

RESEARCH ARTICLE

Assessing pragmatic language difficulties using the Revised Children's Communication Checklist-2. Exploratory structural equation modeling and associations with restricted and repetitive behaviors

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Abstract

In this paper, we investigated the psychometric properties of the Child Communication Checklist-Revised (CCC-R) for the first time with an English-speaking sample. We used a confirmatory application of exploratory structural equation modeling (ESEM) to re-evaluate the CCC-R's psychometric properties. We found strong support for its use as an assessment for pragmatic and structural language. Our second main aim was to explore associations between pragmatic and structural language and restricted and repetitive behaviors (RRBs), two hallmark characteristics of autism. We used the CCC-R and Repetitive Behavior Questionnaire (RBQ-2) to investigate these associations in a diverse non-clinical sample of children, taking a transdiagnostic approach. We intentionally excluded autism and other neurodevelopmental diagnoses to test, (1) the CCC-R in a broad sample and (2) the association between pragmatic language and RRB in children not already selected for that association. The sample comprised two groups of children, one was community sampled ($n = 123$) and the other ($n = 143$) included children with non-specific behavioral, emotional and/or cognitive difficulties referred to an assessment unit by schools. We found clear associations between pragmatic language difficulties and RRBs in both groups. Regression analysis showed that pragmatic language was the only significant contributor to RRBs even after Grammatical-Semantic score, age, sex, and socioeconomic status were controlled. The pattern was the same for both recruitment groups. However, the effects were stronger for the school-referred group which also had more pragmatic difficulties, grammatical-semantic difficulties and RRBs. A robust link between pragmatic language and RRBs, established in autism, has continuity across the broader non-clinical population.

Lay Summary

We studied language difficulties in a diverse child population. First, we tested the brief Revised Children's Communication Checklist-2 (CCC-R) and confirmed that it is a valid assessment tool in an English-speaking sample. Second, we found that CCC-R pragmatic language difficulties were associated with restricted and repetitive behaviors. As no child had a clinical diagnosis, we were able to identify this result in children not already selected to have this association. These findings have implications for understanding how repetitive behavior and language come to be related in autistic children.

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KEYWORDS

autism, exploratory structural equation modeling (ESEM), pragmatic language, restricted and repetitive behavior, revised Children's Communication Checklist-2 (CCC-R)

INTRODUCTION

Pragmatic language difficulties are a hallmark diagnostic symptom for autism. The co-occurrence of pragmatic language difficulties with repetitive behaviors (RRBs) is well established in autism but remains relatively unexplored in non-clinical populations. Given the dimensional nature of both pragmatic language and RRBs we investigated the continuity of this association across a diverse child population. The first step was to confirm the psychometric properties of a measure for pragmatic language. The second step was to use this measure together with a validated measure of RRBs to test the potential association between them.

Measurement of pragmatic language difficulties

Pragmatic language refers to the use and interpretation of language in a contextually appropriate manner or for the purpose of social connection. It encompasses the use of non-literal language, the ability to adapt the amount of information to the listener's needs, and conversational turn-taking and topic management (Levinson, 1983; Matthews, 2023). While definitions of pragmatics are extremely varied, it can be useful to limit this term to the verbal component of social communication (Carruthers et al., 2022; Matthews et al., 2018).

In addition to their significance in autism, pragmatic language difficulties are characteristic of other neurodevelopmental conditions (NDDs) (Geurts & Embrechts, 2008; Norbury, 2014). Pragmatic language difficulties are also common in children who are socially disadvantaged (Law et al., 2014) or who have problems with attention, learning, or memory (Hawkins et al., 2016). Indeed, large-scale population analysis shows that pragmatic language impairment is dimensional rather than categorical as it forms a continuum across clinical groups and general populations (Oi et al., 2017).

The widespread occurrence of pragmatic difficulties requires psychometrically sound assessment tools. A well-established questionnaire method that differentiates the features of pragmatic and structural (grammatical-semantic) language is the Children's Communication Checklist-2 (CCC-2; Bishop, 2003). The CCC-2 is a 70-item validated caregiver questionnaire originally designed as a screening tool for children with language impairments, including those with suspected autism. The CCC-2 has recently been revised, forming a new 39-item Revised Children's Communication Checklist-2 (Wellnitz et al., 2021) that provides a simplified measure of pragmatic and grammatical-semantic language. In developing

the CCC-R, the authors excluded a set of reverse-scored items together with a set of items unrelated to communication from the CCC-2. In doing so, the CCC-R overcame some measurement limitations of the original CCC-2 and increased its applicability for samples likely to have communication problems.

The psychometric properties of the CCC-R have been tested in a large German-speaking sample ($N = 839$). Exploratory factor analyses (EFA) found two factors (pragmatic and grammatical-semantic). Three clinically diagnosed groups, including a group diagnosed with autism spectrum disorder (ASD),¹ had elevated CCC-R total scores compared with a typically developing control group. The ASD group was distinct among these groups in having the highest level of pragmatic language difficulties (Wellnitz et al., 2021). However, the CCC-R has not yet been formally validated in English-speaking samples.

The first aim of the current study was to confirm the psychometric properties of the CCC-R in an English-speaking sample, using a confirmatory application of exploratory structural equation modeling (ESEM). ESEM has advantages over CFA and EFA approaches. It can estimate both CFA and EFA models, allow cross-loadings, enable better fit for the data than EFA and CFA models (Marsh et al., 2014), and is useful for scale construction. ESEM is also suitable when factors are associated with each other as is the case for the pragmatic and grammatical-semantic factors of the CCC-R. The current study therefore tested the reliability of the CCC-R, using a different analysis and testing a different non-clinical sample from a different European country.

Given evidence that pragmatic difficulties are continuously distributed (Oi et al., 2017), and are present, to various degrees, in non-diagnosed, and general populations, we used a different approach to Wellnitz et al. (2021), by testing the CCC-R in a diverse sample instead of in clinical samples. While Wellnitz et al. (2021) assessed specific categorically-defined clinical groups of children from 4 to 17-years, we set aside diagnostic criteria, taking a transdiagnostic approach (Astle et al., 2022). Our non-clinical sample comprised two groups of 6- and 7-year-old children from mainstream schools in different countries of the UK (England and Wales). One was a school-referred group identified by teachers for assessment of their strengths and needs due to behavioral, emotional, and/or cognitive difficulties in

¹Throughout the manuscript, we use identity-first language. However, in describing the study design of Wellnitz et al. (2021), or any study where children are selected and reported according to international diagnostic criteria, we follow the use of the term 'Autism Spectrum Disorder' as used by the researchers to refer to the diagnostic label from the Diagnostic and Statistical Manual of Mental Disorders (American Psychiatric Association, 2013).

the classroom. The other group was an unselected community sample without teacher-identified difficulties. In both groups diagnosed conditions were excluded. Several research studies of pragmatic language difficulties (Hawkins et al., 2016; Norbury et al., 2016) have used examples of these sampling methods to capture new insights into the correlates of pragmatic language difficulties (see Astle, et al., 2022 for the description of these studies). In the current study, we followed this approach, combining our recruitment groups into one sample for the ESEM analysis and then separating them to examine group similarities and differences when analyzing correlates of pragmatic difficulties.

The relationship between pragmatic difficulties and RRBs

The second aim of the study was to explore the association between pragmatic language and RRBs in a diverse non-autistic population. RRBs form a class of behaviors that include repetitive motor movements, preoccupations with objects, sensory over- or under-reactivity, and the strong preference for sameness and routine. The broader class of RRBs forms subtypes that include repetitive sensory and motor behaviors (RSMB) and Insistence on sameness behaviors (IS) (routines, rituals, and fixed interests). This stable two-factor structure has been validated consistently in autistic and in non-clinical community samples (see Uljarević et al., 2023 for systematic review). RRBs are highly prevalent in general child populations. They are adaptive for early physiological development and a mechanism for mastering the environment in early childhood (Evans et al., 2014; Gesell, 1928; Thelen, 1981). They are also included in the diagnostic criteria for autism where, unlike in the general population, they remain frequent and severe throughout the lifespan.

In autism, RRBs and pragmatic language aspects of social communication are found together within the diagnosis (Frazier et al., 2014; Jones et al., 2018; Mandy & Skuse, 2008; Martínez-González et al., 2022). Indeed, a diagnosis of ASD according to the criteria of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5; APA, 2013) cannot be made unless social-communication difficulties co-occur with RRBs. Yet despite co-occurrence of pragmatic language with repetitive behavior in the diagnosis of autism, to our knowledge, no previous research has studied this specific association in non-clinical samples of children. Yet if the association is robust in the absence of diagnosis this finding may have several implications. First for research, the finding of a link between the domains of pragmatic language difficulties and RRBs would warrant further conceptual analysis and further empirical study of how they come to associate and co-develop over-time. Second, for clinical and educational applications, an elevated level of association in some children could be a potential

subclinical signal: even if these children do not qualify for a specific autism diagnosis, they might require a certain level of support.

If we find an association between pragmatic difficulties and RRBs in non-autistic children, it is important to also explore whether the link is specific to pragmatic language or whether it can be explained by grammatical-semantic aspects of language. Evidence on the association between language and RRBs in non-autistic children is sparse. While one study with two-year-olds found that semantic language (vocabulary comprehension) was associated with one subtype of RRB (RSMB) (Larkin et al., 2017), another study with older children found no association (Keating et al., 2023). Pragmatic language was not tested in either of these studies. Therefore, in the current study we used regression analyses to assess the relative contributions of both pragmatic language and grammatical-semantic language difficulties to RRBs. We also controlled for other relevant factors such as socioeconomic status (SES), age, and sex.

The association between pragmatic difficulties and RRBs in children with and without behavior and emotional difficulties

Many RRBs (e.g., hand-flicking, repetitive use of language) are associated with elevated arousal levels in autistic children (Heathers et al., 2019; Willemsen-Swinkels et al., 1998). Where RRBs are linked to stress (for example if RRBs are blocked or disrupted) this might trigger difficult-to-manage behavior (Goodwin et al., 2022; Grahame et al., 2021; Lau et al., 2020). It is well known from research with both autistic and non-clinical samples that pragmatic language difficulties are associated with behavior difficulties measured by the Strengths and Difficulties Questionnaire (SDQ, Goodman, 1997) including hyperactivity, conduct, and peer problems (Helland et al., 2014; Miranda et al., 2023). Furthermore, the association with behavior difficulties is much stronger with pragmatic language difficulties than it is with structural (e.g., grammatic or semantic) language difficulties (Hawkins et al., 2016; Ketelaars et al., 2010; Law et al., 2014).

RRBs are not defined as behavior difficulties. However, they may raise the risk for them, via stress and anxiety even in non-autistic children, especially if they have language difficulties. In the current study, we would expect that the school-referred group with behavioral and emotional difficulties would show a stronger association between RRB and pragmatic language difficulties compared with the community-sampled group. The study used the Repetitive Behavior Questionnaire-2 (RBQ-2) to measure RRBs, a questionnaire widely validated across clinical and non-clinical populations (Keating et al., 2023; Leekam et al., 2007; Lidstone et al., 2014; Uljarević et al., 2017) with a robust two-factor structure.

The current study

In summary, using the confirmatory application of the ESEM, we aimed to confirm the reliability of the CCC-R in a diverse child population and then sought to use the CCC-R to test the potential association between pragmatic language and RRBs. We investigated whether two distinct types of RRBs, repetitive sensory and motor behaviors (RSMB) and Insistence on sameness behaviors (IS), would associate more strongly with pragmatic than with structural language difficulties and whether RSMB and IS might show a distinct pattern of associations. We were interested in evidence of similarity in the pattern of association across recruitment groups that would indicate continuity across the broader population. We were also interested in a differential association; whether this association would be stronger in a group recruited with school-referred behavioral and emotional difficulties, than in a community-sampled group.

METHOD

Participants

Participants were 266 children aged between 6; 0–7; 11 ($M = 6.75$, $SD = 0.49$) whose parents had completed both the CCC-2 and the RBQ-2. The sample encompassed two groups from different geographical regions in the UK.

Community sampled group

Participants were taking part in a longitudinal study of child development involving 206 mothers and children first recruited from health clinics and community organizations in the North-East of England, when their infants were 8 months of age. The current study focuses on the 126 (63 females, 63 males) whose parents consented to participate in a follow-up postal questionnaire study when children were 6–7 years of age (Mean age = 6.48 years, $SD = 0.14$). All children had been born full-term with no diagnosed medical conditions or developmental delay. At 6–7 years, three children had neurodevelopmental conditions, including one with autism, described clinically as Asperger syndrome, one with speech problems, and one with written language impairment described as dyslexia. These three children were excluded from analyses. This resulted in a final sample of 123 (62 females, 61 males).

The sample reflected the ethnic and socioeconomic background of the local and regional area with the majority (97%) White and British, and the full range of SES were represented from deprived to affluent. Hollingshead scores (Hollingshead, 1975) ranged from 11 to 66, with 44% of families classed as low SES

TABLE 1 Demographic information for sample.

	Community sampled group ($N = 123$)	School-referred group ($N = 143$)
Age	6.48 (0.14)	6.89 (0.56)
Sex	61 Male, 62 Female	87 Male, 56 Female
SES % of children in areas of deprivation	44%	42.8%

Note: SES was calculated using Hollingshead scores for the community sampled group and the Welsh Index of Multiple Deprivation for the school-referred group.

(parents with no post-16 education and no/menial/manual employment).

Assessment of the language skills of the sample was carried out at age 2 years using the Preschool Language Scales Third Edition (PLS; Boucher & Lewis, 1997) and at age 4–5 years using the British Picture Vocabulary Scale Second Edition (BPVS; Dunn et al., 1997). These assessments found mean standardized scores consistent with general population norms (e.g., BPVS mean score 103.20) ($SD = 12.99$) (see Larkin et al., 2017, Table 1). No information was available about parents' language difficulties or the presence of broader autism phenotype in relatives.

Repetitive behavior data from the sample at different ages has previously been used to answer different research questions (Larkin et al., 2017; Uljarević et al., 2017) but this is the first-time analysis has been made of CCC-2 scores and their relationship with RRBs.

School-referred group (children with emotional, cognitive, and/or behavioral difficulties)

Participants were referred by schools to the Neurodevelopment Assessment Unit (NDAU) at Cardiff University, Wales (<https://www.cardiff.ac.uk/neurodevelopment-assessment-unit>). The NDAU sees children between 4 and 7 years old who are referred for assessment by teachers and Additional Learning Needs Coordinators (ALNCOs) for having non-specific emotional, cognitive, and/or behavioral difficulties in the classroom. Referrals are based on teachers' judgment of children's classroom difficulties and are not based on any evidence of the children's RRBs. The NDAU is not a clinical unit, but instead assesses children across multiple psychological domains in line with the Research Domain Criteria framework (Cuthbert, 2014) with the aim of capturing the full profile of strengths and needs of each child. All children were referred from mainstream schools and no child had a clinical diagnosis of neurodevelopmental and/or learning disorders at the time of assessment. No data are collected on whether children eventually receive a clinical diagnosis because the NDAU is concerned with identifying patterns of psychological functioning and

behavior to help schools understand each child's profile. Language data for these children have previously been reported (Keating et al., 2023; Paine et al., 2021) showing that most children have verbal and non-verbal ability in the average range within 1 SD from the mean (see Keating et al., 2023, Table 1 and Figure S1). Among 4-to 7 year-olds, 20.9% had below average BPVS scores (1 SD below mean), 6.4% had below average Lucid Verbal Reasoning scores (Singleton, 2001) GL Assessment (2014). Lucid ability [computer software]. Lucid Research Limited., and 29.4% below average Lucid Non-verbal reasoning scores.

Data for children aged 6 and 7 years were selected for this study, to facilitate the same age range across the two samples. The NDAU sample contained 146 children (mean age = 6.88 years, $SD = 0.56$), three of whom were subsequently excluded due to missing data on the CCC-2, resulting in a final sample of 143 (56 females, 87 males). Of these 89.9% of mothers and 89.5% of fathers identified as Welsh, English, Scottish, or Irish. 42.8% had low SES as measured by being within the two highest quintiles of the Welsh Index of Multiple Deprivation (WIMD, Mean = 903.61, $SD = 567.89$) (Welsh Government, 2019). As with the community-sampled group, no information was available on whether relatives showed signs of a broader autism phenotype or had neurodevelopmental conditions or language difficulties.

Measures

Child Communication Checklist-2 revised

The Child Communication Checklist-2 Revised (CCC-R), a caregiver questionnaire, was developed by Wellnitz et al. (2021) as a concise, shortened, and simplified version of the CCC-2 (Bishop, 2003). Caregivers are asked to rate the strengths and weaknesses of their child's communication from 0 ("less than once a week") to 3 ("every day"). The development of the CCC-R involved removing CCC-2 item numbers 50–70 (reverse scored items) and items unrelated to communication (e.g., "has one or more overriding specific interests (e.g., computers, dinosaurs), and will prefer doing activities involving this to anything else"). This resulted in a final questionnaire of 39 items. Exploratory factor analysis by Wellnitz et al. (2021) revealed a two-factor structure, a "pragmatic" subscale of 26 items ($\alpha = 0.96$) derived from items in the original CCC-2 subscales of D-J and a "grammatical-semantic language" subscale of 13 items derived from items in the original CCC-2 subscales of A-C ($\alpha = 0.93$). The CCC-R uses raw item scores, and therefore higher scores indicate a greater incidence of language difficulties (max score for Pragmatic: 78, max score for Grammatical-semantic: 39).

Further psychometric analysis was carried out as part of the current study and described below.

The Repetitive Behavior Questionnaire-2

The Repetitive Behavior Questionnaire (RBQ-2) (Leekam et al., 2007) is a caregiver-report measure consisting of 20 items scored 1, 2, or 3 (never/rarely, mild/occasional, or marked/notable). Items include motor behaviors (e.g., rocking, repetitive hand/finger movements), sensory behaviors (e.g., special interest in the feel of surfaces), restricted interests (e.g., repeatedly playing the same music, game, or video), and routines (e.g., insisting that aspects of daily routine must remain the same). Parents are asked to rate behaviors shown in the previous month. Higher scores represent increased level of and/or impact of the RRB. The internal consistency of the total RBQ-2 scale is $\alpha = 0.85$ for neurotypical samples (Leekam et al., 2007) and $\alpha = 0.86$ for autistic samples (Lidstone et al., 2014). The two subscales of RSMB and IS also have excellent internal consistency in samples of young neurotypical children (Larkin et al., 2017), Cronbach's alpha (α) = 0.80 and $\alpha = 0.76$, respectively, and in older samples of autistic children (Lidstone et al., 2014), $\alpha = 0.79$; $\alpha = 0.83$, respectively. In the current study, the RSMB and IS subscales were calculated using items reported in the original factor analysis (Leekam et al., 2007 (Table 2)) which comprise items 1–6 and 8–10 for RSMB and 11 and 13–19 for IS. Scores for each two-factor subscale are averaged across the valid items completed to account for missing data. Internal consistency for the community sample in the current study was: $\alpha = 0.85$ for Total score, $\alpha = 0.84$ for RSMB, $\alpha = 0.75$ for IS. Cronbach's alpha for the school-referred sample in the current study was $\alpha = 0.91$ for Total score, $\alpha = 0.86$ for RSMB, and $\alpha = 0.88$ for IS.

Procedure

Data from the community sampled group were collected during a postal study. The RBQ-2 and CCC-2 were sent to parents who were asked to return the questionnaires by prepaid envelope. A follow-up phone call was made after the questionnaire submission, which gave the opportunity to ask about any missing questions. Ethical approval was granted from Local Health Service Ethics committees and University Ethics Committees, and parents provided written consent for their child to participate in the study.

Children from the school-referred group were seen at the assessment unit. Each child was given a battery of task-based assessments and their parents/guardians completed questionnaire measures including the RBQ-2 and the CCC-2. The research procedures were approved by

TABLE 2 Description of RBQ-2 and CCC-R scores for both datasets.

	Whole sample (<i>N</i> = 266)	School-referred group (<i>N</i> = 143)	Community sampled group (<i>N</i> = 123)	Max. Possible score	Score range (<i>N</i> = 266)	<i>p</i>
RSMB	1.46 (0.47)	1.57 (0.50)	1.29 (0.35)	3	1–3	<0.001**
IS	1.57 (0.51)	1.70 (0.56)	1.38 (0.35)	3	1–3	<0.001**
RBQ-2 Total	1.55 (0.44)	1.66 (0.48)	1.39 (0.30)	3	1–2.9	<0.001**
Grammatical-Semantic	5.27 (6.84)	7.70 (8.21)	2.41 (3.03)	39	0–37	<0.001**
Pragmatic	21.36 (15.98)	28.05 (16.79)	13.20 (9.62)	78	0–7	<0.001**
CCC-R Total	25.53 (19.20)	33.85 (20.59)	15.73 (11.08)	117	0–98	<0.001**

Note: RSMB: Repetitive sensory motor behavior; IS: Insistence on sameness, RBQ: Repetitive Behavior Questionnaire, CCC-R: Child Communication Checklist-Revised. *p*-value denotes significance of difference between school-referred and community-sampled groups.

the University's Ethics Committee. Parents/guardians gave written informed consent on behalf of the child and the child gave their assent.

Data analysis plan

Analyses were completed using SPSS 26 (IMB) and MPlus. Following data screening and summary of demographic characteristics, descriptive data were summarized for Wellnitz et al.'s (2021) CCC-R factor scores and RBQ-2 subscale scores (see Tables 1 and 2 for details of sample and descriptive statistics). Nonparametric statistics were also carried out to support the findings, as these variables were not normally distributed.

Confirmatory application of the ESEM (Asparouhov & Muthén, 2009; Marsh et al., 2014) was used to test the fit of the CCC-R two-factor model previously derived by Wellnitz et al. (2021) that comprised a pragmatic and a grammatical-semantic language factor. Robust maximum likelihood estimator was used. Analyses were also re-run using the polychoric correlations with the weighted least square estimator. Model fit was evaluated using the following recommended set of fit indices (Marsh et al., 2014): the comparative fit index (CFI); the Tucker-Lewis Index (TLI); the root mean square error of approximation (RMSEA); the standardized root mean square residual (SRMR). The following cut-offs were applied: (i) CFI and TLI values >0.90 indicating adequate and >0.95 excellent fit; (ii) RMSEA and SRSM values of <0.08 indicating adequate and <0.06 excellent fit, with RMSEA 90% confidence intervals <0.08 and the close fit-test with a *p*-value >0.05.

To explore the association between pragmatic and grammatic-semantic factors of the CCC-R and subtypes of RRBs, correlational and regression analyses were carried out. For the correlations between CCC-R and RBQ-2 scores, both Pearson's and Spearman's correlations were run. Bonferroni correction was applied for multiple comparisons ($0.05/4 = 0.0125$) and Meng's *Z* test was used to examine the relative strength of association between each language factor (grammatic/semantic

vs. pragmatic). A series of hierarchical regression analyses was conducted to examine the effect of each language factor on RRB. In the first analysis, Grammatical-Semantic scores were entered at Step 1, with Pragmatic scores entered at Step 2, the last step. In the second analysis, Pragmatic scores were entered in Step 1 instead of Grammatical-Semantic. In the third analysis, to check for the effect of demographic variables, Age, Sex, and SES were entered in Step 1, Grammatical-Semantic scores at Step 2 and Pragmatic scores at Step 3. Finally, scatter plots and summaries of inter-item coefficients were run to provide more detail of the nature of the associations found between RBQ-2 and CCC-R.

RESULTS

Data screening

There were no missing RBQ-2 data for the community sampled group. Two participants from this group (1.6%) were missing one item from the CCC-R and so the lowest possible score (0) was entered for these items, as per the scoring instructions. For the school-referred group, 19 participants (13.01%) were missing one item, and 2 participants (1.37%) were missing two items from the RBQ-2 and scores were averaged across the valid items to account for missing data. Fourteen participants (9.59%) had one missing item for the CCC-R, and two participants (1.37%) had two missing items and so the lowest possible score (0) was entered. Three participants from the school-referred sample provided no data for CCC-R. These three were removed from analysis leaving a sample size of 143.

Participant characteristics and descriptive statistics

Demographic details are shown in Table 1.

Descriptive statistics for RBQ-2 and CCC-R are in Table 2. As shown, there were group differences with

higher CCC-R scores (greater difficulties) for the school-referred group, both for pragmatics ($t(254) = -8.63$, $p < 0.001$) and for “structural” (Grammatical-Semantic) language ($t(262) = -6.80$, $p < 0.001$). This same pattern was found for Mann Whitney U-Tests. For RBQ-2 scores, the school-referred group also had higher levels of RRBs (RSMB: $t(270) = -5.21$, $p < 0.001$; IS: $t(270) = -5.59$, $p < 0.001$, total: $t(270) = -5.55$, $p < 0.001$). Again, the same result was found using Mann-Whitney U tests.

Exploratory structural equation modeling

The confirmatory application of the ESEM analysis focused on CCC-R item-level data. This showed that the two-factor model derived by Wellnitz et al. had good to excellent fit in the current dataset (CFI = 0.943; TLI = 0.937; RMSEA = 0.052 [90% CI: 0.047, 0.057], $p = 0.29$; SRMR = 0.065), and that individual items loaded onto the hypothesized factors, with the exception of the item 36, (“leaves off past tense-ed endings on words”), which in the original exploratory analysis loaded onto the Grammatical-Semantic language factor but in the current analysis showed higher loading on the Pragmatic factor. Given that item 36 had a better conceptual fit to the grammatical-semantic language factor, we retained the original placement of this individual item. All item loadings are reported in Table S1. The factor correlation was $r = 0.45$, $p < 0.001$, and $\alpha = 0.89$ for Pragmatic and 0.73 for Grammatical-Semantic subscale.

Correlational and regression analyses

The Wellnitz et al. (2021) CCC-R items (raw scores) calculated as CCC-R Pragmatic and Grammatical-Semantic subscales were adopted for all analyses. First, an initial series of correlational analyses between CCC-R and RBQ-2 subscales were conducted. Higher levels of RRB were associated with a greater incidence of language difficulties across both CCC-R subscales. Correlation tables and analyses are reported in full in Tables S2, S3a, and S3b. Table S2 shows that RRB scores correlated with demographic variables for the school-referred recruitment group only, with significant correlations between RRB higher scores and younger age (RSMB and IS) and male sex (RSMB only). Tables S3a and S3b show the correlations between RBQ-2 and CCC-R subtypes for both groups. There were moderate-sized correlations between the Pragmatic subscale and both subtypes of RRB (RSMB: $r = 0.48$, (community sampled) $r = 0.64$; (school-referred group) and IS: $r = 0.46$, (community sampled) $r = 0.63$ (school-referred group). However, for the Grammatical-Semantic subscale these correlations were weaker (RSMB: $r = 0.18$, $r = 0.29$; IS: $r = 0.31$, $r = 0.22$) (results for community and school-referred groups

presented respectively). Scatterplots (Figures S1–S4) provide further detail the bivariate relationships between each RBQ-2 subscale and CCC-R subscale showing the relative pattern of association for each group.

The hierarchical linear regression analyses reported in Tables 3a and 3b below show the contribution of both the Grammatical-Semantic and Pragmatic subscales to children’s RSMB and IS scores. While for both groups the Grammatical-Semantic scores make a significant contribution to both types of RRB, once the Pragmatic score is entered the contribution of the Grammatical-Semantic score becomes non-significant. This pattern applied to the IS subtype of RRB for both groups of children, and additionally to the RSMB subtype for the school-referred sample. For the community sample, grammatical-semantic scores also significantly contributed to RSMB though the contribution is smaller than that made by pragmatic scores.

Further regression analysis entering Pragmatic scores in reverse order at Step 1 (Table S4a and S4b), confirmed the same pattern as above. As before, for the community sample group only, Grammatical-Semantic scores also predicted RSMB; however, the contribution was smaller than that made by pragmatic scores. Finally, an analysis entering Age, Sex, and SES at Step 1, Grammatical-Semantic scores at Step 2 and Pragmatic scores at Step 3 (Table S5a and S5b) showed that demographic variables did not change the pattern of results in Table 3a and 3b above. Although there were significant additional effects at Step 1 of sex (boys with higher RRB scores) and age (younger children with higher RRB scores) in community and school-referred groups respectively, these effects became non-significant once Pragmatic scores were entered and only Pragmatic language predicted RRB.

Summaries of Pearson’s inter-item coefficients did not reveal any significant clusters of items that might explain the association. Positive item correlations were higher in the school-referred group and negative item correlations were negligible in size for both groups and rarely found in the school-referred sample. However no specific patterns were found among the items themselves.

DISCUSSION

The study aimed (1) to investigate the psychometric properties of the CCC-R with an English-speaking sample for the first time and (2) to characterize the association between pragmatic language difficulties and RRBs in a diverse child population. The findings indicate potential methodological and conceptual relevance for autism research, firstly in the use of the CCC-R as an assessment tool, and secondly in relation to the robust link found between pragmatic language and RRBs which shows continuity with autism.

This study added to the methodological development of the CCC-R (Wellnitz et al., 2021), a parent

TABLE 3 A Regression of Grammatical-semantic and Pragmatic subscales predicting RBQ-2 subtypes in community-sampled group.

Multiple regression	Repetitive sensory motor behaviors			
	B	SE	B	p
Step 1				
CCC-R Grammatical Semantic	0.048	0.010	0.401	<0.001**
R ²	0.161			
Adjusted R ²	0.153			
Step 2				
CCC-R Grammatical Semantic	0.027	0.010	0.226	0.008**
CCC-R Pragmatic	0.016	0.003	0.423	<0.001**
R ²	0.309			
Adjusted R ²	0.298			
Multiple regression	Insistence on sameness			
	B	SE	B	p
Step 1				
CCC-R Grammatical Semantic	0.026	0.011	0.221	0.015*
R ²	0.049			
Adjusted R ²	0.041			
Step 2				
CCC-R Grammatical Semantic	0.008	0.011	0.071	0.446
CCC-R Pragmatic	0.013	0.003	0.366	<0.001**
R ²	0.160			
Adjusted R ²	0.146			

Abbreviations: CCC-R, Child Communication Checklist-Revised; IS, insistence on Sameness; RSMB, repetitive sensory motor behaviors.

* $p < 0.05$; ** $p < 0.01$.

questionnaire that shows autism-specific elevated pragmatic difficulties compared with other clinical groups. We demonstrated its reliability and generalizability with data from a diverse sample of non-autistic children drawn from different geographical locations. The confirmatory application of the ESEM indicated that the two-factor model derived by Wellnitz et al. (Pragmatic, Grammatical-Semantic) had good to excellent fit even when tested with younger children than previously studied. All but one item loaded onto the hypothesized factors. The internal consistency was in the good to excellent range for both factors ($\alpha = 0.89$ for Pragmatic and 0.73 for Grammatical-Semantic subscale), albeit somewhat lower than that reported by Wellnitz et al. in clinical samples of German-speaking children ($\alpha = 0.96$ and $\alpha = 0.93$ respectively). Moderate correlations were found between the factors for both this study ($r = 0.45$, $p < 0.001$) and the Wellnitz et al., study ($r = 0.52$, $p < 0.001$). These results indicate support for the CCC-R as a psychometrically sound and concise measure of child language suitable for a diverse population sample.

The findings are also relevant to conceptual and clinical discussions about the association between social communication and RRBs in autism. We found that by setting aside diagnosis, a pattern of association intrinsic to autism diagnosis, was also found in two non-clinical

groups—a school-referred group with teacher-identified difficulties and a community-sampled group without difficulties. Correlations between pragmatic language difficulties and RRBs were sizable ($r = 0.6$ for school-referred sample and $r = 0.4$ – 0.5 for the community sample). Moreover, the regression analysis showed that even though Grammatical-Semantic scores made a significant contribution to RRBs, once the pragmatic scores were entered into the regression, the contribution of grammatical-semantic difficulties became non-significant. This pattern was the same for both groups for the Insistence on Sameness RRB subtype even after all other variables including Grammatical-Semantic language, age, sex, and SES were entered into the regression model. For the school-referred group, this effect was also found for the Repetitive and Sensory Motor RRB subtype.

The domains of RRB and social communication co-occur in those who are selected with a diagnosis of autism. By taking an agnostic approach to diagnostic categories in this study, we were able to address the question of whether there is a meaningful association between them for children not selected to have this co-occurrence. While some general population studies showed weak correlations between social-communication impairment and RRBs (Happé et al., 2006), other later studies have found moderate (Mandy et al., 2014, $r = 0.4$) or high correlations

TABLE 3B Regression of Grammatical-semantic and Pragmatic subscales predicting RBQ subtypes in the school-referred group.

Multiple regression	Repetitive sensory motor behaviors			
	B	SE	B	p
Step 1				
CCC-R grammatical semantic	0.012	0.006	0.192	0.031*
R^2	0.037			
Adjusted R^2	0.029			
Step 2				
CCC-R Grammatical Semantic	-0.008	0.005	-0.129	0.103
CCC-R Pragmatic	0.019	0.002	0.685	<0.001**
R^2	0.403			
Adjusted R^2	0.393			
Multiple regression	Insistence on sameness			
	B	SE	B	p
Step 1				
CCC-R Grammatical Semantic	0.020	0.006	0.265	0.003**
R^2	0.070			
Adjusted R^2	0.063			
Step 2				
CCC-R Grammatical Semantic	-0.001	0.006	-0.019	0.816
CCC-R Pragmatic	0.020	0.003	0.606	<0.001**
R^2	0.357			
Adjusted R^2	0.346			

Abbreviations: CCC-R, Child Communication Checklist-Revised; IS, insistence on Sameness; RSMB, repetitive sensory motor behaviors.

* $p < 0.05$; ** $p < 0.01$.

(Frazier et al., 2014, $r = 0.66$ – 0.95) supporting the proposal that these domains are connected (Constantino & Charman, 2015; Williams & Bowler, 2014).

The findings raise the question of why RRBs and pragmatic difficulties co-occur outside of an autism diagnosis. Previous research with siblings of autistic children who do not have a diagnosis themselves has found significant impairments on a range of CCC-2 subscales (Bishop et al., 2006). Furthermore, the children in the Bishop et al. (2006) study who performed poorly on the CCC-2 also had lower verbal IQs and some had diagnoses of language disorder. While it is difficult to conclude whether the children in the current samples might be part of the broader autism phenotype, due to lack of information about autistic relatives, neither the community-sampled group nor the school-referred group appear to have significant levels of language impairments.

Nevertheless, future research would benefit from more detailed investigation of subgroups of children who have language difficulties in grammatical-semantic ability to understand better the nature of the pragmatic language and RRB relationship. This is particularly important because the CCC-R does not yet have standardized scores for its two language subscales. Furthermore, while correlational analyses at the item level did not identify obvious clusters of high or negative correlations, more systematic analysis, especially with children of different

ages, could help identify subsets of items that consistently correlate.

Finally, the research offers preliminary evidence that may have practical implications for parents and school professionals who are managing children's general behavior difficulties. While we emphasize a similar autistic-like associative pattern in both recruitment groups, we also draw attention to the significance of the group differences. For example, the school-referred group of children with behavioral and emotional difficulties had mean pragmatic difficulties score that were twice that of the community sample. Likewise, they scored significantly higher than the community sample on the RRB measures. These findings have relevance to previous literature indicating that children with behavioral difficulties, (Donno et al., 2010; Law et al., 2014; Roy & Chiat, 2014; Saul et al., 2023) also have pragmatic difficulties. It is worth noting that the two groups also differed significantly on the grammatical-semantic measure (which was also twice as high in the school-referred than in the community sample).

RRBs differ from behavioral difficulties. From the beginning, RRBs are considered as an adaptive and regular part of early typical development (Evans et al., 2014; Thelen, 1981). They also continue to serve as tools for adaptation for autistic individuals for the purpose of regulating emotion, sensation, and information processing (Burack et al., 2021; Kapp et al., 2019; Keating

et al., 2023). Currently, there is insufficient evidence from community samples of children without NDDs or without existing behavioral and emotional difficulties to clarify the exact relationship between RRBs and behavior difficulties. However, our working proposal is that individual differences in behavior difficulties reflect the limits of a child's RRB adaptive strategy, which in turn, is highly influenced by cognitive and emotional development as RRBs serve different functions with age. The regulatory purpose of RRBs may break down under circumstances of overwhelm when other adaptive tools of cognitive and emotional flexibility are not available to regulate behavior. Further research is needed to investigate this and to consider how adaptive strategies change with age and cognitive development.

Limitations

While the recruitment of a diverse sample was a strength for testing the psychometric properties of the CCC-R, some aspects of the results did not match the findings of Wellnitz et al. (2021). To facilitate a more similar age comparison, the results from Wellnitz's data for the 4–8-year-old sample were accessed from the authors and summarized in Table S6. However, the means and standard deviations are still higher in the current sample than in the Wellnitz et al. (2021) samples.

There are also limitations in relation to the methods used. Both CCC-R and RBQ-2 are parent-informant questionnaires. Therefore, the associations found between CCC-R scores and RBQ-2 scores may simply reflect common variance in reporting by parents.

Conclusion

Our findings offer new evidence to support the CCC-R as a reliable language assessment, suitable for use in a diverse non-clinical population, as well as suitable for autistic and other NDD groups. By taking a dimensional approach, we found that children who have not been selected to have autistic co-occurrence of RRB and social communication by diagnosis, also show an association between pragmatic difficulties and RRB, and that this association remains significant independently of other language abilities or demographic features. These findings indicate that the CCC-R can now be widely applied to assess children in diverse populations. They also indicate the need for further understanding about how RRBs may both contribute to and be influenced by pragmatic language difficulties as children develop.

AUTHOR CONTRIBUTIONS

SL, JK and MU had the idea and designed the study. KAS, DH contributed to the conceptualization. SL led the article preparation. JK curated the data and JK and MU

carried out the statistical analysis. MU and DH contributed to the analysis plan and interpretation. The sample recruitment and data collection were led by SvG for Study 1 and by SL for Study 2. JK, MU, DH, KAS, and SvG contributed to the manuscript drafts. SL, SvG, and DH contributed to funding acquisition. All authors contributed to the article and approved the submitted version.

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CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/Supplementary material, further inquiries can be directed to the corresponding author.


ETHICS STATEMENT

The studies were reviewed and approved by School of Psychology Ethics Committees in two universities. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

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