

Do caregiving-related stress and emotion regulation impact treatment adherence to a caregiver-mediated intervention for toddlers at risk for ASD?

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Abstract

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Caregivers of toddlers with autism spectrum disorder (ASD) experience increased levels of caregiving-related stress, which can impede gains their toddlers make from participating in interventions. Treatment adherence (i.e., the degree to which caregivers implement interventions in the manner intended) predicts improvements in toddlers' gains from treatment. In addition, caregivers' emotion regulation (ER) strategies predicts treatment adherence, suggesting that this may be a primary way in which caregiving-related stress is managed.

The current study examines the extent to which caregiving-related stress and ER strategies are associated with caregivers' treatment adherence to Project ImPACT while delivering it to their toddlers at familial risk for ASD. It was expected that: 1) high levels of caregiving-related stress would predict low levels of treatment adherence; 2) high levels of adaptive ER strategies would predict high levels of treatment adherence; and 3) ER strategies would moderate the relation between caregiving-related stress and treatment adherence, such that the relation between caregiving-related stress and treatment adherence would be stronger in caregivers with less optimal ER strategies than in caregivers with more optimal ER strategies.

Participants were thirty-eight caregivers who had a child with ASD and a later-born toddler between 11 and 18 months and were assigned to the treatment condition of a randomized controlled trial at the University of Washington and Vanderbilt University. Caregiving-related stress and ER strategies (i.e., reappraisal, suppression, and rumination) were measured via self-report surveys at Baseline (i.e., prior to beginning Project ImPACT training). Treatment adherence was measured via video coding of caregivers' use of modeling and expanding their toddler's language at Baseline and T2 (after completing Project ImPACT training).

Descriptive analyses indicated that caregivers demonstrated low levels of caregiving-related stress and that treatment adherence increased significantly from Baseline to T2. Reappraisal and suppression were negatively correlated with caregiving-related stress, indicating that higher stress was associated with less optimal use of these two ER strategies. Linear regressions indicated that neither caregiving-related stress nor ER strategies predicted treatment adherence. Reappraisal was the only ER strategy that moderated the relation between caregiving-related stress and treatment adherence. The association between caregiving-related stress and treatment adherence was stronger in caregivers with high levels of reappraisal than caregivers with low levels of reappraisal. This pattern of results was in the opposite direction than predicted; however, this result should be interpreted with caution, as the region of significance represented only a small number of participants ($n = 3$). In sum, results suggest that caregiving-related stress and emotion regulation strategies did not affect caregivers' treatment adherence in implementing Project ImPACT in this sample. It is likely that low levels of caregiving-related stress and normative use of ER strategies underlie the null findings in this sample. Future studies should consider the ways in which high levels of caregiving-related stress affect caregivers' treatment adherence and ER strategies in other samples.

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ABBREVIATION KEY

ASD	Autism spectrum disorder
CBT	Cognitive behavioral therapy
CDC	Center for Disease Control
CERQ	Cognitive Emotion Regulation Questionnaire
ER	Emotion regulation
ERQ	Emotion Regulation Questionnaire
ImPACT	Improving Parents as Communication Teachers
IRB	Institutional Review Board
NDBI	Naturalistic developmental behavioral intervention
PCFP	Parent-child free play
PSI-SF	Parenting Stress Index-Short Form
RCT	Randomized controlled trial
RSQ	Response Style Questionnaire
UW	University of Washington
VU	Vanderbilt University

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DEDICATION

This dissertation is dedicated to all caregivers of children with autism spectrum disorder.

Thank you for fighting tirelessly every day so that your children can lead a better life.

Introduction

Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterized by impairments in two core domains: social interaction and communication, and restricted interests and repetitive behaviors (American Psychiatric Association, 2013). The Center for Disease Control and Prevention (CDC) currently estimates that 1 in 59 children in the United States has ASD, which represents a 15% increase from prevalence estimates released in 2012 (Baio, 2018; Christensen et al., 2019). There is no single biological cause for ASD, but research has revealed that ASD is likely attributable to a combination of genetic and environmental risk factors (e.g., Abrahams & Geschwind, 2008; Froehlich-Santino et al., 2014; Gardener, Spiegelman, & Buka, 2009, 2011). A number of interventions have been developed to address the core features of ASD, many of which are effectively delivered to young children 36 months of age and younger (see “ASD-Specialized Interventions for Young Children” section below for further information). The effectiveness of intervention at young ages is thought to be a function of the rapid developmental changes in the brain that occur during the early years, which makes learning and remediation more effective and efficient (Dawson, 2008). One way to study ASD early is to enroll younger siblings of children with ASD in research. These later-born siblings of children with ASD are at an increased risk of developing ASD; recurrence estimates range from 7% -19% (Grønberg, Schendel, & Parner, 2013; Ozonoff et al., 2011). An additional 20% of these later-born siblings who do not receive an ASD diagnosis may demonstrate subclinical ASD symptoms or other types of language or cognitive delays (Messinger et al., 2013). These later-born siblings are the focus of the present study.

Caregivers of children with ASD and caregivers of later-born siblings experience unique challenges associated with having a child with ASD, or ASD risk. These challenges are

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attributable to strains on caregivers' financial resources (Buescher, Cidav, Knapp, & Mandell, 2014; Sharpe & Baker, 2007) and the presence of challenging behaviors (e.g., Baker et al., 2003; Lecavalier, Leone, & Wiltz, 2006; Estes et al., 2013; Neece, Green, & Baker, 2012). The estimated lifetime cost of supporting an individual with ASD in the United States is between \$1.4 and \$2.4 million. These costs stem from special education services, loss of parental income, as well as fees for residential and supportive care into adulthood (Buescher et al., 2014). Many caregivers of children with ASD even experience bankruptcy while ensuring that their children obtain intervention services (Sharpe & Baker, 2007). In addition to the strain on resources, caregivers of children with ASD also report significant challenges due to the unique combination of behavioral deficits and excesses that children with ASD exhibit. Many children with ASD demonstrate limited social interest and understanding, difficulties communicating, understanding the communication of others, challenging behaviors, and emotion regulation deficits (Baker et al., 2003; Baker-Ericzn, Brookman-Fraze, & Stahmer, 2005; Estes et al., 2013; Hastings, 2003; Seltzer et al., 2010; Lecavalier et al., 2006; Neece et al., 2012). Taken together, the strain on caregivers' resources and the presence of behavioral challenges necessitate an examination of how caregivers respond to these stressors, commonly referred to as caregiving-related stress. Caregiving-related stress is crucial to consider in caregivers of later-born toddlers as their caregiving-related stress may already be heightened because of the behavioral demands associated with having at least one child already diagnosed with ASD and a younger toddler who is considered at genetic risk for developing ASD.

Caregiving-Related Stress

Caregiving-related stress, or parenting stress, arises when an individual perceives that the challenges associated with being a parent outweigh one's abilities and resources (e.g., financial

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resources, time, and knowledge; Abidin, 1992; Bornstein, 2002; Deater-Deckard, 1998, 2004; Rao & Beidel, 2009). The extent to which one experiences caregiving-related stress is influenced by caregivers' characteristics, children's characteristics, the degree of conflict within the caregiver-child relationship, and the context in which the caregiver and child live (Abidin, 1992). Caregiving-related stress is fluid, such that levels of caregiving-related stress change over time in response to shifting parenting demands (e.g., caregiving-related stress increases as children transition from early childhood to adolescence; Putnick et al., 2010; Stone, Mares, Otten, Engels, & Janssens, 2016; Williford, Calkins, & Keane, 2007). Caregivers of children with challenging behavior and children with psychopathology are more likely to experience caregiving-related stress than caregivers of children without these same difficulties (Baker et al., 2003; Barroso, Mendez, Graziano, & Bagner, 2017; Briggs-Gowan, Carter, Skuban, & Horwitz, 2001; Deater-Deckard, 1998; Donenberg & Baker, 1993; Neece et al., 2012; Theule, Wiener, Tannock, & Jenkins, 2013). High levels of caregiving-related stress have also been found in caregivers of children with chronic medical conditions (Cousino & Hazen, 2013; Golfenshtein, Srulovici, & Deatrick, 2016; Moreira & Canavarro, 2016; Streisand, Swift, Wickmark, Chen, & Holmes, 2005), caregivers of children with challenging behavior (Barroso et al., 2017; Benzies, Harrison, & Magill-Evans, 2004; Eyberg, Boggs, & Rodriguez, 1993; Neece et al., 2012), and caregivers of children with developmental disabilities and ASD (Baker et al., 2003; Barroso et al., 2017; Emerson, 2003; Hauser-Cram et al., 2001; Hayes & Watson, 2013; Lecavalier et al., 2006).

The construct of caregiving-related stress stems from the general model of stress as defined by Folkman and Lazarus (1985; Hayes & Watson, 2013). As with caregiving-related stress, general stress is a consequence of an imbalance between perceived environmental stressors and an individual's ability to manage and respond to those stressors. This imbalance is a

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direct result of maladaptive or insufficient coping strategies (Folkman & Lazarus, 1985; Hayes & Watson, 2013). High levels of both caregiving-related stress and general stress are associated with negative mental health and physical health symptoms including depression, fatigue, restlessness, and an increased risk for heart disease and ulcers (Carpenter & Steffen, 2004; Folkman & Lazarus, 1985; Hayes & Watson, 2013).

Though high levels of general stress can be harmful, there is evidence that moderate levels of general stress can actually be adaptive (Selye, 1974). This phenomenon is best demonstrated by the inverted U-shaped curve, or the “Yerkes-Dodson law” (Broadbent, 1965; Mendl, 1999; Schilling et al., 2013; Yerkes & Dodson, 1908). This theory posits that, while high and low levels of stress may be maladaptive, there are situations in which moderate levels of stress actually improve performance. This relation is best illustrated during memory tasks in which individuals who are experiencing moderate levels of stress demonstrate improved performance on these tasks compared to individuals who are experiencing high or low levels of stress (Domes, Rothfischer, Reichwald, & Hautzinger, 2005; Young, Drevets, Schulkin, & Erickson, 2011). This relation between moderate levels of stress and performance has not been observed for caregiving-related stress. Instead, even moderate levels of caregiving-related stress are associated with negative physiological (i.e., poorer physical health), behavioral (i.e., increased use of negative parenting practices), and mental health consequences (i.e., increased rates of depression and anxiety; Abidin, 1992; Bornstein, 2002; Coldwell, Pike, & Dunn, 2006; Crnic, Gaze, & Hoffman, 2005; Deater-Deckard, 1998; Eisenhower, Baker, & Blacher, 2009; Gutermuth et al., 2005; Hastings, Daley, Burns, & Beck, 2006; Oelofsen & Richardson, 2006; Rao & Beidel, 2009).

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The measurement of general stress and caregiving-related stress also differs. General stress is often examined via both physiological (e.g., heart rate variability, blood pressure, and cortisol) and self-report measures (Kirschbaum & Hellhammer, 1994; Kudielka, Buske-Kirschbaum, Hellhammer, & Kirschbaum, 2004; Luecken, Appelhans, Kraft, & Brown, 2006; Smith & Allred, 1989). Though recent studies have begun to examine these same physiological markers of caregiving-related stress (e.g., [Factor, Swain, & Scarpa, 2017](#)), the vast majority of research has relied solely on self-reported caregiving-related stress (Hayes & Watson, 2013).

Caregiving-related stress in ASD.

Caregivers of children with ASD experience higher caregiving-related stress than caregivers of children who are typically developing, caregivers of children who have developmental disabilities, and caregivers of children who have ASD. This pattern is well-documented and may be attributable to the unique challenges associated with ASD described previously, including challenging behavior and strain on resources (e.g., Baker-Ericzn et al., 2005; Barroso et al., 2017; Dunn, Burbine, Bowers, & Tantleff-Dunn, 2001; Estes et al., 2013; Hayes & Watson, 2013; Hoffman, Sweeney, Hodge, Lopez-Wagner, & Looney, 2009; Karst & Van Hecke, 2012; Kasari & Sigman, 1997; Lecavalier et al., 2006; Schieve, Blumberg, Rice, Visser, & Boyle, 2007).

In addition to the negative impact of caregiving-related stress on caregivers' physical health, mental health, and parenting practices, there also is evidence that caregiving-related stress affects their child's access to and benefits from ASD-specialized interventions. First, there is preliminary evidence that children whose caregivers experience high levels of caregiving-related stress receive fewer intervention services than children whose caregivers experience low levels of caregiving-related stress (Karp et al., 2018). In addition, there is emerging evidence that

caregiving-related stress is related to child gains from the intervention. Children of caregivers with high levels of caregiving-related stress have been found to make fewer gains from treatment when caregivers are directly involved in the delivery of the intervention (Stadnick, Stahmer, & Brookman-Frazee, 2015; Weiss, Vecili, & Bohr, 2014). In fact, even when the caregivers are not directly involved in all aspects of the intervention, caregivers with high levels of caregiving-related stress have children who make fewer gains in intellectual and social functioning than children of caregivers with low levels of caregiving-related stress (Osborne, McHugh, Saunders, & Reed, 2008a; Robbins, Dunlap, & Plienis, 1991). Taken together, these results indicate that the effects of caregiving-related stress must be considered when enrolling children in interventions, as this stress may inhibit anticipated improvements from treatment.

The current study focuses on the extent to which caregiving-related stress affects caregivers' implementation of an intervention with their later-born toddler. Later-born toddlers are the focus of the present study as they can be identified early and may benefit from the receipt of ASD-specialized early intervention (Dawson, 2008; Green et al., 2013). It is important to note that, by virtue of having an older child with ASD, caregivers of later-born toddlers may already be experiencing high levels of caregiving-related stress related to their older child and any challenging behavior that child may be experiencing. This caregiving-related stress may be compounded by additional stressors associated with monitoring and evaluating the development of their later-born toddler. To date, no intervention studies that have enrolled later-born toddlers have examined caregiving-related stress related to the younger toddler when the caregiver is directly involved in the implementation of the intervention.

ASD-Specialized Interventions for Young Children

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Although there is no known cure for ASD, ASD-specialized early interventions may ameliorate core symptoms. Different research groups have developed ASD-specialized early interventions for children under 36 months of age (e.g., Dawson et al., 2010; Ingersoll 2012; Landa et al., 2011). Though many of these interventions share certain characteristics, they have long been examined by independent research groups. Recently, however, in an effort to unify the field of ASD-specialized intervention research, a seminal manuscript identified the principles shared by each of these programs and coined a new term to describe this class of interventions: Naturalistic Developmental Behavioral Interventions (NDBIs; Schreibman et al., 2015). All NDBIs focus on teaching in natural settings, and are based on a blend of developmental and behavioral principles (Schreibman et al., 2015).

All NDBIs share the following characteristics: 1) utilizing three-part contingencies (i.e., using the antecedent-response-consequence pattern to teach skills); 2) implementing manualized practices; 3) expanding the child's focus to new play routines or activities; 4) creating individualized goals for treatment; 5) tracking progress throughout the course of treatment; 6) focusing on teaching children within their attentional field; 7) arranging the child's immediate environment to promote social-communication; 8) focusing on increasing children's motivation using natural reinforcement; 9) utilizing prompting hierarchies; 10) taking turns with a play partner; 11) modeling new behaviors; 12) imitating child behavior, vocalization, and gestures; and 13) examining what is referred to as "fidelity" (Schreibman et al., 2015).

With regard to the final characteristic, fidelity, it is crucial to note that within the field of ASD research, the term "fidelity" is frequently used to reflect the extent to which the interventionist is implementing skills in the manner designed and intended by the treatment developers (i.e., treatment adherence; Perepletchikova & Kazdin, 2005; Perepletchikova, Treat,

& Kazdin, 2007; Waltz, Addis, Koerner, & Jacobson, 1993). Within the treatment development literature, however, “fidelity” reflects multiple aspects of the intervention delivery, only one of which is treatment adherence (Dane & Schneider, 1998; Durlak & DuPre, 2008; Perepletchikova & Kazdin, 2005; Southam-Gerow & McLeod, 2013; Waltz et al., 1993). The different aspects of fidelity are described in further detail below. To maintain consistency and bridge terminology across fields of research, the term “treatment adherence” will be used to reflect interventionists’ implementation of the intervention in the way in which it was designed and intended by treatment developers (Perepletchikova & Kazdin, 2005; Perepletchikova et al., 2007; Waltz et al., 1993).

Though all NDBIs share common principles, each intervention differs in the skills that are addressed and who is responsible for delivering the intervention. NDBIs are either targeted or comprehensive. Targeted interventions are those that teach the development of specific skills such as joint attention, play, or imitation (Ingersoll, 2012; Kasari, Gulsrud, Wong, Kwon, & Locke, 2010; Kasari, Paparella, Freeman, & Jahromi, 2008). Comprehensive interventions address a range of skills that include adaptive behavior, language, and social engagement (Dawson et al., 2010; Marcus, Schopler, & Lord, 2000; Rogers et al., 2012). Both targeted and comprehensive NDBIs have been shown to be effective when compared to treatment-as-usual or waitlist control conditions (e.g., Dawson et al., 2010; Kasari et al., 2010). Though there is evidence that both targeted and comprehensive NDBIs can be effectively delivered by trained professionals and by caregivers of children with ASD (Ben-Itzhak & Zachor, 2007; Dawson et al., 2010; Ingersoll, 2012; Kasari, Freeman, & Paparella, 2006; Kasari et al., 2008; Landa et al., 2011; Remington et al., 2007), the field of NDBI research has shifted towards including

caregivers in the delivery of both targeted and comprehensive NDBIs (Bearss, Burrell, Stewart, & Scahill, 2015).

Involving caregivers in ASD-specialized interventions.

There are several known benefits associated with including caregivers in interventions. First, caregivers' involvement promotes generalization of skills taught in intervention programs to the home environment (Kasari et al., 2010). Additionally, because toddlers spend a great deal of time at home, it is considered best practice for caregivers to be involved in their toddler's intervention. Involving caregivers allows for these skills to be taught in a cost-effective manner in a naturalistic setting (e.g. Green et al., 2010; Kasari et al., 2010; National Research Council, 2001; Wong et al., 2015). The vast majority of NDBIs include caregivers in the delivery of the intervention in some capacity (Schreibman et al., 2015).

Caregivers can be included in intervention delivery in different ways. ASD research has long used inconsistent terminology across studies to describe caregivers' involvement in interventions. In an effort to create a comprehensive and consistent taxonomy to describe caregiver involvement in interventions, Bearss and colleagues (2015) defined different types of caregiver¹ involvement in interventions. The goal of caregiver support interventions are to provide caregivers with knowledge about and support with ASD and its related symptoms. Children are the indirect beneficiaries of caregiver support interventions. There are two subtypes of caregiver support interventions: care coordination and psychoeducation. By contrast, the goal of caregiver implementation interventions is to provide caregivers with hands-on training in skills and techniques so that they can act as the agents of change for their child's behavior.

¹ Bearss and colleagues (2015) used the term "parent" in their manuscript. To be inclusive of all participants, including grandparents and other primary caregivers, this review will use the term "caregiver" when describing the adult who participates in the intervention program.

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Children are the direct beneficiaries of the intervention from caregiver implementation interventions. There are two subtypes of caregiver implementation interventions: caregiver training interventions for the management of maladaptive behaviors associated with ASD and caregiver-mediated interventions for core symptoms of ASD. Caregiver training for managing maladaptive behaviors are those that target unique challenges often seen in children with ASD, including challenging behavior, difficulties with sleep, toileting, and feeding. Caregiver-mediated interventions for core symptoms are those that focus on improving children's skills in areas including social communication, imitation, and play as each of these areas is challenging for many children with ASD (Bearss et al., 2015). Many intervention programs designed for children under 36 months old are caregiver-mediated, which is the focus of the proposed study (e.g., Carter et al., 2011; Kasari et al., 2010; Koegel, Koegel, Bruinsma, Brookman, & Fredeen, 2003; Koegel et al., 1989).

Several caregiver-mediated NDBIs have been implemented with young children with ASD. Each of these interventions requires trained professionals to teach caregivers to implement NDBI strategies with their young children. Different research groups have developed different caregiver-mediated NDBIs. The NDBIs with the strongest evidence base include Project ImPACT (Improving Parents as Communication Teachers; Ingersoll & Dvortcsak, 2010), ESDM (Early Start Denver Model; Rogers, Herbison, Lewis, Pantone, & Reis, 1986), JASPER (Joint Attention, Symbolic Play, Engagement, and Regulation; Kasari et al., 2006, 2008), and PRT (Pivotal Response Treatment; Koegel, Koegel, Harrower, & Carter, 1999).

While each of these programs share overarching NDBI principles, they differ in terms of the setting of intervention delivery, the duration of the intervention, and the follow-up period after intervention completion. With regard to setting, some NDBIs are delivered in the home

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(e.g., Dawson et al., 2010; Kasari et al., 2014), while others are delivered in labs (e.g., Ingersoll & Wainer, 2013a; Kaiser, Hancock, & Nietfeld, 2000; Kasari et al., 2010; Kasari, Gulsrud, Paparella, Hellemann, & Berry, 2015), or community clinics (e.g., Ingersoll & Gergans, 2007; Stadnick et al., 2015). There also is great variability in the duration of the intervention (Oono, Honey, & McConachie, 2013). Most intervention programs range from eight to 12 weeks (e.g., Kasari et al., 2010, 2014), though one study has examined the efficacy of a two-year NDBI (Dawson et al., 2010). After intervention completion, many studies include follow-up visits which range from two months to two years (e.g., Estes et al., 2015; Kasari et al., 2010).

Efficacy of caregiver-mediated interventions.

Individual studies of different caregiver-mediated NDBIs have demonstrated gains for both children and caregivers after they participate in these interventions. For example, children show gains in joint attention, engagement, play skills (Kasari et al., 2010, 2015), language and communication (Hardan et al., 2015; Minjarez, Williams, Mercier, & Hardan, 2011), and reductions in autism symptoms (Rogers et al., 2014). In addition, caregivers show gains in empowerment (Minjarez, Mercier, Williams, & Hardan, 2012), interaction skills with their child (Harrop, Gulsrud, Shih, Hovsepyan, & Kasari, 2016; Rogers et al., 2012), and reductions in caregiving-related stress (Estes et al., 2014; Minjarez et al., 2012).

Although individual studies have demonstrated improvements in child and caregiver outcomes after participating in caregiver-mediated interventions, when examined via meta-analyses and systematic reviews, there is less empirical support for the efficacy of these interventions. Two meta-analyses and one systematic review of caregiver-mediated interventions for young children (i.e., children seven years old and younger) have been conducted. Findings highlight that, as with many interventions for young children, no one intervention program is

effective for all families (Beaudoin, Sébire, & Couture, 2014; Nevill, Lecavalier, & Stratis, 2016; Oono et al., 2013). While some studies have found improvements in children's communication, socioemotional skills, autism symptom severity, and socialization when they participated in a caregiver-mediated NDBI, other studies did not find these same improvements. These conflicting results are primarily due to small sample sizes, coders who are not blind to treatment condition, risk of bias from participant attrition, and inconsistent outcome measures across studies (Oono et al., 2013).

The fact that no single intervention is effective for all children or caregivers raises a critical question: What are the characteristics of children and caregivers who *do* benefit from specific NDBIs? To address this question, many studies, including both meta-analyses and the systematic review, have called for the examination of predictors and moderators of treatment efficacy. Predictors are pretreatment characteristics that are associated with gains from treatment (e.g., Vivanti, Prior, Williams, & Dissanayake, 2014). Moderators are pretreatment characteristics that interact with a specific treatment to identify those who are likely to respond to the treatment (Beaudoin et al., 2014; Kraemer, Wilson, Fairburn, & Agras, 2002; Nevill et al., 2016; Oono et al., 2013; Schreibman, 2000; Vivanti et al., 2014). Examining predictors and moderators of treatment efficacy allows for interventions to be individualized to meet each family's needs (Howlin & Charman, 2011; McConachie, Fletcher-Watson, & Working Group 4, COST Action 'Enhancing the Scientific Study of Early Autism,' 2014; Nevill et al., 2016; Oono et al., 2013; Oosterling et al., 2010).

Moderators and predictors of the efficacy of caregiver-mediated interventions in ASD.

To date, only four studies have examined moderators of treatment efficacy of caregiver-mediated NDBIs (Carter et al., 2011; Lecavalier et al., 2016; Scahill et al., 2016; Siller, Hutman,

& Sigman, 2013). For children under the age of eight with ASD and challenging behavior, the presence of additional comorbidities (i.e., ADHD, ODD, anxiety, and hyperactivity) leads to fewer treatment gains on caregiver-reported irritability and noncompliance in children randomly assigned to a parent training condition compared to children randomly assigned to a parent education condition (Lecavalier et al., 2016). Within the same sample, however, IQ does not moderate treatment efficacy (Scahill et al., 2016). For children six-years-old and younger with ASD, Siller and colleagues (2013) found that child baseline expressive language ability and caregiver insightfulness moderate treatment efficacy. Children with low levels of expressive language show greater improvement when randomly assigned to the caregiver-mediated intervention compared to children randomly assigned to the no-treatment control condition. Further, insightful mothers (i.e., those with awareness and expression of emotion about children's motivations) demonstrate improved interactions with their child when assigned to the caregiver-mediated intervention compared to insightful mothers assigned to the control condition.

Within NDBI research for young children under the age of three, to our knowledge, only one study has examined moderators of treatment efficacy of a caregiver-mediated intervention. Carter and colleagues (2011) found that toddlers' pretreatment level of object interest moderates outcome, such that toddlers with low levels of object interest show greater gains in social-communication skills (i.e., initiating joint attention, initiating behavioral requests, nonverbal communication, and intentional communication) when randomized to the intervention condition compared to those randomized to the routine care condition.

Predictors of treatment efficacy have been more thoroughly examined than moderators of treatment efficacy of caregiver-mediated NDBIs. These predictors include child factors,

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caregiver factors, and factors associated with caregivers' delivery of the intervention. With regard to child factors, there is evidence that high nonverbal problem-solving, cognitive abilities, and adaptive behavior are associated with gains seen from treatment (Hardan et al., 2015; Hayward, Eikeseth, Gale, & Morgan, 2009; Remington et al., 2007; Smith et al., 2015). With regard to caregiver factors, as previously described, there is evidence that high caregiving-related stress is negatively associated with children's gains from treatment (Osborne, McHugh, Saunders, & Reed, 2008b; Robbins et al., 1991; Stadnick et al., 2015). With regard to factors associated with caregivers' delivery of the intervention, there is evidence that caregiver- and interventionist-rated caregiver buy-in and involvement, and caregiver use of intervention strategies, each predict treatment outcomes (Gulsrud, Helleman, Shire, & Kasari, 2015; Stadnick et al., 2015; Weiss et al., 2014).

It is crucial to consider the extent to which factors associated with intervention delivery predict treatment efficacy. Within the field of treatment development and efficacy, these factors are collectively referred to as "treatment integrity" or "treatment fidelity" (Margison et al., 2000; McLeod, Southam-Gerow, & Weisz, 2009; Perepletchikova & Kazdin, 2005; Perepletchikova, et al., 2007; Waltz et al., 1993). There are four aspects of treatment integrity: relational elements, treatment differentiation, treatment adherence, and therapist competence. Relational elements refer to the extent to which clients are involved in treatment and the quality of their bond with the therapist. Treatment differentiation represents the extent to which one intervention differs from other interventions, whereas treatment adherence is the extent to which the intervention is delivered in the manner designed and intended by intervention developers. Finally, therapist competence is defined as the extent to which the therapist delivers the intervention skillfully and responsively (Dane & Schneider, 1998; Margison et al., 2000; Perepletchikova & Kazdin, 2005;

Perepletchikova et al., 2007; Southam-Gerow & McLeod, 2013; Waltz et al., 1993). Research has examined the ways in which each aspect of treatment integrity relates to treatment efficacy for interventions delivered by professionals (e.g., teachers and therapists) and caregivers (Carroll et al., 2007; Dusenbury, Brannigan, Falco, & Hansen, 2003; Forgatch, Patterson, & DeGarmo, 2005; Wainer & Ingersoll, 2013). The proposed study focuses on one aspect of treatment integrity: treatment adherence.

Treatment adherence is the focus of the current study for three reasons. First, it is the aspect of treatment integrity that has been most often examined in treatments for children with ASD (Wainer & Ingersoll, 2013). Second, it allows for an understanding of the extent to which changes in client behavior are due to the treatment itself or other factors and plays a crucial role in the replication of results from intervention studies across samples and settings (Carroll et al., 2007; Dane & Schneider, 1998; Dusenbury et al., 2005; Gitlin & Paris, 2016; Kazdin, 1979; Mandell et al., 2013; McArthur, Riosa, & Preyde, 2012; McConachie et al., 2014; Schoenwald, 2011). Third, there is evidence that treatment adherence is directly related to treatment efficacy for behavioral interventions, as described below.

The Relation between Treatment Adherence and Treatment Efficacy

The relation between treatment adherence and the efficacy of these interventions is well-documented for individuals with internalizing and externalizing disorders (e.g., Frank, Kupfer, Wagner, McEachran, & Cornes, 1991; Gillespie, Huey, & Cunningham, 2017; Hogue et al., 2008; Löfholm, Eichas, & Sundell, 2014; Robbins et al., 2011; Spanier, Frank, McEachran, Grochocinski, & Kupfer, 1996; Springer & Reddy, 2010). This relation has been examined for therapist-delivered interventions (e.g., Chapman & Schoenwald, 2011; Gillespie et al., 2017; Henggeler, Melton, Brondino, Scherer, & Hanley, 1997; Hogue et al., 2008; Huey, Henggeler,

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Brondino, & Pickrel, 2000; Löfholm et al., 2014; Robbins et al., 2011; Schoenwald, Carter, Chapman, & Sheidow, 2008), and, to a lesser extent, for caregiver-mediated interventions (e.g., Nock & Ferriter, 2005; Patterson & Chamberlain, 1994; Stoolmiller, Duncan, Bank, & Patterson, 1993).

Within caregiver-mediated interventions for children with externalizing disorders, high caregiver treatment adherence is associated with positive treatment outcomes including improvements in parenting practices and reductions in future child behavior problems such as arrests, out-of-home-placements, and antisocial behavior (Eames et al., 2009; Patterson & Chamberlain, 1994; Reardon, Cukrowicz, Reeves, & Joiner, 2002; Stoolmiller et al., 1993). There also is evidence that treatment adherence to caregiver-mediated interventions for children with ASD is associated with improvements in child outcomes. Two studies have found that high levels of treatment adherence are associated with improvements in child language and communication (Ingersoll & Wainer, 2013a; Vismara, Colombi, & Rogers, 2009). The relation between treatment adherence and treatment efficacy supports the continued examination of treatment adherence within caregiver-mediated NDBIs.

Caregiver-Mediated NDBIs and Treatment Adherence

Intervention developers examine treatment adherence to the key components of the treatment. Identifying these key components can result in the delivery of individualized, cost-effective interventions (Pellecchia et al., 2015). These components can be identified through a priori selection of hypothesized mechanisms of change (Kasari et al., 2010; Rogers et al., 2014; Vismara et al., 2009), or through the use of systematic dismantling, which requires statistical examination of the active ingredients of a treatment (Chaffin, Funderburk, Bard, Valle, & Gurwitch, 2011; Damschroder et al., 2009; Micco, Choate-Summers, Ehrenreich, Pincus, &

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Mattis, 2007; Pellecchia et al., 2015; Van Brunt, 2000). For caregiver-mediated NDBIs, only the former method has been used, whereby developers select theorized aspects of the intervention that are most likely to be associated with changes in child and caregiver behavior (e.g., caregiver modeling and expanding their toddler's language in Project ImPACT; Ingersoll & Dvortcsak, 2006) as a measure of treatment adherence.

Within NDBI research, treatment adherence can be measured via caregiver-report or video-recorded sessions of caregivers and their children (Ingersoll & Wainer, 2013a, 2013b; Ingersoll, Wainer, Berger, Pickard, & Bonter, 2016; Kasari et al., 2010; Rogers et al., 2014; Rogers et al., 2012; Symon, 2005; Vismara et al., 2009). Kasari and colleagues (2010) measured caregiver-reported treatment adherence via a four-item Likert-style questionnaire that required caregivers to report on the extent to which they adhered to treatment protocol over the course of the week. Though this questionnaire demonstrated good internal consistency (Cronbach's $\alpha = .82$), it is possible that caregivers were susceptible to social desirability bias, which may have influenced their responses. When treatment adherence is coded by individuals who are blind to treatment condition, this risk of bias is removed (Hogue, Liddle, & Rowe, 1996). These video-recorded sessions involve direct observation of the caregiver implementing the intervention with their child.

Treatment adherence can be coded as it relates to discrete or global behaviors (Ingersoll & Wainer, 2013a, 2013b; Ingersoll et al., 2016; Rogers et al., 2012; 2014; Vismara et al., 2009). Some checklists require coders to rate caregiver behavior in discrete areas related to the intervention. These behaviors often include rating caregivers' prompting strategies, as these are considered to be essential aspects of many interventions (Coolican, Smith, & Bryson, 2010; Ingersoll & Wainer, 2013a, 2013b; Minjarez et al., 2011; Symon, 2005). By contrast, Rogers and

colleagues (2012; 2014) and Vismara and colleagues (2009) rated caregivers based on global behaviors that are qualitative in nature and represent the style caregivers were taught through the intervention. For example, caregivers may be rated on their general management of unwanted behaviors using positive approaches (Vismara et al., 2009). Coding discrete behaviors offers unique advantages compared to coding global behaviors. First, coding these discrete behaviors allows for flexibility in later analyses, as behaviors may be examined in a temporal manner, or may be combined in different ways. Second, coding discrete behavior may allow for more reliable coding than coding global behaviors, as it removes some elements of human judgement inherent in global coding (Chorney, McMurtry, Chambers, & Bakeman, 2015; Margolin et al., 1998). For these reasons, the current study examines treatment adherence via a discrete behavior.

In the current study, treatment adherence is examined as it relates to a discrete behavior that is a hypothesized key ingredient of a specific NDBI: Project ImPACT (Ingersoll & Dvortcsak, 2006). Specifically, the extent to which a caregiver models and expands their toddler's language was coded and used to define treatment adherence. This discrete behavior was selected because improving toddlers' language skills is one of the primary goals of Project ImPACT, and caregivers are taught to do so by building upon what a toddler already does and by providing examples of the language their toddler could use. The concept of modeling and expanding a toddler's language has been identified as one of the common features of all NDBIs (though is sometimes referred to as simply "modeling"; Schreibman et al., 2015). As such, caregivers who participate in the Project ImPACT intervention are taught "modeling and expanding their toddler's language" as a single skill, which is measured as a discrete behavior (Ingersoll & Dvortcsak, 2006; Schreibman et al., 2015). Previous research illustrates that caregivers' use of modeling and expanding their toddler's language increases after participating

in Project ImPACT (Ingersoll & Wainer, 2013). For these reasons, modeling and expanding a toddler's language will be considered together as a discrete variable in the current study.

The critical role that treatment adherence plays in ensuring treatment efficacy suggests that it is necessary to complete a nuanced evaluation of the factors that promote increased treatment adherence (Nock & Ferriter, 2005). Doing so will allow intervention developers to identify caregiver factors that are associated with their high or low levels of treatment adherence. Once these factors are identified, treatments can be individualized to address barriers to treatment adherence in the service of increasing the quality of caregivers' implementation of interventions (Nock & Ferriter, 2005; Vivanti, 2017; Vivanti et al., 2014).

Predictors of treatment adherence to caregiver-mediated NDBIs.

Few studies have examined factors that predict treatment adherence to caregiver-mediated NDBIs. Both Carr and colleagues (2015) and Moore and Symons (2011) examined caregiver factors as predictors of caregiver-reported treatment adherence. Moore and Symons asked caregivers to rate their adherence to six categories of skills they were taught to use in the intervention. The authors found that self-reported treatment adherence was positively associated with three caregiver beliefs: 1) the intervention was effective; 2) they were able to act as an effective agent of change; and 3) their child was accepted in their community. Moore and Symons also found that higher levels of perceived intervention efficacy predicted higher levels of treatment adherence compared to lower levels of perceived intervention efficacy. Carr and colleagues examined caregiver-reported treatment adherence by asking questions related to time and effort extended to use intervention strategies. Carr found that high levels of caregiver-reported treatment adherence were predicted by high levels of caregiving-related stress, and low child nonverbal IQ. The direction of this result was surprising, especially in light of research that

indicates that caregiving-related stress is negatively associated with gains children make from treatment (Stadnick et al., 2015; Weiss et al., 2014). This result may be attributable to the fact that treatment adherence in Carr's study was measured solely via caregiver report of time and effort spent using intervention strategies, rather than a measure of the extent to which intervention strategies are implemented in the manner intended by treatment developers. As such, caregivers who feel high levels of caregiving-related stress may also feel pressure to put forth greater time and effort to help their toddler (Carr & Lord, 2013; Carr et al., 2015). Carr's study does not provide information about whether caregivers were implementing intervention strategies correctly.

To our knowledge, Carr and colleagues (2015) and Moore and Symons (2011) are the only studies to have examined predictors of treatment adherence to caregiver-mediated NDBIs. It is difficult to draw conclusions from these two studies because they each used different criteria to define treatment adherence. While Moore and Symons asked caregivers to rate their adherence to specific behaviors, Carr asked caregivers to report on the time and effort spent on using intervention strategies. Future studies should define treatment adherence in the same way so that comparisons across studies can be made. Despite these limitations, however, both Carr's and Moore and Symons' work lend support to considering caregiver factors as predictors of treatment adherence to caregiver-mediated NDBIs. The current study builds upon the work of Carr and Moore and Symons by examining additional caregiver factors that may relate to blindly-coded treatment adherence.

The relation between caregiving-related stress and treatment adherence in ASD.

Though there are clear benefits to including caregivers in their child's intervention, the shift towards caregiver-mediated NDBIs must be considered in the context of what is known

about caregiving-related stress in this population. In fact, as previously described, one of the few studies that examined the deleterious effects of caregiving-related stress on child gains was examined within the context of Project ImPACT (Stadnick et al., 2015). The evidence that caregiving-related stress can reduce children's gains not only from therapist-delivered interventions but also from caregiver-delivered interventions, highlights the importance of assessing caregiving-related stress prior to the beginning of intervention. Doing so has the potential to improve outcomes for both caregivers and children, in that adaptations to the intervention can be made and/or provision of adjunctive stress management programs may be offered (McConnell, Parakkal, Savage, & Rempel, 2015). Thus, within caregiver-mediated NDBIs, it is critical to understand the relation between caregiving-related stress and treatment adherence and, importantly, whether the deleterious effects of caregiving-related stress can be attenuated. The current study examines one factor that may ameliorate caregiving-related stress: caregivers' emotion regulation strategies.

Emotion Regulation

Emotion regulation (ER) is one way in which individuals can attempt to manage stress, including caregiving-related stress. ER has been defined as, "the extrinsic and intrinsic processes responsible for monitoring, evaluating, and modifying emotional reactions" (Thompson, 1994, p. 28). The theory behind how emotions are regulated has sparked debate, with different research groups defining ER, or aspects of ER, differently. ER (Gross, 1998), self-regulation (Rothbart & Rueda, 2005), effortful control (Rothbart & Rueda, 2005), and inhibitory control (Diamond, 2013) are some of the most common constructs that different research groups have used to attempt to quantify and measure the regulation of one's emotions. Gross' model will be used in the current study because it conceptualizes ER in a stage-like, temporal manner, with direct

implications for interventions that are either antecedent or response focused (Gross, 1998, 2002, 2015).

According to Gross' (1998) process model, emotion regulation occurs across five distinct stages. The first stage, *situation selection*, refers to the regulation that occurs when an individual approaches or avoids certain settings as a means of regulating his emotions. The second stage, *situation modification*, refers to one's attempts to change his environment to either enhance or reduce an emotional response. The third stage, *attentional deployment*, refers to one's attempts to focus attention and select certain aspects of a situation, while avoiding others. After these aspects have been selected during attentional deployment, the fourth stage occurs: *cognitive change*. Cognitive change describes the process by which an individual associates meaning with the situation. The fifth and final stage of emotion regulation, *response modulation*, occurs after one has an emotional response and attempts to modify his response to this emotion. This modification can include either enhancement or suppression of this response. The first four stages are antecedent-focused strategies because they occur prior to an emotional response, whereas response modulation is a response-focused strategy (Gross, 1998, 2002).

ER can be measured via physiological as well as self-report measures. One physiological measure of ER is respiratory sinus arrhythmia (RSA; Porges, 2001, 2003). RSA is a measure of the interbeat interval of the heart and is influenced by the parasympathetic nervous system (Gentzler, Santucci, Kovacs, & Fox, 2009; Porges, 2001). RSA has been examined in both adults and children and can provide critical information related to ER (Porges, 2001, 2007). In addition to collecting RSA, ER strategies can also be measured via self-report measures in adults. These measures typically collect information on discrete ER strategies. For example, the Emotion Regulation Questionnaire (ERQ; Gross & John, 2003) provides information on two ER

strategies: reappraisal and suppression. Reappraisal, an antecedent-focused strategy, is an example of cognitive change. Reappraisal is defined as the process by which neutral or positive interpretations are produced as a means of decreasing distress when one is confronted with a stressful situation (Gross, 1998, 2002). By contrast, suppression, a response-focused strategy, is an example of response modulation. Suppression is defined as the inhibition of an emotion, a thought, or a behavior congruent with an emotion (Aldao, Nolen-Hoeksema, & Schweizer, 2010; Gross & Thompson, 2007; Gross, 2002; John & Gross, 2004; Thompson, 1994).

ER and psychopathology.

Maladaptive ER is a key component of many mental health diagnoses and may represent a transdiagnostic feature of psychopathology (Aldao & Nolen-Hoeksema, 2010; Aldao, Nolen-Hoeksema, & Schweizer, 2010; Berenbaum, Raghavan, Le, Vernon, & Gomez, 2003; Campbell-Sills & Barlow, 2007; Ehring & Watkins, 2008; Greenberg, 2002; Gross & Munoz, 1995; Gross & John, 2003; Harvey, Watkins, Mansell, & Shafran, 2004; Kring & Sloan, 2010; Kring & Bachorowski, 1999; McLaughlin & Nolen-Hoeksema, 2011; Mennin & Farach, 2007; Mennin, Holaway, Fresco, Moore, & Heimberg, 2007; Moses & Barlow, 2006; Watkins, 2008). Disorders including major depressive disorder (Nolen-Hoeksema, Wisco, & Lyubomirsky, 2008; Rottenberg, Gross, & Gotlib, 2005), borderline personality disorder (Linehan, 1993; Lynch, Trost, Salsman, & Linehan, 2007), social anxiety disorder (Kashdan & Breen, 2007), generalized anxiety disorder (Mennin et al., 2007), and substance use disorders (Fox, Axelrod, Paliwal, Sleeper, & Sinha, 2007; Linehan et al., 2002; Sher & Grekin, 2007; Tice, Bratslavsky, & Baumeister, 2001) are all characterized by maladaptive ER.

The fact that ER represents a core feature of such a wide range of psychopathologies is why it is often a targeted skill during mental health interventions. Interventions that target ER

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strategies include cognitive-behavioral therapy (CBT; Beck, 1976; Linehan, 1993), acceptance and commitment therapy, and mindfulness-based therapies (Hayes, Strosahl, & Wilson, 1999; Roemer, Orsillo, & Salters-Pedneault, 2008; Segal, Williams, & Teasdale, 2002). Certain ER strategies (e.g., rumination, and reappraisal) have been shown to be associated with anxiety and stress such that high levels of rumination and low levels of reappraisal predict high levels of stress (Garnefski, Kraaij, & Spinhoven, 2001; Martin & Dahlen, 2005).

The relation between ER strategies and caregiving-related stress.

The relation between caregiving-related stress and ER strategies has been most widely examined in caregivers of individuals with traumatic brain injuries, dementia, and cancer (Chronister & Chan, 2006; Haley, Levine, Brown, & Bartolucci, 1987; Pearlin, Mullan, Semple, & Skaff, 1990; Weitzner, Haley, & Chen, 2000). Results from these studies reveal that certain ER strategies, including reappraisal, may serve as protective factors against the deleterious effects of caregiving-related stress including depression, physical illness, and poor life satisfaction (Cooper, Katona, Orrell, & Livingston, 2008; Haley, LaMonde, Han, Burton, & Schonwetter, 2003; Haley et al., 1987; Morano, 2003; Weitzner et al., 2000).

The aim of the current study is to expand upon previous research that has examined the relation between caregiving-related stress and ER strategies. Research from other populations suggests that, even when high, caregiving-related stress may be ameliorated by adaptive ER strategies (e.g., Haley et al., 2003). This relation has not yet been examined in caregivers of children with ASD or caregivers of later-born toddlers, yet is of critical import as these caregivers represent another population with high levels of caregiving-related stress. The current study aims to address this gap in the literature.

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The current study focuses on three self-reported response-focused ER strategies: *reappraisal*, *suppression*, and *rumination*. These three strategies have been selected because of their relevance to mental health and emerging evidence of how they affect caregivers. The following review will begin by describing the relation between reappraisal, suppression, rumination and mental health. Next, each of these aspects will be described as they relate to caregiver-specific factors. To date, no studies have examined these constructs in caregivers of young children with ASD and later-born toddlers, and as such, research with children without ASD will be described.

Reappraisal, an antecedent-focused strategy, is commonly considered an adaptive ER strategy (Aldao et al., 2010; Beck, 1976; Clark, 1988; Salkovskis, 1998). Because anxiety and depression are often associated with a decreased use of reappraisal, CBT often focuses on teaching reappraisal strategies as a key component of the intervention (Beck, 1976; Beck, Rush, Shaw, & Emery, 1979; Clark, 1988; Salkovskis, 1998). When an individual uses reappraisal strategies to regulate his or her emotions, the use of these strategies is associated with the expression of positive emotions, strong interpersonal interactions, and well-being (Gross, 1998; Gross & John, 2003).

By contrast, suppression and rumination, both response-focused strategies, are associated with less positive outcomes than reappraisal (Aldao et al., 2010). Suppression includes attempts to inhibit the expression of emotions or thoughts (Aldao et al., 2010; Gross, 1998; Wenzlaff & Wegner, 2000). Although suppressing emotional expression and thoughts may be helpful short-term, particularly in cultures in which large expression of emotions are not considered socially appropriate, there is evidence that suppression has negative long-term impacts (Gross & Thompson, 2007; Gross, 1998; John & Gross, 2004). Suppression of emotions, whether positive

or negative, is associated with reductions in memory capacity (Richards & Gross, 2000), the use of less effective interpersonal functioning, the presence of negative emotions, and low levels of well-being (Gross & John, 2003). Rumination, or the passive and repetitive focus on one's distress, also is considered maladaptive (Nolen-Hoeksema et al., 2008; Trapnell & Campbell, 1999; Watkins, 2008). Rumination actively interferes with the use of effective and helpful ER strategies (e.g., reappraisal; Hong, 2007; Ward, Lyubomirsky, Sousa, & Nolen-Hoeksema, 2003), and is associated with depression, anxiety, eating disorders, and substance use disorders (Nolen-Hoeksema, Stice, Wade, & Bohon, 2007).

ER strategies and caregiving experiences and practices.

There is emerging evidence that ER strategies are related to caregiving experiences and practices. Of particular relevance to the proposed study, when ER strategies were added to an evidence-based parenting program, children whose caregivers were taught ER strategies demonstrated greater gains than children whose caregivers received the standard program (Sanders, Markie-Dadds, Tully, & Bor, 2000). One study found that reappraisal is negatively associated with caregiving-related stress such that caregivers who experience high levels of caregiving-related stress experience low levels of reappraisal (Solem, Christophersen, & Martinussen, 2011). Another study found that suppression is associated with caregivers' emotional expression and parenting strategies such that caregivers who suppress emotions demonstrate less positive emotion and less supportive parenting strategies compared to caregivers who do not suppress emotions (Hughes & Gullone, 2010; Meyer, Raikes, Virmani, Waters, & Thompson, 2014). Results of these two studies suggest that suppression and reappraisal should continue to be examined in caregivers of young children, as they may relate to caregiving experiences and practices.

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Rumination also has been examined as it relates to caregiving (Moreira & Canavarro, 2018; O'Mahen, Boyd, & Gashe, 2015; Stein et al., 2012). Experimenters have induced rumination by instructing mothers with psychopathology (i.e., generalized anxiety disorder and major depressive disorder) to think of a topic that worries them most and then worry about that topic as intensely as possible. After rumination occurs, mothers' interactions with their infants change, whereby mothers who ruminate are less responsive to their infants' vocalizations than mothers who do not ruminate. Researchers hypothesize that the cognitive load associated with rumination is responsible for this decreased responsiveness in mothers (Stein et al., 2012). There is further evidence that dysphoric mothers who exhibit high levels of rumination demonstrate poor effectiveness and confidence about problem-solving related to their infant (O'Mahen et al., 2015). This is critical to consider within the frame of caregiver-mediated interventions, as the cognitive load associated with learning a new intervention may be compounded if one also is engaging in rumination. A recent study found that rumination is positively associated with caregiving-related stress such that caregivers who exhibit high levels of rumination also exhibit high levels of caregiving-related stress (Moreira & Canavarro, 2018).

The relation between ER strategies and treatment adherence.

There also is emerging evidence that ER strategies are related to treatment adherence, which is particularly relevant to the current study. One study found that caregivers who exhibit adaptive ER strategies implement behavioral interventions with their young children with higher levels of treatment adherence than caregivers who exhibit less adaptive ER strategies (Maliken & Katz, 2013). Though a nascent area of research, results of studies that examine ER strategies and caregiving suggest that ER strategies may directly influence parenting experiences such as caregiving related stress, and practices such as intervention delivery and treatment adherence.

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The relation between ER strategies, caregiving-related stress, and treatment adherence has not yet been examined for caregivers of later-born toddlers; however, results from other populations suggests that ER strategies are critical to consider when examining factors that are associated with treatment adherence (Maliken & Katz, 2013). Successful intervention implementation may be related not only to caregiving-related stress but also to the strategies used to manage caregiving-related stress. For example, a caregiver with high caregiving-related stress who uses adaptive ER strategies may be more effective than one with high caregiving-related stress who uses maladaptive ER strategies.

Consequently, studying ER strategies in conjunction with caregiving-related stress may provide a more complete picture of factors that contribute to caregivers' treatment adherence in implementing interventions. These associations have not yet been examined for caregivers implementing a preventative intervention with their toddlers who are at genetic risk for ASD. The importance of studying both caregiving-related stress and ER strategies in this sample of caregivers lies in the fact that there are evidence-based strategies that can be used to improve both areas (e.g., Gruber, Hay, & Gross, 2014; Scarpa & Reyes, 2011; Watkins et al., 2011). If ER strategies ameliorate the deleterious effects of caregiving-related stress on treatment adherence, then ER strategies may represent malleable moderators (i.e., moderators that can be addressed and potentially changed through interventions such as CBT) of treatment efficacy.

The Current Study

The current study examines the relation between caregiving-related stress, ER strategies, and treatment adherence in caregivers of later-born toddlers. As suggested by the previous review, caregiving-related stress and ER strategies will be examined as predictors of treatment adherence, and ER strategies also will be examined as moderators of the effects of caregiving-

related stress on treatment adherence. Caregiving-related stress and ER strategies will be examined via self-report. Treatment adherence to a caregiver-mediated NDBI (i.e., Project ImPACT; Ingersoll & Dvortcsak, 2006) will be coded by treatment-blind observers by measuring caregivers' modeling and expanding of their child's language.

The current study will provide novel information about the relation between caregiving-related stress, ER strategies, and treatment adherence in caregivers of toddlers at risk for ASD and/or social-communication delays. Results will have implications for determining whether adjunctive interventions may be beneficial for improving caregivers' ER to improve their treatment adherence, which may have implications for the social-communication skills of their very young toddlers at risk for ASD.

Aim 1: To replicate and extend previous findings that caregiving-related stress predicts treatment adherence at T2. Though the relation between caregiving-related stress and treatment adherence has been examined in caregivers implementing interventions with their children with ASD, to our knowledge, the current study is the first to investigate this relation in caregivers implementing an intervention with their later-born toddler.

Hypothesis 1: Caregiving-related stress will predict treatment adherence such that high levels of caregiving-related stress will predict low levels of treatment adherence.

Aim 2: To examine the extent to which ER strategies predict treatment adherence at T2. One previous study has suggested that ER strategies predict treatment adherence (Maliken & Katz, 2013). As such, the current study will aim to replicate this previous finding. Reappraisal, suppression, and rumination were selected because of their relevance to mental health, their implications for intervention, as well as preliminary evidence that they are related to parenting.

Hypothesis 2: Treatment adherence will be positively associated with reappraisal, such that high levels of reappraisal will predict high levels of treatment adherence. Treatment adherence will be negatively associated with suppression and rumination, such that high levels of suppression and rumination will predict low levels of treatment adherence.

Aim 3: To examine the extent to which caregivers' ER strategies moderate the relation between caregiving-related stress at Baseline and treatment adherence at T2. The extent to which caregivers' ER strategies may moderate the relation between caregiving-related stress at Baseline and treatment adherence at T2 has not yet been examined in caregivers of later-born toddlers.

Hypothesis 3: ER strategies will moderate the relation between caregiving-related stress and treatment adherence such that the relation between caregiving-related stress and treatment adherence is stronger for caregivers with low levels of adaptive ER strategies than for caregivers with high levels of adaptive ER strategies. For example, if caregivers report high levels of caregiving-related stress and high levels of suppression or rumination, it is hypothesized that their treatment adherence will be lower than caregivers who report high levels of caregiving-related stress and high levels of reappraisal. It is hypothesized that reappraisal may serve as a protective factor against the deleterious effects of caregiving stress on caregivers' treatment adherence (see Figure 1). This hypothesis is built upon preliminary evidence that adaptive ER strategies are associated with high levels of treatment adherence (Maliken & Katz, 2013).

Method

Participants

Participants were 38 caregivers of children with ASD who had a later-born toddler between 11 and 18 months old. All caregivers were part of a larger NIH-funded multi-site randomized controlled trial (RCT) conducted at the University of Washington (UW) and

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Vanderbilt University (VU). Inclusion criteria for families in the RCT were: 1) having a toddler between 11 and 18 months of age with at least one older sibling diagnosed with ASD; 2) living within 30 miles of UW or VU; 3) no known neurological or genetic conditions in the toddler; and 4) speaking English at least 50% of the time in the home. An additional inclusion criterion for the current study was that caregivers were randomly assigned to the treatment condition of the RCT in which they learned how to implement the Project ImPACT intervention with their later-born toddler (i.e., Project ImPACT condition). All study activities received Institutional Review Board (IRB) approval at UW and VU.

A total of 44 caregivers in the Project ImPACT condition at both sites were initially approached about participating in the current study, which entailed completing 3 self-report measures of ER strategies. Of those approached, 41 (93.18%) consented to the additional study procedures. Three participants (all from UW) chose not to participate: one was too busy to complete additional measures and the other two did not want to complete measures of ER strategies. Of the 41 participants who consented to participate in the study, one family did not complete measures of ER strategies and two did not complete the T2 measure of treatment adherence (all families were from VU). As such, these three participants were excluded from the final sample. Therefore, the final sample comprises 38 participants, all of whom completed all study measures of interest.

The majority of primary caregivers were mothers (81.58%) and were highly educated (81.58%), with a college degree or higher. Caregiver age, marital status, and race were not collected. Half of the sample of later-born toddlers was female. Toddlers ranged in age from 11.43 months to 18.43 months at study entry and 78.95% of toddlers were white. Most families

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(89.47%) had only one older child diagnosed with ASD. Additional demographic characteristics of families are presented by site in Table 1.

Procedure

Caregiving-related stress and ER strategies (i.e., suppression, rumination, and reappraisal) were collected from caregivers prior to beginning the Project ImPACT intervention (i.e., Baseline). Treatment adherence (i.e., modeling and expanding their toddler's language) was measured at Baseline and after caregivers completed the 12-week, in-home Project ImPACT curriculum with a study interventionist (i.e., T2; see Figure 2 for study timeline). Caregivers had the option of completing the self-report questionnaires either via paper-and-pencil or online via REDCap (<https://redcap.vanderbilt.edu>), a secure online data collection and storage program.

Overview of the Project ImPACT intervention.

Project ImPACT is an NDBI (Schreibman et al., 2015) designed to teach caregivers to improve the social-communication skills of children at risk for or diagnosed with ASD (Ingersoll & Dvortcsak, 2006). Sessions are used to promote social-communication skills during both structured activities (e.g., snack time) and unstructured activities (e.g., free-play) in a naturalistic setting (i.e., the home). Children whose caregivers receive training in Project ImPACT demonstrate increases in blindly-coded language from pre-treatment to post-treatment (Ingersoll & Wainer, 2013a, 2013b) as well as improvements in caregiver-reported communication and social-communication skills both via questionnaires (Stadnick et al., 2015) and a qualitative interview (Pickard, Wainer, Bailey, & Ingersoll, 2016). Project ImPACT can be implemented effectively by caregivers, teachers, and early intervention providers working in the community (Ingersoll & Dvortcsak, 2006; Ingersoll & Wainer, 2013a; Ingersoll & Wainer,

2011). In the current study, caregiver coaching in the Project ImPACT approach occurred in caregivers' homes approximately twice per week over the course of 12 weeks. During sessions, caregivers received *in vivo* coaching and were assigned homework. They were encouraged to practice strategies throughout the week. For the RCT, caregiver and later-born toddler behaviors were measured during clinic visits at four assessment periods (before treatment, after treatment, and at two follow-up visits).

Measures

Information regarding constructs, time points, procedures, and variable metrics for all study variables (i.e., predictor and outcome variables) can be found in Table 2. Each study measure is described below.

Caregiving-related stress.

Caregiving-related stress was measured at Baseline using the Parenting Stress Index-Short Form (PSI-SF; Abidin, 1995), a self-report questionnaire. The PSI-SF is one of most commonly used measures of caregiving-related stress and has been used to examine changes in caregiving-related stress in studies examining the effectiveness of ASD-specific interventions (e.g., Stadnick et al., 2015; Minjarez et al., 2012). The PSI-SF has 36 items, which examine caregiving-related stress about a specific child on three distinct dimensions: Parent-Child Dysfunction, Difficult Child, and Parental Distress. A total caregiving-related stress score was derived by summing the three subscales. Totals can range from 36-180, with higher scores indicating higher levels of stress. The PSI-SF has strong face validity and Cronbach's α for the total caregiving-related stress score is .91, demonstrating strong internal consistency (Abidin, 1995). Caregivers' total scores were also converted into percentiles to characterize caregivers' levels of stress. Scores between the 15th and 80th percentile are classified as Typical Stress,

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scores between the 81st and 89th percentiles are classified as High Stress, and scores between the 90th and 100th percentiles are classified as Clinically Significant Stress (Abidin, 1995); these categories were used for descriptive purposes only.

Emotion regulation.

Caregivers' ER strategies—reappraisal, suppression, and rumination—were measured via three self-report questionnaires at Baseline. Each of these aspects was selected because not only are they considered malleable using CBT strategies (Beck, 1976; Beck et al., 1979; Clark, 1988), but there also is preliminary evidence that each of these strategies is related to caregiving practices (e.g., Hughes & Gullone, 2010; Meyer et al., 2014; Sanders et al., 2000; Solem et al., 2011; Stein et al., 2012).

The Cognitive Emotion Regulation Questionnaire (CERQ; Garnefski & Kraaij, 2007) is a 36-item measure used to identify nine distinct ER strategies: self-blame, other-blame, rumination, catastrophizing, putting into perspective, positive refocusing, positive reappraisal, acceptance, and planning. The CERQ has strong face validity and Cronbach's α for all subscales ranges from .75 to .87, demonstrating adequate to strong internal consistency (Garnefski & Kraaij, 2007). The current study examined the total scores from the positive reappraisal subscale and the rumination subscale. These subscales each comprise 4 questions. A total score for each subscale was calculated by summing responses on each of the 4 items, such that higher scores reflect increased use of positive reappraisal or rumination (Garnefski & Kraaij, 2007).

The Emotion Regulation Questionnaire (ERQ; Gross & John, 2003) is a 10-item measure that examines ER strategies in two domains: suppression and reappraisal. The suppression domain is comprised of 4 items and the reappraisal domain is comprised of 6 items. Cronbach's α is .79 for reappraisal and .73 for suppression (Gross & John, 2003). The ERQ is one of the

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most commonly used measures of self-reported ER strategies. A total score for each domain was calculated, such that higher scores reflect increased use of strategies. Both of these subscales were included because suppression is associated with negative long-term outcomes, and, as previously described, reappraisal is a key component of CBT (e.g., Beck et al., 1979).

The Response Style Questionnaire (RSQ; Nolen-Hoeksema & Morrow, 1991) contains 22 items used to examine a person's use of ruminative strategies and 11 distractor items, which are not scored. The RSQ has strong internal consistency (Cronbach's $\alpha = .90$; Nolen-Hoeksema, Parker, & Larson, 1994), and acceptable convergent and predictive validity (Butler & Nolen-Hoeksema, 1994; Nolen-Hoeksema & Morrow, 1991). A composite score for rumination was calculated from the 22 test items, with higher scores reflecting higher use of rumination than lower scores. This questionnaire was included because of its strong psychometric properties and because it is one of the most commonly used measures of rumination (Treyner, Gonzalez, & Nolen-Hoeksema, 2003).

Prior to conducting study analyses, the suppression subscale of the ERQ, the rumination subscale of the RSQ, and the rumination subscale of the CERQ were reverse scored such that high scores on each of these subscales reflect more optimal use of ER strategies (i.e., lower use of rumination and suppression) than low scores. In doing so, high scores on all ER variables reflect optimal use of ER strategies compared to low scores.

Treatment adherence.

Treatment adherence was measured as the caregivers' modeling and expanding their toddler's language during a lab-based parent-child free play (PCFP) assessment at Baseline and T2. For ease of data reporting, caregivers' use of these strategies at Baseline will be referred to as treatment adherence at Baseline, and their use of these strategies after they complete the

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ImPACT intervention will be referred to as treatment adherence at T2. The PCFP is a parent-child interaction that was selected to assess caregivers' treatment adherence because it provides a standardized way to measure caregiver behavior in a naturalistic manner. Previous intervention studies, including those that have assessed the effectiveness of Project ImPACT, have used similar protocols during which a caregiver is asked to play with their toddler using a standardized set of toys (e.g., Stadnick et al., 2015; Kasari et al., 2010). The PCFP is 15 minutes in duration. Prior to the start of the PCFP, caregivers were provided with a box of toys and given the following instructions: *"We want to see how you and your toddler play. We have some toys for you. You can set them up the way you would like. Your toddler can play with all or just some of the toys. The camera is located behind this window."*

Specific behaviors for modeling and expanding their toddler's language included caregivers' labeling, describing, or commenting on what the toddler is looking at, touching, communication about, or doing, expanding of their toddler's gestures, sounds, or words, as well as self-narration of their own activities. Nominal interval coding procedures were used, such that a caregiver received a "yes" score if they modeled and expanded their toddler's language during each five-second interval of the PCFP. Treatment adherence scores at Baseline and T2 were calculated by dividing the total number of "yes" intervals by the total number of codable intervals. Higher scores reflected greater use of modeling and expanding a toddler's language, and thus high levels of treatment adherence as compared to lower scores.

Six coders who were blind to treatment condition and time point independently coded the PCFP for caregivers' modeling and expanding their toddler's language. Coders were trained by one lead coder at VU. Prior to coding videos for the current study, coders met reliability cutoffs (i.e., interobserver agreement $\geq 80\%$) on three consecutively coded videos. Coders

began coding independently once they established reliability. After initial reliability was met, coders were randomly assigned to overlap on 20% of the videos they coded, which were used to assess reliability between coders using intraclass coefficients (ICCs); ICCs were consistently above .80 for the entire RCT sample. The final ICC for treatment adherence in the present sample at Baseline was .94, and the ICC for T2 was .74, reflecting moderate-to-good reliability.

Results

Preliminary Analyses

Prior to conducting the main study analyses, preliminary analyses were conducted. First, data were screened and participant attrition was examined. This process included assessment of normality and internal consistency of study measures, imputation of missing data, and decision-making regarding participant inclusion. Next, data reduction for ER strategies was conducted, whereby measures of the same construct were combined to create an aggregate score. Finally, demographic characteristics and site differences were examined.

Data screening and attrition.

Assessment of normality revealed that skewness and kurtosis were within acceptable ranges for all study variables (i.e., skewness $> |.8|$ and kurtosis $> |3.0|$; Tabachnick & Fidell, 2001; See Table 3). Internal consistency was strong for all study questionnaires. Cronbach's α for the PSI-SF was .93. Cronbach's α for the CERQ was .88 for positive reappraisal and .83 for rumination. On the ERQ, Cronbach's α was .91 for reappraisal and .86 for suppression. On the RSQ, Cronbach's α was .90 for rumination.

Two participants failed to answer one question on the PSI-SF. These missing values were imputed using person-mean imputation, such that the individuals' responses on the same

subscale were averaged and used to replace the missing item (Mazza, Enders, & Ruehlman, 2015; Schafer & Graham, 2002).

Participants differed in the amount of Project ImPACT “content” they received. The number of intervention sessions completed was determined not to be an accurate way of distinguishing between “treatment completers” and “non-completers,” because it was sometimes possible for interventionists to address multiple topics in one session. To be considered a “treatment completer”, all Project ImPACT content must have been delivered to the families. If families did not receive all Project ImPACT content, they were considered “non-completers”. Four families were determined to be “non-completers;” one did not receive all content because they moved out of state, and the other three had poor attendance. Data from all 38 participants were included in the analysis, regardless of whether they were deemed “treatment completers” or “non-completers”.

Data reduction for ER strategies.

To examine the feasibility of creating robust measures of reappraisal and rumination (Cohen & Cohen, 1984; Rushton, Brainerd, & Pressley, 1983; Stone & Yoder, 2001), correlations between these similar constructs were examined across measures. The correlation between the rumination subscales from the CERQ and the RSQ was $.58, p < .001$, and the correlation between the positive reappraisal subscale on the CERQ and the reappraisal subscale on the ERQ was $.62, p < .001$. Because these correlations were $.40$ or greater (Cohen & Cohen, 1984), an aggregate score was created for each construct. Because the different measures employ different scales, Percent of Maximum Possible Score (POMP; Cohen, Cohen, Aiken, & West, 1999) was used to create an aggregate score for each of these constructs. Though the CERQ uses the term “positive reappraisal”, and the ERQ uses the term “reappraisal”, both

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questionnaires are assessing the same process by which neutral or positive interpretations are produced to decrease distress. As such, the aggregate strategy was labeled “reappraisal”. Scores on the reappraisal aggregate variable and the suppression variable could range from 0-100. Scores on the suppression variable could range from 4-28. High scores on the reappraisal aggregate variable, the rumination aggregate variable, and the suppression variable reflect greater use of adaptive emotion regulation strategies than low scores on these aggregate variables.

Demographic and site differences.

Analyses were conducted to examine: 1) differences on demographic characteristics by site and 2) differences on treatment adherence at T2 by site and demographic characteristics. The sex of the later-born toddler was significantly different from UW than VU, with VU enrolling significantly more females than UW ($\chi^2 = 5.16, p = .02$). However, toddler sex did not predict treatment adherence ($p = .91$), and was therefore not included as a predictor in the primary analyses.

There were no other site differences on demographic characteristics, predictor variables, or the outcome variable $ps = .06-1.00$. The association between certain key demographic variables (i.e., who the primary caregiver was, primary caregiver education level, later-born toddler race, and number of older siblings with ASD per family) and treatment adherence (T2) could not be examined due to the small number of participants at each level. Later-born toddler sex, age at study entry, the number of older siblings with ASD per family, and the total number of older siblings in the family did not predict treatment adherence (T2; $ps = .34-.47$); therefore, no demographic characteristics were included as predictors in the primary analyses.

Descriptive Statistics, Treatment Adherence over Time, and Correlations between Study Variables

Descriptive statistics for all study variables are presented in Table 3. Caregiving-related stress was generally low; for 34 of 38 (89.47%) caregivers, caregiving-related stress fell within typical levels (Abidin, 1995). Scores for 3 caregivers (7.89%) fell in the High Stress range, and one caregiver (2.63%) reported caregiving-related stress in the Clinically Significant range. Caregivers' treatment adherence increased significantly from Baseline to T2 ($t = 5.84, p < .001$). It also is noteworthy that caregivers' treatment adherence at Baseline ranged from 9.50%-53.01%, suggesting that caregivers were using some Project IMPACT strategies upon study entry.

Pearson product-moment zero-order correlations between study variables are presented in Table 4. Of note, caregiving-related stress was negatively correlated with reappraisal ($r = -.53, p < .001$), suggesting that higher caregiving-related stress is associated with less optimal (i.e., lower) use of reappraisal strategies. Caregiving-related stress also was negatively correlated with suppression ($r = -.35, p < .05$) such that higher caregiving-related stress is associated with less optimal (i.e., higher) use of suppression strategies. Caregiving-related stress was not correlated with rumination. An unexpected finding was that caregiving-related stress was not correlated with treatment adherence either at Baseline or T2. Treatment adherence at Baseline was positively correlated with treatment adherence at T2 ($r = .54, p < .001$).

Main Analyses

Multiple linear regression models were used to address all study aims. When moderation was examined, variables were grand-mean centered prior to being included in the model. All analyses were conducted using RStudio (RStudio Team, 2016) and interaction

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models were plotted using the interActive web application (McCabe, Kim, & King, 2018). Each ER strategy (i.e., reappraisal, suppression, and rumination) was examined in a separate model for Aim 2 and Aim 3.

Aim 1. The first aim was to replicate and extend previous findings that caregiving-related stress predicts treatment adherence at T2. Caregiving-related stress at Baseline did not predict caregivers' treatment adherence at T2 ($\beta = -.15, p = .30$) when controlling for treatment adherence at Baseline (see Table 5).

Aim 2. The second aim was to examine the extent to which ER strategies predict treatment adherence at T2. Suppression ($\beta = .17, p = .23$; Table 6), rumination, ($\beta = .16, p = .26$; Table 7), and reappraisal ($\beta = .18, p = .21$; Table 8) did not significantly predict treatment adherence at T2 after controlling for treatment adherence at Baseline.

Aim 3. The third aim was to examine the extent to which caregivers' ER strategies moderate the relation between caregiving-related stress at Baseline and treatment adherence at T2. Suppression ($\beta = -.04, p = .27$; See Table 9 and Figure 3) and rumination ($\beta = -.002, p = .76$; See Table 10 and Figure 4) did not moderate the relation between caregiving-related stress and treatment adherence at T2 after controlling for treatment adherence at Baseline. The shaded region of Figure 3 and Figure 4 represent the error bars. Reappraisal was a significant moderator of the relation between caregiving-related stress and treatment adherence at T2 after controlling for treatment adherence at Baseline ($\beta = -.02, p = .047$; See Table 11). Figure 5 illustrates that caregivers with low caregiving-related stress and reappraisal above the median had higher levels of treatment adherence at T2 than caregivers with low caregiving-related stress and reappraisal below the median. However, as caregiving-related stress increased, caregivers with reappraisal above the median demonstrated lower levels of treatment adherence at T2 than caregivers with

reappraisal below the median. When reappraisal was examined as a continuous variable, two patterns emerged (see Figure 6). First, the majority of the participants are represented between -1 and 1 standard deviation from the mean on reappraisal. However, the interaction between caregiving-related stress and reappraisal occurred approximately 2 SD above the mean. Only three participants (i.e., those with high reappraisal) were responsible for the interaction. This was confirmed by examining the marginal effects graph, which provides a visual representation of the region of significance of the interaction between caregiving-related stress and reappraisal (see Figure 7). The region of significance for the interaction is 1.6 standard deviations above the mean of reappraisal. That is, for those participants who are 1.6 standard deviations below the mean of reappraisal, the interaction between reappraisal and caregiving-related stress should not be interpreted. As such, only three participants provide data for the statistically significant interaction.

Discussion

The current study was the first to examine the relation between caregiving-related stress, emotion regulation strategies, and treatment adherence in caregivers participating in a caregiver-mediated NDBI (i.e., Project IMPACT) with their later-born toddler. Results revealed that neither caregiving-related stress (Aim 1) nor ER strategies (Aim 2) predicted caregivers' treatment adherence. With respect to Aim 3, neither suppression nor rumination moderated the relation between caregiving-related stress and treatment adherence. However, reappraisal emerged as a significant moderator of the relation between caregiving-related stress and treatment adherence, such that the relation between caregiving-related stress and treatment adherence is strongest at high levels of reappraisal as compared to low levels of reappraisal. The

direction of this finding was contrary to that predicted. However, caution should be used when interpreting this result, as discussed in further detail below.

Predictors of Treatment Adherence at T2

Caregivers in the current study demonstrated treatment adherence even at Baseline, prior to learning the Project ImPACT intervention. During free play interactions, caregivers used modeling and expanded their toddler's language an average of 31.31% of the time. After treatment, the average rose to 44.19%. The presence of treatment adherence at Baseline, and the increase in treatment adherence from Baseline to T2 is consistent with other studies of the efficacy and effectiveness of Project ImPACT (Ingersoll & Wainer, 2013a; Stadnick et al., 2015). The fact that caregivers entered the study using some of these strategies is promising, as it may suggest that Project ImPACT teaches caregivers to increase strategies that they are already using instead of teaching caregivers an entirely new set of strategies.

Caregiving-related stress did not predict treatment adherence at T2.

The first aim of this study was to examine the extent to which treatment adherence at T2 is predicted by caregiving-related stress after controlling for treatment adherence at Baseline. Results from this study indicate that caregiving-related stress at Baseline did not predict treatment adherence at T2 after controlling for treatment adherence at Baseline. In addition, caregiving-related stress at Baseline also was not correlated with treatment adherence at Baseline or T2. These unexpected results may be due to the low levels of caregiving-related stress in the current sample (i.e., 89.47% of caregivers demonstrated typical levels of caregiving-related stress). These low levels of caregiving-related stress were surprising, given that high levels of caregiving-related stress are well-documented in caregivers of children with ASD (Baker-Ericzn et al., 2005; Barroso et al., 2017; Dunn et al., 2001; Estes et al., 2013;

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Hayes & Watson, 2013; Hoffman et al., 2009; Karst & Van Hecke, 2012; Kasari & Sigman, 1997; Lecavalier et al., 2006; Schieve et al., 2007).

There are several possible reasons why caregiving-related stress in the present study may have been lower than that reported in other studies. First, although later-born siblings of children with ASD are at increased risk of developing ASD or subclinical symptoms (Grønberg et al., 2013; Messinger et al., 2013; Ozonoff et al., 2011), a significant portion of these later-born siblings will not be diagnosed with ASD. Therefore, it is likely that many toddlers in the current sample are not demonstrating early ASD symptoms and may follow a typical course of development. As such, their caregivers may not have been concerned about their development or experiencing caregiving-related stress about their behavior.

Second, even if caregivers were concerned about their later-born toddler, it is possible that these toddlers were not demonstrating behavior that is often associated with caregiving-related stress (i.e., challenging behavior). Research related to caregiving-related stress in children with ASD indicates that high levels of caregiving-related stress are associated with the presence of challenging child behavior (e.g., Baker et al., 2003; Baker-Ericzn et al., 2005; Lecavalier et al., 2006; Estes et al., 2013; Hastings, 2003; Neece et al., 2012; Seltzer et al., 2010). Though their older child with ASD may be exhibiting challenging behavior, it is possible that caregivers' later-born toddlers may not be exhibiting this same challenging behavior. As such, this may explain why their caregiving-related stress about their later-born toddler is low. In fact, research indicates that at early ages, caregivers are most often concerned about their later-born toddler's language and development, not their challenging behavior (Herlihy, Knoch, Vibert, & Fein, 2015; Ozonoff et al., 2009).

Finally, caregivers in the current study may have learned strategies to help support their older child with ASD, which they are now using with their later-born toddler. If caregivers have learned effective strategies to use with their later-born toddler, it is possible that challenges associated with their later-born toddler do not outweigh their perceived resources and abilities (Abidin, 1992; Bornstein, 2002; Deater-Deckard, 1998, 2004; Rao & Beidel, 2009). Thus, their caregiving-related stress may be low because they feel equipped to address their later-born toddler's behavior.

Emotion regulation strategies did not predict treatment adherence at T2.

The second aim of this study was to examine the extent to which treatment adherence at T2 is predicted by emotion regulation strategies, after controlling for treatment adherence at Baseline. Results from this study indicate that none of the emotion regulation strategies measured (i.e., suppression, rumination, and reappraisal) predicted treatment adherence at T2 after controlling for treatment adherence at Baseline. Possible explanations for this finding are: 1) the normative levels of emotion regulation in the current sample and 2) the way in which treatment adherence was measured.

First, caregivers in the current sample demonstrated normative levels of suppression, rumination, and reappraisal (i.e., means and standard deviations) and were comparable to other published, non-clinical samples (i.e., individuals without a psychiatric diagnosis) of caregivers and adults (Garnefski and Kraaij, 2007; Hughes & Gullone, 2010; Nolen-Hoeksema & Morrow, 1991). Though ER strategies were not associated with treatment adherence in the current sample, they may still be of interest to examine as a component of overall well-being in caregivers of later-born toddlers.

Second, it may be that the lack of association between ER and treatment adherence is attributable to the way in which treatment adherence was measured. In the current study, one discrete behavior was used to measure treatment adherence: modeling and expanding their toddler's language. This discrete behavior was selected because it is hypothesized to be a key ingredient of Project ImPACT (Ingersoll & Dvortcsak, 2006; Ingersoll & Wainer, 2013a; Schreibman et al., 2015). Though treatment adherence was intentionally measured in this way, there are two possible problems with this form of measurement. First, modeling and expanding their toddler's language may represent only one of many active ingredients of Project ImPACT. Future dismantling studies will provide critical guidance regarding the active ingredients of NDBIs and the ways in which treatment adherence should be measured (Chaffin et al., 2011; Damschroder et al., 2009; Micco et al., 2007; Pellecchia et al., 2015; Van Brunt, 2000). Second, while the discrete nature of this variable is critical from a treatment efficacy perspective, it may have been too specific to capture broad aspects of caregiving practices. Studies that found evidence of an association between ER strategies and caregiving practices operationalized caregiving practices in broad ways (e.g., emotional expression and problem-solving) and therefore may have captured greater variability in caregiving practices (Hughes & Gullone, 2010; O'Mahen et al., 2015).

ER Strategies as Moderators of the Relation between Caregiving-Related Stress and Treatment Adherence at T2

The third aim of this study was to examine the extent to which ER strategies moderated the relation between caregiving-related stress and treatment adherence at T2 after controlling for treatment adherence at Baseline. Results indicate that suppression and rumination did not moderate the relation between caregiving-related stress and treatment adherence. In contrast,

reappraisal emerged as a significant moderator of the relation between caregiving-related stress and treatment adherence, such that the relation between caregiving-related stress and treatment adherence was stronger for caregivers with higher use of reappraisal than for caregivers with lower use of reappraisal. This pattern of results is different from the hypothesized pattern, which was that the relation between caregiving-related stress and treatment adherence would be stronger for caregivers with *lower use* of reappraisal. However, these results should be interpreted with caution. As depicted in Figure 7, the region of significance of the interaction between caregiving-related stress and reappraisal is quite small and captures a limited portion of the dataset (i.e., only three participants with reappraisal levels 1.6 standard deviations above the mean). As such, results from this study indicate that the relation between caregiving-related stress and treatment adherence is moderated only when caregivers demonstrate reappraisal 1.6 SD above the mean. Future studies with larger datasets could continue to examine these factors to better characterize the relation between ER strategies, caregiving-related stress, and treatment adherence.

Suppression and Reappraisal were Associated with Caregiving-Related Stress

The present study found a significant correlation between caregiving-related stress and two ER strategies: suppression and reappraisal. Though not a primary study aim, the current study replicates the association between suppression and reappraisal and caregiving-related stress (e.g., Cooper et al., 2008; Haley et al., 1987; 2003; Solem et al., 2011) in a new sample of caregivers: caregivers of later-born toddlers. Results revealed that higher caregiving-related stress was associated with less optimal use of reappraisal and suppression. That is, higher caregiving-related stress was associated with lower use of reappraisal and higher use of suppression. These associations are important for two reasons. First, the relation between these

ER strategies and caregiving-related stress suggests that reappraisal and suppression may play a crucial role in the way in which caregiving-related stress is managed. Second, since these ER strategies may contribute to the management of caregiving-related stress, it also is possible that they may impact other sequelae of caregiving-related stress, including physical and mental health symptoms (Carpenter & Steffen, 2004; Folkman & Lazarus, 1985; Hayes & Watson, 2013). As such, future studies should examine the extent to which these ER strategies protect against the negative effects of stress on other caregiver outcomes including physical and mental health. This examination could provide valuable information about ways in which these negative effects may be prevented.

Limitations and Future Directions

This study had several limitations. First, all variables, with the exception of treatment adherence, were measured via self-report. The use of self-report measures increases the possibility of the social desirability bias, which may influence caregivers to report on their experiences in a more positive light because they know that others will be reviewing their responses. Relying on self-report alone without the use of physiological measures of stress and emotion regulation limits the objectivity of the results (Factor et al., 2017). It is possible that caregivers were experiencing higher levels of caregiving-related stress than they reported via self-report. Physiological measures may provide insight into the objective experiences of caregivers.

Second, the current study was focused on the relation between caregiver characteristics and treatment adherence and did not consider the effect of these factors on toddlers' gains from treatment. Previous research has indicated that there is a relation between caregiving-related stress and gains toddlers with ASD make from interventions (Stadnick et al., 2015; Weiss et al.,

2014). The relation between caregiving-related stress and gains from treatment should also be examined in later-born toddlers. Additionally, if later-born toddlers make gains regardless of their caregivers' treatment adherence, then it is likely that other unmeasured factors are responsible for these gains. Future research should measure caregiver characteristics, treatment adherence, and toddlers' gains from treatment to fully understand the impact of these factors on intervention efficacy and effectiveness.

Third, the current sample was comprised primarily of caregivers who were white, highly educated, and mothers. As such, this sample is not representative of all caregivers of children with ASD and the generalizability of the results of the current study is limited. These caregivers may have greater support and resources than other caregivers in the community and thus demonstrate different patterns of caregiving-related stress and emotion regulation strategies. Future studies should include caregivers who are more diverse racially and educationally. Additionally, caregivers in this sample sought out intervention and monitoring for their later-born toddler. As such, this sample may represent a unique group of caregivers who are motivated to enroll in research with their later-born toddler and have the time to do so. In the future, it will be critical to characterize caregivers who are approached to participate in RCTs and choose not to participate. Their reasons for not enrolling could shed light on modifications that are needed in order to enroll a more representative and inclusive sample.

There are also several future directions which may help address questions raised by the current study. First, in addition to examining caregiving-related stress, it also may be useful to examine caregivers' self-efficacy. Caregiving-related stress and self-efficacy are closely related constructs, such that high levels of caregiving-related stress are associated with low levels of self-efficacy (Karp, Ibañez, Warren, & Stone, 2017; Kuhn & Carter, 2006). Self-efficacy

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provides information about the extent to which caregivers feel competent in their role as a caregiver. Examining caregivers' self-efficacy could provide a nuanced picture of how caregivers view themselves as effective in managing the demands associated with caring for a child with ASD or ASD risk.

Second, future studies could examine additional aspects of treatment integrity in addition to treatment adherence. The field of ASD most often examines treatment adherence (Wainer & Ingersoll, 2013) and occasionally examines dosage and density of treatment (Carr et al., 2015). However, including other aspects of treatment integrity, such as relational elements, treatment differentiation, and therapist competence (Margison et al., 2000; Mcleod et al., 2009; Perepletchikova & Kazdin, 2005; Perepletchikova et al., 2007; Waltz et al., 1993) will assist in the replication of existing research and interpretation of results across NDBIs.

Third, future studies could collect information about the older child(ren) with ASD in the family to provide a context for the caregiving-related stress caregivers report about their later-born toddler. The severity of ASD-related symptoms in the older child with ASD may impact caregiving-related stress related to their later-born toddler. For example, it is possible that caregiving-related stress in the current sample was low because the ASD severity of the older sibling was high, thus providing caregivers with a stark point of comparison between their older child and their later-born toddler. This will be particularly crucial if the PSI-SF (Abidin, 1995) is used, as this measure addresses caregiving-related stress about a specific child rather than general caregiving-related stress. As such, caregiving-related stress on the PSI-SF (Abidin, 1995) could be different depending on the child about which the caregiver is reporting.

Fourth, future studies could measure treatment adherence across multiple contexts. These contexts could include structured (i.e., a meal) and unstructured (i.e., free-play) activities as well

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as different settings (i.e., at a caregivers' home in addition to the lab). Caregivers may demonstrate different levels of treatment adherence depending on the context in which they are interacting with their later-born toddler.

In sum, results from the current study add to the literature about caregivers of later-born toddlers who participate in caregiver-mediated NDBIs. The current study provides valuable insight into the patterns of caregiving-related stress, emotion regulation strategies, and treatment adherence in caregivers of later-born toddlers. Caregivers in the current study demonstrated emotion regulation strategies similar to those reported for other samples, and some of these strategies (i.e., reappraisal and suppression) were associated with caregiving-related stress. All caregivers improved significantly in their treatment adherence from Baseline to T2, even though caregiving-related stress and emotion regulation strategies did not predict treatment adherence at T2. The current study provides support for additional examination of caregiving-related stress, emotion regulation strategies, and treatment adherence in caregivers who participate in caregiver-mediated NDBIs.

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Table 1

Demographic Characteristics of Sample, by Site

Caregiver, later-born toddler, and family characteristics	UW (<i>n</i> = 19)	VU (<i>n</i> = 19)	Total
Primary caregiver: <i>n</i> (%)			
Mothers	17 (89.47)	14 (73.68)	31 (81.58)
Fathers	2 (10.52)	3 (15.79)	5 (13.16)
Other	0 (0)	1 (5.26)	1 (2.63)
Missing	0 (0)	1 (5.26)	1 (2.63)
Primary caregiver education level: <i>n</i> (%)			
High School or GED	0 (0)	3 (15.79)	3 (7.94)
Technical degree or some college	2 (10.52)	2 (10.52)	4 (10.53)
Four-year college degree or higher	17(89.47)	14 (73.78)	31 (81.58)
Later-born toddler sex female: <i>n</i> (%)	6 (31.58)	13 (68.42)	19 (50)
Later-born toddler race: <i>n</i> (%)			
Asian	3 (15.79)	0 (0)	3 (7.94)
White	11 (57.89)	19 (100)	30 (78.95)
Black or African American	1 (5.26)	0 (0)	1 (2.63)
Mixed	3 (15.79)	0 (0)	3 (7.94)
Missing	1 (5.26)	0 (0)	1 (2.63)
Later-born toddler age at study entry (mos.)			
<i>M</i> (<i>SD</i>)	13.68 (2.26)	13.68 (1.89)	13.68 (2.06)
<i>Range</i>	11.53-18.43	11.43-17.90	11.43-18.43
Number of older siblings with ASD per family: <i>n</i> (%)			
One	18 (94.74)	16 (84.21)	34 (89.47)
Two	1 (5.26)	2 (10.53)	3 (7.94)
Three	0 (0)	1 (5.26)	1 (2.63)
Number of other siblings in the family			
<i>M</i> (<i>SD</i>)	1.89 (0.94)	1.47 (0.70)	1.68 (0.84)
<i>Range</i>	1-4	1-3	1-4

Note. UW = University of Washington; VU = Vanderbilt University

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Table 2

Constructs, Measures, and Variables Assessed at Each Time Point

Construct	Time point	Measure and	Variable Metric
Caregiving-related stress	Baseline	PSI-SF Total	Total score from the PSI-SF. High scores indicate high levels of stress.
Suppression	Baseline	ERQ suppression subscale	Total score from the suppression subscale of the ERQ, reverse scored, such that high scores represent more adaptive emotion regulation strategies than low scores.
Reappraisal	Baseline	1) ERQ reappraisal subscale and 2) CERQ positive reappraisal subscale	Aggregate of reappraisal strategies as reported on the CERQ and ERQ. Aggregate created using Percent of Maximum Possible (POMP). Scores range from 0-100 with high scores reflecting greater use of adaptive emotion regulation strategies than low scores.
Rumination	Baseline	1) CERQ rumination subscale and 2) RSQ rumination subscale	Aggregate of rumination strategies as reported on the CERQ and RSQ. Aggregate created using Percent of Maximum Possible (POMP). Scores range from 0-100 with high scores reflecting greater use of adaptive emotion regulation strategies than low scores.
Treatment adherence (Baseline)	Baseline	PCFP Modeling and expanding a toddler's language (Baseline)	Percent of the time caregivers modeled and expanded their toddler's language out of the total number of codable intervals. High scores represent greater treatment adherence than low scores.
Treatment adherence (T2)	T2	PCFP modeling and expanding a toddler's language (T2)	Percent of the time caregivers modeled and expanded their toddler's language out of the total number of codable intervals. High scores represent greater treatment adherence than low scores.

Note: PSI-SF = Parenting Stress Index-Short Form (Abidin; 1995); ERQ = Emotion Regulation Questionnaire (Gross & John, 2003); CERQ = Cognitive Emotion Regulation Questionnaire; (Garnefski & Kraaij, 2007); RSQ = Response Style Questionnaire (Nolen-Hoeksema & Morrow, 1991); PCFP = Parent-Child Free Play

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Table 3

Descriptive Statistics for Study Variables Included in Final Models

Construct	<i>M (SD)</i>	Range	Skewness	Kurtosis
Caregiving-related stress ^a	73.5 (19.71)	40-114	0.27	2.59 2.34
Reappraisal	41.74 (17.70)	2.5-72.5	-0.13	
Rumination	50.92 (17.92)	16.7-84.9	0.21	2.23
Suppression	15.95 (5.79)	4-25	-0.57	2.60
Treatment adherence (Baseline; %)	31.31 (11.73)	9.50-53.01	0.14	2.07
Treatment adherence (T2; %) ^b	44.19 (15.60)	8.94-75.42	-0.38	2.63
Participants completing all session content <i>n (%)</i>	34 (89.5)			

Note: PSI-SF = Parenting Stress Index-Short Form (Abidin, 1995); ERQ = Emotion Regulation Questionnaire (Gross & John, 2003)

^a 34 caregivers (89.47%) reported typical stress, 3 caregivers (7.89%) reported high stress, and 1 caregiver (2.63%) reported clinically significant stress

^b Caregivers' treatment adherence increased significantly from Baseline to T2 ($t = 5.84, p < .001$)

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Table 4

Pearson Product-Moment Zero-Order Correlations Among Study Variables Included in Final

Models

	1	2	3	4	5	6
1. Caregiving-related stress	--	-.53**	-.11	-.35*	-.09	-.20
2. Reappraisal	--	--	.21	.18	.23	.30
3. Rumination	--	--	--	.12	.12	.22
4. Suppression	--	--	--	--	-.09	.12
5. Treatment adherence (Baseline)	--	--	--	--	--	.54**
6. Treatment adherence (T2)	--	--	--	--	--	--

* $p < .05$; ** $p < .01$

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Table 5

Linear Regression with Caregiving-Related Stress as a Predictor of Treatment Adherence (T2)

Predictors	B	SE	β	$t(p)$
Intercept	.23	.06		3.60 ($p < .001$)
Caregiving-related stress	< -.001	< .001	-.15	-1.06 ($p = .30$)
Treatment Adherence (Baseline)	.69	.19	.52	3.69 ($p < .001$)

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Table 6

Linear Regression with Suppression as a Predictor of Treatment Adherence (T2)

Predictors	B	SE	β	$t(p)$
Intercept	.21	.06		3.43 ($p < .01$)
Suppression	< .01	< .01	.17	1.23 ($p = .23$)
Treatment Adherence (Baseline)	.73	.19	.55	3.92 ($p < .001$)

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Table 7

Linear Regression with Rumination as a Predictor of Treatment Adherence (T2)

Predictors	B	SE	β	$t(p)$
Intercept	.16	.08		1.88 ($p = .07$)
Rumination	< .01	< .01	.16	1.13 ($p = .26$)
Treatment Adherence (Baseline)	.69	.19	.52	3.65 ($p < .001$)

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Table 8

Linear Regression with Reappraisal as a Predictor of Treatment Adherence (T2)

Predictors	B	SE	β	$t(p)$
Intercept	.17	.07		2.31 ($p = .03$)
Reappraisal	< .01	< .01	.18	1.29 ($p = .21$)
Treatment Adherence (Baseline)	.66	.19	.49	3.45 ($p < .01$)

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Table 9

Linear Regression with Suppression as a Moderator of the Relation between Caregiving-Related Stress and Treatment Adherence (T2)

Predictors	B	SE	β	$t(p)$
Intercept	.21	.06		3.22 ($p < .01$)
Caregiving-related stress	< -.001	< .001	-.06	-0.39 ($p = .70$)
Suppression	< .01	< .01	.11	0.75 ($p = .46$)
Treatment Adherence	.71	.19	.54	3.77 ($p < .001$)
(Baseline)				
Caregiving-related stress*Suppression	< -.001	< .001	-.04	-1.12 ($p = .27$)

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Table 10

Linear Regression with Rumination as a Moderator of the Relation between Caregiving-Related Stress and Treatment Adherence (T2)

Predictors	B	SE	β	$t(p)$
Intercept	< .001	< .001		1.83 ($p = .08$)
Caregiving-related stress	< -.001	< .001	-.01	-.02 ($p = .99$)
Rumination	< .001	< .001	.15	1.05 ($p = .30$)
Treatment Adherence	< .001	< .001	.51	3.52 ($p < .01$)
(Baseline)				
Caregiving-related stress*Rumination	< -.001	< .001	-.002	-0.31 ($p = .76$)

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Table 11

Linear Regression with Reappraisal as a Moderator of the Relation between Caregiving-Related Stress and Treatment Adherence (T2)

Predictors	B	SE	β	$t(p)$
Intercept	< .001	< .001		2.51 ($p = .02$)
Caregiving-related stress	< .001	< .001	.69	1.71 ($p = .10$)
Reappraisal	< .001	< .001	.16	1.0 ($p = .33$)
Treatment Adherence	< .001	< .001	.39	2.65 ($p = .01$)
(Baseline)				
Caregiving-related stress*Reappraisal	< .001	< .001	-.02	-2.06 ($p = .047$)

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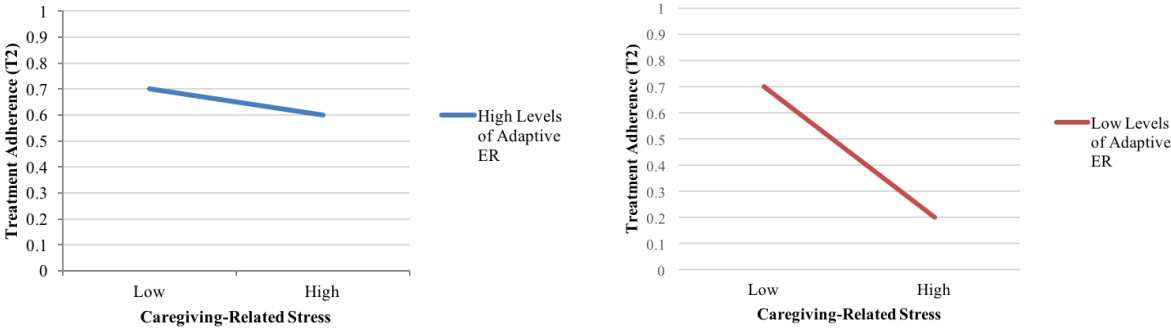


Figure 1. Hypothesized model of moderation such that the relation between caregiving-related stress and treatment adherence is stronger for caregivers with less optimal ER strategies than for caregivers with more optimal ER strategies.

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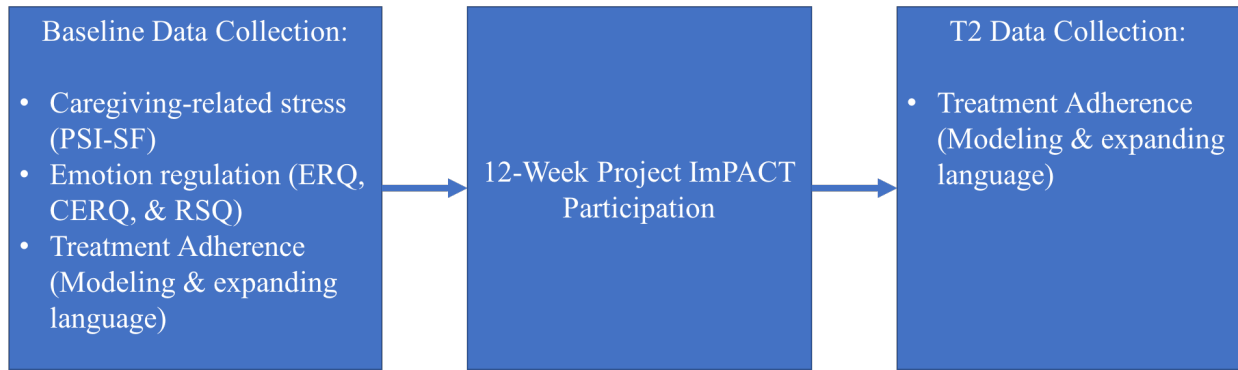


Figure 2. Description of study timeline for Participants in the Project ImPACT Condition. PSI-SF=Parenting Stress Index-Short Form (Abidin; 1995); ERQ= Emotion Regulation Questionnaire (Gross & John, 2003); CERQ= Cognitive Emotion Regulation Questionnaire; (Garnefski & Kraaij, 2007); RSQ=Response Style Questionnaire (Nolen-Hoeksema & Morrow, 1991); PCFP=Parent-Child Free Play

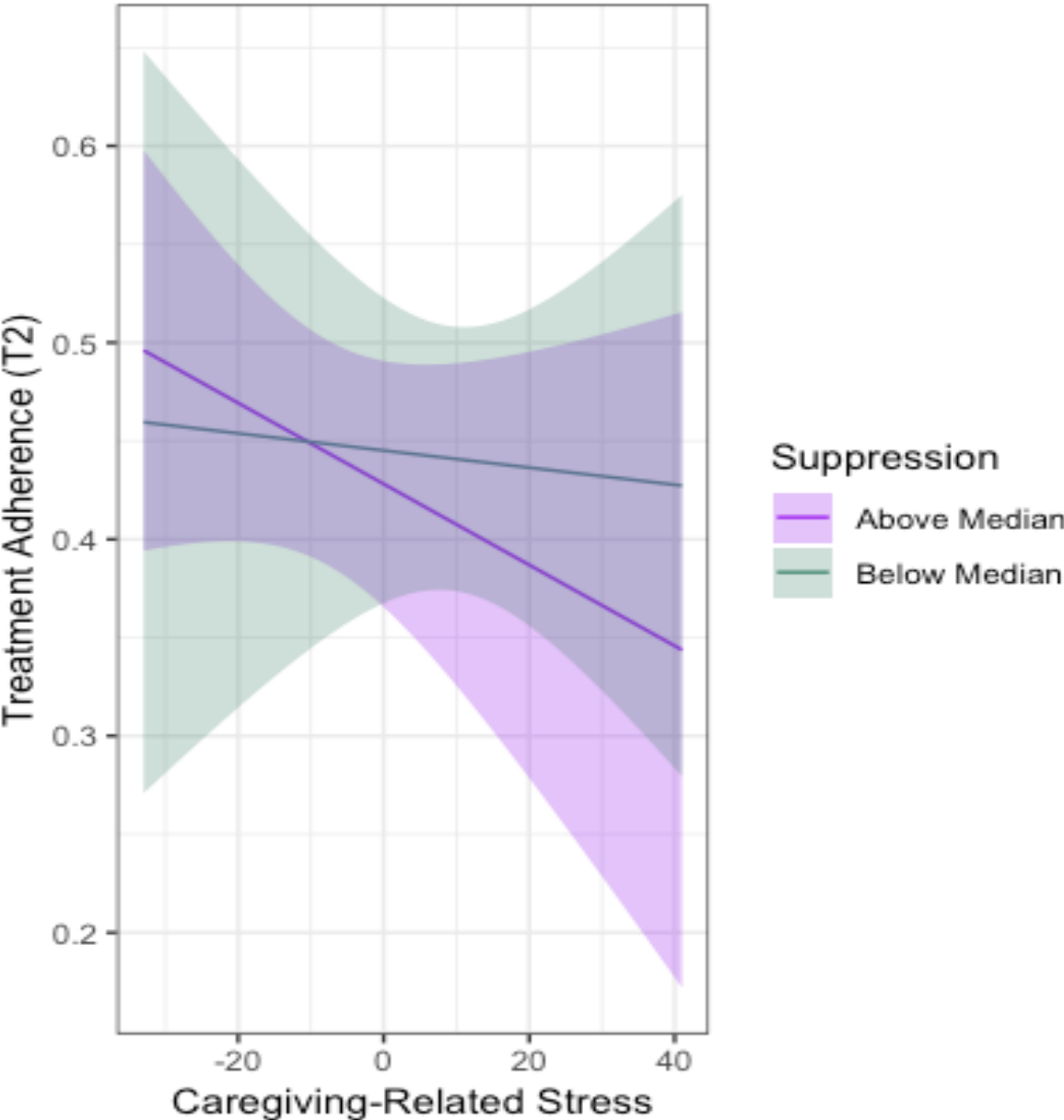


Figure 3. Suppression does not moderate the relation between caregiving-related stress and treatment adherence (T2). Shaded regions represent the error bars for each group (i.e., those above the median on suppression and those below the median on suppression).

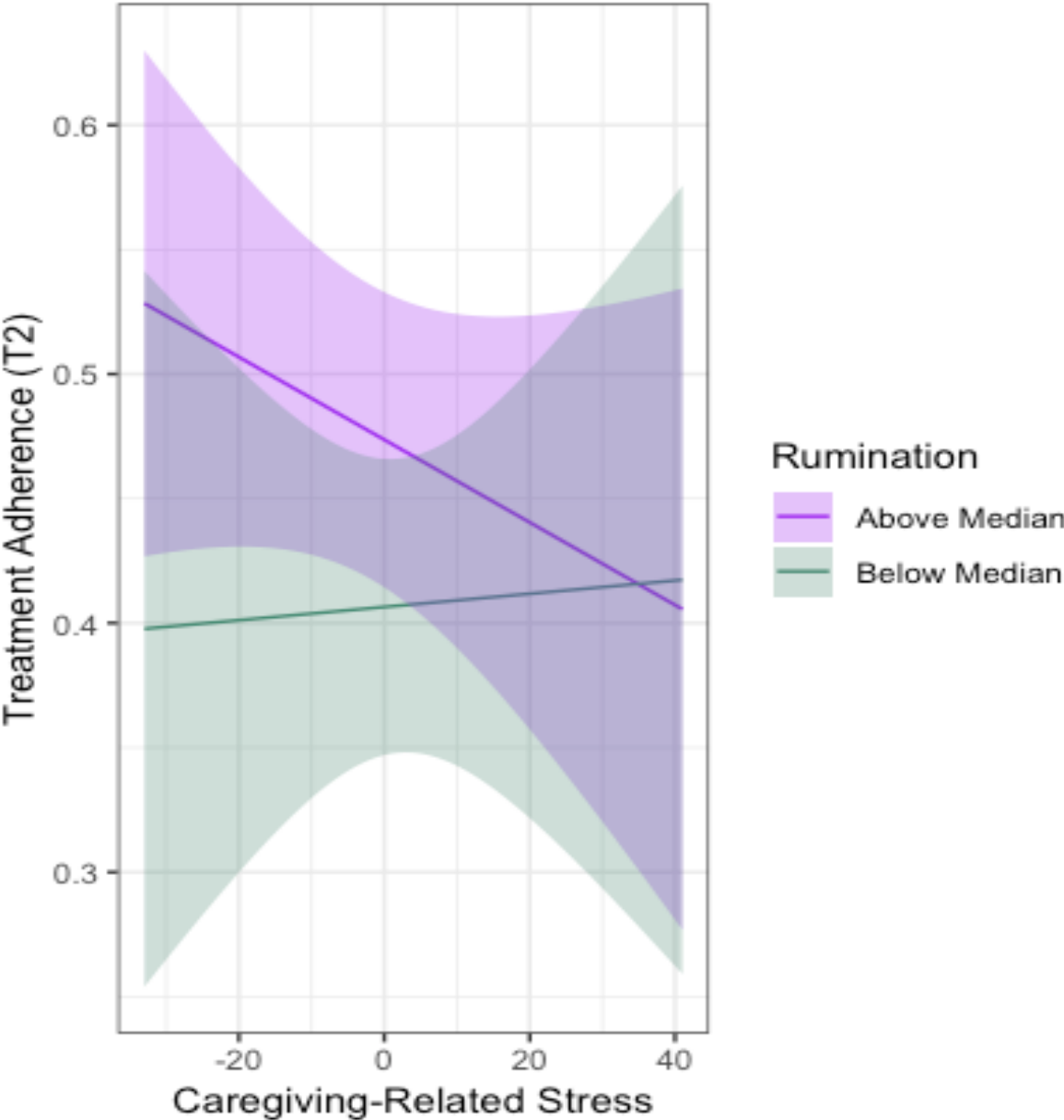


Figure 4. Rumination does not moderate the relation between caregiving-related stress and treatment adherence (T2). Shaded regions represent the error bars for each group (i.e., those above the median on rumination and those below the median on rumination).

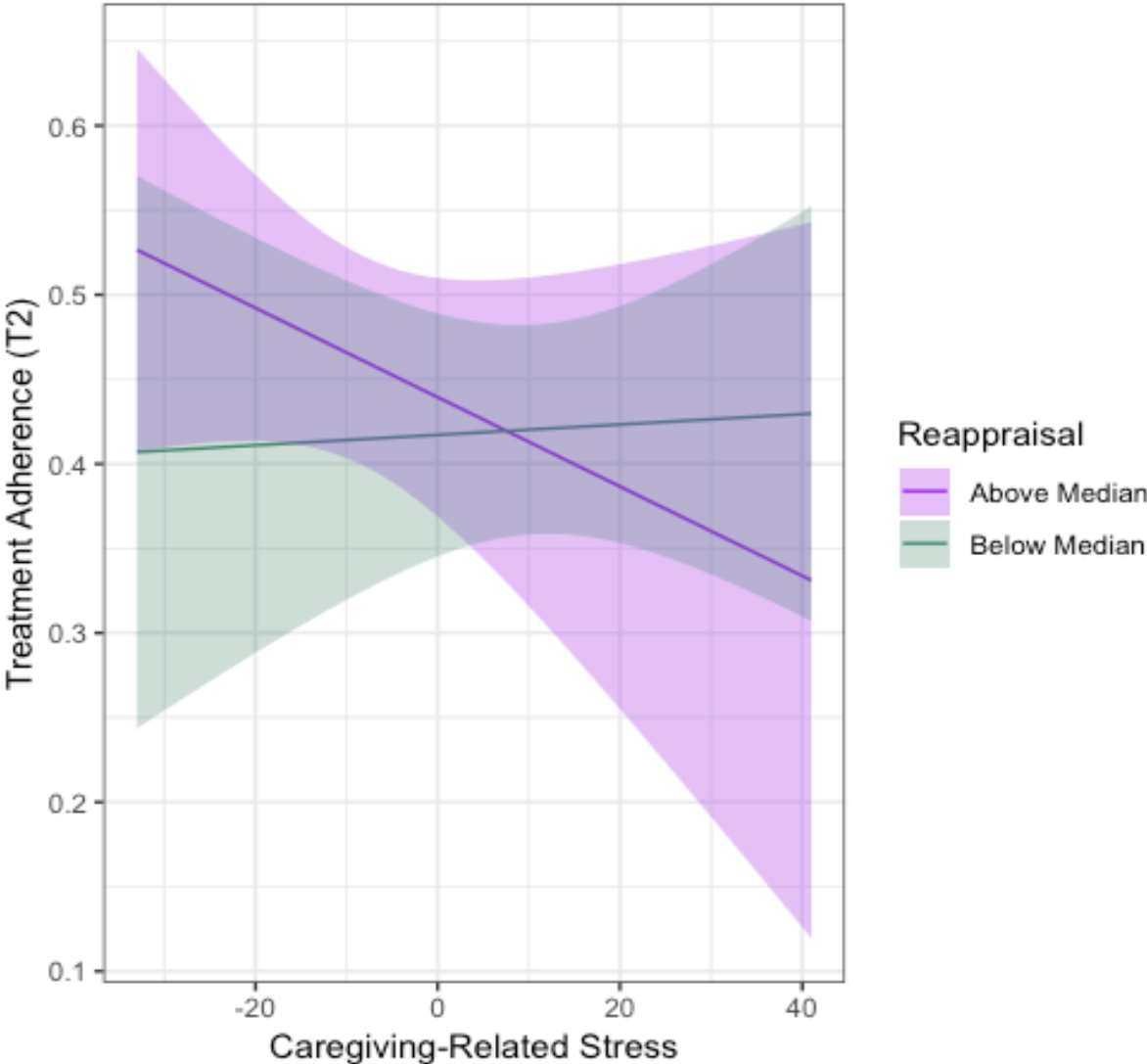


Figure 5. Reappraisal is a significant moderator of the relation between caregiving-related stress and treatment adherence (T2). Shaded regions represent the error bars for each group (i.e., those above the median on reappraisal and those below the median on reappraisal).

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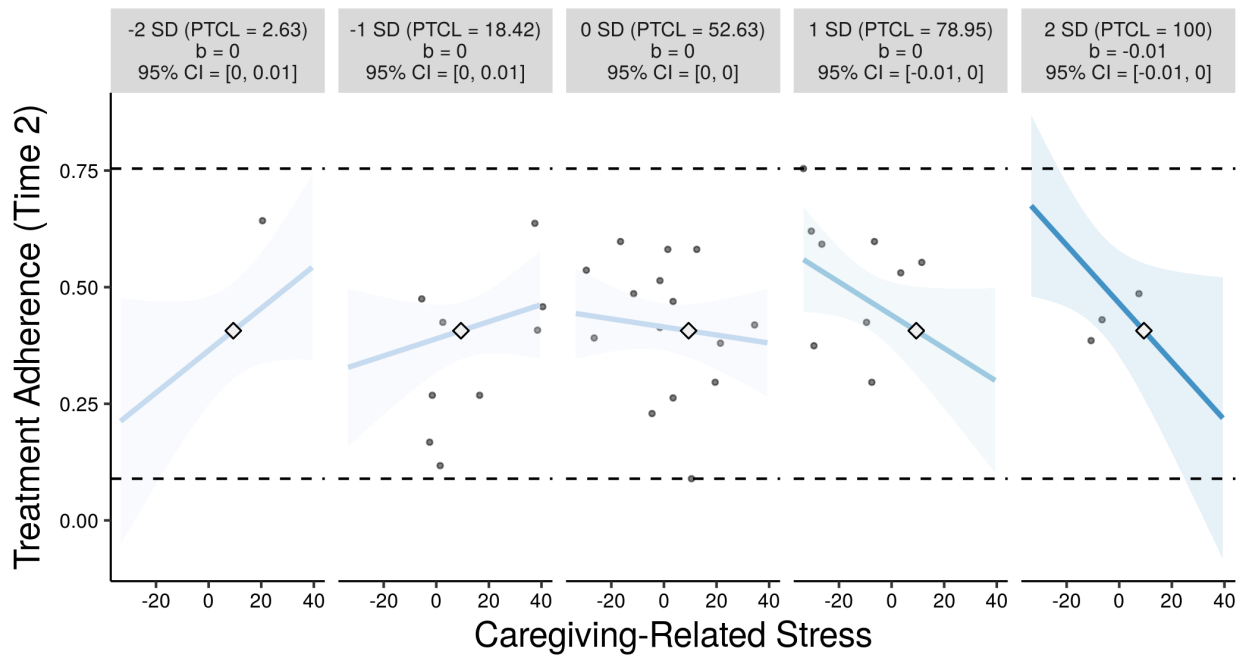


Figure 6. The interaction between caregiving-related stress and reappraisal when reappraisal is modeled as a continuous variable. Each of the five graphs represents reappraisal at the mean, and one and two standard deviations from the mean. Each individual participant is represented by a single data point on the graph. The dark blue line 2 SD from the mean represents the level at which a statistically significant interaction occurred.

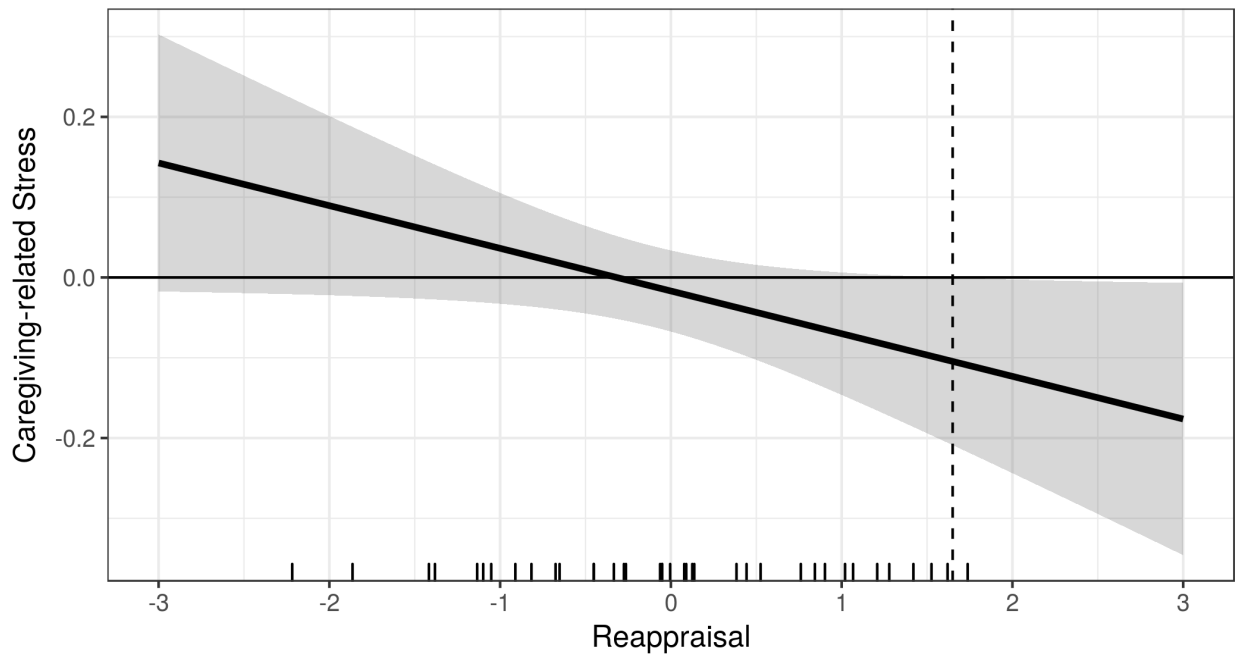


Figure 7. Marginal effects of the interaction between caregiving-related stress and reappraisal. The dotted vertical line represents the region of significance for the interaction between reappraisal and caregiving-related stress. The region of significance exists 1.6 SD above the mean.

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