

ARTICLE

Developmental costs associated with early maternal withdrawal

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Abstract

Neglect is the most prevalent form of maltreatment, but it has been understudied relative to abuse. Additionally, developmental outcomes associated with early maternal withdrawal have been understudied relative to outcomes associated with harsh treatment. However, a large body of studies on rodents has documented the causal effect of low maternal care on altered stress responses in offspring. Other evidence from human studies links early maternal withdrawal to clinical levels of neglect. Studies of both rodents and humans suggest that, rather than the aversive responses (e.g., fight, flight, freeze) modeled in relation to threat of attack or harsh treatment, early maternal withdrawal is associated with increased calling and contact seeking to mothers. Moreover, two longitudinal studies indicate that early maternal withdrawal, but not negative-intrusive interaction, contributes to adolescent borderline psychopathology. The field needs prospective studies with well-operationalized constructs of maternal withdrawal to delineate the distinct developmental pathways that may be associated with neglect.

KEY WORDS

deprivation, low maternal nurturance, maternal withdrawal, neglect, threat

Neglect is the most prevalent form of maltreatment, yet it has not been studied extensively: A meta-analysis identified 31 studies of neglect compared to more than 200 studies of sexual abuse (Stoltenborgh et al., 2015). Another meta-analysis, of observed maltreatment-related parenting behavior, found 18 studies of abuse but only eight studies of neglect (Wilson et al., 2008), with neglect associated with low parental involvement and abuse with harsh parenting. In both reviews, researchers called for more work to operationalize the assessment of caregiving behavior associated with neglect and delineate neglect-related outcomes in children (Stoltenborgh et al., 2015; Wilson et al., 2008).

In this article, we aim to increase understanding of the developmental pathways associated with infant neglect. These pathways have been understudied relative

to pathways associated with abuse. Lack of involvement by many neglecting parents has not been defined consistently, but two literatures with well-operationalized assessments of neglect-related caregiving provide evidence pertaining to children's developmental adaptations to low maternal care. A large body of randomized studies of rodents has explored the neurobiological consequences of low maternal care in rat pups. A smaller set of human studies has explored the longitudinal correlates of maternal withdrawal in infancy for children's adaptation in infancy, middle childhood, and adolescence. We expand on these points, trace the developmental trajectories related to maternal withdrawal, and suggest research to augment our understanding of the developmental consequences of early neglect.

Abbreviations: BPD, borderline personality disorder; HFPS, Harvard Family Pathways Study.

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ISSUES IN THE OPERATIONALIZATION OF NEGLECTING PARENTING

Much research has relied on social service classifications that define neglect dichotomously (Lau et al., 2005). However, natural variation in caregiving is continuous, leading researchers to suggest assessing the continuum of compromised caregiving related to neglect, rather than relying on dichotomous social service classifications (Cuartas et al., 2021; Lau et al., 2005). Yet studies of neglect-related caregiving have often been poorly operationalized, resulting in a plethora of differently labeled constructs. These constructs have been labeled as low maternal involvement (Wilson et al., 2008), detached parenting (Jones-Harden et al., 2014), emotional unavailability (Sturge-Apple et al., 2012), or parental disengagement (Doom et al., 2020). Additionally, many studies have relied on parent reports (Wilson et al., 2008), which also lack consistent definition and measurement. It remains unclear how these varied constructs map onto one another as well as how well they map onto a well-articulated construct of neglect.

Central to the construct of emotional neglect is a failure to meet the basic emotional needs of the child. Key indices of early emotional neglect include both a pervasive parental failure to respond to children's signals, particularly those of stress or distress, and a pervasive failure of parental initiative in proactively structuring the interaction with the child in protective and developmentally enhancing ways. Developing well-validated parenting measures that operationalize key aspects of neglecting parenting using clearly specified behavioral criteria would advance our understanding of neglect-related developmental trajectories. Later, we provide an overview of studies with rodents and humans that have used well-specified behavioral criteria to assess parental low nurturance/withdrawal and have identified potential neglect-related developmental outcomes.

A related construct, severe deprivation, has received attention within a framework of differentiating the neurobiological consequences of threat from deprivation (McLaughlin et al., 2014). Researchers framed harsh treatment/abuse as activating the limbic threat system, while deprivation/neglect differentially affects cognitive systems underlying language pathways and executive function (McLaughlin et al., 2014). In this framework, the primary model for deprivation has been the experience of institutional rearing, characterized by rotating caregivers, minimal care, and a bare and unstimulating environment. It remains unclear whether naturally occurring variations in withdrawing behavior by a parental figure would be included in a framework that highlights severe deprivation. Considering deprivation in terms of a continuum of caregiving would allow assessment of whether effects at lower levels of deprivation are on a continuum with those at higher levels.

RODENT STUDIES OF LOW MATERNAL NURTURANCE

As noted, researchers have framed harsh treatment/abuse as primarily activating limbic threat pathways, while deprivation/neglect affects language pathways and executive function differentially (McLaughlin et al., 2014). However, a large body of studies on rodents has documented the causal effects of low maternal nurturance (LMN) on increased stress responses of offspring. These studies have used a variety of paradigms to randomize pups' exposure to LMN (exhibited as low licking, grooming, and arched-back nursing), including randomly cross-fostering pups to less nurturing dams (Turecki & Meaney, 2016) and providing inadequate nesting material to mothers (Drury et al., 2016). Offspring of low-nurturing rodent mothers have shown increased contact seeking and calling, increased release of stress hormones, increased anxiety-like behavior, earlier puberty, and less nurturing behavior toward their own offspring. Pups have also shown marked alterations in stress-responsive limbic regions (amygdala and hippocampus). These alterations have been further tied to changes in the expression of genes that guide the development of stress-sensitive brain regions (Drury et al., 2016; Turecki & Meaney, 2016).

This work highlights the degree of survival threat associated with LMN among young rodents. LMN is associated with increased calling and contact seeking toward the mother, rather than with aversive fight, flight, or freeze responses (Meaney, 2001). This threat to the pup from LMN might be termed *threat of abandonment* to differentiate it from the more commonly studied *threat of attack or injury* that motivates fight, flight, or freeze behaviors (Sapolsky, 2004). The causal relation between LMN and activation of the threat system in young rodents raises the question of whether maternal withdrawal in young humans is also associated with altered threat responses, as well as with potential cognitive deprivation effects (see McLaughlin et al., 2014, for more on the effects of cognitive deprivation).

HUMAN STUDIES OF EARLY MATERNAL WITHDRAWAL

Studies with well-operationalized assessments of maternal withdrawal are rare in human developmental research. However, two longitudinal studies that included parenting observations from infancy suggest that distinct developmental trajectories may be associated with early maternal withdrawal. In this section, we summarize findings from two longitudinal studies with early observational data—the Harvard Family Pathways Study (HFPS), conducted in our laboratory, and the National Institute of Child Health and Human Development (NICHD) Study of Early Child Care and Youth Development (SECCYD)—and we cite other relevant studies.

Maternal withdrawal in the HFPS and related studies

The HFPS followed a relatively small group of infant-mother dyads (15% Black/Latina) from the first year of life in a cohort of low-income families (Lyons-Ruth et al., 1991). Half were referred by community providers to parenting services before children were 9 months old because of concerns about the quality of care. The aim of the HFPS was to assess risk factors and outcomes associated with infant attachment disorganization. Families were followed up in middle childhood (at 8 years) and adolescence (at 19 years).

In attachment studies, maternal sensitivity is only weakly associated with disorganized attachment (van IJzendoorn et al., 1999). Therefore, the HFPS developed a more detailed coding system for disrupted maternal interaction: the Atypical Maternal Behavior Instrument for Assessment and Classification (AMBIANCE; Lyons-Ruth et al., 1999). The AMBIANCE codes five aspects of disrupted interaction: affective communication errors, role confusion, disorientation, negative-intrusive behavior, and withdrawal. This instrument has been validated extensively in relation to infant disorganization (Madigan et al., 2006). Additionally, both negative-intrusive behavior and maternal withdrawal in the laboratory were significantly related to similar constructs coded in home observations (Lyons-Ruth et al., 1999). While sequelae of negative-intrusive parenting were the focus of early publications (e.g., Lyons-Ruth et al., 1993), distinct results related to early maternal withdrawal also emerged.

Maternal withdrawal, as defined in the AMBIANCE, comprises a coherent set of behaviors notable for the caregiver's abdication of a parental role in greeting, approaching, and comforting the infant and for the caregiver's reluctance in responding fully and promptly to the infant's cues for engagement (Lyons-Ruth et al., 1999; see Table 1). Withdrawing caregivers were reluctant to be in close contact with the infant, standing across the room or hesitating to respond when approached by the infant. The infants of withdrawing mothers tended to be insistent in their approaches and requests for contact. Often withdrawing mothers did finally respond to the infant's concerted efforts, albeit as minimally as possible. In the HFPS, maternal withdrawal was coded during the Strange Situation, a mild stressor for the infant, so the caregiver's lack of initiative and response to distress went beyond low involvement per se and was aligned more closely with constructs of low nurturance and emotional neglect. Withdrawing mothers showed little hostility or intrusiveness, so infants were undeterred in their continued pursuit of the caregiver for care, even if the resulting responses were inadequate.

Maternal withdrawal and maternal risk

A mother's withdrawal in interaction with her infant has been associated with maltreatment in the mother's

TABLE 1 Withdrawing profile of maternal behavior

- | |
|--|
| A. Lack of parental initiative around attachment (e.g., does not initiate approach, greeting, or comforting to infant) |
| B. Distanced interaction (e.g., interacts from across the room, backs away from approaching infant) |
| C. Delayed responding (e.g., hesitates before responding to infant cue, tries to deflect infant overture for contact) |
| D. Cursory responding (e.g., "hot potato" pickup and putdown, moves away quickly after responding) |
| E. Directs infant away from self to toys (e.g., uses toys instead of self to comfort infant) |
| F. Little or no hostility or intrusion |

Note: The full set of 26 behavioral items contributing to the coding of maternal withdrawal on the AMBIANCE, as well as a more detailed description of the AMBIANCE coding procedures, are available in Haltigan et al. (2019).

own childhood. In the Mother-Infant Neurobiological Development Study (35% Black/Latina/other minority), maternal withdrawal assessed at 4 months in the Still-Face Procedure was associated uniquely with mothers having experienced multiple forms of childhood maltreatment, including emotional and physical neglect (Khoury et al., 2021). In contrast, negative-intrusive behavior was associated uniquely with mothers' childhood physical abuse. In a Danish study, maternal withdrawal was also associated with greater severity of childhood maltreatment (Nyström-Hansen et al., 2019).

Clinician referral in infancy

Maternal withdrawal has also been associated with risk in the next generation. In the HFPS, maternal withdrawal in the lab was associated significantly with community providers' independent referrals to clinical parent-infant services (Lyons-Ruth et al., 2013). With each additional withdrawing behavior, the relative risk of clinical referral increased 50% (Lyons-Ruth et al., 2013). Since almost all referrals were for infant neglect, these data link withdrawal in the laboratory to clinician-assessed neglect. Furthermore, among mothers involved with Child Protective Services (74% Black/21% Latina), decreased maternal withdrawal, but not other aspects of interaction, mediated the association between randomized assignment to intervention and lower rates of infant disorganization (Yarger et al., 2020). These two studies point to the ecological validity and clinical relevance of early maternal withdrawal as a potential indicator of risk for infant neglect.

Infant disorganized/secure attachment classification

In the HFPS, maternal withdrawal was specifically associated with infant attachment behavior classified as disorganized/secure (Lyons-Ruth et al., 1999). This type of infant behavior is characterized by infant distress at

separation, and proximity to and contact seeking toward the mother on reunion, without marked avoidant or ambivalent behavior but accompanied by the contradictory, out-of-context behaviors characteristic of disorganized attachment. Thus, caregivers' distancing and hesitation in response to infants' cues may be salient signals prompting calling and contact seeking by infants. In two large studies, 55%–62% of disorganized infants were classified as disorganized/secure (Main & Solomon, 1990; Olsen, 2012). Descriptively, infants of withdrawing mothers differentially assumed the responsibility for reaching their caregivers and making physical contact, while the mothers showed little initiative. However, withdrawal also appears to create uncertainty, conflict, and disorientation in infants, as shown by concomitant disorganization. This form of infant behavior may have a functional basis in the infant's attempts to elicit care from a reluctant caregiver and decrease the possibility of serious neglect. In contrast, negative-intrusive maternal interaction was associated with infants' disorganized behavior that included avoidant and ambivalent elements (Lyons-Ruth et al., 1999), with a possible functional basis in infants' need to fight or flee threat of attack or harsh treatment.

Children's controlling/caregiving behavior in middle childhood

Few studies have examined outcomes associated with maternal withdrawal in middle childhood. In the HFPS, maternal withdrawal in infancy predicted children's controlling/caregiving (role-confused) behavior toward the

mother at age 8, assessed during the modified Strange Situation (Bureau et al., 2009). Controlling/caregiving behavior is considered a form of disorganized attachment and involves focusing unduly on the parent's needs by guiding the parent–child interaction, entertaining and praising the parent, and defusing hostility (Main & Cassidy, 1988). In contrast, early withdrawal did not predict punitive/controlling or disorganized behavior at age 8; these were predicted by the overall level of disrupted caregiving in infancy (including negative-intrusive behavior). Thus, in middle childhood, children of withdrawing mothers continue to be hypervigilant to their caregivers' needs and take more initiative in interaction than the caregivers, possibly as a continued attempt to keep the attention and involvement of a disengaged parent.

Adolescent borderline psychopathology

When the children in the HFPS were 19, researchers assessed the five dimensions of parenting in infancy as predictors of psychiatric symptomatology. Only early maternal withdrawal significantly predicted adolescent psychopathology, including features of elevated borderline personality disorder (BPD; e.g., impulsive self-damaging behaviors), suicidality, and related disorders (see Figure 1). Maternal withdrawal accounted for 20% of the variance in borderline features ($\beta = .45$) after controlling for gender, depression, and the four other parenting dimensions (Lyons-Ruth et al., 2013).

One hypothesis linking withdrawal to later risky, self-damaging behavior is that undue responsibility

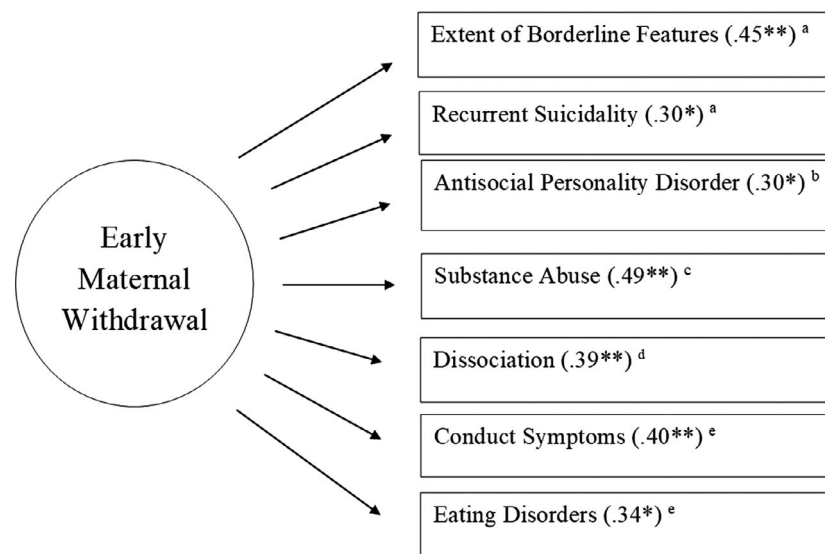


FIGURE 1 Maternal withdrawal and adolescent psychopathology on the structured clinical interview for diagnosis and the dissociative experiences scale. *Note:* Effect sizes for continuous outcomes are regression beta; effects sizes for dichotomous outcomes are eta, after adjustment for covariates. ^aFrom Lyons-Ruth et al. (2013); ^bShi et al. (2012); ^cPechtel et al. (2012); ^dDutra et al. (2009); ^eUnpublished data available from the authors. * $p < .05$ and ** $p < .01$. The structured clinical interview for diagnosis is from First et al., 1997a, b, and the dissociative experiences scale is from Bernstein & Putnam, 1986

associated with a child's hypervigilance to the needs of the caregiver might also lead to guilt, distress, and hopelessness in the child, culminating in suicidality as a way of amplifying signals of distress and eliciting care from an emotionally unavailable caregiver. Furthermore, the lack of external regulation from the primary caregiver, seen in his or her lack of initiative and structuring, may contribute to the child's pervasive lack of self-regulation when given more autonomy and responsibility in adolescence.

Potential mediators

Such long-term associations are likely to involve a number of mediators over time. As expected (e.g., Zanarini et al., 1989), maltreatment was also a significant predictor of features of BPD, but maltreatment did not mediate the effects of early maternal withdrawal (Lyons-Ruth et al., 2013; see Figure S1 in online materials). Maternal withdrawal remained an independent predictor of late adolescent suicidality, dissociation, features of BPD, and antisocial personality disorder (APD; Dutra et al., 2009; Lyons-Ruth et al., 2013; Shi et al., 2012). Additionally, neither disorganized/controlling attachment in middle childhood nor genetic stress vulnerability (number of short serotonin alleles) accounted for the effect of early maternal withdrawal on features of BPD and APD (Lyons-Ruth et al., 2007, 2013; Shi et al., 2012). Thus, early maternal withdrawal appears to contribute independently to a variety of psychopathologies by age 19, which suggests that it may undermine multiple child competencies related to self-regulation over time.

Maternal withdrawal and adolescent features of BPD in the SECCYD

The modest size of the HFPS makes replication of the association between early withdrawal and adolescent features of BPD important. Therefore, this association was also assessed using the public access dataset of the NICHD SECCYD, a longitudinal study of families (19.6% identified as African American or other minority) varying in socioeconomic status from 10 sites across the United States (NICHD Early Child Care Research Network, 2001). In the original study, researchers assessed children and families at multiple points from infancy to age 15. In the subsequent work of relevance here (Brumariu et al., 2020), a 10-item indicator of features of BPD at 15 years was developed. Then, prediction of these features of BPD was assessed from maternal behavior observed when the infants were 54 months old. Maternal behavior at 54 months was chosen because coding became more detailed at that age and included six scales (see Figure 2) rather than the three scales used at earlier ages.

Researchers conducted a latent class analysis on the six scales (Brumariu et al., 2020), which yielded four parenting profiles (see Figure 2). One profile was labeled *withdrawn* because mothers' lack of supportive involvement was clear, while hostility remained low; the withdrawn group accounted for 15% of parents. Compared to mothers in the parenting class labeled *optimal* in Figure 2, only adolescents of withdrawn mothers had significantly elevated BPD-related features at age 15 (Brumariu et al., 2020). Thus, the importance of early maternal withdrawal for adolescents' risky, self-damaging behavior was replicated in a large cohort of families at much lower social risk than families in the HFPS.

Maternal withdrawal and infant neurobiological response to threat

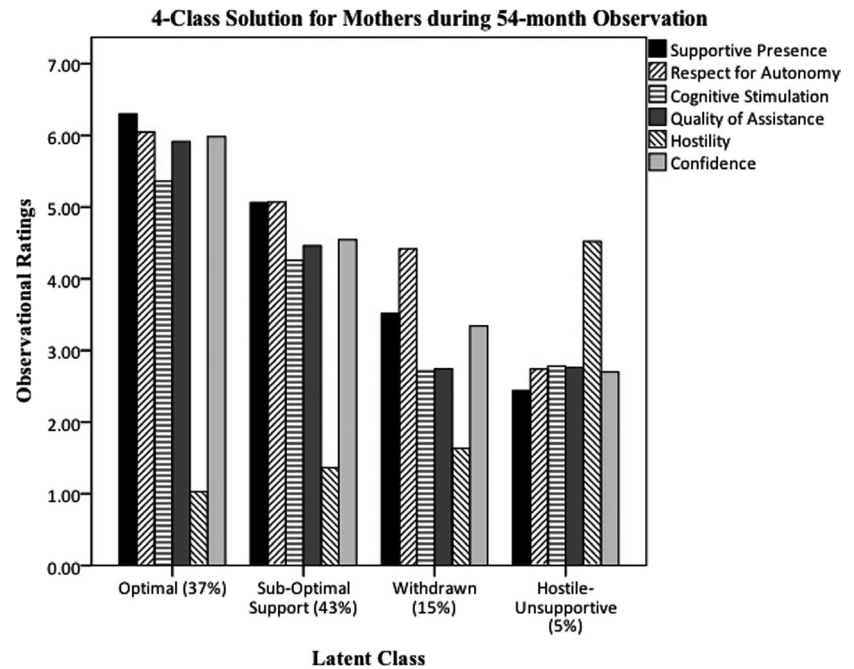
The degree to which effects on the infant threat response system in humans might contribute to the trajectory toward pathology associated with maternal withdrawal remains unknown. In a comprehensive review of the effects of maltreatment on brain structure and function (Teicher et al., 2016), larger amygdala volumes were found among children adopted from institutional settings, children of depressed mothers, and young adults with disrupted attachments in infancy in the HFPS, suggesting that low caregiver involvement may be associated with enlarged limbic volumes. Additionally, lower morning cortisol levels have been observed in young neglected children (Bernard et al., 2017). Finally, in the Fragile Families Study, lower cortisol levels in hair and increased depression at age 15 were predicted by greater parental disengagement assessed by interviews at 1 year, but not at 3, 5, 9, or 15 years (Doom et al., 2020). Thus, these data from studies of humans, together with data from studies of rodents cited earlier, suggest that early neglect may influence not only cognitive outcomes (McLaughlin et al., 2014) but also the development of infants' stress response systems.

GAPS IN KNOWLEDGE AND LOOKING AHEAD

The studies we have reviewed raise important questions about the developmental trajectories associated with neglect and reveal significant gaps in our understanding. Among the questions: How do the problematic relational constellations in infancy and middle childhood intersect with possible altered stress responding and with deprivation-related cognitive effects (McLaughlin et al., 2014) to lead to serious psychopathology in adolescence? We suggest five steps to address these questions and gaps.

First, researchers need to advance the development of scalable observational tools for indexing withdrawing/neglecting parenting. Validation of such tools in a

FIGURE 2 Results of latent class analysis of six maternal interaction scales rated at 4½ years in the SECCYD. *Note:* Four latent class model: BIC = 21882.03, Entropy = .90, LMR = $p < .01$. From Brumariu et al. (2020)



variety of home and laboratory settings is needed, as is their application to diverse samples.

Second, more work is needed to map the limbic responses associated with threat of loss/abandonment versus threat of attack/harsh treatment. The fight, flight, or freeze responses posited by traditional models of threat (Sapolsky, 2004) would be clearly maladaptive as responses to caregiver withdrawal. Instead, in the studies we have reviewed, infants responded to caregiver withdrawal with calling and contact seeking toward the caregiver. Therefore, different forms of threat appear to activate different behavioral systems. Researchers need to assess early maternal withdrawal in relation to developing infant cortisol levels, limbic brain volumes (e.g., Khoury et al., 2019), corticolimbic connectivity, and differential expression of genes regulating early stress responsiveness.

Third, children's hypervigilance to their withdrawing caregivers seems to extend from infancy to middle childhood. More work is needed to identify factors that maintain hypervigilance over time, such as caregiver vulnerabilities (e.g., substance abuse, depression) that increase children's anxiety about parents' well-being. Also, we know little about how early hypervigilance to caregivers might affect children's emerging self-regulation and executive functioning. Studies are needed to assess maternal withdrawal and infant hypervigilance in relation to neuroendocrine adaptations, and to cognitive and self-regulatory development.

Fourth, to map developmental trajectories toward adolescent psychopathology more effectively, we need more studies in middle childhood. Middle childhood data from the HFPS were based on a small sample, and other major longitudinal studies have not assessed

children's controlling behavior toward parents at this age (e.g., NICHD Early Child Care Research Network, 2001; Sroufe et al., 2005). Moreover, by middle childhood, the peer group has become an important influence that is likely to alter or augment links between early parenting and adolescent psychopathology (e.g., Brumariu et al., 2020). Increased understanding of how parent and peer relationships at this stage contribute to or modify these trajectories is needed.

Finally, interventions to decrease maternal withdrawal are promising in ameliorating infants' disorganized behavior (Yarger et al., 2020). Whether such interventions also interrupt trajectories toward adolescent self-damaging behavior is a question for the next generation of longitudinal studies on the developmental costs of neglect.

In summary, neglect is the most prevalent form of maltreatment, yet we know relatively little about its effects on child development. Important advances could be made by specifying more clearly the parenting behaviors that define neglect and by following children at high and low risk for neglect over time, focusing on the outcomes and gaps noted earlier. Most importantly, early maternal withdrawal may be one contributor to long-term trajectories toward impulsive self-damaging behaviors in adolescence, behaviors with high mortality rates and public health costs (Grant et al., 2008). Identifying children at risk for self-damaging behavior earlier in the developmental process offers the possibility of developing interventions to prevent these serious outcomes.

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