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Fluency patterns in narratives from children with localization related epilepsy

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ABSTRACT

This study assessed the relationship between fluency and language demand in children with epilepsy, a group known to demonstrate depressed language skills. Disfluency type and frequencies were analyzed in elicited narratives from 52 children. Half of these children had localization-related epilepsy (CWE), while the others were age- and gender-matched typically-developing (TD) peers. CWE were found to be significantly more disfluent overall than their matched TD peers during narrative productions, and demonstrated a higher proportion of stutter-like disfluencies, particularly prolongations. The current study adds to an emerging literature that has found depressed language skills and listener perceptions of verbal ability in children with chronic seizure activity, and contributes to the small but growing literature that suggests that disfluency during spoken language tasks may be a subtle marker of expressive language impairment.

Educational objectives: The reader will be able to (a) describe why children with epilepsy might be at greater risk for language delays and or increased levels of disfluency; (b) describe profiles of fluency that differentiated children with chronic and recent-onset epilepsy from their age and gender matched peers; and (c) apply this information to monitoring of children with seizure disorder on their caseloads.

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1. Introduction

1.1. Speech and language characteristics of children with epilepsy

Epilepsy is one of the most common neurological disorders that occur during childhood and adolescence. Approximately 326,000 school children (up to age 15) have epilepsy, and by age 20, about 1% of the United States population will develop epilepsy (Epilepsy Foundation, 2012). A recent epidemiological study estimated the rate of current epilepsy/seizure disorder at above 6 cases per 1000 children (Russ, Larson, & Halfon, 2012). According to the Epilepsy Foundation of America 45,000 new cases are diagnosed in children under the age of 15 each year. Despite the fact that epilepsy is such a frequent disorder of childhood, there are virtually no reports of the speech and language problems seen in this population in any of the major communication disorders journals, with the exception of Landau–Kleffner Syndrome (LKS), whose major clinical feature is language regression (Rapin, 2006). Camfield and Camfield (2002) place LKS among the "catastrophic" epilepsy syndromes.

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However, it has been documented that even the general population of children with epilepsy (CWE) are at an increased risk for developing speech and language disorders (see review by Pal, 2011, discussion in Caplan et al., 2009, 2010). In one study documenting the potential educational consequences of seizure disorder, more than 37% of the children with epilepsy who participated were found to have previously undiagnosed language impairments after undergoing thorough psycho-educational and language testing (Parkinson, 2002). In a survey of the parents of CWE conducted 8–9 years after initial diagnosis of epilepsy, Benn et al. (2010) found that CWE (even those with normal anatomical findings) were almost 6 times more likely to have been referred for speech/language problems than their unaffected siblings. The increased risk for CWE referrals to SLP remained even after children with CWE who had one specific syndrome (BECTS, another syndrome already known to convey higher risk for language impairment) were removed from the analysis.

There is great variability in the manifestation of speech and language problems among the larger population of children with epilepsy. They may demonstrate a variety of symptoms that impair their ability to comprehend and produce language (Svoboda, 2004). Caplan and colleagues have conducted a number of studies that detail a broad range of language impairments in children with typically seen etiologies of seizure disorder. They include both structural differences, as well as differences in the cohesion of spoken discourse. We discuss these findings in greater detail below.

For individual children, it has been difficult to ascertain whether any detected communication problems are the result of anatomical lesions. In such cases, seizures are not thought to cause language problems directly, but stem jointly from the underlying condition that produces the seizure activity (Berg, Hesdorffer, & Zelko, 2011). Treatment with antiepileptic medications (also known as antiepileptic drugs or AEDs) is also thought to impact development of speech and language problems (Austin & Caplan, 2007; Sechi, Cocco, Donofrio, Deriu, & Rosati, 2006; Svoboda, 2004). Thus, at this time, it is unknown whether, for individual children, speech and language impairments exist at the time of seizure onset, or result from a more chronic decline in cognitive skills over time, as a result either of continued seizure activity, or medications used to control seizures.

Nevertheless, the mounting data suggesting that CWE are at risk for increased incidence of psycho-educational and communicative impairments has prompted some disciplines (pediatrics, neurology) to recommend routine baseline testing of skills in children at the time that first diagnosis of seizure disorder is made (Austin & Fastenau, 2010; Loring, 2010), in much the same way that baseline testing is being recommended for children at risk for head trauma due to sports participation. These recommendations have not yet been disseminated in the speech-language pathology literature, which has virtually no reports on communicative abilities in CWE.

Thus, one clear trend has emerged in the literature on CWE: many of these children go on to be diagnosed with problems that impact their educational achievement. However, despite a recent call to benchmark psycho-educational skills at onset of seizure activity, there is continued dispute as to whether later impairments emerge as a consequence of chronic seizure activity or its medical management, or reflect the generalized neurological impairments that give rise to seizure activity in the first place (Strekas et al., in press). For this reason, we have been tracking a cohort of CWE, divided into two groups: children whose seizure disorder is of recent onset (abbreviated CWE-R) and those with long-standing, chronic seizure activity (CWE-C). Virtually unique to the assessment literature in this population, we have conducted language sample analysis and standardized testing, as well as experimental testing of language skills while children are imaged using functional Magnetic Resonance Imaging. Our results thus far are congruent with the growing literature on language impairment in childhood seizure disorder, in finding that children with chronic epilepsy generate narratives that are less complex and less well-structured than those generated by age- and gender-matched peers. Listener judgments provided added evidence that the children with chronic epilepsy produced samples that were judged less positively along a wide number of listener dimensions (Strekas et al., in press) In this paper, we ask whether the fluency of CWE differs from that of typical peers, given their lower performance on a number of linguistic measures. In the sections below, we explore why we might expect fluency impairments in children with lowered levels of language performance, and, if differences in fluency profiles emerge, how they might inform other measures used to gauge language ability in clinical populations.

1.2. Fluency and language

Language production and language fluency are extremely intertwined linguistic processes. Spoken language production involves a number of discrete and concurrent stages (Levelt, 1989). The speaker must have the intent to communicate an idea, develop it into a nonlinguistic representation (conceptualization), translate the nonlinguistic representation into a linguistic message composed of the appropriate semantic and syntactic components (formulation), and finally, convert the linguistic message into an articulatory plan that can be executed (articulation). Monitoring occurs at all stages of language production, and malfunctions in the encoding system may arise and manifest as disfluencies in the production of speech and language (Guo, Tomblin, & Samelson, 2008; Postma & Kolk, 1993).

A variety of hypotheses have been proposed to explain the origins of speech disfluencies in the language production process. Models such as WEAVER++ (Levelt, Roelofs, & Meyer, 1999), attempt to account for typical types of disfluencies in all speakers that may arise either during conceptual mapping, syntactic formulation or word retrieval and encoding. Other speech production models such as the Covert Repair Hypothesis (Postma & Kolk, 1993), Neuropsycholinguistic theory (Perkins, Kent, & Curlee, 1991), and Demands and Capacity model (Bernstein Ratner, 2000; Starkweather, 1987) were developed as theories to explain how disfluencies arise in persons who stutter. Although each model proposes a different

mechanism for the production of stuttered disfluencies, they all include aspects of language formulation as a primary cause of speech disfluencies (Yaruss, Newman, & Flora, 1999).

Disfluencies in spoken language may be used as strategies to "buy time" and repair errors. In addition, different types of disfluencies may reflect different types of processing breakdowns (Boomer, 1965; Clark & Fox Tree, 2002; Goldman-Eisler, 1972; Levelt, 1989; Maclay & Osgood, 1959; Postma & Kolk, 1993). For example, a silent pause may occur if the speaker has difficulty formulating concepts or retrieving semantic or syntactic information, while a filled pause or revision may occur if the incorrect concept or linguistic information was activated (Guo et al., 2008). Together with other researchers, we believe that insight about the underlying language processes can be gained by studying the type of disfluencies present in spontaneous discourse of various populations (Wijnen, 1990).

1.2.1. Language formulation and fluency in children

To our knowledge, there are no prior studies of speech fluency in children with epilepsy. Thus, we will preface our study with a discussion of what is generally known about language formulation and fluency in children. During typical language development, children undergo a period of normal disfluency between ages 2 and 3, which may be related to increased language formulation demands (Colburn & Mysak, 1982a, 1982b; DeJoy & Gregory, 1985; Hall, Yamashita, & Aram, 1993; Lees, Anderson, & Martin, 1999; Yaruss et al., 1999). At this time, children experience an exponential growth in their language abilities as their lexicon expands and their ability to understand and use more complex syntax improves. When this happens, the children's demands for language exceed their productive abilities and can result in an increase in disfluencies in their speech (Adams, 1990; Starkweather & Gottwald, 1990; Yaruss et al., 1999). Longer and more complex utterances have been associated with increased disfluencies (Bernstein Ratner & Sih, 1987; Bernstein, 1981; Colburn & Mysak, 1982a; Hall et al., 1993; McLaughlin, 1989; Yaruss et al., 1999).

1.2.2. Relationship between fluency and atypical language development

There has been increased interest in studying the relationship between fluency and language, especially in children with compromised language functioning. It has been shown that individuals with weaker language skills, such as those with specific language impairment (SLI), may demonstrate increased levels of disfluencies in their speech (Boscolo, Bernstein Ratner, & Rescorla, 2002; Finneran, Leonard, & Miller, 2009; Guo et al., 2008; Hall, 1977, 1999; Hall et al., 1993; MacLachlan & Chapman, 1988). However, it is also possible that disfluency and impaired language ability may be co-morbid symptoms of another larger overarching disorder or condition and may co-occur without having the same underlying etiology.

Since fluency seems to be correlated with language development and complexity, some researchers have predicted that children with impaired speech and language skills would demonstrate increased number of disfluencies in their spontaneous speech (Boscolo et al., 2002; Finneran et al., 2009; Guo et al., 2008). Although there is some evidence to support this hypothesis, there is also contradictory evidence and the mechanism that produces these findings remains unclear.

One study suggested that children with language impairments demonstrate differences in both the number as well as the type of disfluencies in their conversational speech. Hall et al. (1993) studied spontaneous speech samples from 60 children who were between 4 and 6 years old. The children were diagnosed with speech and language impairments, but were not diagnosed as children who stutter. After coding the speech samples, the children were separated into "high disfluency" (HD) and "normal disfluency" (ND) groups based on the total number of disfluencies in their speech sample. Hall et al. (1993) reported that, although both stutter-like and normal disfluencies were seen in both groups, the 10 participants in the HD group had a greater frequency of stutter-like disfluencies (e.g., part-word repetitions, prolongation, broken words and tense pauses). The HD group also scored significantly lower on vocabulary tests (e.g., PPVT, EOWVT) and other language assessments. Based on these findings, Hall et al. proposed that disfluencies in children with language disorders may result from a "dysynchrony" between speech processes and language processes and from the "mismatches between speaking demands and capacities" (p. 568).

Hall (1996) conducted a follow-up longitudinal study of 9 of the 10 children from the HD group in her 1993 study, when they were between 7 and 9 years old, to see if disfluency could be used as a marker of a change in linguistic abilities over time. She found that all of the participants demonstrated decreases in their overall frequency of disfluencies when they were older, although some participants had an increase in the frequency of stutter-like disfluencies. Increased fluency with improved language skills was also noted in these children, despite great variability among individual participants.

Findings from Boscolo et al. (2002) provide further support for the relationship between disfluency and language abilities found in Hall's 1996 study. Types and frequency of disfluencies in narrative productions from 22 pairs of 9-year-old children with and without a history of specific expressive language impairment (HSLI-E) were compared. Children with HSLI-E were significantly more disfluent than their typically-developing peers. Although stutter-like disfluencies were relatively rare in both groups, children with HSLI-E had significantly more than those without HSLI-E. It is important to note that at the time of the study, the children with HSLI-E had matured to levels of language skills within the normal range, suggesting that "subtle language formulation difficulties can contribute to fluency breakdown" (Boscolo et al., 2002).

Guo et al. (2008) further examined the types, frequencies and distribution of speech disruptions, including pauses and vocal hesitations, in a comparison of narrative productions from children with and without specific language impairment. Guo et al. found that children with SLI produced more speech disruptions than the age-matched group, but not the language-matched group; suggesting that speech disfluency is related to expressive language level regardless of age. This finding is consistent with previous studies of language and fluency (Boscolo et al., 2002; Dollaghan and Campbell, 1992). Additionally,

Guo et al. noted that children with SLI produced disfluencies mostly at phrase boundaries rather than before sentences, clauses or words. The authors suggest that these disruptions may result from less developed semantic and syntactic systems in children with SLI (Guo et al., 2008). However, findings from Finneran et al. (2009) demonstrated that children with SLI produced increased levels of disfluency as compared to their TD peers, even when grammatical accuracy was controlled, suggesting that disfluency may represent subtle language formulation difficulties, and may be a general feature of compromised expressive language skill.

1.3. Summary and research questions

Previous research demonstrates that children with epilepsy are a group at high risk to demonstrate depressed language skills (Caplan et al., 2009; Parkinson, 2002). However, very few of the studies of language skills in CWE have used language corpus analysis to examine other features of spoken language in this population.

A notable exception is the large series of studies by Caplan and colleagues, who have not analyzed speech fluency but have analyzed complex language skills using naturalistic language samples, as well as standardized language 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 discussion in Caplan et al., 2006) as well as lowered scores on the *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.

Prior research suggests that children with weaker language skills (e.g., those with SLI,) may demonstrate increased levels of disfluencies. Because our ongoing work with a cohort of children with CWE has identified depressed language skills in children with chronic seizure disorder, we extended our analyses of their spoken language skills by examining fluency in narrative productions from the children with epilepsy and their age- and gender-matched peers.

The current study thus sought to determine if children with epilepsy (CWE) differ in the number and types of disfluencies in their narrative speech productions as compared to age- and gender-matched typically-developing peers. Additionally, differences between children with chronic epilepsy and children with recent-onset epilepsy were examined to test the hypothesis that differences in language abilities between these cohorts, as measured both by standardized language test scores, and narrative performance, will also be mirrored in their fluency profiles.

2. Method

Participants were part of a larger National Institutes of Health (NIH)-funded study[1] designed to examine effects of localization-related epilepsy (LRE) on children's language performance, as well as dynamic language processing as measured by functional Magnetic Resonance Imaging (fMRI). The project sought to investigate how history and duration of seizure activity appear to influence brain substrates of language processing in children with epilepsy (profiles of language dominance and intra-hemispheric activation profiles during language processing tasks (see earlier reports by Berl et al., 2005, 2010; Gaillard et al., 2007; Mbwana et al., 2009)). All participants in the larger study underwent a neurological examination and standardized psycho-educational testing as well as fMRI scanning during language tasks.

2.1. Participants

From the larger cohort, fifty-two (52) participants, consisting of 26 children with epilepsy (CWE) and 26 typically-developing (TD) peers were evaluated, because they could be matched closely enough by gender and age (±3 months) to permit the naturalistic language-based analyses to be reported here. These participants were further subdivided into four groups. The first group contained 10 children with recent-onset (<1 year following second seizure) epilepsy (CWE-R) while another group contained 10 typically-developing peers, who were age- (within 3 months) and gender-matched to the children with recent-onset epilepsy (TD-R). A third group contained 16 children with chronic (>3 years) epilepsy (CWE-C), who was matched with another group which contained 16 age- (within 3 months) and gender-matched typically-developing peers (TD-C).

A cut-off of three years with epilepsy was used to divide the CWE cohorts. There is a high rate of recovery from first seizure activity, and it may take a number of years for both the symptoms and attempts to manage them make it clear that "the epilepsy will not go away" (Camfield & Camfield, 2002). Because approximately 90% of recurrent seizures appear within two years of the first episode (Camfield & Camfield, 2008), three years post-onset was chosen as the boundary to demarcate

[1] POLER: Plasticity of Language in Epilepsy Research, PI: William Davis Gaillard NINDS R01 NS44280.

Table 1
Demographic profiles of participants.

Groups	CWE-R	TD-R	CWE-C	TD-C
Number of participants	10	10	16	16
Age (months) mean (stdev)	92 (29.0)	92 (28.6)	117 (25.0)	117 (24.8)
Full-Scale IQ mean (stdev)	100.7 (16.0)	112.7 (10.7)	96.6 (13.5)	117.3 (18.0)
Verbal IQ mean (stdev)	102.7 (17.4)	110.4 (12.6)	98.4 (13.2)	119.1 (19.0)
Age (months) at 1st seizure mean	74		55	
Total lifetime seizures[a]	4.5		7.1	

[a] 2 = 2 seizures, 3 = 3 seizures, 4 = 4 seizures, 5 = 5 seizures, 6 = 6–10 seizures, 7 = 11–20 seizures, 8 = >20 seizures.

the two groups of CWE. Most children in the CWE-R group had experienced more than two seizures. A non-linear coding scale was used to record the total lifetime number of seizures for all participants in CWE groups (see Table 1), with scores capped at 8 for children who experienced more than 20 seizures.

CWE-R and TD-R each contained 4 females and 6 males; the mean age of the children in these groups was 92 months (range 50–139 months). CWE-C and TD-C each contained 8 females and 8 males, with a mean age of 117 months (range 75–155 months). The average age at which seizure onset occurred in CWE-R was 74 months and in CWE-C was 55 months. The participants in CWE-C and CWE-R had electroencephalogram (EEG) or other clinical evidence that suggested a left hemisphere focus of seizure activity, and all were diagnosed with localization-related epilepsy localized to the temporal lobe. All children also had normal brain anatomy, as ascertained by imaging. All participants reported here were right-handed; there were left-handed children in the larger study, but they provided too few potential matches to allow analysis. No child was reported as receiving speech-language services at time of initial testing. See Table 1 for demographic profiles.

2.2. Psycho-educational testing

Standardized psycho-educational testing for the children in this study was performed as part of the larger protocol at Children's National Medical Center (CNMC). The *Wechsler Abbreviated Scales of Intelligence* (*WASI*, Psychological Corp, 1999) or the *Differential Ability Scales* (*DAS*, Elliot, 1990), for children less than 6 years of age, were used to obtain IQ scores. The *Clinical Evaluation of Language Fundamentals, Fourth Edition* (*CELF-4*, Semel, Wiig, & Secord, 2003), or the *Clinical Evaluation of Language Fundamentals, Preschool Edition* for children less than 5 years of age (*CELF-P*; Wiig, Secord, & Semel, 2004), and the *Expressive One Word Vocabulary Test* (EOWVT) were used to measure language abilities. Only the core subtests were administered on the *CELF-4* and *CELF-P* (Concepts and Following Directions; Word Structure; Recalling Sentences; Formulated Sentences). One CWE-C and 1 TD-C did not complete IQ testing and *EOWVT*, and therefore only 50 participants, instead of 52, were included in these analyses. Three CWE-C, 1 TD-C and 1 CWE-R did not complete testing to allow computation of the Core Language quotient. For this reason, only 47 child participants (instead of 52) were included the *CELF* analysis.

IQ, *EOWVT* and *CELF* test scores were entered as standard scores and 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); $F(3, 46) = 4.7$; $p = .006$, $h^2 = .235$). Performance was poorest by children with chronic epilepsy (M = 98.4; SD = 13.17), which differed significantly only from their typically-developing peers (M = 119.13, SD = 19.05). CWE-R also differed from the CWE-C's typical peers (M = 102.7; SD = 17.04), but not from their own age-matched peers (M = 110.4; SD = 12.58), and the two groups of CWE did not differ from each other in verbal IQ (MSE = 253.74, $p = .05$).

Performance IQ scores also showed an effect of group (F (3, 46) = 4.59; $p = .007$; $h^2 = .23$). By Fisher's LSD Multiple-Comparison Test (Lindman, 1974), 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 typically-developing children differ from each other (MSE = 198.91, $p = .05$). CWE-C achieved a mean standard score of 93.13 (SD = 13.1); CWE-R achieved a mean standard score of 97.7 (SD = 13.17). Typically-developing peers of CWE-C had average scores of 111.5 (SD = 15.7); peers of CWE-R had average scores of 111.6 (SD = 13.3).

Scores on the *CELF* and *CELF-P* showed effects of group as well. Group difference for the Core Language subtests was significant (F (3, 43) = 9.06, $p = .0009$; $h^2 = .36$). However, on this instrument, CWE-R showed the lowest average scores (M = 86.78; SD = 15.5), significantly different from performance by their typically-developing peers (M = 114.4; SD = 1.7) and the peers of CWE-C (M = 111.8; SD = 14.5). The same profile was shown by children with chronic epilepsy (M = 95.9; SD = 15.1). The two groups of CWE did not differ from each other on *CELF* performance (MSE = 201.56, $p = .05$).

2.3. Narrative elicitation

At CNMC, as part of the larger protocol that included standardized psycho-educational testing, and experimental measures obtained during fMRI scanning, researchers elicited stories using the wordless picture book, *Frog, Where Are You?* by Mercer Mayer (1969). Each participant was handed a copy of the book and asked to make up a story based on the pictures. *Frog, Where Are You?* was selected for narrative elicitation because it has been utilized before in many studies involving typical

Table 2
Characteristics of narratives.

Group	Number of words			Mean length of utterance in words		
	Mean	SD	Range	Mean	SD	Range
TD-C	266.19	81.94	117–474	8.41	1.52	5.32–11.07
TD-R	210.1	90.94	87–411	6.82	1.76	3.35–8.29
CWE-C	242.75	96.47	92–364	7.31	2.03	3.24–10.4
CWE-R	218	91.26	63–351	6.2	2.05	2.74–10.59

and language-impaired children from numerous linguistic communities (Berman and Slobin, 1994) and has large reference samples available in the Child Language Data Exchange System (CHILDES) archive database (MacWhinney, 2000).

Narrative has also been used to examine disfluency frequency in children who stutter (CWS) (Byrd, Logan, & Gillam, 2012). In that study CWS were more disfluent in narrative than in conversation, leading the researchers to recommend narrative as an efficient way to elicit disfluency in CWS.

The narratives were digitally recorded and transferred electronically to researchers at the University of Maryland, College Park (UMCP), along with a de-identified database containing psycho-educational test scores and medical information, such as seizure history and drug regimens. The audio-recordings were labeled using numerical codes, and the participants' identities were concealed from UMCP researchers, with the exception of age, gender and patient group information.

2.4. Coding of narratives

Samples were transcribed into Codes for Human Analysis of Transcripts (CHAT) using the CHILDES conventions (MacWhinney, 2000). The total corpus consisted of 12,424 words. CHAT provided a calculation for total number of utterances and total number of words for each participant. Mean length of utterance per word (MLUw) was calculated by dividing the total number of words by the total number of utterances. Table 2 provides descriptive characteristics by group regarding means and ranges for number of words and MLUw. We note that the children with chronic seizures and their peers tended to produce longer narratives, in terms of number of words, and length of utterance, as would be expected, since this cohort was older than the group of children with recent onset seizures and their peers. This is one reason why comparisons in the following sections are performed for each cohort separately.

Narrative samples were coded and analyzed for frequency and type of disfluencies. The disfluencies within the samples were separated into two categories: "typical disfluencies" and "stutter-like disfluencies" (SLD), since previous research suggests that narratives from children with language impairments contain a higher percentage of disfluencies that are more characteristic of persons who stutter (Boscolo et al., 2002; Hall et al., 1993). Typical disfluencies included whole-word repetitions, phrase repetitions, revisions, and interjections/filled pauses, while stutter-like disfluencies included part-word repetitions, prolongations, and broken words. Unfilled pauses longer than 250 ms were also coded and tabulated separately, since these pauses may reflect time needed for linguistic planning (Guo et al., 2008). Pauses shorter than 250 ms were considered to be related to articulation rather than cognitive function during speech production (Guo et al., 2008; Goldman-Eisler, 1961). To avoid double counting, disfluencies that were contained within revisions were classified only as revisions. However, unfilled pauses contained within revisions were noted, as this may reflect time needed to reformulate the linguistic message. Repetitions that were used purposely for emphasis (e.g., "*the big big rock*") were not coded as disfluencies. In addition, utterances that were directed toward the experimenter or were unrelated to the participant's narrative were excluded from the analysis.

To compute the frequency of disfluencies for each participant, the total number of each type of disfluency was divided by the total number of intended words in the narrative (excluding words in mazes, restarts and repetitions). Although a variety of methods have been used to compute the frequency of disfluencies in previous studies (e.g., by number of C-units, by total number of words), Dollaghan and Campbell (1992) suggested that using the total number of words to compute disfluency frequency was the most sensitive to language impairments. The frequency of unfilled pauses was also computed by dividing the total number of unfilled pauses by the total number of words.

2.5. Reliability measures

Measurement reliability was calculated for the four variables: total disfluency frequency, typical disfluency frequency, stutter-like disfluency frequency, and pause frequency. A second transcriber, who was blinded to condition, identified the occurrence of disfluencies, classified them as either typical or stutter-like, and identified pauses in the narratives for approximately 23% of the total sample. Reliability data were collected from 3 participants in each group. Pearson product-moment correlation produced reliability agreement with Cronbach's alpha at .991 for total disfluency frequency, .980 for typical disfluency, .908 for stutter-like disfluency frequency, and .707 for pause frequency. Given the relatively strong reliability found between coders, the first author's (MS's) coding was use for all statistical analyses to maintain consistency across samples. However, since reliability agreement for pause frequency was less than 90%, utterances included in speech rate measures

were imported into Praat (www.praat.org) software program to determine their length objectively and ensure that pauses were at least 250 ms or longer.

2.6. Analysis of disfluency measures

Although there was an overlap in age range between CWE-R and CWE-C, there was a significant group difference in age, with the chronic group and their peers, on average, 2 years older than the recent-onset pairs. As a result, some analyses were made only between children with recent-onset epilepsy and children with chronic epilepsy compared to their respective gender-matched typically-developing peers. Other analyses combined children with recent onset epilepsy with children with chronic epilepsy and compared them to all of the age- and gender-matched typically-developing peers to look more generally at differences between these groups.

To determine whether CWE (both recent-onset and chronic) had a higher percentage of disfluencies and unfilled pauses in their narrative productions than their typically-developing peers, Mann–Whitney *U* tests (converted to Wilcoxon *Z*) were used to compare total disfluency frequency, typical disfluency frequency, stutter-like disfluency frequency and pause frequency. Since variances were unequal, nonparametric statistics were used to analyze group comparisons (e.g., CWE vs. TD). Correlations between standardized language tests and disfluency frequency measures were performed using parametric statistics; however, disfluency frequency percentages were converted to rationalized arcsine units (RAU) since percentages are not normally distributed (Gaussian).

3. Results

3.1. Comparison of total disfluency and unfilled pause means: CWE and TD children

Analyses (Mann–Whitney *U* converted to Wilcoxon *Z*) revealed that CWE (mean frequency = 6.0%) had a significantly higher frequency of total disfluencies than their TD peers (mean frequency = 4.6%, $z = 1.98$, $p = 0.048$, $d = 0.466$, small effect) as shown in Table 3. However, CWE did not have a higher frequency of either typical disfluencies (mean frequency = 5.0%) or stutter-like disfluencies (mean frequency = 1.0%) than their TD peers (typical disfluency mean frequency = 4.1%, $z = 1.57$, $p = 0.116$; SLD mean frequency = 0.5%, $z = 1.95$, $p = 0.052$). However, differences in SLD frequency approached significance level with a *p*-value of 0.052. Unfilled pauses were also not significantly different between CWE (mean frequency = 10.5%) and their TD peers (mean frequency = 8.9%, $z = 1.63$, $p = 0.103$).

Analyzes (Mann–Whitney *U* converted to Wilcoxon *Z*) of specific disfluency types was conducted between the CWE and TD groups. The means and ranges are shown in Table 4. The *p*-value was set at $p < 0.01$ to adjust for multiple comparisons within disfluency categories. Overall, the distribution of disfluencies was similar between groups, with the exception of prolongations. CWE had significantly more prolongations as compared to the TD group ($z = 3.27$, $p = 0.001$, $d = 0.998$, large effect).

Table 3
Comparison of disfluency means and unfilled pauses of CWE and TD (in percentages).

	CWE		TD		*Z*	*p*
	Mean	SD	Mean	SD		
Total disfluencies	6.0	2.9	4.6	3.1	1.98	0.048*
Typical disfluencies	5.0	2.4	4.1	2.8	1.57	0.116
Stutter-like disfluencies	1.0	0.9	0.5	0.5	1.95	0.052
Unfilled pauses	10.5	3.0	8.9	3.2	1.63	0.103

* Significant at $p < .05$.

Table 4
Disfluency type means, standard deviations (SD) and ranges by group (in percentages).

Type	CWE			TD			*Z*	*p*
	Mean	SD	Range	Mean	SD	Range		
Typical disfluencies								
Whole-word repetitions	0.80	0.008	0–2.58	0.48	0.006	0–1.61	1.37	0.169
Phrase repetitions	0.52	0.006	0–2.35	0.49	0.009	0–3.75	1.43	0.154
Revisions	2.31	0.015	0–6.88	2.14	0.012	0–5.41	0.53	0.596
Interjections/filled pauses	0.85	0.015	0–5.29	0.82	0.013	0–3.96	1.51	0.131
Stutter-like disfluencies								
Part-word repetitions	0.65	0.007	0–2.27	0.44	0.004	0–1.21	0.56	0.578
Prolongations	0.29	0.005	0–1.92	0.01	0.007	0–0.34	3.27**	0.001
Broken words	0.05	0.002	0–0.28	0.03	0.009	0–0.38	0.20	0.984

** Significant at $p < .01$.

Table 5
Comparison of disfluency means and unfilled pauses of CWE-C and TD-C (in percentages).

	CWE-C		TD-C		*Z*	*p*
	Mean	SD	Mean	SD		
Total disfluencies	6.3	2.9	4.1	2.0	2.32	0.022*
Typical disfluencies	5.2	2.3	3.7	1.9	2.11	0.035*
Stutter-like disfluencies	1.1	0.9	0.4	0.3	1.98	0.048*
Unfilled pauses	10.0	3.1	8.6	2.7	1.34	0.19

* Significant at $p < .05$.

Table 6
Comparison of disfluency means and unfilled pauses of CWE-R and TD-R (in percentages).

	CWE-R		TD-R		*Z*	*p*
	Mean	SD	Mean	SD		
Total disfluencies	5.5	3.1	5.4	4.3	0.42	0.677
Typical disfluencies	4.5	2.7	4.7	3.9	0.15	0.880
Stutter-like disfluencies	1.0	1.0	0.7	0.7	0.61	0.540
Unfilled pauses	11.3	3.0	9.3	4.0	1.36	0.174

*Significant at $p < .05$.

3.2. Comparison of total disfluency and unfilled pause means: CWE-R and CWE-C

Disfluency frequencies in narratives from CWE-C were significantly higher than disfluency frequencies in narratives from TD-C in terms of: total disfluency frequency ($z = 2.32$, $p = 0.022$, $d = 0.883$, large effect), typical disfluency frequency ($z = 2.11$, $p = 0.035$, $d = 0.711$, medium effect), and stutter-like disfluency frequency ($z = 1.98$, $p = 0.048$, $d = 1.043$, large effect). There was no significant difference in unfilled pause frequency ($z = 1.34$, $p = 0.19$). See Table 5.

There were no significant differences in disfluency or pause frequencies for CWE-R vs. TD-R (see Table 6).

3.3. Relationship between age and disfluency means

Although not a focus of our study, correlations between age and disfluency frequencies support the prediction that older children, who are assumed to be more proficient in using more complex language, have lower frequencies of disfluencies (Starkweather, 1987; total disfluency frequency $r = -.37$, $p = 0.01$; typical disfluency frequency $r = -0.33$, $p = 0.02$; stutter-like disfluency frequency $r = -.21$, $p = 0.13$; see Table 7). This finding confirms the wisdom of comparing a small subset of the original sample for our current hypotheses; the current sample of 52 children were more closely matched for age between groups of CWE, rather than the entire sample, in which age was allowed to vary across a much wider range for imaging analyses.

3.4. Relationship between disfluency frequency means and standardized assessments

In this analysis, Pearson's product-moment correlations were used to assess the relationship between disfluency means and formal measures of language and IQ (see Table 8). Age was not added as an exploratory variable, because all scores had been age-normalized as standard scores. To adjust for multiple correlations, the *p*-value was set at 0.01. Results indicated that total disfluency frequency was not correlated with any standardized measure of IQ or language. As shown in Table 8, without the correction for multiple correlations, *WASI* Verbal IQ and *EOWVT* standard scores would correlate with total disfluency at the $p < 0.05$ level. Additionally, typical disfluency frequency and stutter-like disfluency frequency were not correlated with any standardized measure of IQ or language at $p < 0.01$. However, as noted in the Section 2, CWE scored significantly lower than their TD peers on standardized language testing, narrative values, and listener judgments. Thus, correlations among these values are not as meaningful as group differences reported earlier.

Table 7
Correlation of age and disfluency means in CWE and TD children.

	Age	
	Correlation (*r*)	*p*-Value
Total disfluency	−0.37	0.01**
Typical disfluency	−0.33	0.02*
Stutter-like disfluency	−0.21	0.13

* Significant at $p < .05$.
** Significant at $p < .01$.

Table 8
Intercorrelations between total disfluency mean and formal measures.

Measure		Typical disfluency mean	Stutter-like disfluency mean	CELF CL	EOWVT SS	Verbal IQ	Performance IQ
Total disfluency mean	*r*	.952**	.578**	−.240	−.335*	−.311*	−.159
	p	<0.001	<0.001	0.104	0.017	0.028	0.270
Typical disfluency mean	*r*		0.340*	−.183	−.334*	−.260	−.134
	p		0.014	0.219	0.018	0.068	0.355
Stutter-like disfluency mean	*r*			−.206	−.143	−.211	−.117
	p			0.164	0.321	0.142	0.420
Verbal IQ	*r*						.649**
	p						< 0.001

* Significant at $p < .05$.
** Significant at $p < .01$.

4. Discussion

The purpose of this study was to examine fluency in narratives produced by children with localization-related epilepsy, a group previously shown to have depressed language skills (Caplan, 2009; Parkinson, 2002; Strekas et al., in press) in a cohort documented to have poorer language ability than their age- and gender-matched peers. Prior research has shown that children with impaired speech and language abilities may demonstrate increased levels of disfluencies (Boscolo et al., 2002; Guo et al., 2008; Hall, 1996) in their spontaneous narrative speech, which may result from underlying differences in the way these children process and produce language. Differences between children with recent-onset epilepsy and children with chronic epilepsy were also explored, to further assess the impact of epilepsy and duration of seizure activity on language abilities.

4.1. Disfluency means in CWE and TD children

In the present study, CWE, who scored more poorly on a variety of language measures, were found to be significantly more disfluent overall than their matched TD peers during narrative productions. Although no significant differences were seen in the proportion of typical or stutter-like disfluencies between groups, there was a non-significant trend for CWE to produce more stutter-like disfluencies than children with typical development, and a significantly higher proportion of prolongations in the expressive language samples from these children. These observations are consistent with those of Hall (1996), Boscolo et al. (2002), Guo et al. (2008), and Finneran et al. (2009), who found that children with language impairments produced significantly more SLDs than their typically-developing peers. Consistent with Boscolo et al. (2002), part-word repetitions and prolongations were the most frequently observed SLDs in CWE, with a significant difference noted in the frequency of prolongations. No differences were observed for frequency of unfilled pauses between groups. In sum, increased rate of disfluency is a feature of impaired language that may, in the future, add to a better understanding of the underlying areas of encoding difficulty in these children, and add to a fuller description of the markers of language impairment.

Why prolongation frequency is elevated in subgroups of children with relatively weaker language skills is not clear. Prolongations can be seen as a form of “stalling” (Rispoli, Hadley, & Holt, 2008), in which children linger on a sentence constituent or pause while formulating downstream elements for production. McDaniel, McKee, and Garrett (2010) have observed similar patterns across early child language development, likening such behaviors to a frog resting on a lilypad as he crosses the pond (full target utterance).

Childhood disfluency may arise from non-linguistic origins, as well; this study did not analyze the speech-motor abilities of the CWE or their peers. Moreover, disfluency in language impairment may be the result of neither isolated language formulation weaknesses nor speech-motor deficiencies, but rather a complex interaction between the two systems, as has been shown in work with people who stutter by Anne Smith and colleagues (Kleinow & Smith, 2000).

We did discover differences within our population of CWE that merit discussion. When CWE were divided into recent-onset and chronic epilepsy groups, an interesting pattern emerged. No differences were noted between the types of disfluencies in CWE-R and TD-R; however, CWE-C had significantly more total disfluencies, typical disfluencies and stutter-like disfluencies than TD-C. These patterns were consistent with what had been found in analysis of their standardized test results, narratives, and listeners’ judgments of their language abilities, where the most marked differences were found between children with chronic epilepsy and their peers, with less obvious differences seen in children with a more recent diagnosis. Similarly, narratives produced by children with chronic epilepsy contained more disfluencies overall, while no differences in disfluency were observed in narratives produced by children with recent-onset epilepsy. As noted, this pattern is in accordance with differences in language skills, as measured both by standardized testing and narrative analysis. In Strekas et al. (2007), both CWE-R and CWE-C obtained significantly lower *CELF-4* expressive language and *WASI* verbal IQ scores than did TD-R and TD-C. In addition, listeners provided significantly poorer quality scores for narratives produced by CWE-C as compared to TD-C, while no differences in quality scores were seen in CWE-R and TD-R. Findings from these studies demonstrate that performance on standardized and naturalistic measures of language, as well as perceptions of language performance, are impaired in some CWE-C.

Our findings, in addition to results from previous cross-sectional studies, suggest that perhaps the chronic nature of epilepsy and ongoing seizure activity may be characterized by lower language performance over time, which results in increased disfluencies. However, most work that has examined spontaneous language performance has been cross-sectional, rather than longitudinal, making any causative links quite tentative. Moreover, it is important to note that anti-epileptic drugs (AED) are often used to treat children with chronic epilepsy and children taking AEDs are at a greater risk for developing speech and language problems (Svoboda, 2004). The medications themselves have not been extensively evaluated for acoustic/speech impacts; the one report we found on speech effects of an AED in pediatric treatment did not appear to impact speech in any measurable way (Yun, Choi, Eun, Seol, & Kim, 2011). Of more general concern, AEDs, most of which have not been evaluated extensively in the pediatric population, can have documented effects on speed of processing and attention that might be reflected in the fluency of speech production (Lagae, 2006), As a result, it is difficult to parse out the role of ongoing seizure activity vs. the role of AEDs in the language performance of children with chronic epilepsy. Yet, in this study children with both recent-onset and chronic epilepsy were taking AEDs during the period of time in which they completed behavioral and fMRI testing. Since the average number of AEDs did not statistically differ between groups (CWE-C mean = 0.5, CWE-R mean = 0.1, $t = 1.44$, $p = 0.16$), we feel that it is less likely that AEDs were responsible for differences seen in this study.

4.2. Limitations

One limitation of this study is the cross-sectional design. Analyzing language skills at the onset of epilepsy and comparing performance over the course of childhood in a longitudinal study would better inform if seizure disorder (epilepsy) is the source of speech and language impairments. Longitudinal research could more specifically assess whether ongoing seizure activity, anti-epileptic drugs and/or other treatments, or some underlying neuropathy is responsible for differences in performance on measures of cognition and language in children with epilepsy.

Although this group was carefully selected to be more homogenous than other studies, the participants with epilepsy still differed in some ways. First, some children experienced simple seizure activity in addition to complex seizures, while other children only experienced complex seizures. Also, although CWE-C and CWE-R were separated by length of time since diagnosis, there was an overlap between groups in the number of total lifetime seizures. For example, a few children in the recent-onset group actually experienced more seizures than the children in the chronic group. Additionally, a non-linear coding scale was used to record the total lifetime number of seizures, with scores capped at 8 for children who experienced more than 20 seizures. Although originally done to separate the CWE with a poorer prognosis, this scale may have actually obscured the effect of large numbers of seizures over time on speech and language performance. Finally, sample sizes were relatively low compared to prior studies.

4.3. Future directions

The first two authors of this manuscript wish to emphasize that the growing literature on linguistic performance in children with chronic seizure disorder, none of which has to date appeared in the speech-language pathology literature, further motivates the baseline testing of all children recently diagnosed with seizure disorder. This testing should include both standardized testing and naturalistic sampling, consistent with our best professional practices, and probably the only way in which fluency of language production can be measured. Only with such baselining will any “declines” in speech and language skill, including speech fluency, be detected and addressed, if necessary, through therapy to address areas in which children appear to be falling behind their peers developmentally. The recent move to baseline children playing contact sports (Fjordbak, 2011) recognizes the utility of assessing children at risk for language problems; in this case, medical referral should trigger such an evaluation.

This study analyzed disfluency frequency and type, but did not consider the location of disfluency within the utterances. Considering locus of disfluency and type of words (e.g., content vs. function words) on which disfluency occurred may provide additional information about underlying syntactic and semantic processing. Preliminary reviews of narrative transcripts in Boscolo et al. (2002) found that although the frequency of SLDs was low for all participants, in typically-developing children SLDs most frequently occurred on longer, content words while in children with a history of expressive specific language impairment SLDs most frequently occurred on function words and in the initial position of phrases. These findings suggest that while disfluencies from typically-developing children may be related to semantic and lexical retrieval issues, disfluencies in children with language impairments may result from difficulties with syntactic processing. Additionally, if this is true, it may be interesting to compare disfluency measures from children with and without speech and language impairments to fMRI data obtained while these children perform different syntactic or semantic tasks.

For various methodological reasons, disfluencies were tallied discretely, and “nested” or complex “disfluency clusters” were not treated as they have been in some other studies (Logan & LaSalle, 1999); this may have served to minimize the calculated rates of disfluency seen in this study. Future analyses may wish to examine fluency clusters as well as isolated disfluencies.

Our task may have inadvertently disadvantaged the CWE relative to the TD children in terms of likelihood of fluent speech production. Byrd et al. (2012) recently reported that, while children who stutter were more disfluent in narrative than in conversation, the reverse was true for typically-developing children. Other research outlined earlier suggests that children

with language problems also experience fluency problems during narrative generation. Future research should contrast the fluency demands of multiple speaking tasks in order to isolate possible determinants of fluency failure during conversation as opposed to narrative production.

Only children with localization-related epilepsy with a seizure focus in the left-hemisphere were included in this study. Although the left hemisphere is considered the dominant side for most language functions, and speech and language impairments are typically more severe in children who have a left hemisphere seizure focus, it would be interesting to analyze fluency and speech rate in children with a right hemisphere focus, since prosody, an important component of speech rate and fluency, is typically thought to be controlled by the right hemisphere.

4.4. Conclusion

This study suggests that children with seizure disorder who have lower expressive language skills than typically developing age- and gender-matched peers also show a higher level of disfluency, particularly a higher proportion of SLDs, and in particular prolongations. The current study adds to the existing literature suggesting that disfluency during expressive language tasks may be a subtle feature of language impairment. As the field of communication disorders searches for ecologically valid markers of formulation difficulty in children with expressive language impairment, it may be useful to continue and extend studies of the fluency abilities of children whose language skills are impaired relative to their peers. In addition to the total number of disfluencies as a potential marker of formulation difficulty, it may be possible for future investigations to use the loci of such fluency breakdowns to pinpoint specific areas of weakness in expressive language production in children with compromised language skills.

CONTINUING EDUCATION

QUESTIONS

(1) The fluency of children with epilepsy (CWE) is relevant because:
   a. Epilepsy is a very rare neurological condition in school-aged children;
   b. CWE often display severe speech and language deficits;
   c. Atypical fluency profiles in CWE may be a subtle feature of underlying language impairments;
   d. There is a great deal known about fluency in CWE;
   e. CWE tend to have fewer disfluencies than their age- and gender- matched peers.

(2) Previous investigations of the fluency profiles of children with language delay/disorder suggest that:
   a. There is no systematic effect of language delay on the types and frequency of disfluency in children's spontaneous speech;
   b. Children with language delay/disorder tend to have more normal disfluencies than language-matched peers, but do not display a greater number of stutterlike disfluencies (SLDS);
   c. Children with language delay/disorder may have elevated frequency of both normal disfluencies and SLDs;
   d. Children with language delay or disorder are likely to present with co-morbid stuttering;
   e. Children with language delay or disorder tend to have fewer disfluencies, perhaps as a result of slower speech rate.

(3) Which of the following was NOT found in this study?
   a. Children with epilepsy (CWE) display fluency profiles that are not significantly different from their age-matched peers;
   b. Children with epilepsy are more disfluent than their age-matched peers;
   c. Children with chronic epilepsy are more disfluent than children whose epilepsy was more recently diagnosed;
   d. Frequency of disfluencies in spontaneous speech was not significantly correlated with any of the standardized language scores;
   e. Older children were less disfluent than younger children, across both groups.

(4) Children with chronic epilepsy:
   a. Are not at risk for depressed language skills;
   b. Appear to show a lower rate of disfluency than typical peers;
   c. Are often on anti-epileptic drugs (AEDs) that do not affect their speech and language skills;
   d. Display fluency and language profiles that are more impaired, relative to age-matched peers, than children with recent-onset epilepsy;
   e. Generate narratives with the same complexity and structure as age-matched peers;

(5) Which of the following is not likely to account for the elevated rates of disfluency seen in CWE in this study?
   a. The CWE had lower levels of language performance than did their age-matched peer;
   b. Ongoing seizure disorder may compromise language formulation in CWE;
   c. AEDs may compromise language formulation in CWE;
   d. Only children with right hemisphere focal seizures were studied;
   e. Underlying differences in the way CWE process and produce language.

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