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Research Report

Narrative abilities of children with epilepsy

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Abstract

Background: There is a noticeable publication gap in the speech–language pathology literature regarding the language abilities of children with common types of epilepsy. This paper reviews studies that suggest a high frequency of undetected language problems in this population, and it proposes the need for pragmatically based assessment of children with epilepsy that includes analysis of spontaneous language skills.
Aims: To compare the language skills of two groups of children with epilepsy—those with recent onset seizures and those with more chronic seizure activity (>3 years' duration)—using a mix of standardized tests, analysis of elicited narratives and listener judgments of the children's narratives.
Methods & Procedures: Twenty-five children with epilepsy, divided into two groups (recent onset versus chronic), were age- and gender-matched to 25 typically functioning peers. In addition to standardized IQ and language testing, children produced narratives to accompany the book *Frog, Where Are You?* (1969). Narratives were analysed for syntax, vocabulary and narrative components. Forty-five adult listeners each blindly rated nine narratives to create a large pool of listener judgments.
Outcomes & Results: Children with chronic epilepsy showed the greatest and significant differences in both language skill and listener judgments from their unaffected peers. Differences were smaller for children whose epilepsy was of more recent onset and their matched peers.
Conclusions & Implications: Although based on cross-sectional data rather than on longitudinal study, the current results raise the possibility that continued seizures, or prolonged exposure to the medications used to control them, produce decrements in children's language performance over time. Thus, the authors strongly urge that there is greater awareness of seizure disorder among speech-and-language pathologists/therapists, and they strongly recommend baseline testing at first diagnosis so that changes over time can be reliably documented.

Keywords: children, narrative skills, epilepsy (seizure disorder), listener judgments.

What this paper adds

What is already known about this subject?

Reports in the medical and educational psychology literature suggest that children with chronic epilepsy may present with educational difficulties, some of which appear to be language based. However, there are virtually no reports of the language skills of children with epilepsy in the speech–language pathology literature.

What this paper adds

Analysis of the standardized test performance and narrative production skills of children with recent onset and chronic epilepsy confirms prior reports that this population is at risk for impaired language skills. Although significant differences were found between children with epilepsy and their unaffected peers, it is unlikely that these children would be referred for follow-up assessment or therapeutic intervention/support if baseline measurement of their skills at first diagnosis was not available.

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Introduction

Epilepsy is the most common of all childhood neurologic disorders; in the United States, a recent epidemiological study estimated a prevalence of reported lifetime diagnosis of 1%, and of current epilepsy/seizure disorder at above six cases per 1000 children (Russ *et al.* 2012). According to the Epilepsy Foundation of America (see http://www.epilepsyfoundation.org), 45 000 new cases are diagnosed in children under the age of 15 each year, leading to a prevalence of approximately 325 000 school-aged children under 15 years with a seizure disorder. Epilepsy is associated with difficulties in academic, social and emotional function (see the review by Pal 2011), including verbal and linguistic deficits on general neuropsychological testing. Berg *et al.* (2011) recently found that children with epilepsy (CWE) were more likely to be referred for special education services, even after controlling for cognitive abilities, than their case controls and siblings. Despite the literature that places CWE at elevated risk for a broad range of academic problems, including language use (Caplan *et al.* 2009, 2010), there is a startling lack of attention in the speech–language pathology (SLP) literature to this large constituency. A search of the major journals in the field (e.g. American Speech–Language–Hearing Association journals, *JCD*, *IJLCD*) produced only a single article having the root terms epilep* or seizure* in its title, suggesting a dearth of guidance to practising SLPs about potential risk for language impairment in CWE. The one article (Broeders *et al.* 2010) identified a higher incidence of pragmatic communication deficits in a cohort of 30 children having one of three major types of epilepsy.

The growing literature on academic performance in CWE now includes prospective longitudinal studies. However, many studies include children with varying types of epilepsy, which can be associated with variable profiles of communication impairment (Svoboda 2004). The current study specifically limits itself to the language profiles seen in localization-related epilepsy (LRE), localized to the temporal lobe. Further, the majority of reports track children using standardized psycho-educational measures not typically employed by SLPs to identify or monitor children suspected of language impairment. Additionally, there is limited information regarding functional language performance on typical communicative tasks encountered in school settings, such as narrative production. Finally, there are virtually no data regarding listener perceptions of the verbal skills of CWE. Each of these concerns is addressed in turn.

For the SLP seeking a concise overview of major epilepsy types and associated profiles, Camfield and Camfield (2002a, 2002b) are useful references. Much of the literature on language in epilepsy has concentrated on a limited number of syndromes, such as Rasmussen's syndrome (the seizure disorder most often leading to hemispherectomy) or Landau–Kleffner syndrome, well known to some SLPs because of its primary symptom of progressive language loss (Svoboda 2004). Although these syndromes can have major effects on language, they are not among the most typical forms of epilepsy likely to be seen by SLPs in most work settings, as they are among the rarer epilepsy syndromes; in contrast, localization-related temporal lobe epilepsy, the focus of the present study, is among the most common forms (see http://www.epilepsy.com).

The literature addressing speech/language problems in the more general population of CWE has been growing over recent years, although it continues to be confined to paediatric medical journals and journals specializing in epilepsy research. Benn *et al.* (2010) found that CWE without obvious brain atypicalities were almost six times more likely to have been referred for speech/language problems than their unaffected sibling. Parkinson (2002) found that children with focal, localization-related epilepsy (LRE) were more likely to have evaluation results suggestive of language impairments than other CWE subtypes. However, none of the children identified as language-impaired in Parkinson's study had been previously identified as needing intervention services. In contrast, children with focal epilepsy scored significantly higher than children with generalized seizures on the Vocabulary subtest of the Kaufman Brief Intelligence Test (K-BIT) in an analysis by Bhise *et al.* (2009) that did not include non-impaired peers. Rantanen *et al.* (2011) found that even in a small subgroup of children with 'uncomplicated epilepsy', verbal IQ and verbal short-term memory were significantly lower than in non-affected peers. Similarly, in a recent report by Jones *et al.* (2010), CWE with average IQ still scored an average of almost 10 points lower than their peers on the Test of Language Development (TOLD-2), although still within normal range.

Byars *et al.* (2007) found almost 15% of their sample of CWE to have significant structural brain abnormalities; these were associated with lowered scores on a number of assessments, including two subtests of the Clinical Evaluation of Language Fundamentals (CELF). Caplan *et al.* (2010), whose CWE participants were diagnosed with one of two major forms of seizure disorder, found measurable differences in brain anatomy in children showing differential performance on their language tasks. However, Dunn *et al.* (2010) found the aetiology of epilepsy to be associated more strongly with math achievement during the school years than with language skills; over time, changes in language skills more directly moderated reading and writing achievement, as might be expected.

In addition to the fact that most reports of language function in CWE do not appear in journals likely to be read by practising SLPs, the majority of studies on language function in CWE have been conducted using general psycho-educational measures, such as single-word picture naming, verbal fluency, colour naming or word definitions. Commonly used measures include the Wechsler scales, K-BIT and/or the Neuropsychological Assessment (NEPSY). Thus, assessment of language skills in CWE tends to use neuropsychological measures not optimally suited to the assessment of functional language.

A notable exception is a large series of studies by Rochelle Caplan and colleagues; they have analysed complex language skills using naturalistic language samples as well as testing using the Test of Language Development (TOLD) (Caplan *et al.* 2001, 2002, 2004, 2006, 2010, Drewel and Caplan 2007, Jones *et al.* 2010). They have found measurable deficits in discourse skills in their cohort of CWE (see the discussion by Caplan *et al.* 2006) as well as lowered scores on TOLD (Caplan *et al.* 2009); over time most language measures showed declines associated with continued duration of seizures. Using conversational language samples, they have reported (Caplan *et al.* 2002) that CWE may experience difficulty in repairing communication breakdown, maintaining conversational topics during interactions and/or using cohesive devices to link ideas across continuous discourse. These patterns may be perceived as instances of illogical thinking, loose and tangential associations, unpredicted topic changes, and a lack of cohesion (e.g. inappropriate use of synonyms or pronouns to link ideas over stretches of discourse or text). Caplan *et al.* (2002) also observed a relationship among conversational skills, lowered academic achievement and parental reports of school difficulties.

There is ongoing theoretical and practical debate regarding whether or not any deficits observed in CWE pre-existed diagnosis and reflect the underlying brain dysfunction leading to seizure activity, or evolve over time as children experience repeated seizures and are treated with anti-epileptic drugs (AEDs). While the current study design cannot fully answer this question, we offer an exploratory comparison of children with newly diagnosed epilepsy and children who have a more long-standing (chronic) history of seizure disorder. The impact of continued seizure activity is controversial, but suggests that seizure frequency further diminishes children's abilities across a range of functions (Camfield and Camfield 2002b). However, other studies have found depressed psycho-educational function in CWE even at the onset of seizure activity (Byars *et al.* 2007, Hermann *et al.* 2006, Oostrom *et al.* 2003). This would imply that language was likely to have been impaired before the diagnosis was made. Continued seizure activity will generally require use of AEDs, which are thought to impact neuropsychological function negatively as well (Hermann *et al.* 2010, but see Mandelbaum *et al.* 2009).

Summary and research questions

We sought to clarify the impact of epilepsy on children's language function by comparing both standardized, commonly administered speech–language test scores and language sample analysis results in a carefully described subset of CWE with a single, homogeneous diagnosis (LRE involving the temporal lobe with a left hemisphere focus). We hypothesize that, even at onset, CWE will have lower profiles on such measures, and that impairment may be greater for children with longer histories of seizure disorder.

We also hypothesize, given past research, that narrative abilities may be impaired in CWE, since the formulation of narrative involves language skills that have been measurably impaired in some past studies. We next extended the ecological validity of language appraisal in CWE by utilizing listener judgments of the language samples. A prior study by Newman and McGregor (2006) of teacher and SLP judgments of children with specific language impairment (SLI) found robust listener sensitivity to subtle differences in language use by children with SLI and typical peers.

Finally, we also sought to conduct an exploratory test of the hypothesis that any observed differences in language profiles between CWE and peers are likely to be the result of continued seizure activity rather than the underlying deficit that provoked these seizures. If so, language performance would be worse in the cohort of children who have lived with seizures for a longer period of time and it would then be important for CWE to receive baseline assessment of language use and follow-up, to detect any evolving decrements in language skill that merit attention.

Method

The Plasticity of Language in Epilepsy Research (POLER) initiative (Gaillard *et al.* 2007) is designed to examine effects of LRE on children's language performance, as well as dynamic language processing as measured by functional magnetic resonance imaging (fMRI) (see the earlier reports by Berl *et al.* 2005, and Mbwana *et al.* 2009).

Participants

Twenty-five CWE diagnosed with LRE,[1] with normal MRI (as determined by a standard, sequenced imaging procedure for epilepsy evaluation at 1.5 T, including

a high-resolution three-dimensional T1 weighted sequence) participated in the study. All children with CWE were referred to the Children's National Medical Center, Washington, DC, for assessment and management of their seizure disorder. They were selected from the larger cohort followed in the POLER project because they could be divided into two major subgroups—those who had recently been diagnosed (CWE-R; $n = 10$) and who had little prior seizure experience compared with those with a chronic history of continued seizure activity (CWE-C; $n = 15$)—and each of whom could be closely matched to typically developing peers on age and gender (TD-R and TD-C; $n = 10$ and 15, respectively. The larger study, which primarily focused on functional brain imaging profiles, did not closely match participants on these variables). TD peers were recruited by distribution of flyers; the study was described as one designed to locate brain centres responsible for language understanding. These children were provided with a 'picture of their brain' (personal brain scan printout) as reward for participation.

The full CWE group consisted of 11 females and 14 males (age range = 50–155 months). Each child was then matched by search through the larger sample (approximately 140 children) by gender and age within 3 months to a TD child having no prior diagnosis of seizure activity, or academic or language concerns, as reported by parents on a standard case history questionnaire. Figure 1 shows the age distribution of the matched pairs.

Exclusionary criteria were abnormal imaging findings (including prior surgery) or concomitant developmental disorder (other neurological conditions or syndromes, autism spectrum disorder and previously identified communication disorder, since the intent was to identify language profiles unique to a diagnosis of TLE). Participants were then divided into four groups. Group 1 contained ten children with recent-onset (less than 1 year following second seizure) epilepsy (CWE-R). Group 2 contained ten TD peers, who were age- (within 3 months) and gender-matched to the children with recent-onset epilepsy (TD-R). Groups 1 and 2 each contained four females and six males; the children's mean age in each group was 92 months (range = 50–139 months). Group 3 contained 15 children with chronic (>3 years) epilepsy (CWE-C). Group 4 contained their 15 age- and gender-matched TD peers (TD-C). Groups 3 and 4 each contained seven females and eight males, with a mean age in each group of 116 months (range = 75–155 months).

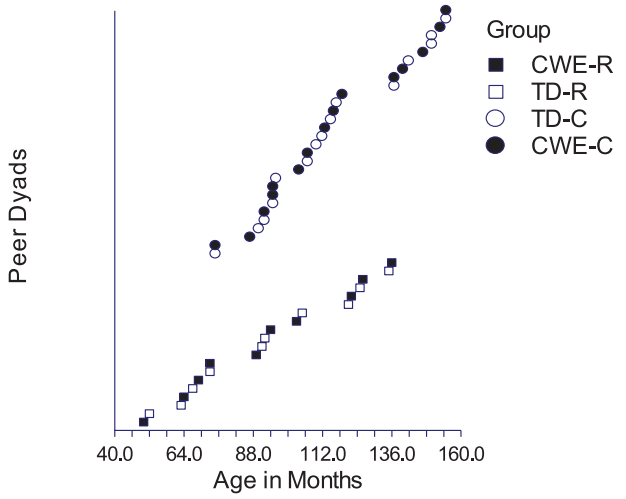

Figure 1. Participant scattergram: age distribution.

The average age of seizure onset in Group 1 (CWE-R) was 74 months; for Group 3 (CWE-C) it was 55 months. The range of total number of lifetime seizures (as reported by parents and/or in referring physicians' reports) in Group 1 varied between two to more than 20 seizures.[2] Forty per cent had experienced six or more seizures. In Group 3, the total number of lifetime seizures ranged from five to more than 20. Approximately 80% had experienced six or more seizures.

All child participants for the analyses reported here were selected from the larger recruitment sample and were right-handed native English speakers with normal anatomy as determined by MRI and standard electroencephalography (EEG). Structural lesions and mesial temporal sclerosis (MTS) were exclusionary to participation. Seizure focus for the CWE was determined on the basis of EEG and/or other clinical evidence that suggested a left hemisphere focus of seizure activity.

Because the original focus of the research had been functional imaging of language processing, some demographic variables were not gathered at assessment and were obtained through follow-up. Although not all parents provided information regarding race or education, the CWE groups were generally similar for these demographic variables. Racial composition was 57% Caucasian, 43% African-American for the CWE-R group, and 50% Caucasian/50% African-American for the CWE-C group. Both groups of typical peers were 62% Caucasian, 30% African-American and 10% 'other'. Mean parental education for the CWE-R group was 15 years for mothers and 14 years for fathers; for the CWE-C group it was 13.5 and 16 years, respectively. The typical peer groups' parents both averaged 18 years of education for mothers and 17 years for fathers. Thus, as might be expected, the clinically referred sample showed a slightly higher minority racial composition, and significantly lower parental educational levels ($p < 0.001$) than the child volunteers in the TD group, although the two groups of CWE did not differ markedly on these variables ($p > 0.6$).

Psycho-educational and language tasks

Each child was given the Clinical Evaluation of Language Fundamentals, Fourth Edition (CELF-4; Semel *et al.* 2003) or the Clinical Evaluation of Language Fundamentals, Preschool Edition for children less than 5 years of age (CELF-P). Only the core subtests were administered.

An elicited narrative was obtained using the wordless picture book *Frog, Where Are You?* (Mayer 1969). Each child was handed the book, led through its pages and asked to create a story. The examiner stopped at each page to prompt the child. If the child did not say anything, s/he was prompted with 'and now . . . ?' Testing and language sample elicitation were performed by clinicians not blinded to the child's diagnosis. However, audio recordings of the language samples were de-identified, and transcribed and coded by researchers blinded as to group.

Samples were transcribed and coded using CHILDES conventions (MacWhinney 2000). Stories were coded for narrative elements (Trabasso and Rodkin 1994): setting, initiating events, high-order goals, attempts (to locate the frog) and outcome. Narratives were also analysed for narrative length in words and utterances, lexical diversity, syntactic complexity and use of cohesion devices (Strong 1998). Narrative length was measured both in terms of total number of words and total number of C-units ('communication units'; one C-unit is defined as an independent clause and its modifiers). Lexical diversity (range of word types used) was measured using the Child Language Analysis (CLAN) Vocabulary Diversity (VOCD) program. The VOCD utility adapts the more classic type-token ratio calculation, and uses resampling procedures to minimize artefacts caused by sample size differences. A higher *D*-value represents higher lexical diversity.

Syntactic complexity was defined as the number of complex C-units (containing at least one dependent clause) proportioned against the total number of C-units per narrative to derive an average value for each child's story. Finally, use of cohesive devices (e.g. conjunctions, referential pronouns, word repetition, as defined and illustrated by Strong 1998) was proportioned against the total number of words per narrative.

A team of three individuals, including the first two authors, and a third transcriber transcribed and reviewed each child's transcript to achieve consensus on coding conventions. In the relatively rare event that consensus was not reached, the code selected by two of the three coders was used.

Listener judgments of narrative quality

Narrative production requires the integration of linguistic, cognitive and socio-behavioural skills (Norbury and Bishop 2003, Reilly *et al.* 2004). Clarity, charm and creativity are difficult to examine quantitatively and are often overlooked when using traditional objective measures (McFadden and Gillam 1996). We employed a design utilized by Newman and McGregor (2006). Listeners assigned a numerical score to stories and then completed a questionnaire rating the extent to which various specific factors (e.g. story grammar, syntax, fluency/articulation, 'sparkle') influenced their ratings.

Listener participants

Adult participants included 45 undergraduates, aged 18–22 years (37 women and 8 men) from the University of Maryland, College Park, and George Washington University, Washington, DC. Exclusionary criteria included hearing loss and English as a second language, by self-report.

Rating scale

Survey ratings addressed seven different narrative features: overall quality, vocabulary usage, story structure, grammatical complexity, speech fluency, colour/interest and prosody (see Appendix A). Listeners were instructed to base their overall quality rating on the clarity and ease of presentation, and vocabulary rating based on the level, variety and relevance of the words used in the story. Listeners were instructed to rate based on inclusion of critical parts of the story, and on whether the child followed the theme of the story. Grammar ratings referred to length and complexity of sentences, and fluency ratings to the smoothness of speech. Finally, participants were instructed to base colour/interest judgments ('sparkle') on inclusion of emotion and humour, and prosody judgments on whether the narrative sounded monotonous or expressive. Each quality rating was made using a seven-point interval scale.

Rating task

Adult listeners/raters first examined a copy of the illustrations used to elicit the narratives (which they could refer to as they listened to the stories) and the survey packet used to rate the narratives. Next, they reviewed procedural directions (see Appendix A), and were instructed to disregard articulation errors and recording quality. Participants each listened to ten different narratives, delivered through an iSymphony speaker for iPod. Each audio-recording was identified by numerical code, and participants were unaware of the group designation for any child.

Fifty narratives were used, and judged by 45 adult listeners. Nine different participants listened to and rated each narrative. The presentation order of the narratives

was counterbalanced. Each playlist of ten narratives was also designed to assure that all children on a playlist were of roughly similar ages (e.g. within a 2-year age span) to prevent younger children's narratives from being compared unfavourably with those from older children.

Results

Overview

We report a large number of comparisons on two relatively small peer cohorts who were specifically selected to be tightly matched in age and gender (known to impact language performance profiles) and to rule out typical confounds in prior studies, such as type of seizure disorder and localization focus; thus, all results should be viewed as exploratory. We adjusted all p-values on a per-hypothesis basis using the Bonferroni correction, and report effect sizes for all results meeting significance.

Psycho-educational standardized test scores

IQ and CELF test scores (standard scores) were compared across groups using a one-way analysis of variance (ANOVA). Group was a significant determinant of verbal IQ score on the WASI (or DAS for children less than 6 years of age); $F(3, 46) = 4.7$; $p = 0.006$, $\eta^2 = 0.235$). Performance was poorest by children with chronic epilepsy (mean = 98.4, SD = 13.17), which differed significantly only from their TD peers (mean = 119.13, SD = 19.05). CWE-R also differed from the CWE-C's typical peers (mean = 102.7, SD = 17.4), but not from their own age-matched peers (mean = 110.4, SD = 12.58), and the two groups of CWE did not differ from each other in verbal IQ (MSE = 253.74, $p = 0.05$). It should be noted that the TD cohorts performed at or above the mean standard score, while the CWE were functioning within normal limits, but at a lower level than their peers.

Performance IQ scores also showed an effect of group ($F(3, 46) = 4.59$; $p = 0.007$; $\eta^2 = 0.23$). By Fisher's LSD multiple-comparison test, both groups of CWE differed from both groups of TD peers, but children with recent-onset seizures did not differ from chronic children, nor did the two groups of TD children differ from each other (MSE = 198.91, $p = 0.05$). CWE-C achieved a mean standard score of 96.13 (SD = 13.1); CWE-R achieved a mean standard score of 97.7 (SD = 13.7). TD peers of CWE-C had average scores of 111.5(SD = 15.7); peers of CWE-R had average scores of 111.6 (13.3). Once again, the CWE performed at average levels, while the TD peers performed somewhat above average levels for age.

Scores on the CELF and CELF-P showed effects of group. Group difference for the Core Language subtests was significant ($F(3, 43) = 9.06$, $p = 0.0009$; $\eta^2 = 0.36$). However, on this instrument, CWE-R showed the lowest average scores (mean = 86.78, SD = 15.5), significantly different from performance by their TD peers (mean = 114.4, SD = 10.7) and the peers of CWE-C (mean = 111.8, SD = 14.5). The same profile was shown by children with chronic epilepsy (mean = 95.9, SD = 15.1). The two groups of CWE did not differ from each other on CELF performance (MSE = 201.56, $p = 0.05$). Two CWE-C and one CWE-R did not complete testing to allow computation of the Core Language quotient. For this reason, only 47 child participants (instead of 50) were included in this analysis. Although there are no firm cut-offs for using CELF scores to define a clinical language disorder, both groups of CWE probably would be judged as falling within average to low-average performance. However, gender- and age-matched children from their general region and educational systems did perform significantly better.

Narrative analyses

Narratives were analysed for a range of variables, including those that are micro-structural (grammatical and lexical) as well as those considered to be macro-structural (e.g. incorporation/integration of story elements, as described by Justice *et al.* 2006). All narrative codes were assigned by one of two trained judges and then juried in conjunction with a third judge to reach consensus judgments. Narrative data were compared cross-sectionally in two sets of comparisons: children with recent-onset epilepsy (CWE-R) compared with their age- and gender-matched TD peers (TD-R), and children with chronic epilepsy (CWE-C) compared with their age- and gender-matched TD peers (TD-C). Outcomes for these analyses were not standard scores and thus reflected the influence of age as well as gender, which were not constant across the four groups. Although age ranges overlapped between CWE-R and CWE-C and their peers, the chronic group and their peers were, on average, 2 years older than the pairs of children with recent-onset epilepsy and their peers, a significant difference ($t = 2.1$, $p = 0.047$). Thus, all comparisons in the following sections are made between the CWE and their respective peer-matched group only. Additionally, for many of the comparisons, lack of homogeneity of variance in scores among groups forced non-parametric statistical analysis. In these cases, effect size was appraised using Cliff's delta.

The number of words used to tell the narratives did not differ within matched cohorts. CWE-R used an average of 227.5 words to tell their stories, while their peers used an average of 222.4 ($t = 0.1166$, $p = 0.91$). CWE-C used a mean of 251 words in telling the *Frog*

narrative, while their peers used an average of 256.7 words ($t = -0.1576$, $p = 0.88$). Narrative length, in C-units, did not distinguish between either group of CWE and their peers. In fact, both groups of CWE produced slightly longer narratives than did their TD matches (recent-onset/TD comparison means 34.4 (SD = 7.9) and 30.2 (8.5), respectively ($t = 1.1455$, $p = 0.27$); chronic/TD means 33.6 (11.7), 31.8 (7.3), respectively ($t = 0.5061$, $p = 0.62$).

Lexical diversity also showed few differences between the groups. CWE-R demonstrated mean VOCD scores of 28.9 (SD = 7.9), while their peers achieved similar scores of 31.5 (SD = 12.4) ($t = 0.5581$, $p = 0.58$). CWE-C had mean VOCD values of 30.05 (SD = 13.3), while their peers had a mean value of 31.44 (SD = 5.7). There was large variability seen within the CWE-C group, forcing a non-parametric statistic. Nevertheless, no statistically significant differences emerged between the groups for this variable (Mann–Whitney U converted to Wilcoxon $z = -1.2651$, $p = 0.21$).

Syntactic complexity, as measured by proportion of complex C-units, did not statistically differentiate between groups. CWE-R and their peers used an average of 7.7% and 8.7% complex C-units ($t = -0.328$, $p = 0.75$), while CWE-C and their peers showed a larger difference (3.4% and 7.4%, respectively, $t = -1.6730$, $p = 0.11$); however, this difference was not statistically significant. We did not find differences among groups in use of cohesive devices (e.g. conjunction, referential pronouns). There was little range in the proportional use of these devices. CWE-R used the fewest, at 1.8%, while CWE-C used the most, at 2.1%. Both groups of TD peers used an average of 1.9%. Neither contrast reached significance (CWE-C and peers, $t = 0.5932$, $p = 0.56$; CWE-R and peers, $t = -0.5868$, $p = 0.56$).

In contrast, narrative structure distinguished the two groups of CWE and their TD peers (table 1). For total number of narrative elements, mean scores were fairly close when CWE-R and their peers were compared, at 8.1 (SD = 3.98) and 9.3 (3.4), respectively ($t = 0.7244$, $p = 0.48$) and no individual story component showed significantly differing level of inclusion, except for specifying the story setting appropriately. For that story component, CWE-R included a mean of only 0.9 (SD = 0.74) elements, while their peers included 1.5 (SD = 0.52) ($t = -2.4579$, $p = 0.03$; not significant after Bonferroni correction).

However, CWE-C included only an average of 8.3 (SD = 3.14) elements per story, while their TD peers, as expected of a slightly older group of children, produced an average of 12.2 (SD = 2.19) elements; homogeneity of variance was violated, forcing a non-parametric analysis ($Z = -3.546$, $p = 0.0002$; Cliff's delta = 0.623). CWE-C differed from their peers in specifying the story setting (mean = 1.1876 (0.7), 1.6876 (0.6), respectively; $Z = -2.3004$, $p = 0.02$; Cliff's delta = 0.41), initiating events (mean = 2.813 (1.3), 3.813 (0.99), respectively; $Z = -2.62$, $p = 0.01$; Cliff's delta = 0.46), and attempts (mean = 3.1875 (2.1), 5.25 (1.28), respectively; $Z = -2.832$, $p = 0.002$; Cliff's delta = 0.5). For specification of character goals, differences were also observed (CWE-C mean = 0.13 (0.35), peers' mean = 0.4375 (0.51); $Z = -1.935$, $p = 0.05$; Cliff's delta = 0.34). Story outcomes did not distinguish CWE-C and their peers, primarily because both groups of children virtually always included this component (mean = 0.94 (0.26), 1.0 (0), respectively; Z = −1.0, $p = 0.35$). Of all story subcomponents, only attempts differentiated the CWE-C from their typical peers after correction for multiple comparisons.

Table 1. Narrative components during the story retell task

	CWE-R ($N = 10$)	TD-R ($N = 10$)	CWE-C ($N = 15$)	TD-C ($N = 15$)
Narrative total	8.1 (3.98)	9.3 (3.4)	8.13 (3.14)	12.3 (2.19)*
Setting	0.9 (0.74)	1.6 (0.52)	1.2 (0.68)	1.7 (0.59)
Initiating events	2.6 (1.35)	3.1 (0.99)	2.8 (1.3)	3.9 (0.99)
Goals	0.1 (0.32)	0.4 (0.52)	0.13 (0.35)	0.4 (0.51)
Attempts	3.8 (2.2)	3.5 (2.5)	3.1 (2.1)	5.3 (1.28)**
Outcome	0.7 (0.48)	0.7 (0.48)	0.93 (0.26)	1.0 (0)

Notes: Differences for narrative total were significant for CWE-C/TD-C. For the story subcomponents, after Bonferroni correction, only differences between attempts (CWE-C/TD-C) met the threshold for significance.
*< 0.00025; **< 0.0016.

Listener perceptions

The nine ratings for each narrative were averaged. Data were compared across participant groups using non-parametric *T*-tests (Mann–Whitney *U*-values converted to Wilcoxon *Z*-scores) appropriate to analysis of non-ratio survey data. A separate Mann–Whitney *U*-test was used for each one of the seven rating areas and were compared cross-sectionally in two sets of comparisons: CWE-R compared with TD-R, and CWE-C compared with their TD peers (TD-C). Significance level was adjusted to 0.008 due to multiple comparisons.

In terms of their overall quality, listeners scored CWE narratives (mean = 3.92) lower than TD narratives (mean = 4.7, $Z = -2.10$, $p = 0.04$, Cliff's delta = 0.27) (figure 2 and table 2). Scores for use of grammar were significantly depressed (mean = 4.33) for the CWE (mean = 3.46, $Z = -2.66$, $p = 0.008$; Cliff's delta = 0.345). Vocabulary ($Z = -2.42$, $p = 0.02$; Cliff's delta = 0.31) and fluency ($Z = -1.98$, $p = 0.05$; Cliff's delta = 0.26) ratings were depressed to a non-significant degree.

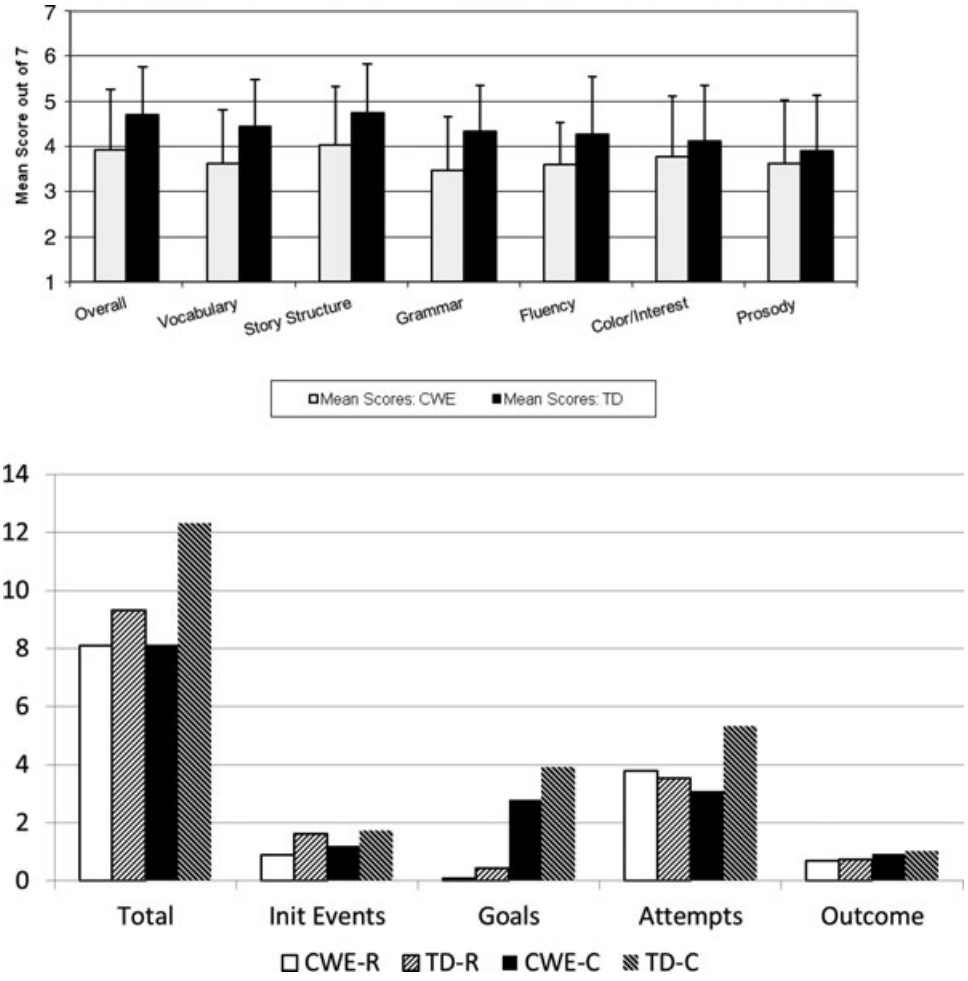

Figure 2. Mean scores of CWE and TD listener ratings.

Table 2. Comparison of listener ratings of children with epilepsy (CWE) and typically-developing children (TD)

	Mean Scores: CWE	Mean Scores: TD	Standard Deviation: CAVE	Standard Deviation: TD	*Z*	*p*
Overall	3.92	4.70	1.34	1.07	2.10	*0.0360
Vocabulary	3.63	4.45	1.19	1.04	− 2.42	0.0156
Story Structure	4.03	4.75	1.30	1.07	− 1.76	0.0789
Grammar	3.46	4.33	1.19	1.01	− 2.66	**0.0078
Fluency	3.59	4.26	0.93	1.29	− 1.98	0.0476
Color/ Interest	3.77	4.12	1.35	1.24	− 1.09	0.2769
Prosody	3.63	3.90	1.40	1.23	− 0.85	0.3928

*Significant at $p < .05$.
**Significant at $p < .008$.

Table 3. Correlations between age and listener ratings

		Age
Child Participants (n = 50}		
Overall	*r*	0.33
	p	*0.0198
Vocabulary	*r*	0.33
	p	*0.0181
Story Structure	*r*	0.34
	p	*0.0149
Grammar	*r*	0.33
	p	*0.219
Fluency	*r*	0.38
	p	*0.006
Color/Interest	*r*	0.19
	p	0.1837
Prosody	*r*	0.10
	p	0.0485

*Significant at $p < .01$.

Mean ages of CWE-C and CWE-R differed by 2 years, which had motivated two groups of age-matched TD children. Across the combined typical and CWE samples, correlation between age and overall quality listener score was significant ($r = 0.33$, $p = 0.02$). Correlations between age and story subcomponents were also significant for vocabulary, story structure, grammar and fluency, with *r*-values ranging from 0.33 to 0.38 (table 3), but there was no evident correlation

between age and colour/interest ($r = 0.19$) or prosody ($r = 0.10$). Although significant, these correlations were modest, suggesting that age did not strongly predict listener judgments when narrative task was held constant. The decision to limit any stimulus playlist to stories generated by children of roughly similar ages may have prevented a strong effect of age on listener judgments.

A one-way ANOVA to examine overall listener perceptions was significant for group at $p = 0.04$, $F(3, 46) = 2.92$; $\eta^2 = 0.16$). Fisher's LSD multiple-comparison test revealed that CWE-C received significantly lower scores than age-matched peers with no other group differences significant.

We hypothesized that differences in listener scores would be greater in the chronic comparison (CWE-C and TD-C) than in the recent-onset comparison (CWE-R and TD-R). Narrative ratings assigned to TD-C were significantly higher than narrative ratings assigned to CWE-C in four areas: overall quality ($Z = -3.13$, $p = 0.002$; Cliff's delta = 0.55), vocabulary ($Z = -3.28$, $p = 0.001$; Cliff's delta = 0.58), story structure ($Z = -2.91$, $p = 0.004$; Cliff's delta = 0.51), and grammar ($Z = -3.69$, $p = 0.000$; Cliff's delta = 0.65) (figure 2 and table 1). After adjustment for multiple comparisons, there were no significant differences between groups in ratings assigned for fluency ($Z = -2.39$, $p = 0.02$), colour/interest ($Z = -2.30$, $p = 0.02$), or prosody ($Z = -1.97$, $p = 0.484$). In contrast, and consistent with our predictions, there were no significant or noticeable differences in ratings assigned to CWE-R versus ratings assigned to TD-R (figure 2 and table 1) for any of the response variables.

Pearson correlations revealed that overall listener quality ratings were highly correlated with vocabulary ($r = 0.93$, $p < 0.008$), story structure ($r = 0.94$, $p < 0.008$), and grammar ratings ($r = 0.96$, $p < 0.008$). Overall quality ratings also correlated with fluency ($r = 0.70$, $p < 0.008$) and colour/interest ratings ($r = 0.59$, $p < 0.008$), but not prosody ratings ($r = 0.38$, $p = 0.0759$, n.s.).

Overall quality scores correlated with *CELF* Core Language scores ($r = 0.47$, $p < 0.01$), WASI verbal IQ scores ($r = 0.51$, $p < 0.01$), number of story grammar elements (N total, $r = 0.64$, $p < 0.01$) and vocabulary diversity (VOC-D; $r = 0.44$, $p < 0.01$). The only measure that did not significantly correlate with overall listener rating was total number of C-units ($r = 0.17$, $p < 0.01$) (table 4). It appears that listeners detect differences in stories that are also apparent in clinical language sample analysis and standardized assessments.

Table 4. Intercorrelations between listeners' overall quality scores and formal measures

Measure		*CELF CL*	Verbal IQ	N total	VOC-D	Total C units
			CWE (n = 25)			
Overall	*r*	0.47	0.51	0.64	0.44	0.17
	p	*.0009	*.0002	*.0000	*.0021	0.2622
CELF CL	*r*		0.75	0.31	0.28	−0.26
	P		*0.0000	*0.0337	0.0531	0.0806
Verbal IQ	*r*			0.38	0.22	−0.16
	p			0.0077	0.1399	0.2888
N total	*r*				0.28	0.17
	p				0.0541	0.2598
VOC-D	*r*					0.05
	p					0.7299

Note: CWE, all children with CWE; CELF CL, CELF Core Language Quotient; *N* total, number of words in the narrative; VOC-D, vocabulary diversity.
*Significant at $p < .01$.

Discussion

Caplan *et al.* (2009) estimate that up to 25% of CWE ages 6–8 are linguistically impaired, 33% of those 8–12 years, and up to half of CWE ages 12–15. Fastenau *et al.* (2008) found that almost half of their CWE from ages 8 to 15 years would meet criteria for a diagnosis of learning disability. This is a large number of school-aged children, given the high incidence of epilepsy in this age group, yet there is virtually no coverage of this problem in the professional literature addressed to practising SLPs.

In the current study, CWE, who were carefully matched to typical peers by gender and on age, scored significantly lower than did TD peers on the CELF, a standardized language assessment. On average, however, scores fell within 1 SD of the normative mean and, therefore, would not normally trigger a clinical diagnosis of language impairment, with referral for treatment. The same was true of performance IQ scores for both groups of CWE and for verbal IQ scores in children with chronic epilepsy (CWE-C); thus, we cannot rule out that the language differences we detected in this study were specific and not a reflection of subtle cognitive impairments that either predated or accompanied development of seizure disorder in the chronic group. Critically, focal epilepsy may negatively impact standardized test scores, but not to such an extent that the CWE would obviously qualify for speech/language or educational interventions, according to most guidelines, particularly if no baseline measures of performance prior to diagnosis were available.

No professional guidelines appear to suggest a speech–language evaluation for CWE at diagnosis, although comparable recommendations for baseline neuropsychological evaluation are emerging (Loring 2010). This gap in baseline assessment contrasts with the growing number of American states that now require preseason baseline assessment of student athletes at risk

for concussion to measure possible skill losses over time (Fjordbak 2011).

The largest differences between groups were seen in the functional measure: storytelling, both in terms of structural analysis and listener judgments. Narratives produced by CWE-C were scored much more poorly than the others, although their narratives contained more utterances. However, their typical utterances were shorter, and did not contain as many scorable story elements. CWE-C talked as much, if not more, than peers, but conveyed less information to listeners.

Listener ratings of narratives from CWE and matched TD peers were not greatly different when epilepsy was of recent onset. However, CWE-C narrative ratings were significantly lower than TD-C ratings, specifically in the areas of overall quality, vocabulary, story structure and grammar. Listeners perceived stories produced by CWE-C as less well put together and less syntactically and lexically complex, but no different in prosody, fluency or interest.

Listeners provided significantly lower overall quality and story structure ratings to narratives produced by CWE-C than TD-C. This finding is consistent with Caplan *et al.*'s (2002) finding that CWE produced more disorganized narratives than healthy peers. Conversational or narrative samples that lack cohesion or specificity are also typical of children with frank language disorders, such as SLI (for an extensive review, see Leonard 1998).

In the current investigation, children whose epilepsy was more recent in onset did not demonstrate the same functional deficits in language production that children with chronic seizures demonstrated. The chronic group was older than the recent-onset group (mean ages = 116 and 92 months, respectively). Story production is a higher-order task that requires executive functions such as planning and organization. As a general rule, the older children's narratives were longer and more complicated, thus requiring more planning and better organization. Deficits in this age range may have been more apparent to listeners than at younger ages, when stories were less well-elaborated.

Findings from the narrative task thus provide evidence of a functional language deficit associated with LRE, particularly in children who have lived with the disorder for more than 3 years. Our study results strongly suggest the utility of assessing CWE soon after diagnosis, using a basic battery of IQ and language assessments (including collection and analysis of a spontaneous language sample), and reassessment on a periodic basis to monitor cognitive and language status over time. Thus, we agree with Caplan *et al.*'s (2004) recommendation that CWE be given thorough language assessments, with particular emphasis on tracking children whose seizures are poorly controlled, who receive AED polytherapies and who have other risk factors for subtle linguistic differences (such as lower socio-economic status or non-native language proficiency). We note that baseline psycho-educational assessment of CWE is increasingly frequently recommended in the medical literature to monitor for potential disorder at time of onset, as well as gradual decline in function (see most recently the calls by Austin and Fastenau 2010, and Loring 2010), although our review did not find any mention of this issue in the psycho-educational or speech–language literature. Loring (2010) specifically suggests that schools treat children with newly diagnosed epilepsy similarly to those who experience traumatic brain injury, and we endorse this recommendation.

Although the elicitation and scoring of expressive language samples can be a time-consuming task, language sample analysis taps the real-world contexts of expressive language performance. Some standardized language sampling assessment tools have become available recently, e.g. the Strong Narrative Assessment Procedure (Strong 1998), and the Expression, Reception and Recall of Narrative Instrument (ERNNI) (Bishop 2004); they are less time-consuming and provide normative scores to be used in clinical referrals for intervention, if necessary.

CWE may present with difficulties in the areas of learning, memory, attention and executive functioning (Oostrom *et al.* 2005, Drewel and Caplan 2007), skills critical to conversational discourse and narrative production. Such basic cognitive impairments also can contribute to higher-level linguistic deficits because these problems can make it difficult to process and respond to language demands across different contexts (Drewel and Caplan 2007).

All of the CWE in this study were receiving AEDs. We could not systematically evaluate the effects of specific drugs or mono- versus polytherapy on the observed differences in language and psycho-educational function, particularly in the CWE-C, who, by definition, had taken them for longer periods of time. While untangling this relationship is important, we note that, whether as a function of their seizures *or* their medications, CWE appear to be at risk for the development of perceptible changes in speech and language production and merit periodic base-lining and monitoring by a speech–language pathologist.

A major limitation of this study is its cross-sectional and retrospective design, as well as limited sample size. A prospective design that would have more closely matched children for age, gender, SES, as well as IQ would have better informed the findings. Although we believe that the language differences that we found are specific to the children's history of seizures, both groups of children under-scored their peers on verbal IQ tasks, and we cannot rule out the possibility that

the CWE-C, in particular, were not verbally impaired BEFORE their seizures began. This is why a longitudinal study specifically tracking language abilities in children diagnosed with seizure disorders is so critically needed.

Additionally, because the original focus of the larger project was neuroimaging, a number of potentially influential factors were not balanced across the sample (e.g. as it was unclear how SES should affect brain imaging results, these data were not systematically gathered or controlled). Thus, the findings should be considered preliminary. However, their congruence with a number of other recent reports showing CWE at risk for language and psycho-educational deficits should be considered cautionary, and supportive of the need for further research by communication disorders specialists as well as a higher level of baseline assessment for CWE after initial diagnosis.

A longitudinal study that contrasts expressive and receptive language skills at the onset of seizures with these skills over the course of childhood epilepsy would better ascertain if seizures lead to diminished language abilities. It could address whether lower performance on measures of cognition and language is caused by seizures, their treatment or some underlying neuropathology. However, few such prospective studies had been reported in the recent literature (Benn *et al.* 2010, Dunn *et al.* 2010, Jones *et al.* 2010, Oostrom *et al.* 2005, Hermann *et al.* 2008) and they did not use the specific language measures that were the focus of the current study.

Second, a number of socio-economic variables may affect language performance, as well as moderate outcomes in childhood epilepsy (Fastenau *et al.* 2004). We had to obtain racial and parental education information *post hoc* and could not match or control for these variables. However, the two groups of CWE did not differ substantially on these values, despite differences in their language performance profiles. Clearly, such factors should be carefully controlled in follow-up studies. Fastenau *et al.* (2004) also note that additional variables, such as family support and home environment, may moderate academic outcomes in CWE.

The majority of studies describing language profiles in epilepsy involve adults. The current study focused on children having LRE with focus in the left hemisphere. It is also important to track language performance in children with more severe profiles of epilepsy. While a strength of this study was that we excluded symptomatic causes of epilepsy that confound some other studies, such narrow inclusion criteria necessarily limit the generalizability of our findings to the larger population of CWE. We suspect that children with more complex profiles may show even greater levels of impairment.

We recommend the use of language samples in future work examining the language skills of CWE. Spontaneous language samples capture functional aspects of language and tap into communicative skills that standardized tests do not. Studies that report only standardized test scores may fail to capture the impact of epilepsy on communicative ability. They examine language in a decontextualized manner and are not as sensitive or ecologically valid as language samples in detecting the subtle expressive language differences that may distinguish CWE from their seizure-free peers. The converging evidence provided by the present study, which paired standardized test scores with both clinical language sample analysis, and then added listener judgments, suggest measurable consequences of living with a chronic seizure disorder which merit further investigation. Baseline measurements of the language and psycho-educational performance of CWE appear to be strongly motivated, so that any discernible losses in proficiency over time can be referred for intervention.

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Notes

1. The abbreviation 'CWE' will be used to refer to the clinical samples; however, all children had the more specific diagnosis of LRE.
2. Based on Camfield and Camfield (2002b), a ceiling of 'more than 20 seizures' was applied because a cut-off at fewer than 20 seizures best predicts eventual outcomes in childhood-onset epilepsy and minimizes imprecision; parents experience difficulty in precise recall above this number.

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Appendix A: Listener rating sheet

INSTUCTIONS: You will listen to 10 child-narrated stories based on the wordless picture book, *Frog, Where Are you*? Each recording is approximately 4 minutes long. Your task is to rate the quality of narratives based on the criteria listed in the attached surveys. After listening to each narrative, you will be provided with as much time as needed to complete a survey. Please do not hesitate to ask any questions.

Before you begin, please answer the following questions:

Do you have any history of hearing loss? Y N

Are you a native speaker of English? Y N

Note: In some of these stories, children may not pronounce their words correctly (articulation errors). Please do not allow this to affect your ratings.

OVERALL QUALITY

☹ 1 Very low quality 2 3 4 5 6 7 Very high quality ☺

For example, you might base your judgment on:
- Amount of information conveyed
- Clarity of the story
- Ease of presentation

VOCABULARY

☹ 1 Very low quality 2 3 4 5 6 7 Very high quality ☺

For example, you might base your judgment on:
- Level of vocabulary
- Variety of vocabulary
- Relevance of vocabulary to the story

STORY STRUCTURE

☹ 1 Very low quality 2 3 4 5 6 7 Very high quality ☺

For example, you might base your judgment on:
- Inclusion of critical parts of the story
- Child followed the theme of the story

GRAMMAR

☹ 1 Very low quality 2 3 4 5 6 7 Very high quality ☺

For example, you might base your judgment on:
- Use of correct grammar/complete sentences
- Complexity of sentences
- Length of sentences

FLUENCY/FLOW

☹ 1 Very low quality 2 3 4 5 6 7 Very high quality ☺

For example, you might base your judgment on:
- Smoothness of speech

(or does the child pause, hesitate, and/or use words such as *um* or *uh*?)

COLOR/INTEREST

☹ 1 Very low quality 2 3 4 5 6 7 Very high quality ☺

For example, you might base your judgment on:
- Does the child sound as if he or she is telling a story (vs. having a conversation)?
- Inclusion of emotion
- Inclusion of humor

PROSODY

☹ 1 Very low quality 2 3 4 5 6 7 Very high quality ☺

For example, you might base your judgment on:
- Rhythm & Intonation
- Stress Patterns
- Does the narrative sound expressive (high quality) and filled with emotion, or does it sound "flat" (low quality)?