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TECHNICAL REPORT

Phonological Deficits in Children with Perinatal Stroke: Evidence from Spelling

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PHONOLOGICAL DEFICITS IN CHILDREN WITH PERINATAL STROKE: EVIDENCE FROM SPELLING

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Abstract

Children with perinatal stroke (PS) show attenuated linguistic deficits relative to adults with comparable insults, but lag behind their typically-developing (TD) peers on many pre-scholastic linguistic measures. Acquisition of literacy is a crucial academic skill but little is known about written language outcomes in the PS group. This study is the first to conduct formal analyses on the spelling of children with PS. Two writing tasks and two standardized tests (spelling and word reading) were administered to 43 children with PS and 42 age-matched children with TD, ages 7 to 17. The descriptive task had a referent (i.e., a picture) which provided a constrained framework, whereas the narrative task had a minimal obligatory context. Spelling was assessed by two indices: frequency of errors and phonological accuracy. Results showed effects for age (older > younger) in both experimental tasks, and population (TD > PS) in the standardized and descriptive tasks, suggesting that children with PS employed compensatory strategies when given the freedom provided by the narrative task. Unlike children with TD, children with PS showed no effect of age in phonological accuracy, suggesting persistent deficits in phonological encoding.

Introduction

Insights into Literacy: Spelling in Children with Perinatal Stroke

The primary linguistic task facing a school-age child is mastery of literacy. This task is demanding cognitively and important socially: it has been called "the supreme achievement of schooling" (Bialystok, 2007, p. 46). Phonological decoding (word reading) and encoding (spelling) are early components of literacy which correlate highly with one another (Ehri, 2000). Phonological impairments in spoken language that appear to resolve with age reassert themselves as deficits in written language, particularly in spelling (for a review, see Silliman, Bahr, & Peters, 2006). A great deal of research has focused on the spoken language of children with perinatal stroke (PS). This research has revealed a variable pattern of initial delay (Bates et al., 1997; Thal et al., 1991) and apparent later improvement (Reilly, Bates, & Marchman, 1998; Reilly, Losh, Bellugi, & Wulfeck, 2004), demonstrating the plasticity of the brain in the wake of early insult (Bates et al., 2001). However, assessments of the visuospatial skills of children with PS show a contrasting profile of subtle deficits that do not seem to resolve with age (Akshoomoff, Feroleto, Doyle, & Stiles, 2002; Lidzba, Staudt, Wilke, & Krageloh-Mann, 2006; Stiles et al., 2003; Stiles, Stern, Trauner, & Nass, 1996). Spelling is a relatively unstudied domain in children with PS (Aram, 1991). Underlying language impairments that no longer manifest in the spoken domain may appear in writing. Such impairments would be expected based on the visuospatial performance of these children and suggest limitations to neural plasticity.

The Spoken Language Profile Subsequent to Stroke

PS occurs in approximately 1 of every 4000 live births (Nelson & Lynch, 2004), and is often diagnosed after presentation of seizures or mild hemiparesis. The majority of cases are caused by infarct of the middle cerebral artery (Lynch & Nelson, 2001), more frequently in the left hemisphere (Kirton & Deveber, 2006). Children with left-hemisphere lesions (LHL) rarely show signs of the agrammatic, dysfluent speech often seen in adults (Bernhardt, 1896; Cotard, 1868; Guttmann, 1942). Children with right-hemisphere lesions (RHL) do not manifest the fluent but "unconnected" speech of adults with comparable lesions (Bates et al., 2001). Both groups are delayed in language acquisition but hemispheric effects are not often seen in their linguistic performance past age seven (e.g., Reilly & Wulfeck, 2004). Overall, children with PS appear to show an iterative profile of delay and subsequent recovery with each new linguistic challenge (Reilly, Levine, Nass, & Stiles, 2008). Unlike children with PS, adults do not show the same apparent plasticity (Bates & Roe, 2001). In studies that compare the two age groups directly, the PS group shows attenuated deficits relative to adults with lesions (Kempler, Van Lancker, Marchman, & Bates, 1999).

The Visuospatial Profile Subsequent to Stroke

Hemispheric differences are seen in the visuospatial domain in both adults and children following stroke. LHL in adults can lead to local-level, detail deficits and RHL create more integrative, global-level deficits (e.g., Delis, Robertson, & Efron, 1986; Robertson & Lamb, 1991). A subtler version of this same profile is seen in children with PS and does not change with age (Reilly, Levine, Nass, & Stiles, 2008; Stiles, Nass, Levine, Moses, & Reilly, 2009; Stiles et al., 2008). These deficits may impede literacy acquisition even if spoken language is spared, as writing is language brought into the visual domain.

Longitudinal studies of PS have tested the same children in multiple domains (Aram, 1991; Levine, Kraus, Alexander, Suriyakham, & Huttenlocher, 2005; Stiles, Bates, Thal, Trauner, & Reilly, 1998). The differential profiles of deficit and development for language and visuospatial functioning manifest within individual children. One explanation for this discrepant performance is language has privileged status in the brain, and is spared at the expense of visuospatial processing (i.e., the "crowding hypothesis"; see Lidzba, Staudt, Wilke, & Krageloh-Mann, 2006; Stiles, 2000; Woods & Teuber, 1978). Alternatively, previous spoken language tasks may not have been challenging enough to reveal subtle deficits. Assessing written ability in children with PS has the potential to uncover such deficits.

The Written Language Profile Subsequent to Stroke

Agraphia[1] was first thought to be co-morbid with aphasia in adults (e.g., Hughlings-Jackson, 1879), despite cases of agraphia with spared speech (Ogle, 1867; Pitres, 1884). A review of 307 patients with LHL found that nearly half were agraphic (Hecaen, Angelergues, & Douzenis, 1963), indicating it is a common outcome (see also Henry, Beeson, Stark, & Rapcsak, 2007). LHL may lead to phonological agraphia, where spelling of familiar words is spared relative to spelling of unfamiliar or invented words (Alexander, Friedman, Loverso, & Fischer, 1992). RHL may result in spatial agraphia (Hecaen & Marcie, 1974), characterized by deficits in organization of words (such as writing on a slant, writing on the right of the page, etc.) rather than problems in representing phonology (Rode et al., 2006).

As mentioned above, the writing of children with PS is understudied. Spelling may be a vulnerable domain in children with PS, but methodological issues make clear conclusions from prior studies difficult. Previous research has conflated early and late acquired lesions (e.g., Alajouanine & Lhermitte, 1965), looked exclusively at LHL (Pitchford, 2000; Woods & Carey, 1979), or looked only at standardized measures (Aram, 1991; Ballantyne, Spilkin, Hesselink, & Trauner, 2008; Frith & Vargha-Khadem, 2001). These studies found that spelling is vulnerable in children with PS, suggesting either phonological encoding impairments or difficulty in graphically representing spoken language. Neuroimaging studies of the neural substrates of spelling suggest it is largely a linguistic process carried about by the left hemisphere in adults (Beeson et al., 2003) and children (Deutsch et al., 2005). However, the visuospatial deficits of children with PS are likely to have an impact on the processing and production of writing. Before we assess these competing claims, we provide an overview of spelling development.

Acquiring Literacy in an Opaque Orthography

English has an opaque orthography: it is rule-based (Venezky, 1970), but phoneme-grapheme correspondences are irregular. Transparent orthographies, like Spanish, have (nearly) one-to-one phoneme-grapheme correspondences (Sainz, 2006). Only the vowel-less variants of scripts like Hebrew or Arabic are more opaque than English (Perfetti, 2008). Recent cross-linguistic research has provided support for the orthographic depth hypothesis: transparent orthographies are learned more quickly than opaque ones (Aro & Wimmer, 2003; Goswami, Gombert, & de Barrera, 1998). One study found that 86% of Spanish first graders were no longer making errors after one year of explicit spelling instruction (Defior & Serrano, 2005). The researchers found the developmental trajectory was the same across languages, but Spanish children performed 1-2 years ahead of their English peers (see also Aro & Wimmer, 2003).

[1] Agraphia and dysgraphia are often used interchangeably, as are alexia and dyslexia. We use agraphia here to refer to both writing disorders. For clarity, we use alexia to refer to disorders resulting from a neurological insult, and dyslexia for developmental disorders in the absence of frank brain damage.

Many models of spelling are stage-based, although there is debate regarding how strict these stages are (e.g., Treiman, Cohen, Mulqueeny, Kessler, & Schechtman, 2007). Pre-literate spellers may attempt to encode physical characteristics of the referent rather than the spoken form of the word (Ferreiro & Teberosky, 1982). Early spelling productions that seem arbitrary may reflect sensitivity to phonology, as spelling *chruck* for "truck," or use a letter name strategy such as *ne* for "any" (Read, 1971; Treiman & Bourassa, 2000). Such misspellings are precursors to acquisition of the "alphabetic principle" in first and second grade (ages 7-9): knowledge that letters in words are meant to encode its phonology. This principle is crucial to literacy development (e.g., Moats, 1995). Children begin incorporating orthographic and morphological rules in their spelling around 9 years of age (Snow, Burns, & Griffin, 1998).

Analysis of Spelling Errors

Typically, children misspell words because they improperly segment phonemes or are unfamiliar with spelling conventions (Apel & Masterson, 2001). These misspellings can be analyzed using phonological or orthographic measures to gauge spelling proficiency. Phonological accuracy measures evaluate how well the production encodes the phonology of the target form (also called phonological plausibility; for reviews, see Lennox & Siegel, 1996; Moats, 1993; Silliman, Bahr, & Peters, 2006). Orthographic accuracy measures compare letter pairs (digraphs) in the target to those in the production.

Such measures can be constrained or unconstrained and dichotomous or phoneme-by-phoneme. Constrained measures account for orthographic constraints and show better sensitivity (Silliman, Bahr, & Peters, 2006). Likewise, phoneme-by-phoneme systems can be used to provide greater resolution (e.g., Treiman & Kessler, 2004), indicating not just whether a misspelling is phonologically accurate, but also the *extent* to which it is a viable substitute. Thus, misspelling "cat" as *kta* would score a 67% phonologically accuracy rating (for representing two out of three target phonemes in the appropriate order) but *kol* would score 33%. As deficits in children with PS are often subtle (e.g., Stiles, Reilly, Paul, & Moses, 2005), we opted for a constrained, phoneme-by-phoneme analysis of their misspellings.

The Current Study

PS is an accident of nature that allows researchers the opportunity to analyze the effect of early insult on cognitive development. As mentioned above, there is a contrast between the linguistic and visuospatial profile of children with PS. Previous research on writing in this group has been limited, despite the fact that writing is language brought into the visuospatial domain.

The current study aims to assess the phonological abilities of children with PS, as reflected in their spelling. We have outlined two competing hypotheses:

1) Due to the linguistic nature of writing, the spelling of children with PS will mirror their spoken language profile. There will be no evidence of hemispheric differences; spelling deficits in the younger age groups will not be apparent in the older groups.

2) The visuospatial deficits seen in children with PS will interfere with mapping written language onto spoken language. Their spelling will reflect the visuospatial profile. LHL will lead to local spelling errors indicated by lower phonological plausibility; RHL will lead to global deficits of organizing the words on the page but largely sparing spelling. Persistent deficits will be seen in the older age groups.

Methods

Participants

Existing data from a larger longitudinal study of typical development and developmental disorders were used (e.g., Reilly & Wulfeck, 2004). Participants were 43 children with PS (24 LHL, 19 RHL), aged 7-17 years, and 42 children with TD matched for age, gender, and SES. Children in both groups had normal hearing, normal or assisted vision, and mean IQ within the normal range; all children were English monolinguals with the exception of 2 children with PS.

TD children were recruited from the community, had no history of developmental delay and were neurologically intact as confirmed by neurological exam. The criterion for inclusion in the PS group was a single, unilateral focal lesion in the absence of other more diffuse pathology (see Appendix A for lesion data). The insult occurred in either the last trimester of pregnancy or the first 4 weeks after birth, and was confirmed by MRI or CT scan. Lesions were given a qualitative severity rating on a five point scale (adapted from Vargha-Khadem, O'Gorman, & Watters, 1985), with one being the smallest lesion and five representing a lobe impacting multiple lobes. Mean severity ratings are shown in table 1.

In order to examine development, the children were split into three age groups: 7-9 (n = 18), 10-13 (n = 15), and 14-17 years (n = 9). Children with TD often acquire the alphabetic principle between 7 and 9 years old, and focus on incorporating orthographic and morphological rules around age 10, as mentioned above. At age 14 many children begin high school and writing tasks become a more regular part of the curriculum.

Materials and Design

Standardized Measures

Children were administered the letter-word (reading) and spelling sub-tests of the Woodcock-Johnson 3 (WJ3; Woodcock & Mather, 1989) as part of a larger battery of standardized and experimental measures. The reading test from the WJ3 asks younger children to point to letters and words first; then all children read lists of words aloud. The spelling test asks younger children to copy shapes and write letters; then all children spell words from dictation. Each test yields a standardized score with a mean performance for age level of 100 and a standard deviation of 15.

Experimental Measure 1: Picture Description

Two writing tasks were administered during different testing sessions and were always presented in the same order. The first was a description of the "Cookie Theft" picture from the Boston Diagnostic Aphasia Exam (Goodglass & Kaplan, 1983). This picture depicts a child trying to steal cookies while water spills out of the sink where his mother is distractedly washing dishes. The experimenter prompted the child to describe "what's going on" in the picture. This task provided an obligatory context (i.e., the picture) for the child to use as a framework.

After providing a spoken description of the picture, the child then wrote her description. When the child was finished, she was given a different color pen and the chance to revise. The experimenter asked the child to read her description aloud to clarify what the child intended to write. Children were given as much time as they needed to complete the tasks. Experimenters prompted children having trouble with the task with open-ended questions ("How did it start?").

The child was audio- and video-recorded while performing the task. The spoken description was transcribed in CHILDES format (MacWhinney, 2000). Transcribers also used a word-processing program to create a "mirror" of the written description, including all errors and corrections made by the child. Examples are shown in Figure 1.

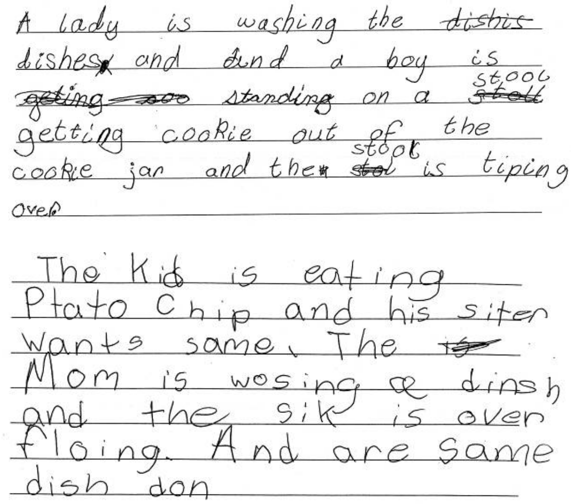


Figure 1. *Picture description from a child with TD (upper, age 9; 6) and with PS (lower, age 9;8). The description on the upper text reads* A lady is washing the dishis dishes and and a boy is getting soo standing on a stoll stool getting [0 article] cookie out of the cookie jar and the stol stool is tiping over. *The description on the lower text reads:* The kid is eating Ptato Chip and his siter wants same. The Mom is wasing a dinsh and the sik is over floing. And are same dish don. *The last line was read back as "*And there are some dishes done.*" The TD description scored a PWC of 96.4% and a PPA of 100%, while the PS description scored 64.3% and 76.7%, respectively.*

Experimental Measure 2: Spontaneous Narrative

The second writing task was an autobiographical spontaneous narrative, a de-contextualized discourse task that put greater cognitive demands on the child. The task is described in detail elsewhere (Reilly & Woolpert, in preparation) and has been used in numerous cross-linguistic studies (e.g., Berman, 2002). Children were asked to talk about a time when someone made them "mad or sad" and then write their narrative. Otherwise, the methods were comparable to those used in the first task. Example narratives are shown in Figure 2 (next page).

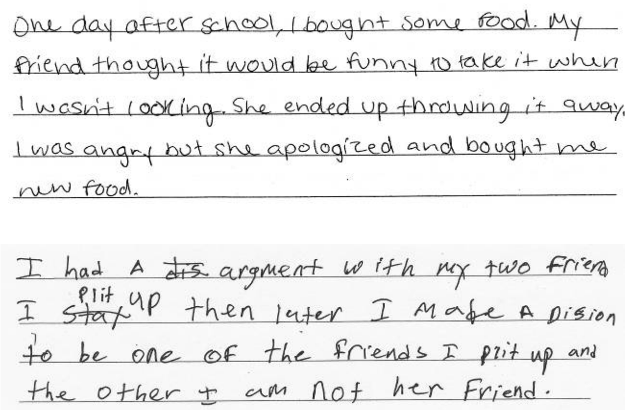


Figure 2. *Narrative from a child with TD (left, age 15;7) and with PS (right, age 15;4). The narrative on the left reads:* One day after school, I bought some food. My friend thought it would be funny to take it when I wasn't looking. She ended up throwing it away. I was angry but she apologized and bought me new food. *The narrative on the right reads:* I had a ~~dis~~ **argment** with my two friend. I ~~stay~~ **plit** up then later I made a **Dision** to be one of the friends I **plit** up and the other I am not her Friend. *The TD description scored a PWC of 100% and a PPA of 100%, while the PS description scored 88.2% and 80.1%, respectively.*

Evaluating Spelling Performance: Error Frequency and Phonological Accuracy

Children's writing samples were evaluated on three measures: number of words, percentage of words correct (PWC), and percentage of phonological accuracy (PPA). The number of words was used as a denominator for the other two measures. Thus, a child who wrote 50 words and misspelled five would score 90% on PWC. PWC provides a quantitative measure of the spelling errors, whereas PPA provides a qualitative one.

Spelling errors were classified as either morphological or non-morphological. Morphological errors were violations of morphosyntactic rules (such as using the wrong tense) or inappropriate applications of morphological rules (such as producing *saveing* for "saving"). All other misspellings were non-morphological errors and were analyzed for percentage of phonological accuracy (PPA), a measure adapted from the Automated Measure of Phoneme Representation, or AMPR (Treiman and Kessler, 2004). AMPR is designed for evaluating beginning spellers' productions, and is unconstrained (i.e., does not take orthographic context into account). We employed a constrained system for its greater discriminatory ability. All the scoring on the PPA measure was done by the first author. Children who made no spelling errors scored 100% on PPA for their writing sample.

Two coders worked independently to determine the production (what the child wrote) and the target (what the child *intended* to write) for each error. Inter-coder agreement was greater than 90% and discrepancies were discussed until a consensus was reached. Spelling errors were counted only if not corrected later by the child.

The total number of phonemes in the target word was counted as a denominator. Diphthongs (e.g., /aʊ/ as in "p<u>ou</u>t"), affricates (e.g., /tʃ/ as in "<u>ch</u>ase"), and rhotic vowels (e.g., /ɚ/ as in "h<u>ur</u>t") were counted as single phonemes. One point was awarded for each phoneme that was represented in the proper order. No points were awarded for phonemes represented out of order (e.g., *muisc* for "music" scored a PPA of 5/6 = 83%).

An error that spelled a different word was scored using the phonological representation of that word; otherwise, letter combinations with multiple correspondences were scored as accurate. Thus, *dose* for "does" scored 1/3 = 33% on PPA, as "dose" /doʊs/ and "does" /dʌz/ only have one phoneme in common, even though *s* can represent /z/ between *o* and *e* (e.g., "nose"). A spelling of *tou* for "too" (or its homophones) would yield a PPA of 2/2 = 100%; *ou* is often /aʊ/ but can be /u/ in words like "you." Representing phonemes not present in the target was penalized by increasing the denominator by one. For example, *dinsh* for "dish" represents the three target phonemes and an additional /n/, so scored a PPA of 3/4 = 75%.

A Note on Outliers

An *a priori* decision was made to exclude children from analysis who scored more than 2 SDs below the mean for their group. One of the children in the PS group was excluded based on this criterion: she scored more than 2 SDs below the PS mean on both PWC and PPA. Her writing was severely agraphic and generally only function words ("of," "the") were spelled correctly. Misspellings were consistent when there were multiple instances of the same word, however. The full text of her narrative and description are included in Appendix B.

	PS Means (SE)			TD Means (SE)		
Age	8.7 (.2)	11.9 (.3)	15.4 (.4)	8.7 (0.2)	11.8 (0.3)	15.7 (0.3)
Spelling	101.5 (4.2)	98.2 (5.1)	84.6 (5.3)	114.1 (2.3)	111.8 (2.5)	102.9 (1.9)
Reading	97.9 (3.1)	98.2 (4.4)	81.4 (6.6)	115.2 (3.5)	113.1 (2.9)	106.9 (3.4)
Words - Desc	28.7 (6.2)	39.8 (9.4)	57.7 (7.4)	37.2 (5.3)	44.3 (4.6)	47.6 (8.8)
PWC - Desc	80.0% (4.6)	86.6% (4.1)	94.5% (1.7)	88.4% (2.5)	96.3% (2.5)	98.6% (1.0)
PPA - Desc	78.2% (4.3)	89.1% (2.9)	84.3% (4.5)	85.2% (3.3)	91.5% (3.8)	99.3% (0.7)
Words - Narr	33.1 (5.0)	58.0 (10.4)	72.0 (14.8)	49.2 (6.1)	90.2 (19.3)	74.7 (9.6)
PWC - Narr	86.6% (3.4)	94.1% (2.1)	95.3% (1.7)	87.5% (3.0)	97.4% (1.3)	98.6% (0.7)
PPA - Narr	83.3% (4.7)	88.5% (4.0)	86.4% (3.5)	90.5% (2.4)	91.5% (3.2)	95.4% (4.6)
VIQ	93.6 (5.3)	82.1 (5.3)	86.9 (5)	-	-	-
PIQ	93.5 (5)	85.5 (6.5)	88.6 (6)	-	-	-
FS IQ	87 (7.2)	78.3 (8)	81 (7.7)	-	-	-
Severity	4.1 (0.35)	3.9 (0.4)	4.1 (0.4)	-	-	-

Table 1. *Mean ages and outcomes by population and age group.*[2]

Results

ANOVAs analyzing the PS group's performance on the two tasks returned no effect for side of lesion (for results of tests see Appendix C). Results are collapsed for all children with PS. Mean ages and results for each group are shown in Table 1.

Standardized Measures of Spelling and Reading

WJ3 scores were not available for 2 children with TD and 12 children with PS. A 2 x 3 ANOVA (Population x Age Group) of the standardized WJ3 scores yielded significant effects of age group on spelling, $F(2,64)=6.59$, $p < .01$, and reading, $F(2,64)=4.91$, $p = .01$; and of population on spelling, $F(1,65)=24.65$, $p < .001$ and reading, $F(1,65)=33.71$, $p < .001$. Mean standard scores are shown in Table 1.

Results from the Descriptive Task

Data on the descriptive task were not available for 4 children with PS. There were 214 spelling errors in the 80 remaining descriptions. Children in the youngest age group produced 125 errors (58.4% of the total). Twenty errors were morphological. Eighteen children with TD (6/9 from the oldest group) and 6 children with PS (1/9 from the oldest group) made no errors. Mean performance on the descriptive task is shown in Table 1 (above).

A 2 x 3 ANOVA (population x age group) yielded significant effects of population on PWC, $F(1,78)=6.50$, $p = .013$, and PPA, $F(1,78)=5.80$, $p = .019$, but not on number of words written, $F(1,78)=.03$, $p > .86$. Significant effects of age group were found on all measures: number of words,

[2] Means are for age, in years; standard scores from the WJ3 Spelling (Spelling) and Letter-Word Identification (Reading) subtests; number of words (Words), percentage of words correct (PWC), and percentage of phonological accuracy (PPA) for the description (Desc) and narrative (Narr) tasks; verbal (VIQ), performance (PIQ) and full scale (FS IQ) IQ scores; and lesion severity (Severity) scale ratings. Two children with TD and 12 with PS did not have standardized test scores available. In the PS group, four children did not have data available from the description task, and three were unable to produce a written narrative. Nine children with PS did not have IQ scores available and seven did not have lesion severity data available.

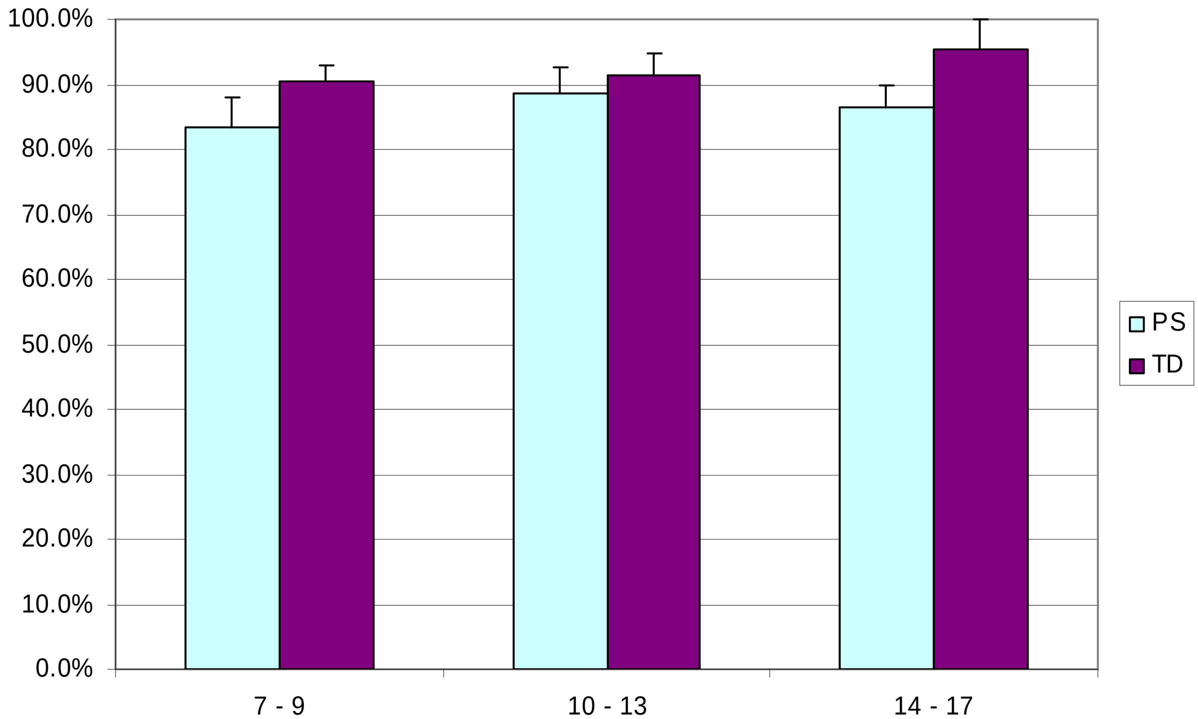


Figure 4. *Mean percentage of phonological accuracy (PPA) scores from the narrative task. There was an effect for age in the TD group but no such effect in the PS group.*

$F(2,77)=3.91$, $p = .024$; PWC $F(2,77)=6.39$, $p < .01$; and PPA, $F(2,77)=4.30$, $p = .017$. There was no significant population by age group interaction on any measure.

Mean PPA by age group and population is shown in Figure 3. A post-hoc ANOVA revealed significant effects of age on both PWC and PPA ($p < .05$) in the TD group. There was a trend for an effect of age group in the PS group on PWC ($p = .08$) but not PPA ($p > .16$).

Results from the Narrative Task

Of the children with PS who completed a written description, 3 were unable to write a narrative. In the remaining 81 narratives, 228 spelling errors were counted, 153 of which were from the youngest age group; 14 were morphological. Fifteen children with TD (5/9 from the oldest group) and 11 children with PS (1/9 from the oldest group) made no spelling errors in their narratives. Mean performance on the narrative task is shown in Table 1.

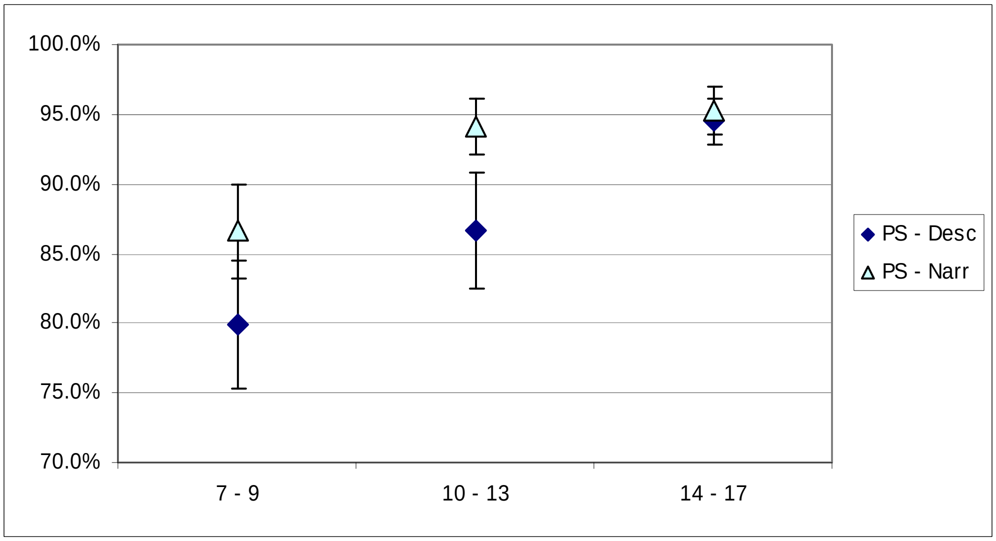
A 2 x 3 ANOVA (population x age group) yielded no population effects on number of words, $F(1,79)=2.91$, $p = .09$; PWC, $F(1,79)=1.32$, $p = .26$. There was a trend for an effect of population on PPA, $F(1,79)=3.33$, $p = .07$. There were significant effects of age group on number of words, $F(2,78)=5.55$, $p < .01$, and PWC, $F(2,78)=9.34$, $p < .01$. The effect of age group on PPA was not significant, $F(2,78)=.82$, $p > .40$. Mean PPA by age group and population is shown in Figure 4. There were no significant population by age group interactions.

Post-hoc tests showed an effect for age in the TD group on PWC ($p < .01$) but not PPA ($p > .60$). Children with PS showed a trend for age on PWC ($p = .06$), but not PPA ($p > .60$).

Follow-Up Tests

In the PS group there were seven children who had only one writing sample. To analyze whether this impacted the results on the two tasks, we re-ran the ANOVAs with those 7 cases excluded. The pattern for both tasks was similar, with a significant effect ($p < .05$) for population on PWC and PPA on the descriptive task. There were no significant effects on the narrative task.

We also analyzed for an effect of task on PWC with two-tailed paired samples *t*-tests on PWC. In the TD group, performance was not significantly different between the two tasks, $t(40) =.01$, $p > .99$. For the PS group, PWC was significantly different between the two tasks, $t(33) = -2.29$, $p = .029$. Mean PWC for the two groups by task is shown in Figure 5 (next page). Note that only children with data for both tasks were included in this analysis.



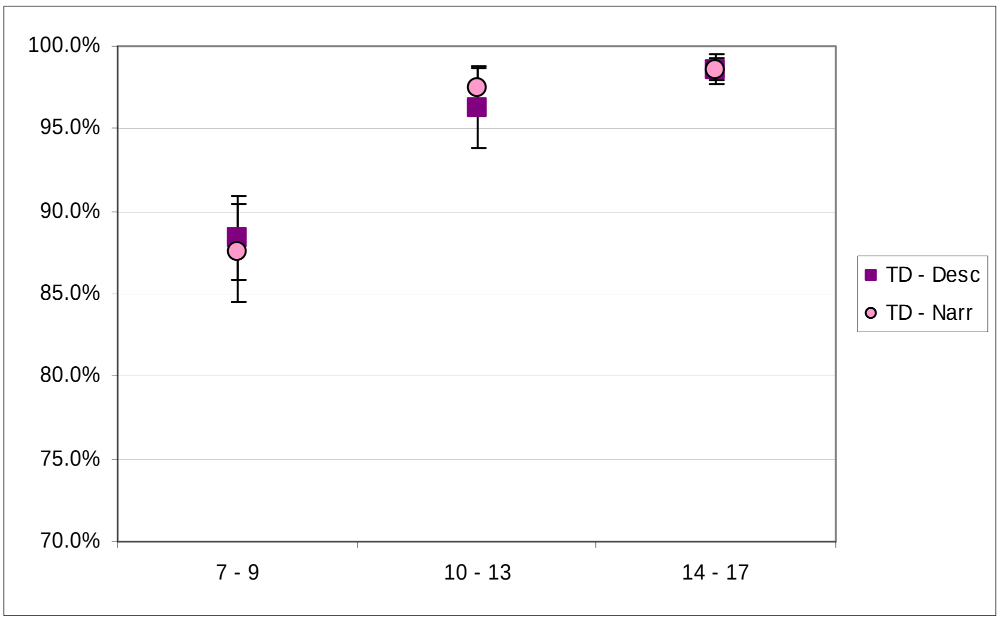


Figure 5. *Mean percentage of words correct (PWC) on the descriptive (desc) and narrative (narr) tasks. There was a significant difference in task performance for the PS (top) but not the TD (bottom) group.*

Since our data are cross-sectional there was concern regarding cohort effects – specifically, that the older children in the PS group may have been more cognitively impaired overall than the younger children. Six children did not have IQ scores available (3 in the youngest group, 2 in the middle, and 1 in the oldest). A univariate ANOVA run on verbal, performance, and full-scale IQ scores of the remaining PS children yielded no effect for age group on any of the IQ scores, $p > .58$. IQ scores were thus evenly distributed across age groups.

Higher mean lesion severity ratings in the older vs. younger age groups would also create cohort effects. Seven children did not have severity data available (5 in the youngest group, 1 in the middle, and 1 in the oldest). A univariate ANOVA for lesion severity returned no effect for age group, $p > .67$. The three age groups had comparable lesion severities.

Discussion

Written Language of Children with PS

Literacy is crucial to success in school and beyond. Spelling performance reflects fundamental skills such as word decoding and phonemic awareness. Our competing hypotheses were that spelling performance in children with PS would either be consistent with the spoken language profile (i.e., deficits in the younger groups; no hemispheric differences) or the visuospatial profile (subtle deficits in all age groups; hemispheric differences). The results suggest a mix of the two. No clear hemispheric differences are seen in performance, consistent with the children's linguistic profile. The largest performance gap between PS and TD is seen in the oldest age group, indicating persistent deficits such as those seen in the visuospatial domain.

Phonological Deficits in Children with PS

The most striking aspect of our findings is the evidence for persistent deficits in phonological encoding in children with PS. The children in the oldest PS group were not significantly better at phonological encoding, as measured by PPA, than beginning PS spellers. In fact, the oldest PS children's mean performance on PPA for both tasks was below that of the youngest TD group. Likewise, only one of the nine oldest children in the PS group made no errors on either task, compared to more than half of the children with TD in that age group. This suggests persistent phonological deficits for the PS group similar to those seen in the visuospatial domain.

There is other evidence that phonology is problematic for children with PS. Novel word learning, serial recall, and non-word repetition were tested in 11 children with perinatal LHL and 61 age-matched peers (Gupta, MacWhinney, Feldman, & Sacco, 2003). Children with TD outperformed the children with LHL on all three measures, but the effect was greatest on the non-word repetition task, which taps phonological memory. Furthermore, the correlation between age and higher scores was similar for the PS and TD groups on the word learning and serial recall tasks, but not the non-word repetition task. These data are cross-sectional, but suggest an underlying phonological deficit in the spoken domain, just as our results indicate such a deficit in their writing.

If a child has reached high school without mastering phonological encoding, is she likely to ever master it? Evidence of persistent phonological deficits is routinely found in studies of adults with histories of dyslexia (e.g., Bruck, 1990; Snowling, 1995). Children with PS may have similar intractable phonological problems. These problems suggest the possibility of a limitation to neural plasticity.

Since our data are cross-sectional, claims regarding plasticity must be qualified. Stronger assertions come from two longitudinal studies of children with PS (Ballantyne, Spilkin, Hesselink, & Trauner, 2008; Levine, Kraus, Alexander, Suriyakham, & Huttenlocher, 2005), although the results conflict. In the first study, Levine and colleagues gave 15 children with PS (4 RHL) an IQ test, once before age 7 and again anywhere from 1 to 14 years later. The mean IQ score at time 2 was significantly below that at time 1, and all children had lower scores aside from 2 children who showed increase and 1 child who showed no change. The authors conclude that early lesions may allow initial development of a skill but may interfere with later refinement of that skill.

In the second study, Ballantyne and colleagues found no significant differences in IQ scores over two testing sessions in 19 children with PS (6 RHL). The PS group's longitudinal profile was parallel to that of 24 children with TD matched for age and SES. Children with PS were found to have comparable verbal and non-verbal IQs. Those with seizures were found to have significantly poorer outcomes at both data points than those without seizures, and were not significantly parallel to the non-seizure or TD groups. The authors attribute the differences between this study and that of Levine and colleagues to a more heterogeneous sample in the latter study. The age range between data points is narrower in the former study, however, which reduces the probability of detecting deficits that take more time to manifest.

The Importance of an Appropriate Task: Compensatory Strategies

Another key finding was the effect of task in the PS group on PWC. The PS group made significantly fewer errors on the narrative than the descriptive tasks, while the TD group performed comparably on both tasks. Likewise, the TD group scored significantly higher on PPA than the PS group on the descriptive task, but demonstrated only a trend in that direction on the narrative task. This trend might have been significant had the three children who were unable to perform the narrative task had been able to produce a story. Regardless, the task effect suggests that children with PS were able to take advantage of the freedom afforded by the narrative task to use words of their own choosing, but when obligated to write about a specific set of lexical items, their problems with spelling surface.

This finding parallels outcomes seen for children with PS, where subtle visuospatial deficits only emerge during challenging tasks (e.g., Stiles, Reilly, Paul, & Moses, 2005). Additionally, the work of Kolb and colleagues has shown that apparent recovery in animals subsequent to cortical ablation may actually be due to compensatory strategies (Kolb, 1995).

Lack of Hemispheric Differences: the Linguistic Nature of Spelling

No evidence was seen of the adult profile of phonological agraphia in LHL and constructional agraphia in RHL. Given that the PS children's spelling performance showed evidence of persistent spelling deficits, similar to their performance on visuospatial tasks, hemispheric differences might also be expected. However, the negative finding is consistent with research on spoken language in children with PS (reviewed above) and the current view that spelling maps phonology and is not merely based on visual memory.

Hemispheric differences in the written language of the PS group may be uncovered by further research. Measures of non-word spelling and reading could more directly tap the phonological encoding and decoding abilities of these children. Likewise, children with PS could be administered measures of phonological awareness such as phoneme elision and blending to examine evidence of phonological deficits in their spoken language.

Conclusions

This study was the first to look in-depth at the spelling abilities of children with PS. The children in the older age groups made fewer errors than those in the younger group. Deeper analysis revealed comparable phonological deficits across all PS age groups. Research on the reading and writing abilities of children with PS is necessary to further evaluate the nature of their deficits in phonological conversion. The data suggest limits on neuroplasticity and underscore the importance of examining later development in children with PS, as consequences of their lesions may not be evident in the early childhood.

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Appendix A: Lesion Data for the PS Group

	Side	Sever	Lobes	Other Regions
1	Left	2	Frontal	Subcortical
2	Left	4	Frontal	Subcortical, Broca's,
3	Left	4	Frontal	Subcortical
4	Left	5	Parietal, Occipital	None
5	Left	5	Frontal, Temporal, Parietal	Subcortical, Thalamic, Basal Ganglia, Broca's, Wernicke's
6	Left	4	Frontal	None
7	Left	5	Frontal, Temporal, Parietal, Occipital	Subcortical, Broca's, Wernicke's
8	Left	5	Frontal, Parietal, Occipital	Subcortical,
9	Left	5	Frontal, Temporal, Parietal	Subcortical, Basal Ganglia, Broca's, Wernicke's
10	Left	4	Temporal	Wernicke's
11	Left	2	Subcortical only	Subcortical
12	Left	4	Frontal	Unknown
13	Left	4	Frontal	None
14	Left	2	Subcortical only	Subcortical
15	Left	*	Frontal, Temporal	None
16	Left	5	Frontal, Parietal	Subcortical, Thalamic, Basal Ganglia, Broca's, Wernicke's
17	Left	2	Temporal	Subcortical, Thalamic, Basal Ganglia,
18	Left	4	Temporal	None
19	Left	*	Frontal, Temporal, Parietal	None
20	Left	*	Frontal, Parietal	None
21	Left	5	Frontal, Parietal, Occipital	Basal Ganglia
22	Left	2	Frontal	Broca's
23	Left	5	Frontal, Temporal, Parietal	Basal Ganglia, Broca's, Wernicke's
24	Left	5	Parietal	Basal Ganglia
25	Right	5	Frontal, Temporal, Parietal	Subcortical, Thalamic, Basal Ganglia, Broca's, Wernicke's
26	Right	5	Frontal, Temporal, Parietal	Subcortical, Thalamic, Basal Ganglia, Broca's, Wernicke's
27	Right	2	Frontal, Temporal, Parietal	Subcortical, Wernicke's
28	Right	5	Frontal, Parietal, Occipital	Subcortical, Thalamic, Basal Ganglia, Broca's
29	Right	5	Frontal, Temporal, Parietal, Occipital	Subcortical, Broca's, Wernicke's
30	Right	3	Frontal, Temporal, Parietal, Occipital	Subcortical, Basal Ganglia,
31	Right	2	Frontal	Subcortical
32	Right	5	Frontal, Temporal, Parietal	None
33	Right	5	Frontal, Temporal, Parietal	None
34	Right	*	Frontal	Subcortical, Caudate
35	Right	2	Subcortical only	Subcortical

* Severity not known.

Appendix A cont'd: Lesion Data for the PS Group

36	Right	5	Frontal, Parietal	None
37	Right	*	Parietal	Subcortical
38	Right	5	Frontal, Temporal, Parietal, Occipital	Wernicke's
39	Right	5	Subcortical only	Subcortical, Thalamic, Basal Ganglia
40	Right	5	Frontal, Temporal, Parietal, Occipital	Subcortial, Broca's, Wernicke's
41	Right	*	Frontal, Temporal, Parietal, Occipital	Subcortical
42	Right	*	Unknown	Unknown

* Severity not known.

Appendix B: Narrative and Description for PS 43, Excluded from Statistical Tests

Age: 11 years, 0 months

Left frontal, temporal, and parietal lesion, severity 5.

__Narrative__:

My **sincer sain** a **bix wond** to me and it **outd** my **filling went hie sain** a **bix wond** to me, **thern** my **sincer** <**sain**> [added during readback] **sromre** to me.

My sister said a bad word to me and it hurt my feelings when she said a bad word to me, then my sister <said> sorry to me.

28 words / 15 spelling errors = 46.4% words correct. 54.4% phonological accuracy.

__Description__:

The **gime wis wing** the **dinme**. And the **whined cime** out of the **sin**. The **biy wes giying** a **cooing** to the **sinrer sand thim** you for the **cooing** and the **biy fill dille** on the **gine**.

The girl was washing the dishes. And the water came out of the sink. The boy was giving a cookie to the sister [*] said thank you for the cookie and the boy fell down on the ground.

37 words / 19 spelling errors = 48.6% words correct. 53.9% phonological accuracy.

Appendix C: Results for Statistical Tests of an Effect for Lesion Side

2 x 3 (lesion side x age group) ANOVA – Description Task

Number of words – $F(1,29)=.88$, $p = .36$

PWC – $F(1,29)=.04$, $p = .85$

PPA – $F(1,29)=3.12$, $p = .09$

2 x 3 (lesion side x age group) ANOVA – Narrative Task

Number of words – $F(1,33)=.32$, $p = .73$

PWC – $F(1,33)=.44$, $p = .65$

PPA – $F(1,33)=2.21$, $p = .13$