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ORIGINAL PAPER

Narrative Performance of Optimal Outcome Children and Adolescents with a History of an Autism Spectrum Disorder (ASD)

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Abstract Autism Spectrum Disorders (ASDs) have traditionally been considered a lifelong condition; however, a subset of people makes such significant improvements that they no longer meet diagnostic criteria for an ASD. The current study examines whether these "optimal outcome" (OO) children and adolescents continue to have subtle pragmatic language deficits. The narratives of 15 OO individuals, 15 high-functioning individuals with an ASD (HFA), and 15 typically developing (TD) peers were evaluated. Despite average cognitive functioning, the ASD group produced narratives with fewer central "gist" descriptions, more ambiguous pronominal referents, idiosyncratic language, speech dysfluency (more repetitions and self-corrections), and were less likely to name story characters. The OO participants displayed only very subtle pragmatic and higher-level language deficits (idiosyncratic language and self-correction dysfluency).

Keywords Autism · Outcome · Optimal · Narrative · Language · Pragmatics · Dysfluency

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Introduction

Autism Spectrum Disorders are a heterogeneous group of neurodevelopmental disorders that have been behaviorally defined by impairments in reciprocal social interaction and communication, and restricted interests and repetitive behaviors (American Psychiatric Association 2000). ASDs have traditionally been considered a lifelong condition; however, there appear to be a subset of people who make such significant improvements that they no longer meet diagnostic criteria for an ASD (e.g., Fein et al. 2013; Fein et al. 2005; Helt et al. 2008; Kelley et al. 2006, 2010; Sutera et al. 2007). This "optimal outcome" (OO) group generally scores in the average range in measures of cognition, language, adaptive behavior, and social skills (Fein et al. 2013; Helt et al. 2008), but may have subtle residual deficits in higher-order language functions. The current study examines such aspects of pragmatic language functioning in OO children and adolescents by comparing their narrative performance to that of high-functioning peers with an ASD (HFA) and peers with a history of typical development (TD).

ASD Prognosis and "Optimal Outcome"

There has been debate over whether recovery is truly possible in ASDs, as it had previously been conceptualized as a life-long neurological disorder. Longitudinal studies of ASDs have discovered groups with good outcomes (e.g., Lovaas 1987; Sallows and Graupner 2005; Sutera et al. 2007; Szatmari et al. 1989), cross-sectional studies have specifically recruited OO children (e.g., Fein et al. 2013; Kelley et al. 2010, 2006), and detailed clinical case studies have presented clear examples of children who formerly

had an ASD diagnosis who no longer meet diagnostic criteria for an ASD (Fein et al. 2005; Perry et al. 1995; Zappella 2010).

Lovaas (1987) was the first to use the term "recovery" to describe the outcome of a series of children with an ASD. He found that 9 out of 19 children achieved IQ scores in the average range and successfully completed first grade after intensive Applied Behavior Analysis (ABA) treatment. However, social functioning was not evaluated: it is possible the children continued to have significant socialization and communication deficits associated with ASDs. Szatmari et al. (1989) added measures of social functioning and adaptive functioning and found that 4 out of 16 individuals no longer met criteria for an ASD and had adaptive functioning in the average range.

Limitations to OO studies have been that experimenters were not blind to diagnosis in the cross-sectional studies (Fein et al. 2013; Kelley et al. 2010, 2006), and not all studies used gold-standard instruments such as the Autism Diagnostic Observation Scale (ADOS) and Autism Diagnostic Interview (ADI-R). However, there have been several studies that have used the ADOS or ADI-R to evaluate an initial ASD diagnosis (Sallows and Graupner 2005; Sutera et al. 2007), or to confirm whether participants still met criteria for an ASD (Fein et al. 2013; Sallows and Graupner 2005; Sutera et al. 2007). Although the precise factors that contribute to OO are unclear, there are features that have been associated with better outcomes, including: earlier diagnosis and treatment; higher initial receptive language, non-verbal problem-solving and motor scores on standardized tests; greater imitation skills; and a previous diagnosis of Asperger Disorder (AD) or Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS) (as opposed to a diagnosis of Autistic Disorder) (Helt et al. 2008; Sallows and Graupner 2005; Sutera et al. 2007).

Recent studies have employed more stringent criteria to define a subset of individuals as having an "optimal outcome" (Fein et al. 2013; Helt et al. 2008; Kelley et al. 2010; Sallows and Graupner 2005; Sutera et al. 2007). Helt et al. (2008) proposed an operationalized definition, where individuals must: no longer meet diagnostic criteria for an ASD; no longer be considered by the school system to have an ASD; have a full scale IQ greater than 77; and be mainstreamed in a regular classroom and not receive more than 1 h per week of speech, occupational or special educational services. It is estimated that between 3 and 25 % of children originally diagnosed with an ASD lose the diagnosis and may meet these criteria for OO (Helt et al. 2008).

Using these stringent criteria, studies have compared the profiles of OO, HFA, and TD children to explore whether autistic features persist in the OO group and to establish which skills lag behind those of their peers. Residual deficits reported in a range of studies include attention problems, mild social deficits associated with impulsivity and immaturity, anxiety and tics, mild perseverative behaviors and interests, and subtle deficits in language functioning (Fein et al. 2005; Kelley et al. 2006; Naigles et al. 2013; Piven et al. 1996; Sallows and Graupner 2005; Zappella 2010). It is unclear whether some of these subtle deficits are associated with a phenotypic expression of an already-present genetic susceptibility to an ASD, called the Broader Autism Phenotype (BAP) (Landa et al. 1991; Piven et al. 1997), or whether there are some OO individuals who do not display any phenotypic characteristics associated with ASDs and are truly indistinguishable from their peers.

Language Functioning in ASDs

Communication deficits are one of the core features of ASDs. However, there is considerable heterogeneity in language functioning among people with an ASD: a subset are nonverbal or do not acquire functional speech (30–40 %; Tager-Flusberg et al. 2005), whereas others become verbally fluent but reach language milestones significantly later than their peers (Mayo et al. 2013). Studies show that, in higher-functioning individuals with an ASD, phonology (producing and understanding different patterns of speech sounds), vocabulary (using and understanding a variety of words), and grammar (the combining of words and morphemes into phrases) may initially show significant delays (Eigsti et al. 2007; Rapin et al. 2009) but are more likely to attain average proficiency later in development (Kjelgaard and Tager-Flusberg 2001; Naigles et al. 2013; Naigles et al. 2011; Tager-Flusberg 2001). In contrast, pragmatic language difficulties are a defining feature of ASDs and have consistently been found to be more resistant to remediation (Kelley et al. 2006; Tager-Flusberg 2001). Pragmatic language involves an ability to use language appropriately in a social context and to account for the knowledge and interests of the listener (Diehl et al. 2006; Geurts and Embrechts 2010; Kjelgaard and Tager-Flusberg 2001). Deficits in pragmatic skills could contribute to social misunderstanding and impede effective communication, leading to fewer positive social interactions and greater difficulty in social relationships.

Spontaneous language samples, such as narratives, have been used to capture in vivo language abilities (Botting 2002). As typically developing children develop more language skills, their narratives increase in length, include a greater variety of words, and have more complex syntax (Botting 2002). Children and adults with an ASD (aged 6–22), when matched for language abilities to peers without an ASD, have been found to produce narratives that are similar in length, structure, and syntactic complexity (Diehl et al. 2006; Tager-Flusberg and Sullivan 1995). However,

children and adults with an ASD have been found to incorporate fewer story components and include irrelevant details in their narratives, such as when using a picture book (e.g., Losh and Capps 2003) or writing about an observed film clip (Barnes and Baron-Cohen 2012). Detailed scrutiny of narratives have also revealed difficulties with pronoun use, restricted or unconventional vocabulary, dysfluencies, and fewer causal references (e.g., Capps et al. 2000; Colle et al. 2008; Lake et al. 2011; Rapin and Dunn 2003).

The production of pronouns and their antecedents involve an integrated use of social and linguistic knowledge. When a character in a story is first introduced, the first mention typically gives the character's name or a specific description: subsequent mentions can adhere to Gricean principles of pragmatics (Grice 1975) by then referring to that character by pronoun (*he*, *she*, *it*). When it is unclear to whom a pronoun is referring (e.g., 'he was chasing him'), that pronoun's reference is ambiguous. Typically-developing children produce unambiguous pronouns in narration by 10 years of age (Karmiloff-Smith 1985). When telling a story, a speaker must simultaneously narrate while tracking the information that he has already conveyed to the listener. This representation, known as "common ground" (Clark 1996), taps social as well as linguistic knowledge, the integration of which also involves executive demands (Schuh 2012). A preliminary study of children (Edelson 2012) and a study of adults (Colle et al. 2008) with an ASD found that, when telling a story from a picture book, the ASD groups produced more ambiguous pronouns than TD controls. It was conjectured that the ASD groups were less able to attend to what the listener did and did not know, resulting in less effective communication.

People with an ASD are more likely to use words in unconventional ways and to have speech that is repetitive and restricted in content (Eigsti et al. 2007; Ghaziuddin and Gerstein 1996; Rapin and Dunn 2003; Tager-Flusberg 2001). When people communicate, they must use words that are appropriate to the context and the age and formality level of the listener. Failure to do so could result in discourse that is harder to understand and perceived as odd. Idiosyncratic use of language, which includes stereotyped speech (such as lines from movies said in a consistent tone), neologisms (words invented by the individual), and overly pedantic (formal) speech, has been one of the features that has distinguished people with an ASD from language-impaired individuals without an ASD (Rapin and Dunn 2003). A study by Loveland et al. (1990) found that, when recalling a story from a puppet show or a video, children with an ASD produced more inappropriate utterances than verbal ability-matched children with Down Syndrome.

In addition to ambiguous and idiosyncratic references, studies have reported more dysfluent speech in people with ASDs when compared to typically-developing peers (Belardi and Williams 2009; Lake et al. 2011). Dysfluent speech is speech that is interrupted by *repetitions*, *self-corrections*, or *filler words*. *Repetitions* are repeated words or part-words with no functional purpose (e.g., "*They all*-they all were flying"). *Self-corrections* occur when speech is stopped in the middle of an utterance and revised (e.g., "*They were all*-those crows were all sitting on the wires"), whereas *fillers* are sounds with no semantic meaning (e.g., 'uh,' or 'um'). A study by Lake et al. (2011) evaluated speech dysfluencies in high-functioning adults with an ASD during a structured conversation. They found that *repetitions* were produced more often in the ASD group, whereas *self-corrections* and *fillers* were produced more often by non-affected peers. It was postulated that *repetitions* do not serve a communicative function and are *speaker-oriented*, whereas *self-corrections* and *fillers* serve *listener-oriented* functions, as self-corrections could indicate a desire to clarify information for the listener, and fillers could indicate that a speaker has not finished talking and is thinking of what to say [also consistent with work by Fox Tree (2001) and Heeman et al. (2010)]. However, a preliminary study of adults with an ASD and non-affected controls found no differences in production of self-corrections or fillers on the Creating a Story task on the ADOS (a task that requires making up a story using five unrelated objects), although, consistent with previous research, the ASD group produced significantly more repetitions (Belardi and Williams 2009).

The use of mental state verbs (words that indicate what another person is thinking or feeling) in narratives is thought to reflect a speaker's insight into others' mental states. Findings have been mixed, with some studies of ASDs reporting narratives with fewer mental state verbs, such as when telling a story using a story book (e.g., Losh and Capps 2003), or when conversing during a home visit (Tager-Flusberg 1992). However, other studies have found no group differences, including when telling a story using a story book (e.g., Capps et al. 2000; Norbury and Bishop 2003; Tager-Flusberg and Sullivan 1995) or cards from the Thematic Apperception Test (TAT) (Beaumont and Newcombe 2006). Nevertheless, even children with an ASD who produced a similar number of mental state verbs as controls were less likely to give a causal explanation as to *why* a character was thinking or feeling a particular way (Beaumont and Newcombe 2006; Capps et al. 2000).

Evaluation of Language Abilities in OO Children

Considering the social nature of learning and using language (Baldwin and Meyer 2007) and the central role that

communication impairment plays in the diagnosis of ASDs, it is critical to systematically evaluate language functioning in a group of OO individuals. There has been little systematic research to date on language functioning in OO. The most detailed analyses have come from Kelley et al. (2006), who studied OO children (aged 5–9). While OO children had intact grammatical abilities and did not significantly differ from controls on most lexical variables, they struggled with aspects of pragmatic language during a narrative task: they were less likely to identify the goals and motivations of the characters, gave fewer causal explanations, and were more likely to misinterpret story events. These activities require clearly communicating the story to the listener as well as an understanding and interest in the social dynamics of the story and characters. There were also two trends: the OO group tended to make more ambiguous references to events and characters, and to repeat story events more often than their TD peers (Kelley et al. 2006, p. 816).

A later study by Kelley et al. (2010) of a group of older OO children (aged 8–13, many of whom were in the 2006 sample) found no language deficits on standardized language assessments, suggesting that these OO children were able to overcome some semantic and pragmatic deficits when they got older (see also Naigles et al. 2013). However, the study did not evaluate spontaneous language samples. Because standardized language measures have been limited in their ability to capture subtle language deficits (Diehl et al. 2006; Young et al. 2005), the current study utilized spontaneous language samples to better characterize an older group of children and adolescents to evaluate whether pragmatic language deficits could truly remediate over time.

The current study evaluated the narratives of a group of OO, HFA, and TD children and adolescents (ages 9–15). The primary aim was to better characterize the language abilities of a group of OO children and adolescents, with a focus on potentially subtle residual pragmatic language deficits. A secondary goal was to probe for pragmatic language deficits in a well-defined group of individuals with HFA with cognitive scores in the average range (within 1.5 standard deviations from the mean). We hypothesized that this older group of OO individuals would continue to display subtle pragmatic impairments [as suggested by the study by Kelley et al. (2006)], including producing *more ambiguous pronouns* and giving *fewer causal explanations* to characters' mental states. We hypothesized that the HFA group would demonstrate these pragmatic impairments in addition to: inclusion of *fewer story elements*, production of *idiosyncratic language*, and *greater dysfluency* in the form of *repetitions*.

Methods

Participants

The narrative abilities of 15 OO, 15, HFA, and 15 TD peers were evaluated. The participants in the study were selected from a larger study of OO at the University of Connecticut that recruited children from 8 to 21 years old. The sample from this study ranged in age from 9 years, 3 months to 15 years, 8 months old, with an average age of 12.4 to 13 years old. The groups from the current study were matched on age, and gender and verbal IQ (VIQ) did not significantly differ among the groups. The participants were predominantly Caucasian, with only three individuals in the TD group reporting other races or ethnicities. See Table 1 for participant characteristics.

The study was approved by the Institutional Review Board of the University of Connecticut, the Institute of Living Hartford Hospital, Children's Hospital of Philadelphia, and Queens University. Recruitment was done through media outlets (newspaper stories, radio interviews), private practices, and clinic referrals. In some cases, therapists contacted parents of children known to have OOs, and in some cases, parents saw media reports and contacted the investigators. Data were collected as part of a larger study of OO at the University of Connecticut.

Inclusion Criteria

All participants had verbal, nonverbal, and full-scale IQ standard scores greater than 77 (within 1.5 standard deviations (SDs) of the average of 100). Each group had additional specific inclusion criteria.

For the OO group:

1. Participants had a documented ASD diagnosis made by a physician or psychologist specializing in ASDs before the age of 5, verified in a written diagnostic report provided by parents. Early language delay (no words by 18 months or no phrases by 24 months) documented in the report was required. As a second step in confirming diagnosis, the report was edited to remove information about diagnosis, summary, and recommendations but leaving descriptions of behavior. One of the co-investigators (MB), an expert in diagnosis of ASDs and Director of the University of Connecticut Psychological Services Clinic, reviewed these reports, blind to early diagnosis and current group membership. In addition to potential OO participants, she reviewed 24 "foil" reports for children with non-ASD diagnoses, such as global

Table 1 Demographic, IQ, and Vineland Adaptive Behavior Scores for HFA, OO, and TD participants

	HFA (SD) (n = 15)	OO (SD) (n = 15)	TD (SD) (n = 15)	F	*p*	η^2	Post-hoc
Age	12.9 (1.6)	12.4 (1.8)	13.0 (1.6)	.50	.61	.03	
(range)	(10.5–15.7)	(9.3–15.6)	(9.9–15.6)				
Sex ratio (M/F)[a]	14/1	12/3	13/2		.56		
Full scale IQ (FSIQ)	104.6 (11.0)[b]	112.0 (15.2)	116.3 (12.1)	3.00	.06	.13	
(range)	(80–117)	(82–134)	(101–142)				
Nonverbal IQ (NVIQ)	105.9 (12.9)[b]	110.6 (15.6)	118.4 (12.7)	3.06	.06	.13	
(range)	(78–120)	(87–131)	(89–139)				
Verbal IQ (VIQ)	102.6 (14.6)[b]	110.8 (16.1)	112.1 (14.1)	1.70	.20	.08	
(range)	(81–133)	(80–137)	(93–136)				
Vineland communication	85.2 (12.4)	94.4 (11.9)	93.9 (7.9)	3.27	.04	.14	TD > HFA
(range)	(51–100)	(79–110)	(85–115)				OO > HFA
							OO = TD
Vineland socialization	80.9 (16.7)	98.8 (7.6)	103.6 (8.5)	15.67	<.001	.43	TD > HFA
(range)	(54–119)	(80–109)	(91–119)				OO > HFA
							OO = TD

[a] Pearson Chi square

[b] Data only available for n = 14 participants

delay or language disorder. Four potential OO participants were rejected for insufficient early documentation, and were dropped from the study. All 24 foils were correctly rejected.

2. Participants could not currently meet criteria for any ASD according to the Autism Diagnostic Observation Schedule (ADOS; Lord et al. 2000), which was administered by a research-reliable interviewer. In addition, the ADOS of all potential OO cases was reviewed by a clinician with more than 15 years of ASD diagnostic experience (IME, MB, or DF) who confirmed that ADOS scores were below ASD thresholds and that in their expert clinical judgment, an ASD was not present.
3. Participants' scores on the Communication and Socialization domains of the Vineland had to be greater than 77 (within 1.5 SDs of the mean of 100).
4. Participants had to be fully included in regular education classrooms with no one-on-one assistance and no special education services to address autism deficits (e.g., no social skills training). However, participants could be receiving limited special education services or psychological support to address impairments not specific to an ASD, such as attention or academic difficulties.

For the HFA group:

1. Following Collaborative Programs of Excellence in Autism diagnostic guidelines, participants had to meet criteria for an ASD on the ADOS (both Social and Communication domains and total score) and according to best estimate clinical judgment.

For the TD group:

1. Participants could not meet criteria for any ASD at any point in their development, by parent report.
2. Participants could not have a first-degree relative with an ASD diagnosis.
3. Participants could not meet current diagnostic criteria for an ASD on the ADOS, or by clinical judgment. There was no attempt to exclude children with learning or psychiatric disorders, but all 15 TD children in our sample were reading within 1.5 SD of average or above.
4. Scores on the Communication and Socialization domains of the Vineland had to be greater than 77.

Exclusion Criteria

Potential participants for any group were excluded from the study if (1) at the time of the telephone screening they exhibited symptoms of major psychopathology (e.g., active psychotic disorder) that would impede full participation, (2) they had severe visual or hearing impairments per parent report, or (3) they had a history of seizure disorder, Fragile X syndrome, or significant head trauma with loss of consciousness. Further details of the methods from the larger study, including a flow chart of participant inclusion and exclusion, are reported in Fein et al. (2013).

Procedure

Phone screenings based on study criteria were conducted with parents of each potential participant. Those who passed screening were scheduled for an assessment. Informed consent and assent were obtained, as appropriate, prior to testing. The evaluation was administered in a quiet room over the course of two or three testing sessions at the University of Connecticut, the Institute of Living of Hartford Hospital, Queens University, or in the home. Testing lasted approximately 6 h. In most cases, parent interviews were conducted concurrently by a second examiner and lasted approximately 3 h for the OO and HFA groups and 1.5 h for the TD group. Participants received a monetary incentive for participation, even if the testing could not be completed.

Measures

ASD symptomatology was evaluated using the Autism Diagnostic Observation Schedule-Revised (ADOS; Lord et al. 2000), which assesses social and communication skills, play behaviors, and repetitive and stereotyped interests. All individuals in this sample completed Module 3. Participants' stories from the *Tuesday* book (Wiesner 1991) were used as a measure of narrative storytelling skills. Instructions for the *Tuesday* book were modified from standardized ADOS instructions, with the child completing the story rather than the examiner. The examiner gave the child the book and introduced the activity with the following prompt:

> This is a book called *Tuesday*. It has a lot of pictures, but not a lot of words. I am going to start telling the story, and then I want you to tell me the rest of the story: 'It was Tuesday evening around eight, and Mr. Turtle was sitting on a log when suddenly he heard a sound. What could it be?'

The participants were encouraged to finish telling the rest of the story, with minimal interruptions or prompting from the examiner.

Cognitive functioning was evaluated using the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler 1999) to provide a measure of nonverbal reasoning and verbal ability. Parents completed the Vineland Adaptive Behavior Scale (VABS; Sparrow et al. 1984), to assess adaptive functioning.

Narrative Analysis

The *Tuesday* narratives were transcribed from digital video using the Child Language Data Exchange System (CHILDES; MacWhinney 2000) and analyzed (details given below) for a number of characteristics that were identified as important indices of pragmatic language competence.

Narrative Length and Lexical Diversity Narrative length was measured in three ways: (1) total number of words (word tokens), (2) total number of utterances, and (3) mean length of utterance. An utterance was defined as "an independent clause and all the clauses that modify it." (Hunt 1965). The mean length of utterance in words (MLUw) was calculated by dividing *the total number of words* by the *total number of utterances*. Comments extraneous to the story ("that was cool!") and dysfluencies (repetitions, self-corrections, and filler words) were not included in the total word count. *Lexical diversity* was calculated by dividing the *number of different words* (*word types*) by *total number of words* in the narrative (*word tokens*). This is the *type/token ratio*: a larger ratio is associated with more varied language and vocabulary.

Story Elements The number of *story elements* in each narrative was counted. Story elements, or the events representing the "essential features" of the narrative, were determined by the first author prior to analyses (see "Appendix"). This enabled us to analyze how comprehensively participants described the story.

Ambiguous Pronoun References Ambiguous pronouns are pronouns for which it is unclear to whom the pronoun is referring. Reference analysis was conducted by coding the total number of pronouns and the percentage that were ambiguous.

Dysfluency The number of *repetitions*, *self-corrections*, and *fillers* were tallied. *Repetitions* were defined as the repeated use of words or parts of words with no functional purpose. Each repetition, regardless of the number of words involved, was considered one unit (e.g., "*They all*-they all were flying" was a single repetition). *Self-corrections* were operationalized as the use of words to self-correct one's speech. As above, each occurrence was counted as a single event (e.g., "*They were all*-those crows were all sitting on the wires" was one self-correction). *Filler words* were defined as words with no semantic meaning (e.g., 'uh' or 'um').

Mental State Expressions and Causal References Mental state expressions were references to mental states (e.g., 'thought', 'realized', 'didn't know') as well as emotional states ('scared,' angry,' 'excited'). A *causal reference* was defined as the number of references to a *causal connection* between events (e.g., 'the frog was mad *because* he could no longer fly').

Idiosyncratic Language and Unusual References Idiosyncratic language was defined as language that is used in an unconventional manner, such as overly formal speech (e.g., 'congregating around a human suburb'), scripted language ('stay tuned for the sequel!'), or made-up words (e.g., 'Aquamaratia Jacksonpetina Jr.'). This variable was dichotomized to "present" or "absent," as there were several children who so consistently produced idiosyncratic language that averaging the number of idiosyncratic words per group would inflate group differences. *Unusual references* were operationalized as references to people, things, or events that were not directly relevant to the story (e.g., 'Department of Homeland Security,' 'Men in Black,' 'late-night Jeopardy'). Likewise, this variable was also coded in a dichotomous manner, as the goal on a theoretical level was to examine whether the children's narratives contained *any* idiosyncratic or unusual characteristics.

Naming Characters We coded whether participants named the characters in their narratives in a dichotomous manner, as we were interested in whether or not the children followed the evaluator's lead and named the characters.

Reliability

The narratives were coded by the first author, who was naïve to group membership. Twenty percent of the narratives were transcribed by a second transcriber. Comparing each word in the two transcriptions, there was agreement with 96 % of the words from the first transcription. One-third of the narratives were coded for multiple pragmatic qualities (including number of story elements, dysfluency, mentalizing and emotion terms, and causal references) and all transcriptions for unclear pronoun use by another naïve coder. Reliability was computed for each of the coded variables. Type A intraclass correlations (ICC) were used for continuous variables, using an "absolute agreement" definition. We chose a two-way random model, with a single-measure intraclass correlation. All ICCs were >0.70, indicating "strong agreement" (Shrout and Fleiss 1979).

All coding for idiosyncratic language and unusual references were consensus coded by the first author with a second rater, and coded separately by a third rater who was naïve to group. Cohen's kappa (κ) was calculated between the consensus coding and the third rater and found to be greater than or equal to .80, indicating "almost perfect agreement" (Landis and Koch 1977).

Results

To control for narrative length, ambiguous pronouns, dysfluencies, mental state expressions, and causal references were divided by the total number of utterances. To avoid running multiple individual ANOVA's for each variable, narrative factors were grouped into seven overall clusters based on narrative characteristics: (1) narrative length/lexical diversity (word types, word tokens, number of utterances, MLU, type/token ratio); (2) story elements; (3) ambiguous pronoun use; (4) dysfluency (repetitions, self-corrections, fillers); (5) mental state/causal references; (6) idiosyncratic narrative characteristics (idiosyncratic language, unusual references); and (7) naming of story characters. A multivariate analysis of variance (MANOVA) was utilized to evaluate differences among the HFA, OO, and TD groups on the narrative clusters with continuous dependent variables (length/lexical diversity, dysfluency, and mental state/causal reference). One-way analyses of variance (ANOVAs) were performed on clusters that displayed significant group differences, as well as for story elements and ambiguous pronoun use, which did not fit into any of the clusters. Logistic regression was used to evaluate whether group membership predicted the presence of idiosyncratic narrative characteristics or character names.

Narrative Length/Lexical Diversity

There was no significant group difference for the narrative length/lexical diversity cluster, $F(10, 76) = .220$, $p = 43$; Wilks' Lambda = .945; partial eta squared = .03 (see Table 2 for *means*, *standard deviations*, and *ranges*).

Story Elements

There was a main effect for group for story elements (see Table 2 for F, p, and η^2 values). Post-hoc tests showed that TD participants produced significantly more story elements than the HFA group ($t = -2.89$, $p = .01$), whereas the OO group did not significantly differ from either the HFA or TD groups ($ps = .32$ and .82 respectively).

Ambiguous Pronoun References

All three groups produced a similar number of total pronouns (m = 19.8, 23.0, and 22.7 for the HFA, OO, and TD groups respectively, $p = .70$). The total number of ambiguous pronouns was divided by the total number of pronouns to calculate the percent of pronouns that were ambiguous. There was a main effect for group, with an average of 31, 18, and 17 % of pronouns that were ambiguous for the HFA, OO, and TD groups respectively (see Table 2 for F, p, and η^2 values). Post-hoc analyses showed that the HFA group produced a significantly larger percentage of ambiguous pronouns than either the TD ($p = .03$) or OO ($p = .02$) groups, whereas there was no significant difference between the OO and TD groups ($p = .81$).

Table 2 Narrative characteristics of HFA, OO, and TD participants

	HFA (SD) (n = 15)	OO (SD) (n = 15)	TD (SD) (n = 15)	F	*p*	η^2	Post-hoc
Total number of words (word tokens)	247.1 (83.3)	279.5 (89.2)	276.3 (113.1)				
(range)	(154–453)	(182–493)	(127–576)				
Number of different words (word types)	124.5 (34.5)	134.1 (31.2)	133.3 (40.2)				
(range)	(66–196)	(99–201)	(77–223)				
Number of utterances	27.4 (8.3)	29.2 (8.8)	28.4 (8.6)				
(range)	(14–47)	(19–56)	(14–43)				
Mean length of utterance (MLU)	9.2 (2.0)	9.6 (1.6)	9.7 (1.8)				
(range)	(5.9–11.8)	(6.9–12.4)	(6.2–13.4)				
Type/token ratio	.513 (.056)	.493 (.069)	.501 (.056)				
(range)	(.43–.62)	(.37–.61)	(.39–.61)				
Story elements	16.6 (3.7)	18.4 (2.6)	19.9 (2.5)	4.74	.01	.18	HFA < TD
(range)	(11–25)	(15–24)	(15–24)				OO = TD
							OO = HFA
Ambiguous pronouns ([a])	31 % (16.6 %)	18 % (10 %)	17 % (15 %)	4.21	.02	.17	HFA > TD
(range)	(8–70 %)	(6–40 %)	(0–50 %)				HFA > OO
							OO = TD
Repetitions	.33 (.39)	.14 (.14)	.11 (.14)	3.19	.05	.13	HFA > TD
(range)	(0–1.3)	(0–.48)	(0–.44)				OO = TD
							OO = HFA
Self-corrections	.19 (.16)	.24 (.16)	.10 (.10)	3.76	.03	.15	HFA > TD
(range)	(0–.71)	(.06–.67)	(0–.35)				OO > TD
							OO = HFA
Filler words	.04 (.13)	.15 (.20)	.10 (.12)	1.69	.20	.07	
(range)	(0–.52)	(0–.74)	(0–.39)				
Mental state expressions	.17 (.10)	.20 (.10)	.17 (.12)				
(range)	(.04–.37)	(0–.32)	(.04–.38)				
Causal references	.03 (.04)	.05 (.06)	.03 (.06)				
(range)	(0–.13)	(0–.21)	(0–.18)				

[a] Percentage of pronouns that were ambiguous

Dysfluency

There was a statistically significant difference between groups for the dysfluency cluster (repetitions, self-corrections, fillers), F(6, 80) = 2.89, $p = .01$; Wilks' Lambda = .675; partial eta squared = .18. When the results for the dependent variables were considered separately, the factors that showed a main effect for group were repetitions and self-corrections (see Table 2 for F, p, and η^2 values). Specifically, the HFA group produced significantly more repetitions than the TD group ($t = 2.01$, $p = .05$), whereas the OO group did not significantly differ from either the HFA ($p = .54$) or TD groups ($p = .10$). Both the HFA ($p = .05$) and OO ($p < .01$) groups made significantly more self-corrections than the TD group, whereas there was no significant difference between the HFA and OO groups ($p = .45$). There was no group difference in use of filler words (see Table 2 for F, p, and η^2 values).

Mental State/Causal References

There was no significant group difference for the mental state/causal references cluster, F(4, 82) = .414, $p = .80$; Wilks' Lambda = .961; partial eta squared = .020 (see Table 2 for *means*, *standard deviations*, and *ranges*).

Narrative Analysis

In the above analyses, we did not covary for VIQ, following the argument by Dennis et al. (2009) that covarying for IQ could produce overcorrected and anomalous findings in studies of neurodevelopmental disorders. However, when we did run the analyses described covarying for IQ, our findings were largely similar, with the one exception that no significant group difference was found for number of repetitions among the groups. We also did not make Bonferroni corrections for the number of comparisons, as

the main thrust of this project is to explore whether the OO group has become indistinguishable in their functioning from the TD group; therefore, we considered it most conservative to probe the data for even small differences between these groups. Correcting for multiple comparisons using the Bonferroni procedure provides a reduction of false positives only, when in the present context, the goal is to reduce the risk of false negatives. Therefore, we adopted the approach of clustering data in composite variables where possible (Perneger 1998). However, when we assessed the effect of Bonferroni corrections, the results were unchanged, with the exception that the difference in self-corrections between TD and HFA was no longer significant ($p = .23$).

Idiosyncratic Narrative Characteristics

The logistic regression model was significant, $\chi^2 = (2, N = 45) = 9.69, p < .01$, indicating that the model was able to distinguish between participants who did and did not produce narratives with idiosyncratic characteristics (idiosyncratic language or unusual references). More specifically, 10 out of 15 HFA, 7 out of 15 OO, and 2 out of 15 TD participants produced narratives with idiosyncratic characteristics. Further analysis was conducted on those who produced idiosyncratic language and those who made unusual references. The logistic regression model for idiosyncratic language was significant, $\chi^2 = (2, N = 45) = 10.85, p < .01$, indicating the model was able to distinguish between participants who did and did not produce idiosyncratic language in their narratives. The model explained between 21.4 % (Cox and Snell R square) and 29.4 % (Nagerlkerke R squared) of the variance and correctly classified 71.1 % of the cases. The HFA group was 21 times more likely than the TD group to have idiosyncratic language in their narratives ($p < .01$), and the OO group was 9.33 more likely than the TD group to have idiosyncratic language ($p = .05$). There was no significant difference between the OO and HFA groups ($p = .29$). Group membership did not predict whether the participants introduced odd specific references not apparent from the pictures ("unusual references"), $\chi^2 = (2, N = 45) = 3.83, p = .15$ (Table 3).

Table 3 Number of people who produced idiosyncratic language, made unusual references, and named the characters in their narratives

	HFA (SD) (n = 15)	OO (SD) (n = 15)	TD (SD) (n = 15)	
Idiosyncratic language	9/15[a]	6/15[b]	1/15[a,b]	HFA > TD OO > TD OO = HFA
Unusual references	5/15	4/15	1/15	
Named characters	2/15[a]	6/15	11/15[a]	HFA < TD OO = TD OO = HFA

[a] $p < .01$

[b] $p = .05$

Naming Characters

The logistic regression model was significant, $\chi^2 = (2, N = 45) = 11.92, p < .01$, indicating that it was possible to distinguish between participants who did and did not name the characters. The odds ratio between the TD and HFA groups was 17.88, suggesting that the TD group was almost 18 times more likely to name the characters in their narratives than the HFA group ($p < .01$). The OO group did not differ from either the HFA ($p = .11$) or TD ($p = .07$) groups.

Discussion

This study examined the narrative performance of a group of children and adolescents with a previous diagnosis of an ASD who no longer meet diagnostic criteria for an ASD (optimal outcome, or OO), a group of well-defined high-functioning children with an ASD (HFA), and their typically developing (TD) peers. In a previous study, Kelley et al. (2010) found that OO and TD performance did not significantly differ in standardized language measures. However, because standardized testing may not reveal subtle language deficits, spontaneous narratives were analyzed for potentially subtle aspects of pragmatic language.

As predicted, the HFA, OO and TD groups did not differ with respect to narrative length and lexical diversity. All three groups had well-developed morphological and syntactic language skills. Furthermore, all groups produced narratives of similar sophistication in many respects; thus, consistent with previous research, there were no differences in the number of references made to characters' emotions and cognitive states, or in the total number of causal references made in the narratives. This latter finding reflects the fact that there were few causal references in general: across groups, there was an average of zero to one causal reference per narrative. This floor effect prevented an exploration of causal attribution of characters' emotional and cognitive states, which was previously found to occur less frequently in the narratives of children with an ASD (Beaumont and Newcombe 2006; Capps et al. 2000).

There were two language domains for which both OO and HFA groups showed deficits relative to TD peers: self-correction dysfluency, and idiosyncratic language. Lake et al. (2011) have postulated that self-corrections are for

the benefit of the listener, or *listener-oriented*. However, whereas they found that TD adults produced more self-corrections than adults with an ASD, TD participants in the current study produced *fewer* self-corrections than either HFA or OO groups. There are several possible interpretations of this finding: self-corrections reflect general language ability rather than pragmatic deficits (Sherratt 2007); repetitions and self-corrections reflect greater impulsivity and deficits in other executive processes [a profile consistent with case studies that found attention deficits in OO children (Fein et al. 2005)]; or, a combination of the two, where the OO and HFA groups have more opportunities to self-correct because of higher-level language and executive deficits but are socially adept enough to notice they are not being clear and correct themselves for the benefit of the listener. If so, this is a promising finding, as it shows that some OO and HFA individuals develop these higher-level social skills as they grow older.

The OO group was 9 times more likely to produce narratives with idiosyncratic language than the TD group, whereas the HFA group was 21 times more likely to do so relative to their TD peers. Idiosyncratic language potentially reflects a reduced familiarity with conventional ways of speaking. It should be noted that a qualitative analysis of idiosyncratic language indicated similarities in quality for OO and HFA participants (e.g., a reference to a "human resistance team" by an OO child or "human suburb" from an HFA participant). OO individuals also produced overly formal language (e.g., "excited by this new phenomenon"), neologisms (e.g., "electronical wires") or scripted speech (e.g., "stay tuned for the sequel!"). Furthermore, although there was no significant group difference for unusual references, a qualitative examination found that a subset of OO and HFA children and adolescents produced unusual references, including references to movies and TV shows such as "A Bug's Life," "Jeopardy" and "Men in Black," and references to specific institutions such as "the *Department of Homeland Security*;" only one TD participant made such reference. These findings suggest that although individuals in the OO group no longer meet criteria for an ASD, a significant minority (7 out of 15 OO participants) continue to produce narratives with idiosyncratic characteristics. The converse should also be noted, however, in that 8 out of 15 OO children (and 5 out of 15 HFA children) produced narratives without *any* idiosyncratic characteristics.

There were four analyses in which the HFA group differed significantly from their TD peers, though the OO group did not: (1) inclusion of central story elements, (2) repetition dysfluency, (3) ambiguous pronoun use, and (4) naming of story characters. These findings are largely consistent with previous studies. First, the HFA participants in our study were more likely to stray from the story as shown in the picture book and instead tell stories containing extraneous aspects and themes, suggesting they were more likely to incorporate personal interests or to misinterpret elements in the book (e.g. mistake the grandmother as being awake when she was sleeping (Barnes and Baron-Cohen 2012; Losh and Capps 2003). Second, the HFA individuals produced more repetition dysfluencies. Prior research has established the role of multiple factors in speech dysfluency, including syntactic complexity (Ratner and Sih 1987), individual differences in inhibitory control (Engelhardt et al. 2013), and planning difficulty (Bortfeld et al. 2001). Further analyses of this dataset are underway; for now, we conjecture that the group differences in dysfluency observed in this study could reflect group differences in the efficiency of processing the demands of the syntactic planning required by this narrative task. Third, children and adolescents with HFA included significantly more pronouns for which the antecedent was ambiguous, a potentially salient and subtle marker of pragmatic language deficits. Fourth, TD and OO participants were more likely to name the characters in the story, an approach that can heighten listener engagement. In contrast, HFA individuals were more likely to refer to the characters as "the man" or "the human," an approach that is more formal and distancing (Losh and Capps 2003). To our knowledge, no study to date has evaluated whether participants with an ASD adopt an examiner's name for a character, or follow the examiner's cue and name the other characters in a story.

Limitations and Directions for Future Research

While a strength of the study was that the participants told the story while looking at the book, thus limiting working memory demands, because the child did not first get to explore the book, and the story unfolds at the same time the teller describes it for the listener, we were not able to assess how the narrator would have structured a story were he familiar with it before telling it. This prevented an evaluation of socio-cognitive factors such as how a child may organize a story around a pivotal event for the benefit of the listener (Goldman 2008), an important area for future study. Furthermore, while evaluation of a story provides important consistency and permits comparison across narrations, the structure inherent in a story limited our ability to note pragmatic deficits that may manifest in less-structured settings, such as when recounting personal events, or during a conversation with peers. We hypothesize that the HFA group benefits most from structured interaction with an adult, and that a less structured task with peers may better reveal pragmatic difficulties. Additionally, we were not able to assess pre-morbid language functioning and did

not specifically evaluate the relationship between different language skills (e.g., receptive, expressive language) and narrative production.

There are also sample characteristics that could serve as limitations. First is the small sample size of 15 participants per group and the relatively large number of narrative comparisons. We addressed this by grouping narrative characteristics into clusters when theoretically indicated. While most of our significant findings produced medium to large effect sizes, decreasing the likelihood of a Type I error, we may not have had enough power to detect group differences for fillers, which had a medium effect size, with the trend that OO and TD produced more fillers than HFA. Additionally, a larger OO sample would allow a researcher to subgroup OO individuals by evaluating their pattern of strengths and weaknesses.

Furthermore, although we rigorously checked that each OO child received a prior diagnosis of an ASD from a specialist in ASDs and evaluated blinded reports to confirm diagnosis, not all children were given ADOSes or ADI-Rs at the time of their initial diagnosis. Moreover, while we used ADOSes, we did not use ADI-Rs to confirm current diagnostic status (HFA, OO, or TD). Additionally, experimenters were not blind to group membership, as they were also involved in the recruitment process.

Strengths of the study are, first, that the OO, HFA, and TD groups were very well characterized and defined. Furthermore, the OO, HFA and TD groups were high functioning, such that verbal IQ was generally in the average range or above, allowing for a cleaner investigation of how pragmatic and language factors (rather than cognitive factors) impact the quality of a narrative.

Furthermore, few studies have examined narrative abilities of high-functioning children and adolescents with an ASD, and only one other study (Kelley et al. 2006) has been conducted on narrative abilities in OO children. Therefore, this study adds to our understanding of the pragmatic functioning of a high-functioning group of children and adolescents with an ASD, as well as of a strictly-defined group of children and adolescents who had a former diagnosis of an ASD but no longer meet diagnostic criteria for an ASD.

Implications for Treatment

We found that OO children have few residual pragmatic and language deficits as measured by their production of narratives. They were indistinguishable from their TD peers even in subtle pragmatic language characteristics such as ambiguous pronoun use. However, OO individuals produced stories that contained significantly more self-correction dysfluencies and were more likely to contain idiosyncratic language than the stories of TD peers. There were also variables (story elements, repetition dysfluency) in which, although the OO group did not significantly differ from their TD peers, they also did not differ from their HFA peers: their performance fell between the two groups. It is unclear how these findings will manifest in day-to-day life. Idiosyncratic language could be interpreted as distancing and formal; or, OO children and adolescents may rely on adaptive compensatory strategies, such as humor, and be more likely to be perceived as "quirky" or "creative" rather than "odd." Future research should explore whether these factors impact the daily functioning of these OO individuals, including how OO children are perceived by peers and the quality of their peer relationships. It is possible that the differences as described in this study may not even rise to the standard of "deficits," as it is not known whether these discourse characteristics are socially impairing.

In contrast, HFA group differences were more pronounced than those in the OO group in this study. In addition to more idiosyncratic language and self-correction dysfluencies, children with HFA also produced more repetition dysfluencies, paid less attention to the cues in the book and incorporated fewer "story elements," produced more ambiguous pronouns, and were less likely to name the characters in their stories, indicating continued deficits in pragmatic functioning despite verbal IQ in the average range. This also suggests that these language factors are the aspects of language either most resistant to remediation or least likely to be targeted for intervention. Therefore, there is a need for abundant opportunities for practice of narrative production, with feedback. This includes encouraging clarity of pronoun use, limiting references to personal interests, fostering an understanding and inclusion of central elements in stories, and explicitly teaching story grammar skills (e.g., Petersen et al. 2010). Fostering these skills could facilitate more effective communication and socialization with others.

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Appendix: Coding of Story Elements

Page 1

1. Something about Mr. Turtle (how he feels, what he does)
2. Frogs flying on lily pads

Page 2

3. Frogs doing tricks/having fun on lily pads
4. Frogs scaring/chasing birds

Page 3

5. Frogs flying/floating (toward houses, etc.)

Page 4

6. Man eating a sandwich
7. Man sees frog
8. Frog waves at man

Page 5

9. Frog flying into clothesline

Page 6

10. Frog with cape
11. Frogs flying through window
12. Frogs flying through chimney

Page 7

13. Grandmother sleeping
14. Frogs watching TV
15. A frog changing the channel with his tongue

Page 8

16. A frog flying
17. A dog chasing the frog

Page 9

18. Frogs chasing the dog

Page 10

19. Frogs and lily pads falling/frogs landing on houses

Page 11

20. Frogs fall
21. Frogs are back in the water

Page 12

22. Detective investigating/trying to figure out what happened
23. Dog sniffing lily pad (or mention of the dog)
24. Mention of police, ambulance, other dogs.
25. Man telling the newswoman what had happened

Page 13

26. Shadow by the barn
27. The sun is setting

Page 14

28. Pigs are flying

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