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When an Early Diagnosis of Autism Spectrum Disorder Resolves, What Remains?

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Abstract

A chart review was performed of 38 children diagnosed with autism spectrum disorder (ASD) by 3 years of age at an inner-city developmental program who subsequently experienced resolution of ASD symptomatology and no longer met diagnostic criteria for ASD at follow-up an average of 4 years later. Demographic, developmental/cognitive data, Childhood Autism Rating Scale, and Autism Diagnostic Observation Schedule data as available were reviewed from the initial diagnostic evaluation and at the time of follow-up. Services received by the children between the time of diagnosis and follow-up, educational setting at the time of follow-up, and emotional/behavioral and learning diagnoses made by the multidisciplinary team at follow-up were reviewed. The findings indicate that residual emotional/behavioral and learning problems were present at follow-up in the vast majority of children in this group and that the majority continued to require educational support.

Keywords

optimal outcomes, autism recovery, residual effects

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Although autism spectrum disorder (ASD) has generally been considered a lifelong condition, it has been acknowledged for more than 40 years that some individuals with an early diagnosis of ASD do not meet criteria for the diagnosis at a later age.[1-3] Lovaas used the term *recovery* to characterize the outcome of this group of children who received intensive behavioral intervention and later could be educationally mainstreamed and had average Intelligence Quotient (IQ).[2] The term *optimal outcome* was coined by Fein to characterize a group of 34 individuals with early ASD whose later social functioning could not be distinguished from typical controls and who appeared to be cured of ASD.[4]

Follow-up studies have spoken of varying degrees of learning or emotional vulnerability that continue in the "positive outcome" populations, including attention problems[5-7] and language problems.[8,9] More recently, a long-term follow-up study of 198 children diagnosed with ASD at ages 2 to 4.5 years found that 17 children no longer met the criteria for ASD at the 2-year follow-up. Later, when the children were about 10 years of age, parents were interviewed by phone regarding the children's school age needs. Based on parent report, all 17 children continued to have some type of ongoing developmental and/or neuropsychiatric challenge.[10]

The goals of the current study were to further characterize the residual learning, cognitive, and emotional/behavioral diagnoses as well as the range of educational supports required at school age in a group of children with a history of an early diagnosis of ASD that resolved.

Methods

The setting for the current study was a university-affiliated inner-city diagnostic Early Intervention program where children with developmental disabilities continue to be followed and receive intervention through adulthood. The clinical database indicated that 569 children

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younger than age 3 were diagnosed with ASD by the program between 2003 and 2013. Review of the database indicated that there were 38 children who had an ICD9 diagnostic code of 299.81, "ASD residual," as was the clinic practice for indicating that follow-up re-evaluation had been carried out and demonstrated that the child no longer met criteria for ASD. This study was a chart review of those 38 children.

Original and follow-up diagnoses were made by an experienced and stable multidisciplinary team that included bilingual developmental and behavioral pediatricians, psychologists, psychiatrists, speech and language pathologists, and special educators, and were based on *Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition* (DSM IV), criteria.

As a clinical program, the evaluations on the children were carried out on the basis of clinical need (such as diagnostic clarification and/or educational planning) rather than on the basis of a research protocol. As such, not all children received a full battery of assessments at either the initial or follow-up points. For example, DSM IV criteria and the Childhood Autism Rating Scale were used for ASD diagnosis for all children. The Autism Diagnostic Observation Schedule was used for clinical purposes only, such as when children were less clear-cut due to very young age (<24 months), had mild/questionable symptomatology, or in order to support parental understanding of their child's social communicative deficits. As such, an Autism Diagnostic Observation Schedule is not available for all 38 children at either baseline or follow-up. Similarly, developmental testing initially and cognitive testing at follow-up were not available on all of the children. There were no differences in demographics, specifically ethnic group, language spoken at home, socioeconomic status, and maternal education, between those who had an Autism Diagnostic Observation Schedule initially or at follow-up and those who did not as well as those who had developmental or cognitive testing and those who did not. In the course of receiving clinical care at the center, all of the children in this sample were offered ongoing follow-up visits. Some children were seen by a developmental pediatrician every 6 months and others much less regularly by parent choice, such as at the time of transition to kindergarten or later when the parents developed questions or concerns. Data from the visit documenting that the child no longer met ASD criteria and subsequent evaluations were used for follow-up findings. This was on average 4 years after the original diagnosis (range: 3-9.5 years).

The available data from both the original and follow-up evaluations was reviewed, including demographics, Childhood Autism Rating Scale scores, Autism Diagnostic Observation Schedule results (when available), developmental quotients and cognitive scores, and developmental and emotional/behavioral diagnoses made by the multidisciplinary team. Developmental quotients and cognitive scores included the Bayley Scales of Infant Development (Second and then Third Edition) at initial diagnosis and Wechsler Preschool and Primary Scale of Intelligence, Wechsler Intelligence Scale for Children, or Stanford Binet IV at follow-up when available. Therapeutic services received by the child since initial diagnosis were determined based on parent report and review of available records. Current educational setting was determined as per the child's current Individualized Educational Plan (IEP).

Results

Demographics

The 38 subjects of this study represented 7% of the 569 children receiving an early diagnosis at the center in a 10-year period. (In order to provide a real denominator for calculating the percentage of children no longer meeting criteria for ASD at follow-up, we need to know the number of children of the 569 who actually came for follow-up. Unfortunately, a change in our program's clinical affiliation since the original cohort was studied with resultant changes in the charting system does not allow us to establish the exact number of the 569 children who came for follow-up. Our best estimate of the follow-up rate is on the basis of a prior study involving a subset of 108 children from this group of 569 who were diagnosed by 24 months. The follow-up rate for that group was 71%. Utilizing the 71% follow-up rate, the 38 children no longer meeting criteria for ASD would represent 9.4% of the sample.) The mean age of the sample was 2.6 $\pm$ 0.9 years of age at initial diagnosis and 6.4 $\pm$ 2.8 years at follow-up. The sample was 80% male with a diverse demographic representative of the community served: 36% self-identifying as Caucasian, 44% Hispanic, and 10% African American. Forty-six percent of the sample had Medicaid, and 42% were bilingual (Spanish and English). Eighty percent of the children received Early Intervention services (most commonly weekly special instruction for 1 hour per week and twice-weekly speech and occupational therapies) and 39% had received Applied Behavioral Analysis.

Table 1. Autism and Developmental/Cognitive Testing: Initially and at Follow-up.

	Initial	Follow-up
Mean age, y	2.6 $\pm$ 0.9	6.4 $\pm$ 2.8
Mean CARS score	32 $\pm$ 3	25 $\pm$ 4
ADOS classification/ diagnosis		
Available in	21/38	23/38
Autism, %	29	
ASD, %	70	
Non-ASD, %	–	100
Cognitive/developmental level		
Available in	29/38	33/38
Severe developmental delay, %	33	–
Moderate developmental delay, %	23	–
Mild developmental delay, %	27	–
No delay, %	17	–
Borderline, %	–	6
Low average, %	–	27
Average, %	–	67

Abbreviations: ADOS, Autism Diagnostic Observation Schedule; ASD, autism spectrum disorder; CARS, Childhood Autism Rating Scale.

Initial Evaluation Results

The results of Autism assessments and developmental/cognitive testing at initial and follow-up evaluations can be found in Table 1 and Figure 1. All of the children in the sample were initially diagnosed with ASD (as opposed to a specific DSM-IV diagnosis) as per the standard clinical practice at our clinic for children under age 3 years. The mean initial Childhood Autism Rating Scale total score was 32 $\pm$ 3, in the range of

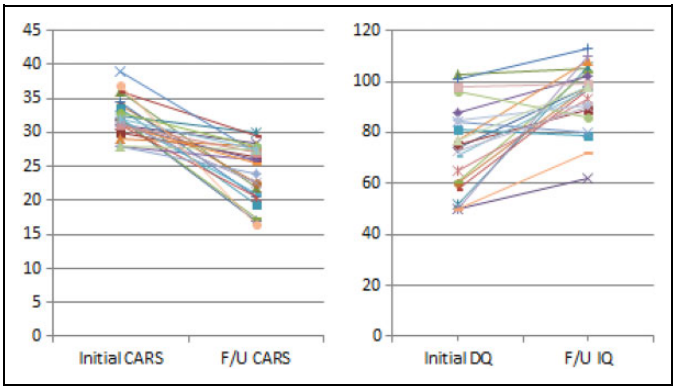

Figure 1. Individual children's initial and follow-up Childhood Autism Rating Scale and Developmental Quotient/Full-Scale IQ.

mild-moderate symptoms of autism, and initial Autism Diagnostic Observation Schedule classification (available in n = 21) categorized 29% as having autism and 70% as having ASD. Initial developmental testing with the Bayley Scales of Infant Development Ed. II or III, available in 29 of the 38 participants, revealed that 33% functioned in the range of severe developmental delay (Mental Development Index on the Bayley-II or Cognitive Composite on the Bayley-III <55); 23% in the range of moderate developmental delay (Mental Development Index or Cognitive Composite 55-69); 27% in the range of mild developmental delay (Mental Development Index or Cognitive Composite 70-84); and 17% in the range of no delays.

Follow-up Evaluation Results

At follow-up, it was the clinical impression of the multidisciplinary team using DSM IV that none of the participants continued to meet criteria for an ASD diagnosis. Mean Childhood Autism Rating Scale score at follow-up for the group was 25 ± 4, with 30 being the cutoff for ASD (Table 1). On the Autism Diagnostic Observation Schedule (available in 23 of the 38), all available scores were in the nonautistic range.

However, other diagnoses were present (Table 2). Only 8% (n = 3) of the children warranted no diagnosis other than having had a history of ASD. Sixty-eight percent (n = 26) had language/learning disabilities, 49% (n = 19) of the children were diagnosed with externalizing behavior problems (attention-deficit hyperactivity disorder [ADHD], oppositional defiant disorder, disruptive behavior disorder), 24% (n = 9) were diagnosed with internalizing problems (mood disorder, anxiety disorder, obsessive compulsive disorder [OCD], selective mutism), and 5% (n = 2) were given a significant mental health diagnosis (psychotic disorder not otherwise specified). Sixty percent (n = 23) of the children received 2 diagnoses, with the most common combination being language/learning disability and ADHD.

Follow-up cognitive testing, available in 33 of the 38 participants (Table 1), revealed that none of the children functioned in the range of intellectual disability, including those who previously tested as delayed on the Bayley. The average Full Scale IQ of the sample at follow-up was 93 ± 14; 6% (n = 2) scored in the borderline category; 27% (n = 9) in the low average range; and 67% (n = 22) in the average range (Table 1). The mean Verbal IQ of the sample at follow-up was 92 ± 14 and mean Nonverbal IQ was 95 ± 12 (Figure 1).

Table 2. Residual Diagnoses and Educational Setting at Follow-Up.

Diagnoses at follow-up	% (n)
Language or learning disability	68 (26)
Externalizing behavior problems (ie, ADHD, ODD, disruptive behavior disorder)	49 (19)
Internalizing problems (ie, mood disorder, anxiety disorder, OCD, selective mutism)	24 (9)
Thought disorders (ie, psychosis nos)	5 (2)
No diagnoses	8 (3)
Educational/academic setting at follow-up	
Mainstream academic setting without support	26 (10)
Mainstream academic setting with assistant teacher support	13 (5)
Integrated setting/resource room	29 (11)
Self-contained class	21 (8)

Abbreviations: ADHD, attention-deficit hyperactivity disorder; OCD, obsessive-compulsive disorder; ODD, oppositional defiant disorder.

Follow-Up Educational/Academic Setting

Information regarding academic setting at follow-up was available for 34 of the 38 children: 26% of the children (n = 10) were in a mainstream class, 13% (n = 5) were in a mainstream class with assistant teacher support, 29% (n = 11) were in an integrated co-teaching (ICT) or collaborative team teaching (CTT) class/or received resource room, and 21% (n = 8) were in a self-contained classroom (Table 2).

Discussion

A given for this study is that there is a subset of children with an early ASD diagnosis who show a categorical improvement in their original social communicative impairment, such that they no longer manifest a social communicative impairment impacting on their functioning at some later point. Such a phenomenon has been repeatedly documented for a small group of children with ASD. Less clear has been whether children with this history continue to experience residual learning and emotional/behavioral problems. The findings of this small sample suggest that at least in the early elementary years, such residual problems are very common for this group. In this sample, although the loss of the ASD diagnosis was associated with gains in cognition, the vast majority of these children who experienced resolution of ASD continued to manifest symptoms of other emotional/behavioral and/or learning diagnoses (92%) and continued to require educational supports (74%).

Though it has been reported in multiple studies that a small subset of children with early ASD improve in terms of their social functioning and no longer warrant a diagnosis of autism,[11] this persistent finding continues to beg several

questions: Was autism overcalled in these children to begin with? Are some children better able to respond to intervention? Does the specific intervention the child receives contribute to outcome? All are possible. Based on our experience, our sense is that the symptoms evolve—in some children in response to intervention, in others due to their individual developmental trajectories.

And who is most likely to evolve in this positive direction? Those with the mildest symptoms to begin with. This study as well as previous studies[11] all support that it is the children with milder autistic symptoms who are most likely to follow this pattern of resolution of autistic symptoms. The milder forms of autism likely serve as a holding area. There, children await the emergence of the signs and symptoms of a more specific developmental or emotional-behavioral condition—whether that is language/learning disability or emotional-behavioral disorders that require more language, cognition, and increased behavioral expectations in order to be specifically identified and diagnosed. A 2-year-old simply does not have sufficient language or cognition to manifest the signs of schizophrenia, or to give voice to anxiety.

The strengths of the current study lie in the clinical context in which the study was carried out. The children were seen in a diagnostic center serving a diverse local population. They were assessed by the same highly experienced clinical team for both the initial ASD diagnosis as well as the subsequent clinical visits during which the children were determined to no longer meet ASD criteria and were diagnosed with other academic and/or emotional behavioral diagnoses. The multidisciplinary clinical team possesses the expertise to diagnose both learning disabilities and emotional/behavioral disorders. The previous studies on children experiencing an optimal outcome or resolution of ASD have had limitations. In Fein's studies on optimal outcome, the research group did not complete the initial evaluations in the very young toddlers with optimal outcome, but instead reviewed records documenting the initial clinical findings and used record review to determine whether the children met ASD criteria initially.[4] In the Olsson study, the follow-up regarding current needs was based on telephone interview.[10] The children were not seen, nor were their current educational or emotional/behavioral diagnoses clarified. In the current study, the same clinical team who made the diagnosis originally saw the children in follow-up. The initial diagnoses represented our best effort at a valid early ASD diagnosis. Our follow-up assessment was based on reevaluating the children and in addition to clarifying whether they continued to meet ASD criteria, the evaluation clarified whether they met criteria for other educational or emotional/behavioral diagnoses.

Study limitations include that this was a chart review with a small number of cases and a limited follow-up time frame. It is also possible that there is an inherent bias in characterizing residual deficits in this sample, as it is likely that children without ongoing difficulties would be less likely to be brought back by their parents for follow-up. It is possible that some children followed in the clinic were not identified or coded as having residual ASD and were missed. These factors may impact on our ability to generalize these findings. In the future, we plan to further evaluate differences between those who do and do not move out of the diagnosis over time. These preliminary results do, however, highlight the importance of ongoing monitoring and therapeutic support as needed, in this scenario. This is important information for parents, clinicians, and the educational system as they continue to strive to meet the needs of this unique group of children.

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Author Contributions

LHS and ED wrote the first draft of the manuscript. LHS developed the research question and subsequent revisions of the manuscript. RS, ET, DM, and NT carried out the testing that generated the data on the children. ED and SL created the database and analyzed the data under the supervision of MVM. All of the authors contributed, read, and approved of this manuscript. No honorarium, grant, or other form of payment was given to anyone to produce the manuscript.

Declaration of Conflicting Interests

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Ethical Approval

This study protocol was approved by the Internal Review Board of Albert Einstein College of Medicine, Bronx, New York (IRB no. 2007-546).

References

1. Helt M, Kelley E, Kinsbourne M, et al. Can children with autism recover? If so, how? *Neuropsychol Rev*. 2008;18(4):339-366.
2. Lovaas OI. Behavioral treatment and normal educational and intellectual functioning in young autistic children. *J Consult Clin Psychol*. 1987;55(1):3.
3. Rutter M. Autistic children: infancy to adulthood. Paper presented at: Seminars in psychiatry, 1970.
4. Fein D, Barton M, Eigsti IM, et al. Optimal outcome in individuals with a history of autism. *J Child Psychol Psychiatry*. 2013; 54(2):195-205.
5. Fein D, Dixon P, Paul J, Levin H. Brief report: pervasive developmental disorder can evolve into ADHD: case illustrations. *J Autism Dev Disord*. 2005;35(4):525-534.

6. Zappella M. Familial complex tics and autistic behaviour with favourable outcome in young children. *Infanto-Rev Neuropsiq Infanc Adolesc*. 1999;7:61-66.
7. Kelley E, Naigles L, Fein D. An in-depth examination of optimal outcome children with a history of autism spectrum disorders. *Res Autism Spectr Disord*. 2010;4(3):526-538.
8. Kelley E, Paul JJ, Fein D, Naigles LR. Residual language deficits in optimal outcome children with a history of autism. *J Autism Dev Disord*. 2006;36(6):807.
9. Turner LM, Stone WL. Variability in outcome for children with an ASD diagnosis at age 2. *J Child Psychol Psychiatry*. 2007; 48(8):793-802.
10. Olsson MB, Westerlund J, Lundström S, Giacobini M, Fernell E, Gillberg C. "Recovery" from the diagnosis of autism—and then? *Neuropsychiatr Dis Treat*. 2015;11:999.
11. Lord C, Risi S, DiLavore PS, Shulman C, Thurm A, Pickles A. Autism from 2 to 9 years of age. *Arch Gen Psychiatry*. 2006;63(6): 694-701.