An in-depth examination of optimal outcome children with a history of autism spectrum disorders

Elizabeth Kelley *, Letitia Naigles, Deborah Fein

University of Connecticut, Department of Psychology, Storrs, CT 06269-1020, USA

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ABSTRACT

Previous research has suggested that some children with autism spectrum disorders (ASD) may improve to such an extent that they lose their diagnosis, yet little research has examined these ‘optimal outcome’ children in depth. We examined multiple aspects of functioning in a group of 13 optimal outcome (OO) children, matched on age, gender, and non-verbal IQ to a group of typically developing children (N = 14) and a group of high-functioning children with ASD who still retained a diagnosis on the autism spectrum (N = 14). These children were tested on average about eight years after they had been diagnosed (OO = 93 months, HFA = 94 months). Unlike their high-functioning peers with ASD, the OO group's adaptive and problem behavior scores fell within the average range. They also showed average language and communication scores on all language measures. The HFA group, however, continued to show pragmatic, linguistic, social, and behavioral difficulties. The OO children tended to have been diagnosed at younger ages and were significantly more likely to have received intensive early intervention. Although the high-functioning children with ASD continued to show difficulties in the behavioral realm, the individuals in the OO group were functioning within the average range on all measures. Future research should address how this optimal outcome is achieved.

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1. An in-depth examination of optimal outcomes in children with a history of autism spectrum disorders

An extensive debate exists in the literature as to whether children with autism spectrum disorders (ASD) ever reach a point where they lose their diagnosis, so that they are no longer considered to be on the autism spectrum. Although outcome for children with ASD, particularly for those with an IQ in the normal range of functioning, is thought to be better than it was 30 years ago (Howlin, Goode, Hutton, & Rutter, 2004), there is much disagreement in the field as to what the general range of outcome is for these children and whether children can actually lose their diagnosis over the course of development (Eaves & Ho, 1996; Sallows & Graupner, 2005). Some authors have hypothesized that there are children with ASD who are indeed recovered, or who appear to have lost the behavioral manifestations of a genetically based, biological disorder (see Helt et al., 2008). However, others state that while these children may improve to such an extent that they are
functioning well intellectually and academically, they may continue to experience the core deficits of this disorder, such as social and communicative problems (Mundy, 1993). To date, it has been difficult to address this debate empirically, as many studies have simply used IQ or school placement as an outcome variable (Charman & Howlin, 2003; Shonkoff, Hauser-Cram, Krauss, & Upshur, 1988). As Mundy (1993) points out, integrated school placement or IQ within the normal range is not sufficient to consider a child to have moved off the autism spectrum. The current study was designed to assess a group of optimal outcome children (OO) and compare them to both their typically developing (TD) peers and high-functioning peers with ASD (HFA) on a wide range of variables in order to better delineate their profiles of functioning. We expected that the HFA group would continue to experience problems in communication, social adjustment and adaptive behavior, whereas the OO group would be functioning within the average range on these measures, but we considered it possible that some residual cognitive or behavioral difficulties would be seen in the OO group.

Optimal outcome children were those who, as in previous studies, were mainstreamed into regular classrooms and had, according to their records, a full-scale IQ within the average range (i.e. greater than 70). However, we imposed additional criteria for optimal outcome: Unlike previous studies, the optimal outcome children in the current study were no longer receiving extra help in the classroom, had lost their diagnosis according to the school system, and no longer met criteria for a diagnosis on the autism spectrum according to their ADOS-G scores. The HFA group also had a full-scale IQ within the average range, according to their records, but, unlike the OO group, retained their diagnosis in the school system and continued to meet criteria for a diagnosis on the autism spectrum according to the ADOS-G.

1.1. Previous outcome studies in children on the autism spectrum

Many of the outcome studies reported in the literature suffer from a restricted set of outcome variables, as pointed out by Charman and Howlin (2003) and Shonkoff et al. (1988). Some of these studies report that the children have been mainstreamed into regular classrooms but do not address whether the children are receiving continued support in terms of an aide or other extra help (Kasari, 2002). Since in many school systems there is a tendency to mainstream all but the most impaired children, we cannot determine with any certainty whether or not these children are truly functioning at the level of their typically developing peers (Schopler, Short, & Mesibov, 1989).

Other researchers have argued that while IQ and school placement are of interest, outcome studies need to focus more on how the children are performing communicatively, socially and adaptively because these areas are commonly the most resistant to treatment (Mundy, 1993; Tsatsanis, Foley, & Donehower, 2004). That is, there may be children who are performing fairly well in the academic realm but have difficulty with navigating the social world, effectively communicating with others, and being flexible in their behaviors. This study is only the second that we know of (see also Sallows & Graupner, 2005) to address levels of autistic symptomatology, language and communication, adaptive behavior, and problem behaviors in a group of optimal outcome children. However, unlike those studied by Sallows and Graupner, the children in the current study were tested well beyond the time they had finished treatment; thus, we investigate whether the children’s improved status has been maintained for a number of years.

The current study is not, strictly speaking, an outcome study as it did not follow a sample of children over a specific period of time. However, it is relevant to the outcome literature in that we examined two groups of children with a history of ASD whose behavioral outcomes differ despite their being matched on non-verbal intelligence at the time of the study. While most researchers seem to agree that early diagnosis and early intervention in children with ASD are critical (Bibby, Eikeseth, Martin, Mudford, & Reeves, 2002), little agreement exists as to what the best outcomes for children with ASD can be. This disagreement is complicated by the fact that for a long time most outcome studies either tended to focus on very specific treatment outcomes (e.g., learning of irregular past tense forms) over very short periods of time (see Goldstein, 2002, for a review), or very general outcomes (e.g., ability to live independently in adulthood) (Billstedt, Gillberg, & Gillberg, 2005; Szatmari, Bartolucci, Bremner, Bond, & Rich, 1989).

Only a few studies have examined a wide variety of outcome variables over the course of childhood and these studies have reported mixed results. Stevens et al. (2000) found that ASD children who were high- or low-functioning in preschool generally remained so into the school years on a wide variety of measures, with some high-functioning children showing improvement on standard scores of language and cognitive functioning while the low-functioning children tended to show a decline in these same areas. In that study, almost all of the low-functioning children stayed in the low-functioning group from preschool to school-age, while the high-functioning preschool group split into two groups, one of which scored in the low-functioning range at follow-up and the other (relatively small) had scores in the normal range on most cognitive variables. The researchers did not investigate the treatments received by the children, however, nor did they report autistic symptomatology or problem behaviors.

Sigman and Ruskin (1999) conducted a longitudinal study of a number of children with autism and found considerable variability in the trajectories of development. While overall the group means for intelligence did not change a great deal over time, many of the children showed either marked improvement or a decline in intelligence scores over the course of the study. However, the vast majority of the children in this study progressed very little in the language domain and continued to remain socially isolated and unable to relate to their peers. Interestingly, Sigman and Ruskin found that 17% of these children lost their diagnosis in late childhood or early adolescence. Finally, a few studies have examined outcome after several years of behavioral treatment. Bibby et al. (2002) followed a group of children with ASD who had received home-based behavior modification programs, and found that little change transpired in these children on a variety of
cognitive, language, and adaptive measures approximately two years after beginning treatment in preschool; gains were made in adaptive skills, though. Eaves and Ho (1996, 2004) found that behavioral and sensory symptoms often improved more than social and communicative symptoms, and IQ improved more than adaptive functioning between the ages of two and four.

In sum, the literature is mixed as to what the outcome of children on the autism spectrum is over time. One reason for the disparate findings is likely that many studies included both high- and low-functioning children. These two groups of children may exhibit different developmental trajectories, and thus the effects of change may be occluded by the large variability which results when the two groups are combined. The current study comprised only initially high-functioning children and thus a clearer picture of outcome for this particular group of children was expected.

For many years the only group of studies that had yielded a large number of optimal outcome children was that of Lovaas (1987) and McEachin, Smith, and Lovaas (1993). Although Lovaas reported that 47% of the children who participated in an intensive 40-h-a-week program of behavioral therapy improved to such a degree that they were mainstreamed into regular classrooms, his methodology has been criticized by some (Schopler et al., 1989). The current study does not directly address treatment comparisons as Lovaas (1987) and McEachin et al. (1993) did, but rather whether or not children truly can improve to such an extent that they no longer appear to be on the autism spectrum.

More recently, a handful of studies have suggested that an optimal outcome for some high-functioning children with ASD is indeed a possibility. Sallows and Graupner (2005) found that 48% of their sample reached optimal outcomes after an intensive program of behavioral therapy; that is, these children had been mainstreamed into regular classrooms and were functioning at a normal intelligence level. This study found, however, that general language ability and adaptive behavior did not increase to the same extent as IQ. Moreover, the children in this study were still relatively young and had only recently completed an intensive behavioral treatment program. The current study extends this work by examining older children who had not been receiving treatment for several years.

Kelley, Paul, Fein, and Naigles (2006) demonstrated that some children with ASD reached a level of behavioral and cognitive functioning where they no longer met criteria for any ASD and were mainstreamed into a regular classroom without any extra help, though they continued to experience some subtle social and communicative difficulties. That is, the OO children in this study experienced continuing difficulties in semantic and pragmatic areas of language, such as understanding the meaning of mental state verbs, understanding theory of mind, engaging in a narrative, and being able to induce categories properties using semantic information. It should be noted that 11 of the children in the current study were also part of the Kelley et al. study, although the current study is not a longitudinal study because very different measures were used in the different studies. Moreover, the Kelley et al. study did not measure autism symptomatology, adaptive functioning, or problem behaviors as the current study did.

We hypothesized that the children in the OO group would fall within the normal range of functioning on all measures, including those measures not used in defining them as a group, while the HFA group would continue to show difficulties with adaptive behavior, semantic and pragmatic language skills, as well as adjustment problems.

2. Method

2.1. Participants

Three groups of children were examined in this study. After several children were eliminated for the reasons given below, the final sample included 13 children with a history of ASD who had reached an optimal outcome level (the OO group), 14 typically developing children (the TD group), and 14 children on the autism spectrum who were of average intelligence (the HFA group).

The OO group was chosen a priori by the following criteria:

1. They had been mainstreamed into a regular classroom.
2. They had a full-scale IQ on their last school assessment which was greater than 70.
3. They were receiving no more than 1 h per week of service overall (e.g. 1 h of speech therapy, or 1 h of occupational therapy) and did not have an educational aide in the classroom.
4. They were considered by the school system to no longer be on the autism spectrum.
5. They had previously been diagnosed on the autism spectrum by a clinician who specialized in ASD and they met criteria on the ADI-R based on their ‘ever’ scores.
6. They no longer met criteria for an ASD diagnosis on the ADOS-G.

The HFA group was chosen a priori by the following criteria:

1. If they had been mainstreamed into a regular classroom, they continued to receive extra help. Two of the HFA children were being home-schooled, two were half-time in special needs classrooms and half-time mainstreamed with a one-on-one aide, three were mainstreamed with a one-on-one aide, and seven were mainstreamed with a shared aide.
2. They had a full-scale IQ on their last school assessment which was greater than 70.
3. They retained their diagnosis of an ASD in the school system.
4. They had previously been diagnosed on the autism spectrum by a clinician who specialized in ASD and they met criteria on the ADI-R based on their ‘ever’ scores.

5. They currently met criteria for an ASD diagnosis on the ADOS-G.

The typically developing children had no history of academic, neurological, or psychological problems as reported by their parents. Children in all of the groups were from middle- to upper-middle class families that resided in suburban and rural areas of the northeastern US.

All of the parents from our previous study with these optimal outcome children (Kelley et al., 2006) were contacted and asked to take part in the current study. Two of the optimal outcome children from the previous study were lost to attrition as the parents no longer wished to participate; however, two more were gained through the clinical files of the third author. One of the children from the Kelley et al. study was not included because, although no longer considered on the spectrum by the school system, he met the current criteria for an ASD on the ADOS-G. Five of the typically developing children were lost to attrition but five more were gained through the local school system. These testing sessions took place approximately three years after they had participated in the previous study (Kelley et al., 2006). Finally, the 14 children still on the autism spectrum were obtained through the clinical practice of the third author or were participants who had previously engaged in research with our lab, and were known to have a full-scale IQ score within the normal range.

A number of children were tested but not included in the current study: one typically developing child and one OO child, each of whom did not want to participate further after the first testing session; two OO children who were too young to complete the current protocol; one child thought to be OO, but who met criteria for an ASD diagnosis on the ADOS-G (see above); two HFA children who did not meet criteria on the ADOS-G or ADI-R yet still retained a diagnosis from the school system; and one HFA child who experienced difficulty during both testing sessions and was found to test with below average intelligence. Information on the children’s age, gender, and non-verbal IQ scores can be found in Table 1. There were no significant differences between the groups on gender, age, or non-verbal intelligence as measured by the Matrix Reasoning subtest of the Wechsler Intelligence Scale for Children-IV (Wechsler, 2003).

2.2. Materials

2.2.1. Autistic symptomatology

The Autism Diagnostic Interview-Revised (ADI-R) (Lord, Rutter, & LeCouteur, 1995) is a structured interview conducted with the parent(s) that is based in part on the DSM-IV’s and ICD-10’s diagnostic criteria for autistic disorder. In the current study all parents were instead asked to describe their child’s behavior when their child’s behavior was the most autistic, whatever age that might be. This methodological adjustment was made because many of the optimal outcome children had already improved significantly by the age of four or five. Data from one child from the OO group was not available as the parents did not want to be interviewed. Her clinical files, however, showed that she had indeed been on the autism spectrum according to the DSM-IV symptoms and clinical reports.

Data from two children in the OO group were unavailable due to experimenter error; however, these children met all of our other criteria for the OO group and did not meet the criteria for an ASD on the ADI-R current functioning diagnostic algorithm. One child in the HFA group was unwilling to complete the ADOS-G, yet he still clearly met criteria for an ASD on the ADI-R current functioning diagnostic algorithm.

<table>
<thead>
<tr>
<th>Table 1</th>
<th>Age, gender and non-verbal intelligence of the groups.</th>
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</thead>
<tbody>
<tr>
<td></td>
<td>OO group N = 13</td>
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<tr>
<td></td>
<td>HFA group N = 14</td>
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<tr>
<td></td>
<td>TD group N = 14</td>
</tr>
<tr>
<td></td>
<td>F-test</td>
</tr>
<tr>
<td>Age</td>
<td>Mean 10 years, 5 months</td>
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<tr>
<td></td>
<td>SD = 17.5 months</td>
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<tr>
<td></td>
<td>Range 99–163 months</td>
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<tr>
<td></td>
<td>Mean 11 years, 5 months</td>
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<tr>
<td></td>
<td>SD = 25.9 months</td>
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<tr>
<td></td>
<td>Range 98–170 months</td>
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<td></td>
<td>Mean 10 years, 3 months</td>
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<td></td>
<td>SD = 15.5 months</td>
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<tr>
<td></td>
<td>Range 95–154 months</td>
</tr>
<tr>
<td>Gender</td>
<td>10 boys, 3 girls</td>
</tr>
<tr>
<td></td>
<td>9 boys, 5 girls</td>
</tr>
<tr>
<td></td>
<td>11 boys, 3 girls</td>
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<tr>
<td></td>
<td>$\chi^2(2) = 0.864, n.s.$</td>
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<tr>
<td>Matrix reasoning scaled score (NVIQ)</td>
<td>Mean = 11.17</td>
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<tr>
<td></td>
<td>SD = 3.1</td>
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<tr>
<td></td>
<td>Range 5–17</td>
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<td></td>
<td>Mean = 9.85</td>
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<td></td>
<td>SD = 3.0</td>
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<td></td>
<td>Range 4–14</td>
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<tr>
<td></td>
<td>Mean = 11.21</td>
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<tr>
<td></td>
<td>SD = 2.8</td>
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<tr>
<td></td>
<td>Range 4–16</td>
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<tr>
<td>Time since diagnosis (in months)</td>
<td>Mean = 92.67</td>
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<tr>
<td></td>
<td>SD = 20.68</td>
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<tr>
<td></td>
<td>Range 57–131</td>
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<tr>
<td></td>
<td>Mean = 94.00</td>
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<tr>
<td></td>
<td>SD = 34.26</td>
</tr>
<tr>
<td></td>
<td>Range 32–140</td>
</tr>
</tbody>
</table>

2.2.2. The autism diagnostic observation schedule-generic (ADOS-G)

Lord et al. (2000) is a standardized diagnostic instrument used to assess and diagnose children on the autism spectrum. Every child was given module 3 as they were all verbally fluent. Data from two children in the OO group were unavailable due to experimenter error; however, these children met all of our other criteria for the OO group and did not meet the criteria for an ASD on the ADI-R current functioning diagnostic algorithm. One child in the HFA group was unwilling to complete the ADOS-G, yet he still clearly met criteria for an ASD on the ADI-R current functioning diagnostic algorithm.
2.2.3. Adaptive functioning

The Vineland Adaptive Behavior Scales (Sparrow, Balla, & Cicchetti, 1984) is a frequently used parent interview which addresses three major aspects of the child’s adaptive functioning: Communication, Daily Living Skills, and Socialization. Two parents in the typically developing group did not wish to complete this interview.

2.2.4. Problem behaviors

The Behavior Assessment System for Children (BASC) (Reynolds & Kamphaus, 1992) was designed to address children’s problem behaviors between the ages of 2 and 18. Four of the OO children, two of the TD children and four of the HFA children did not have complete BASC data as parents did not return the questionnaire through the mail.

2.2.5. Language and communication

To assess general language verbal understanding, the Similarities, Vocabulary, and Comprehension subscales of the Wechsler Intelligence Scale for Children-Fourth Edition (WISC; Wechsler, 2003) were used and the Verbal Composite Index (equivalent to verbal IQ) was calculated. Children were also tested on the Peabody Picture Vocabulary Test-Third Edition (PPVT; Dunn & Dunn, 1997) to assess vocabulary knowledge. To assess pragmatic language ability, the Making Inferences and Figurative Language subtests from the Test of Language Competence (TLC; Wiig & Secord, 1989) and the Interpreting Intentions standard score was calculated. To address a wider range of pragmatic language abilities, the Test of Pragmatic Language (TOPL: Phelps-Terasaki & Phelps-Gunn, 1992) was also administered. Children were also administered the Comprehensive Assessment of Spoken Language—Third Edition (CELF; Semel, Wiig, & Secord, 1995) to assess both semantic and syntactic aspects of language. All tests were administered in the standard procedure outlined in the manuals.

2.2.6. Differences between autism groups on history

To assess possible reasons for differences in trajectories between the groups, a number of questions from the ADI-R were explored. We examined whether the OO or HFA groups were more likely to be taking medication and were more likely to have a history of autism in the family. We also determined from the ADI-R the age at which the child was diagnosed. Furthermore, we coded the amount and type of intervention the children had received as follows: a zero was coded if the child had received no ABA therapy, a one was coded if the child had received an eclectic type of therapy (which may have had some ABA included), a two was coded if the child had received primarily ABA therapy but for fewer than 20 h per week, and a three was coded if the child had received more than 20 h per week of ABA therapy.

2.3. Procedure

Informed consent was obtained from all parents and written assent was obtained from all children in the study before testing began. This research study was approved by the university’s Institutional Review Board.

The children were administered the tasks during one of two sessions in which the children were given the battery tasks in random order (as well as three experimental tasks that are not reported here). In order to make the testing proceed as quickly as possible, children were videotaped and videotapes were scored after the fact. It should be noted that where missing data occurred it was never because the child scored at floor; rather, in most instances the testing sessions ran too long and children could not complete the final test or there was experimenter error or videotape malfunction. A demographic survey including information about family structure, other children affected by ASD or learning or psychiatric difficulties, and SES was gathered from all of the families.

2.4. Analyses

To determine the differences between the three groups on the dependent measures, one-way ANOVAs were run in SPSS. Alpha level was set at .01 as our sample size was relatively small. Moreover, effect sizes are reported for all ANOVAs to ensure that differences between groups did not occur by chance. Scheffe’s post-hoc tests were conducted to determine specific group differences; the alpha for these tests was set at .01 to correct for multiple comparisons. Effect sizes are also reported for the ANOVAs to demonstrate that effect sizes in most cases are quite large; we believe this justifies our use of a relatively liberal multiple-comparison correction.

3. Results

3.1. Autistic symptomatology

Given that our samples were determined in part by the OO group not meeting criteria on the ADOS-G and the HFA group still meeting criteria, it is not surprising that there were highly significant differences between the groups on the Communication (F(1,22) = 62.85, p < .001) and Social Interaction (F(1,22) = 67.87, p < .001) subscales.

However, there were no significant differences on the ADI-R algorithm scores, which are based on earlier functioning (at the times when the child showed the highest level of symptoms in this study, as mentioned in Section 2). The Qualitative Abnormalities in Reciprocal Social Interaction subscale showed no significant difference between the groups (OO = 20.5 (2.11),
HFA = 21.64 (3.23); F(1,25) = 1.10, n.s.), nor did the Qualitative Abnormalities in Communication subscale (OO = 18.58 (2.47), HFA = 17.86 (4.37); F(1,25) = .260, n.s.) or the Restricted, Repetitive, and Stereotyped Patterns of Behavior subscale (OO = 5.00 (2.45), HFA = 5.50 (2.44); F(1,25) = .298, n.s.). On the Current Behavior Algorithm, however, which assesses current functioning, the expected large differences between the groups were found and confirmed the differences in autistic diagnostic status between the groups. On the Qualitative Abnormalities in Reciprocal Social Interaction subscale the OO group had an average of 2.00 (2.52) and the HFA mean was 8.36 (3.78) (F(1,25) = 26.07, p < .001; partial $\eta^2 = .51$), on the Qualitative Abnormalities in Communication subscale the OO group had a mean of 1.08 (1.50) and the HFA mean was 6.36 (3.88) (F(1,25) = .21.15, p < .001; partial $\eta^2 = .46$.) and on the Restricted, Repetitive, and Stereotyped Patterns of Behavior subscale the OO group had a mean of 1.08 (1.19) and the HFA mean was 3.07 (2.13) (F(1,25) = 8.838, p < .01; partial $\eta^2 = .26$).

3.2. Adaptive functioning

Unlike previous research with high-functioning children with ASD, the three groups scored within the average range on all subscales except Socialization. On the Socialization scale, as would be expected, the HFA group mean did fall in the impaired range. It should be noted, however, that in both the OO and HFA groups there were individual children who fell in the impaired range on all three subscales. While there were no significant differences between the groups on the Daily Living Skills subscale, the OO group was not significantly different from the TD group on both the Communication and Socialization subscales, and the OO group was significantly higher on the Socialization subscale than the HFA group (Table 2).

3.3. Problem behaviors

The BASC $t$-scores can be seen in Table 3. None of the $t$-score means for any of the groups fell within the clinical range. It should be noted, however, that the HFA group’s means were within the at-risk range on the Adaptability, Atypicality, Withdrawal, Social Skills, and Leadership measures. The OO group was just into the at-risk range on Attention problems. Again, however, it is important to note that individual children in both the OO and HFA groups fell within the clinical range on some of these subscales. Unlike previous research (e.g. Lindner & Rosen, 2006), there were no significant differences between the groups on Hyperactivity, Aggression, Anxiety, Somatization, and Adaptability. On a number of other subscales, the OO group was not significantly different from the HFA group (i.e., Conduct Problems, Depression, Atypicality, and Withdrawal); however, only the Atypicality subscale showed any signs of elevation from the normal range.

3.4. Language and communication

The OO group fell within the normal range on all of the language and pragmatic language measures (see Table 4). While the OO group performed significantly better than the HFA group on the PPVT, WISC, TOPL and TLC, no firm conclusions could be made about the CELF-3 composite measures as the group differences between the OO and HFA group were not significant. However, as can be seen in Table 5, the OO group did perform at a significantly higher level than the HFA group on the WISC-IV Vocabulary and Comprehension subtests, as well as on the Concepts and Directions and Formulated Sentences subtests of the CELF-3. Moreover, while the HFA group had several individuals who fell well within the impaired range on individual subtests, the majority of the OO group fell well within the normal range.
3.5. Differences between autism groups on history

Demographic comparisons obtained from the ADI-R interview were used to investigate differences between the OO and HFA groups. The OO group was found to have been diagnosed approximately one year earlier than the HFA group (see Table 6), but the difference between the groups was only approaching significance. Chi-square tests of significance were conducted to determine if there were statistical differences between the OO group and the HFA group on measures of history of ASD within the family, whether the children were currently on medication, and an approximate measure of what type of intervention they had received (see Table 6). There were no significant differences between the groups on the measures of family history of autism or current medication use. The categorical measure of intervention obtained from the ADI-R history showed that the OO group was significantly more likely to have received an intensive program of behavioral intervention than the HFA group.

4. Discussion

The purpose of this study was to investigate how children who had been diagnosed with ASD as toddlers, but now in late childhood no longer carried an ASD diagnosis, compared with typically developing children and high-functioning children who still carried the ASD diagnosis on early and current autism symptoms, adaptive functioning, problem behaviors, and language and communication skills. Moreover, we examined some aspects of the OO and HFA children's interventions in a preliminary attempt to ascertain why they developed different outcomes. We consider each of these in turn.
4.1. Autism symptomatology

There were no differences between the OO and HFA groups on the number of overall autism symptoms in early childhood as measured by the ADI-R. This is consistent with the results of Sutera et al. (2007), who found that few symptoms or skills observed at age 2 predicted movement off the spectrum at age 4. Of course, there were significant differences between the groups on their ADOS scores given that one way the groups were ascertained was by choosing children for the OO group that no longer met diagnostic criteria on this measure. The HFA group also had higher scores than the OO group on all the subscales of the ADI-R Current Diagnostic Algorithm, which is consistent with the fact that the former children are still on the spectrum.

4.2. Adaptive functioning

The OO group scored within the normal range on all subscales of the Vineland, and did not differ significantly from the TD group on any subscale. In contrast, as expected, the HFA group scored significantly lower than the TD group on the Vineland Communication and Socialization subscales. Interestingly, though, only on the Socialization subscale did the OO group score significantly higher than the HFA group. There were no differences between the groups on the Daily Living Skills subscale. The HFA group was within the normal range on the Communication subscale but the Socialization subscale was in the impaired range, which is consistent with previous studies reported that socialization is generally lower than expected for mental age in children with ASD (Liss et al., 2001). The OO group, however, appears to have overcome their difficulties on this measure, at least according to parent report.

4.3. Problem behaviors

Lindner and Rosen (2006) used the BASC to assess the level of functioning of a group of individuals with Asperger syndrome. Although they found no differences between the Asperger group and the typically developing group only on Aggression and Somatization, we found no significant differences between the groups on those subscales plus Hyperactivity, Anxiety and Adaptability, perhaps because of our more stringent alpha criterion. The OO group scored within the normal range on all of the

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**Table 4**

<table>
<thead>
<tr>
<th></th>
<th>TD group N = 14 unless otherwise indicated</th>
<th>OO group N = 13 unless otherwise indicated</th>
<th>HFA group N = 14 unless otherwise indicated</th>
<th>F-test, significance, effect size and group comparison</th>
</tr>
</thead>
<tbody>
<tr>
<td>PPVT standard score</td>
<td>121.15 (13.30)</td>
<td>115.54 (16.00)</td>
<td>95.38 (15.25)</td>
<td>F(2,36) = 10.76** Partial $\eta^2 = .37$ OO &gt; HFA&quot; TD &gt; HFA&quot;</td>
</tr>
<tr>
<td></td>
<td>N = 13</td>
<td>Range 97–140</td>
<td>N = 13</td>
<td>Range 5–31</td>
</tr>
<tr>
<td>TOPL standard score</td>
<td>110.00 (9.79)</td>
<td>98.54 (10.46)</td>
<td>78.38 (16.89)</td>
<td>F(2,37) = 21.21** Partial $\eta^2 = .33$ OO &gt; HFA&quot; TD &gt; HFA&quot;</td>
</tr>
<tr>
<td></td>
<td>Range 91–26</td>
<td>Range 75–12</td>
<td>N = 13</td>
<td>Range 58–25</td>
</tr>
<tr>
<td>WISC verbal composite index</td>
<td>120.79 (16.70)</td>
<td>111.62 (17.21)</td>
<td>88.85 (14.60)</td>
<td>F(2,37) = 13.71**</td>
</tr>
<tr>
<td></td>
<td>Range 100–52</td>
<td>Range 79–40</td>
<td>N = 13</td>
<td>Range 55–119</td>
</tr>
<tr>
<td>CELF receptive language standard score</td>
<td>122.29 (18.64)</td>
<td>112.62 (18.86)</td>
<td>91.64 (17.81)</td>
<td>F(2,38) = 10.09**</td>
</tr>
<tr>
<td></td>
<td>Range 92–150</td>
<td>Range 84–150</td>
<td>Range 53–120</td>
<td>Partial $\eta^2 = .35$ TD &gt; HFA&quot;</td>
</tr>
<tr>
<td>CELF expressive language standard score</td>
<td>118.07 (14.46)</td>
<td>102.00 (14.24)</td>
<td>87.69 (15.25)</td>
<td>F(2,37) = 14.54**</td>
</tr>
<tr>
<td></td>
<td>Range 96–143</td>
<td>Range 82–128</td>
<td>N = 13</td>
<td>Range 50–114</td>
</tr>
<tr>
<td>TLC interpreting intentions standard score</td>
<td>109.25 (18.32)</td>
<td>99.80 (17.75)</td>
<td>78.21 (10.66)</td>
<td>F(2,33) = 13.62**</td>
</tr>
<tr>
<td></td>
<td>N = 12</td>
<td>Range 85–135</td>
<td>Range 65–94</td>
<td>Partial $\eta^2 = .45$ OO &gt; HFA&quot; TD &gt; HFA&quot;</td>
</tr>
</tbody>
</table>

*p < .01  
"p < .001.
subscales of the BASC, although they fell in between the TD and HFA groups on a number of measures and were borderline at-risk on the Attention subscale. The borderline at-risk attention scores are consistent with the existence of a subgroup of optimal outcome children who present with ADHD as they lose their autism symptoms (Fein, Dixon, Paul, & Levin, 2005). The HFA group, in contrast, was in the at-risk range for their mean t-scores on the Adaptability, Atypicality, Withdrawal, Social Skills, and Leadership subscales, indicating that they continue to experience behavioral difficulties in these areas. It should be noted, however, that the OO group was not significantly different from the HFA group on Conduct Problems, Depression, Atypicality, and Withdrawal. Moreover, on all of these subscales we were unable to determine (using our strict alpha criterion) whether or not the OO group was statistically distinguishable from the TD group. Because some of the OO children’s scores were elevated in the current study at 8–13 years of age, it is quite possible that problems with internalizing or externalizing behaviors might emerge as they enter adolescence. Furthermore, these data are based on a parent checklist rather than a parent clinical interview, direct psychiatric evaluation or other informant information, and these parents might be invested in a fully “normal” outcome for their children.

In summary, although the individuals with high-functioning ASD in this sample are continuing to display adjustment difficulties in middle childhood and early adolescence, for the most part the individuals with optimal outcome are experiencing few adjustment problems, with the exception of attention difficulties.

4.4. Language and communication

The OO group did not perform significantly differently from the TD group on all the composite measures of language and pragmatic language given, and performed at a significantly higher level than the HFA group on all composite measures
except for the CELF. This is in contrast to the previous study, which included some of these OO children, which had found that the OO group experienced pragmatic difficulties (Kelley et al., 2006). The current finding also contrasts with those of Sallows and Graupner (2005), who found that language abilities were not improved to the same extent in their optimal outcome group as behavioral and cognitive measures. However, the OO children here are a few years older than those in the Sallows and Graupner sample, which suggests that language abilities may continue to improve in optimal outcome children long after treatment has been terminated.

It is worth noting here that the HFA group also scored within the normal range on all language measures, despite performing more poorly than the other two groups. Although their TOPL and TLC scores were significantly lower than those in the TD and OO groups, and were the low points in their profile (an expected finding since they assess pragmatic language abilities), their group mean did fall within the low normal range.

A slightly different pattern of findings emerged when individual subtests of the composite language tests were examined. The OO group scored significantly higher than the HFA group on the WISC-IV Vocabulary and Comprehension, as well as the CELF-3 Concepts and Directions and Formulated Sentences. Moreover, while there were several individual children in the impaired range on each of the language subtests in the HFA group, the OO group included no children in the impaired range on most subtests, and only one or two children in the impaired range on the CELF-3 Recalling Sentences and Listening to Paragraphs subtests, and on the Comprehension subtest of the WISC-IV. This pattern of findings indicates that while many children in the HFA group may still be experiencing subtle language and communication impairments, the children with OO appear mostly unimpaired, with subtle difficulties only in remembering orally presented information, as well as in the more general social knowledge that is assessed by the Comprehension subtest of the WISC.

4.5. Differences between autism groups on history

There were no differences between the HFA and OO groups on having a history of ASD in the family or currently being on medication. Except for one HFA child on Risperidol, the medications were all for attention problems or depression. Although there were no statistically significant differences between the groups on the age at diagnosis, this is probably due to the small sample size of the current study as the group means were approximately one year apart and there was a trend toward significance \( p = .095 \). The OO group was significantly more likely to have received intensive behavioral intervention, which is likely to have contributed to their optimal outcome. Given the retrospective and categorical nature of the measure of early intervention, as well as the small sample size, however, this hypothesis must be interpreted with caution. It is also worth noting that both the OO and HFA groups contained several children who were currently taking psychoactive medications of various sorts (most for attention or depression/anxiety). Thus, while the OO group seemed to be functioning within the average range according to their parents on problem behaviors, it is clear that at least a subset of these youth experience problems with attention or depression and anxiety.

4.6. Comparisons with previous outcome studies

This study is comparable to those that find there are individuals with ASD who go on to achieve relatively normal functioning (Lovaas, 1987; McEachin et al., 1993; Sallows & Graupner, 2005). Unlike those other studies, however, the current study cannot address what proportion of children has the potential to reach this optimal outcome. Moreover, because of the retrospective nature of this study we are unable to pinpoint precisely which early variables predicted the children's membership in either the OO or HFA groups. There was an indication of treatment differences between the OO and

<table>
<thead>
<tr>
<th>Table 6</th>
<th>Other ASD variables.</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>OO group</td>
</tr>
<tr>
<td>Age at diagnosis in months</td>
<td>32.15 (11.73)</td>
</tr>
<tr>
<td>History of ASD in family</td>
<td></td>
</tr>
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<td>None</td>
<td>6</td>
</tr>
<tr>
<td>Present in extended family</td>
<td>1</td>
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<td>Present in immediate family</td>
<td>5</td>
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<tr>
<td>Medication</td>
<td></td>
</tr>
<tr>
<td>No</td>
<td>9</td>
</tr>
<tr>
<td>Yes</td>
<td>4 Strattera, Concerta, Zoloft, and Adderal</td>
</tr>
<tr>
<td>Type of treatment</td>
<td></td>
</tr>
<tr>
<td>No ABA</td>
<td>0</td>
</tr>
<tr>
<td>Eclectic with Some ABA</td>
<td>3</td>
</tr>
<tr>
<td>&lt;20 h per week ABA</td>
<td>2</td>
</tr>
<tr>
<td>&gt;20 h per week ABA</td>
<td>8</td>
</tr>
</tbody>
</table>
HFA groups but a more in-depth measure would need to be collected to give more weight to the findings, and of course a prospective study would be ideal for determining the effect of intervention on outcome.

A major strength of the current study is the wide variety of outcome measures given to the children. Several authors have criticized the field of outcome research for focusing mainly on IQ tests (Charman & Howlin, 2003; Shonkoff et al., 1988). Moreover, few studies have addressed both symptomatology and social and adaptive functioning as the current study did (Tsatsanis et al., 2004). By focusing on these measures we can show that although all children in this study showed intelligence within the average range, the HFA group continued to display residual autism symptoms, including problems in social relationships, communication problems, atypical behaviors, and pragmatic language difficulties. In contrast, the OO group showed no group elevations on BASC scores, and their social and communication patterns were well within the normal range. Granted, these results were from parent checklist responses and were filled out for the most part before adolescence. We are continuing to follow these children into adolescence and in fact are observing elevated rates of anxiety and depression in the OO children, which might be due to their advancing into adolescence and/or the use of more sensitive measures.

Another strength of this study is that it was undertaken several years after the OO children had stopped receiving treatment and was thus able to show that the children were continuing to display an optimal outcome. Moreover, it is the first study that we know of in which the outcome assessment was not conducted by the same researchers who conducted the treatment; we had little vested interest in the outcome of this study.

4.7. Limitations of the current study

This study was not able to address on any level what percentage of children diagnosed with an ASD may reach this optimal outcome level. Although the large majority of the children came from the clinical files of the third author, there is no way to ascertain what proportion of children with ASD these children represent.

The children were assigned to one of four categorical levels with regard to the type of treatment they had received as young children. This was a somewhat crude measure of intervention, though obtaining accurate and specific information retrospectively is very difficult (Eaves & Ho, 2004; Shonkoff et al., 1988). Some have argued that the type of intervention the child receives is not as important as that s(he) receives it early and intensively (Mundy & Stella, 2000). Moreover, many have asserted that not all interventions work for all children and there is a need for research on types of interventions other than behavioral interventions (Beglinger & Smith, 2005). The current study was not able to address these issues, particularly because most of the children received at least some level of behavioral intervention (ABA).

Lack of power was also an issue in this study. While this was inevitable due to the relative scarcity of optimal outcome children, it does mean that the data must be interpreted with some caution. We did our best to ensure that the group differences obtained were indeed group differences by only attributing group differences to measures that showed an alpha of .01 or lower. While this is not a complete solution, we believe that the consistent pattern of results across all measures (where the OO group means were higher than those of the HFA group means and closer to the TD group means) indicates that the children in the OO group are indeed performing quite well on most of the measures.

Mental retardation is present in more than half of children with ASD (Fombonne, 2005). Relationships between variables found in this study may not be at all applicable to low-functioning children with ASD as these children likely follow different developmental pathways (Stevens et al., 2000). However, it has been suggested that low-functioning and high-functioning children with ASDs be studied separately as they should be conceptualized as substantially different disorders (Fein et al., 1999). It should be noted that the HFA group studied here were for the most part quite mildly affected. Had we used an HFA group with a higher level of autism symptoms, presumably we would have found greater differences between the OO and HFA groups.

5. Conclusions

The optimal outcome children in this study have, to a large extent, overcome their social, communicative, and behavioral difficulties and have become engaged in the social world. Although the OO children had lower group means than the TD group on a few of the standardized tests and behavioral measures, their group means were still well within the normal range on all measures. How they and their families and therapists accomplished this cannot be answered definitively through a retrospective study. The treatment data, however, suggest that the proportion of children receiving intensive behavioral treatment was a significant factor: eight of the 13 OO children for whom we had treatment data (62%), but only two of the 14 HFA children (13%) received more than 20 h per week of ABA treatment, and all of the OO children experienced some form of ABA intervention. We do not mean to suggest that ABA treatment alone can accomplish this outcome; clearly, there are child characteristics, including most importantly, potential for normal IQ and lack of an intractable language disorder, that probably are necessary as well (see Dawson, 2008; Helt et al., 2008; and Mundy & Crowson, 1997, for discussion of this issue).

Whether the difference between the OO and HFA children is a categorical difference or a matter of degree remains to be determined; the sample sizes in this study are too small and too variable to clarify this issue. However, it is apparent that these OO children have experienced something more dramatic than simply a reduction in symptoms across development (e.g. Klinger & Renner, 2000). While both the OO and HFA groups demonstrated verbal ability within the normal range, the HFA group was found to experience continuing difficulty to a much greater extent than their OO peers, who were
indistinguishable from the TD group on many measures and within the average range on all of them. Further research is needed to address more subtle aspects of social-cognitive functioning, including how the children interact with others.

Another issue that is clear from the current study is that there is a definite change in the symptom profile of individuals with high-functioning ASD across development. This study lends support to research that suggests that many children with HFA improve to a great extent in middle childhood, even if they do not attain optimal outcome status (Fein et al., 1999; Starr, Szatmari, Bryson, & Zwaigenbaum, 2003; Stevens et al., 2000). The fact that the HFA group means were within the normal range on all of the language measures and several of the BASC subscales suggests that at least some high-functioning children with autism may improve much more in late childhood and early adolescence than was previously thought.

What does the existence of this optimal outcome group tell us about ASD and about development? It can be argued that optimal outcomes in these children suggest that the course of this disorder may be halted or even reversed, at least for high-functioning children with ASD. The current study does not provide an answer to how this change in course was accomplished; prospective studies are needed to address this issue. The existence of these optimal outcome children would also seem to suggest that atypical development, and development itself, can be altered significantly by the environment. While there are limiting factors (and mental retardation in children with ASD is probably the most common and obvious limiting factor), for some children the trajectories of development can be altered quite dramatically (see Helt et al., 2008 for theoretical speculations about the mechanisms by which behavioral treatment can accelerate development and reverse symptoms).

It is clear that at least some children with ASD are able to achieve an excellent outcome; further research must delineate the pathways by which this outcome is achieved. Perhaps one day optimal outcomes will be, at least for higher-functioning children, more common than the relatively rare occurrence that they are today. Until then it is important to gain as much information about these children as possible, to determine which factors have placed them on such a positive developmental trajectory.

Acknowledgments

We would like to thank all of the families who gave so generously of their time to participate in this research and all of the undergraduate research assistants who helped collect and enter this data.

References


