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ORIGINAL PAPER



A Comprehensive Examination of Reading Heterogeneity in Students with High Functioning Autism: Distinct Reading Profiles and Their Relation to Autism Symptom Severity

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Abstract The goal of this study was to identify unique profiles of readers in a sample of 8-16 year olds with higher functioning autism spectrum disorders (HFASD) and examine the profiles in relation to ASD symptom severity. Eighty-one students were assessed utilizing a comprehensive reading battery that included basic word reading, language, and comprehension. Using Latent Profile Analysis, four empirically distinct profiles of readers emerged. Next, using the Autism Diagnostic Observation Schedule, Second Edition (Lord et al., Autism diagnostic observation schedule, 2nd edn, Western Psychological Services, Torrance, CA, 2012), analyses were conducted to determine if significant differences existed between profiles as a result of ASD symptomatology. Findings demonstrate the heterogeneous nature of reading profiles in students with HFASD and significant differences between the reading profiles and ASD symptom severity.

Keywords Reading · Language · Reading profiles · Higher functioning autism

Introduction

Current science emphasizes the need to not only distinguish groups of children affected by autism spectrum disorders (ASD) from comparison children but also to investigate

Nancy S. McIntyre nsmcintyre@ucdavis.edu the nature of heterogeneity in development among children affected by ASD (Georgiades et al. 2013; Happé et al. 2006). The expectation and need to understand psychological heterogeneity is especially true for older children because as cognitive, language, and emotional processes develop and differentiate with age it is likely that there are greater degrees of freedom for the expression of heterogeneity. There are many approaches to studying heterogeneity in ASD including, for example, the study of differences in social engagement (Wing and Gould 1979), differences in social attention (Rice et al. 2012), differences in executive function (Tager-Flusberg and Joseph 2003; Geurts et al. 2004), sensory processing (Lane et al. 2010), and language (Pickles et al. 2014; Rapin et al. 2009; Tager-Flusberg 2006), as well as multidimensional approaches (Beglinger and Smith 2001; Insel 2014).

There are many reasons why understanding heterogeneity in ASD is of great interest. One of these is that the identification of valid subgroups holds the promise of enabling a more precise alignment of treatments and educational plans for affected individuals with ASD (Beglinger and Smith 2005; Miles et al. 2005). This is an especially important consideration for school-aged children who begin to experience their kindergarten through 12th grade classrooms as their primary venue for intervention. Currently we have very little information on the heterogeneity of ASD that informs and advances contemporary educational practices for elementary and secondary students with ASD (Dingfelder and Mandell 2011; Kasari and Smith 2013; Machalicek et al. 2008). This is particularly true for higher functioning children for whom very little evidencebased information is available to guide optimal education in inclusive regular education classrooms (Machalicek et al. 2008). This is unfortunate for two reasons.

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First, current epidemiological data not only indicate that 1 in 66 children in second grade throughout the nation are affected by ASD, but 47% of these children have average to above average intellectual ability and 25% have borderline IQ. Only 28% of these second grade children are affected by intellectual disabilities (Christensen 2016). Many school-aged children with ASD function in a range of intelligence that allows them to receive their education in regular education classrooms (de Bruin et al. 2013; Fleury et al. 2014), which would suggest that these students should gain benefit from general education instruction, with individualized special education supports.

Second, although children with higher functioning ASD (HFASD) are capable of receiving their education in regular education classrooms, they are at risk for academic difficulties. One specific difficulty that has been empirically demonstrated in the literature is in reading; these difficulties share similarities to students identified as reading disabled. Multiple studies report symptomatology similar to reading comprehension disability, or individuals who are able to fluently decode words, yet have difficulties understanding the meaning of written text, in 33-65% of the samples (Estes et al. 2011; Jones et al. 2009; Nation et al. 2006; Norbury and Nation 2011; Ricketts et al. 2013). Furthermore, poor reading abilities have been shown to be substantially discrepant from IQ in many children, leading to the concern that many students are underachieving (Jones et al. 2009). This provides evidence that current school-based reading instruction does not sufficiently prevent negative reading comprehension outcomes for children with HFASD. Less is known about the development of basic word recognition skills in students with HFASD. Some studies have suggested that word decoding or word reading is not a specific deficit that is more prevalent in this population of students (e.g. Brown et al. 2013), however, it has not been thoroughly examined with a comprehensive word recognition skills battery in large samples.

With a deeper, evidence-based understanding of the nature and variability of reading difficulties, or disturbances, in school-aged children with HFASD, beyond what is already known about reading comprehension disability, and including the development of word recognition, it may be possible to develop more targeted methods of instruction for this population (Reutebuch et al. 2015). In this study, we extend the current knowledge about the reading development and disability in students with HFASD in three ways. First, we provide a deeper understanding of reading impairments in this sample, beyond reading comprehension difficulties, with a close examination of basic reading development. Second, we attempt to determine if meaningful subgroups or profiles of reading strengths and weaknesses can be identified in a sample of students with HFASD.

Third, we investigate the relation between HFASD reading subgroups and ASD symptomatology.

Development of Reading: Subcomponent Skills and Profiles of ASD Struggling Readers

Reading for meaning develops over time and builds upon two brain regions already present in infancy: the visual object recognition and oral language systems (Dehaene 2009). By the time children are 5 or 6 years old, key visual recognition processes are well developed but still maximally plastic. Children's vocabulary grows 10-20 words per day by the end of their second year, and by the time they are 6 years old, most have expert knowledge of phonology, basic grammar rules, and a vocabulary of several thousand words (Dehaene 2009). In the phonological stage of reading (Frith 1985), children develop letter-sound correspondence requiring proficient letter recognition skills and phonological awareness, or the ability to discern individual speech sounds. They learn to decode words, progressing from the simple to the complex. Morphemic awareness develops as well and children learn that prefixes, root words, and suffixes are associated with pronunciation and meaning. In the orthographic stage (Frith 1985), the lexical pathway used to identify words by sight develops and progressively supplements the decoding/phonological pathway. Oral language processing creates meaning from the words. These two processes may develop relatively independently (Adlof et al. 2010) and the relationship between these factors and reading comprehension changes over time. Cain and Oakhill (2008) noted that for younger children decoding is more important and the correlation between reading and listening comprehension is low. By high school however, decoding differences are generally small and the correlation between reading and listening comprehension is high. Therefore, they posited that based on this model one should expect a person's reading comprehension to develop to the same level as their listening comprehension once word reading is fluent.

The Simple View of Reading describes successful reading comprehension as the result of sufficient decoding and linguistic comprehension skills (Gough and Tunmer 1986). These two component skills are described as multiplicative in nature and therefore both sets of skills must be operating sufficiently for successful reading comprehension. The Component Model of Reading (Joshi and Aaron 2000), based upon the Simple View, incorporated processing speed, as measured by speed of letter naming, as a third predictor of reading comprehension. Prior studies have used the Component Model to investigate how poor readers may fall into subgroups that differ across the components of word decoding and linguistic comprehension (Aaron 1997; Catts et al. 2003; Catts and Kamhi 1999; Gough and Tunmer 1986). At least three subgroups of poor readers are predicted by this model: (a) poor readers with word recognition problems only (e.g. dyslexics), (b) poor readers with linguistic comprehension problems only (e.g. poor comprehenders and/or hyperlexics), and (c) poor readers with difficulties in both components [e.g. garden variety poor readers (Gough and Tunmer 1986), mixed reading disabled (Catts and Kamhi 2005), or language-learning disabled (LLD; Berninger and May 2011)]. Furthermore, Aaron et al. (2008) demonstrated the utility of the Component Model in identifying the facet(s) of reading that is(are) the source of a child's reading difficulty in order to better target intervention efforts.

A deeper examination of the two component skills, word recognition and linguistic comprehension, reveals many essential sub skills. For example, poor readers who struggle with accurate lower-level word recognition skills typically demonstrate deficits in phonological processing, or the processing of speech sounds. There is empirical evidence that demonstrates a correlational relation between facets of phonological processing and word recognition including: phonological awareness (Bradley and Bryant 1983; Goswami and Bryant 1990; Share 1995; Swanson et al. 2003; Wagner et al. 1994), phonological decoding (Rastle and Coltheart 1998; Rey et al. 2000; Swanson et al. 2003), and rapid automatized naming (Kirby et al. 2003; Manis et al. 2000; Swanson et al. 2003). Vocabulary development has also been linked with word recognition skill (Biemiller 2007; Biemiller and Boote 2006; Chiappe et al. 2004; Nation 2009; National Reading Panel 2000; Ouellette and Beers 2010: Perfetti 2007).

Linguistic comprehension, or the oral language processing that creates meaning from words, has a profound effect on the comprehension of written texts (e.g., Nation and Snowling 2004; Roth et al. 2002). Empirical evidence suggests that higher-level linguistic comprehension skills are underpinned by the depth and breadth of one's vocabulary (Ouellette and Beers 2010; Perfetti 2007; Ricketts et al. 2007; Roth et al. 2002; Sénéchal et al. 2006), syntax and grammar (Cain and Oakhill 2006; Muter et al. 2004; Nation et al. 2004), verbal reasoning and integration of background knowledge during reading to generate inferences (Hannon and Daneman 2001; Long and Lea 2005; McNamara 2001), and narrative recall (Fuchs et al. 1988; Leslie and Caldwell 2009). All of these facets of oral language support the construction of a globally coherent situation model of a text; semantic, grammatical and syntactic information provide the foundation of the text-based mental model, then continuous connections between prior knowledge, inferences, and text ideas are made to create the situation model required for proficient reading comprehension (Kintsch 1988; Van Dijk and Kintsch 1983).

Empirical evidence exists to demonstrate that both word recognition and linguistic comprehension account for substantial unique variance in reading comprehension for children with ASD, supporting the Simple View in this population (Jones et al. 2009; Lindgren et al. 2009; Nation et al. 2006; Norbury and Nation 2011; Ricketts et al. 2013). Similar to the subtypes described by Catts et al. (2003) with a typically developing reading sample, many samples with ASD have displayed profiles comparable to poor comprehenders, or hyperlexics, who demonstrate adequate word decoding alongside poor language and reading comprehension (e.g. Brown et al. 2013; Jones et al. 2009; Nation et al. 2006; Newman et al. 2007; Huemer and Mann 2010; Wei et al. 2015; Zuccarello et al. 2015).

Other researchers have reported evidence that subgroups of children with poor comprehension have significant concomitant lower-level phonological, rapid naming, and/or word decoding deficits (Åsberg and Dahlgren Sandberg 2012; Nation et al. 2006; White et al. 2006). White et al. (2006) found that, similar to those with typical development, phonological skills were a strong predictor of word recognition and spelling in 8-12 year-olds with ASD. However, Gabig (2010) found that while phonological awareness was delayed in development for 5-7 year olds with ASD in their sample, it was not significantly related to word reading or decoding, but it was significantly correlated with receptive vocabulary. Similarly, in several other studies, word recognition skills have been shown to correlate with language abilities, reporting subgroups of children with poor word recognition associated with poor oral language skills (e.g. Brown et al. 2013; Jacobs and Richdale 2013; Lindgren et al. 2009; Nation et al. 2006; Norbury and Nation 2011; Ricketts et al. 2013). Three studies have reported that children with ASD who have age-appropriate language skills scored significantly higher than those with language impairments on standardized measures of reading comprehension, word recognition, and decoding (Lindgren et al. 2009; Lucas and Norbury 2014; Norbury and Nation 2011). None of the studies reviewed reported a subgroup of children displaying a dyslexic profile, or one in which impaired word recognition is concomitant with proficient linguistic comprehension.

Several studies have placed emphasis on exploring higher-level linguistic comprehension factors in more detail. For example, participants with ASD have been shown to have difficulty integrating background knowledge and inferred knowledge explicitly with global text (Saldaña and Frith 2007), using background knowledge to interpret and remember specific information or resolve ambiguities in discourse (Wahlberg and Magliano 2004), or responding to questions about inferred emotions (Tirado and Saldaña 2016). Language impairment in adolescents with ASD was associated with poorer performance on a passage-level inference measure (Norbury and Nation 2011), and in elementary school-aged children verbal ability was the strongest predictor of performance on inferential reading comprehension questions (Lucas and Norbury 2015). Norbury and Nation (2011) suggested that difficulties integrating information from different sources for global coherence and inference generation might be highly dependent on variance in the language skills of students with ASD.

Reading Comprehension and ASD Symptomatology

Reading for meaning is fundamental for accessing social, cultural, and political milieus through written documents, and is a cognitively complex process. Current research suggests that the risk for reading comprehension disability may be related to ASD symptomatology and be a component of the social-communicative and cognitive phenotype of school-aged children with ASD; several studies have reported significant associations between individual differences in reading development and diagnostic status, social functioning, or autistic symptom severity in samples of school-aged children with ASD (Åsberg et al. 2010; Estes et al. 2011; Jones et al. 2009; Norbury and Nation 2011; Ricketts et al. 2013). Reading is a written form of communication between the author and the reader, and as such, is likely to be impacted by deficits in social communication abilities such as understanding an author's intentions or purpose for writing a text, which impedes learning from the text. Furthermore, impairments in social communication skills may impact reading comprehension through impeding the development of rich networks of semantic and episodic knowledge typically developed through sociallymediated learning. Additionally, challenges in understanding social norms may lead to difficulty developing skills that rely on social knowledge such as understanding characters' intentions, inference generation, and understanding of narrative elements.

The cognitive characteristics of many children with ASD include the tendency to focus on details rather than global meaning (Booth and Happé 2010), leading to particular problems generating global coherence or processing at the gist level across a text (e.g., Pellicano 2010), which in turn leads to difficulty recalling, retelling, and comprehending stories (Diehl et al. 2006; Williams et al. 2006). Furthermore, this local processing bias, or weak central coherence (Happé and Frith 2006), has been posited to lead to particular difficulty integrating information both from the text and from background knowledge for inference generation (Norbury and Nation 2011) and global comprehension (Ricketts et al. 2013). Another cognitive characteristic associated with ASD is the tendency to have restricted or fixated interests, and this can limit exposure to situations where individuals learn about a wide variety of topics and develop oral language skills across multiple contexts. This restricts vocabulary growth except in fields of specific interests, and leads to more literal, less flexible understanding of words and phrases. Combined, these difficulties constrain creation of a coherent mental model of text that draws on a reader's ability to combine text-based information with relevant background knowledge to generate inferences about things not explicitly stated in the text (Kintsch 1988; McNamara 2001). Overall, it may be that the severity of the social communication and ASD-specific cognitive difficulties align with the severity of reading comprehension deficits for many children affected by ASD.

In summary, there have been several attempts to unpack the relation between ASD symptomatology, language, and reading performance. The existing empirical literature suggests individuals with ASD have particular difficulties with reading comprehension and those difficulties may be associated with both language and the symptom severity of individuals with ASD. The extant data also presents some evidence that profiles of struggling readers exist similar to those seen in typically developing populations, with perhaps the exception of a reading profile that exhibits a dyslexic profile of poor word recognition alongside discrepantly proficient linguistic comprehension. However, very few studies of reading in ASD have utilized comprehensive reading batteries that assess the Simple View of Reading and include the key sub skills supporting linguistic comprehension and word recognition. Therefore, we still know very little about the potential difficulties individuals with ASD have on tasks related to word recognition and how these lower level reading variables interact with linguistic and reading comprehension. Furthermore, measurement of reading ability varies depending on the assessments used (Cutting and Scarborough 2006), and different findings from previous studies could be the result of using different reading measures. Therefore, use of multiple measures of each component of reading would be beneficial.

Current Study

Previous research has started to investigate reading development in school-aged children with ASD. To our knowledge, no studies have analyzed subgroups, or distinct profiles, of school-aged readers with ASD using an extensive reading battery that includes the sub skills of lower-level word recognition abilities, including phonological processing and processing speed measures, and higher-level linguistic and reading comprehension skills. In the current study, a comprehensive reading and language assessment battery was collected with school-aged children with HFASD to answer the following research questions: (a) do individuals with HFASD exhibit distinct reading profiles? (b) how do distinct reading subgroup profiles relate to ASD symptom severity? Based on previous literature, we hypothesize that HFASD readers are heterogeneous in nature, with relative strengths and weaknesses, therefore distinct profiles will emerge. We also hypothesize that similar to previous research, individuals with HFASD who demonstrate more severe reading difficulties will also display more severe ASD symptomatology.

Method

Participants

This research was conducted in compliance with the Institutional Review Board and written parental consent and child assent was obtained prior to data collection. Participants were 81 (66 male) children, aged 8-16 years, who had a community diagnosis of ASD (see descriptive statistics in Table 1). Enrolled subjects were recruited from the local community through school districts, a university research subject tracking system, and word of mouth. Individuals were included in the HFASD sample if they had a community diagnosis of ASD that was confirmed by trained researchers using the Autism Diagnostic Observation Schedule, Second Edition (ADOS-2; Lord et al. 2012), and if they had had a full-scale IQ (FIQ) estimate \geq 75 as measured on the Wechsler Abbreviated Scales of Intelligence-II (WASI-2, Wechsler 2011). A total of 93 individuals with ASD were recruited for this study; they all met criteria for ASD on the ADOS, but 12 individuals were ineligible for the study due to FIQ <75. Exclusionary criteria included an identified syndrome other than ASD or ADHD (e.g. Fragile X), significant sensory or motor impairment (e.g. visual impairments), a neurological disorder (e.g. epilepsy, cerebral palsy), psychotic symptoms (e.g. hallucinations or delusions), or any major medical disorder that could be associated with extended absences from school. Twentyeight percent of the children with HFASD also met criteria for ADHD according to parent report of a community diagnosis. Most of the children in this sample spent much, or all, of their school day in a general education classroom setting: 65% were in general education 81-100% of the day, 12% were in general education 41-80% of the day, 10% were in general education 1-40% of the day, 10% were not in general education at all, and three percent did not report placement. Eighty-four percent of the children attended public schools, and 91% had an IEP or 504 Plan.

Measures and Procedures

Data reported are from assessment sessions that were conducted by members of a trained research group in a university-based child assessment laboratory over two 2.5-h sessions.

Diagnostic Measures and Sample Description

The ADOS-2 (Lord et al. 2012) is a semi-structured diagnostic assessment shown to have strong predictive validity compared to best estimate clinical diagnoses (Charman and Gotham 2013). Scores were utilized to both confirm ASD diagnosis and as a distal measure in the second research question. Modules 3 and 4 were administered, providing scores for Social Affect (SA) and Restricted and Repetitive Behavior (RRB). The Module 3 vielded a raw subscore for SA and for RRB that combined to create the Total Score. Intraclass correlations for interrater reliability for Module 3 were reported as 0.92 for SA, 0.91 for RRB and 0.94 for overall total raw score. Intraclass correlations for interrater reliability for Module 4 were reported to be 0.93 for Social Interaction, 0.84 for Communication, 0.92 for Communication+Social Interaction, and 0.82 for Stereotyped Behaviors and Restricted Interests (Lord et al. 2012). Module 4 scores were converted via the modified Module 4 algorithm per Hus and Lord (2014).

IQ

The WASI-2 (Wechsler 2011) provided an estimate of verbal and nonverbal cognitive ability. Two verbal subtests, Vocabulary and Similarities, measured expressive vocabulary and abstract semantic reasoning and formed the verbal composite (VIQ). Two nonverbal subtests, Block Design and Matrix Reasoning, measured spatial perception, visual abstract processing and problem solving with motor and non-motor involvement and formed the performance composite (PIQ). Combined, the four subtests yielded an agenormed standard score measurement of full-scale IQ (FIQ). The FIQ index has established internal consistency (0.96)and test-retest reliability for children ages 6-16, r=0.94(Wechsler 2011). In this sample, internal consistency Cronbach's alpha coefficients were 0.89 for Vocabulary; 0.88 for Similarities; 0.87 for Block Design; and 0.92 for Matrix Reasoning.

Phonological Processing and Rapid Automatized Naming

The Elision and Nonword Repetition (NWR) subtests were administered from the Comprehensive Test of Phonological Processing, Second Version (CTOPP-2; Wagner et al. 1999) that yielded age-normed scaled scores (M=10, SD=3) measuring phonological awareness (PA) and expressive phonology/phonological memory respectively. The internal consistency Cronbach's alpha coefficient from our sample for Elision (alpha=0.93) was consistent with publisher

reported alphas (alphas = 0.81-0.91; Wagner et al. 1999). The internal consistency Cronbach's alpha coefficient from our sample for NWR (alpha=0.78) was consistent with publisher reported alphas (alphas=0.73-0.80). The speed at which participants were able to connect orthographic and phonological representations was measured using two rapid automatized naming (RAN) tasks from the CTOPP-2; Rapid Letter Naming and Rapid Digit Naming subtests yielded separate age-normed scaled scores (M=10, SD=3), and combined for an age-normed RAN index score (M=100, SD=15). Alternate-form reliability coefficients from our sample for Rapid Letter Naming (0.87) were consistent with publisher reported alternate-form reliability coefficients (0.70-0.93).

Word Recognition

The Test of Word Reading Efficiency, Second Edition (TOWRE-2, Torgesen et al. 2012) provided an age-normed standard score (M = 100, SD = 15) measuring accuracy and fluency of sight word recognition (Sight Word Efficiency: SWE) and phonemic decoding (Phonemic Decoding Efficiency: PDE). Participants read as many real words (SWE) or decodable nonwords (PDE) as they were able to in 45 s per subtest. Internal consistency Cronbach's alpha coefficients from our sample for SWE (alpha=0.97), and PDE (alpha=0.87) were generally consistent with publisher reported alphas for both subtests (alphas > 0.90; Torgesen et al. 2012). Text-level reading accuracy was assessed with age-normed scaled scores (M=10, SD=3) from the Gray Oral Reading Tests-Fifth Edition (GORT-5; Wiederholt and Bryant 2012). Publisher (Wiederholt and Bryant 2012) reported Cronbach's alpha coefficients for GORT-5 Accuracy scores ranged between 0.85 and 0.94 in the normative sample, and 0.93 in an ASD subgroup.

Linguistic Comprehension

The Recalling Sentences subtest from the Clinical Evaluation of Language Fundamentals, Fourth Edition (CELF-4; Semel et al. 2003) provided an age-normed scaled score (M=10, SD=3) assessing sentence-level semantic and syntactic expressive language skills. In order to accurately recall increasingly longer and more complex sentences, one must strategically utilize language structure (e.g., syntax) and meaning. Publisher (Semel et al. 2003) reported Cronbach's alpha reliability coefficients ranged from 0.86–0.93 in the normative sample and 0.97 in an ASD subsample. Expressive vocabulary was measured with the Vocabulary subtest from the WASI-II (Wechsler 2011), which yielded an age-normed T-score (M=50, SD=10). This subtest was designed to measure verbal concept formation and semantic knowledge by asking the participant to orally define words of increasing complexity. The Auditory Reasoning subtest of the Test of Auditory Processing Skills. Third Edition (TAPS-3; Martin and Brownwell 2005) provided an age-normed scaled score (M=10, SD=3) assessing higher order linguistic processing related to listening comprehension, understanding implied meanings and idioms, and to making inferences. Participants are read short vignettes (approximately 2-3 sentences each) and asked to respond to one question for each vignette. In order for an answer to receive credit, a participant must either make the correct inference, or correctly interpret an abstraction or idiom. Internal consistency Cronbach's alpha from our sample for Auditory Reasoning (alpha=0.87) was generally consistent with publisher reported alphas (alphas = 0.91-0.96; Martin and Brownwell 2005). The Story Recall subtest of the Wide Range Assessment of Memory and Learning, Second Edition (WRAML2, Sheslow and Adams 2003) tapped the ability to listen to and utilize narrative structure to organize and retell gist and verbatim details of two orally presented narratives and yielded an age-normed scaled score (M = 10, SD=3). Internal consistency Cronbach's alpha from our sample for Story Recall (alpha=0.95) was generally consistent with publisher reported alphas (alphas = 0.91 - 0.92; Sheslow and Adams 2003).

Reading Comprehension

The Gray Oral Reading Tests—Fifth Edition (GORT-5; Wiederholt and Bryant 2012) provided a standardized measurement of reading comprehension that yielded agenormed scaled scores (M=10, SD=3). The individually administered test is comprised of 16 progressively more difficult reading passages read aloud by the child, each followed by 5 open-ended comprehension questions given orally by the tester with the passage removed from view. Question types vary, from those asking for recall of details to those requiring higher order processing such as synthesis of the main idea, understanding of causal relations, or ability to make predictions. Publisher (Wiederholt and Bryant 2012) reported Cronbach's alpha reliability coefficients for Comprehension scores range between 0.90 and 0.96 in the normative sample, and 0.97 in an ASD subsample.

Analytic Strategy

Differentiated Profiles of Reading Skills

All analyses were conducted using Mplus 7.11 (Muthén and Muthén 1998–2015). To answer the first research question, we began by iteratively fitting a series of unconditional latent profile analyses beginning with a one-profile model and increasing the number of profiles by one with each subsequent run. The twelve reading-related measures (i.e. RAN, NWR, Elision, PDE, SWE, GORT Accuracy, Recalling Sentences, Expressive Vocabulary, Auditory Reasoning, Story Recall, and GORT Comprehension) were used as latent profile indicators. See Fig. 1 for a conceptual diagram of the full model. As the 12 indicators represented the 4 broader constructs, including the indicators simultaneously allows profiles to reflect differences across the 4 constructs concomitantly. This analysis also provides an empirical method of deriving reading profiles as opposed to using relatively arbitrary cutoff scores. Finally, examining the results in light of the 12 indicators (and, consequently, the 4 broader constructs) simultaneously enabled us to identify the greatest discrepancies across constructs among the emergent profiles.

Multiple fit indices were used to compare the models as no single fit index has been shown to perfectly identify the optimal model (Nylund et al. 2007). First, we utilized the Bayesian Information Criterion (BIC; Schwarz 1978) and adjusted BIC (ABIC) with lower values indicating a preferred model. Additionally, we used two likelihood ratio based indices, the Lo-Mendell-Rubin (LMR) test and the bootstrapped likelihood ratio test (BLRT). Both tests assess whether adding a profile significantly improves model fit such that a non-significant p-value for a k-class model indicates the model with k-1 classes is preferred. For further information on these three fit indices, see Nylund et al. (2007). Finally, we employed two information-heuristic indices, the Bayes Factor (BF) and correct model probability (cmP) that have only recently been applied to mixture modeling (Masyn 2013). The BF provides pairwise comparisons of adjacent models that provides a ratio of the probability of a model with k classes being preferred compared to a model with k+1classes. Values between 1 and 3 are weak evidence for the k-class model, 3-10 are moderate evidence, and values greater than 10 indicate strong evidence. The cmP provides a probability that each model is preferred compared to all of the models under consideration. While not considered a fit index, we also examined entropy, which provides a measure of the strength of classification, with values between 0.80 and 1.00 or greater indicative of good



Fig. 1 Conceptual diagram of heterogeneous reading profiles and relation to ASD symptomatology

classification (Ram and Grimm 2009). While fit statistics aided us in identifying a chosen model, we also considered the substantive interpretation of the latent profiles in each model to ensure the chosen model was theoretically viable (Muthén 2003).

Linking Reading Profiles to ASD Symptomatology

After choosing the preferred unconditional model, we examined differences in ASD symptomatology based on latent profile membership to answer the second research question. This was accomplished by estimating profilespecific means. This process has been shown to result in a shift in the latent profiles, thereby altering the substantive interpretation of them (Asparouhov and Muthén 2014a; Nylund-Gibson et al. 2014). Therefore, we implemented the BCH approach (Asparouhov and Muthén 2014b; Bakk et al. 2013; Bolck et al. 2004; Vermunt 2010) in order to account for classification error and avoid profile shifts. This method does so by applying weights to individuals based on posterior probabilities of profile membership. Finally, the BCH approach estimated profile-specific means of ADOS and conducted all pairwise comparisons. For technical details of the BCH approach, see Asparouhov and Muthén (2014b), Bakk et al. (2013), and Bolck et al. (2004).

Results

This section is divided into three subsections reflecting the model building steps. First, we provide descriptive statistics to compare the present sample to national norms. Next, we describe the latent profile enumeration process and label and interpret the emergent profiles. Finally, we present the results of the relation between the reading profiles and ASD symptomatology severity.

Descriptive Statistics

Descriptive statistics, all reported as standard scores can be seen in Table 1. The descriptive statistics demonstrate that the sample met criteria for ASD on the ADOS-2; measures of IQ show normal range. A range of scores was seen on the reading related measures. On average, the overall sample scored at least one standard deviation below the normed mean on all reading measures. The one exception is the word decoding measure, PDE and SWE where the sample scored closer to the normed average of 100. In order to determine if the heterogeneity of the samples reading abilities, next we conducted a series of latent profile analyses.

Table 1 Descriptive statistics for the full samp	le
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Measure	М	SD	Range
PP and RAN			
RAN ^c	85.68	21.92	1-145
NWR ^a	7.50	2.15	1–13
Elision ^a	9.94	3.08	1-15
Word recognition			
PDE ^c	94.89	14.81	58-127
SWE ^c	93.29	14.75	57–136
GORTAcc ^a	8.03	2.69	2-16
Linguistic comprehension			
CELFrs ^a	7.36	3.15	
Evocab ^b	46.96	9.89	24-69
AudReas ^a	6.04	2.77	1-11
StryRec ^a	7.94	3.31	1-15
Reading comprehension			
GORTComp ^a	7.37	2.61	1-13
ASD symptomatology			
ADOS-2 total	10.94	3.65	7–24
IQ			
FIQ ^c	100.00	14.00	76–132
VIQ ^c	96.00	15.00	60–136
PIQ ^c	105.00	16.00	71–150
Age	11.24	2.19	8-16

PP phonological processing, *RAN* rapid automatized naming, *NWR* CTOPP nonword repetition, *PDE* phonemic decoding efficiency, *SWE* sight word efficiency, *GORTAcc* GORT text accuracy, *CELFrs* CELF recalling sentences, *EVocab* expressive vocabulary, *AudReas* auditory reasoning, *StryRec* story recall, *GORTComp* GORT reading comprehension, *FIQ* full-scale IQ

^aScaled score, M = 10, SD = 3

^bT-score, M = 50, SD = 10

^cStandard score, M = 100, SD = 15

Identifying Differentiated Reading Profiles

Fit statistics of the six latent profile models can be seen in Table 2. Values in boldface indicate the preferred model for a particular fit index. The BIC reached a minimum value at the 4-profile model. However, the only statistically significant LMR value occurred with the 2-profile model. The BLRT never became non-significant and, thus, was non-informative in choosing a preferred model. Both the BF and cmP supported the 4-profile model. The entropy value for the 4-profile model was 0.90 (Table 3).

While statistical evidence was clear for the 4-profile model, we also examined the profile plot to ensure theoretical viability. Though the analysis was conducted using age-normed standardized scores, these were rescaled to z-scores for the profile plot to foster interpretability. The four profiles were characterized by their performance on the reading and language measures. The profile plot for the 4-profile model can be seen in Fig. 2. The profile demarcated by a dashed line with square markers was labeled Readers with Comprehension Disturbance and accounted for approximately 20% of this sample. These students were characterized by average rapid automatized naming, phonological awareness, word decoding and word recognition, text reading accuracy, and expressive vocabulary, alongside low-average phonological memory, sentence-level syntactic expressive language skills, and story recall. Concomitant deficits in auditory reasoning/inference, and reading comprehension typified this profile. The profile demarcated by a dotted line with triangle markers accounted for about one-third of the sample and was distinguished by poor performance (approximately 1 SD below average) across all language and reading variables, so we termed this profile *Readers with Global Disturbance*. The profile at the bottom of the plot depicted by a solid line with diamond markers accounted for about 14% of the sample and was marked by very poor performance on all language and reading variables, this subgroup was called Readers with Severe Global Disturbance. In particular, RAN, sentence-level syntactic expressive language skills, auditory reasoning/inference, narrative retelling, and reading comprehension were very low with scores approximately 2 SD or more below average. The final profile with a solid line with circular markers accounted for about 32% of the sample and was delineated by scores in the average range on all language and reading variables; this subgroup is called Average Readers.

Table 2	Fit statistics of the six	
LPA mod	lels	

Profile	LL	BIC	ABIC	LMR p value	BLRT p value	BF	cmP	Min n
1	-2687.81	5472.31	5402.93	_	_	< 0.001	< 0.001	_
2	-2580.87	5311.15	5203.92	0.006	< 0.001	1.56	0.17	34
3	-2554.94	5312.03	5166.96	0.683	< 0.001	0.16	0.11	13
4	-2526.75	5308.38	5125.46	0.181	< 0.001	34.81	0.69	11
5	-2503.93	5315.48	5094.72	0.37	< 0.001	_	0.02	10
6	Did not converge							

LL log-likelihood, BIC Bayesian information criterion, ABIC adjusted BIC, LMR Lo-Mendell-Rubin likelihood ratio test, BLRT bootstrapped likelihood ratio test, BF Bayes factor, cmP correct model probability

Table 3Profile-specific means(standard errors) of all indicatorvariables

Measure	Severe global	Comprehension	Global disturbance	Average readers	
	disturbance	disturbance		-	
PP and RAN					
RAN ^c	71.70 (8.12)	96.79 (4.39)	80.24 (4.50)	90.31 (4.51)	
NWR ^a	6.25 (0.54)	7.60 (0.50)	7.22 (0.44)	8.27 (0.45)	
Elision ^a	7.58 (1.24)	11.93 (0.78)	8.34 (0.79)	11.35 (0.38)	
Word recognition					
PDE ^c	86.20 (4.64)	105.81 (3.99)	82.33 (3.32)	104.89 (2.09)	
SWE ^c	76.70 (3.37)	101.22 (3.95)	86.64 (2.98)	102.66 (2.60)	
GORTAcc ^a	5.61 (0.70)	8.56 (0.66)	6.63 (0.30)	10.26 (0.58)	
Linguistic comprehension					
CELFrs ^a	2.67 (0.75)	7.18 (0.53)	6.74 (0.54)	10.28 (0.41)	
EVocab ^b	35.36 (2.56)	47.22 (2.48)	42.94 (1.39)	55.74 (1.53)	
AudReas ^a	2.75 (0.53)	4.66 (0.97)	6.71 (0.67)	7.67 (0.47)	
StryRec ^a	3.78 (0.76)	7.40 (1.05)	8.00 (0.63)	9.86 (0.58)	
Reading comprehension					
GORTComp ^a	3.50 (0.45)	5.85 (0.64)	7.42 (0.26)	10.00 (0.40)	

PP phonological processing, *RAN* rapid automatized naming, *NWR* CTOPP nonword repetition, *PDE* phonemic decoding efficiency, *SWE* sight word efficiency, *GORTAcc* GORT text accuracy, *CELFrs* CELF recalling sentences, *EVocab* expressive vocabulary, *AudReas* auditory reasoning, *StryRec* story recall, *GORTComp* GORT reading comprehension

^aScaled score, M = 10, SD = 3

 ${}^{b}\text{T-score}, M = 50, SD = 10$

^cStandard score, M = 100, SD = 15





ciency, GORTAcc GORT text accuracy, CELFrs CELF recalling sentences, EVocab expressive vocabulary, AudReas auditory reasoning, StryRec story recall, GORTComp GORT reading comprehension We also examined potential differences in age and gender among the emergent profiles using the three-step method (Asparouhov and Muthén 2014a; Nylund-Gibson et al. 2014). There were no effects of either age or gender. Readers of any age or either gender were equally likely to be assigned to any of the latent profiles.

Relating Reading Profiles to ASD Symptomatology

The final step in this analysis was to relate ASD symptomatology (i.e. ADOS-2 total score) to the heterogeneous reading profiles using the BCH approach. Results can be seen in Fig. 3. The *Readers with Severe Global Disturbance* (M=14.38) had the highest level of ASD symptomatology and this was significantly higher than both the *Readers* with Global Disturbance (M=10.15) and Average Readers (M=9.98) profiles. Readers with Comprehension Disturbance (M=11.31) did not significantly differ from any of the other three profiles and there were no other significant differences across profiles.

Discussion

There is converging evidence that many individuals with ASD demonstrate difficulties with reading; the majority of previous studies have concentrated specifically on reading comprehension disturbance (Estes et al. 2011; Jones et al. 2009; Nation et al. 2006; Norbury and Nation 2011; Ricketts et al. 2013). There is some evidence that beyond reading comprehension disturbance, there are different profiles of readers in school-aged children with ASD (e.g., Brown et al. 2013; Jones et al. 2009; Nation et al. 2006). In addition, research has delineated language subgroups in children and adolescents with ASD (Rapin et al. 2009;



Fig. 3 Means (standard deviations) of ADOS-2 scores by reading profile. An *asterisk* indicates significantly different (p < .05) ADOS-2 means between a given pair of reading profiles

Tager-Flusberg and Joseph 2003), and language impairments have been linked to reading difficulties in this population (Lindgren et al. 2009; Lucas and Norbury 2014; Norbury and Nation 2011). However, the relation between these language and reading subgroups was previously unexamined using comprehensive reading and language batteries. In the present study, the first research question probed the heterogeneity of reading and language performance for individuals with HFASD based upon a comprehensive battery of assessments of phonological processing, word recognition, and linguistic and reading comprehension measures. The inclusion of both lower-level reading sub skills that are related to word recognition and variables related to higher-level linguistic comprehension allowed simultaneous consideration of the relation between the two domains outlined by the Simple View of Reading and their sub skills. Four distinct profiles emerged from the sample of students with HFASD: Readers with Comprehension Disturbance, Readers with Global Disturbance, Readers with Severe Global Disturbance and Average Readers. The second research question investigated the relation between the subgroups of readers and ASD symptomatology in order to further understand the relation between the socialcommunicative and cognitive phenotype of ASD and reading related skills in sample of individuals diagnosed with HFASD.

HFASD Reading Subgroups

The Readers with Comprehension Disturbance typified the poor comprehender or hyperlexic reading disability profile predicted by the Component Model of Reading. This subgroup has been frequently reported in prior studies of reading with individuals with ASD (e.g., Brown et al. 2013; Jones et al. 2009; Nation et al. 2006; Newman et al. 2007; Huemer and Mann 2010; Wei et al. 2015; Zuccarello et al. 2015) and shares characteristics with a language subgroup reported by Rapin et al. (2009) whose members demonstrated adequate phonology and vocabulary alongside linguistic comprehension deficits. Grigorenko et al. (2003) noted disagreement in the literature as to whether hyperlexia is synonymous with a reading comprehension disorder, or whether it is a unique condition characterized by an almost obsessive interest in letters and words, precocious and unprompted emergence of word decoding, and an extreme degree of discrepancy between word recognition and other cognitive skills that emerges between 3 and 5 years of age (Healy 1982). Individuals in this profile demonstrated strong phonological awareness, decoding, and word reading skills; it is possible that some of the children in this group may have been considered hyperlexic earlier in their development. We do not have data depicting the sample's early reading development prior to age 8, but even if some of the children demonstrated a precocious and circumscribed interest in word reading and decoding when very young, they are now functioning in the average range, similar to findings reported by Newman et al. (2007).

Single word expressive vocabulary for children in the Readers with Comprehension Disturbance profile was in the average range. However, the two measures reported to be sensitive markers of language impairment, Nonword Repetition and Recalling Sentences (Condouris et al. 2003; Norbury and Nation 2011; Rapin et al. 2009), posed a challenge for many of these children. Therefore, in this subgroup, word recognition abilities did not necessarily align with structural language abilities as reported in prior studies of reading and language in ASD (Lindgren et al. 2009; Lucas and Norbury 2014; Norbury and Nation 2011). Furthermore, children in this profile displayed higher-level linguistic comprehension deficits ranging from approximately 1 to nearly 2 standard deviations below average across the auditory reasoning/inference, story recall, and reading comprehension measures. In summary, while children in this subgroup demonstrated adequate word recognition skills and single word vocabulary and therefore may appear to be proficient readers if other sub skills are not assessed, their moderate to profound structural language and linguistic comprehension difficulties significantly impaired reading for meaning.

Readers with Global Disturbance corresponded with the garden variety poor reader (Gough and Tunmer 1986), or mixed reading disability subtype (Catts and Kamhi 2005), and has also been reported in prior studies of reading in ASD samples (Åsberg and Dahlgren Sandberg 2012; Gabig 2010; White et al. 2006; Nation et al. 2006). Children in this subgroup shared characteristics with a language subgroup reported by Rapin et al. (2009) who struggled with phonology, vocabulary, and linguistic comprehension. Similarly, Tager-Flusberg and Joseph (2003) identified an impaired language subtype of children with ASD who tended to have phonological processing deficits and scores 1-2 SD below the mean on most language tests. Unlike Readers with Comprehension Disturbance, Readers with Global Disturbance demonstrated overall low word reading and decoding abilities commensurate with their poor language skills. Similar to children in the Readers with Global Disturbance, the Readers with Severe Global Disturbance resembled Rapin et al. (2009) and Tager-Flusberg and Joseph's (2003) language impaired subtypes previously described, but with far more severe impairment. The distinction between the two latter profiles may be thought of as categorically distinct areas of a continuum, such as the difference between the terms "below average" and "far below average" that are sometimes used in diagnostic measures. This is consistent with longitudinal evidence from Pickles et al. (2014) that oral language impairments in ASD present in parallel patterns of development and proficiency levels after the age of seven. Together these two subtypes comprised about 47% of the sample.

The individuals in the Average Readers subgroup did not struggle with the reading or language measures, and in fact exhibited intact reading skills overall. Their performance across the language measures was similar to that of Rapin et al.'s (2009) subgroup that demonstrated average or above performance on all language and cognitive measures. Similarly, this subgroup shared many characteristics with Tager-Flusberg and Joseph's (2003) description of a group of children with ASD with normal linguistic abilities who have intact phonological skills, fluency, syntax and morphology, expressive language, and average to large lexicons. They noted however, that comprehension may still be impaired at the discourse level, as well as for more open-ended questions such as "why, when, and how". Therefore, more complex measures of reading and linguistic comprehension that require increased demands on cognitive resources, inferential thinking, and social knowledge might still pose a challenge for those in the Average group.

Many of the children in all subgroups performed poorly on the auditory reasoning/inference measure. This is consistent with Tager-Flusberg and Joseph's (2003) finding regarding difficulty with open-ended questions, as well as research indicating that children with ASD often have difficulties integrating information from background knowledge with that from the text for global coherence and inference generation (Norbury and Nation 2011; Wahlberg and Magliano 2004). However, some studies have demonstrated that there are aspects of inferencing which may be preserved in children with ASD such as automatic inference generation between sentences in very short passages (Saldaña and Frith 2007) and inferring emotions of main characters in short texts (Tirado and Saldaña 2016). However, Tirado and Saldaña (2016) also found that their participants had difficulty responding to questions about those inferred emotions. It is possible that for individuals with ASD, there is particular difficulty with a deep understanding of inferences in situations that are more abstract such as in the context of reading unknown text, and that these difficulties may be exacerbated in longer texts. This is an important area to target for explicit instruction.

This study found both similarities and differences compared to the subgroups of neuro-typical readers reported by Catts et al. (2003). The most prominent difference was that no dyslexic profile emerged in our study whereas this subgroup made up 35.5% of the poor readers in their sample. This finding is consistent with previous studies of reading in samples with ASD (Lindgren et al. 2009; Lucas and Norbury 2014; Norbury and Nation 2011). Similar to these prior studies, poor word reading and decoding in our sample was generally associated with structural language difficulties as well as language and reading comprehension impairments, not as a stand-alone dyslexic profile.

However, both the Readers with Global Disturbance and the Readers with Severe Global Disturbance profiles resembled the Catts et al. (2003) language-learning disabilities subgroup. Combining our two Global Disturbance profiles would account for 47.3% of our sample compared to 35.7% in the Catts et al. sample. Thus, while we identified a similar subgroup, the prevalence rates differed between the two studies. The differences might be a result of the younger age (i.e., second grade) used in the Catts et al. (2003) study. However, language delay and impairment is common in children with ASD (Pickles et al. 2014) and it is probable that children with ASD who struggle with reading are more typified by impairments in either language comprehension alone or language comprehension coupled with word reading difficulties. This could explain the lack of a dyslexic profile along with a greater prevalence of children who resembled the language-learning disabilities subgroup in Catts et al. (2003).

Relation of Reading Profiles to ASD Symptom Severity

Previous research has provided evidence that reading comprehension is negatively associated with ASD diagnosis and symptom severity (Åsberg et al. 2010; Estes et al. 2011; Jones et al. 2009; Norbury and Nation 2011; Ricketts et al. 2013). Results of this study are consistent with these previous findings: reading comprehension scores were highest when ASD symptomatology as measured by the ADOS-2 was lowest. Readers with Severe Global Disturbance, who demonstrated the poorest linguistic and reading comprehension abilities, had significantly higher levels of ASD symptomatology than children in the Average Reader and Global Disturbance subgroups. They also struggled the most with sight word recognition and text reading accuracy, consistent with individuals referred to in the typically developing literature as having a language-learning disability (LLD; Berninger and May 2011). The current study provides additional evidence that the social communicative and cognitive phenotype of ASD impacts both linguistic and reading comprehension for many students with ASD throughout the school-age years.

Potential Implications for Treatment of Reading Disturbance

The majority of students with HFASD are educated in general education classrooms. Extant data suggests that these students are being underserved in these settings in the area of reading development, with many of them scoring at least one grade level below their typically developing peers on reading assessments. In this sample of higher-functioning children with ASD, 65% of the students were in general education classes 81–100% of the day, and an additional 12% were in these classes 41–80% of the day, yet almost 68% of the students demonstrated various profiles of moderate to severe language and reading difficulties. Furthermore, these profiles were related to the severity of social communication and cognitive characteristics associated with ASD. This has important implications for educating students with an ASD diagnosis, particularly in socially-mediated, language-based learning contexts. It is difficult to expect general education teachers to know how to meet the reading instructional needs of individuals with HFASD when very little is known about the development of the sub skills necessary for successful comprehension in this population of students.

In order to address the unique instructional needs of students with ASD, and to be able to develop the most effective reading intervention protocols, a more in depth investigation into reading profiles in this population was necessary. The results of this study demonstrate that assessment and intervention methods must be tailored to meet the specific reading needs of individual students, and the specific skill deficits depicted in these profiles can be addressed. Thorough assessment of both word recognition and linguistic comprehension sub skills is important; for example, average single word expressive vocabulary was higher for all subgroups than was auditory reasoning ability and an overreliance on vocabulary skill level could lead to overlooking a key domain for intervention. When planning intervention, students in the Readers with Comprehension Disturbance subgroup, who demonstrate dissociation between word recognition skills and comprehension, would benefit from explicit structural language intervention and linguistic comprehension instruction. However, students in the Severe Global and Global Disturbance subgroups would benefit from explicit phonological processing, word recognition, and linguistic comprehension intervention. The Severe Global Disturbance subgroup would likely benefit from a much more intense intervention in these areas and may require additional behavioral scaffolding to sufficiently engage with the intervention. These types of targeted interventions could be implemented through collaborations with various school professionals including reading specialists, speech and language pathologists, and special education or general education teachers.

Conclusions

The proportion of individuals with reading disturbance has been shown repeatedly to be greater in samples of individuals with ASD than in the general population. The data in the present study concurred with previous literature that a large percentage of individuals with ASD demonstrate reading disturbance and that this disturbance is associated with language impairments. Furthermore, this study provided additional evidence that phonological awareness is associated with word decoding for school-aged children with ASD, as is seen in typically developing samples, and word recognition deficits were concomitant with language deficits. It has also been argued and shown empirically that there is a significant relation between the social-communicative and cognitive phenotype of ASD and reading performance. The present study demonstrates support for this finding in a much more specific way, by showing that ASD symptom severity is related differentially to specific profiles of readers.

Limitations and Future Directions

A limitation of the current study is that while the sample size was relatively large, the developmental span across elementary and secondary school years was extensive. Future studies would benefit from even larger samples at each age and grade level to more fully understand reading profiles in students with ASD. Future studies would also benefit from data collection with students with a broader range of ASD severity, as this study only included individuals with HFASD. We would also suggest that text reading fluency be collected as a part of future reading batteries as it is possible that while this sample showed relative strength on word reading, it may not translate to fluent reading of connected text. In addition to text reading, we also note limitations related to the language measures used in this study. In the future, we would suggest collecting more robust measures of language development in order to gain a better understanding of the role language plays in reading comprehension. Another limitation of this study is that the standardized measures used may not have been robust enough to adequately capture the extent of higher-level linguistic and reading comprehension challenges. A future study would benefit from the inclusion of additional reading and language comprehension measures that are more complex and would demand more cognitive resources, inferential thinking, narrative retelling, and social knowledge. Longer texts, both fiction and nonfiction, that are similar to those used in classrooms, as well as other genres such as persuasive essays or satire might also uncover additional targets for intervention even for those in the Average Reader group. Longitudinal studies would also contribute further to our understanding of patterns of subgroup membership and how they may change with intervention and maturation. Finally, future investigations are needed to further probe the specific aspects of ASD symptomatology that are associated with reading and language-based learning in structured, multifaceted social contexts such as classrooms in order to develop effective interventions for school-aged children with ASD.

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Authors' Contributions NM participated in the design and coordination of the study, acquired data, and oversaw statistical analyses, interpretation, and drafting and revision of the manuscript; ES participated in the design and interpretation of statistical analyses, and made a substantial contribution to the drafting and revision of the manuscript; RG designed, directed and performed statistical analyses, created tables and figures, and participated in the drafting and revision of the manuscript; LL participated in the design and coordination of the study and acquired data; JG collaborated on paper methodology and performed statistical analyses; PM conceived of the study, directed its design and coordination, and participated in the drafting of the manuscript. All authors read and approved the final manuscript.

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Compliance with Ethical Standards

Conflict of interest All the authors declare that they have no conflict of interest.

Ethical Approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed Consent Informed consent was obtained from all individual participants included in the study.

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