Comprehensive Psychiatry 90 (2019) 21–29

Contents lists available at ScienceDirect

Comprehensive Psychiatry

journal homepage: www.elsevier.com/locate/comppsych

Parenting behavior and the development of children with autism spectrum disorder

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ARTICLE INFO

ABSTRACT

Autism spectrum disorders (ASD) are neurodevelopmental disorders in which multiple genetic and environmental factors play roles. Symptoms of deficits in social communication and restrictive, repetitive behavioral patterns emerge early in a child's development. While parents do not cause these difficulties, impairments in social relatedness can strain parent child interactions and parental stress can have negative transactional effects that impede children development. Conversely, as with typically developing children, parental behavior can also enhance development in ASD and parents play a role in many interventions. In this review we examine parental contributions to the development of children with ASD, focusing on social communication and emotion regulation. We address parent and family characteristics that may impede development so they can be identified in families and interventions developed to target them.

> "… *mistaken stereotypes* [*of psychiatric and behavioral genetics include the idea that*] *strong effects mean that environmental influences must be unimportant…*"
>
> [(Rutter, 1991 p. 125)]

Since Leo Kanner described autism in 1943, there has been drive to discover the etiology of and contributing factors to the development of the autism spectrum disorders [1]. Many theories have emerged, looking at both nature and nurture. Currently, autism spectrum disorders (ASD) are classified as neurodevelopmental disorders in which multiple genetic and environmental factors play roles in the expression of the disorders. There has been progress in identifying genetic markers for autism, yet there is much still to be determined. There have been multiple theories on environmental factors contributing to autism, with only few showing consistent evidence for their validity.

Until quite recently, there has been a relative lack of careful investigation into the role of parenting behavior on the development and functioning of children with ASD, especially in comparison with the vast body of literature on parenting effects on typically developing (TD) children. Indeed, Schopler and Reichler (1971) presciently stated that "… little of the care lavished on classifying and describing the [autistic] children was spent on understanding the parents…." [2] (p. 89). A number of researchers have speculated that the limited investigations of parent-driven effects on development in children with autism have been in reaction to the refrigerator parent concept expounded by Bruno Bettelheim [3]. There has been a sense that such research would add to the guilt many parents of children with ASD feel about what they could or should have done differently to prevent or correct their children's problems [4,5].

Although there are clearly child-driven effects on parental behavior, this review predominantly focuses on the less studied parent-driven effects in ASD. Of course, transactional effects are always present: parents and children influence each other throughout the course of development and these dynamics make it very difficult to parse purely parent-driven effects. We initiated this review because the parents of children with ASD we see clinically often feel ineffective in parenting, and yet very much want to positively impact their children's development [6].

We review the history of parental influence on development of children with ASD, including observations of Leo Kanner and the impact of the ideas of Bruno Bettleheim, as well as the early attachment literature. We address more recent investigations of parental behavioral influences on development in children with ASD. We describe studies that aim to get at mechanisms, including longitudinal studies and those of infants at high and low risk of development of ASD. We describe the broad autism phenotype (BAP) in parents, and given that there is little overlap in studies of BAP and those assessing parental influence, we speculate on how this could impact parenting behavior and child development. Lastly, we review select intervention studies with children

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https://doi.org/10.1016/j.comppsych.2018.11.007

with ASD that include parents as co-therapists as these also suggest mechanisms of influence and illustrate particular issues.

1. Early observations about parents of children with ASD

Leo Kanner (1943) noted that a core feature of the original 11 children he described was their "extreme autistic aloneness" with preference for being alone and a lack of social awareness (1, p. 241). He reported that according to their mothers, they did not orient to being picked up and held, and the parents' comings and goings did not seem to register with the children. He also made the statement that became so controversial, that the parents of the children, although very intelligent, were not "warm-hearted". He wrote that the parents were strongly "preoccupied with abstractions of a scientific, literary, or artistic nature and limited in genuine interest in people" with marriages that were formal at best (1, p. 249). He raised the question of whether these parental characteristics had contributed to the "condition of the children" (1, p. 249). However he went on to say that the "children's aloneness from the beginning of life" made it hard to attribute to the disorder "exclusively" to the quality of the parent-child relationship (p. 249). He concluded that the children were born with an "innate inability to form the usual, biologically provided, affective contact with people…" (1, p. 250). Over time his writings indicated that he believed autism was innate, at the same time acknowledging that a child's environment can affect development [7]. Later retrospective accounts state that Kanner's descriptions of parental characteristics laid the foundation for identifying the broad autism phenotype described below [8].

2. Questions of etiology and their implications over time

The term "refrigerator" mother or parents emerged in the 1950s. Lack of parental warmth and a dysfunctional parent-child relationship were identified as the factors connected to the child's impaired social and emotional functioning. Bruno Bettelheim was the most well-known proponent of this theory [9]. In 1967, in the face of changing and increasingly neurodevelopmental perspectives on the syndrome, Bettelheim summarized his ideas that cold and rejecting parenting was causal in the emergence of autistic behaviors and childhood psychosis in infants and children [3,10]. His hypothesis was that a lack of proper early emotional stimulation damaged the developing nervous system, leading to developmental deficits in personality formation and intellectual functioning [11]. Bettelheim subsequently went so far as recommending "parentectomy" for autistic children. He postulated that an alternative environment with more emotionally nurturing conditions would allow the children to give up their autistic coping or defensive strategies [11].

In the face of this damaging hypothesis, the probability and then certainty that autism spectrum disorders (ASD) and childhood schizophrenia/psychosis (these terms were often used interchangeably in the past) were neurodevelopmental, highly heritable disorders steadily emerged [12–15]. In 1964, Bernard Rimland published his book, "*Infantile Autism*: *The Syndrome and its Implications for a Neural Theory of Behavior*" [14]. Rimland himself had a child diagnosed with autism, and the book sought to examine biological theories of development. The idea that poor parenting was a cause of autism was repeatedly challenged and eventually rejected. This evolution, led by highly effective lobbying from parents, led to an explosion of research into neurobiology, cognition, genetics, and behavioral and medical interventions in the autism spectrum disorders [9].

3. Links between parenting and autism spectrum disorders

Transactional associations between parenting and their children diagnosed with autism have been discussed for some time [16–19]. First, it has been hypothesized that a child who does not offer reciprocal affection or communication may shape the parenting style to appear "cold." Furthermore, the severity of the child's disorder, including both primary symptoms and co-occurring disorders, can lead to a cascade of stress proliferation that eventually manifests in anger and depression in parents [20]. Indeed, multiple studies have examined the stress of parenting a child with ASD – child-driven effects- and the need to support these parents, due to their distress associated with having a child who may have profound social communication, emotional, and behavioral difficulties, as well as the challenges of finding services. In addition to these psychological reactions to parenting a child with ASD, a parent who appears distant and exhibits her/his own social deficits supports the idea of a genetic basis for autism [21]. These characteristics are referred to as the broad autism phenotype (BAP) that is described more fully in Section 5 below.

Nevertheless, evidence increasingly shows that parental behavior plays a role in the development of children with ASD as it does with TD children. Some of the earliest studies examining parent-driven effects and parenting behavior addressed the role of attachment in infants with ASD. Thus, early research in parenting and parent child relationships not only described the bidirectional negative influences described above [16–19], but suggested that parental sensitivity, synchrony, and support also mattered; that children with ASD could form attachments, and furthermore could be securely attached to their mothers [22–24].

3.1. Parental sensitivity and attachment in ASD

Margaret Mahler and Michael Rutter both addressed the concept of attachment in autism [25,26]. Mahler hypothesized that the children's development had not advanced to a level to support an attachment relationship. Rutter described a "lack of attachment behavior and relative failure of bonding" [26] (p. 9). The idea that attachment relationships were incompatible with the autism diagnosis persisted for some time. A core criterion of autism in the DSM-III [27] was a "pervasive lack of responsiveness to other people." Children with autism were said to be unable "to demonstrate differential attachment to familiar people in contrast with unfamiliar adults…" [28](pg. 229). However, there was a line of evidence that not all children on the autism spectrum were so interpersonally impaired. Wing & Gould [29] identified three groups of children with ASD: 1) those who were aloof and socially remote, 2) those who moved away from social overtures but could be engaged, and 3) those who engaged in interactions in unusual or odd ways.

In the 1980s, a number of studies reported that children with ASD showed the hallmarks of secure attachment relationships, that is, they were more oriented to their caregivers than other adults and sought proximity after separation [23,30,31]. Subsequently, studies using the Strange Situation procedure (SSP) have shown that approximately 50% of young children with autism are securely attached to their mothers, that is, they show proximity seeking when stressed by the separation, controlling for difficulties with eye contact and stereotypies such as hand flapping and rocking [22,32,33]. With respect to attachment, the mothers of secure children with autism were found to be more sensitive and responsive than those of insecure children, as is true of mothers of typically developing (TD) children [22,32,34]. Because of the large effect size found between attachment security and maternal sensitivity in these cross-sectional studies, the authors of these early studies concluded somewhat prematurely that the SSP was a valid measure in young children with ASD and that the attachment system operated in children with ASD as it did in TD children.

Attachment security in autism has been correlated with language comprehension and joint attention [22]. Secure children with ASD, independent of developmental quotient, have been found to be more socially interactive with their mothers and more interested in their mothers' and the experimenter's bids for joint attention. Security of attachment has also been correlated with play behavior: attachment relationship quality of children with ASD was a better predictor of the level of play behavior than the child's disorder, controlling for developmental level [35]. Similarly, pre-school children's language development has

been linked to both parental and children's responsiveness in earlier play settings, with each variable being independent of the other [24].

It appears that fathers and mothers differ in their strategies with their children with ASD. Fathers are less active in engaging their young children with ASD [36,37]. Fathers are reported to be more directive in general [38]. However, in a cross-sectional study, those fathers who were more sensitive and verbally responsive, that is, those who were positive and contingent in response to their children with ASD, had children with higher language scores [36].

Nevertheless, it is very important to note that these assessments of parental sensitivity and attachment in children with ASD were largely been cross-sectional and therefore did not rule out the idea that higher functioning/more social children are 1) more capable of showing attachment behaviors and 2) elicit more responsive parenting.

3.1.1. Does the attachment system develop differently in children with ASD?

Evidence has been accumulating that the attachment system does not develop in the same ways in children with ASD as it does in TD children. For example, a longitudinal study of separation and reunion behaviors in children with ASD, mixed non-ASD disorders (e.g., intellectual disability, attention deficit hyperactivity disorder (ADHD), speech and language disorders), and TD children [39], found that separation behaviors were very similar for children in all the groups. The children in the ASD group's reunion behavior, in particular, their pro-social acknowledgement of the parent's return (smiling, looking, holding arms out) lagged behind the other children in the expression of these behaviors at the <13 month assessment, but they had essentially the same levels of pro-social greetings by the time they reached 24 months of age. More impairment as measured with the Autism Diagnostic Observational Schedule (ADOS) [40] was associated with fewer pro-social reunion behaviors. A small number of the children in the ASD group never appeared to react to the separation and reunion.

van IJzendoorn and colleagues (2007) also reported that two year-old children's social interaction scores on the ADOS [40] were highly correlated with attachment security in the SSP [41]. The mothers of children with ASD were as sensitive as mothers of other children (both TD and intellectually disabled (ID)). However, attachment security in the children with ASD was not correlated with maternal sensitivity, as was the case with both the TD and ID groups. The authors suggested that parents of children with ASD are less able to impact the children's constitutional deficits in mirroring, and that parental "sensitivity", at least as it is commonly assessed, does not impact children with ASD as attachment theory would suggest.

Another study of attachment behaviors assessed in middle childhood (age 7–14) also found that children with ASD had difficulty with using their caregivers as a secure base and assisting them with emotional regulation [42]. This finding implies that despite the finding that children with ASD manifest attachment behaviors in response to stressful situations, some of the adaptive benefits of attachment security with regard to emotion regulation are not fully developed in these children. This hypothesis is further investigated in studies of parental stress buffering mechanisms as described below [43,44].

3.2. Emotion regulation

Another line of research examines emotion regulation (ER) and the stress buffering behaviors of parents of children with ASD. These behaviors are closely linked to investigations of attachment behavior and parental sensitivity insofar as the secure base function of the attachment system promotes recognizing and managing potential dangers and distress in balance with supporting exploration and mastery [45,46].

Gottman and Katz (1989) defined ER as the ability to "(a) inhibit inappropriate behavior related to strong negative or positive emotion, (b) self-soothe any physiological arousal that the strong affect has induced, (c) refocus attention, and (d) organize for coordinated action in the service of an external goal" [47](p. 373). ER is conceptualized as an executive function that plays a role in regulating and inhibiting behaviors.

While this executive function has a neurobiological basis, parents play a central role in the development of ER via soothing, organizing and refocusing, and they enhance the development of more independent regulatory strategies in TD children. Parental co-regulation is defined as, "a parent's support of their child's development through motivational or emotional scaffolding, and using strategies to help their children modulate their emotions…" [48]. Emotional scaffolding includes parents' ability to make an experience a positive and/or safe one for the child, including protection, valuing the child's involvement in the activity, and maintaining sensitivity and support with respect to the child's emotions. Motivational scaffolding refers to parents' ability to help the child maintain enthusiasm toward an activity, including praise and encouragement, and redirecting attention back to the task at hand [49,50].

Poor ER in individuals with ASD appears to be associated with impaired cognitive appraisal of emotion-eliciting stimuli [51]. Hypersensitivity to sensory stimuli and poor coping skills such as avoidance also are factors contributing to the observed increased intensity of emotion frequently exhibited by individuals with ASD. Previous conceptualizations of the poor ER associated with ASD have focused on co-occurring psychiatric disorders; i.e., many children with ASD have anxiety and attention difficulties that also are associated with impaired ER. More recently, ER dysregulation has been hypothesized to be an intrinsic feature of ASD that renders individuals with ASD at greater risk of developing comorbid disorders [51].

ASD symptoms in the social communication and interaction domain negatively impact parental sensitivity, joint attention, and dyadic attunement, factors that are core to parental ER strategies with young children. Greater severity of the child's symptoms has been associated with less attunement and synchrony between parents and children [52]. Guo and colleagues [53] found that mother-child dyadic interactions with ASD children had more mismatched emotion-engagement states (e.g., child negative/mother positive) and the children spent more time during parent-child interactions engaged with objects compared to TD children.

3.2.1. Parental sensitivity and emotion regulation in ASD

In a series of observational investigations of ER, children with ASD and their parents were compared with TD dyads. The children in the ASD and TD groups were matched on all domains of the Stanford-Binet Intelligence Test as well as age (although the children in the ASD group were slightly older), gender, and family demographics. The parents of preschool children with ASD did not differ from those of TD children with respect to support and discipline qualities, despite the greater degree of withdrawal and more negative emotion expressed by their children [54]. The children with ASD were observed to seek help from their parents when distressed, and child cooperation was associated with supportive parenting. In comparison to the more complex strategies used by parents of TD preschoolers, e.g., reframing, diverting attention, and reappraisal techniques, the mothers of the children with ASD were more likely to use simpler strategies of physical comfort and holding gaze as means of regulating emotions.

In a related study, the children were presented with a still face paradigm executed by each parent 1 month apart [44]. During the still-face episode, the children with ASD were distressed and increased their social gaze toward their parents. They used simpler emotion regulation strategies than the TD children, including physical and verbal self-soothing with various repetitive behaviors. The mothers and fathers of children with ASD showed more positive affect before and after the still-face and more social gaze after the still-face episode compared with the parents of the TD preschoolers. The authors hypothesized that this was indicative of the great parental regulatory effort in the moment and throughout development required for children with ASD.

This study also assessed cortisol responses to the still face stressor [44]. Interestingly, the children with ASD did not show any variability in cortisol levels when with their mothers, whereas with their fathers they showed the same responses as TD children, i.e., higher at baseline with a recovery period. The authors suggest that the ER strategies of the mothers of children with ASD, and the lack of cortisol response to novelty shown in their children, indicate "that the typical cortisol response in children with ASD … is suppressed by maternal presence… [This phenomenon is] observed during the transition period before the mature self-regulated HPA function develops in rodents. … one mechanism in the pathophysiology of ASD may relate to abnormal extension of the HPA sensitive period, with mothers exerting social buffering effect for much longer periods and the system not maturing to its full adult profile for extended intervals" [44](p. 20).

3.2.2. Parental emotional state and emotion regulation in ASD

As is true for parents of TD children, parents of ASD children report that their own emotional states impact their children's emotions and behavior [55]. Cortisol levels can be used to assess linked emotional stress in dyads. Closely linked cortisol levels between members of a parent-child dyad indicate similar stress levels and are considered to be indicators of less effective ER by the parent. That is, child distress and parental distress are mirroring each other rather than parent down-regulating and soothing child stress responses [43]. Two groups of preschoolers (TD and those with ASD) were seen with each parent for a series of play activities. Examination of the linkage between cortisol levels of children with ASD and their parents, show different results for mothers and fathers [43]. For all children, paternal cortisol levels predicted child cortisol levels indicating less effective ER by fathers overall. The correlation was weaker for more sensitive fathers. In contrast, mothers' cortisol did not predict children's cortisol levels, indicating more effective ER from mothers.

Parents' self-reported difficulties in ER strategies predicted lower levels of positive and collaborative dyadic behaviors with their 4- to 6-year-old children with ASD [56]. Furthermore, parental self-reports of negative parenting behaviors are associated with their reports of children's externalizing behavior, a manifestation of difficulty with ER [57]. Ting & Weis [48] conducted an observational study of parental scaffolding (e.g., prompting, following the child's lead, reassurance) for children with ASD aged 8–12 years. In an emotion discussion task in which the dyad had to discuss times when the child felt happy, anxious, and angry, more scaffolding behaviors by the parents were associated with lower parental reports of anxiety and anger in the children, controlling for child age or IQ.

In non-ASD families, negative emotion and criticism by parents have been shown to predict behavioral problems in children [57]. The same association between negative emotion in mother and offspring behavioral problems has been reported for teens and adults with ASD (aged 10–47 years) across an 18-month interval in a sample of over 400 families [58]. A follow-up study of this older sample with four assessments over seven years aimed to clarify mechanisms and bi-directional effects, using trajectories of maternal criticism and offspring behavior [4]. Changes in maternal criticism were found to predict behavioral problems, but change in behavior problems did not predict maternal criticism. Intellectual disability level was not related to criticism. Interestingly, graduation from high school was associated with increased maternal criticism at the time of this transition, but in this case, the teen behavior problems did not increase. The phenomenon was hypothesized to be due to maternal anxiety about the teens' futures.

A subset of this sample was studied to examine the effects of mother-child relationship quality as reported by the mother as well as mothers' warmth and praise captured in a 5-minute speech sample, on offspring behavior across a two-year period [18]. Mothers frequently spoke highly of their children and expressed considerable warmth both in verbal utterances and in tone. Relationship quality, warmth, and praise were associated not only with reduction in problem behaviors across the two years, but in a reduction in autism symptoms of repetitive behaviors, controlling for children's IQ.

3.3. Parenting behavior and social and communication development

Social communication impairments are a hallmark of ASD especially in the domains of joint attention, imitation, and language development. In TD infants, parents support language development by engaging in familiar interactions that focus the child's attention on aspects of the environment and shared experience [59]. In addition, parents who track children's attention and activities and give contingent language input in those situations, have children who acquire language more rapidly.

Children with ASD spend much less time in joint-attention activities than TD children, that is, 30% of interaction time versus 76% for TD children and 78% for children with Down Syndrome [60]. In a study of children with ASD followed over four waves of data collection during the preschool years, children who were more responsive to bids for joint attention acquired language more rapidly [59]. The same was true when their parents were more responsive to the children's activities and focus. These two predictors of language development were independent of each other and were not explained by the children's initial language skills or IQ.

Similarly with respect to social skills, in a study of preschool children in a joint wordless picture book task, emotional support and cohesive structuring by the mothers were associated with pre-school children's social skills for both TD children and those with ASD, whereas mother's positive affect was not [61]. In a third study by Patterson and colleagues, maternal responsiveness, that is, behavior that is sensitive, contingent, and followed the child's lead, predicted child-initiated joint attention in 2–3 year olds with ASD, whereas mother-initiated joint attention was predicted by maternal directiveness [60].

3.4. Parenting and the development of empathy

Although empathy is a heritable trait, it is also influenced by the environment [62]. Children with ASD have been described as having impairments in empathy as well as theory of mind. In a short longitudinal study, Rozga, et al. (2018) examined empathic responses in young children with ASD a year after their initial assessment of attachment [32]. Both attachment security and language development predicted empathic responses at a one-year follow-up of these children. Controlling for language development, the children classified as secure showed gains in empathic responses after one year whereas the insecure children showed no gains.

In contrast, in an investigation of toddlers at high risk (HR) for ASD, parental interaction behaviors did not explain differences in the children's empathic responses [63]. However, there was an interaction effect: children were less empathic when they had a genetic polymorphism of a common oxytocin receptor gene, rs53576, *and* the parent interaction behaviors were of low quality.

4. Looking at the dyad from infancy in high risk samples

Taking partner behavior into account over time may clarify the contributing role of each member of the dyad [64,65], and is critical to understanding mechanisms. As is true for the studies discussed above, the focus of dyadic studies examining the effects of parenting on infant development has focused on behaviors of parental sensitivity, responsiveness, directiveness, and emotion regulation strategies. All of the studies described above were conducted after the parent-children relationship has played out for years. Parent-infant studies aim to avoid this confound.

Most of these investigations utilize the younger siblings of children with autism as participants, so the ASD diagnosis has not yet been made in these children. These siblings are at 20-fold higher risk for

autism than infants in the general population and thus are referred to as high-risk (HR) infants [66]. Assessing and following these children to determine their early presentations and the behaviors associated with development of ASD, can avoid the problem of effects over time as well as biased recall in both parent and child behaviors [67], and thus aid in understanding direction of effects.

4.1. Parental sensitivity, responsiveness, and directiveness

Parents of children with ASD tend to be more directive and less sensitive in their interactions and this is also observed with the ASD children's high-risk (HR) siblings, possibly due to the parents' greater stress and/or feeling less effective as parents [60,65,68,69] or because of parental manifestation of the broad autism phenotype described below. Two aspects of parent-child interactions in a study of HR infants independently predicted an ASD diagnosis at age 3 years: 1) the parents of the HR infants were less sensitive, and 2) the children were less active and attentive to parental affect compared with low-risk dyads and those dyads in which the child did not go on to have an ASD diagnosis [69]. These findings suggest a complex transactional component in the development of the children with ASD.

In general, maternal responsiveness is associated with more social smiling for all infants [65]. A study of HR infants showed impaired social smiling compared with TD infants regardless of whether they went on to be diagnosed with ASD [70]. The mothers of the HR 9-month-olds were more directive, that is, more skill focused and instructive, but not less responsive than mothers of LR infants. When the mothers were more directive, there was a decline in social smiling 9 months later. However, when controlling for maternal directiveness, the HR-infants had *greater growth* in social smiling than LR-infants. As with the study of cortisol response [44], the authors understood this finding to mean that there is maintenance of more infantile social behavior in HR infants (versus development of complex social behaviors of gestures and language), a phenomenon that may be masked by maternal behavior.

5. The broad autism phenotype

In addition to the idea that parental stress and the social impairments of children with ASD lead to a more directive, less sensitive parenting style, it is also possible that parental characteristics lead to a distinct parenting style, especially in families with more than one child with ASD. A number of studies have shown that a sub-set of parents and siblings of children with ASD show a broad autism phenotype (BAP), that is, a sub-clinical phenotype of ASD. The BAP or "aloof personality," is consistent with Kanner's original description of the parents and Rimland's suggestion that parents of children with autism might have a mild form of the disorder [14,71].

Losh, Childress, Lam, and Piven (2009) defined the key features of the BAP by comparing parents who had two or more children with ASD with parents with a single child with ASD, and those with children with Down Syndrome [72,73]. The parents in the multiple-incidence ASD group were observed to be more aloof and to have fewer friendships, to have pragmatic language impairments, and to be more sensitive to criticism and more anxious compared with the parents in the other two groups [74]. The children with parents with BAP were more severely affected with ASD symptoms than children without an affected parent, but there was no difference in severity for children with one versus two affected parents [75].

In a small study using self-report, parents of children with ASD differed from control parents in describing their impairments in the communication and social skills domains of the Autism Quotient (AQ) self-report measure [58]. In a larger sample follow-up study, parents of children with ASD reported greater impairments all AQ subscales: communication, social skills, attention switching, imagination, and attention to detail [76,77]. A small sample of French parents of children with ASD also reported differences in the social domain of the French version of the AQ [78]. Japanese mothers of children with ASD who reported difficulty with communication and attention switching on the AQ had children with greater social impairment as assessed with the Social Responsiveness Scale [79].

Neuropsychological testing shows that parents, and especially fathers of children with ASD, have impaired executive function skills in the area of attention shifting and planning skills [80]. With respect to executive functioning, parents of children with ASD compared with control parents were found to have a "generativity deficit" as demonstrated on a Patterns Meaning task that requires the participant to create novel responses [81]. The fathers of the children in that sample also had difficulty with shifting sets whereas their mothers did not. Parents of children with ASD have been found to process facial stimuli differently, with more emphasis on the mouth than on the eyes in identifying faces as happy or fearful [82]. The parents of ASD children do not appear to differ in inhibitory control [82] or in assessments of thought disorder [21].

These studies of parents suggest that many infants at genetic risk are also exposed to parents who have deficits in some of the same domains as the children, e.g., generating novel responses and shifting sets, potentially further impacting parent-child interactions and development transactionally from a very early age [68]. However, there is very little overlap in the studies cited above that examine the behavior of parents and their children with or at-risk for ASD, and those studies regarding the BAP [69]. For example, we do not know if the parental directiveness common in interactions with children with ASD is related to parental BAP because these studies either did not assess for the BAP or did not include it in the analyses. In these families with parental BAP and/or HR siblings, there is the strong probability of gene-environment interaction. For example, parents with BAP increase the genetic predisposition for their children to have ASD, and they are more likely to have problems with sensitive attention and responsiveness to children's cues, which in ASD may be more difficult to interpret than in TD children. Both of these factors may be associated with more impaired functioning in the child.

6. Treatment as a means of understanding mechanisms

Bettelheim and others, including Margaret Mahler, advocated the use of psychotherapy for both the children and their parents, although not as a dyad or triad [15,71,83]. Indeed, as noted above, Bettelheim [3] advocated a 'parentectomy' or prolonged separation of the child from the family. In 1971, Eric Schopler and Robert Reichler wrote, "[These] children have been exposed to a remarkable array of therapies in the last three decades [the 1940s, 50s and 60s], including custodial isolation, electro-convulsive shock, drug therapies, psychoanalytic therapy, operant conditioning, electronic typewriters, and megadose vitamin therapy" [2](p. 87). Indeed, ASD has been described as a "fertile ground for quackery" [9] because 1) the children themselves usually appear physically normal, setting up the idea that there is a hidden, typically developing child within, 2) the parents are highly motivated to take action and find a cure for the pervasive and impairing condition, and 3) there is such extreme variability of treatment response in individuals with ASD that it can be difficult to identify effective treatments. Despite the many non-effective interventions, parents have been incorporated as therapists from the earliest days of the behavioral therapies advocated by Rimland [84] and Lovaas [85].

6.1. Psychotherapeutic options that involve parents

As noted above, many studies of ASD and parenting are cross-sectional, making direction of effects impossible to establish. Intervention studies provide one means of assessing the direction of effects including parent-driven effects [86,87]. Currently, evidence-based interventions for ASD are aimed at 1) improving the quality of reciprocal social

interactions, which include those addressing parental sensitivity, responsiveness, and emotion regulation strategies, 2) enhancing language and communication skills, that include foundation skills of imitation, affective sharing, and social motivation, 3) naturalistic applications of applied behavior analysis, 4) social skills and competence training, and 5) cognitive behavioral interventions [88]. The first three of these intervention goals incorporate parents as therapists.

Starting from a very young age it is possible to enhance the development of infants with symptoms of autism or those at risk for ASD. Five key ingredients have been identified in successful intervention programs for infants with autism symptoms [89,90]. Two of these ingredients specifically involve parents. The first is parent coaching which includes a therapist modeling an intervention and then the parent practicing the strategies on a daily basis. The second are efforts to increase parental sensitivity and responsiveness to the infant.

Over the past three decades, intervention research in ASD has increasingly incorporated findings from the large body of research in infant and child development, especially incorporating findings about the development of communication and social learning in TD infants and toddlers. This intervention work has focused on using fundamentals of language and social development such as joint attention, affective engagement, and imitation. Collectively, these intervention approaches are known as Naturalistic Developmental Behavioral Interventions (NDBI) [91]. They are a progression from Applied Behavioral Analysis (ABA) that used operant learning approaches to both teach skills and reduce difficult and 'interfering' behaviors, and NDBIs use the principles of ABA [85, Strain, 1979 #142, 92]. They draw on concepts of engaging and rewarding the child to be an active learner rather than passive recipient, as well as using scaffolding techniques that target skill just above the existing skill set or developmental level [91]. Importantly, the intervention techniques are applied in natural settings, such as at meals and at play, so they are useful and comfortable for parents as well as being ecologically valid. They follow the child's lead and choice, incorporating toys and activities that the child likes.

One example of an NDBI is the Early Start Denver Model (ESDM) for toddlers and preschool children with ASD. ESDM is a well-investigated therapeutic approach that involves both therapists and parental involvement and support [93,94]. Parents are trained by a therapist to use ABA teaching strategies that are sensitive and responsive to the child's cues, and focused on communication and shared engagement. The parents are instructed to use these strategies in daily life, e.g., in play, in the bath, and at mealtimes. The child also has a therapist multiple hours a week. In a study of toddlers who received the ESDM compared with those who were assessed and monitored, the ESDM group had significantly enhanced IQ scores. In addition, they had greater improvement in communication skills on Mullen Scales of Early Learning, in adaptive behavior, and in ADOS severity score two years later [93].

Infant Start began as a pilot program based on these concepts [89] but aimed at extending the ESDM strategies to HR infants. The program entails weekly sessions for parents and their infants, 6 to 11 months. The focus is placed on enhancing parental sensitivity and responsiveness. For example, a parent is trained to follow the child's lead in finding objects of interest and then to share in the emotion regarding that object. Furthermore, parents are taught to engage their children in early social games that optimize attention and positive emotion to promote acquisition of social-communication skills. In another intervention designed to address relationship development, it was found that autism severity as assessed with the ADOS was associated with lower quality of interactions at baseline [95]. However, improvement in severity after one year was predicted by initial interaction quality. The emphasis on parental responsiveness in programs such as *Infant Start* establishes a parent-child dynamic that can have benefits later in development.

6.1.1. What works?

How these NDBI programs targeting parental responsvieness are effective is not entirely clear [96]. There are limited research clinical trials (RCT) of parent-mediated treatment of ASD and understanding of efficacy, effectiveness, and mechanisms is generally lacking [97]. Aldred and colleagues (2012) examined both change in ADOS score and social communication improvements as the outcome measures in a study of 3–5 year old children with ASD. They reported that change in parental synchrony, that is, an increase in parents' contingent responses to the children's activities and attention, predicted more social communication and lower ADOS scores over a 12-month intervention.

The JASPER intervention is a play based therapy that is used to improve "joint engagement" between parent and child with ASD. It strongly focuses on teaching the parent to follow the child's lead in play and incorporates four components in that approach. These components are 1) toy selection and environment, 2) mirrored pacing, 3) prompting, and 4) communication expansion. Gulsrud and colleagues [98] investigated which of these components was most predictive in child outcome. Interestingly, given children with ASD's interest in objects, they found that mirrored pacing, i.e., imitating the way the child played with the selected toys, was the most important element in supporting children's joint attention.

Despite these and other promising interventions that involve parents and their young children, a variety of factors such as sample size, general lack of RCTs, and the heterogeneity of the population make it difficult to draw conclusions [97]. Not all outcomes are equally affected – some studies suggest that parent-child interaction and language development are more positively impacted than are core symptoms of autism [99,100]. However, a meta-analysis of 19 parent-mediated RCTs described outcomes in four areas of child impairment: ASD severity, socialization, communication-language, and cognition [101]. Only small improvements in symptoms in these domains were observed as was true for an earlier similar meta-analysis [97]. Parents differed only slightly from clinicians in their reports of gains.

The variability of the outcome measures and length of time to outcome assessed in these studies also make it difficult to assess the impact of parenting interventions. For example, the Preschool Autism Communication Trial (PACT) was designed to help parents adapt their communication style and increase sensitivity and responsiveness to their children [100,102,103]. There is an emphasis on synchrony and shared attention as well as developmentally appropriate language. However, no group treatment effect was detected in autism severity in the PACT study early on, and the authors were doubtful of its benefits as an intervention [100]. Yet, follow-up of the participants approximately 6 years later revealed that the PACT group displayed less severe ASD symptoms [102]. Thus the impact of interventions may be slow to manifest in children in whom flexible responsiveness is impaired, and yet may be quite valuable if efforts are sustained over years [104].

6.2. Emotion regulation as a target of treatment

Regarding the key role played by ER in maladaptive functioning of ASD individuals, all of these therapies require psycho-education for the parents. Mazefsky writes: "If it is determined that poor ER is an explanatory factor for the clinical presentation, psycho-education for the patient and family on ER and adaptive and maladaptive strategies can be useful. In some cases, caregivers may not realize their reliance on a maladaptive strategy and that these strategies are modeled for the patient. Psycho-education about ASD and ER can promote a validating environment and acceptance of the individual's difficulties … as well as recognition of the individual's strengths" [51] (p. 9).

Mindfulness represents a popular and research-supported approach to stress reduction. While being present-minded is emphasized, individuals are encouraged to also consider long-term goals and values in relation to situations they are facing. The principles inherent in this approach have great potential to help parents and individuals with ASD with their emotion dysregulation, in honing skills for getting "unstuck" [105] from negative emotion, and transitioning to a state of mind in which they are motivated to generate solutions to problems. [106].

Thus, the parents' desire to cultivate closer relationships with their children may supersede the power of negative affect at a given moment.

7. How do the parents' adaptation to the ASD diagnosis and their own abilities impact outcomes?

While the core symptoms of ASD, as well as related ER difficulties, have a strong basis in neurobiology, the impact that parents have on the course and outcome of individuals with ASD is demonstrable if somewhat challenging to untangle. In parent-mediated interventions, parents learn to deliver services directly and are co-therapists facilitating generalization of skills. While service providers often take the role of providing general emotional support to caretakers, providing guidance to advance parents' own coping skills is not always at the forefront of evidence-based intervention [107]. Parents are not all equally skilled, motivated, or capable of delivering the interventions with the needed intensity. Family processes and cultural/community factors may impede parents' ability to apply the interventions. Parents may not fully accept or understand the disorder; they may be distressed and have multiple competing demands on their resources, e.g., other children, jobs, marital issues. They may also share some of their children's symptoms, i.e., the BAP.

Parents of children with ASD tend to report lower parental self-efficacy or belief in their ability to effectively parent their child [6]. They also tend to have a decreased sense of agency, that is, they assume they can play a less active role in child's development than do parents of TD children. These cognitive implications are accompanied by emotional distress, including guilt, depressive symptoms, and feelings of helplessness.

Increased parental stress appears after an ASD diagnosis is made regardless of how high functioning intellectually the individual with ASD is or how capable the parent is [16,108]. A number of factors are associated with increased parental stress: co-occurring ADHD and/or anxiety which further confound the parent's ability to be sensitively responsive, low adaptive functioning, social-relatedness, and sleep issues, as well as the BAP in parents [20,109,110]. Parental stress has been associated with diminished positive outcomes for behavioral interventions [111], and decreased parental responsiveness has been associated with language delays and joint attention [112]. Certain coping styles have been associated with increased negative affect in parents of children with ASD: blaming, worrying, withdrawal coping, and helplessness [113]. Interestingly, problem-focused coping, or solving problems in a concrete, organized manner, has also been implicated in greater parental negative emotions [113]. The authors posited that the problems faced by parents of children with ASD are better approached in a flexible manner rather than with concrete ideas that may lead to parental frustration.

An example of an NDBI that evaluated parental sense of efficacy is the Focused Playtime Intervention (FPI) study. FPI is an attachment and joint attention-focused treatment for parents and their toddlers and preschool children with ASD [114,115]. It developed out of the research showing the benefits of improved sensitivity and synchronization in parent-child dyads, thus fitting the NDBI model. The mothers who were classified as insightful, e.g., understanding of the child's motives and feelings, early on became more responsive with the intervention. The children in the FPI who had low language abilities at baseline showed marked gains in expressive language with the intervention. They also showed an increase in attachment behaviors and joint attention, both reported by the parents and observed. The children who did not receive the intervention became more avoidant over the course of the study duration. The FPI enhanced the mothers' sense of efficacy as parents as they saw the increase in responsiveness of their children. The goal for the intervention is not solely in the short run, but aims to lay the foundation for a long-term positive relationship between parent and child.

8. Conclusion

ASD is a neurodevelopmental disorder with complex genetic components that are influenced by the environment. Deficits in social-communication skills and a restricted, repetitive repertoire of behaviors emerge early in a child's development. While parents do not cause these difficulties, impairments in social-relatedness can strain parent-child relationships and interactions and parental stress can have transactional effects that impede development. Parents with the BAP could have trouble with parenting because of their own characteristics. Furthermore, children with ASD are prone to co-occurring diagnoses of ADHD, anxiety, specific phobias, and depression as well as medical problems [116,117] that can further strain parent-child relations and impact emotional well-being and goal achievement. Despite these many challenges, as with most parents, those with children with ASD want to feel effective and put tremendous energy into being a source of support and growth potential in their children.

Evidence suggests that as with TD children, parental sensitivity and synchronization do enhance development in a number of domains of development. Parents play a major role in many interventions, from carrying out interventions that enhance parents' responsiveness to infant cues and following a child's lead, to serving as coaches and co-therapists. Evidence suggests that these interventions take time and may have long-range positive effects that can be difficult to capture in research studies.

Studies conducted around the world, e.g., in the United States, the United Kingdom, Scandinavian countries, France, Israel, the Netherlands, Japan and China, provide insights into the relations between parenting behaviors and child development in children with ASD. These insights have provided and will continue to provide direction for treatments that involve parents and families. In particular, parent and family characteristics that may negatively impact treatment efficacy, including the BAP found in many families, need to be identified and factored into interventions.

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