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ORIGINAL PAPER

Language Skill Differences Further Distinguish Social Sub-types in Children with Autism

Weihua Zhao[1] · Qin Li[2] · Xiaolu Zhang[1] · Xinwei Song[1] · Siyu Zhu[1] · Xiaojing Shou[3] · Fanchao Meng[3] · Xinjie Xu[3] · Rong Zhang[3] · Keith M Kendrick[1]

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Abstract
This study investigated heterogeneity in language skills of children with autism and their relationship with different autistic social subtypes. Data from 90 autistic and 30 typically developing children were analyzed. Results showed that autistic social subtypes varied in language skill problems (aloof > passive > active-but-odd). There was a negative association between aloof dimension scores and language performance but positive for the active-but-odd dimension and no association in the passive one. Moreover, aloof dimension score was the main contributor to language performance. A receiver operating characteristic analysis suggested language vocabulary as an additional component in differentiating autistic social subtypes. These findings demonstrate that variations in language skills in autistic children provide additional information for discriminating their social subtype.

Keywords Heterogeneity · Autistic subtype · Language skill · Vocabulary test · Aloof

Introduction

Autism Spectrum Disorder (ASD) is markedly heterogeneous and characterized by two core symptoms: difficulties in social interaction and communication and restricted, repetitive behaviors or interests (Diagnostic and Statistical Manual of Mental Disorders, DSM-V) and ASD prevalence estimates have been one in 44 children aged 8 years from the latest Autism and Developmental Disabilities Monitoring Network report (Maenner et al., 2021). Unlike other psychiatric disorders (e.g. schizophrenia, anxiety), individual variation in autism goes beyond core symptoms and its heterogeneity varies across some factors, such as intellectual abilities (Charman et al., 2011, i.e. intellectual disability, average intelligence; above average) and neuro-subtypes (Hong et al., 2020; i.e. intrinsic functional connectivity decreased between networks but increased within networks for neurosubtype 1 relative to neurosubtype 2). There have been an increasing number of studies attempting to parse the heterogeneity among individuals with autism using person-centered approaches (Kaneko et al., 2022). For example, four distinct subgroups were identified based on the degree of parent-teacher informant discrepancy about autism symptom severity and this also helped for characterizing the autistic phenotype (Lerner et al., 2017). In addition, the siblings of individuals with autism also exhibited diversity and heterogeneity (i.e. unaffected, normative development, receptive language delay, widespread delay) (Landa et al., 2012). Autism Diagnostic Observation Schedule (ADOS) severity scores revealed 4

Weihua Zhao, Qin Li, Rong Zhang and Keith M Kendrick contributed equally to this work.

✉ Rong Zhang
zhangrong@bjmu.edu.cn

✉ Keith M Kendrick
k.kendrick.uestc@gmail.com

1 MOE Key Laboratory for NeuroInformation of Ministry of Education, Center for Information in Medicine, The Clinical Hospital of Chengdu Brain Science Institute, University of Electronic Science and Technology of China, 611731 Chengdu, China

2 Chengdu University of Traditional Chinese Medicine, 611137 Chengdu, China

3 Neuroscience Research Institute, Key Laboratory for Neuroscience, Key Laboratory for Neuroscience, Department of Neurobiology, School of Basic Medical Sciences, Ministry of Education of China, National Committee of Health and Family Planning of China, Peking University, 100191 Beijing, China

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different classes including severe persistent (no changes in social affect score over time), worsening (increased over time), improving (decreased over time) and non-spectrum (marginal decreased) (Lord et al., 2012). Moreover, a similar 4-class model also reported (persistent high, moderately severe, increased and decreased) using longitudinal ADOS severity scores (Gotham et al., 2012).

In addition to the factors mentioned above, language skills are also a key aspect of heterogeneity in autism. Language skills in individuals with autism can range from mutism to adequate speech with poor conversational skills (Fombonne, 1999). Pickles et al. suggested that individuals with autism could be sub-grouped into mild delay, late delay, marked delay and very low development in expressive and receptive language (Pickles et al., 2014). Kang et al. proposed three subgroups: a stable subtype (stable pragmatic language difficulties), mostly current-only subtype (current-only pragmatic language difficulties and perseveration) and little professors (slight pragmatic language difficulties) according to atypical communication characteristics (Kang et al., 2020). Most studies have focused on general quantitative language performance or structural observations, such as the number of words produced (Spek et al., 2009). In particular, profiles of communication and language problems can be traced back to the stage of infancy and even earlier. In general, onset of symptoms can occur at a very early stage although it is not until the parents notice there are certain delays or regressions in the children's development of speech that they are taken to the hospital for further possible diagnosis with ASD (Short & Schopler, 1988). Individuals with autism show differences not only in terms of universal difficulties with pragmatics, but also with structural aspects of language (Eigsti et al., 2011). For example, individuals with autism frequently invent novel words (i.e. neologisms) with a specific idiosyncratic meaning (Eigsti et al., 2007). Given that language delay has been associated with a greater incidence of autism (Eigsti et al., 2011; Howlin, 2003), a more in depth and systematic focus on language development (i.e. atypicality of quantity, specific aspects of verbal performance) could help with diagnosis and early intervention for children with autism.

Typically developing (TD) children start producing single words at about 12-months old and develop increasingly complex conversational speech afterwards (Tager-Flusberg, 2006). However, children with autism typically show little intention of engaging in social interaction at an early stage and many develop insufficient functional language. On average, children with autism only start speaking at around 38 months old, which represents a significant delay compared with 8–14 months in TD children (Howlin, 2003), and exhibit significant problems in both semantic and phonemic tasks (Eigsti et al., 2007; Ehlen et al., 2020; Kenworthy et al., 2009). However, children with autism are comparable with TD in producing novel verbs (Shulman & Guberman, 2007) or verbal fluency (Tóth et al., 2022) and individual differences (heterogeneity) in the vocabulary skills of children with autism have frequently been observed with around a third failing to develop sufficient language for even basic communication with others (Tager-Flusberg & Kasari, 2013).

Previously, Tager-Flusberg et al. have divided autistic children (7–14 years old) into two subgroups: children with normal linguistic abilities and children whose language proficiency is similar to children diagnosed with specific language impairment. These two autism subgroups show distinctive performance across language measures (i.e. vocabulary, phonological processing, syntax and semantics) (Tager-Flusberg, 2006; Tager-Flusberg & Joseph, 2003). A recent study has also shown that these two language subtypes can even be distinguished by differential responses to vocal stimuli in brain language processing regions as early as 12 months of age (Lombardo et al., 2018). Additionally, individuals with autism who have language problems also exhibit pronounced difficulties in executive control (i.e. suppressing reflective shifts of gaze and maintaining fixation on a target) (Kelly et al., 2013). On the other hand, Wing & Gould (1979) have proposed subtyping based more generally on how children interact with others socially (i.e. ranging from social aloofness to awkward social approaches) and have distinguished three subtypes: (1) aloof: indifferent and uninterested in various forms of social context; avoiding social interaction and communication; (2) passive: lack of social overture and little spontaneous initiation in communication. Although children in this category do respond to others advances or requests, they appear to derive little pleasure from such interactions; (3) active but odd (ABO): spontaneous social contact with poor or deficient quality (Bonde, 2000). Autistic symptom severity is generally greatest in the aloof subtype and least in the ABO (Meng et al., 2018) and notably, individuals with the different autism social subtypes can even show different responses to the same intervention, i.e. aloof children compared to the other two subtypes showed smaller changes in IQ after a period of early, intensive behavioral intervention (Beglinger & Smith, 2005). In addition, verbal communication subscales of the Child Autism Rating Scale (CARS –Schopler et al., 1980) and Autism Behavior Checklist (ABC – Volkmar et al., 1988) also indicate that use of expressive language is lowest in aloof and greatest in ABO subtypes (Meng et al., 2018). However, no studies to date have assessed language skills in the three autism social subtypes using language-specific measures or determined the differences between each autism social subtype and TD individuals. Such a cross-subtype study will help provide a better understanding of the status of language development in autistic children with different social phenotypes and identify which ones might particularly benefit from language training interventions.

Against this background, we aimed to investigate the heterogeneity language skills in young Chinese children (3–6 years) with autism and check the difference (i.e. quantity and specific aspect) between each autism social subtype and TD children using the Chinese vocabulary test (Hao et al., 2008) as follows: (1) using a categorical approach, with the three subgroup categories assessed via the Beijing Autism Subtyping Questionnaire (BASQ; aloof, passive, ABO - Meng et al., 2018) and compared to a TD group in their language-related skills; (2) using a dimensional approach, where the magnitudes of scores on each of the three individual categories of social symptoms are associated with severity of language skill problems. Based on previous findings we hypothesized that language skill would be most affected in the aloof subtype and additionally that scores on the aloof dimension across subjects would generally exhibit the strongest association with language problems.

Methods

Participants

A sample of 120 children aging from 3 to 6 years participated in the study, 90 diagnosed with ASD (10 girls (11%), mean age = 4.62 years, SD = 0.58) and 30 age-matched typically developing (TD) children (6 girls (20%), mean age = 4.68 years, SD = 0.45, equal variances not assumed ($F = 4.06$, $p = 0.046$), $t(64.01) = 0.66$, $p = 0.51$). Children with autism were recruited through pediatric psychiatric clinics and autism rehabilitation training centers in Beijing. Age and gender (gender: 11% vs. 20%, Chi-Square Test, $\chi^2 = 2.61$, $p = 0.11$) matched TD children were also recruited through online social platforms or day care centers in Beijing. A priori power calculation using G*Power (version 3.1.9.2) showed that 100 subjects would be sufficient to achieve 95% power for a medium effect size of 0.3 at $\alpha = 0.05$ with repeated ANOVAs. All children in the autism group met the diagnostic criteria of Diagnostic and Statistical Manual of Mental Disorders IV-Text Revision (DSM-IV-TR) (American Psychiatric Association, 1994) or DSM-V (American Psychiatric Association, 2013) and International Statistical Classification of Diseases and Related Health Problems 10th revision (ICD-10) (Brämer, 1988) and diagnosis was confirmed using the Autism Diagnostic Observation Schedule (ADOS) (Lord et al., 2009; Molloy et al., 2011).

Additional measures used included the ABC (Volkmar et al., 1988), CARS (Schopler et al., 1980), and Gesell Developmental Schedules (GDS) (Gesell Institute of Child Development, 2012; Jin et al., 2007) which all contain a language subscale and the Beijing Autism Subtyping Questionnaire (BASQ) (Meng et al., 2018). Four children with autism having the same highest scores in two BASQ subcategories (2 with equal scores on aloof and passive and 2 with equal scores on passive and ABO) were allocated to one of them based on their autism symptom severity measured by the total ADOS score. This resulted in 27 children being classified as aloof, 39 as passive and 24 as ABO. Language proficiency and skills (performance) were evaluated using a validated Chinese vocabulary test for young children (Hao et al., 2008). The TD group were only administered GDS and the vocabulary test. The experiment was approved by the ethics committee of the Peking University Institutional Review Board (approval no. IRB00001052-13079) and study procedures were in accordance with the latest revision of the declaration of Helsinki. Caregivers of all children provided written informed consent.

Measures

Chinese Vocabulary test

The Chinese vocabulary test developed by Hao et al., (2008) is an adaptation of the Communicative Development Inventory (CDI) (Dale & Fenson, 1996) and is composed of 710 items in total, including food and drink (82 items), people (32 items), quantifiers and articles (9 items), animals (51 items), games and routines (30 items), body parts (39 items), descriptive words (75 items), helping verbs (6 items), places to go (19 items), vehicles (14 items), connecting words (6 items), small household items (63 items), outside things (35 items), toys (24 items), action words (124 items), furniture and rooms (33 items), clothing (27 items), pronouns (19 items), question words (9 items) and words about time (13 items). A caregiver is asked which of the words the child uses and understands and the number of words used in each category is counted by the experimenter. This test was originally applied to 884 Chinese subjects aged from 12 to 30 months and the results were well matched with the CDI (Hao et al., 2008).

Beijing Autism Subtyping Questionnaire (BASQ)

The Chinese BASQ (Copyright © 2016 Peking University, No.2016-L-00275072) is a validated adaptation of the original Autism Subtyping Questionnaires (Wing & Gould, 1979) and is composed of 40 items illustrating a child's social behaviors with a good internal consistency (Meng et al., 2018). The 40 items are divided into 4 groups of descriptions designed to evaluate a specific type of behavior (e.g., communication, social approach, and social response). Items in each group correspond to three autistic subtypes or to non-autistic behaviors. The subtype of an autistic child is determined by the highest among the three scores (aloof, passive and ABO). BASQ subtyping can be administered as early as 2 years old. It has good internal consistency (Cronbach's α: aloof-0.891; passive-0.836; ABO-0.821) and test-retest reliability (correlation value: aloof-0.650; passive-0.723; ABO-0.884) (Meng et al., 2018).

Autism Behavior Checklist (ABC)

The ABC is composed of 57 items evaluating autistic behaviors including sensory, relating, body/object use, language, social/self-help (Volkmar et al., 1988). Among these, language scores were used for further analysis based on our study aim. The discrimination validity is 0.78 (Rellini et al., 2004) and the Cronbach alpha coefficient is 0.426 (Chu et al., 2022).

Childhood Autism Rating Scale (CARS)

The CARS is a diagnostic assessment that rates individuals on a scale ranging from normal to severe, and yields a composite score ranging from non-autistic to mildly autistic, moderately autistic, or severely autistic (Schopler et al., 1980). The scale is used to differentiate children with autism from those with other developmental delays, such as intellectual disability and used by clinically trained individuals to observe and subjectively rate fifteen items and is widely used in China. One of the 15 subscales (verbal communication) was used for further analysis based on our aims. The sensitivity of CARS is 100% (Rellini et al., 2004) and the Cronbach alpha coefficient is 0.772 (Chu et al., 2022).

Autism Diagnostic Observation Schedule (ADOS)

The ADOS is a semi-structured, standardized assessment of communication, social interaction, play and imaginative use of material for individuals (from toddlers to adults) suspected of having autism administered by individuals following extensive training. The ADOS has strong inter-rater and test-retest reliability for individual items, excellent inter-rater reliability within domains and excellent internal consistency. It is generally regarded as the gold standard for ASD diagnosis (Lord et al., 2009; Molloy et al., 2011). Inter-rater (kappa $w = 1$) and retest reliability (kappa $w = 0.62$) are good. The concordance of judgment and diagnosis is 77%, with a sensitivity of 90.4% (Bölte & Poustka, 2004). The sensitivity of the ADOS with clinical diagnosis was 97.9% and the positive predictive value was 95.9%. Inter-rater reliability test showed very good results (inter-rater reliability = 0.94, $p < 0.005$, 95% confidence interval (CI) [0.86, 1.00]) (Sun et al., 2015).

Gesell Developmental Schedules (GDS)

The GDS is a standardized instrument to evaluate a child's developmental performance across age including gross motor, fine motor, adaptive, language and social domains with good reliability and validity. It is designed for children from 2.5 to 9 years (Gesell Institute of Child Development, 2012; Jin et al., 2007). It has been validated in a Chinese population and is widely used for assessing child development in China (Ke et al., 2004).

Statistical Analysis

An exploratory factor analysis was performed using principal component (PCA) extraction to identify factors in the vocabulary test. Kaiser-Meyer-Olkin (KMO) and Bartlett's test results showed that the overall of KMO measure of sampling adequacy was 0.956 (values between 0.8 and 0.9 are meritorious, values above 0.9 are superb, Field 2009) and Bartlett's test of sphericity was significant (Chi-square = 3691.91, $p < 0.001$) suggesting that such an analysis was suitable. Subsequently an exploratory PCA was conducted using varimax rotation with Kaiser normalization and the number of components were determined with eigenvalues > 1. Only variables with rotated loadings > 0.7 were included and rotation converged in 3 iterations to generate components with unique variables, which help increase orthogonality and interpretability of the components. Screen plot was performed to check the eigenvalues with corresponding number of factors. The number of factors was identified from the rotated component matrix and total variance explained was reported.

Based on the PCA results, the number of words in the same category (word category: concrete and abstract) were combined and averaged. Subsequently, two-way repeated ANOVA was performed with group (TD, passive, aloof and ABO) as between subject factor and word category from PCA results (concrete and abstract) as within subject factor, and the language performance (the number of words used from the Chinese vocabulary test) in different word categories as the dependent variable. Furthermore, a Pearson's correlation analysis was performed to check the relationship between language skills via vocabulary test and language (or communication) differences in children with autism via ABC, CARS, ADOS (both language and reciprocal social interaction scores) and GDS, as well as the autism subtype scores. Autism subtype scores for each individual subscale were expressed as a percentage of the total score on all subscales (i.e. aloof/ total BASQ scores) to decrease the

Table 1 Demographic and questionnaire measures

Measurements	Aloof	Passive	Active but odd	Statistic	P value
Age	4.60 ± 0.61	4.57 ± 0.57	4.72 ± 0.55	0.54	0.582
Sex(girls)	27(3)	39(4)	24(3)	0.08+	0.963
ABC-total	68.11 ± 27.06#*	45.44 ± 20.86#	28.92 ± 18.50	19.94	< 0.001
ABC-language	16.59 ± 7.83#	12.72 ± 6.83#	7.50 ± 7.03	10.18	< 0.001
CARS-total	37.50 ± 4.27#	35.51 ± 5.25#	30.43 ± 5.10	13.47	< 0.001
CARS-verbal communication	3.07 ± 0.66#*	2.58 ± 0.68	2.22 ± 0.52	11.49	< 0.001
ADOS-total	22.22 ± 3.93#	19.65 ± 5.95#	14.92 ± 4.71	13.52	< 0.001
ADOS-communication	6.22 ± 1.76#	5.81 ± 1.88#	4.42 ± 1.47	7.50	0.001
GDS-total	56.13 ± 13.45#	62.26 ± 16.82#	77.83 ± 14.85	12.59	< 0.001
GDS-language	39.79 ± 15.89#*	52.54 ± 17.31#	72.30 ± 17.27	22.10	< 0.001

Table 1 Demographic and questionnaire measures from the 90 autistic children who divided into autism subtypes (aloof, passive and active but odd). Data are mean and SD (in brackets) for the age, Autism Behavior Checklist (ABC) including total and language scores, Childhood Autism Rating Scale (CARS) including total and verbal communication scores, and Autism Diagnostic Observation Schedule (ADOS) including total and communication scores, Gesell Developmental Schedules (GDS) including total and language scores. The total sample size and the number of girls (in brackets) are presented. Statistic (F-values) and p values for ANOVA analysis. + Chi-square test. # indicating significant differences between aloof and active but odd, passive and active but odd (ps < 0.02); * indicating significant differences between aloof and passive (ps < 0.009)

variance due to large individual differences in total scores and conducted Pearson's correlation analysis to investigate the associations between subtype scores and language performance via the vocabulary test using a dimensional approach. Differences in correlation coefficients for the autism subtypes were tested with bootstrap analysis (Wilcox, 2016). Moreover, to confirm the contribution of three subtype scores in language performance for abstract and concrete words separately, we performed two linear regression models using an enter method (F values and coefficients are reported). In addition, Bayesian ANOVAs (including post hoc tests) and Bayesian correlations were conducted and BF_{10} reported. All statistical analyses were performed using SPSS 25.0 and JASP 0.14.10 (https://jasp-stats.org/). Effect sizes (partial eta-squared for F statistics) were also reported. Partial eta squared was interpreted as > 0.01(small effect), > 0.06 (moderate effect) and > 0.14 (large effect) (Cohen, 1988). Receiver operating characteristic (ROC) plots were used to investigate the ability of language performance on the Chinese vocabulary test to discriminate between autism subtypes or between autism subtypes and TD. Accuracy, sensitivity, specificity and area under ROC curve (AUC) are presented separately for concrete vs. abstract words. Sensitivity (the proportion of true positive results) is shown on the y axis (from 0 to 1) and 1-specificity (the proportion of false positive results) is shown on the x axis (from 0 to 1). An AUC > 0.9 indicates high accuracy, while 0.7–0.9 a moderate accuracy and 0.5–0.7 a low accuracy, with 0.5 indicating chance (Akobeng, 2007). In addition, some other ROC plots including the sensory subscale of ABC/anxiety response subscale of CARS /restrictive behaviors subscale of ADOS were performed to further check whether these indices could differentiate the autism subtypes.

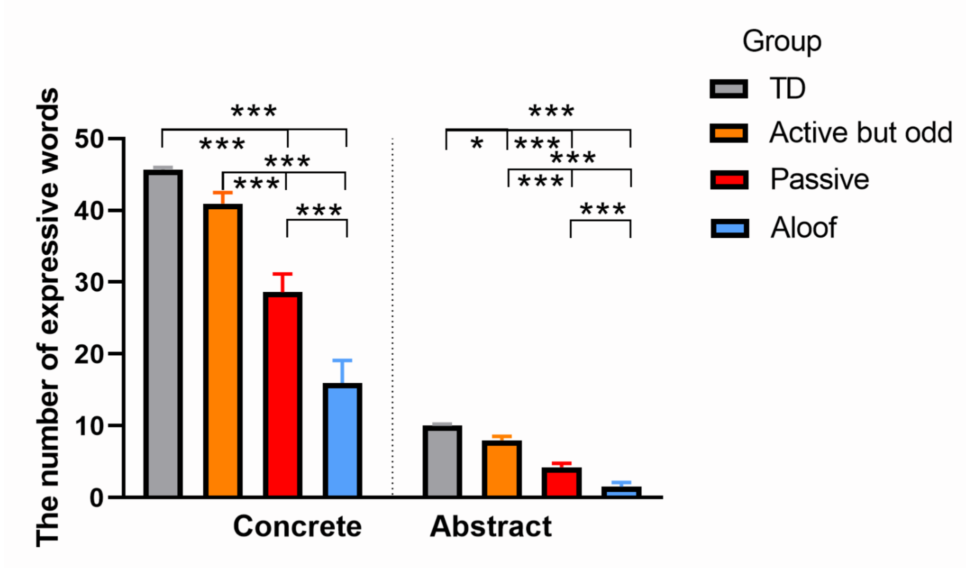
Results

Table 1 shows there were no significant differences between the three autism subtypes in terms of age and gender (ps > 0.58). In addition, these three subgroups showed different levels of language skills (see Table 1) using ABC-language (aloof > ABO: p < 0.001, passive > ABO: p = 0.019), CARS-verbal communication (aloof > ABO: p < 0.001; aloof > passive: p = 0.008), ADOS-communication (aloof > ABO: p = 0.001; passive > ABO: p = 0.009) and GDS-language (aloof < passive < ABO, ps < 0.017) related language or communication subscales and different levels of severity (ABC-total: aloof > passive > ABO, ps < 0.017; CARS-total: aloof > ABO, passive > ABO; ps < 0.001; ADOS-total: aloof > ABO, passive > ABO, ps < 0.002; GDS-total: aloof < ABO, passive < ABO, ps < 0.001). The TD group showed better performance in the language domain and total scores using GDS compared to each autism subtype (ps < 0.001).

Vocabulary test PCA Results

A two-component model was identified with total variance explained of 90.37% for all items. Screen plot also supported a two-component model. These components were conceptualized as concrete words (Factor 1: food and drink, people, animals, games and routines, body parts, descriptive

Fig. 1 The performance in language skills (mean ± SEM) across three autism subtypes (aloof, passive and active but odd) and typically developing group (TD). Differences are shown for concrete words (A) and abstract words (B). ***p < 0.001, *p < 0.05 post-hoc Bonferroni corrected tests for three autism subtypes and TD group



words, places to go, vehicles, small household items, outside things, toys, action words, furniture and rooms, clothing) and abstract words (Factor 2: quantifiers and articles, helping verbs, connecting words, pronouns, question words, words about time). Based on these PCA results, we averaged the number of specific category words for further analysis.

Language Skills via the Vocabulary test in Different Autism Social Subtypes

Two-way repeated ANOVA with between-subject factor group (TD, aloof, passive and ABO) and within-subject factor word category as independent variables and the performance of language skill via the vocabulary test as the dependent variable showed significant main effects of word category [F (1,116) = 793.00, $p < 0.001$, partial $\eta^2 = 0.87$, $BF_{10} > 100$, better performance in concrete words relative to abstract words] and group [F (3,116) = 23.93, $p < 0.001$, partial $\eta^2 = 0.38$, $BF_{10} > 100$, TD > aloof, TD > passive, ABO > passive > aloof, ps < 0.001], as well as interaction [F (3,116) = 23.93, $p < 0.001$, partial $\eta^2 = 0.38$, $BF_{10} > 100$] indicating aloof performed worse than passive (concrete: $M_{aloof} \pm SD = 15.99 \pm 16.18$, $M_{passive} \pm SD = 28.60 \pm 15.85$, $p < 0.001$, $BF_{10} = 14.45$; abstract: $M_{aloof} \pm SD = 1.54 \pm 2.85$, $M_{passive} \pm SD = 4.22 \pm 3.43$, $p < 0.001$, $BF_{10} = 23.50$) and passive performed worse than ABO (concrete: $M_{passive} \pm SD = 28.60 \pm 15.85$, $M_{ABO} \pm SD = 40.94 \pm 7.46$, $p = 0.001$, $BF_{10} = 41.48$; abstract: $M_{passive} \pm SD = 4.22 \pm 3.43$, $M_{ABO} \pm SD = 7.98 \pm 2.75$, $p < 0.001$, $BF_{10} = 686.66$) and aloof performed worse than ABO (concrete and abstract: $p < 0.001$, $BF_{10} > 100$) in both word categories using Bonferroni correction multiple comparisons. In addition, individuals in the TD group exhibited better performance compared to aloof and passive autism subtypes for concrete words ($M_{TD} \pm SD = 45.68 \pm 1.59$, TD > aloof, TD > passive: $p < 0.001$, $BF_{10} > 100$; TD vs. ABO: $p = 0.982$) and compared to all three subtypes for abstract words ($M_{TD} \pm SD = 10.07 \pm 0.86$, TD > aloof, TD > passive, ps < 0.001, $BF_{10} > 100$; TD > ABO: $p = 0.035$, $BF_{10} = 99.40$, see Fig. 1).

The Relationship Between Autism Subtype Score and Language Skills

Firstly, Pearson's correlation analyses showed that ABC language scores, CARS verbal communication scores, ADOS communication scores were negatively associated with the average number of concrete (r = -0.388, $p < 0.001$, $BF_{10} = 147.26$; r = -0.595, $p < 0.001$, $BF_{10} > 100$; r = -0.448, $p < 0.001$, $BF_{10} = 1667$) and abstract words (r = -0.527, $p < 0.001$, $BF_{10} = 163{,}991$; r = -0.567, $p < 0.001$, $BF_{10} > 100$; r = -0.516, $p < 0.001$, $BF_{10} = 61{,}554$), respectively, indicating that the more severe autistic symptoms in language and communication, the worse is performance in language skills. This suggested that individuals with autism individuals showed different levels of language skills. In addition, GDS language scores are highly associated with vocabulary language scores in the autism group (concrete: r = 0.838, $p < 0.001$, $BF_{10} > 100$; abstract: r = 0.868, $p < 0.001$, $BF_{10} > 100$) but not in the TD group (concrete: r = -0.05, p = 0.80; abstract: r = 0.11, p = 0.56). Subsequently, Pearson's correlation analyses also indicated that scores for the aloof dimension negatively correlated with the number of concrete words (r = -0.597, $p < 0.001$, $BF_{10} > 100$, see Fig. 2 A) and abstract words (r = -0.623, $p < 0.001$, $BF_{10} > 100$, see Fig. 2B), however ABO scores were correlated in the opposite direction (concrete: r = 0.488, $p < 0.001$, $BF_{10} = 1728$,

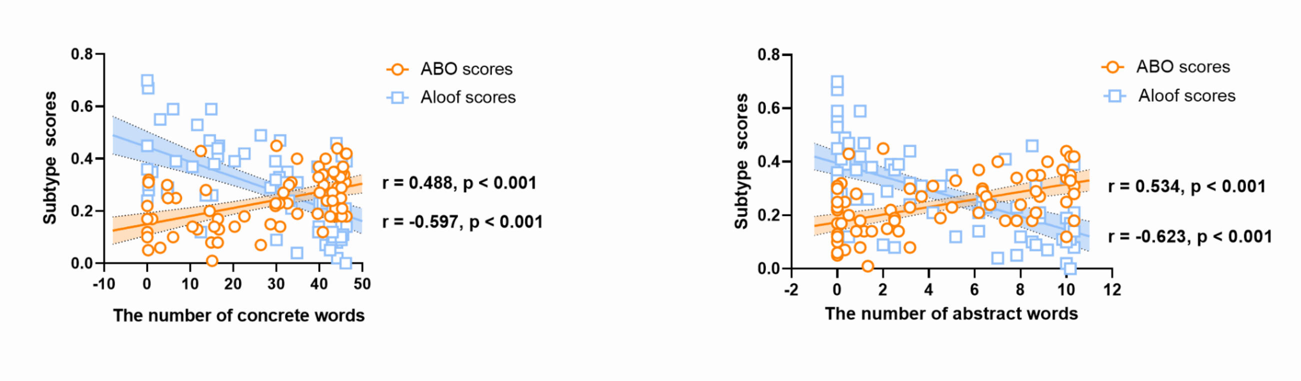


Fig. 2 Correlation analysis between aloof /active but odd (ABO) dimension scores and the average number of words (A, concrete; B, abstract) via the Chinese vocabulary test. Pearson's r and p values are presented

Table 2 Linear regression model results

Independent variable	Unstandardized Coefficients (B)	SEM	Standardized Coefficients	p
Concrete words				
Aloof score	-50.94	17.05	-0.48	0.004**
Passive score	16.69	23.75	0.08	0.49
ABO score	24.54	27.92	0.16	0.38
Abstract words				
Aloof score	-12.67	3.95	-0.51	0.002**
Passive score	0.53	5.50	0.01	0.92
ABO score	5.93	6.47	0.16	0.36

Table 2 Linear regression model results from the autistic children indicating the contribution of three subtype scores (aloof, passive and ABO) in language skills for concrete and abstract words separately. Statistic (both unstandardized and standardized coefficients B values) and p values for linear regression model analysis. SEM displays the standard error. ** p < 0.01

Fig. 2 A; abstract: $r = 0.534$, $p < 0.001$, $BF_{10} = 15{,}731$, Fig. 2B). There were significant differences between these coefficients (concrete: $p < 0.001$, 95% CI [-1.36, -0.78]; abstract: $p < 0.001$, 95% CI [-1.43, -0.84]) with bootstrap procedure (bootstrap = 10,000). There were no significant correlations between scores on the passive dimension and the language performance (concrete: $r = 0.001$, $p = 0.99$; abstract: r = -0.07, $p = 0.55$). Furthermore, we identified the contribution of these three subtype scores in language performance using a linear regression model and the results suggested that aloof scores significantly contributed to language performance in both abstract items ($F(3,69) = 15.20$, $p < 0.001$; Language performance = 6.46–12.66×aloof) and concrete items ($F(3,69) = 13.20$, $p < 0.001$; Language performance = 31.43–50.94×aloof, for more details see Table 2). In addition, we also found that reciprocal social interaction subscale scores on the ADOS negatively correlated with the number of concrete words (r = -0.521, $p < 0.001$, BF10 > 100,) and abstract words (r = -0.588, $p < 0.001$, BF10 > 100), similar to the pattern seen with language subscales of other assessments.

Discrimination Utility of Language Performance via the Vocabulary Test

Based on the above results, ROC analyses were first conducted to discriminate autism social subtypes and TD in language performance via the vocabulary test and good results were achieved (concrete: TD vs. aloof, AUC = 0.96, accuracy = 93%, sensitivity = 1, specificity = 0.85 [95% CI (0.71–0.97)], $p < 0.001$; TD vs. passive, AUC = 0.97, accuracy = 94%, sensitivity = 0.87 [95% CI (0.73–0.97)], specificity = 1, $p < 0.001$; TD vs. ABO, AUC = 0.88, accuracy = 85%, sensitivity = 0.87 [95% CI (0.74–0.97)], specificity = 0.83 [95% CI (0.65–0.96)], $p < 0.001$; abstract: TD vs. aloof, AUC = 0.96, accuracy = 95%, sensitivity = 1, specificity = 0.89 [95% CI (0.75-1)], $p < 0.001$; TD vs. passive, AUC = 0.98, accuracy = 94%, sensitivity = 0.87 [95% CI (0.73–0.97)], specificity = 1, $p < 0.001$; TD vs. ABO, AUC = 0.81, accuracy = 83%, sensitivity = 0.80 [95% CI (0.64–0.93)], specificity = 0.88 [95% CI (0.73-1)], $p < 0.001$).

Moreover, additional ROC analyses were conducted to differentiate between the three autism social subtypes in language performance via the vocabulary test. For concrete words, ROC analysis revealed an area under the ROC curve of 0.90, accuracy of 88%, sensitivity of 0.92, 95%CI (0.78-1) and specificity of 0.85, 95% CI (0.71–0.97), $p < 0.001$ between aloof and ABO; an AUC of 0.72, accuracy of 71%, sensitivity of 0.79, 95%CI (0.65–0.91) and specificity of 0.59, 95% CI (0.38–0.78), $p = 0.0008$ between aloof and passive; an AUC of 0.78, accuracy of 76%, sensitivity of 0.63, 95%CI (0.41–0.82) and specificity of 0.85, 95% CI (0.72–0.95), $p < 0.001$ between ABO and passive (see Fig. 3 A).

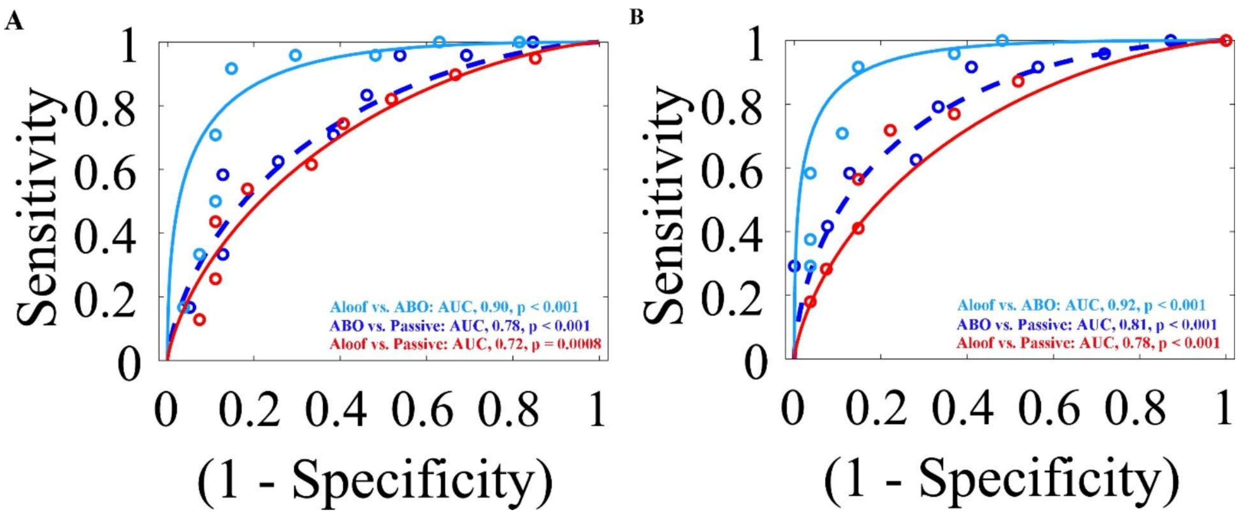


Fig. 3 Receiver operating curve (ROC) for discrimination of autism subtypes (light blue: aloof vs. ABO, red: aloof vs. passive; dark blue: ABO vs. passive) according to the performance in concrete (A) words and abstract (B) words. Active but odd, ABO; area under the ROC curve, AUC

For abstract words, ROC analysis revealed an AUC of 0.92, accuracy of 88%, sensitivity of 0.92, 95%CI (0.79-1) and specificity of 0.85, 95% CI (0.71–0.97), $p<0.001$ between aloof and ABO; AUC of 0.78, accuracy of 77%, sensitivity of 0.77, 95%CI (0.63–0.89) and specificity of 0.78, 95% CI (0.62–0.92), $p<0.001$ between aloof and passive; an AUC of 0.81, accuracy of 76%, sensitivity of 0.58, 95%CI (0.38–0.78) and specificity of 0.87, 95% CI (0.76–0.97), $p<0.001$ between ABO and passive (see Fig. 3B). Performance in differentiating between aloof and ABO was better than between aloof and passive in both concrete and abstract words ($z>2.04$, $ps<0.05$). In addition, a ROC analysis was also conducted to discriminate autism subtypes in behaviors not directly related with language skill, including the sensory subscale of ABC/anxiety response subscale of CARS /restrictive behaviors subscale of ADOS, and found these were unable to differentiate between them (ACC < 61%, $ps>0.10$).

Discussion

The present study firstly confirmed that the validity of a two-component model (concrete and abstract items) of vocabulary test is strongly supported by the high proportion of total variance (90.37%). According to this, negative correlations were found between autism symptom severity (via ABC, CARS, ADOS) but positive correlations between GDS and expressive language skill. The BASQ identified social subtypes exhibited different levels of expressive language skills (aloof > passive > active but odd). Using a categorical approach, for concrete items the aloof and passive subtypes exhibited worse performance compared to the TD group but not the active but odd subtype. For abstract items all three subtypes were impaired relative to the TD group. Furthermore, with a dimensional approach, while there was a strong negative association between dimension scores and language expressive performance across subjects on the aloof dimension, the opposite was found for ABO dimension scores and for the passive dimension there was no association with language skill at all. Linear regression models indicated that the aloof dimension contributed most compared to the other two dimensions in terms of language performance. A ROC analysis showed that language skill performance for both concrete and abstract words could effectively discriminate between the different autism subtypes.

The onset of language skill represents a special developmental milestone, however autism is characterized by the presence of problems or delays in both language and communication as well as social interactions (Eigsti et al., 2011). For example, individuals with autism have been reported to generate fewer words relative to TD in the first 30 s due to a probable initiation deficit (Carmo et al., 2017) and better performance on semantic fluency is associated with fewer autistic communication symptoms (Kenworthy et al., 2009). A recent meta-analysis has suggested that the decreased differences between individuals with autism and TD individuals over time might be associated with a narrow definition of autism which does not take into account the heterogeneity across individuals with autism (Rødgaard et al., 2019). Individuals with autism were divided into subgroups across three core symptoms separately via the social communication questionnaire (high vs. low severity, Whitten et al., 2018) or adaptive behavior trajectories (minimally verbal, verbal and atypical, Cohen & Flory 2019) and these subgroups showed distinct patterns. Our current categorical analysis has also shown heterogeneity in language development across autism social subtypes in young children, with the ABO subtype showing only minor problems, the passive subtype moderate ones and the aloof subtype the most severe ones both in the vocabulary test and other assessments (i.e. ABC, CARS, ADOS and GDS). These findings are consistent with an earlier study, which showed that aloof and passive subtypes showed severe communication problems (no speech: aloof 89%; 35% passive, 6% ABO) and repetitive behaviors and restricted interests (Wing & Gould, 1979).

More interestingly, the aloof and passive subtypes exhibited worse performance compared to the TD group but not the ABO subtype for concrete items, however all three subtypes were less proficient than the TD group for abstract items. Concrete word representations are richer to some extent than abstract ones (Kousta et al., 2011) and are assumed to have greater contextual associations (West & Holcomb, 2000). One study reported no significant difference between individuals with autism without intellectual disability or language impairment and TD individuals in word productivity in both imageability and concreteness characteristics (Tóth et al., 2022). Considering that children with the ABO social subtype of ASD are associated with higher intellectual abilities, better adaptive functioning and lower autism severity compared to the other two subtypes (Eagle et al., 2010), we did not find any difference in the number of concrete words these individuals produced relative to the TD individuals in our current study. On the other hand, not only do individuals with the aloof social subtype have severe problems with all aspects of language but also they have been reported to show fewer improvements after an intensive intervention (Beglinger & Smith, 2005), suggesting that different intervention strategies may be needed for this specific autism subtype.

While the different forms of subtyping for social phenotypes have primarily been used to classify individuals with autism as one of three specific subtypes based on which has the highest score, our analysis shows that using the individual scores of the different dimensions may also be informative. Overall, we found negative associations between scores on the aloof dimension and scores on the language vocabulary test across all autistic individuals and which were also consistent with communication/language subscale scores in ADOS, ABC and CARS assessments. On the other hand, there was a corresponding positive association with scores on the ABO dimension. This suggests that high scores on the aloof dimension of the BASQ are indicative of low language skill in individuals with autism whereas high scores on the ABO dimension are indicative of a level of language skill development seen in TD children. Possibly this may reflect the ABO dimension and subtype symptoms being more aligned with Asperger's (Ghaziuddin, 2008). Interestingly, scores on the passive dimension showed no association at all with language skill, although importantly children subtyped as passive were more likely to have language skill problems if they also had high scores on the aloof dimension. Indeed, the linear regression model used clearly indicates that aloof dimension scores contribute most to lower language skills irrespective of which categorical sub-type is exhibited.

There are some limitations to the present study. Firstly, the sample size is relatively small, especially for the ABO social subtype. Future studies with a larger sample size are needed to both replicate and refine our findings on the different language performances in the three social subtypes. Secondly, pragmatic language impairments are considered universal to individuals with autism, although there is substantial heterogeneity and complexity in structural language and verbal fluency difficulties (Brignell et al., 2018). The current study mainly focused on one specific aspect of language using a vocabulary test (expressive language), and future studies should include more aspects of pragmatic language such as sentence construction to validate the results. In the future, it will also be important to determine differential responses and generalization effects in the three autism social subtypes to the same speech and language therapy/intervention, for example aquatic speech and language therapy (Sourvinos et al., 2021).

Overall, the results demonstrate that the different social subtypes in autism are associated with different levels of language skill and can also be accurately identified using this metric. Importantly, our findings also suggest that use of patterns of individual scores on the dimensions of the three subtypes may be informative for predicting language problems with scores on the aloof dimension positively associated with them while ABO scores predict the opposite. In addition, considering that different social subtypes exhibited different levels of expressive language skills (aloof > passive > active but odd), future language interventions should be considered, particularly for individuals displaying the aloof subtype, or scoring high on the aloof dimension. Furthermore, early prediction of a trajectory of problematic language development is important for both parents and therapists to consider a greater focus on training language skills, especially expressive language. Importantly, the BASQ can be applied at the age of 2 years before the severity of problems with language skills has been fully realized in children with autism. Potentially some computer-based interventions (i.e. serious games and virtual learning environments) may help in this respect (Khowaja et al., 2020).

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