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The Role of Negative Affectivity in Concurrent Relations Between Caregiver Psychological Distress and Social-Emotional Difficulties in Infants With Early Signs of Autism

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**Manuscript Title:** The Role of Negative Affectivity in Concurrent Relations between Caregiver

Psychological Distress and Social-Emotional Difficulties in Infants with Early Signs of Autism

**Short Title:** Temperament and Social-Emotional Difficulties

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**Abstract:** Recent evidence suggests the link between caregiver psychological distress and offspring social-emotional difficulties may be accounted for by offspring temperament characteristics. However, existing studies have only focused on neurotypical children; thus, the current study sought to provide an initial examination of this process among children with varying levels of early autism features. Participants included 103 infants aged 9-16 months ( $M = 12.39$ ,  $SD = 1.97$ ; 68% male) and their primary caregiver (96% mothers) referred to a larger study by community healthcare professionals. We utilized caregiver-reported measures of psychological distress [Depression Anxiety Stress Scales], infant temperament [Infant Behavior Questionnaire-Revised] and internalizing and externalizing symptoms [Infant-Toddler Social and Emotional Assessment] and administered the Autism Observation Schedule for Infants (AOSI) at an assessment visit to quantify autism features. Infant negative affectivity was found to mediate positive concurrent relations between caregiver psychological distress and infant internalizing and externalizing symptoms, irrespective of the infants' AOSI score. While preliminary and cross-sectional, these results replicate and extend previous findings suggesting that the pathway from caregiver psychological distress to negative affectivity to social-emotional difficulties might also be apparent among infants with varying levels of autism features. More rigorous tests of causal effects await future longitudinal investigation.

**Lay Summary:** Offspring of caregivers experiencing psychological distress (i.e., symptoms of depression, anxiety, and/or stress) may themselves be at increased risk of poor mental health outcomes. Several previous studies conducted with neurotypical children suggest that this link from caregiver-to-child may be facilitated by children's temperament qualities. This study was a preliminary cross-sectional exploration of these relationships in infants with features of autism. We found that infants' elevated negative emotions were involved in the relation between caregiver

heightened psychological distress and children's mental health difficulties, consistent with neurotypical development.

**Keywords:** autism spectrum disorder; infant; caregivers; temperament; social-emotional difficulties.

There is a well-established link between caregiver psychological distress and heightened risk towards internalizing (anxiety and/or depression) and externalizing (inattention/hyperactivity, oppositional, and/or aggressive behavior) symptoms among offspring (for meta-analyses, see Goodman et al., 2011, and Lawrence, Murayama, & Creswell, 2018). However, the nature of these associations is currently unclear. One potential mechanism that may account for the relation between caregiver psychological distress and child social-emotional difficulties is children's individual temperament characteristics, defined as biologically-based differences in reactivity and self-regulation (Rothbart & Derryberry, 1981). While early theories of temperament emphasized the genetic etiology and stability of traits across developmental periods (Goldsmith et al., 1987), there is growing recognition that temperament is malleable to environmental experience. Indeed, research has shown that caregiver psychological distress symptoms are associated with children's temperamental difficulties (Hanington, Ramchandani, & Stein, 2010), which in turn may confer risk towards child social-emotional difficulties (Hankin et al., 2017). Nevertheless, few existing studies have specifically tested this pathway.

Among 97 mother-child dyads, Suveg, Shaffer, Morelen, and Thomassin (2011) found that the links between maternal psychological distress and children's internalizing and externalizing symptoms were mediated by child self-regulation (i.e., the capacity to suppress or modulate emotions and behavior). Similar results were garnered by Choe, Shaw, Brennan, Dishion, and Wilson (2014) in a large sample of 677 toddlers and their mothers. Specifically, low levels of self-regulation at age 3 years was found to mediate the association between maternal depression at age 2 and toddler oppositionality at age 4. Nevertheless, Choe et al. did not explore the relevance of this pathway to children's internalizing symptoms. Allen, Oshri, Rogosch, Toth, and Cicchetti (2018) found that low child self-regulation mediated the link between maternal depression and child social-

emotional difficulties. High levels of child negative affect also acted as a mediator of this association, although there was no effect of offspring positive affect/sociability. Nevertheless, the composite measure of both internalizing *and* externalizing utilized by Allen et al. may have obscured the presence of specific internalizing versus externalizing pathways. Indeed, emerging evidence suggests that low positive affect/sociability may confer internalizing-specific risk (Hankin et al., 2017).

The current study is an initial attempt to extend empirical work on this topic to the context of autism. Our primary objective was to examine whether variation in child temperament is relevant to the links between contemporaneously measured caregiver psychological distress and child social-emotional difficulties in a sample of young infants with features of autism. Examining the relevance of this pathway to autism is important given there is a higher prevalence of internalizing and externalizing symptoms/disorders among autistic individuals than in the general population. Indeed, it is estimated that over 90% of individuals with autism meet DSM criteria for a co-occurring psychiatric disorder (Joshi et al., 2010; Salazar et al., 2015), and although these are not typically diagnosed in children under the age of 2, associated social-emotional difficulties can be identified at a very young age (Briggs-Gowan, Carter, Bosson-Heenan, Guyer, & Horwitz, 2006) suggesting potential for pre-emptive intervention.

While yet to be empirically tested, several lines of evidence suggest that the aforementioned pathway identified in neurotypical children – from caregiver psychological distress to child social-emotional difficulties through child temperament – might extend to young infants with features of autism. Symptoms of psychological distress are higher among caregivers of autistic children than comparison samples (Hayes & Watson, 2013; Yirmiya & Shaked, 2005), and positively associated with children's internalizing and externalizing symptomatology (Carter, Martínez-Pedraza, & Gray, 2009; Herring et al., 2006). Moreover, autistic children demonstrate higher negative affect, lower

positive affect/sociability, and lower self-regulation than non-autistic comparison groups (Chetcuti et al., 2019) – a temperament pattern associated with heightened levels of both caregiver psychological distress (Britton, 2011; Olino et al., 2011) and child social-emotional difficulties (Eisenberg et al., 2001, 2009). Prospective studies of infants at higher familial likelihood of developing autism (by virtue of having an older autistic sibling) indicate that a temperament profile consisting of higher negative affect, lower positive affect/sociability, and lower self-regulation might predict subsequent autism diagnosis in toddlerhood (Clifford et al., 2013; Garon et al., 2015; Del Rosario, Gillespie-Lynch, Johnson, Sigman, & Hutman, 2014; Paterson et al., 2019; for a review, see Chetcuti et al., 2019). These findings suggest there may be an early-emerging profile of temperamental susceptibility towards caregiver psychological distress and social-emotional difficulties in autism. However, the relation of temperament characteristics to social-emotional difficulties or caregiver psychological distress has yet to be explored early on in development, among infants with early autism signs.

The establishment of temporal or causal processes is beyond the scope of cross-sectional research. Nevertheless, caregiver-to-child effects were hypothesised and modelled, as studies with longitudinal measures more consistently found an effect of early caregiver attributes on subsequent child temperament than the reverse, when cross-sectional associations and stability of constructs were controlled for (Hanington, Ramchandani, & Stein, 2010; Pesonen et al., 2008), particularly in early life (Eisenberg et al., 2010). In light of evidence suggesting temperament traits function similarly across clinical and non-clinical groups (Burrows, Usher, Schwartz, Mundy, & Henderson, 2016; Schwartz et al., 2009), we expected to replicate the results obtained by Suveg et al. (2011), Choe et al. (2014), and Allen et al. (2018) in a sample of infants with autism features. Specifically, infant negative affectivity and self-regulation were expected to mediate the concurrent relation

between caregiver psychological distress and both internalizing and externalizing symptoms, while we anticipated a mediating effect of surgency only for internalizing. A secondary objective was to examine whether results generalize across infants with varying levels of autism features, though we predicted no such effects.

## Method

### Participants

Participants were 103 infants aged 9-16 months ( $M = 12.39$ ,  $SD = 1.97$ ; 68% male) and their primary caregivers recruited into a larger study [reference withheld for blinded review]. Referral to the study was by community healthcare providers, on the basis of infants showing  $\geq 3$  of 5 behavioural markers autism on the Social Attention and Communication Surveillance-Revised (SACS-R) tool (i.e., atypical/absent pointing, waving, imitation, eye contact, response to name; Barbaro & Dissanayake, 2013). The SACS-R is a revised version of the SACS (Barbaro & Dissanayake, 2010) designed as an autism surveillance tool for implementation during routine well-child checks. The SACS-R has an estimated positive predictive value of 72% when used with 12-month-olds for subsequent autism diagnosis (Barbaro, Dissanayake, & Sadka, 2018). Other inclusion criteria were child chronological age between 9- and 14-months 31 days (corrected for prematurity) and caregivers having sufficient English to understand study requirements and participate fully. Exclusion criteria were diagnosed comorbidity known to affect infant neurological and developmental abilities (including gestation  $< 32$  weeks) or family intention to relocate within 2 years of enrolment. Caregivers were on average 34.28 years old ( $SD = 5.05$ ) and predominantly biological mothers (3% biological fathers, 1% guardians). Most infants ( $n = 80$ ; 78%) had no family history of autism and, among others, an autism diagnosis was reported for an older sibling/s ( $n = 20$ ) or cousin ( $n = 3$ ).

### Procedure and Measures

This study draws on a subset of the data collected at the baseline assessment for the larger study, for which ethical approval was granted by institutional review boards. Baseline assessments occurred an average of 2.53 weeks (SD = 1.50) after eligibility screening. Caregivers provided informed consent and completed a series of questionnaires.

A short form of the Depression Anxiety Stress Scales (DASS-21; Lovibond & Lovibond, 1995) was used to measure caregiver self-reported psychological distress. DASS-21 items (21) are rated on a 4-point Likert scale, ranging from 0 (*Did not apply to me at all*) to 3 (*Applied to me very much or most of the time*). Responses across three subscales (depression, anxiety, stress) were summed to yield an overall score (range 0 to 63).

The Infant-Toddler Social and Emotional Assessment (ITSEA; Carter & Briggs-Gowan, 2006) was used to assess infant internalizing and externalizing symptoms. ITSEA items (170) are rated by caregivers on a 3-point Likert scale ranging from 0 (*Not true/rarely*) to 3 (*Very true/often*), and domain subscale mean scores are averaged to form composite internalizing (consisting of depression/withdrawal, general anxiety, separation distress, and inhibition to novelty) and externalizing (consisting of activity/impulsivity, aggression/defiance, peer aggression) scores (range 0 to 2).

The Infant Behavior Questionnaire-Revised (IBQ-R; Gartstein & Rothbart, 2003) was used to measure child temperament. IBQ-R items (191) are rated by caregivers on a 7-point Likert scale for frequency, ranging from 1 (*Never*) to 7 (*Always*), and fine-grained subscales are averaged to form three higher-order dimension scores: surgency/extraversion (consisting of activity level, smiling and laughter, high intensity pleasure, vocal reactivity, approach, perceptual sensitivity), negative affectivity (consisting of distress to limitations, fear, sadness, falling reactivity) and

orienting/regulation (consisting of duration of orienting, low intensity pleasure, cuddliness, and soothability).

Overlapping item content between the ITSEA and IBQ-R was removed to reduce measurement confounding, including the entire inhibition to novelty subscale which measures a temperament-based construct (for more details, see [reference withheld for blinded review]).

Autism features were measured by the Autism Observation Scale for Infants (AOSI; Bryson, Zwaigenbaum, McDermott, Rombough, & Brian, 2008), a direct observational measure that includes a standard set of semi-structured activities. Examiner ratings of (16) target social-communicative, sensory-motor, attentional, and play behaviours, ranging from 0 to 2 or 3, are summed to create a total score (maximum 38). Higher scores on all metrics denoted greater expression of the measured construct(s), including more autism-related behaviour.

## Results

Descriptive statistics and bivariate correlations are presented in Table 1. Child age and sex were considered as potential covariates but were mostly unrelated to other variables (see Table 1)<sup>1</sup>. There were significant inter-correlations among caregiver psychological distress, child social-emotional difficulties (internalizing, externalizing), and negative affectivity in the expected direction. Orienting/regulation and child internalizing and externalizing were negatively correlated, although neither orienting/regulation nor surgency/extraversion were related to parent depression.

<Table 1>

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<sup>1</sup>Child age was significantly correlated with externalizing,  $r = .26, p < .05$ . Inclusion of age as a covariate in the mediation model with externalizing as the dependent variable and AOSI Total score as a moderator did not substantively change the results,  $F(2,88) = 6.35, R^2 = .36, p < .01$ . The mediation effect remained significant ( $B = 0.004, 95\% \text{ bootstrap CI } 0.001 \text{ to } 0.008$ ), and the direct effect of caregiver psychological distress remained non-significant ( $B = 0.005, 95\% \text{ bootstrap CI } -0.002 \text{ to } 0.011$ ). Child age had a significant direct effect on child externalizing,  $B = 0.03, t(85) = 2.54, p < .05$ .

Analyses were performed using the PROCESS macro for SPSS (Hayes, 2018). Internalizing and externalizing symptoms were examined in separate models as the dependent variable, with caregiver psychological distress as the independent variable and child negative affectivity as the mediator. Listwise deletion of missing values resulted in a sample size of 91 for the internalizing model and 88 for the externalizing model. Since significant relations between the proposed mediator(s) and both the dependent and independent variable is a necessary precondition for testing mediation (Hayes, 2018), surgency/extraversion and orienting/regulation were not included in the models.

The full model accounted for a significant proportion of the variance in both infant internalizing symptoms,  $F(2, 88) = 25.30, R^2 = .37, p < .001$ , and externalizing symptoms,  $F(2, 85) = 11.11, R^2 = .21, p < .001$ . There was a significant positive indirect effect ( $a \times b$ ) of caregiver psychological distress on infant internalizing ( $B = 0.005$ , 95% bootstrap CI 0.002 to 0.008) and externalizing ( $B = 0.004$ , 95% bootstrap CI 0.001 to 0.008) through infant negative affectivity. The direct effect ( $c'$ ) of caregiver psychological distress was significant for infant internalizing ( $B = 0.005$ , 95% bootstrap CI 0.002 to 0.011), and non-significant for externalizing ( $B = 0.003$ , 95% bootstrap CI -0.003 to 0.010).

Next, AOSI Total score was included in the model as a moderator of the association between negative affectivity and social-emotional difficulties (path  $b$ ) in order to test the equivalence of temperament pathways across the spectrum of autism expression (i.e., AOSI Total scores ranging from 1, signifying little-to-no autism features, to  $\geq 9$  predictive of clinical diagnosis; Zwaigenbaum et al., 2005). The mediation effect remained statistically significant in both the internalizing ( $B = 0.01$ , 95% bootstrap CI 0.002 to 0.008) and externalizing models ( $B = 0.004$ , 95% bootstrap CI 0.001 to 0.009) when AOSI Total score was added as a moderator. However, the direct effect of caregiver psychological distress on infant internalizing was no longer significant. AOSI Total score did not

interact with negative affectivity in either model, and there was no direct effect; thus, there was no evidence that the mediating effects of infant negative affectivity – on the relation between caregiver psychological distress and infant social-emotional difficulties – were contingent on infants' autism expression.

Results from these models are depicted in Figure 1.

<Figure 1>

### **Discussion**

This study explored whether the relation of caregiver psychological distress to child social-emotional difficulties through temperament identified previously among neurotypical children extends to young infants with early signs of autism. Consistent with Allen et al. (2018), infant negative affectivity was found to mediate the positive association between caregiver psychological distress and concurrent infant internalizing and externalizing symptoms. There was no moderating effect of AOSI score on these indirect effects; hence, the pathway from caregiver psychological distress to infant negative affectivity to infant social-emotional difficulties may be shared across young children irrespective of whether they have autism features.

Infant orienting/regulation was negatively correlated with internalizing and externalizing symptoms, though unrelated to caregiver psychological distress. This finding contrasts with prior prospective (Hoffman, Crnic, & Baker, 2006) and mediational analyses (Allen et al., 2018; Choe et al., 2014; Suveg et al., 2011) conducted in later childhood, which have shown that caregiver psychological distress predicts children's subsequent emotional dysregulation and, in turn, social-emotional difficulties. The negative impact of caregiver psychological distress on children's self-regulation might thus be dependent on child developmental stage; such that an effect may be

apparent for later-developing top-down (deliberate) aspects of self-regulation (e.g., attention switching) but not early-emerging bottom-up (automatic) processes (e.g., attention capture).

Surgency/extraversion was unrelated to caregiver psychological distress and infant social-emotional difficulties in our sample. This is not surprising given previous inconsistencies in the literature linking children's positive affect/sociability to the family environment and developmental outcomes (for reviews, see Putnam, 2012, and Davis & Suveg, 2014). Indeed, Allen et al. (2018) found that positive affect/sociability during childhood did not mediate relations between maternal depression and offspring social-emotional difficulties. Nonetheless, a more nuanced examination of the various facets of positive affect/sociability may help resolve inconsistencies across studies relating these traits to environmental factors and child outcomes.

Level of infant autism features was positively correlated with internalizing symptoms, but unrelated to externalizing symptoms. The latter non-significant association might be due to the young age and limited behavioural repertoire of our sample, as a positive correlation between autism and externalizing symptoms been reported among autistic pre-schoolers (Tureck, Matson, Cervantes, & Turygin, 2015). Further, level of autism features was negatively correlated with temperamental surgency but unrelated to orienting/regulation and negative affectivity; consequently, the nonsignificant interaction of AOSI Total and negative affectivity in the mediation models was not all that surprising.

The results from this study should be interpreted in light of some methodological limitations. First, the cross-sectional design of this study precludes causal inference. We formulated our hypotheses under the assumption that effects flow from caregiver-to-child, although the reverse might also be true. Indeed, Choe et al. (2014) found that toddlers' oppositionality predicted subsequent difficulties with self-regulation and more depressive symptoms for mothers. Studies that

have tested bidirectional relations between child temperament and caregiver psychological distress, however, provide more consistent evidence of caregiver evocative effects than vice versa in early childhood (Hanington et al., 2010; Pesonen et al., 2008). The presence and magnitude of caregiver-to-child and child-to-caregiver effects should be elucidated in future studies through use of repeated measures multivariate modelling (e.g., structural equation modelling); specifically, evaluating whether initial child temperament and/or caregiver psychological distress predict subsequent levels of the other construct over and above cross-sectional between-construct associations and within-construct stability over time. Controlling for potential shared genetic influences on child temperament and caregiver psychological distress (e.g., through a genetically informed research design such as illustrated by Micalizzi, Wang, & Saudino, 2017) would further the robustness of this approach.

Next, the use of a single informant and method of assessment may have inflated observed relations between measures. It seems unlikely that the current results were solely due to method variance, however, given associations between caregiver and child outcomes have been observed across different methods of assessment (Goodman et al., 2011). Nonetheless, our results should be interpreted with caution until they are replicated using multiple informants and measurement methods.

Finally, it remains unknown what proportion of infants in our sample will go on to receive an autism diagnosis and/or other clinical diagnoses. The equivalence of the indirect effect of caregiver psychological distress on child social-emotional difficulties (through child temperament) across categorical diagnostic groups should be addressed in future work.

In conclusion, this study is one of few – and, notably, the first in the context of autism – to have explored temperament as a potential mechanism underlying the concurrent relation between

caregiver psychological distress and offspring social-emotional difficulties. While preliminary and cross-sectional, these findings suggest the pathway from caregiver psychological distress to child negative affectivity to child internalizing and externalizing identified in neurotypical children might also extend to young infants with early signs of autism. It is hoped that this work will provide impetus for future replications using multiple methods of assessment and longitudinal designs, as the establishment of causal relations would permit clinical translation of these findings. A tentative implication is that child and caregiver affective symptoms should be treated concurrently to promote well-being in the entire family system. Should a caregiver-to-child flow of effects indeed be borne out in longitudinal analyses, the provision of mental health support to caregivers of children with autism symptoms could reduce strain on the caregiver-child relationship and improve children's affective tolerance to, in turn, promote positive social-emotional functioning.

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Table 1. Descriptive statistics and bivariate correlations between study variables ( $N = 103$ ).

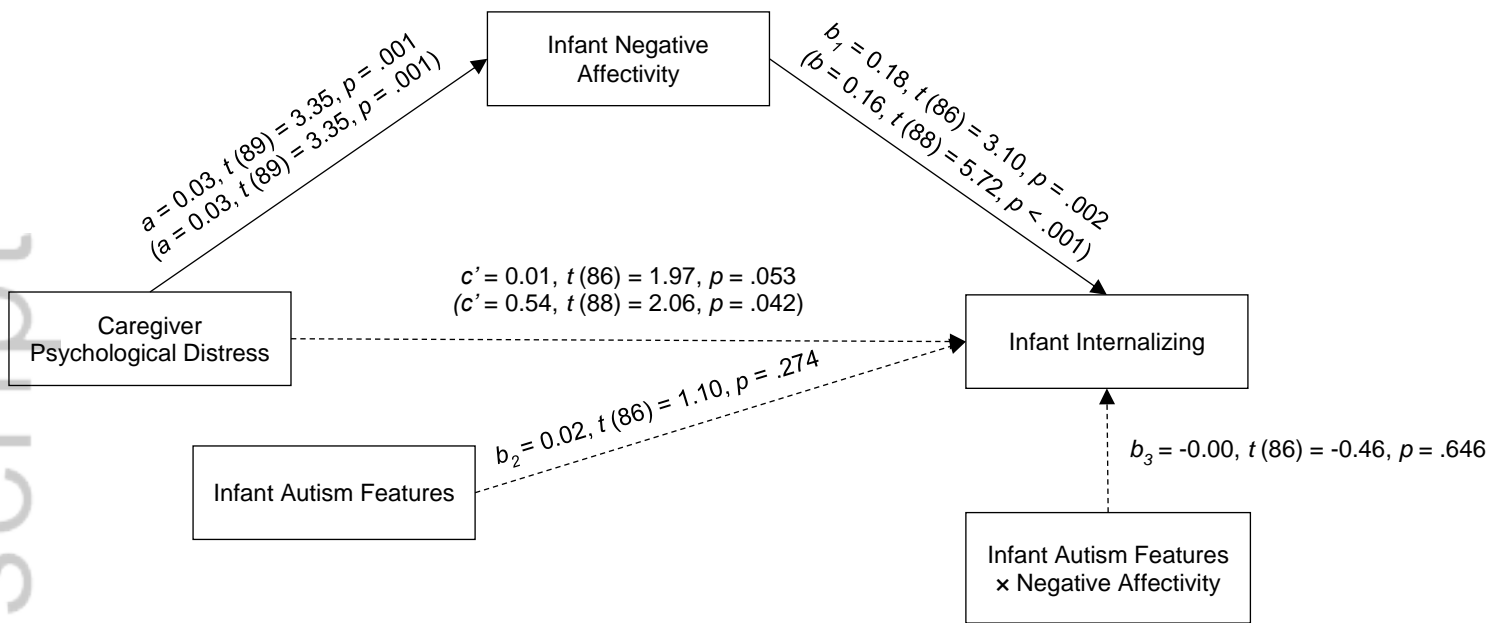
	$n$ data	$\alpha$	$n$ (%) or $M$ (SD)	Range	1	2	3	4	5	6
Child sex (male)	103	-	70 (68%)	-	.05 [-.17, .26]	.02 [-.19, .24]	-.01 [-.22, .20]	-.03 [-.24, .18]	-.01 [-.21, .20]	.02 [-.19, .24]
Child age at baseline (months)	103	-	12.39 (1.97)	9.10-16.30	-.10 [-.32, .09]	.06 [-.15, .25]	.26* [.08, .42]	.07 [-.13, .27]	.08 [-.14, .29]	.04 [-.18, .25]
SACS-R	103	-		3-5						
3 markers			32 (31%)							
4 markers			34 (33%)							
5 markers			37 (36%)							
1. DASS-21 Total	95	.86	9.41 (7.84)	0-39	-					
2. ITSEA Internalizing	91	.62 <sup>a</sup>	0.38 (0.23)	0.00-0.99	.37** [.15, .56]	-				
3. ITSEA Externalizing	88	.82	0.32 (0.25)	0.00-1.21	.24* [-.04, .48]	.23* [.04, .43]	-			
4. IBQ-R Surgency/Extraversion	96	.77	4.40 (0.73)	1.88-5.22	-.05 [-.29, .16]	-.11 [-.33, .11]	.07 [-.19, .31]	-		
5. IBQ-R Negative Affectivity	96	.78	3.28 (0.76)	1.88-5.22	.34** [.12, .53]	.58** [.41, .72]	.45** [.28, .60]	.09 [-.11, .29]	-	
6. IBQ-R Orienting/Regulation	96	.63	4.43 (0.62)	2.95-6.41	-.11 [-.30, .07]	-.21* [-.40, -.01]	-.23* [-.45, .01]	.53** [.37, .67]	-.34** [-.51, -.15]	-
7. AOSI Total	103	.66	8.90 (4.31)	1-28	.07 [-.21, .33]	.29** [.11, .45]	-.07 [-.27, .13]	-.36** [-.54, -.15]	.03 [-.19, .22]	-.05 [-.24, .14]

*Note.* Correlation coefficients were bootstrapped (5,000 resamples) to account for distributional non-normality. SACS-R = Social Attention and Communication Surveillance-Revised; DASS-21 = Depression Anxiety Stress Scales-21; ITSEA = Infant-Toddler Social and Emotional Assessment; IBQ-R = Infant Behavior Questionnaire-Revised; AOSI = Autism Observation Scale for Infants.

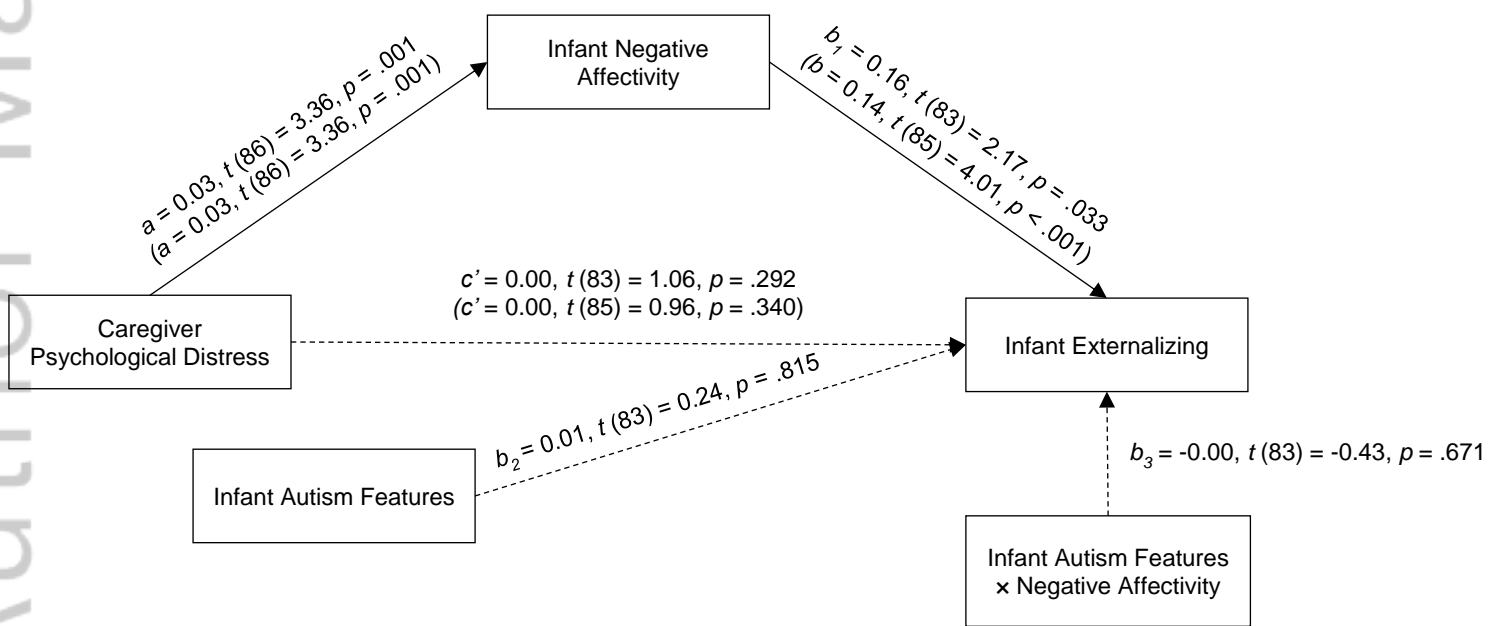
<sup>a</sup> Removal of the item with the lowest corrected item-total correlation increased the value of Cronbach's  $\alpha$  from .56 to .62 for the ITSEA Internalizing domain.

\* $p < .05$ . \*\* $p < .01$ .

*Figure 1.* Moderated mediation models investigating infant negative affectivity as a mediator of the relation between caregiver psychological distress and infant internalizing (a;  $n = 91$ ) and externalizing (b;  $n = 88$ ), including infant autism features as a moderator. Results from the initial mediation model (without the moderator) are also presented within brackets. Regression coefficients are unstandardized, and pathways in bold are significant ( $p < .05$ ).



a) Model predicting infant internalizing symptoms,  $F(4, 86) = 16.71, R^2 = .44, p < .001$



b) Model predicting infant externalizing symptoms,  $F(4, 83) = 5.75, R^2 = .22, p < .001$