Developmental Regression in Autism Spectrum Disorders and the Broad Autism Phenotype

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by

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Abstract

This study examined the relationship between the Broad Autism Phenotype (BAP) and developmental regression in children with Autism Spectrum Disorders. A sample of 2757 children and adolescents with autism spectrum disorders was drawn from the Simons Simplex Collection, an archival database administered by the Simons Foundation Autism Research Initiative (SFARI). The Autism Diagnostic Interview – Revised (ADI-R) was utilized to capture regression status, while the Broad Autism Phenotype Questionnaire (BAPQ) collected information pertaining to parental personality attributes (i.e., aloofness, rigidity, and use of pragmatic language) associated with the BAP. Analysis of Variance demonstrated that parents of children experiencing more significant regressions have a tendency to self-report lower BAPQ ratings than parents of children experiencing either no regression or possible regression. These effects were partially attributable to fathers of other loss children, who may have exhibited a selective blind spot in evaluating their own skill deficiencies. While fathers with BAP traits appear capable of identifying BAP traits in their children, they may lack the insight to recognize these same traits within themselves. Thus, self-reports of father's BAP may require supplementation with informant information or professional clinical evaluation.

Keywords: Autism Spectrum Disorder, Broad Autism Phenotype, developmental regression, Simons Simplex Collection, Broad Autism Phenotype Questionnaire

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APPROVAL OF THE DISSERTATION

This dissertation, "Developmental Regression in Autism Spectrum Disorders and the Broad Autism Phenotype," has been approved by the Graduate Faculty of the Curry School of Education in partial fulfillment of the requirements for the degree of Doctor of Philosophy.

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DEDICATION

This work is dedicated to my wife, Sara.

Thank you for your unwavering support in the face of countless hurdles and for your perpetual belief in my ability to find the finish line.

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This dissertation would never have been written without the support and encouragement of my research advisor and mentor, Dr. Ronald Reeve. I'll never forget the way you welcomed my arrival and guided me throughout this remarkable journey. Every word in this document and every professional achievement thereafter grows from a seed you planted, encouraged, and nourished. My gratitude for all you've done is boundless.

To my dissertation committee, Julia Blodgett, Tim Konold, and Antoinette Thomas, thank you for your helpful feedback and valuable recommendations.

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To my boys, Daniel and Connor, thank you for maintaining my focus on what's truly important in life. The story that will be your life pivots on how you respond not only to adversity, but also to opportunity. Know that I'm here to listen, comfort and encourage no matter how old you are.

Lastly, I would like to thank my mother, Judy Haxter, who above all people, would have wanted to read this document and be there to celebrate the end of this arduous journey. I miss you every day.

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Introduction

Autism Spectrum Disorder (ASD) is a complex neurodevelopmental condition characterized by deficits in social communication and social interaction, as well as the presence of restricted and repetitive behavior and interests (APA, 2013). The significant majority (70%) of children with an ASD have at least one other comorbidly occurring mental health condition, while 40% meet diagnostic criteria for two or more mental health conditions (APA, 2013). Statistics compiled by the Centers for Disease Control and Prevention (CDC, 2014) indicate an overall prevalence rate of 1:68, with boys (1:42) being impacted significantly more often than girls (1:189).

The literature (Kalb, Law, Landa, & Law, 2010; Rogers & DiLalla, 1990; Siperstein & Volkmar, 2004) recognizes three distinct onset patterns for ASD. One third (Volkmar, Stier, & Cohen, 1985) to approximately one half (Short & Schopler, 1988) of all children diagnosed with an ASD exhibit a "congenital" onset in which the symptoms of the condition emerge early in life and become increasingly pronounced and identifiable as the child ages (Rogers, 2004). This onset pattern is consistent with that described in Kanner's (1943) seminal publication on "infantile autism." A second onset pattern involves children developing typically until around two years of age, only to then exhibit a plateauing of skills that decelerates advancement along the anticipated developmental trajectory (Kalb et al., 2010). A final set of children exhibit a regressive onset of symptoms in which caregivers report a clear loss of previously acquired skills (Rogers, 2004). It is this final subset of children that will be the focus of this paper.

In the significant majority of cases, a regressive onset of ASD includes the loss of previously utilized linguistic skills (Rogers, 2004; Rogers & DiLalla, 1990). Given the

mean age of onset for most regression events, the majority of regressed children have ordinarily progressed beyond babbling and proto-word production (Stefanatos, 2008). Therefore, the typical language loss refers to single word loss, as relatively few impacted children progress to the use of multi-word utterances prior to the documented regression (Luyster et al., 2005). Often, language regression is accompanied by loss of early social interaction skills, inclusive of imitation, reciprocal smiling, eye gaze, name orientation, joint attention, social engagement, and initiation (Bernabei, Cerquiglini, Cortesi, & D'Ardia, 2007; Davidovitch, Glick, Holtzman, Tirosh, & Safir, 2000; Goldberg, Osann, & Filipek, 2003; Kurita, 1985; Ozonoff, Williams, & Landa, 2005). During the regression event, parents may report the emergence of stereotypical, repetitive behaviors (Stefanatos, 2008).

While the Diagnostic and Statistical Manual for Mental Disorders – Fifth Edition does not include formal diagnostic criteria for the diagnosis of developmental regression in individuals with an ASD (APA, 2013), the phenomenon was initially identified over 50 years ago (Eisenberg, 1956; Wolff & Chess, 1964). Two decades later, "setback phenomenon" and "late onset autism" initiated a period of more intensive study (Hoshino et al., 1987; Kobayashi & Murata, 1998; Volkmar & Cohen, 1989) that continues to the present day. The earliest efforts to incorporate autistic regression into the diagnostic criteria for ASD began in the early 1960's (Creak, 1961). Recently, a number of researchers (Barger, Campbell, & McDonough, 2013; Meng-Chuan, Lombardo, Chakrabarti, & Baron-Cohen, 2012) voiced support for the addition of a regression specifier for ASD in the DSM-5; however, it was ultimately not included due to concerns

related to the clinician's ability to accurately utilize the specifier with a reasonable level of diagnostic certainty (First, 2008).

An increasing body of research indicates regression status is associated with more negative outcomes, including significantly lower cognitive and adaptive abilities (Bernabei et al., 2007; Goin-Kochel, Esler, Kanne, & Hus, 2014; Wiggins, Rice, & Baio, 2009). While a number of studies have broadly examined causal factors for ASD, including genetic (Abrahams & Geschwind, 2008; Hallmayer et al., 2011) and pre/perinatal (Froehlich-Santino et al., 2014) influences, a paucity of research has been conducted to specifically examine parent characteristics that may be more highly associated with a regressive onset of ASD. Using an existing, large database of probands with ASD diagnoses, the current study seeks to investigate this question. While "proband" refers to the initial, focal subject (i.e., the child diagnosed with ASD) in a research study, use of the term indicates that data was also collected from other sources (i.e., parents and siblings).

Epidemiology, Onset, and Course

The frequency at which regression is reported in children with an ASD is considerably higher by comparison to other neurodevelopmental disorders, as regression is a relatively rare phenomenon otherwise observed in only a few seizure (e.g., Landau-Kleffner Syndrome) and genetic (e.g., Rett Syndrome) disorders (Williams, Brignell, Prior, Bartak, & Roberts, 2015). A large, meta-analytic study using highly structured and stringent inclusion criteria (Barger et al., 2013) that ultimately included research participants from 85 studies produced a prevalence rate for autistic regression of 32%. These results are highly consistent with those (33%) found by Goldberg et al. (2003)

during preliminary research on the Regression Supplement Form (RSF) for the Autism Diagnostic Interview – Revised (ADI-R). However, reported prevalence rates of autistic regression have varied from 15% to 48% (Stefanatos, 2008). Rogers (2004) succinctly states regression is observed in the minority of impacted children, with higher prevalence rates observed in studies utilizing small sample sizes or in studies that utilize clinic referral populations. Additionally, studies that utilize parent report questionnaires instead of structured interviews have a tendency to report higher prevalence rates of regression (Barger et al., 2013; Goin-Kochel et al., 2014).

The fact that prevalence rates for regression have varied so considerably is in large part because the field lacks a clear and consistent definition for the phenomenon (Barger et al., 2013; Ozonoff, Heung, Byrd, Hansen, & Hertz-Picciotto, 2008). While it is generally acknowledged that regression includes a loss of verbal skills, one aspect that creates categorical divergence is the inclusion (or non-inclusion) of prior or simultaneous "other" losses when recording regression. As an illustrative example, Goldberg et al. (2003), during preliminary study of the RSF, identified a total of five regression types: 1) Exclusive language loss, 2) Exclusive other (non-language) skill loss, 3) Simultaneous language and other loss, 4) Language loss followed by other skill loss, and 5) Other skill loss followed by language loss. Other skill losses are often categorized as a loss of some form of social skill (e.g., eye gaze) or social interest (e.g., joint attention) (Bernabei et al., 2007; Ozonoff et al., 2005).

Additionally, it is unclear as to how long a child should exhibit mastery of a given skill before a subsequent loss in order for the loss event to be considered regression. For clinicians using the ADI-R, a child is expected to have exhibited a skill for at least three

months prior to the observed loss in order for the event to be coded as a skill regression. However, the ADI-R RSF lowers the threshold for skill mastery from three months to a single month, resulting in the identification of significantly more regressive events. Recent studies (Goin-Kochel et al., 2014; Haxter, Hall, & Reeve, 2015) have indicated that while the intellectual and adaptive functioning of these children is not as impaired as those of children who exhibited a more pronounced regression, they are in fact more impaired than are children who displayed no form of regression.

Another of the more complex aspects of autistic regression involves the disentanglement of "age of onset" from "age of recognition" (Volkmar et al., 1985), as age of onset is inextricably linked to the parent's ability to recognize symptomology (Goldberg et al., 2003). Furthermore, as articulated by Stefanatos (2008), "in the absence of a full and clear appreciation of the child's developmental history, it is conceivable that, in some circumstances, a *regression* may be difficult to differentiate from a failure of developmental *progression"* (p. 308). The results of a recent meta-analytic study (Barger et al., 2013) produced a mean age of onset of 21.35 months, which is consistent with the previously reported range of 18-24 months (Davidovitch et al., 2000; Fombonne & Chakrabarti, 2001; Tuchman & Rapin, 1997). While Goldberg et al. (2003) concluded that children with non-language regression demonstrated an earlier age of onset (18.5 months) than those that exhibited language regression (21 months), other researchers (Barger et al., 2013) have found no differences with regard to the age of onset between regression types. However, an earlier onset of regression does not appear to be associated with more negative outcomes (Haxter et al., 2015).

Home video comparison studies indicate children who later exhibit autistic regression initially demonstrated more advanced linguistic (Werner, Dawson, Munson, & Osterling, 2005) and social (Maestro et al, 2005) skills than their non-regressed ASD peers. A study using the DSM-IV diagnostic conceptualization of ASD indicated regression is more commonly observed in individuals with autism (24%) than in children with either Asperger's disorder or Pervasive Developmental Disorder – Not Otherwise Specified (8%)(Fombonne et al., 2004). The significant majority (65%) of regressions are reported to unfold gradually over several months, whereas the remainder of cases are revealed abruptly over a period of days or a few weeks (Ozonoff et al., 2005).

Etiology

After examining the existing pool of empirical study regarding the cause of autism, two conclusions are firm: 1) autism has a strong genetic basis (Abrahams & Geschwind, 2008), and 2) is caused by a multitude of factors (Happé, Ronald, & Plomin, 2006). In their seminal investigation, Folstein and Rutter (1977) found significantly higher ASD concordance rates in monozygotic twins compared to dizygotic twins. Observing that the consideration of subclinical symptoms resulted in marked increases in concordance rates, the authors opined not only the heritability of ASD, but also subthreshold symptomology. These findings were subsequently replicated, most convincingly using a sample of 3,400 twins (Ronald, Happé, Price, Baron-Cohen, & Plomin, 2006). This milder, subclinical expression of symptoms consistent with ASD in undiagnosed siblings, parents, and relatives (Piven, 2001) is currently referred to as the Broader Autism Phenotype (BAP).

The genetic and neurocognitive underpinnings of the BAP are becoming increasingly clear. A multitude of studies have confirmed the disproportionate presence of the BAP in parents of children with an ASD (Piven et al, 1994; Piven, Palmer, Jacobi, Childress, & Arndt, 1997; Sasson et al., 2013a), while BAP characteristics are more prevalent in multiplex families (Bernier, Gerdts, Munson, Dawson, & Estes, 2012; Gerdts, Bernier, Dawson, & Estes, 2013). However, ASD severity does not differ relative to the number of parents (i.e., one or both) manifesting clinically significant BAP characteristics (Sasson, Lam, Parlier, Daniels, & Piven, 2013c). Neuroscientific advances have also permitted study of the BAP in parents and siblings of impacted children. Using fMRI with siblings of children with an ASD, Bullmore et al. (2011) found reduced activation in the superior temporal sulcus and fusiform face area in response to facial expressions, while Baron-Cohen et al. (2006) discovered reduced visual attention capacity in the parents of children with an ASD. An EEG study by Dawson, Webb, and McPartland (2005) concluded that ASD family members are less persistent in their evaluation of facial stimuli, while structural MRI research (Dalton, Nacewicz, Alexander, & Davidson, 2007) suggests decreased amygdala volume in ASD siblings. Indeed, evidence collected from a variety of neuroscientific methodologies continues to confirm the heritable nature of the BAP.

Parent Relationships

While the impact of a typically developing child's behavior and disposition on the nature and quality of their interactions with caregivers is well documented (De Mol & Buysse, 2008; Sameroff, 2009), the manner in which ASD impacts the transactional nature of the parent-child dyad is less clear. Campbell, Leezenbaum, Mahoney, Day, and

Schmidt (2015) found that infants, subsequently diagnosed with an ASD, exhibited fewer reciprocal social interactions with parents during unstructured play. During play, parents of children with ASD have also demonstrated less reciprocal smiling (Dawson, Hill, Spencer, Galpert, & Watson, 1990), a reduced capacity for shared emotional experiences and co-regulation (Larkin, Guerin, Hobson, & Gutstein, 2015), an increased focus on management of the physical environment and child containment (Kasari, Sigman, Mundy, & Yirmiya, 1998), and a more directive interaction style (Wan et al., 2015).

The experience of parenting a child with an ASD is associated with deleterious effects on parental stress and mental health. By comparison to mothers of typically developing and developmentally delayed children, mothers of children with an ASD report higher levels of depressive symptoms and overall stress (Baker-Ericzén, Brookman-Frazee, & Stahmer, 2005; Estes et al., 2009; Pisula & Kossakowska, 2010). By comparison to parents of both typically developing children and to those with other developmental disabilities, ASD parents are also more likely to express higher levels of anxiety, to demonstrate behavioral rigidity, to exhibit social aloofness, and to report fewer quality social relationships (Losh & Piven, 2007; Murphy et al, 2000; Piven, Palmer, Landa, Santangelo & Jacobi, 1997). ASD mothers experiencing depression, mediated by their own interpersonal experiences, are less likely to follow through with treatment recommendations for their children or to participate in training aimed to improve outcomes for their children (Hutchings, Bywater, Williams, Lane, & Whitaker, 2012).

However, little is known about the impact the presence of the BAP may have on these parenting interactions. Sasson, Nowlin, and Pinkham (2013b) have identified

deficits in facial identification, theory of mind, and facial recognition amongst individuals with BAP deficits, while Losh and Piven (2007) have associated emotional recognition impairments with the BAP. While deficits of this kind may have negative consequences, other research indicates positive effects. For instance, while the children of high BAP parents develop an understanding of fewer words than children of low BAP parents, (Parr, Gray, Wigham, McConachie, & Le Couteur, 2015), rigidity amongst high BAP parents may contribute to increased adherence to treatment recommendations (Parr, Wittemeyer, & Le Couteur, 2011).

Purpose of the Study

The author is aware of a single study that has directly examined the relationship between the BAP and a regressive onset of autism. Lainhart et al. (2002) evaluated the presence of BAP characteristics using three instruments; the Modified Personality Assessment Schedule – Revised, the Pragmatic Rating Scale, and the Friendship Interview. Using these instruments with a small sample (n = 47), the results of this study indicated there was not a meaningful difference in the BAP among parents who did and did not have a child with a regressive onset of autism.

The present study examined the relationship between regression status and BAP characteristics utilizing a large sample of probands with an ASD. Use of the ADI-R to identify regressed individuals not only allows for the identification of autistic regression using an empirically validated assessment instrument (Gray, Tonge, & Sweeney, 2008; Lecavalier et al., 2006), but also permits impacted individuals to be identified in accordance with the type of skill reportedly lost. The simultaneous use of the RSF allows for more subtle regression events to be documented and considered. Parent BAP

information was collected using the Broad Autism Phenotype Questionnaire (BAPQ), which was designed to measure personality and language characteristics believed to be inherent to the BAP in non-ASD parents of individuals diagnosed with an ASD (Hurley, Losh, Parlier, Reznick, & Piven, 2007). Finally, the present study utilizes the Simons Simplex Collection (SSC), a database consisting of approximately 2800 children diagnosed with an autism spectrum disorder. Use of a larger database that encompasses a more representative range of cultural and ethnic backgrounds constructs a more characteristic sample while minimizing the potential impact that outliers have on the determination of statistical significance. The present study aims to answer the following questions:

1. Do the overall BAP ratings of parents with a regressed child differ significantly from the BAP ratings of parents with a non-regressed child?
2. Are there meaningful differences between the parents of regressed children and non-regressed children with regard to their BAP subscale (aloof, rigid, pragmatic language) ratings?
3. Do the BAP characteristics of mothers and fathers of regressed children with an ASD differ significantly?
4. Among parents of children who have experienced regression, are higher BAP scores associated with more characteristically severe forms of regression?
5. Are there meaningful differences between the parents of regressed children in accordance with the type (word loss, other loss, and possible loss) of reported regression?

Methods

Participants

Research participants for the study were drawn from the Simons Simplex Collection (SSC), an archival database of children administered by the Simons Foundation Autism Research Initiative (SFARI). The SSC sample was drawn from a large, multisite network of 12 university clinics in diverse geographical locations throughout the United States (SSC; http://sfari.org/resources/simons-simplex-collection). The term "simplex" indicates the impacted child has no immediate relatives with either a confirmed or suspected ASD diagnosis. After an initial screening procedure was completed to ensure research participants met basic inclusion criteria, a comprehensive battery of assessments was completed with the impacted family. Proband assessment protocol included the ADI-R and the Autism Diagnostic Observation Schedule (ADOS), as well as a variety of cognitive, adaptive, linguistic, and social-emotional assessments, including the BAPQ. Those ultimately included in the SSC were between ages 4-17 and determined to have met diagnostic criteria for an ASD (including having met established clinical thresholds on the ADOS and ADI-R), to have no primary relatives on the autism spectrum, to have a mental age above 18 months, and to be free of medically significant perinatal events (Fischbach & Lord, 2010). The SSC database contained a total of 2858 probands, with the percentage of male participants (83.4%) approximating established prevalence rates (CDC, 2014). The significant majority of the sample was Caucasian (78.5%), and the mean age of initial ADOS administration was nine years of age (9.02).

Measures

Regression status was captured using the ADI-R (Rutter, LeCouteur, & Lord, 2003) and the RSF (Goldberg et al., 2003). The 93 item ADI-R is a comprehensive, highly structured parent interview utilized to collect historical and developmental information about a child suspected of having an ASD. Regression events are initially coded on Item 11 (language regression) and Item 20 (other skill regression), with subsequent questions detailing the specific nature of the loss. Positive endorsement of Item 11 was indicative of having exhibited "language" regression, whereas positive endorsement of Item 20 was coded as having exhibited an "other" regression event. Individuals meeting criteria for both loss groups were coded as having language regression, as priority is given to the more severe form of loss. In order to code a loss event on the ADI-R, a child must have demonstrated a skill for a minimum of three months, after which the skill is lost for a period of three or more months. Non-regressed children included those that did not endorse a loss on either Item 11 or 20.

More subtle regression events were captured by the 31-item Regression Supplement Form. The RSF lowers the threshold for inclusion by counting regression events in which the child had previously shown, and subsequently lost, either skill type for a single month. Losses operationalized using this one month criteria were coded as "possible losses." Individuals in the "possible loss" category failed to meet the ADI-R criteria for a "language" or "other" loss, but demonstrated language and/or social loss of shorter duration.

BAP characteristics were measured using the Broad Autism Phenotype Questionnaire (Hurley et al., 2007), an empirically validated (Broderick, Wade, Meyer,

Hull, & Reeve, 2015; Hurley et al., 2007; Ingersoll, Hopwood, Wainer, & Donnellan, 2011) 36-item self-report form providing information along three dimensions of personality; aloofness, rigidity, and pragmatic language. Respondents were asked to answer questions using a six-point continuum similar to a Likert scale. Information may be collected about the informant (self-report) or about the child's other biological parent (informant report). The questions on the two versions of the BAPQ only differ according to the type of personal pronouns utilized. Parents participating in the SSC completed the self-report version of the BAPQ.

The psychometric properties and clinical utility of the BAPQ have been evaluated (Ingersoll et al., 2011) in comparison to two other self-report BAP measures, the Social *Responsiveness Scale – Adult Form* (SRS-A) and the *Autism Spectrum Quotient* (AQ). All three subscales of the BAPQ demonstrated adequate internal consistency (Cronbach's alpha > .70), while exploratory factor analysis extracted three factors (consistent with the BAPQ's three factor theoretical structure) explaining nearly 39% of the variance across BAPQ items. Subsequently, Sasson et al. (2013a) confirmed the robust three-factor structure of the BAPQ. Despite being the shortest of the three instruments, the BAPQ proved the only instrument, following clinical assessment, to accurately discriminate between individuals who did and did not manifest the broad autism phenotype. Thus, Ingersoll et al. (2011) concluded the BAPQ was the "best of the three measures considered."

Procedures and Statistical Analyses

All analyses were conducted using IBM SPSS Statistics (SPSS), Version 23. First, descriptive analyses (mean, median, range, standard deviation, kurtosis, skewness)

were conducted to ensure the distribution of the dataset was normal. One-way Analysis of Variance (ANOVA) was employed to investigate all research questions, with all post-hoc analyses conducted with Tukey's test, thus permitting comparison of all possible two group combinations. As only mother and father BAPQ scores were provided, an aggregate "parent" BAPQ score was created by averaging the mother and father scores. Thus, analyses were conducted on "parent," mother, and father BAPQ scores.

To investigate the first research question, ANOVA was utilized to compare the mean parent BAPQ scores of all four regression groups. Next, ANOVA was employed to examine mean differences amongst BAPQ variables (aloofness, rigidity, pragmatic language, and total score) amongst all regression groups for parents, mothers, and fathers. Subsequent analyses focused exclusively on the three regressed groups. Question 3 utilized ANOVA to compare the mean BAPQ domain scores of mothers and fathers of children exhibiting any form of regression. To answer the final research questions, ANOVA was utilized to investigate differences in mean BAPQ domain scores amongst parents, mothers, and fathers of children with different regressive presentations. Effect sizes approximating .01 were considered small, .06 medium, and .15 large (Cohen, 1988).

Results

Of the probands that reported scores on at least one BAPQ measure, a total of 101 were removed (n = 2757) because regression status information was not recorded. Information from both parents was available for the substantial majority of remaining cases, with data completely missing from four mothers and ten fathers. Thirty-nine mothers and fifty fathers were considered to have partially missing data (i.e., missing the answer to a single BAPQ question, presumably due to accidental omission). To maintain

use of these cases, the average BAPQ score was utilized in subsequent analyses instead of the total BAPQ score.

Skewness and kurtosis values were within acceptable limits (+/- 1) for all dependent variables and no statistically significant outliers were present. Although the results of the Shapiro-Wilk test revealed violations of normality across all BAPQ domains, a normal distribution within a sample composed entirely of individuals with autism is not to be expected. Levene statistics were computed to evaluate homogeneity of variance assumptions. Analysis revealed a single violation of homogeneity of variance, occurring within the father rigidity variable, F (3, 2724) = 2.76, p = .041. However, ANOVA has demonstrated to be robust to homogeneity of variance violations when the largest cell variance ratio is less than or equal to 3:1(Dean & Voss, 1999).

Overall, parental BAPQ ratings were influenced by regression status, F (3, 2753) = 9.22, p < .001. Follow up Tukey tests indicated statistically significant differences between non-regressed probands and those exhibiting word losses (p = .02) and other losses (p <.001), respectively, as well between the other loss and possible loss group (p = .01). Parents of children exhibiting more severe regressive events rate their own phenotype as less consistent with ASD then do parents of children exhibiting less severe regressive presentations. Significant overall F statistics were observed on all four BAPQ domains on the aggregate parent scale (Table 1). While mothers of word loss children produced lower overall, aloof, and rigid scores in comparison to no loss mothers, fathers of other loss children produced significantly lower ratings in all domains in comparison to fathers of no loss children.

Significant BAPQ differences were observed between mothers and fathers of regressed children on the overall BAPQ score, F (1, 967) = 132.99, $p < .001$, as well as on the Aloof F (1, 967) = 148.123, $p < .001$, Rigid F (1, 967) = 32.46, $p < .001$, and Pragmatic F (1, 967) = 81.59, $p < .001$ domains. Fathers of regressed children produced significantly higher mean BAPQ scores than mothers of regressed children across all aspects of the BAPQ. A significant overall difference was observed between the average parent BAPQ score of children exhibiting word, other, and possible losses, F (2, 973) = 5.40, $p = .005$. Specifically, the mean score of parents of other loss children ($\bar{X} = 2.36$) is significantly lower than the mean score of parents of possible loss children ($\bar{X} = 2.51$). The mean scores of mothers of regressed children were not significantly different along the Aloof, Pragmatic, or Rigid domains of the BAPQ, while the mean scores of fathers were significantly different only within the Pragmatic domain, F (2, 965) = 3.31, $p = .037$, as fathers of possible loss children ($\bar{X} = 2.33$) had higher scores than did fathers of other loss children ($\bar{X} = 2.18$).

Discussion

The present study sought to investigate the relationship between parental broad autism phenotype and a regressive presentation of ASD. The rationale for exploring this relationship was straightforward: If elevated expressions of the BAP were indeed associated with autistic regression, and consequently, more deleterious child outcomes, screening and subsequently educating high BAP parents about future risk would be advantageous. In much the same way that women of advancing age receive counseling about the increased risk for certain genetic conditions (e.g. Down syndrome), high BAP

parents, after completion of a simple checklist, could then factor any increased risk for having a regressed ASD child into their family planning decision.

This study was conducted using a large, simplex sample of probands and their parents, 35% of which reported some form of regression. The percentage of regressed children contained within the sample is highly consistent with samples utilized to complete other research studies (Goldberg et al., 2003). Regression was identified by caregiver responses to interview questions on the ADI-R and RSF, resulting in the creation of three loss categories: word loss, other loss, and possible loss. Inclusion in the first two categories required the impacted child to first exhibit, and then ultimately lose the designated skill for a period of three months, whereas possible losses also required skill onset and diminution, but for only a single month. Thus, possible losses were conceptualized as more subtle regressive events. Analyses of BAPQ responses were conducted using the individual responses of mothers and fathers, and by using an aggregate "parent" score that combines the scores of mothers and fathers, thus creating a variable that accounts for the total expression of BAP across both parents.

While results confirming that a regressive presentation is associated with differences in BAPQ presentation were anticipated, the direction of the mean score differences was unexpected. Maxwell et al. (2013) found an association between higher maternal and paternal BAP characteristics and heightened levels of ASD symptomology in offspring, while Sasson et al. (2013a) established that the absence of BAP characteristics in parents leads to less severe expressions of ASD symptoms in children. As the presence of a regression event is associated with lower adaptive and intellectual functioning (Bernabei et al., 2007; Goin-Kochel et al., 2014; Wiggins, Rice, & Baio,

2009), this researcher anticipated finding higher expressions of BAP features among parents of children experiencing more serious regression events.

However, these results indicate parents of children experiencing more significant regressions in fact have a tendency to self-report lower BAPQ ratings than parents of children experiencing either no regression or possible regression. Specifically, subsequent analyses indicate these tendencies were isolated to fathers of other loss children (across all three BAPQ domains) and mothers of word loss children (across the aloof and rigid domains). As the overall effects observed in the parent variable can be isolated to these two groups, the utility of the parent aggregate variable is limited, with the observed effects best accounted for by examination of the individual mother and father BAPQ scores.

Indeed, these effects could be partially attributable to the inclusion criteria for the SSC, which prohibit the inclusion of children with parents or immediate relatives with an ASD. Thus, parents with the highest expressions of BAP, or those meeting the criteria for clinical diagnosis, are excluded from the SSC. Moreover, BAPQ information collected for the SSC was obtained exclusively through self-report. Prior research (Carlson, 2013; Vazire & Carlson, 2010) has consistently indicated self-reporters lack self-awareness, in contrast to informant report, when self-reporting personality characteristics and pathology. Several large research studies (Maxwell et al., 2013; Sasson et al., 2013a) examining BAP characteristics have alleviated the problems inherent in the self-reporting of pathology by averaging BAPQ self-reports scores with those provided by the other parent's informant report. Sasson et al. (2014) found selective disagreement between parent and informant reports on the BAPQ when the self-reporting parent was positive

for the BAP characteristic in question. Observing such disagreement as trait specific, and that it applied only to trait positive self-reporters (not trait positive informants), Sasson et al. (2014) concluded "parents with BAP traits may be capable of identifying these traits in highly familiar others, but lack insight into recognizing these same traits in themselves (p. 736)." Subsequent analysis revealed this finding was driven largely by the "selective blind spots" of fathers. Whereas only 47% of fathers would have met the clinical cutoff for a positive overall BAP based on self-report, informant report would have driven the classification rate up to 82%.

These findings offer some explanation to account for the consistently observed tendency of fathers of other loss children to report significantly lower BAPQ ratings than parents of non-regressed children within all four BAPQ domains. As the most severe loss experienced by other loss children was a loss of social or play skills (e.g., not a word loss), this group, using the DSM-IV diagnostic system utilized at the time of data collection, is likely composed primarily of children diagnosed with Asperger's Syndrome, or in current vernacular, Social Communication Disorder. Acceptance of the premise that fathers with social impairments transmit these deficits to their offspring, it is reasonable to opine that these same fathers, given their "selective blind spot," would underreport symptoms of their own rigidity, aloofness, and pragmatic deficits.

These findings must be considered within the context of several limitations. First, this research relied on retrospective parent reporting to ascertain establish regression status. In a longitudinal study of parental reporting, Hus, Taylor and Lord (2011) found that an increasing number of children met criteria for language delay as they aged despite the fact that initial reports were not suggestive of delay. Additionally, retrospective

parent reports of developmental information have also been shown to be influenced by knowledge of autism diagnosis (Zwaigenbaum et al., 2007). Secondly, BAPQ ratings were derived exclusively from self-reports. In order to minimize bias inherent in considering only one source of information, future researchers may choose to rely on an average of informant and self-report, or to validate self-reports with direct clinical assessment. Third, the resultant effect sizes of the present study are small, and with the availability of such a large sample, the study may be overpowered. Finally, while use of the SSC affords access to a large sample, it is composed of exclusively of simplex families that are predominantly affluent and Caucasian.

Conclusion

The predominant conclusions drawn by the current study indicate: 1) parents of children experiencing more severe regression events report overall lower parental BAPQ ratings, 2) fathers of other loss children consistently self-report lower BAPQ ratings than fathers of children experiencing either no regression or other ASD onset patterns, 3) among children experiencing a regression event, fathers consistently have higher BAPQ scores than mothers, and 4) while there are no significant differences in the BAPQ scores of mothers of a child that experienced regression, fathers of other loss children produce lower BAPQ scores than do fathers of word or possible loss children. Consistent with the findings of Sasson et al. (2014), these results suggest fathers of children with social deficits may underreport their own social deficiencies, possibly due to a "selective blind spot" that restricts self-awareness and their ability to recognize these characteristics within themselves. Thus, self-reports of father's BAP may require supplementation with informant information or professional clinical evaluation.

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Table 1
Means, Standard Errors, and Significance Values for the Effects of Regression/No Regression on BAPQ Scores

BAPQ Dimensions	No Loss/Loss Group Comparisons				ANOVA	Effect Sizes
	No Loss	Word Loss	Other Loss	Possible Loss	Overall F Statistics	ω^2
Parent						
	Mean	Mean (SE)	Mean (SE)	Mean (SE)		
Overall	2.54	2.44 (.06)*	2.36 (.04)***	2.51 (.03)	9.20***	<.01
Aloof	2.86	2.77 (.05)**	2.64 (.08)**	2.82 (.05)	8.60***	<.01
Rigid	2.91	2.82 (.04)**	2.69 (.07)**	2.83 (.04)	8.23***	.01
Pragmatic	2.36	2.31 (.04)	2.18 (.06)*	2.34 (.05)	4.08*	<.01
Mother						
Overall	2.42	2.30 (.03)**	2.29 (.05)	2.37 (.03)	6.57***	.01
Aloof	2.43	2.26 (.04)***	2.27 (.07)	2.38 (.04)	6.9***	.01
Rigid	2.72	2.57 (.04)**	2.58 (.07)	2.67 (.04)	5.83**	.01
Pragmatic	2.10	2.07 (.03)	2.02 (.05)	2.07 (.03)	1.32	>.01
Father						
Overall	2.71	2.63 (.03)	2.50 (.05)**	2.66 (.03)	6.202***	.01
Aloof	2.86	2.77 (.05)	2.64 (.08)*	2.82 (.05)	3.566*	<.01
Rigid	2.91	2.82 (.04)	2.69 (.07)*	2.82 (.04)	5.031*	<.01
Pragmatic	2.36	2.31 (.04)	2.18 (.06)*	2.34 (.04)	3.948*	<.01
(*n*)	1781	427	141	408		

* $p < .05$
** $p \leq .01$
*** $p \leq .001$