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Cognitive Factors Contributing to Spelling Performance in Children with Prenatal Alcohol Exposure

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Abstract

Objective—Heavy prenatal alcohol exposure is associated with impaired school functioning. Spelling performance has not been comprehensively evaluated. We examined whether children with heavy prenatal alcohol exposure demonstrate deficits in spelling and related abilities, including reading, and tested whether there are unique underlying mechanisms for observed deficits in this population.

Method—Ninety-six school-age children comprised two groups: children with heavy prenatal alcohol exposure (AE, *n*=49) and control children (CON, *n*=47). Children completed select subtests from the WIAT-II and NEPSY-II. Group differences and relations between spelling and theoretically-related cognitive variables were evaluated using MANOVA and Pearson correlations. Hierarchical regression analyses were utilized to assess contributions of group membership and cognitive variables to spelling performance. The specificity of these deficits and underlying mechanisms was tested by examining the relations between reading ability, group membership, and cognitive variables.

Results—Groups differed significantly on all variables. Group membership and phonological processing significantly contributed to spelling performance. In addition, a significant group*working memory interaction revealed that working memory independently contributed significantly to spelling only for the AE group. All cognitive variables contributed to reading across groups and a group*working memory interaction revealed that working memory contributed independently to reading only for alcohol-exposed children.

Conclusion—Alcohol-exposed children demonstrated a unique pattern of spelling deficits. The relation of working memory to spelling and reading was specific to the AE group, suggesting that if prenatal alcohol exposure is known or suspected, working memory ability should be considered in the development and implementation of explicit instruction.

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Keywords

fetal alcohol syndrome (FAS); prenatal alcohol exposure; academic achievement; spelling; working memory

Introduction

Fetal alcohol spectrum disorders (FASD) are estimated to occur in 2 to 5 percent of school children and are characterized by lower general intelligence, central nervous system dysfunction, and behavioral and psychiatric disorders (Mattson, Crocker, & Nguyen, 2011). Given that these factors are associated with reduced academic achievement and higher rates of learning disabilities, it is not surprising that children with histories of prenatal alcohol exposure experience higher rates of poor educational outcomes (Streissguth, 1992), including deficits in all three domains of academic achievement: reading, writing, and math (Jirikowic, Carmichael Olson, & Kartin, 2008). Despite an emphasis on describing and treating mathematics deficits (Coles, Kable, & Taddeo, 2009; Crocker, Riley, & Mattson, in press), verbal academic difficulties have not been comprehensively examined. Spelling ability is understudied in this population, despite the view that "spelling correctly is perhaps one of the most valued, yet difficult skills in written communication" (Wanzek et al., 2006). Spelling deficits are under-recognized and under-treated in all populations, and are often associated with life-long negative outcomes (Berninger, Nielsen, Abbott, Wijsman, & Raskind, 2008; Berninger & Swanson, 2013; Stage, Abbott, Jenkins, & Berninger, 2003). Children unable to master writing and spelling skills are at risk for grade retention and lower rates of graduation, as they demonstrate particular difficulty in reading and high-stakes testing situations required for advancement (Berninger & Swanson, 2013; Graham et al., 2012; Jenkins, Johnson, & Hileman, 2004; Wanzek et al., 2006).

In one of the few studies testing the assumption that children with heavy prenatal alcohol exposure have verbal academic deficits, alcohol-exposed school-age children were found to score approximately 1.0–1.5 standard deviations (SD) below the mean of typically developing children on basic spelling and reading (Howell, Lynch, Platzman, Smith, & Coles, 2006). These literacy deficits continue throughout development. As adults, individuals with histories of heavy prenatal alcohol exposure have spelling abilities at a third grade level and reading abilities at a fourth grade level, which can lead to significant functional disability (Streissguth, 1992). The high co-occurrence of behavioral issues and diagnoses of attention-deficit/hyperactivity disorder (ADHD) (Fryer, McGee, Matt, & Mattson, 2007), which are also associated with higher rates of academic difficulties (DeBono et al., 2012; Mautone et al., 2009), may further contribute to difficulties in alcohol-exposed children.

The most effective spelling interventions target spelling directly and spelling outcomes are found to consistently improve when there is explicit instruction with multiple practice opportunities and immediate corrective feedback (Graham et al., 2013; Sampaio & Capellini, 2014; Wanzek et al., 2006). Teaching spelling is not only important to improve the skill itself, but there is a theorized bidirectional relationship between reading and spelling that *may* support the idea that improvement in one skill benefits the other (Clemens et al., 2014; Graham, Harris, & Chorzempa, 2002). This relationship is nuanced, as a longitudinal study of 1st through 4th graders found that a bidirectional model of reading and spelling fit best at the sentence level, however a reading-to-writing model fit better at the word level (Ahmed et al., 2014). The causal relationship between spelling and reading has been investigated through intervention studies, which find that the integration of phonological and orthographic knowledge, both skills specifically related to spelling, facilitates the acquisition of reading (Ouellette & Senechal, 2008). Likewise, direct, explicit encoding instruction led to improvement in both reading and spelling, demonstrating shared underlying mechanisms (Weiser & Mathes, 2011).

In considering skills that contribute to spelling performance, robust convergent evidence supports the importance of phonological processing and rapid naming performance above and beyond the effects of age, reading ability, and verbal IQ (Ackerman & Dykman, 1993; Berninger et al., 2008; Georgiou, 2008; Plaza & Cohen, 2003). There is substantial evidence supporting a developmental trajectory of phonological awareness and phonological processing abilities that predict future spelling performance in both impaired and typically developing populations (Berninger et al., 2006; Savage et al., 2005; Wanzek et al., 2006; Wocadlo & Rieger, 2007). Automatic rapid naming or speeded naming tasks also have been found to consistently contribute to spelling performance and may be due to the ability to quickly process letters and sounds (Berninger et al., 1992; Plaza & Cohen, 2003; Savage et al., 2005). In addition to the strong evidence supporting the importance of phonology and rapid naming, there has been debate regarding the importance of working memory for spelling performance and whether it contributes unique and significant variance (Brandenburg et al., 2014; Georgiou, 2008; McCallum et al., 2006). A study aiming to disentangle the effects of working memory and phonological skills on spelling found that working memory contributes to the prediction of early spelling ability and outperforms that of general intelligence and phonological decoding (Preßler, 2013). The authors also found that phonological awareness mediates the role of working memory on early literacy outcomes, suggesting that both are key cognitive predictors of spelling.

The relative contribution of underlying cognitive skills to spelling performance may differ across developmental populations with differing etiology of dysfunction or the presence of other cognitive difficulties. For example, past studies investigating the contribution of phonological awareness, rapid naming, and working memory to the spelling and reading skills of children with severe reading disabilities and developmental disabilities found both unique and overlapping contributory cognitive skills for reading and spelling, suggesting that there are both shared and independent predictors for these skills (Brandenburg et al., 2014; Byrne, MacDonald, & Buckley, 2002; Fletcher & Buckley, 2002; Kim et al., 2014).

While verbal academic deficits are assumed to be associated with prenatal alcohol exposure, there is a paucity of empirical evidence supporting these impairments and no investigations to our knowledge that have investigated the cognitive mechanisms that contribute to these abilities, which may differ in alcohol-exposed children as they present with a unique neurobehavioral profile. Thus, the current study aimed to (1) examine whether alcohol-exposed children have deficits in spelling and related cognitive abilities (phonological processing, rapid automatized naming, working memory) and (2) determine if a distinct relationship between contributing cognitive factors and spelling achievement exists for this population, compared to typically developing control children. As reading and spelling are related, we will also consider the relations between contributing cognitive abilities and word reading to determine the specificity of the results for spelling. We hypothesized that children with heavy prenatal alcohol exposure would perform significantly worse than control children on all measures and that there would be group differences in the relations between underlying cognitive abilities and spelling. We hypothesized that alcohol-exposed children would demonstrate a diffuse pattern of impairment, and that the relations between all three cognitive abilities and spelling would be more robust in children with prenatal alcohol exposure indicating the need to recruit greater cognitive resources for successful performance as is this case in other populations, such as children with ADHD (Re et al., 2014)

Method

General Method

Children and their primary caregivers were recruited as part of a larger, ongoing research protocol at the Center for Behavioral Teratology. During a 4-hour neuropsychological test battery, subjects were administered select subtests from valid, standardized neuropsychological assessments including tests of achievement and cognitive abilities. As part of this battery, a full scale IQ (FSIQ) score was estimated using the Wechsler Intelligence Scale for Children-Fourth Edition (Wechsler, 2004). Trained examiners who were blind to subject group conducted testing and all tests were scored by at least two individuals, who were also blind to subject group to ensure accuracy and prevent bias. The Institutional Review Board at San Diego State University approved the project. Subjects and caregivers received compensation for participation.

Subjects

Children between the ages of 8 and 16 years (M = 12.47, SD = 2.35) were recruited by word of mouth, clinical referral, and community outreach. Children were excluded from participation if they had suffered a significant head trauma, had loss of consciousness greater than 30 minutes, had other known causes of cognitive deficiency, were not fluent and primary English speakers, were adopted from abroad after the age of five years or two years prior to assessment, or had a psychiatric or physical disability that prevented successful completion.

Two groups of children were included in the present study: children with a history of heavy prenatal alcohol exposure (AE, n = 49) and control children with very minimal or no prenatal alcohol exposure (CON, n = 47). Children in the AE group had confirmed histories of heavy prenatal alcohol exposure, defined as maternal consumption of more than 4 alcoholic drinks per occasion at least once per week or at least 14 drinks per week (on average) during gestation. Prenatal alcohol exposure was confirmed retrospectively using medical history, birth records, social services records, or maternal report, when available (for details, see Mattson et al., 2010). In most cases in the AE group, precise measures of

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alcohol consumption were unavailable. In these cases, mothers were reported to be "alcoholic" or alcohol abusing or dependent during pregnancy. All subjects were evaluated by a dysmorphologist and a diagnosis of FAS was determined by the presence of two or more key facial features (short palpebral fissures, smooth philtrum, thin vermillion border) in addition to microcephaly (head circumference ≤10th percentile), and/or growth deficiency (≤10th percentile for height or weight) (Jones et al., 2006; Mattson et al., 2010). Within the AE group, 10 children (20.4%) met criteria for FAS. Children in the CON group had no prenatal alcohol exposure (approximately 75%) or very minimal exposure (i.e., no more than one drink per week on average and never more than two drinks per occasion during gestation.

Based on the Computerized Diagnostic Interview Schedule for Children Version IV (C-DISC-4.0), children who had 3 or more criteria consistent with ADHD (i.e., clinical or subclinical levels of symptoms) were excluded from the CON group (Shaffer, Fisher, Lucas, Dulcan, & Schwab-Stone, 2000). Children with ADHD symptoms were not excluded from the AE group given the high rates of ADHD in this population (Fryer et al., 2007). Greater than 70% of the AE group had clinical symptoms of ADHD according to the C-DISC-4.0.

Measures and Procedure

Wechsler Individual Achievement Test – Second Edition (WIAT-II) (Wechsler, 2005)—Spelling achievement was assessed using the WIAT-II spelling subtest. The WIAT-II was normed on a national sample representative of the U.S. population and the spelling subtest has an average reliability coefficient of .93 – .96 across our age range (Abbott, Berninger, & Fayol, 2010). The WIAT-II spelling subtest has a moderately high correlation (.79) with the WRAT-3 spelling subtest indicating convergent validity. Written spelling was tested for words of increasing difficulty. Stimuli were pronounced once, then used in a sentence to provide contextual cues, and then pronounced for a final time. There was no time limit during this task, decreasing the chance of processing speed significantly impacting performance. The dependent variable used in the current analysis was an age-standardized scaled score of correctly spelled words and has high ecological validity, as the task is representative of school spelling tests. Subjects also took the WIAT-II reading subtest, which is an oral single word reading untimed assessment with reliability and validity comparable to the spelling subtest.

NEPSY- Second Edition (NEPSY-II) (Korkman, Kirk, & Kemp, 2007)—Selected subtests of the NEPSY-II, described below, were given to assess cognitive variables that are theoretically related to literacy and spelling in both typically developing children and in clinical populations, including children with ADHD and periventricular brain injury and very low birthweight (Berninger et al., 1992; Downie et al., 2005; Korkman & Pesonen, 1994; Plaza & Cohen, 2003; Savage et al., 2005). The dependent variables are all scaled scores and are normed by age. The NEPSY-II is a flexible pediatric neuropsychological assessment measure that has co-normed subtests that demonstrate strong convergent and discriminant validity as well as high internal reliability (Korkman et al., 2007). The normative sample was stratified by age, sex, race, ethnicity, geographic region, and parental education level.

Phonological Processing (PP): This subtest comprises two phonological processing tasks aimed to comprehensively assess phonemic awareness. The first task, Word Segment Recognition, requires identification of words from word segments, and the second task, Phonological Segmentation, is a test of elision (the omission of a vowel, consonant, or syllable in pronunciation) designed to assess phonological processing for syllables and phonemes. The child is asked to repeat a word and then create a new word by omitting or substituting a syllable or phoneme with no time restraint.

Speeded Naming (SN): This timed subtest (300 seconds) aims to assess rapid automatized naming and rapid semantic access to production of names of letters and numbers. Eight-year-old subjects were given an additional 300 second trial, which included the production of the names, sizes and colors of different shapes. The child is shown an array of stimuli and asked to name them as quickly as possible without making mistakes.

Word-List Interference (WM): This subtest assesses verbal working memory, repetition, and word recall following interference. The child is presented with two series of words and asked to repeat each sequence following its presentation, and then he or she recalls each series in order of presentation. Given the nature of the task, both short-term span and working memory are required for successful performance (Korkman et al., 2007). There was no time limit.

Finger Tapping (FT): This subtest assess finger dexterity, motor speed, and rapid motor programming. The child is asked to copy a series of finger motions demonstrated by the examiner as quickly as possible.

Statistical Analysis

Statistical analyses were conducted using SPSS 22 (SPSS, 2013). Data for the WIAT-II spelling were assessed for outliers using boxplot analysis. Two control subjects were identified as outliers and thus were removed from all analyses, resulting in the total of 96 subjects in the final analyses. All cognitive variables were centered prior to conducting the analyses to reduce multicollinearity, particularly in instances where predictor variables interact with one another.

Demographic Information and Group Differences—Demographic information was analyzed using chi-square (race, ethnicity, sex, handedness) and analysis of variance (ANOVA; age, socioeconomic status [SES] as measured by Hollingshead (Hollingshead, 1975), and full scale IQ [FSIQ]) statistical techniques. To confirm the relationship between the cognitive variables and spelling achievement, within-group Pearson correlations were conducted. Relationships between spelling, reading, and the underlying cognitive variables were also assessed using correlations. MANOVAs were used to examine group differences on WIAT-II measures (spelling and word reading) and NEPSY measures (speeded naming, phonological processing, and working memory). Given their relation to academic achievement, age, race, ethnicity, SES, and sex were considered as possible covariates and included if they were significantly related to the dependent variables (WIAT-II spelling and word reading) and did not interact with group membership. Alpha was set at $p < .05$ for all analyses.

Contributing Cognitive Factors—To determine the contributions of group and the cognitive variables (SN, PP, WM) on spelling, a hierarchical regression was conducted with group entered on step one, the main effects entered on step two, and the group × cognitive variable interactions entered on step three to examine how the cognitive variables and their interaction with group membership impacted spelling over and above independent group differences and main effects. This methodology was repeated for the WIAT-II word reading subtest to test the specificity of observed underlying mechanisms of spelling deficits.

Results

Demographic Information

There were no significant group differences on age [F (1, 94) = .785, p = .378], SES [F (1, 94) = 3.676, p = .058], handedness [χ^2 (df = 3) = 1.070, p = .784], ethnicity [χ^2 (df = 2) = 4.855, p = .088], sex [χ^2 (df = 1) = 2.637, p = .104], or race [χ^2 (df = 3) = 4.575, p = .206]. However, the AE group had significantly lower FSIQ scores than the CON group, [F (1, 94) = 53.669, p < .001]. Demographic information is presented in Table 1.

Evaluation of Covariates

The appropriateness of including theoretically implicated demographic variables as covariates was evaluated. Spelling achievement was not significantly associated with any demographic variables (ps > .100). Reading was significantly associated with SES (p = .041), but no other demographic variables (ps > .383). While group differences existed on FSIQ, there are statistical, theoretical, and methodological challenges in using it as a covariate for this sample (Dennis et al., 2009). Therefore, the hierarchical regression analysis of reading performance was repeated with SES included in step one.

Between-Group Differences

The AE group performed significantly worse than CON on spelling, reading, and all NEPSY-II variables (ps < .001). Group means are presented in Table 1.

Correlational Analyses

Correlational analyses are presented in Table 2. Correlations between all variables and spelling were significant in the AE group (rs > .410; ps < .001); however, only PP was significantly correlated with spelling achievement in the non-exposed group (r = .487; p < .001). All cognitive variables were associated with reading for both exposed and non-exposed children (ps < .050).

Hierarchical Regression

To examine the contribution of the cognitive variables in the explanation of spelling performance, a hierarchical multiple regression analysis was performed. Spelling achievement served as the dependent variable. Group (AE, CON) was entered as the independent variable in step one, and the standard scores of the NEPSY-II cognitive variables (PP, SN, WM) were entered in step two, and their interactions with group were entered in step three. Before the analysis was performed, independent variables were examined for collinearity. Results of the variance inflation factor (all less than 4) and collinearity tolerance (all greater than .250) suggested that the estimated betas were well established in the regression model (Hair, Anderson, Tatham, & William, 1995; Neter, Wasserman, & Kutner, 1989).

Regression results are reported in Table 3. The final model including group, cognitive variables and interactions accounted for 52.2% of the variance. In step one, group accounted for a significant amount of variance in spelling performance [$R^2 = .148$, $F(1,92)= 15.921$, $p < .001$]. In step two, the cognitive variables accounted for a significant amount of variance beyond the effect of group [$\Delta R^2 = .322$, $F\Delta\ (3,89)= 18.011$, $p < .001$]. In this second step, PP was a significant contributor, and group no longer accounted for significant variance. The inclusion of the interactions on step 3 was also significant [$\Delta R^2 = .053$, $F\Delta\ (3,86)= 3.155$, $p = .029$]. Only the WM x group interaction was a significant contributor. This interaction was followed up using simple effects to analyze the contribution of WM to spelling at each level of group to determine the amount of variance accounted (R^2). A simple regression was run to examine the zero-order relations and quantify the contribution of WM to spelling in each group, respectively. Working memory significantly contributed to spelling in the AE group ($p < .001$) accounting for 38.6% of the variance, but not in CON ($p = .321$), accounting only for 1.4% of the variance. These results are presented in Figure 1. This was also true in a regression model including PP and SN entered into step 1 and WM entered into step 2. The addition of WM on step 2 contributed significant variance [$\Delta R^2 = .085$, $F\Delta\ (1,44)= 7.759$, $p = .008$] above and beyond the two other variables in the AE group, whereas step 2 was not significant in CON [$\Delta R^2 = .017$, $F\Delta\ (1,42)= 0.951$, $p = .335$]. Of note, there are two individuals in the AE group who had much lower scores (spelling scores of 43 and 46, respectively, while the next spelling score was 67). While they are not considered outliers in the boxplot analysis due to the greater variability with this group, we re-ran the interaction without these two subjects to minimize any potential undue influence and found the same results: a significant contribution for AE (27.4% of variance, $p <.001$), but not CON (same as above).

WIAT word reading and spelling are highly correlated, and thus we were interested in whether the same cognitive mechanisms were implicated in both academic domains or if the observed relations were specific to spelling. The same regression analyses were conducted for word reading as the dependent variable, and the results are reported in Table 4. The final model including group, cognitive variables, and interactions accounted for 65.8% of the variance. In step one, group accounted for a significant amount of variance in spelling performance [$R^2 = .149$, $F(1,92)= 16.063$, $p < .001$]. In step two, the cognitive variables accounted for a significant amount of variance beyond the effect of group [$\Delta R^2 = .469$, $F\Delta\ (3,89)= 36.433$, $p < .001$]. In this second step, PP, WM, and SN were significant contributors, and group no longer accounted for significant variance. The inclusion of interactions on step three was also significant [$\Delta R^2 = .040$, $F\Delta\ (3,86)= 3.322$, $p = .024$]. The WM x group interaction was the only significant contributor at this step. This interaction was followed up using simple effects in the same two models as above, to analyze the contribution of WM to reading at each level of group. In the simple regression, there is a significant relationship between working memory and reading for both groups, however it is considerably stronger in the AE group accounting for 53.1% of the variance, which is driving the interaction (AE: $R^2 = .729$, $p < .001$; CON: $R^2 = .117$, $p = .020$). Examining the effects in the model including PP and SN, the addition of WM on step 2 contributed significant variance above and beyond the two other variables in the AE group [$\Delta R^2 = .123$, $F\Delta$ (1,44)= 18.264, $p < .001$], whereas step 2 was not significant in CON [$\Delta R^2 = .006$, $F\Delta$ (1,42)= 0.360, $p = .552$]. One of the alcohol-exposed individuals who had very low spelling demonstrated very low reading, and the follow-up analyses were rerun without that subject, with the same results; AE accounted for 49.2% of the variance ($p < .001$). The word reading analyses were repeated with SES included as a covariate, and the results also remained the same.

Influence of Sensorimotor Abilities

The main analysis focused on the influence of cognitive variables, and thus the analysis was re-run with a measure of sensorimotor ability. The only sensorimotor NEPSY-II task normed for our age range is Finger Tapping (FT), a measure of finger dexterity, motor speed and rapid motor programming (Korkman et al., 2007). Alcohol-exposed children performed lower than controls (AE: 8.79, CON: 10.32, $p\eta^2 = .061$). Although expected, there was no significant correlation between FT and spelling ($r = .252$, $p = .084$) for alcohol-exposed children, nor was there a significant correlation for FT and reading ($r = .260$, $p = .075$). Of the other cognitive variables, FT was only significantly related to SN ($r = .260$, $p = .035$). In non-exposed children, FT was also not associated with spelling or reading (ps > .780), nor was it significantly related to any of the other variables (ps > .149). To examine the contribution of the sensorimotor ability in the explanation of spelling and performance, the same hierarchical multiple regression analyses were performed. As above, Group was entered as step 1, the standard scores of the centered NEPSY-II variables entered on step 2 (PP, SN, WM, FT), and their interactions with group were entered on step three. The inclusion of FT (as a main effect and the interaction) did not change the results and all three steps for both regressions remained significant. Results of these analyses are found in Supplemental Table 1.

Post-Hoc Behavioral Analysis

Due to the high rate of ADHD in children with prenatal alcohol exposure, we examined the contribution of ADHD-related behaviors to spelling within the alcohol-exposed group, using the Child Behavior Checklist (CBCL) ADHD T-scale (Achenbach & Rescorla, 2001). We found no significant correlation between the CBCL ADHD scale and spelling ($r = -.218$, $p = .136$). The ADHD T-scale was negatively correlated with reading ($r = -.288$, $p = .047$) although to a weaker degree than any of the cognitive mechanisms (see Table 2). We further assessed the effect of ADHD symptomology on spelling and reading within the alcohol-exposed group using a basic regression model that included CBCL ADHD T-score, PP, WM, and SN predicting both academic skills, respectively. We found that ADHD behaviors did not contribute significant variance to spelling (p=.497) or reading (p=.172).

Discussion

These findings substantiate the limited research indicating that children with heavy prenatal alcohol exposure perform significantly worse on spelling, reading, and pre-literacy abilities (phonological processing, rapid automatized naming, and working memory) compared to those without such exposure (Adnams et al., 2007; Carmichael Olson, Feldman, Streissguth, Sampson, & Bookstein, 1998; Howell et al., 2006; Pei, Rinaldi, Rasmussen, Massey, & Massey, 2008; Streissguth et al., 1994). The current study is the first, to our knowledge, to examine underlying cognitive mechanisms of spelling and reading in alcohol-exposed children, a theoretical weakness noted in one of the few previous studies on literacy in this population (Adnams et al., 2007).

Our findings indicate that phonological skills were important for spelling, above the effect of alcohol exposure. These results confirm the importance of phonological processing for spelling across populations (Berninger & Swanson, 2013; Rapcsak et al., 2009) and extend the understanding of how poor phonological processing in alcohol-exposed children may manifest in school settings (Jacobson, 1998; Rasmussen & Bisanz, 2011). In contrast, whereas phonological processing contributes to spelling and reading across groups, working memory uniquely contributed to performance for alcohol-exposed children, but not controls, when considering the variance accounted for by phonological processing and speeded naming. That working memory is more strongly related to spelling and reading deficits in children with heavy prenatal alcohol exposure, along with the executive functioning insufficiency in this population, implies a broader executive dysfunction that interferes with learning (Mattson et al., 2011; Mattson et al., 2013). For word reading, phonological processing, rapid naming, and working memory all contributed significantly across groups, suggesting a potentially more complex and wider set of contributing skills needed for reading versus spelling.

While rapid automatized naming was not a significant contributing factor for spelling, it did significantly contribute with reading. This suggests there may be a weaker relation between rapid naming and spelling that is overshadowed by the effects of other cognitive abilities and may be indicative of impaired processing speed in this alcohol-exposed sample, which could indirectly contribute to poor spelling performance (Burden, Jacobson, & Jacobson, 2005; Glass et al., 2013). Rapid naming was timed, which may further limit its direct relation to the untimed spelling subtest, however it's relation to reading (also untimed) possibly suggests distinct networks of interrelated abilities. We found no differential relation between our measure of fine motor skills and spelling or reading performance. While graphomotor skills and procedural learning are associated more so with spelling deficits than reading deficits, our measure may not have been sensitive enough to demonstrate these differences (Berninger & Swanson, 2013).

Even though there is substantial literature on spoken language and reading deficits (Vaughn & Wanzek, 2014; Wanzek et al., 2010), there are fewer studies that specifically target spelling deficits (Berninger & Swanson, 2013; Graham et al., 2012). Nonetheless, improvement of spelling in children has been achieved (Sampaio & Capellini, 2014), with long-term gains in as little as one semester of behavioral intervention at a first grade level and is most effective for young children at risk for subsequent reading and spelling deficits (Vellutino et al., 1996). An emphasis on instruction of phonemic awareness and alphabetic understanding, including letter-sound correspondences, letter-writing resulting from explicit instruction, letter names, and an integration of both skills (e.g., what is the first sound in this word? now write the letter), has also been effective (Berninger & Swanson, 2013; Graham et al., 2013; Graham et al., 2012; Santoro, Coyne, & Simmons, 2006). The most effective interventions include explicit instruction, multiple practice opportunities, and immediate corrective feedback (Berninger & Swanson, 2013; Graham et al., 2012; Vaughn & Wanzek, 2014; Wanzek et al., 2006; Wanzek et al., 2010). Focusing on phonological awareness and auditory discrimination training for school-age students has resulted in significant gains in those targeted areas (Alexander, Andersen, Heilman, Voeller, & Torgesen, 1991; Berninger, Dunn, Lin, & Shimada, 2004; Berninger & Swanson, 2013; Graham et al., 2013; Graham et al., 2012; Rapp & Kane, 2002). Comparisons of spelling treatments have found that combination programs, interventions with concurrent phonological training and letter-knowledge instruction, are better than focusing on a single ability at a time (Schneider, Roth, & Ennemoser, 2000).

While directly providing spelling and reading instruction is most efficacious, the previous literature suggest the need for future research to address or consider working memory *in addition* to explicit instruction to improve spelling and reading achievement (Byrne, Buckley, MacDonald, & Bird, 1995; Byrne et al., 2002; Cain, Oakhill, & Bryant, 2004; Fletcher & Buckley, 2002; Gathercole, Alloway, Willis, & Adams, 2006; Laws, Buckley, MacDonald, & Broadley, 1995). Interventions aimed only at teaching spelling may be effective in other populations, however such a strategy may fail to succeed to remediate children with FASD to their full potential. Rather, forthcoming investigations should consider poor working memory when planning explicit spelling instruction, as modifications may be necessary for it to be most efficacious. Traditional instruction methods may result in an artificially low ceiling for spelling achievement in FASD.

Understanding the presence of poor working memory and the potential effects on academic learning may contribute to the success of evidence-based practices in this population, which otherwise may not be fully efficacious. Recent studies have demonstrated the importance of addressing and considering working memory capacity in children with ADHD, who make increased spelling errors when facing a high phonological working memory load (Re et al., 2014). Likewise, there is new evidence from a fractions intervention program suggesting differential treatment outcomes for children with very weak working memory (Fuchs et al., 2014). Understanding the components of working memory and their relations to academic success may also be important as they may differ for spelling, compared to reading (Brandenburg et al., 2014).

A longitudinal study found that even after 3–4 years of formal education, children with severe speech impairment performed significantly lower than typically developing children on all literacy indicators, including spelling, despite the development of good phonological abilities (Sandberg, 2006). An analysis of the spelling errors indicated that children did not use their phonological abilities efficiently as a result of poor working memory, demonstrated by poor rehearsal skills. Further, working memory and reading efficiency (i.e., speed of reading a list of words/non-words) accounted for significant variance of spelling performance in children with ADHD (DeBono et al., 2012). Working memory also significantly contributed to spelling, in addition to phonological processing, in children with very low birth weight and periventricular brain injury (Downie et al., 2005). The severity of overall literacy difficulties is significantly associated with working memory and phonological processing abilities, suggesting that working memory may represent a constraint on the development of skills or knowledge in the area of spelling or reading (Gathercole et al., 2006; Re et al., 2014).

In sum, our results support research on intervention development that considers working memory abilities in children with heavy prenatal alcohol exposure who struggle with spelling. This does not discount the importance of teaching spelling directly, but rather offers an additional area in which to take into account while assessing and developing evidence-based interventions, in line with recommendations for broad focus areas for alcohol-exposed children in the classroom (Peadon, Rhys-Jones, Bower, & Elliott, 2009). Typical treatments may not be as effective as alcohol-exposed children may need more repetition or unique strategies to ensure that the information is encoded (Roebuck-Spencer & Mattson, 2004; Re et al., 2014; Schneider et al., 2000) . Working memory may act as a “bottleneck” for learning in classrooms, and effective management and consideration of the working memory load may facilitate instruction for associated problems in spelling and reading (Gathercole et al., 2006). The considerations may include reducing the concurrent working memory load, repetition, increased reminders in different modalities (visual, verbal), spending more time on a task, and providing additional scaffolding in teaching and assessing spelling in this population.

Limitations

This study adds important information to an area that is understudied despite its clinical implications. While it has many strengths including the sample size, the well-described population, and use of standardized and ecologically valid assessments, these results should be interpreted in light of its limitations. First, this study used one measure for spelling and did not have an in-depth error analysis. Spelling is postulated to consist of several components, whereas only one of these components, the ability to spell from dictation, was assessed. We chose this assessment of spelling because of its ecological validity, though future studies should evaluate multiple aspects of literacy and conduct error analysis in a standardized method to holistically determine the most effective treatment recommendations. This study was part of a larger battery focused on broad neuropsychological functioning and therefore we did not have access to a comprehensive database of related spelling skills or functioning or a validated approach to conduct detailed analysis of spelling errors, orthographic or morphological.

Children with fetal alcohol spectrum disorders frequently have concomitant psychological diagnoses, including high rates of ADHD (Fryer et al., 2007). ADHD is also associated with increased rates of disorders of written expression; however, spelling deficits are found to be associated with cognitive processing deficits and not the hyperactive or inattentive behavioral pathology (DeBono et al., 2012). Consequently, we are limited in understanding the precise etiology of the spelling deficits present in exposed-children, as it is difficult to disentangle the effects of prenatal alcohol exposure and ADHD. Our post-hoc analyses indicated that parent ratings of ADHD behaviors were not independently related to spelling performance.

Spelling is often taught in a school environment, and thus quality of education and type of instruction likely contributes to academic achievement. As prenatal alcohol exposure is often difficult to identify, affected children may not receive the individualized instruction they require, which can contribute to poor spelling or reading abilities, above the influence of underlying cognitive mechanisms. Further, we lack information regarding home environment variables important to literacy such as the presence of shared book reading, parenting quality, and exposure to reading in the household that can influence spelling performance (Institute of Child Health and Human Development Early Child Care Research Network, 2005).

Implications and Future Directions

Additional studies should investigate replication of our findings in a larger sample size and use more comprehensive methodology regarding both working memory components and spelling error analysis. Further research is necessary to determine how the link between working memory and verbal academic skills (reading and spelling) can be effectively translated to clinical settings (Savage, Lavers, & Pillay, 2007). There is evidence that behavioral and computerized interventions work in the FASD population for other domains, demonstrating their feasibility in this population (Kalberg & Buckley, 2007; Loomes, Rasmussen, Pei, Manji, & Andrew, 2008; Rasmussen, Pei, Manji, Loomes, & Andrew, 2009). However, solely conducting computerized working memory training does not appear to consistently or successfully generalize to academic settings (Beck et al., 2010; Carretti, Borella, Cornoldi, & De Beni, 2009; Loosli, Buschkuehl, Perrig, & Jaeggi, 2012; Melby-Lervag & Hulme, 2013).

To date there is only one literacy-focused intervention for the FASD population, which took place in South Africa as part of a larger study of third grade children (Adnams et al., 2007). Alcohol-exposed children were randomly assigned to either a treatment group or no treatment group, and were compared to an untreated non-exposed control group. The treatment intervention consisted of 38 hours of therapy (19 hours of phonological awareness and literacy training and 19 hours of language therapy). Post intervention, performance of both FASD groups remained worse than the controls. However, the treatment condition did result in significant gains in syllable manipulation, letter sound knowledge, written letters, word reading and non-word reading, and spelling. Although these results suggest modest efficacy of targeted cognitive interventions in children with FASD, the authors note that these findings are limited, as cognitive mechanisms contributing to spelling were not assessed nor were variables that may mediate literacy, further demonstrating the importance of the current study. This intervention was an important step in the feasibility of disseminating effective remediation to alcohol-exposed individuals and may be augmented by understanding the specific underlying mechanisms, such as those indicated in the current study.

While interventions have continued in other areas of impairment for children with FASD, such as math (Coles et al., 2009), targeted treatments for spelling and reading have not yet been created or widely disseminated (Bertrand, 2009; Kalberg & Buckley, 2007; Peadon et al., 2009). Poor academic achievement and verbal academic difficulties, in particular, are highly correlated with negative long-term outcomes in vocational success, secondary disabilities, and the ability to successfully contribute to society, resulting in a public health concern (Grant et al., 2004; Streissguth, 1992). The development of efficacious interventions for spelling and reading to improve literacy skills, particularly those that consider working memory ability, can have positive implications for increased personal autonomy and overall positive life outcomes throughout development.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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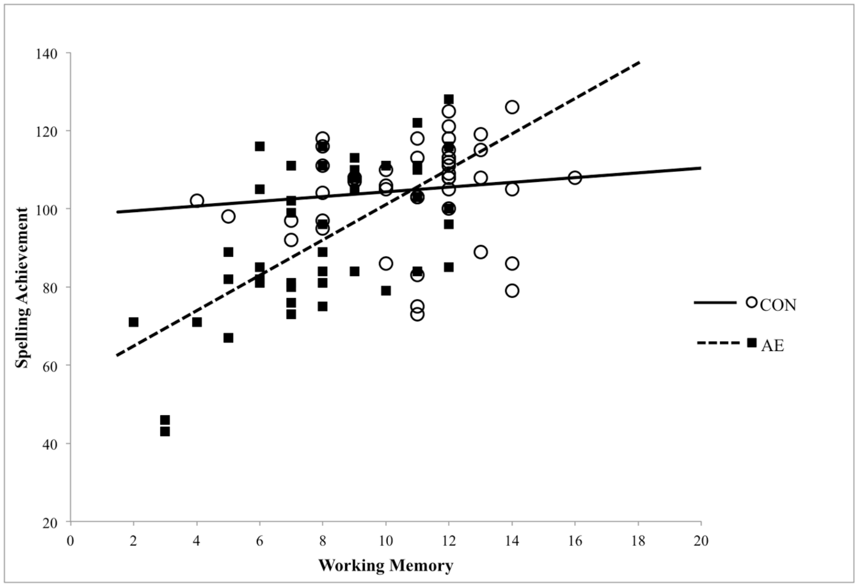

Figure 1.

Relation between working memory (NEPSY-II scaled score) and spelling achievement (WIAT-II standard score) for children with heavy prenatal alcohol exposure (AE), and non-exposed control children (CON). A significant working memory x group interaction and follow up analyses indicated that working memory significantly contributed to spelling achievement in the AE group ($R^2 = 0.386$), but not in the control group ($R^2 = 0.014$). The analyses have been rerun without the two lower AE subjects shown on the graph, and the results remain the same. See text for details.

Table 1

Descriptive data for demographic information, WIAT-II Spelling and Word Reading, and NEPSY-II variables for children with heavy prenatal alcohol exposure (AE) and control children (CON).

Variable	AE (*n* = 49)	CON (*n* = 47)	$p\eta^2$
Sex [*n* (% Female)]	16 (32.7)	23 (48.9)	
Handedness [*n* (% Right)]	42 (85.7)	42 (89.4)	
Race [*n* (% White)]	32 (65.3)	38 (80.9)	
Ethnicity [*n* (% Hispanic)]	6 (12.2)	13 (27.7)	
Age [*M* (SD)]	12.9 (2.27)	12.5 (2.45)	
SES [*M* (SD)]	45.9 (10.69)	50.7 (13.39)	
FSIQ [*M* (SD)]*	87.6 (16.06)	109.5 (12.97)	
FAS [n (%)]	10 (20.4)	0 (0)	
WIAT-II Spelling [*M* (SD)]*	91.7 (18.65)	104.8 (12.55)	.148
WIAT-II Word Reading [*M* (SD)]*	94.3 (18.44)	107.2 (11.87)	.149
NEPSY-II Phonological Processing [*M* (SD)]*	7.3 (3.68)	10.2 (2.31)	.187
NEPSY-II Speeded Naming [*M* (SD)]*	7.8 (2.40)	9.9 (2.07)	.183
NEPSY-II Working Memory [*M* (SD)]*	8.0 (2.57)	10.9 (2.56)	.245

*significant difference, AE < CON, $p < .05$

SES, Socioeconomic status, measured by Hollingshead.

WIAT-II, Wechsler Individual Achievement Test – second edition. Data are presented as standard scores. $p\eta^2$, effect size as partial eta squared (small: .02, medium: .13, large: .26)

FSIQ, Full scale IQ score. Data are presented as standard scores.

NEPSY-II data are presented as scaled scores.

Table 2

Within-group correlations (*r*) between spelling and reading achievement and cognitive variables for children with heavy prenatal alcohol exposure (AE) and non-exposed children (CON).

	Group	Spelling	Reading	Phonological Processing	Speeded Naming	Working Memory
Spelling	**AE**	1	.874**	.622**	.403**	.621**
	CON	1	.782**	.487**	.214	.120
Reading	**AE**		1	.729**	.443**	.729*
	CON		1	.525**	.380**	.343*
Phonological Processing	**AE**			1	.295*	.509**
	CON			1	.332*	.470**
Speeded Naming	**AE**				1	.389**
	CON				1	.309*
Working Memory	**AE**					1
	CON					1

** Correlation is significant at the $p < 0.01$ level (2-tailed)

* Correlation is significant at the $p < 0.05$ level (2-tailed)

Table 3

Unstandardized regression coefficients (B), standardized regression coefficients (β), *t*-values, *p*-values, and 95% confidence intervals (CI) for all cognitive variables and interactions for WIAT-II spelling. All cognitive variables were centered prior to analysis. Bold font indicates significant *p*-values using an alpha of .05.

Step	R^2	Variable	B	β	*t*-value	*p*-value	95% CI
Step 1	.148	**Group**	**6.569**	**.384**	**3.990**	**< .001**	**[3.299, 9.839]**
Step 2	.470	**Phonological Processing (PP)**	**2.421**	**.477**	**4.775**	**<.001**	**[1.414, 3.428]**
		Speeded Naming (SN)	.960	.139	1.499	.137	[−0.312, 2.233]
		Working Memory (WM)	.890	.149	1.414	.161	[−0.360, 2.140]
Step 3	.522	PP x Group	.469	.084	.859	.393	[−0.616, 1.553]
		SN x Group	−.313	−.041	−.500	.618	[−1.559, 0.932]
		WM x Group	**−1.686**	**−.609**	**−2.769**	**.007**	**[−2.896, −0.476]**

Table 4

Unstandardized regression coefficients (B), standardized regression coefficients (β), *t*-values, *p*-values, and 95% confidence intervals (CI) for all cognitive variables and interactions for WIAT-II reading. All cognitive variables were centered prior to analysis. Bold font indicates significant *p*-values using an alpha of .05.

Step	R^2	Variable	B	β	*t*-value	*p*-value	95% CI
Step 1	.149	**Group**	**6.441**	**.386**	**4.008**	**< .001**	**[3.249, 9.633]**
Step 2	.618	**Phonological Processing (PP)**	**2.419**	**.488**	**5.755**	**<.001**	**[1.585, 3.254]**
		Speeded Naming (SN)	**1.179**	**.174**	**2.221**	**.029**	**[0.124, 2.234]**
		Working Memory (WM)	**1.698**	**.291**	**3.255**	**.002**	**[0.661, 2.735]**
Step 3	.658	PP x Group	−.064	−.012	−.142	.887	[−0.961, 0.833]
		SN x Group	.092	.012	.178	.859	[−0.938, 1.122]
		WM x Group	**−1.349**	**−.201**	**−2.679**	**.009**	**[−2.350, −0.348]**