefits.

### ***2.2.5 Sibling Mediated Interventions In ASD***

While a large majority of literature demonstrates adults as intervention agents, there is empirical support for similarly aged, neurotypical peers to implement treatment for individuals with ASD. Peer mediated interventions have been found to result in increased opportunities for socialization, academic gains, independence, higher self-confidence, and expanded peer networks and relationships.<sup>21,22</sup> One significant relationship is that between siblings, as a sibling is often the first peer to whom a child is exposed as well as the first model of interpersonal characteristics.<sup>23,24</sup> The inclusion of siblings as mediators in ASD interventions has not been as common in the literature compared to parent- or peer-mediated interventions, despite siblings being recognized as potential agents of change.<sup>15,25</sup> They are a readily available source of social interaction who can provide a frequent practice schedule for the individual with ASD to develop skills that can further be generalized to peers in other settings than the home setting, such

as school or community.<sup>21,26,27</sup> Sibling involvement in interventions has been demonstrated to have twofold benefit: while children with ASD show improvements in their social, play, or functional skills, their typically developing siblings learn the skills of mediation.<sup>22,27-29</sup> Researchers have hypothesized that sibling participation in interventions can strengthen the sibling relationship, with anecdotal evidence from siblings and parents supporting this.<sup>21</sup> Sibling mediated interventions can foster positive interactions which can lead to long-term care and advocacy.<sup>23</sup>

In a non-concurrent multiple baseline study across child-sibling pairs, typically developing siblings were taught the Natural Language Paradigm (NLP), a play-based speech intervention that incorporates turn-taking, task variation, and highly motivating activities to work towards language acquisition.<sup>30</sup> Prior to this study, the NLP intervention implemented by trained clinicians resulted in an increase in vocal chains and appropriate verbal behavior, and a decrease in maladaptive behavior in children with ASD. In separate studies, parents were also taught to implement NLP through observation, brief training sessions, and practice with feedback, resulting in children with ASD demonstrating improvements in speech and appropriate play. In this study, three sibling dyads were recruited from a behavior management program that the participants with ASD attended on a weekly basis. The children with ASD were between 6 to 9 years old, diagnosed with ASD according to DSM-V criteria, and also displayed a speech/language impairment. Their TD siblings were between 7 to 11 years old. There were several video-recorded baseline sessions where the TD sibling was instructed to “go play and talk” with the sibling with ASD, with a varied number of sessions for each dyad in order to attribute changes in verbal behavior to the intervention. The sibling training

then included the videos of two therapists modeling NLP, which were tailored specifically to the individual level of speech of the child with ASD, and checklists to test the TD sibling periodically for comprehension. The TD siblings were then reintroduced to their sibling with ASD in an NLP treatment session to implement what was seen in the training video. The two sets of dependent measures in this study included sibling verbalization and increases in language production of the children with ASD. Follow up probes were conducted 8 to 12 weeks post-intervention for the two sibling dyads who met learning criterion during intervention. The learning criterion was defined as the average percentage of verbal behavior at least doubled from baseline across two consecutive sessions. This study showed that TD siblings were able to effectively learn NLP and utilize the intervention to appropriately occasion speech from their sibling with ASD. Additionally, two sibling dyads were able to correctly implement NLP in the follow up probes, indicating that sibling implementation of NLP can be maintained over time. Limitations of this study included the limited sample, narrow range of ages, no female children with ASD in this study, short follow up, and intervention conducted only in clinic rather than home settings.<sup>30</sup>

Another concurrent multiple-baseline study trained TD siblings on play strategies to increase positive sibling play in combination with a sibling support group to offer social support for the TD sibling.<sup>31</sup> There were six dyads of siblings in this study, which consisted of baseline sessions, behavioral skills training (BST) sessions for the TD siblings, sibling-implemented play intervention sessions with support group, and maintenance sessions two weeks after intervention for three of the six dyads. This study aimed to determine if there was a functional relationship between the TD sibling BST



focused on play facilitation strategies, and increases in several dependent variables including percentage of time spent in reciprocal play with their sibling with ASD; frequency of TD sibling initiations; and sibling fidelity of intervention implementation. These sessions occurred face-to-face at home or clinic, until switching to telehealth delivery of the intervention package due to the COVID-19 pandemic. The results of this study indicated that, although none of the TD siblings ever reached 100% fidelity of implementation by using all of the strategies most of the time, the brief BST sibling training effectively increased TD sibling use of targeted play strategies and positive reciprocal play between the siblings. The addition of the three-week sibling support group was also highly rated by both the parents and the TD siblings as being beneficial. Limitations of this study included the attrition associated with the switch to telehealth delivery due to the COVID-19 pandemic that limits the internal validity of the study; the low dosage of, and lack of participation in, the sibling support group; lack of social validity measures completed by the sibling with ASD; and lack of long-term maintenance follow-up probes while fading out a dense reinforcement schedule.

In a meta-analysis of studies utilizing siblings as mediators in interventions for their brothers and sisters with ASD, it was found across 16 studies ( $n = 43$ ) that sibling-mediated interventions can be used to teach new or improve on functional skills in the targeted behavior areas.<sup>28</sup> Additionally, the TD siblings were adequately trained in mediation skills that were helpful in mitigating undesired behaviors. Data collection was varied in methodology and included satisfaction surveys, blinded or naïve observers, and interviews. This meta-analysis calculated an overall effect size with non-overlap of all pairs (NAP) methodologies to be 0.83 with a 95% CI [0.76, 0.90], interpreted as a

medium effect size for sibling-mediated interventions. One common limitation found across the reviewed studies was that, despite the effectiveness of the interventions for both children in the sibling dyad, many studies did not assess follow-up. Another limitation was that the roles of individual siblings were not clearly defined in some intervention phases.

A different meta-analysis looked at sibling involvement in therapeutic interventions for individuals with a disability, with 28 out of 31 studies focusing on participants with ASD.<sup>15</sup> This review highlighted the variety of roles that siblings would play in interventions, and a majority of the studies utilized siblings as a playmate, usually receiving some form of training. In studies where specific skills were to be targeted, siblings would fill the role of an instructor, trained specifically to elicit a certain response from the child with ASD. The role of instructor also involved giving feedback, cueing, and specific prompts. This systematic review found that under half of the studies had recorded participant experience or opinion of the study from either the individual with a disability or their sibling, and the studies that did record participant experiences only reported those of the TD sibling.<sup>15</sup>

Sibling mediated interventions have also been explored in other diagnoses, with results demonstrating positive outcomes in young children with ADHD,<sup>32</sup> Down syndrome, Noonan syndrome, speech motor delay, and developmental delay.<sup>15</sup>

### ***2.3 Review of Studies to Identify Possible Confounding Variables***

In this literature review, we identified several potential confounding variables that may threaten the internal validity of our proposed study. Similar to previous studies, these variables include sex, age, symptom severity, education level, and IQ. Females

have demonstrated lower performance in WCST measures compared to their male counterparts.<sup>2,3</sup> Higher levels of education, of both the children and parents, has previously found to be associated with decreased perseveration.<sup>3</sup> Participant differences in age and intellectual ability have been found to increase the variability in EF performance.<sup>6</sup> Our RCT study design will minimize the potential confounding effects that cannot be realistically controlled for through strict exclusion criteria. Difference between the intervention and control groups will be adjusted for with a multivariate analysis of covariance (MANCOVA) similar to previous studies. Other potential confounders, such as medication status and presence of comorbidities, will be accounted for by restricting the eligibility criteria of subjects. All participants with ASD who have an IQ below 80 as measured by the WASI will be excluded from this study as this has been found to interfere with WCST performance in a previous study.<sup>4</sup>

#### ***2.4 Review of Relevant Methodology***

Our proposed study will be a two-arm, single-blinded RCT investigating the effects of a sibling-mediated UOT intervention compared to the standard UOT intervention alone for 1 school year. An RCT design was chosen as RCTs are considered the gold standard for clinical research, and randomization will reduce the potential for bias that is present in other study designs. Additionally, the previous study investigating the effectiveness of UOT also utilized an RCT design and compared this curriculum to a standard social skills intervention in a classroom setting. The only other published study by Elias et al. involving UOT evaluated its feasibility and preliminary efficacy with a multiple baseline design.<sup>13</sup>

Participants enrolled in this study will be assigned to one of two groups: sibling-mediated UOT intervention group, or the UOT intervention group without sibling inclusion. Randomization techniques will be similar to that of the study by Kenworthy et al., where randomization occurred at the level of the school rather than the individual, to prevent cross-contamination of the two treatment groups.<sup>7</sup> Our intervention will be given at the Yale Child Study Center over the span of one school year, similar to the study by Kenworthy et al., with concurrent training sessions for the TD siblings in the dyads assigned to the intervention group.<sup>7</sup> Similar to the concurrent parent groups in the study by Elias et al., these sibling training sessions will provide psychoeducation on ASD phenomenology, UOT content, and reinforcement skills for behaviors targeted by UOT.<sup>13</sup>

Individuals will be considered for our study if they are aged 8 to 11, have been diagnosed with ASD according to DSM-V criteria, have an IQ greater than 80 according to WASI scoring, and have a TD sibling with an age difference of no more than 5 years to reduce possible influences of developmental age differences. Potential participants will also be evaluated with the ADOS, a standardized activity-based assessment that is commonly used in clinical studies to evaluate for and confirm a diagnosis of ASD.<sup>33</sup> This semi-structured, interactive assessment consists of eight tasks designed to elicit certain behaviors characteristic of ASD, which can be found in Table 2.<sup>34</sup> Other measures that provide autism severity ratings, such as the Childhood Autism Rating Scale and the Autism Behavior Checklist, tend to yield scores that are strongly correlated with IQ or do not correspond to standard measures of diagnosis.<sup>35</sup>

**Table 2. Components of Autism Diagnostic Observation Schedule**

| <b>Task</b>                       | <b>Target Behavior(s)</b>                                      |
|-----------------------------------|--|
| Construction task                 | Asking for help  |
| Unstructured presentation of toys | Symbolic play<br>Reciprocal play<br>Giving help to interviewer |
| Drawing game                      | Taking turns in a structured task                              |
| Demonstration task                | Descriptive gesture and mime                                   |
| Poster task                       | Description of agents and actions                              |
| Book task                         | Telling a sequential story                                     |
| Conversation                      | Reciprocal communication                                       |
| Socioemotional questions          | Ability to use language to discuss socioemotional topics       |

*Source:* Lord C, Rutter M, Goode S, et al. Autism Diagnostic Observation Schedule: A Standardized Observation of Communicative and Social Behavior. *Journal of Autism and Developmental Disorders*. 1989;19(2):185-212.

Participants will be recruited through New Haven County schools, Yale Child Study Center, and Yale pediatrician outpatient clinics. Participants with ASD who have co-occurring disorders, an IQ less than 80, are taking pharmacological and/or psychotropic medications, or are undergoing other interventional studies, will be excluded from our study due to potential interference with the validity of our results.<sup>4</sup> Medication classes previously demonstrated to affect cognitive performance include antipsychotics, psychostimulants, antidepressants, and anticonvulsants.<sup>36</sup> Many psychotropic medications target the dopaminergic neurotransmitter pathway, and weakening the effect of dopamine was found to greatly reduce the ability to flexibly adapt behavior to changing tasks in WCST.<sup>37</sup>

Our primary outcome variable will be change in cognitive flexibility as measured by WCST performance pre- and post-intervention. As noted in our literature review, the

WCST is a standardized, norm-reference assessment of cognitive flexibility that has been widely used in clinical studies involving children with ASD, who consistently demonstrate increases in perseverative errors and conceptual responses.<sup>38</sup> The study by Kenworthy et al. which investigated UOT measured its effect on cognitive flexibility with the *Challenge Task*, an assessment created by the authors of the study without normative data for retest reliability or validity.<sup>7</sup> Using a well-normed test such as the WCST to measure changes in cognitive flexibility allows us to draw conclusions from the results with confidence.

Our secondary outcome variable of interest will be change in the quality of sibling relationship as measured by the SRQ pre- and post-intervention. This questionnaire has demonstrated decreased intimacy and nurturance among siblings when one child has ASD, compared to sibling dyads without ASD.<sup>18</sup> Of note, the perception of the sibling relationship on the self-report questionnaire has shown to improve after involving TD siblings in interventions, an important finding that calls for more studies to include TD siblings in interventions for children with ASD.<sup>20</sup> The full SRQ can be found in Appendix E.

## **2.5 Conclusion**

The studies covered in our literature review collectively provide the basis for our hypothesis and justification for our proposed protocol, which will be provided in detail in the following chapter. While UOT has been shown to be efficacious at improving measurements of cognitive flexibility in children with ASD, this intervention has yet to be investigated with the inclusion of siblings as mediators. With the expanding evidence of siblings being promising agents of interventions with multiple benefits for this clinical

population, our proposed study will add to the current literature a novel intervention protocol for children with ASD.

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## **Chapter 3 – Study Methods**

### ***3.1 Study Design***

The proposed study will be a two-arm, single blinded randomized control trial (RCT) to investigate the effects of sibling inclusion as mediators to the *Unstuck and On Target!* (UOT) intervention, compared to the standard UOT intervention without sibling mediators. We will randomize participants to the intervention or control group. Due to the nature of the intervention, it will not be possible to blind participants. Our study aims to primarily examine the change in Wisconsin Card Sorting Task (WCST) performance as a measure of cognitive flexibility pre- and post-intervention. The secondary aim of this study is to evaluate for changes in the relationship quality between siblings using the self-report Sibling Relationship Questionnaire (SRQ) pre- and post-intervention.

### ***3.2 Study Population and Sampling***

The study population of interest will be children aged 6 to 12 years, diagnosed with autism spectrum disorder (ASD) according to DSM-V criteria, with a typically developing sibling with an age difference of no more than 5 years. Potential participants with ASD will be assessed individually with the *Autism Diagnostic Observation Schedule* (ADOS) to evaluate for ASD, and the *Wechsler Abbreviated Scale of Intelligence* (WASI) to measure IQ. Participants will be included in our study if they meet DSM-V diagnostic criteria for ASD, ADOS threshold for ASD, and achieve an IQ score greater than 70. Participants with ASD are not eligible to participate in this study if they have a history of co-occurring disorders such as attention deficit hyperactivity disorder (ADHD), are taking pharmacological therapies for ASD associated symptoms including psychotropic medications, or are undergoing other interventional studies. Typically

developing siblings are not eligible to participate in this study if they have a history of intellectual disability, developmental disability, or any condition with significant cognitive impairment.

### ***3.3 Subject Protection and Confidentiality***

Our proposed research protocol will be submitted to the Yale Institutional Review Board (IRB) and Human Subjects Committee for approval prior to the start of the study. All research personnel will be required to complete the Yale Human Subject Protection and Health Insurance Portability and Accountability Act (HIPAA) compliance training modules prior to the start of the study. Certificates of completion for each mandatory training module must be provided to Yale IRB to ensure protection of all participant health information. All personal participant data and records will be kept on an encrypted computer system accessible only to study investigators. At the conclusion of the study, all participant data will be destroyed.

The policies, required activities, and confidentiality agreement of the proposed study will be reviewed thoroughly with both participating children and their primary caregiver. Parents or guardians of the participants will provide written, informed consent through the parent permission form after discussion of all procedures, risks, and benefits of the study. All participating children will provide their assent through the child and adolescent assent form. Research consent and assent forms can be found in Appendix B and C, respectively.

### ***3.4 Recruitment***

Sibling dyads will be recruited through New Haven County schools, Yale pediatrician offices, and the Yale Child Study Center. We will reach out to participants

using IRB approved advertisements in flyers, posters, local newspapers, email listservs, and word of mouth by clinical healthcare providers in the community. See Appendix D for a sample recruitment flyer. Prior to enrollment, all participating children will be screened and assessed by ASD-expert clinicians.

### ***3.5 Study Variables and Measures***

In our proposed RCT, the independent variable will be our sibling-mediated UOT intervention. Participant dyads, comprising the child with ASD and the TD sibling, will be randomly assigned to one of two groups: sibling-mediated UOT group, and UOT group without sibling mediation.

The primary dependent variable will be change in cognitive flexibility, as measured by the WCST.<sup>1</sup> Specifically, the WCST measures of interest will be perseverative errors and conceptual responses.

The secondary dependent variable will be the change in sibling relationship quality, as measured by the self-report SRQ.<sup>2</sup> The SRQ consists of multiple scales that are grouped into four factors: Closeness/Intimacy, Power, Conflict, and Rivalry. The full questionnaire can be found in Appendix E.

### ***3.6 Blinding***

While it is not possible to blind participants with ASD and their TD siblings to the intervention, the research staff responsible for assessing the primary and secondary outcomes will be blinded to participant group allocation in order to minimize the possibility of information bias.

### ***3.7 Assignment of Intervention***

After baseline screening and evaluation, eligible participants and their TD siblings will be randomly assigned either to the intervention group or to the control group. One member of the research team will use a computerized randomization software to randomly assign sibling dyads, and will not be involved in the remainder of the study. Research staff measuring outcome assessments will be blinded to group allocation.

### ***3.8 Adherence***

Attendance of all participants will be taken by study investigators during bimonthly group sessions with sibling dyads for both groups. To encourage attendance, travel expenses to the Yale Child Study Center for the bimonthly sessions will be reimbursed.

Trained research staff observing intervention sessions for fidelity will monitor participant engagement as well as interventionist adherence to UOT curriculum content.

### ***3.9 Data Collection***

Primary and secondary outcomes will be collected at baseline pre-intervention, immediately post-intervention, and at a 6-month follow-up probe by blinded study investigators to assess maintenance.

### ***3.10 Sample Size Calculation***

This study will utilize a two-sided hypothesis with a statistical significance of alpha of 0.05 and power of 80%. We will base our sample size calculation on the analysis of perseverative responses from the previous study by Pan et al.<sup>3</sup> Using the BioStat Power and Precision 4.1 tool, our estimated sample size was initially calculated to be 32 participants in total. Our complete sample size calculation can be found in Appendix A.

Based on prior studies, we will account for an estimated 20% attrition rate, giving an adjusted sample size of 40 participants in total, with 20 participants with ASD in each group.

### ***3.11 Analysis***

Similar to prior studies, the primary outcome (change in WCST composite performance) and secondary outcome (change in SRQ factor scores) will both be analyzed using multivariate analysis of variance (MANOVA) in relation to the demographic variables identified as potential confounders as outlined in our above literature review.

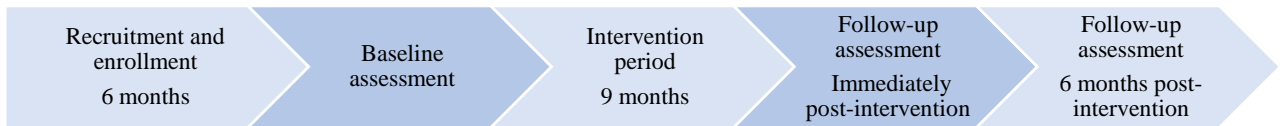
### ***3.12 Timeline and Resources***

Our proposed study will take place within the span of 2 years, from recruiting participants to data analysis of follow-up probes. See Figure 1 for the proposed study timeline. Recruitment and enrollment of participants will take place over the first 6 months, with research assistants at Yale University to collect baseline assessments and questionnaires to determine participant eligibility.

Additionally, all research staff will attend mandatory training sessions to prepare for the study prior to the intervention period. These required training sessions will include Yale HIPAA compliance modules, ASD phenomenology, UOT intervention content and delivery. Treatment integrity will be ensured through bimonthly intervention observations by a trained research staff member and observer ratings of curriculum fidelity. Fidelity checklists will include adherence to intervention content as well as participant engagement. Interventionists will also be given individualized fidelity feedback after each session.

The intervention period will take place over the span of one school year, which is approximately 9 months. Intervention will be carried out at the Yale Child Study Center in bimonthly sessions with the exception of holiday breaks, in a total of 14 sessions lasting 1 hour each.

**Figure 1. Study Timeline**



### **3.13 References**

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## Chapter 4 - Conclusion

### *4.1 Advantages*

Our research proposal provides the background and justification for a study that will add a novel intervention with many advantages to the existing body of research for our clinical population of interest. The prevalence of autism spectrum disorder (ASD) has increased in the past few decades, demonstrated to affect 1 in 44 children aged 8 years in the United States.<sup>1</sup> Aside from individual challenges with executive function and adaptive behavior, ASD can also lead to difficulties with family functioning, such as strained sibling relationships.<sup>2</sup> Our proposed study would provide a way to combine the *Unstuck and On Target!* (UOT) curriculum with the inclusion of typically developing (TD) siblings as mediators, two established and promising interventions for children diagnosed with ASD.<sup>3,4</sup> A notable and important strength of this study is that, by utilizing and involving TD siblings in the treatment program, the teachings and skills gained from this intervention can be generalized to the home setting with improved maintenance and longevity given that siblings are often a long-lasting if not permanent fixture in an individual's life.<sup>5</sup>

An additional advantage of our study is the utilization of a norm-referenced assessment, the WCST, to measure the efficacy of our proposed intervention. The only other RCT to investigate the UOT curriculum assessed cognitive flexibility with the *Challenge Task*.<sup>3</sup> This was an unpublished task designed by the authors at the time of the study, which calls the validity of the results into question given the lack of test reliability. By using an established standardized task such as the WCST which has been widely used



with our clinical population of interest, the results of our proposed study will have good ecological validity and reliability.

Furthermore, this study will assess outcomes not only immediately post-intervention, but also at a 6 month follow up probe after intervention completion. Lack of long-term follow-up for outcome measures was a consistent gap in research studies in this population as noted in our literature review.<sup>3,6-8</sup> Several studies supporting the involvement of TD siblings in interventions only assessed for and demonstrated short-term skill gains for the individuals with ASD with no longitudinal follow up.<sup>8</sup> Because siblings may continue to utilize intervention strategies in the home setting following intervention completion, there is a critical need for additional investigation into this possible maintenance effect. Our proposed research protocol will build on existing literature by measuring the multidimensional impact of our novel intervention and provide direction for future studies regarding the long-term outcomes of a sibling-mediated executive function intervention.

Another advantage of our RCT study is the inclusion of a control group to serve as a comparison for the intervention group. In a 2021 systematic review of sibling-mediated interventions, it was found that only a few studies had utilized a control group to evaluate the overall effects of proposed intervention, which may undermine confidence in the reliability of the reports of effectiveness.<sup>4</sup> An RCT protocol allows for the randomization of participants to limit the influence of confounding variables and minimize possible biases that are inherent in alternative study designs.

## ***4.2 Disadvantages***

Although our proposed intervention and study design has many advantages, there are some disadvantages to be considered. Because our recruitment methods utilize convenience sampling, the findings of our study will ultimately be limited by potential selection bias. Furthermore, the recruitment of participants only from selected school districts and the Yale Health system within the state of Connecticut may potentially lower the external validity of our results, as our sample may not be completely representative of the entire population of children with ASD.

Another limitation of our study lies with the single-blinded design. Although it is not possible to blind participants to their group allocation due to the nature of the intervention, a single-blinded design presents the potential for participant bias. We hope to minimize the effect of participant bias by blinding all children to the hypothesis of our study. Additionally, all research staff involving in measuring and collecting outcome data will be blinded to participant group allocation.

An additional disadvantage to consider is that, by using a self-report questionnaire such as the SRQ to measure the quality of the sibling relationship, there is potential response bias that can limit the internal validity of our findings. Future studies may benefit from including objective assessments, such as behavioral observations and reports by blinded research personnel, to measure this outcome.

## ***4.3 Clinical Significance***

With the increasing prevalence of ASD diagnosis, there is imperative to provide effective intervention in early development given the negative impact of associated executive function deficits not only on an individual's daily functioning, but also on

familial relationships.<sup>9,10</sup> The existing literature strongly supports interventions that focus on improving executive function, such as the UOT curriculum, as well as the inclusion of TD siblings as mediators.<sup>3,4,9</sup> By combining these two effective modalities, this RCT study will be clinically important as it will serve to provide quantitative results on the benefits and efficacy of a sibling-mediated UOT intervention. Given the longitudinal design of our proposed study, we will also be able to determine the long-term effects of our intervention in an additional follow-up probe 6 months after intervention completion, which has been a consistent gap in prior literature for this clinical population. Compared to outside mediators, a TD sibling who represents the longest relationship for an individual can uniquely implement a behavioral intervention such as UOT with greater generalizability and maintenance after intervention completion.<sup>11</sup> If our findings demonstrate significant improvement for cognitive flexibility outcomes as well as sibling relationship quality, our proposed study will add a promising, novel treatment option with multidimensional benefits for children with ASD to the existing body of research.

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# Appendices

## Appendix A: Sample Size Calculation

✓ Power And Precision 4 - [t-test for two independent samples with common variance]

File View Options Tools Scenarios Help



| Group                  | Population Mean | Standard Deviation | N Per Group | Standard Error | 95% Lower | 95% Upper |
|------------------------|-----------------|--------------------|-------------|----------------|-----------|-----------|
| Intervention           | 22.0            | 3.7                | 16          |                |           |           |
| Control                | 18.1            | 3.7                | 16          |                |           |           |
| <b>Mean Difference</b> | 3.9             | 3.7                | 32          | 1.31           | 1.26      | 6.54      |

Alpha= 0.050, Tails= 2      Power

**Welcome**

This interactive guide will lead you through the steps for computing power and precision.

To move this box use the title bar above.

To close or reactivate this panel, select Help from the menu.

  >

**Summary - Power**

For the given effect size (population means of 22.0 vs. 18.1), SD (3.7), sample sizes ( 16 and 16), and alpha (0.050, 2-tailed), power is 0.822.

This means that 82% of studies would be expected to yield a significant effect, rejecting the null hypothesis that the two population means are equal.

    >

## *Appendix B: Parent Consent Form*

### **COMPOUND AUTHORIZATION AND CONSENT FOR PARTICIPATION IN A RESEARCH PROJECT YALE UNIVERSITY SCHOOL OF MEDICINE – YALE-NEW HAVEN HOSPITAL**

**Study Title:** *Sibling Mediation in an Executive Function Intervention for Children with Autism Spectrum Disorder*

**Principal Investigator:**

**Funding Source:** *Yale Child Study Center*

#### **Invitation to Participate and Description of Project**

You are invited to take part in a research study designed to look at the effectiveness of an executive function intervention for children with the autism spectrum, designed to include their typically developing siblings as intervention partners or mediators.

In order to decide whether or not you wish to be a part of this research study you should know enough about its risks and benefits to make an informed decision. This consent form gives you detailed information about the study, which a member of the research team will discuss with you. This discussion should go over all aspects of this research: its purpose, the procedures that will be performed, any risks of the procedures, possible benefits and possible alternative treatments. Once you understand the study, you will be asked if you wish to have your children participate; if so, you will be asked to sign this form.

#### **Description of Procedures**

If you agree to take part in this study, you and your children will first be asked to complete some interviews and questionnaires. These measures will ask questions about your children's knowledge about autism spectrum disorders, and the quality of relationships within your family. Your child with autism will also be asked to complete a card sorting task. After completing these measures, your children will either be assigned to participate in the intervention group, or will be placed in a control group. The control group is our comparison group, which allows us to determine whether receiving the executive function intervention with sibling involvement is more effective than receiving it without sibling involvement. Assignment to the control group is done randomly, and is necessary for the quality of our research. The intervention, called *Unstuck and On Target!*, is an established curriculum designed for children to improve executive function. The intervention sessions will last for one school year on a bi-monthly basis. Typically developing siblings in the intervention group will attend concurrent education sessions in a separate room at the Yale Child Study Center, where teaching will focus on education on autism and how to reinforce the lessons taught in *Unstuck and On Target!* After the intervention is over, you and your children will be asked to complete same measures that

you completed initially. We will then follow up with you and your children to complete the same measures 6 months after the end of the intervention period.

### **Risks and Inconveniences**

There are minimal physical risks associated with this study. Some children, prior to the start of the intervention, may experience some anxiety about what the intervention will be like. Finally, your family relationships may change as a result of the intervention, although we expect that these changes will most often be in a positive direction.

### **Benefits**

There are a number of potential benefits of this study. Your child with autism may improve skills associated with executive functions, namely cognitive flexibility. Your typically developing child may benefit from education about the autism spectrum as well as a variety of strategies for teaching and coping. Your family may also benefit from improved family relationships that result from the child's participation in the intervention. We also hope to learn more about whether our intervention can be helpful to your children and family, which then could be helpful to society at large.

### **Economic Considerations**

There is no cost to you for participation in this study. Any cost associated with travel to the Yale Child Study Center will be reimbursed at the end of the study.

### **Treatment Alternatives/Alternatives**

You may decline to participate in this study, but no alternative treatment is offered by these researchers. An alternative is not to participate in this study. The researchers may provide you with referrals to other intervention studies offered in the community, if any are known to them, or to mental health practitioners who could provide appropriate services for your children.

### **Confidentiality and Privacy**

Any identifiable information that is obtained in connection with this study will remain confidential and will be disclosed only with your permission or as permitted by U.S. or State law. Examples of information that we are legally required to disclose include suspected or known abuse of a child or elderly person, intention to harm oneself or another person, or certain reportable diseases.

All data about you and your child that is collected as part of this study will be labeled with a code number to protect your confidentiality. The principal investigator will keep a link that identifies you to your coded information, but this link will be kept secure and available only to the PI or selected members of the research team. The research team will only give this coded information to others to carry out this research study. Data is stored

in a locked file cabinet and on password protected and secured computers. All data will be destroyed after completion of the study. When the results of the research are published or discussed in conferences, no information will be included that would reveal your identity unless your specific consent for this activity is obtained.

We understand that information about you and your children obtained in connection with your health is personal, and we are committed to protecting the privacy of that information. If you decide to be in this study, the researchers will have some information that identifies you, your children, and your personal health information. This may include information that might directly identify you or your children, such as your names, the names of the members of your family, dates of birth, and diagnostic information. Any information that can identify you or your children will remain confidential. This information until completion of the study, after which time it will be destroyed along with the rest of the study data.

The information about your health that will be collected in this study includes:

- *Research study records / records about your study visits*
- *Clinical records of only those services provided in connection with this study.*
- *Records about phone calls made as part of this research*

Information about you and your health which might identify you may be used by or given to:

- *The U.S. Department of Health and Human Services (DHHS) agencies*
- *Representatives from Yale University and the Human Investigation Committee (the committee that reviews, approves, and monitors research on human subjects), who are responsible for insuring research compliance. These individuals are required to keep all information confidential.*
  - *Those individuals at Yale who are responsible for the financial oversight of research*
  - *The Principal Investigator and Faculty Sponsor*
  - *Co-Investigators of this study*
  - *Study Coordinator and Members of the Research Team*
  - *Other investigators and staff at the Yale Child Study Center*

By signing this form, you authorize the use and/or disclosure of the information described above for this research study. The purpose for the uses and disclosures you are authorizing is to ensure that the information relating to this research is available to all parties who may need it for research purposes.

All health care providers subject to HIPAA (Health Insurance Portability and Accountability Act) are required to protect the privacy of your information. The research staff at the Yale School of Medicine and Yale New Haven Hospital are required to comply with HIPAA and to ensure the confidentiality of your



information. Some of the individuals or agencies listed above may not be subject to HIPAA and therefore may not be required to provide the same type of confidentiality protection. They could use or disclose your information in ways not mentioned in this form. However to better protect your health information, agreements are in place with these individuals and/or companies that require that they keep your information confidential.

You have the right to review and copy your health information in your medical record in accordance with institutional medical records policies.

### **In Case of Injury**

If you or your children are injured as a result of your participation in this study, treatment will be provided at Yale New Haven Hospital or at a treatment facility of your choice. You or your insurance carrier will be expected to pay the costs of this treatment. No additional financial compensation for injury or lost wages is available. By signing this form, you do not waive any legal rights or release the Yale School of Medicine or its agents from liability for negligence.

### **Voluntary Participation and Withdrawal**

Participating in this study is voluntary. You are free to choose not to take part in this study. Refusing to participate will involve no penalty or loss of benefits to which you are otherwise entitled (such as your health care outside the study, the payment for your health care, and your health care benefits). However, you will not be able to enroll in this research study and will not receive study procedures as a study participant if you do not allow use of your information as part of this study.

If you do become a subject, you are free to stop and withdraw from this study at any time during its course. If you sign this authorization, you may change your mind at any time, but the researchers may continue to use information collected before you changed your mind to complete the research. To withdraw, you can call a member of the research team at any time and tell them that you no longer want to take part. This will cancel any appointments in the future. You must also follow up your phone call by sending a written notice to revoke this authorization to the principal investigator.

This authorization to use and disclose your health information will never expire unless and until you change your mind and revoke it.

The researchers may withdraw you from participating in the research if necessary. This may occur if either of your children is not compliant with the treatment or study procedures, or if either of your child's participation in the intervention appears to be harmful to themselves or to the other group members.

Withdrawing from the study will involve no penalty or loss of benefits to which you are otherwise entitled. It will not harm your relationship with your own doctors or with Yale-New Haven hospital. You will still be able to participate in other activities and services offered by the Yale Child Study Center, and where appropriate we will make referrals to a provider who can offer appropriate services for your child.

### **Questions**

We have used some technical terms in this form. Please feel free to ask about anything you don't understand and to consider this research and the consent form carefully – as long as you feel is necessary – before you make a decision.

### **Authorization and Permission**

I have read (or someone has read to me) this form and have decided to participate in the project described above. Its general purposes, the particulars of involvement and possible hazards and inconveniences have been explained to my satisfaction. My signature also indicates that I have received a copy of this consent form.

By signing this form, I give permission to the researchers to use information about me for the purposes described in this form. By refusing to give permission, I understand that I will not be able to be in this research.

Name of Subjects: \_\_\_\_\_

Signature of Parent/Guardian: \_\_\_\_\_

Date: \_\_\_\_\_

Signature of Principal Investigator Date

*or*

Signature of Person Obtaining Consent Date

*If you have further questions about this project or if you have a research-related problem, you may contact the principal investigator at (203) XXX-XXXX . If you have any questions concerning your rights as a research subject, you may contact the Human Investigation Committee at (203) 785-4688. If after you have signed this form you have any questions about your privacy rights, please contact the Yale Privacy Officer at (203) 436-3650.*

*Appendix C: Child Assent Form*

**Child's Assent for Being in a Research Study**

**Yale-New Haven Hospital/Yale University School of Medicine**

For Children Ages 6-12

Title: Sibling Mediation in an Executive Function Intervention for Children with Autism Spectrum Disorder

**Why am I here?**

We are asking you to take part in a research study because we are trying to learn more about children with autism spectrum disorder and their siblings. We are inviting you to be in the study because you have a sibling on the autism spectrum.

**Why are they doing this study?**

We are doing this study to learn about how siblings of kids with autism spectrum disorder can help teach the lessons in our interventions.

**What will happen to me?**

In this study, we will start by having you, your sibling, and your parents fill out some questionnaires. We will also meet with you to ask you some questions about yourself and your sibling. Then you will join our sibling group, where we will learn about autism and how to help our siblings learn the lessons in our intervention. When the group is all over at the end of the school year, you will fill out some more questionnaires and we may ask you some questions again. We will reach out to you again 6 months after finishing the sessions to ask the same questions again.

**Will the study hurt?**

Nothing that you will be doing in this study should hurt. You might be nervous about the sessions before it starts, but we think you will find it to be a lot of fun once you get there. There may be times when talking about your sibling makes you sad. We will never make do anything if you don't want to. You can tell us if something upsets you, and we will stop or make it better for you.

**Will the study help me?**

The study may help you to understand your sibling better, get along better with your sibling, and learn to cope with how difficult it can be to have a sibling on the autism spectrum. This can also help your sibling with autism learn the lessons we are teaching in our study.

**What if I have any questions?**

You can ask any questions that you have about the study. If you have a question later that

you didn't think of now, you can call us at (203) XXX-XXXX or ask the person in charge next time. You may call us at any time to ask questions about the study.

**Do my parents know about this?**

This study was explained to your parents and they said that you and your sibling could be in it. You can talk this over with them before you decide.

**Do I have to be in the study?**

You do not have to be in the study. No one will be upset if you don't want to do this. If you don't want to be in this study, you just have to tell your parents or the researcher. You can say yes now and change your mind later. It's up to you.

Writing your name on this page means that that you agree to be in the study, and know what will happen to you. If you decide to quit the study all you have to do is tell the person in charge.

Signature of Child

Signature of Researcher

Date

Date

*Appendix D: Recruitment Flyer*

**YALE UNIVERSITY SCHOOL OF MEDICINE – YALE NEW HAVEN  
HOSPITAL**

**VOLUNTEERS NEEDED FOR RESEARCH STUDY!**

**Do you have a child diagnosed with autism who has a typically developing sibling?**

We are conducting a research study to investigate whether involving typically developing siblings as mediators to our *Unstuck and On Target!* executive function intervention for children with autism spectrum disorder can help improve cognitive flexibility outcomes as well as the quality of the sibling relationship.

**Who can join the study?**

We are inviting children ages 6 to 12 years diagnosed with autism spectrum disorder who have a typically developing sibling no more than 5 years of age apart.

**What will be asked of study participants?**

Your child with autism will participate in our *Unstuck and On Target!* intervention during the next school year on a bimonthly basis, with or without the involvement your typically developing sibling in concurrent psychoeducation sessions. We will ask your child with autism to perform a card sorting task before and after the intervention. We will also ask both of your children to participate in a survey to determine how they perceive the quality of their sibling relationship before and after the intervention.

**Do you get anything in return?**

Any cost associated with travel to the Yale Child Study Center (230 S. Frontage Road in New Haven) will be reimbursed in full provided your children complete the intervention in its entirety.

**If you and your children are interested in participating or have any questions,  
please do not hesitate to reach out to our clinical research team at:**

**(203) XXX-XXXX or [research@yale.edu](mailto:research@yale.edu)**

*Appendix E: Sibling Relationship Questionnaire*

Sibling Relationship Questionnaire - Revised (Child)

(Furman & Buhrmester, 1985)

|  |   |
|--|---|
| <p>1. Some siblings do nice things for each other a lot, while other siblings do nice things for each other a little. How much do both you and this sibling do nice things for each other?</p> | <p><input type="checkbox"/> Hardly at all<br/> <input type="checkbox"/> Not too much<br/> <input type="checkbox"/> Somewhat<br/> <input type="checkbox"/> Very much<br/> <input type="checkbox"/> EXTREMELY MUCH</p>  |
| <p>2. Who usually gets treated better by your mother, you or this sibling?</p>   | <p><input type="checkbox"/> My sibling almost always gets treated better<br/> <input type="checkbox"/> My sibling often gets treated better<br/> <input type="checkbox"/> We get treated about the same<br/> <input type="checkbox"/> I often get treated better<br/> <input type="checkbox"/> I almost always get treated better</p> |
| <p>3. How much do you show this sibling how to do things he or she doesn't know how to do?</p>   | <p><input type="checkbox"/> Hardly at all<br/> <input type="checkbox"/> Not too much<br/> <input type="checkbox"/> Somewhat<br/> <input type="checkbox"/> Very much<br/> <input type="checkbox"/> EXTREMELY MUCH</p>  |
| <p>4. How much does this sibling show you how to do things you don't know how to do?</p>   | <p><input type="checkbox"/> Hardly at all<br/> <input type="checkbox"/> Not too much<br/> <input type="checkbox"/> Somewhat<br/> <input type="checkbox"/> Very much<br/> <input type="checkbox"/> EXTREMELY MUCH</p>  |
| <p>5. How much do you tell this sibling what to do?</p>  | <p><input type="checkbox"/> Hardly at all<br/> <input type="checkbox"/> Not too much<br/> <input type="checkbox"/> Somewhat<br/> <input type="checkbox"/> Very much<br/> <input type="checkbox"/> EXTREMELY MUCH</p>  |
| <p>6. How much does this sibling tell you what to do?</p>  | <p><input type="checkbox"/> Hardly at all<br/> <input type="checkbox"/> Not too much<br/> <input type="checkbox"/> Somewhat<br/> <input type="checkbox"/> Very much<br/> <input type="checkbox"/> EXTREMELY MUCH</p>  |

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|---|---|
| <p>7. Who usually gets treated better by your father, you or this sibling?</p>  | <p><input type="checkbox"/> My sibling almost always gets treated better<br/> <input type="checkbox"/> My sibling often gets treated better<br/> <input type="checkbox"/> We get treated about the same<br/> <input type="checkbox"/> I often get treated better<br/> <input type="checkbox"/> I almost always get treated better</p> |
| <p>8. Some siblings care about each other a lot while other siblings don't care about each other that much. How much do you and this sibling care about each other?</p>                                 | <p><input type="checkbox"/> Hardly at all<br/> <input type="checkbox"/> Not too much<br/> <input type="checkbox"/> Somewhat<br/> <input type="checkbox"/> Very much<br/> <input type="checkbox"/> EXTREMELY MUCH</p>  |
| <p>9. How much do you and this sibling go places and do things together?</p>  | <p><input type="checkbox"/> Hardly at all<br/> <input type="checkbox"/> Not too much<br/> <input type="checkbox"/> Somewhat<br/> <input type="checkbox"/> Very much<br/> <input type="checkbox"/> EXTREMELY MUCH</p>  |
| <p>10. How much do you and this sibling insult and call each other names?</p>   | <p><input type="checkbox"/> Hardly at all<br/> <input type="checkbox"/> Not too much<br/> <input type="checkbox"/> Somewhat<br/> <input type="checkbox"/> Very much<br/> <input type="checkbox"/> EXTREMELY MUCH</p>  |
| <p>11. How much do you and this sibling like the same things?</p>   | <p><input type="checkbox"/> Hardly at all<br/> <input type="checkbox"/> Not too much<br/> <input type="checkbox"/> Somewhat<br/> <input type="checkbox"/> Very much<br/> <input type="checkbox"/> EXTREMELY MUCH</p>  |
| <p>12. How much do you and this sibling tell each other everything?</p>   | <p><input type="checkbox"/> Hardly at all<br/> <input type="checkbox"/> Not too much<br/> <input type="checkbox"/> Somewhat<br/> <input type="checkbox"/> Very much<br/> <input type="checkbox"/> EXTREMELY MUCH</p>  |
| <p>13. Some siblings try to out-do or beat each other at things a lot, while other siblings try to out-do each other a little. How much do you and this sibling try to out-do each other at things?</p> | <p><input type="checkbox"/> Hardly at all<br/> <input type="checkbox"/> Not too much<br/> <input type="checkbox"/> Somewhat<br/> <input type="checkbox"/> Very much<br/> <input type="checkbox"/> EXTREMELY MUCH</p>  |
| <p>14. How much do you admire and respect this sibling?</p>   | <p><input type="checkbox"/> Hardly at all<br/> <input type="checkbox"/> Not too much<br/> <input type="checkbox"/> Somewhat<br/> <input type="checkbox"/> Very much<br/> <input type="checkbox"/> EXTREMELY MUCH</p>  |
| <p>15. How much does this sibling admire and respect you?</p>   | <p><input type="checkbox"/> Hardly at all<br/> <input type="checkbox"/> Not too much<br/> <input type="checkbox"/> Somewhat</p>   |

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|  | <input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH  |
| 16. How much do you and this sibling disagree and quarrel with each other?   | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH  |
| 17. Some siblings cooperate a lot, while other siblings cooperate a little. How much do you and this sibling cooperate with other? | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH  |
| 18. Who gets more attention from your mother, you or this sibling?   | <input type="checkbox"/> My sibling almost always gets more attention<br><input type="checkbox"/> My sibling often gets more attention<br><input type="checkbox"/> We get about the same amount of attention<br><input type="checkbox"/> I often get more attention<br><input type="checkbox"/> I almost always get more attention |
| 19. How much do you help this sibling with things he or she can't do by him or herself?  | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH  |
| 20. How much does this sibling help you with things you can't do by yourself?  | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH  |
| 21. How much do you make this sibling do things?   | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH  |
| 22. How much does this sibling make you do things?   | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH  |
| 23. Who gets more attention from your father, you or this sibling?   | <input type="checkbox"/> My sibling almost always gets more  |



|  |  |
|--|--|
|  | attention<br><input type="checkbox"/> My sibling often gets more attention<br><input type="checkbox"/> We get about the same amount of attention<br><input type="checkbox"/> I often get more attention<br><input type="checkbox"/> I almost always get more attention |
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| 24. How much do you and this sibling love each other?  | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH |
| 25. Some siblings play around and have fun with each other a lot, while other siblings play around and have fun with each other a little. How much do you and this sibling play around and have fun with each other? | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH |
| 26. How much are you and this sibling mean to each other?  | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH |
| 27. How much do you and this sibling have in common?   | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH |
| 28. How much do you and this sibling share secrets and private feelings?   | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH |
| 29. How much do you and this sibling compete with each other?  | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH |
| 30. How much do you look up to and feel proud of this sibling?   | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH |

|   |   |
|---|---|
| 31. How much does this sibling look up to and feel proud of you?                      | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH |
| 32. How much do you and this sibling get mad at and get in arguments with each other? | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH |

|  |   |
|--|---|
| 33. How much do both you and your sibling share with each other?           | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH   |
| 34. Who does your mother usually favor, you or this sibling?               | <input type="checkbox"/> My sibling almost always is favored<br><input type="checkbox"/> My sibling is often favored<br><input type="checkbox"/> Neither of us is favored<br><input type="checkbox"/> I am often favored<br><input type="checkbox"/> I am almost always favored |
| 35. How much do you teach this sibling things that he or she doesn't know? | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH   |
| 36. How much does this sibling teach you things that you don't know?       | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH   |
| 37. How much do you order this sibling around?                             | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH   |
| 38. How much does this sibling order you around?                           | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH   |
| 39. Who does your father usually favor, you or this sibling?               | <input type="checkbox"/> My sibling almost always is favored<br><input type="checkbox"/> My sibling is often favored<br><input type="checkbox"/> Neither of us is favored   |

|  |   |
|--|---|
|  | <input type="checkbox"/> I am often favored<br><input type="checkbox"/> I am almost always favored  |
| 40. How much is there a strong feeling of affection (love) between you and this sibling?   | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH |
| 41. Some kids spend lots of time with their siblings, while others don't spend so much. How much free time do you and this sibling spend together? | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH |

|  |   |
|--|---|
| 42. How much do you and this sibling bug and pick on each other in mean ways?                    | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH |
| 43. How much are you and this sibling alike?   | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH |
| 44. How much do you and this sibling tell each other things you don't want other people to know? | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH |
| 45. How much do you and this sibling try to do things better than each other?                    | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH |
| 46. How much do you think highly of this sibling?  | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH |
| 47. How much does this sibling think highly of you?  | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat<br><input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH |
| 48. How much do you and this sibling argue with each other?                                      | <input type="checkbox"/> Hardly at all<br><input type="checkbox"/> Not too much<br><input type="checkbox"/> Somewhat  |

|  |   |
|--|---|
|  | <input type="checkbox"/> Very much<br><input type="checkbox"/> EXTREMELY MUCH |
|--|---|

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