ABSTRACT

Title of Thesis:             LISTENER QUALITY JUDGMENTS OF
                            NARRATIVES PRODUCED BY CHILDREN WITH
                            AND WITHOUT LOCALIZATION-RELATED
                            EPILEPSY


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The purpose of this study was to examine if listeners perceive differences
in the narratives of children with and without localization-related epilepsy, and if
these differences are greater in children with a longer history of epilepsy.
Listener ratings were compared cross-sectionally in two sets of comparisons:
children with recent-onset epilepsy (CWE-R) compared to typically-developing
peers (TD-R), and children with chronic epilepsy (CWE-C) compared to another
set of typically-developing peers (TD-C). Listeners assigned significantly lower
overall quality, vocabulary, story structure, and grammar ratings to narratives
produced by CWE-C than to those produced by TD-C, but there were no
significant differences between ratings assigned to narratives produced by CWE-
R and TD-R. These results imply that continued seizure activity, and/or its
management, may impact listener perceptions of expressive language skills in
children.

LISTENER QUALITY JUDGMENTS OF NARRATIVES PRODUCED BY
CHILDREN WITH AND WITHOUT LOCALIZATION-RELATED EPILEPSY


By


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Introduction

*What is Epilepsy?*

Epilepsy is defined as recurrent unprovoked seizures that occur due to abnormal, excessive electrical activity in the brain (Drewel & Caplan, 2007; Goldman & Golby, 2005). There are several different types of epilepsy. In symptomatic epilepsy, the etiology is known (e.g., a traumatic brain injury). On the other hand, in idiopathic epilepsy, the etiology of seizures is not known. Epilepsy can also be characterized by the way in which seizures affect consciousness. In complex epilepsy, the patient loses consciousness during seizures, whereas in simple or primary epilepsy, the patient experiences only a *change* in consciousness, such as cessation of ongoing activity and eye gaze deviations (Caplan et al., 2002). Other subcategories of epilepsy reflect localized versus diffuse origin of onset of seizures. In localization-related epilepsy (also known as partial or focal epilepsy), seizure onset is restricted to a specific, constant region of the brain. On the other hand, in generalized epilepsy, there is a diffuse onset of seizure activity, which cannot be reliably referred back to a single place of origin. Finally, some individuals experience febrile seizures, which are brought on by extremely high fevers.

Convulsive disorders such as epilepsy are amongst the most common neurologic disorders in children (Hauser, 1995). Epilepsy affects approximately 1% of the population (Hauser, 1990) and 20% of cases begin before age five (Epilepsy Foundation, 2007). Children with epilepsy are thought to be at risk for

the development of speech and language deficits (Parkinson, 2002), and yet, many such cases are frequently overlooked (Svoboda, 2004).

*Impact of Epilepsy on School Performance and Peer Relations*

Several studies have confirmed that children with epilepsy (CWE) score lower than typically-developing peers on standardized measures of academic achievement. Additionally, CWE score lower on self-report and parent/teacher-report measures of academic success (see review in Drewel & Caplan, 2007), and the average school grades of adolescents with epilepsy may be significantly lower than those of healthy adolescents (Adewuya, Oseni & Okeniyi, 2006). Specifically, children with Temporal Lobe Epilepsy (TLE; characterized by seizures that initiate in the temporal lobe) have been observed to score more poorly on standardized measures of reading speed and comprehension (Chaix et al., 2006).

It is clear that epilepsy impacts school performance, but how? One could argue that a child living with any chronic health condition is probably at risk for academic difficulties because of illness, absences from school, and so forth. However, some studies have shown that children with epilepsy experience more psychosocial and educational problems than children with other health conditions, such as asthma or juvenile rheumatoid arthritis (Drewel & Caplan, 2007; Wodrich, Kaplan & Deering, 2006).

Caplan et al. (2006) proposed that academic difficulties in CWE are moderated by thought disorder, a psychiatric term that describes difficulty with organizing ideas. In language use, this condition may lead to difficulty in

repairing communication breakdown, maintaining a conversational topic during interactions with others, and using cohesive devices to link ideas across continuous discourse. Thus, the language of an individual with thought disorder may be characterized by instances of illogical thinking (inadequate, inappropriate or contradictory reasoning), loose and tangential associations (often signaled by unpredicted topic change), and may lack cohesion (due to lack of sentence ties such as word repetition or synonym use to link repeated references over stretches of discourse or text). Caplan and colleagues observed a relationship between diagnosis of thought disorder and both lowered academic achievement and parental reports of school difficulties. It is plausible that CWE who have difficulties organizing and expressing their thoughts struggle to communicate their academic knowledge in the classroom.

CWE may also experience social difficulties or problems with peer relations in school. Schoenfeld et al. (1999) found that children with complex partial seizure disorder (CPS) presented with lower scores on measures of social and school competence, as measured by the *Child Behavior Checklist* (Achenbach & Edelbrock, 1991) and the *Wide Range Achievement Test-3* (Jastak & Wilkinson, 1993). Similarly, in a literature review of sixteen studies, Drewel and Caplan (2007) noted numerous reports that CWE (of various types) demonstrate lower social competence and more peer difficulties than typically-developing children or children with non-central nervous system health problems (e.g., diabetes). These researchers' findings also indicated that social difficulties in CWE have been related, in a number of studies, to the cognitive, psychological

and linguistic deficits seen in CWE (e.g., lower IQ, externalizing and anxious behaviors and social communication deficits).

Thus, academic difficulties and social problems in CWE have been linked to difficulties with both neuropsychological functioning and expressive language. But what are the specific effects of epilepsy on cognitive and psychological processes?

*Cognition and Psychopathology*

As noted, children with complex partial seizure disorder (CPS) are at risk for the development of psychopathology and cognitive deficits. Caplan et al. (2004) found that when compared to unaffected children, children with CPS scored significantly lower on the *Wechsler Intelligence Scale for Children-Revised (WISC-R)* on measures of verbal, performance and full scale IQ, even when demographic (e.g., socio-economic status) and perinatal (e.g., delivery complications) variables were controlled for.

Children with epilepsy (CWE) may also present with more attention and internalizing problems (e.g., withdrawal, anxiety, depression, and thought problems) than do typically-developing peers (Austin & Caplan, 2007). In addition, CWE present more frequently with psychiatric problems, such as depression, anxiety disorders and psychotic symptoms (e.g., confusion or delusions, Bortz, 2003).

When compared to their typically-developing peers, CWE present with more difficulties in the areas of memory, attention, and executive functioning (Drewel & Caplan, 2007). These are all areas that are important for academic

performance, conversational discourse and narrative production. CWE also perform significantly worse on measures of learning, memory and sustained attention (Oostrom, van Teeseling, Smeets-Schouten, Peters, & Jennekens-Schinkel, 2005).

Cognitive impairment can lead to higher-level linguistic deficits because problems with memory, attention and executive function make it difficult to process and respond to language (Drewel & Caplan, 2007). In addition, CWE struggle to integrate language and cognition; as noted earlier, the language they use to express their thoughts is often disorganized and illogical (Caplan, 2002).

*Impact of Epilepsy on Language*

As noted, children with CPS present with significantly lower verbal IQs (*WISC-R)* than do typically-developing peers, even when demographic and perinatal variables, such as ethnicity, SES, and pregnancy and delivery complications are controlled for (Caplan et al., 2004). Previous research findings also indicate that children with epilepsy present with diminished language skills when compared to typically-developing children (Parkinson, 2002; Hermann, et al., 2001), either as a result of brain abnormalities that caused epilepsy, seizure activity or disease management (e.g., anti-epileptic medications). However, the relatively few studies of language profiles in children with epilepsy are complicated by enrolling children with a wide variety of types of epilepsy, as well as concomitant developmental problems. For instance, some syndromes characterized by intellectual impairment also demonstrate seizure disorder. Understanding of possible language deficits in epilepsy is also complicated by the

diverse ways in which language outcomes have been measured. Some representative findings include the observation that children with CPS score lower on measures of expressive vocabulary (as measured by the *Kaufman Brief Intelligence Test* and the *Controlled Oral Word Association Test*), category fluency and receptive language (as measured by the *Children's Token Test*) than do their unaffected siblings (Schoenfeld et al., 1999).

Cohen and Le Normand (1988) conducted a longitudinal study with six children having simple partial epilepsy (SPE). Although the children with SPE initially presented with poorer receptive and expressive language skills than unaffected peers, by age seven, the receptive language abilities of children with SPE were equal to those of the comparison group. On the other hand, the expressive language abilities of children with SPE at age seven remained poor. Receptive language skills were measured using pointing tasks (vocabulary comprehension), narrative comprehension, and acting out commands (to test comprehension of prepositions). Expressive language measures included: repetition of thirty-three simple bisyllabic words, Mean Length of Utterance (MLU) and a count of the number of different word types (NDW) in a language sample that was elicited during a play session during which the child participant was asked to verbalize manipulations and actions with dolls in a dollhouse.

In a later study, Dubé, LeNormand and Cohen (2001) examined the diversity and frequency of main verbs, auxiliaries, copulas and nonfinite verbs (infinitive, past participle and modals) in the language samples of three French-speaking children with SPE. They noted a more limited use of auxiliary verbs in

the language samples of children with SPE as compared to a comparison group. Researchers concluded that this deficit reflects a possible global impairment in the use of function words, which help to express grammatical relationships with other words as opposed to serving their own lexical function. The transcripts of language produced by children with SPE mostly contained ideas about the "here and now." It seems that their difficulty with auxiliary verbs (e.g., *have* and *be* forms of the perfect tense) constrained the level of complexity of their utterances.

In investigating the impact of thought disorder on expressive language in children with differing types of seizure disorder, Caplan et al. (2001, 2002, 2006) tallied instances of illogical thinking, tangents, and loose associations in narratives produced by children with CPS and Primary Generalized Epilepsy (PGE; in whom seizure onset is diffuse), using the *Kiddie Formal Thought Disorder* (K-FTDS; Caplan et al., 1989) *Rating Scale*. Language samples were collected with the Story Game from the *K-FTDS*; Caplan, Guthrie, Fish, Tanguay, & David-Lando, 1989). The Story Game involves listening to a story about a ghost and a story about a boy. After listening, the child re-tells the story and is then asked a series of *wh*-questions to measure comprehension. Then, the child makes up their own story about either a good child, a bad child, the Incredible Hulk, or a witch. Caplan et al.(2002) also examined use of cohesive devices based on Halliday and Hasan's (1976) cohesion taxonomy. Results indicated that, when compared to healthy peers, children with CPS used significantly more illogical thinking (e.g., saying, *I forgot my book because the sky is blue*) and exphora (when the speaker interrupts the flow of conversation to refer to

something else in the immediate environment), as well as fewer conjunctions and referential cohesion (e.g., pronouns or definite articles that refer to objects or people in previous parts of the conversation or story). On the other hand, children with PGE only used significantly fewer conjunctions when contrasted with their peers. Researchers concluded that children with CPS present with formal thought disorder characteristics that lead to narrative cohesion deficits.

Children with CPS, especially those whose seizures are focused in the temporal lobe, also have been observed to use significantly more self-corrections of reference and syntax than do children with primary generalized epilepsy and typically-developing comparison peers (Caplan et al., 2001). Children with CPS and PGE who also have frontal or frontotemporal involvement according to electroencephalogram (EEG) findings were also noted to use significantly fewer fillers than unaffected peers. From these findings, researchers concluded that the temporal lobe controls repair of linguistic functions and the frontal lobe controls online processing and planning of language. Therefore, both CPS and PGE are associated with cognitive deficits, such as difficulties with monitoring (involved in repair) and executive functioning (involved in planning) that impact children's communication abilities.

It is generally accepted that a subset of individuals with epilepsy present with cognitive and linguistic impairments. However, what is less clear is whether these impairments reflect some underlying pre-existing neuropathology present during onset of epileptic seizures, or signal continued cognitive decline over time. In adults, there is evidence to support the notion of disease chronicity as a

predictor of cognitive status (Seidenberg, Pulsipher, & Hermann, 2007), which suggests the negative consequences of ongoing seizure activity.  However, there is very little research on children with epilepsy that answers this question. On the other hand, research involving children has confirmed that early age of onset of epilepsy is associated with more impaired cognitive abilities (Caplan, et al., 2002) and poorer language performance (Hermann, Bell, Seidenberg & Woodard, 2001). This suggests that ongoing seizure experience may diminish language skills over time in childhood

*Effects of Age of Onset*

Childhood onset of epilepsy is associated with greater deficits in language and neuropsychological function than is adult-onset epilepsy (Hermann et al., 2002). Similarly, age of onset of seizures is a strong predictor of cognitive functioning; earlier age of onset is associated with poorer cognitive functioning in verbal and non-verbal memory, problem solving, and mental efficiency (Schoenfeld et al., 2007). Specifically, an earlier age of onset of focal epilepsy is related to poorer language performance across the age span (see review in Hermann et al., 2001).

Caplan et al. (2006) found that children with CPS with high externalizing and overall thought disorder propensity scores had earlier onset of seizure activity and a higher rate of prolonged seizures. Externalizing behaviors, such as aggressiveness and hyperactivity, were measured using the *Child Behavior Checklist (CBCL)*.  Overall thought propensity scores were calculated using the *Kiddie Formal Though Disorder Rating Scale (K-FTDS)*.  Older children with

CPS and poor seizure control (i.e., presence of seizures three to six months before the child's participation in study) had increased severity of thought disorder, suggesting that the seizures themselves lead to this problem (Caplan et al., 2004). In addition, seizure frequency was associated with lower spoken language quotient (SLQ) scores from the *Test of Language Development* (*TOLD*; Caplan et al., 2004).

Age of onset and duration of illness are also related to lower grades in school (Adewuya et al., 2006) and school competence as measured by the *Wide Range Achievement Test-3* (Schoenfeld, et al., 1999), which can presumably, in turn, affect standardized language test achievement profiles. This implies that seizure activity impacts cognitive language and language abilities, which in turn leads to communication difficulties at school.

Associations between early onset of epilepsy, poor seizure control, and a longer history of epilepsy with cognitive deficits, poorer language performance and lower academic achievement suggest that seizure activity increases the likelihood of eventual demonstration of such problems in CWE. Following temporal lobectomy, with subsequent control of seizures, improvement in language skills has been noted in adults, again suggesting that seizures themselves negatively impact brain function (Hermann & Wyler, 1988). However, some argue that a primary source of children's deficits is the underlying brain pathology that causes seizure activity.

*Underlying Neuropathy*

Neurobehavioral problems may not necessarily be caused by seizures or their treatment. Byars et al. (2007) discovered structural brain abnormalities during imaging in CWE shortly after diagnosis that potentially exert some influence on behavioral function independent of seizures or their treatment. Atypical language representation (i.e., abnormal activation in the brain for language tasks), as measured by fMRI scans during language processing tasks, is also associated with early onset of localization-related (i.e. partial or focal) epileptic seizures. Berl et al. (2007) proposed that localization-related epilepsy (LRE) "exerts global brain effects on functional organization that may be driven by pathological processes and furthered by adaptive changes" (Berl et al., 2007, p.1610).

Oostrom, van Teeseling, Smeets-Schouten, Peters and Jennekens-Schinkel (2005) found that children with recently-diagnosed idiopathic epilepsy performed significantly worse on measures of learning than gender-matched typically-developing classmates, suggestive of pre-existing processing deficits. Learning was measured by a computerized non-verbal task in which children were required to recall the location of visually presented objects in a matrix, word span backwards, and other tasks requiring sustained attention.  Over time (at diagnosis, and then 3, 12, and about 42 months later), children did not show changes in cognitive and behavioral status. Thus, it is not clear if language and cognitive deficits found in children with epilepsy occur as a result of the brain pathology

that leads seizure activity, are induced by the seizures themselves, or are simply a side effect of the medication used to treat epilepsy.

*Effects of Anti-Epileptic Drugs*

Research has shown that most of the new medications available to treat epilepsy do not produce long-term cognitive side effects, although a few immediate side effects have been noted (Goldstein et al., 2004). However, some AEDs produce side effects including problems with attention or vigilance and when two or more AEDs are used simultaneously; they can produce side effects such as cognitive slowing (Meador, 2002). This finding has been confirmed by several other studies. For instance, children with complex partial epilepsy prescribed an AED polytherapy received higher thought disorder scores than children prescribed an AED monotherapy (Caplan et al., 2006). Caplan et al. (2004) found that a higher number of prescribed AEDs was associated with lower spoken language quotient (SLQ) scores from the *Test of Language Development* (*TOLD*).

*Limitations of Prior Studies*

Much of the research on language profiles in childhood epilepsy is characterized by a small sample size and a heterogeneous participant pool, in terms of epilepsy-type (see Chaix et al., 2006; Cohen & Le Normand, 1988) and focus of seizure activity (see Caplan et al., 2001, 2002, 2004, 2006). In addition, the majority of this research does not utilize matched pairs of children to compare

performance on tasks, but instead, matches by mean age of group. This is likely to be inappropriate due to the rapidity of language growth over childhood.

Many studies also utilized very gross measures of language skill, such as verbal IQ tests (e.g., *Wechsler Intelligence Scale for Children-Revised*). As a body of research, most studies examining language use and skill in CWE have also employed highly diverse measures of language, which makes comparison across studies and appreciation of generalized language skills in this population difficult. Another difficulty may arise when researchers examine a skill in ways that do not permit reference to expected normative values. For example, some studies have used language sample analysis, but used qualitative measures such as total counts of verbs, rather than proportions comparable to normative values (see Cohen & Le Normand, 1988). The confound of quantitative measures (as compared to proportions) is that length of language sample will affect these results (i.e., more instances of various indices occur in longer language samples).

Some studies with large sample sizes contain several problems with statistics that may over-identify language problems in CWE , including the use of numerous ANOVAs without correcting for multiple comparisons (see Caplan et al., 2001, 2002, 2004, 2006).  Conversely, studies with extremely small sample sizes have been limited to descriptive (instead of statistical) analyses (see Cohen & LeNormand, 1998; Dubé, LeNormand, & Cohen, 2001).

*Preliminary Studies of Children with Chronic and Recent-onset LRE: Pilot Data*

*from the POLER Initiative*

Given the limited and complex literature on language profiles of children

with epilepsy, the Plasticity of Language in Epilepsy Research (POLER) initiative

(Gaillard et al., 2007) was designed to narrow investigation of the effects of

localization-related epilepsy on children's language performance and functional

processing of language as measured by functional Magnetic Resonance Imaging

(fMRI). In particular, this project sought to investigate how history and duration

of seizure activity, *per se*, influences behavioral performance and brain substrates

of language processing. To this end, only children with Localization-Related

Epilpespy, as opposed to a broader set of children carrying a diagnosis of

epilepsy, with no evident structural atypicalities in brain structure (absence of

tumors, surgical interventions, etc.) were recruited. All children in this study

experienced complex seizures; however, some also experienced simple seizures.

In Strekas et al. (2007), which used data collected during the POLER

project, researchers specifically examined the effects of epilepsy on children's

narrative skills.  Experimenters transcribed and coded 25 narratives of children

with LRE, whose seizures initiated in the left hemisphere, 10 of which were

recent-onset (CWE-R) patients (< 1 year), and 15 of which were chronic (CWE-

C) patients (> 3 years).  Researchers also transcribed and coded 25 narratives

produced by age- (within 3 months) and gender-matched typically-developing

(TD) peers.  The narratives had been elicited using the wordless picture book,

*Frog, Where Are You?* (Mayer, 1969).  The narratives were formally analyzed for

the following: number of communication units (C-Units, independent clauses plus modifiers, Loban 1976), mean length of turn (MLT), vocabulary diversity (using VOC-D, a type of type-token ratio that statistically controls for the length of the narrative), syntactic complexity (proportion of C-units containing a subordinate clause), and discourse cohesion (proportion of C-units containing cohesive elements, such as conjunctions). The total number of narrative components, taken from Trabasso and Rodkin's (1994) taxonomy, was also computed, using the categories of setting, initiating events, higher-order goals, attempts (to locate frog), and outcome. Finally, the children were administered speech, language, developmental, and psycho-educational assessments, including the *Wechsler Abbreviated Scales of Intelligence* (*WASI,* Psychological Corp, 1999) or the *Differential Ability Scales* (*DAS,* Elliott, 1990) for children less than 6 years of age. The children were also administered the *Clinical Evaluation of Language Fundamentals, Fourth Edition* (*CELF-4,* Semel, Wiig & Secord, 2003) or the *Clinical Evaluation of Language Fundamentals, Preschool Edition* (*CELF-P*; Wiig, Secord & Semel, 2004) for children less than 5 years of age.

The data were compared cross-sectionally in two sets of comparisons: CWE-R compared to a set of age- and gender-matched typically-developing peers (TD-R), and children with CWE-C compared to another set of age- and gender-matched typically-developing peers (TD-C). Significant differences were seen between groups in both sets of comparisons made for full-scale IQ and *CELF* scores in the expressive language subscale. However, mean differences between CWE-C and TD-C were descriptively larger than between CWE-R and TD-R.

Significant differences were seen between CWE-C and TD-C (but not CWE-R and TD-R) in verbal IQ, MLT, and total number of narrative components.

In Strekas et al. (2007), CWE scored significantly lower than did typically-developing peers on narrative structure and standardized language assessments, but, on average, scores fell within one standard deviation of the normative mean (Strekas et al., 2007). Thus, seizure activity may negatively impact language scores, but not to such an extent that children obviously qualify for speech and language services in public schools, according to most current guidelines, particularly if no baseline measures of performance prior to epilepsy diagnosis are available.

These preliminary results suggest that language deficits in CWE can be subtle and difficult to detect, if dependent upon standardized testing in the schools. Perhaps other measures, such as narrative skills, can be used to examine language deficits associated with epilepsy. In turn, research results in this area can be used to develop screening tools and baseline measures for CWE. However, narrative analysis can be time-consuming, and differences observed between the study groups in pilot analyses were not numerous. Because there are a number of different, competing ways of performing a structural analysis of a child's narrative abilities, further exploration of this skill area may be informative in understanding language skills in children with epilepsy.

*Narrative Analysis*

In the present study, we examined listeners' perception of language contained in narrative language samples instead of children's performance on

standardized tests. Narrative production is a complex task that requires the integration of linguistic, cognitive and socio-behavioral skills not addressed in more formal language tests (Norbury & Bishop, 2003). A good narrator uses lexical skills to encode information about the characters and story events; uses cognitive skills to infer characters' motivation for actions and the relationship between the theme of the story and these actions; and uses social skills to relate to the audience while telling a story (Reilly, Losh, Bellugi & Wulfeck, 2004).

Story-telling is a common aspect of all cultures; one that is pervasive and accessible even to young children. Because of their "…'everyday' nature, narratives provide an excellent quasi-naturalistic measure of children's spontaneous language . . ." (Reilly, Losh, Bellugi & Wulfeck, 2004, p. 230). In this sense, narrative performance is a more ecologically valid window into functional language use that may not be tapped through administration of standardized language tests.

Previous research findings suggest that children with focal epilepsy produce narratives that do not differ from typically developing peers in terms of narrative length, diversity of vocabulary used, syntactic complexity or discourse cohesion. However, these narratives do differ in terms of mean length of turn and inclusion of narrative elements (based on Trabasso and Rodkin's 1994 taxonomy), such as setting and initiating events (Strekas et al., 2007). These findings suggest that epilepsy may impact the more functional aspects of language production: structuring a story outline and sentence-length. Results from Strekas et al. (2007) were obtained through formal analyses of narrative sample

transcriptions by trained scorers, but spoken language is not normally transcribed and analyzed. How would these narratives *sound* to the average listener? Would he or she perceive such qualitative differences?

As noted earlier, research findings indicate that children with complex partial seizures (which is a diagnosis similar to the clinical population examined in the present study) produce language that contains illogical thinking and does not effectively link ideas across sentences (Caplan et al., 2006). Therefore, it is reasonable to hypothesize that listeners may find it difficult to comprehend what children with epilepsy are trying to say. This type of communication breakdown could be one source of academic and social difficulties in school for these children. In the current study, we wanted to examine this phenomenon by asking listeners to rate the quality of language produced by this population to see if they could perceive a discernable difference.

*Listener Judgments of Narratives*

There are a number of structural taxonomies that can be used to score children's narratives, including Roth and Spekman (1986), Trabasso and Rodkin (1994), and so forth. However, not all aspects of narrative production are measurable with objective analyses. Clarity, charm, and creativity are difficult to examine quantitatively and are often overlooked when using traditional objective measures (McFadden & Gillam, 1996; Peterson & McCabe, 1983). In order to address this problem), Newman and McGregor (2006) asked listeners to evaluate the quality of narratives produced by children having other diagnosed conditions. Listeners rated narratives from children with and without Specific Language

Impairment (SLI), giving them a score ranging from 1 to 7. The children's narratives were guided by the wordless picture book, *Frog, Where Are You?* (Mayer, 1969). After providing their initial quality rating, listeners completed a questionnaire rating the extent to which various factors (e.g., story grammar, syntax, fluency/articulation, "sparkle") influenced their rating of the narratives. The authors' goal was to examine the functional impact of SLI. They wanted to determine if subjective ratings of narrative quality could differentiate the children with SLI from their typically-developing (TD) peers. They also wanted to determine if teachers and laypersons differed in their quality ratings.

Results indicated that the subjective ratings differentiated the SLI and TD children with 70% non-overlap in scores. Although the lay listeners' and teachers' numeric quality ratings did not differ, teachers reported that they paid more attention to vocabulary and grammar, while laypersons reported that they paid more attention to sparkle. Despite this difference in focus, it was clear that both groups appreciated the impact of language disorder on successful story-telling: " . . . the manifestations of the disorder are noticeable even to laypersons and thereby may limit the successful functioning of the affected child" (p. 1032).

The same may be true in children with epilepsy. As noted earlier, children with epilepsy are at risk for poor peer relations (Drewel & Caplan, 2007) and children with CPS who present with more disorganized language (i.e., thought disorder) have greater difficulty with interpersonal skills and friendships (Caplan et al., 2006). Perhaps the source of this difficulty is the fact that conversational partners of CWE find their language to be confusing. If lay listeners are able to

appreciate a difference in language produced by CWE versus typically-developing children, then conversational partners of CWE probably perceive deficits in their language use.

Additionally, the listening task is useful in terms of comparing the effects of recent-onset and chronic epilepsy. In the current design, all listeners evaluated the 2 sets of matched groups, which otherwise can't be all compared to one another, because the mean age of participants in the chronic comparison (116 months) is slightly older than that of the recent-onset comparison (92 months).

*Summary*

Previous research findings indicate that epilepsy is related to poor school performance (Drewel & Caplan, 2007), a range of cognitive deficits (Oostrom et al., 2005), psychopathology (Austin & Caplan, 2007) and depressed language scores (Caplan et al., 2004). Previous studies also report that CWE whose language is more disorganized struggle with academic success and peer relations (Caplan et al, 2006).

In the present study, we wished to examine listener perceptions of possible language differences present in CWE that may be the source of difficulties in school. Narrative abilities are related to academic performance and social functions in that they tap into the de-contextualized language skills needed to successfully interact with teachers and peers (Price, Roberts & Jackson, 2006). Furthermore, listener judgments of narrative productions capture the functional impact of discourse limitations (Newman & McGregor, 2006).

However, a basic question remains: will listener perceptions of language produced by children with new-onset epilepsy differ from those of language produced by children with chronic seizure activity? Such a difference would strengthen the argument that continued seizure activity can diminish language skills over time. If language deficits and epilepsy both reflect a common underlying neuropathy, there should be minimal difference in language performance and listener perceptions of language skill between children with newly-diagnosed seizure disorder and those with a chronic history of epilepsy.

*Hypotheses*

In this study, we hypothesized that CWE would produce stories that obtained significantly lower listener ratings than those produced by TD children. In addition, we hypothesized that CWE-C would produce stories that obtained the lowest ratings, either as a result of chronic seizures, their management or both. Thus, we sought to compare the performance of children with epilepsy who were divided into two, non-overlapping groups: children with recent-onset ( < 1 year) epilepsy (CWE-R) and children with chronic ( > 3 years) epilepsy (CWE-C).

<div align="center">Method</div>

*Participants*

*Narrative Sample Database.* Researchers at Children's National Medical Center (CNMC) compiled the narratives used in this study. Participant children were part of a larger National Institutes of Health (NIH)-funded study (POLER:

Plasticity of Language in Epilepsy Research, PI: William Davis Gaillard NINDS R01 NS44280).  In addition to producing narrative samples, the child participants in this larger study received speech, language, and psycho-educational testing, as well as functional Magnetic Resonance Imaging (fMRI) scans.  fMRI scan data were not used in the present study.

CNMC researchers elicited stories using the wordless picture book, *Frog, Where Are You?* by Mercer Mayer (1969). These 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. The audio-recordings were labeled using numerical codes, and the participants' identities were concealed to UMCP researchers. Only age, gender and patient group were revealed.

Child participants were divided into four groups.  Group 1 contained 10 children with recent-onset ( < 1 year following second seizure) epilepsy (CWE-R).  The mean full-scale IQ score for this group was 100.7 and verbal IQ score was 102.7 (see Table 1).  The *Wechsler Abbreviated Scales of Intelligence* (*WASI*, Psychological Corp, 1999) or the *Differential Ability Scales* (*DAS,* Elliott, 1990) for children less than 6 years of age were used to obtain IQ scores. Group 2 contained 10 typically-developing peers, who were age- and gender-matched to the children with recent-onset epilepsy (TD-R). The mean full-scale IQ score for this group was 112.7 and verbal IQ score was 110.4. Groups 1 and 2 each

contained 4 females and 6 males; the mean age of the children in these groups was 92 months (range 50-139 months).

Group 3 contained 15 children with chronic ( > 3 years) epilepsy (CWE-C), and Group 4 contained 15 age- and gender-matched typically-developing peers (TD-C). In group 3, the mean full-scale IQ was 96.6 and verbal IQ was 98.4. In group 4, the mean full-scale IQ was 117.3 and the mean verbal IQ was 119.1. Groups 3 and 4 each contained 7 females and 8 males, with a mean age of 116 months (range 75-155 months).

The average age at which seizure onset occurred in group 1 (CWE-R) was 74 months and in group 3 (CWE-C) was 55 months. According to a truncated coding scale, the mean total number of lifetime seizures in group 1 was 4.5 seizures and in group 3 was 7.1 seizures.

The child participants in groups 1 and 3 had electroencephalogram (EEG) or other clinical evidence that suggested a left hemisphere focus of seizure activity. All child participants were right-handed. See Table 1 for demographic profiles.

Table 1

*Demographic Information – Child Participants*

| Groups | 1 | 2 | 3 | 4 |
| --- | --- | --- | --- | --- |
| | CWE-R | TD-R | CWE-C | TD-C |
| Number of participants | 10 | 10 | 15 | 15 |
| Age (months) | 116 | 116 | 92 | 92 |
| Full-Scale IQ | 100.7 | 112.7 | 96.6 | 117.3 |
| Verbal IQ | 102.7 | 110.4 | 98.4 | 119.1 |
| Age (months) at 1st seizure | 74 | | 55 | |
| *Total lifetime seizures | 4.5 | | 7.1 | |
| Poytherapy (number of participants) | 1 | | 4 | |

*2=two seizures, 3=3 seizures, 4=4 seizures, 5=5 seizures, 6=6-10 seizures, 7=11-20 seizures, 8= > 20 seizures

Experimenters from the POLER project collected the children's narrative language samples used in the present study. At Children's National Medical Center, each child 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 and language-impaired children from numerous linguistic communities (Berman & Slobin, 1994) for the CHILDES archive database (MacWhinney, 2000).

*Listener Participants.* Participants included 45 undergraduate students, ages 18 to 22, from University of Maryland at College Park and George Washington University, Washington, DC. Exclusionary criteria included hearing loss and English as a second language, according to self-report. This group of participants included 37 women and 8 men.

Participants were recruited through flyers, class announcements, e-mail list servers, and word of mouth. By participating in this research, individuals enrolled in eligible classes received extra credit points or the opportunity to satisfy a course requirement. All other participants received a $10 Pizza Hut gift card.

*Materials*

*Rating Scale.* Our survey ratings addressed seven different aspects of the narratives: overall quality, vocabulary usage, story structure, grammatical complexity, speech fluency, color/interest and prosody (see Appendix A). The purpose of the survey was to determine if listeners subjectively perceived differences in the narrative abilities of the children across groups. 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. Story structure ratings were based on inclusion of the critical parts of the story, and on whether the child followed the theme of the story. Grammar ratings pertained to the length and complexity of sentences and fluency ratings to the smoothness of speech. Finally, participants were instructed to base color/interest judgments on inclusion of emotion and humor and prosody

judgments on whether the narrative sounded monotonous or expressive. Each quality rating was made using a 7-point interval scale (IS), a valid and reliable method commonly used in psychological and language scaling (Stevens, 1975). It is a time-efficient procedure that can be readily applied by individuals who do not have prior knowledge about language sample analyses (Newman & McGregor, 2006).

*Procedure*

   *Rating Task.* Adult listener participants were tested individually or in groups of up to three. First, they examined a copy of the plates used to elicit the narratives (taken from Mercer Mayer's wordless picture book, *Frog, Where Are You?*) and the survey packet used to rate the narratives. Next, they examined the front of the survey packet (see Appendix A) contained procedural directions, questions to ensure that the participants met the criteria for the study (e.g., native speaker of English with no history of hearing loss), and instructions to disregard articulation errors and recording quality. The front of the survey packet was read aloud to participants by the experimenter. The experimenter also reviewed the seven criteria for rating the narratives and answered participants' questions.

   Participants listened to 10 different narratives, delivered through an iSymphony donut speaker for iPods. The sound files were saved on an iPod mini, and the narratives were played at a comfortable volume that was loud enough for all participants to indicate that they could hear the samples clearly. Each audio-recording was identified using a numerical code, and participants were unaware of the group designation for any child narrator. After listening to each story,

participants were provided with as much time as needed to complete one of the ten surveys contained in each packet.

Fifty child narratives were used in the present study, and 45 adult listeners, in total, participated in the experiment. There were 15 experimental sessions, during which groups of up to 3 participants heard and rated 10 narratives. In this way, 9 different participants listened to and rated each narrative. The presentation order of the narratives was counterbalanced (see Table 2). In addition, each playlist of 10 narratives was designed to assure that all children on a given playlist were of roughly similar ages (e.g., within a two-year age span) to prevent younger children's narratives from being compared unfavorably to those generated by the older children.

Table 2

*Counterbalancing of Presentation Order*

| | Listener Group 1 | Listener Group 2 | Listener Group 3 |
|---|---|---|---|
| Presentation order | 1 | 2 | 9 |
| | 2 | 4 | 5 |
| | 3 | 6 | 1 |
| | 4 | 8 | 8 |
| | 5 | 10 | 4 |
| | 6 | 1 | 7 |
| | 7 | 3 | 3 |
| | 8 | 5 | 10 |
| | 9 | 7 | 6 |
| | 10 | 9 | 2 |

*Analysis*

Nine different listeners provided seven ratings for each narrative (total of 63 scores). The nine ratings for each narrative in each category were averaged and extreme listener scores for any variable that fell two standard deviations below or above the mean of the group average were discarded. A total of 45 ratings (1.4 % of the data) were removed from the analyses and were fairly evenly spread across the child speaker groups. Rating scores were then re-averaged.

Averaged survey 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 areas (overall quality, vocabulary usage, story structure, grammatical complexity, speech fluency, color/interest, and prosody) and were compared cross-sectionally in two sets of comparisons: children with recent-onset (< 1 year) epilepsy (CWE-R) compared to a set of age- and gender-matched typically-developing peers (TD-R), and children with chronic ( > 3 years) epilepsy (CWE-C) compared to another set of age- and gender-matched typically-developing peers (TD-C). Significance level was set to .008 due to multiple comparisons on the relatively large (n = 6) set of measures (Bonferroni correction; Abdi, 2007).

We then conducted a series of *post hoc* analyses. In order to determine which aspect of the CWE narratives influenced listeners' overall quality score, a Pearson's product-moment (*r*) correlation was computed between listener overall

quality scores and subscores (i.e., ratings for vocabulary, story structure, grammar, fluency, color/interest and prosody) for CWE.  We also examined the relationship between listener ratings of CWE language samples and data collected through formal analyses of CWE language samples (e.g., Mean Length of Turn). For this comparison, we used data from POLER project pilot studies (see Strekas et al., 2007).  A Pearson's product-moment correlation matrix was computed to compare listener overall quality scores to Vocabulary Diversity (VOC-D) and total number of narrative components (setting, initiating events, higher-order goals, attempts to locate frog, and outcome (Trabasso & Rodkin, 1994).

In addition, we wanted to examine the relationship between listeners' ratings of language samples and standardized test scores.  For this analysis, overall quality scores were correlated with Core Language Scores from the *CELF-4* or *CELF-P* (for children less than 5 years of age) and verbal IQ scores from the *WASI or DAS (*for children less than 6 years of age).

Finally, in the children with LRE, we examined if seizure history was predictive of listener ratings.  We correlated the age at which each child's first seizure occurred and each child's lifetime number of seizures with listener ratings.

Results

*Listener Perceptions by Group: Children with Epilepsy vs. Typically-Developing Peers*

The primary goal of this study was to determine if listeners perceived differences in narratives produced by children with and without epilepsy.  We

hypothesized that stories produced by children with epilepsy (CWE) would obtain

lower listener ratings than those produced by age- and gender-matched typically-

developing peers (TD).

Analyses (Mann-Whitney $U$ converted to Wilcoxon $Z$) revealed that in

terms of their overall quality, listeners scored CWE narratives (mean score =

3.92) significantly lower than TD narratives (mean score = 4.7, $z$ = -2.10, $p$ =

.0360, see Figure 1, Table 3). Using the Bonferroni correction for multiple

comparisons, scores for use of grammar were significantly depressed (mean =

4.33)  for the children with epilepsy (mean = 3.46, $z$ = -2.66, $p$ = .0078) at a

criterion of .008. We note that without Bonferrnoi correction, vocabulary ($z$ = -

2.42, $p$ = .0156), story structure ($z$ = -1.76, $p$ = .0789), and fluency ($z$ = -1.98, $p$ =

.0476) ratings would have been considered significantly depressed as well.

*Figure 1.* Mean scores and standard deviations of CWE and TD listener ratings
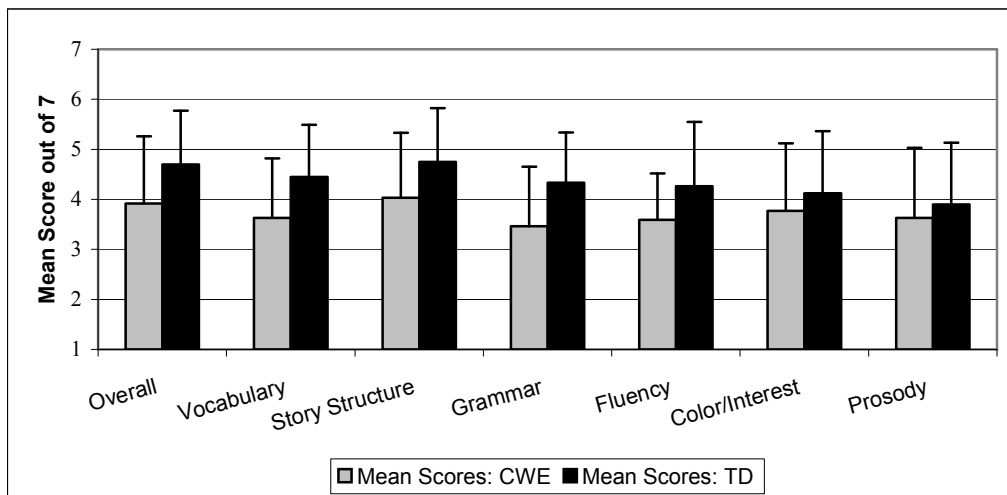
Table 3

*Comparison of Listener Ratings of Children with Epilepsy (CWE) and Typically-*

*Developing Children (TD)*

|  | Mean Scores: CWE | Mean Scores: TD | Standard Deviation: CWE | 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 $p < .008$

However, as noted in the Introduction section, mean ages of CWE-C and CWE-R differed by two years, which was the motivation for compiling two groups of age-matched comparison children. *Post hoc* analysis suggested that this design choice was somewhat appropriate: For the data as a whole, the correlation between age and overall quality listener score was significant ($r = .33, p = .0198$). Correlations between age and story subcomponents were also significant for vocabulary, story structure, grammar and fluency, with *r*-values ranging from .33 to .38 (see Table 4), but there was no evident correlation between age and color/interest ($r = .19$) or prosody ($r = .10$). We note that, although significant, these correlations were somewhat modest, suggesting that age does not strongly predict listener judgments. In addition, use of the Bonferroni correction would

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limit significance to those comparisons meeting $p < .007$, which would make no correlations between listener judgments and age strictly significant. Finally, the decision to limit any stimulus playlist to stories generated by children of roughly similar ages may have prevented a strong effect of age from emerging on listener judgments.

Given this, we conducted a one-way ANOVA to examine overall listener perceptions of the stories across all four groups to ascertain general profiles of performance. Results were significant for group at $p = .04$, $F(3, 46) = 2.92$. Fisher's LSD (Lindman, 1974) revealed children with chronic epilepsy to receive significantly lower scores than their age-matched peers with no other group differences significant. (See Table 5)

Table 4

*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 | *0.0219 |
| Fluency | r | 0.38 |
|  | p | *0.006 |
| Color/Interest | r | 0.19 |
|  | p | 0.1837 |
| Prosody | r | 0.10 |
|  | p | 0.0485 |

*Significant at $p < .01$

Table 5

*Comparison of Listener Ratings across Narrator Groups*

| Sources of Variance | Degrees of Freedom | Sums of Squares | Mean Squares | $F$ | $p$ |
|---|---|---|---|---|---|
| Group | 3 | 12.46 | 4.15 | 2.92 | 0.0440 |
| Error | 46 | 65.49 | 1.42 | | |
| Corrected Total | 49 | 77.96 | | | |

In the sections that follow, we present the results of two separate sets of analyses: children with chronic epilepsy compared to TD peers and children with recent-onset epilepsy compared to their own age-matched set of TD peers.

*Listener Ratings of Children with Recent-onset Epilepsy (CWE-R) and Children with Chronic Epilepsy (CWE-R)*

The secondary goal of this study was to examine listeners' ratings of narratives produced by children who have been diagnosed with epilepsy for longer periods of time. 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 ($z = -3.13$, $p = .0017$), vocabulary ($z = -3.28$, $p = .0010$), story structure ($z = -2.91$, p = .0036), and grammar ($z = -3.69$, $p = .000$, see Figure 2, Table 6). There were no significant differences between groups in ratings assigned for fluency ($z = -2.39$, $p = .0169$), color/interest ($z = -2.30$, $p = .0212$), or prosody ($z = -1.97$, $p = .484$). We note that without Bonferroni correction, a problem in much past work with

children with epilepsy, both fluency and color/interest would also have shown

significant difference from typical child narrative judgments.

*Figure 2.* Mean scores and standard deviations of CWE-C and TD-C listener

ratings.
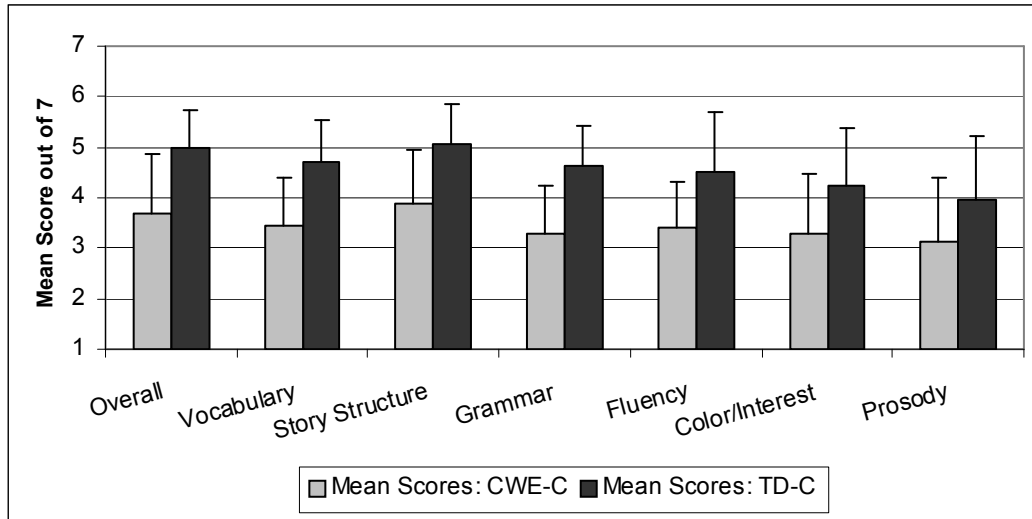


Table 6

*Comparison of Listener Ratings of CWE-C and TD-C*

| | Mean Scores: CWE-C | Standard Deviation: CWE-C | Mean Scores: TD-C | Standard Deviation: TD-C | $Z$ | $p$ |
|---|---|---|---|---|---|---|
| Overall | 3.69 | 1.18 | 4.98 | 0.77 | -3.13 | *0.0017 |
| Vocabulary | 3.44 | 0.95 | 4.70 | 0.83 | -3.28 | **0.0010 |
| Story Structure | 3.87 | 1.09 | 5.07 | 0.80 | -2.91 | **0.0036 |
| Grammar | 3.29 | 0.94 | 4.64 | 0.77 | -3.69 | **0.0002 |
| Fluency | 3.39 | 0.91 | 4.51 | 1.20 | -2.39 | 0.0169 |
| Color/Interest | 3.27 | 1.21 | 4.22 | 1.18 | -2.30 | 0.0212 |
| Prosody | 3.13 | 1.26 | 3.98 | 1.23 | -1.97 | 0.0484 |

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

There were no significant differences in ratings assigned to CWE-R versus ratings assigned to TD-R (see Figure 3, Table 7) for any of the response variables. Moreover, no comparisons remotely approached significance, even without Bonferroni correction.

*Figure 3.* Mean scores and standard deviations of CWE-R and TD-R listener ratings
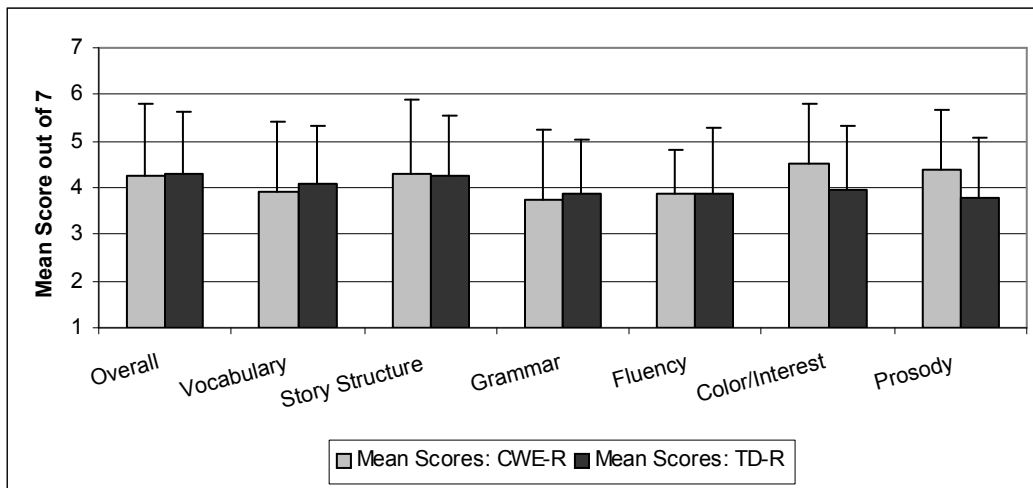


Table 7

*Comparison of Listener Ratings of CWE-R and TD-R*

|  | Mean Scores: CWE-R | Standard Deviation: CWE-R | Mean Scores: TD-R | Standard Deviation: TD-R | $Z$ | $p$ |
|---|---|---|---|---|---|---|
| Overall | 4.26 | 1.55 | 4.28 | 1.33 | 0.11 | 0.9096 |
| Vocabulary | 3.92 | 1.49 | 4.08 | 1.25 | -0.15 | 0.8790 |
| Story Structure | 4.28 | 1.6 | 4.26 | 1.27 | 0.30 | 0.7622 |
| Grammar | 3.73 | 1.5 | 3.86 | 1.19 | -0.11 | 0.9095 |
| Fluency | 3.88 | 0.92 | 3.89 | 1.40 | -0.03 | 0.9697 |
| Color/Interest | 4.51 | 1.27 | 3.95 | 1.38 | 1.10 | 0.2726 |
| Prosody | 4.38 | 1.31 | 3.78 | 1.29 | 1.25 | 0.2112 |

*Factors Influencing Listener Overall Quality Scores*

In order to determine which aspects of the children's narratives (e.g., vocabulary, grammar, fluency, etc.) influenced listeners' scoring of overall quality, we performed a Pearson's product-moment correlation of overall quality scores and listener subscores. Analyses revealed that overall listener quality ratings were highly correlated with vocabulary ($r = .93$, $p < .008$), story structure ($r = 0.94$, $p < .008$), and grammar ratings ($r = 0.96$, $p < .008$). Overall quality ratings also correlated with fluency ($r = .70$, $p < .008$) and color/interest ratings ($r = .59$, $p < .008$), but not prosody ratings ($r = .38$, $p = .0759$, *ns*), see Table 8).

Table 8

*Correlations between Overall Quality and Subscore Ratings*

|  |  | Overall Quality |
| --- | --- | --- |
|  | CWE (N = 25) | |
| Vocabulary | *r* | 0.93 |
|  | *p* | *0.0000 |
| Story Structure | *r* | 0.94 |
|  | *p* | *0.0000 |
| Grammar | *r* | 0.96 |
|  | *p* | *0.0000 |
| Fluency | *r* | 0.70 |
|  | *p* | *0.0002 |
| Color Interest | *r* | 0.59 |
|  | *p* | *0.0030 |
| Prosody | *r* | 0.38 |
|  | *p* | 0.0759 |

*Significant at $p < .008$

*Relationship between Overall Quality Ratings, Formal Language Sample*

*Analyses and Standardized Assessments*

In this analysis, we correlated (Pearson's product-moment) formal measures of story grammar and language from Strekas et al. (2007) with listeners' overall quality scores, which were highly based on their perception of vocabulary, grammar and story structure (see Table 8). The goal was to determine if formal analyses of language samples (e.g., total number of C-units) matched listener perceptions. Additionally, we wanted to determine the construct validity of listener ratings by examining the extent to which standardized test scores were related to listeners' scoring of spontaneous language samples.

Our results indicated that overall quality scores correlated with *CELF* Core Language scores ($r = 0.47$, $p < .01$), *WASI* Verbal IQ scores ($r = 0.51$, $p < .01$), number of story grammar elements included in narratives (N total, $r = 0.64$, $p < .01$) and Vocabulary Diversity (VOC-D; $r = 0.44$, $p < .01$). The only measure that did not significantly correlate with overall listener rating was total number of C-units ($r = 0.17$, $p < .01$, see Table 9). It appears that listeners are able to appreciate most differences in stories that are detected through formal analyses of language samples and standardized assessments.

Table 9

*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 |

*Significant at p < .01

*Relationship between Listener Ratings and Seizure History*

We hypothesized that story production would be more adversely impacted by an earlier onset of epilepsy and a greater number of seizures. The age at which CWE's first seizure occurred was significantly correlated with listener ratings of overall quality ($r$ = .53, $p$ = .0089), vocabulary ($r$ = 0.52, $p$ =.0111), story structure ($r$ = .45, $p$ =.0297), grammar ($r$ = 0.54, $p$ = .0077) and fluency ($r$ = 0.53, $p$ = .0088). However, age at first seizure showed no significant relationship with color/interest ($r$ = 0.52, $p$ = .0356) or prosody ratings ($r$ = 0.35, $p$ =.1041, see Table 10). Contrary to our hypothesis, total number of seizures did not correlate with any listener ratings. This was most likely due to the ceiling effects of our data set (see descriptive statistics in Table 1), in which frequency of seizures was capped at a value reflecting 20 or more seizures.

Table 10

*Correlations between Seizure History and Listener Ratings*

|  |  | Age at 1st Seizure | Number seizures |
|---|---|:---:|:---:|
|  |  | CWE (N = 25) |  |
| Overall | r | 0.53 | -0.09 |
|  | p | *0.0089 | 0.6817 |
| Vocabulary | r | 0.52 | -0.02 |
|  | p | *0.0111 | 0.922 |
| Story Structure | r | 0.45 | -0.08 |
|  | p | *0.0297 | 0.7224 |
| Grammar | r | 0.54 | -0.03 |
|  | p | *0.0077 | 0.8966 |
| Fluency | r | 0.53 | 0.07 |
|  | p | *0.0088 | 0.7347 |
| Color Interest | r | 0.44 | -0.27 |
|  | p | *0.0356 | 0.2200 |
| Prosody | r | 0.35 | -0.30 |
|  | p | 0.1041 | 0.1619 |

*Significant at $p < .01$

*Possible Effects of Anti-Epileptic Drugs (AEDs)*

The fact that greater differences were seen between CWE-C and their unaffected peers could be a result of chronic seizure activity. However, it could also be a result of long-term use of anti-epileptic drugs (AEDs). Out of the twenty-five participants with epilepsy included in the present study, only 5 participants were receiving a polytherapy of AEDs. We performed a *post hoc* analysis (Mann-Whitney *U* converted to Wilcoxon Z) to examine possible differences between these children and those receiving only one AED (i.e., monotherapy). Although children on polytherapy did not perform significantly differently from the other CWE in overall quality rating ($z = -1.36$, $p = .17$, see

Table 11), there exists a trend in which the CWE on polytherapy received lower

overall quality ratings (mean = 3.2) than CWE on monotherapy (mean = 4.1).

This trend is also present for vocabulary, story structure, grammar and fluency

ratings, and is strongest in prosody ratings (see Table 11). Additionally,

color/interest ratings were significantly lower for CWE on an AED polytherapy

than for CWE on an AED monotherapy ($z$ =-2.176, $p$ = .029).

Table 11

*Listener Quality Scores Compared Across AED Groups*

| | Mean Scores | | Standard Deviation | | $Z$ | $p$ |
|---|---|---|---|---|---|---|
| | Monotherapy | Polytherapy | Monotherapy | Polytherapy | | |
| Overall | 4.100 | 3.200 | 1.346 | 1.169 | -1.36 | 0.174 |
| Vocabulary | 3.805 | 2.933 | 1.195 | 0.993 | -1.496 | 0.135 |
| Story Structure | 4.159 | 3.533 | 1.346 | 1.073 | -0.918 | 0.359 |
| Grammar | 3.601 | 2.911 | 1.245 | 0.783 | -1.428 | 0.153 |
| Fluency | 3.642 | 3.356 | 0.887 | 1.174 | -0.341 | 0.733 |
| Color/Interest | 4.042 | 2.664 | 1.358 | 0.577 | -2.176 | *0.029 |
| Prosody | 3.881 | 2.642 | 1.432 | 0.695 | -1.532 | 0.126 |

*Significant at $p < .01$

## Discussion

The purpose of this study was to examine listener perceptions of stories

produced by children with epilepsy. Prior studies of children with epilepsy used

either standardized assessments or structural analyses of language samples, and

employed broadly selected and poorly described samples of children. To

understand how epilepsy impacts the functional aspects of language, we examined

if listeners would perceive differences in narratives produced by children with and

without epilepsy.  A secondary question involved the impact of epilepsy over time, so we examined if differences in listener perceptions appear greater for stories produced by children who have suffered from chronic seizures.

We found that listener ratings of narratives were not greatly different when epilepsy was of recent onset. However, CWE-C narrative ratings were significantly lower than TD-C narrative ratings, specifically in the areas of overall quality, vocabulary, story structure and grammar.  In other words, listeners perceived stories produced by children with chronic epilepsy as less well put together and less complex syntactically and lexically but no different in terms of prosody, fluency or interest.

These findings are in accordance with those from Strekas et al. (2007), which examined the same group of participants included in this study.  In that preliminary study, 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. However, differences in verbal IQ in the chronic comparison were greater than in the recent-onset comparison.  Similarly, Caplan et al. (2004) found that children with complex partial seizures had lower verbal IQs than typically-developing children. Thus, listener perceptions are in agreement with the results of standardized assessments of language production. We also found that listeners' overall quality ratings correlated with verbal IQ and *CELF* Core Language scores. These results further support the notion that listener perceptions are in accordance with standardized assessment results.

In the present study, 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 children with CPS produced more disorganized narratives than did children in the comparison group. However, children whose epilepsy is more recent in onset do not demonstrate the same functional deficits in language production that children with chronic seizures demonstrate. In the closer analysis of the current study, experience with epilepsy plays an important role.  Listeners also provided lower vocabulary ratings in narratives produced by CWE-C, which is consistent with previous findings indicating that children with CPS receive significantly lower scores on standardized measures of expressive vocabulary (Schoenfeld et al., 1999).

Our study results strongly suggest that CWE should be given a basic battery of IQ and language assessments (including collection and analysis of a spontaneous language sample) at baseline, shortly after diagnosis, and on a periodic basis to monitor their cognitive and language status over time. This is consistent with Caplan et al.'s (2004) recommendation that children CPS be given thorough language assessments, with particular emphasis on tracking children whose seizures are poorly controlled, who receive Anti-Epileptic Drug (AED) polytherapies and who have other risk factors for subtle linguistic delays (such as lower socio-economic status or non-native language proficiency).

The fact that listener quality scores of a naturalistic language task were depressed more significantly for the children who had epilepsy longer than three years is of considerable practical utility in monitoring children in academic

settings. Although the elicitation and scoring of expressive language samples is a difficult and time-consuming task, language sample analysis has higher construct validity in tapping the real-world contexts of expressive language performance (Lund & Duchan, 1993). Thus, they should be utilized in conjunction with more standardized measures of language, such as the *Clinical Evaluation of Language Fundamentals, 4th edition* (*CELF-4,* Semel, Wiig & Secord, 2003) for assessment purposes. In addition, some standardized language sampling assessment tools have become available recently, such as the *Strong Narrative Assessment Procedure* (Strong, 1998) and the *Expression, Reception and Recall of Narrative Instrument (ERNNI*, Bishop, 2004).

In addition, we discovered strong correlations among overall quality scores, and vocabulary, story structure and grammar scores. This indicates that listeners' overall quality ratings were most influenced by the narrator's lexical sophistication, inclusion of narrative elements and syntax. It is also possible that lay listeners do not differentiate between these measures. In other words, if they perceive a story as being high quality, then they tend to provide high ratings in all of these areas (overall quality, vocabulary, grammar and fluency) and vice versa if they perceive a story as being low quality. We also discovered a relationship, though not as strong, between listeners' overall quality ratings and their fluency and color/interest ratings. This suggests that the speech fluidity and creativity of narrators had some influence on listeners' overall quality scoring. However, we did not discover a relationship between overall quality and prosody scores, suggesting that this area of narrative production did not strongly influence

listeners' perception of quality and that listeners view these measures as being in a totally separate category.

Our results also indicated a strong correlation between listeners' overall quality ratings and Vocabulary Diversity and number of narrative elements included in the story. Thus, listeners' perceptions are in agreement with certain results of formal language sample analyses. We did not discover a strong correlation between the total number of C-Units contained in the language sample and listener scoring. This suggests that the total length of the narrative did not have a strong influence on listeners' perception of quality.

*Anti-Epileptic Drugs*

Our results support previous research findings indicating that CWE prescribed an AED polytherapy perform worse than CWE prescribed an AED monotherapy (Meador, 2002). This may be due to listener sensitivity to the sedating effects of medication. However, it is also plausible that children who require more complex drug therapies to control seizures have more significant underlying brain pathology. Thus, the direction of the effect on story production cannot be firmly ascertained. Nonetheless, children with epilepsy on more than one AED appear to be at greater risk for the development of perceptible changes in speech and language production and should definitely be screened and monitored by a speech-language pathologist.

*Effects of Seizure History*

An earlier age of onset of epilepsy is associated with poorer cognitive functioning (Caplan et al., 2004; Schoenfeld et al., 2007), poorer language performance (Herman et al., 2002; Hermann, Bell, Seidenberg & Woodard, 2001) and poorer school performance *(*Adewuya et al., 2006; Chaix et al., 2006). Similarly, our analyses revealed that age at first seizure was predictive of listeners' overall quality, vocabulary, story structure, grammar, and fluency scores. In other words, CWE whose first seizure occurred at an older age obtained higher narrative ratings from listeners. Conversely, children who developed epilepsy at younger ages obtained lower narrative ratings. However, we discovered no relationship between total lifetime number of seizures and listener ratings. Therefore, age of first seizure was more predictive of listener ratings than was total number of seizures. This is surprising, given that our other analyses discovered strong effects of chronicity, which imply some relationship with extent and duration of seizures.

Overall, these results suggest that age of onset interacts with duration of experience with seizures (rather than their frequency) to produce perceptible changes in language function. Thus, children who develop epilepsy at a very young age should be carefully screened and monitored, for they are most at risk for perceptible changes in language performance.

*Limitations*

One limitation of this study and Strekas et al. (2007) was their cross-sectional design. A longitudinal study that contrasts expressive language skills at

the onset of epilepsy with these skills over the course of childhood epilepsy would better ascertain if epilepsy is the source of diminished language abilities. This work would more accurately address whether lower performance on measures of cognition and language is caused by seizures, their treatment, or some underlying neuropathy.

Although carefully selected to be somewhat more homogeneous than prior studies, children in the CWE groups still differed in numerous ways. For example, some children experienced simple seizure activity in addition to complex seizure profiles. Finally, eventual sample sizes were relatively low compared to some prior research.

Our inability to find an effect of seizure history on listener judgments may have reflected the non-linear coding scheme for reporting lifetime number of seizures. Scores were capped at 8 for any number of seizures above 20, which may have obscured the effect of large numbers of seizures over time on perceptions of language performance.

*Future Directions*

Research that specifically examines the effects of AEDs on the progression of language and cognitive skills in children are needed. The majority of studies present in the current literature involve adult participants. Although seizure management is the primary goal in prescribing medications to CWE, if there were more information about which medications and which combinations of medications are most detrimental to language and cognitive skills, then doctors could make more informed decisions in selecting drugs for their patients.

The current study only examined listener perceptions of children having localization-related epilepsy with focus in the left hemisphere. Although the left hemisphere is considered dominant for most language functions, and epilepsy with focus in this hemisphere should have produced the largest differences from perceptions of typical children's performance, it would be interesting to observe whether listeners perceive any differences in narratives produced by children with LRE having a focus in the right hemisphere. Candidate differences might be those involving overall story goals, or prosody, since both skills have been observed to be impaired in right hemisphere damage in a pilot study (Strekas et al., 2006), and in the general literature on aphasic syndromes (Springer & Deutsch, 1998).

It is also important for research to track language performance and listener perceptions of language use in children with more severe profiles of epilepsy. The children in this study can be considered to be somewhat mild in terms of seizure disorder when compared to other types of epilepsy, and none had obvious co-morbid conditions, such as intellectual impairment syndromes, autism, and so forth, which would additionally impact language skills both at onset of seizures and over time.

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 skills (e.g. creativity) that standardized tests do not. Studies that examine only standardized test scores may not tell the whole story because they examine language in a decontextualized manner and are not as sensitive as language samples in picking up on the subtle expressive language

differences present in children with epilepsy. In the present study, we examined

standardized test scores and also examined language samples in more than one

way (i.e. through both formal analyses and listener ratings). This provided us with

a variety of data, the type of variety that is needed in future research that

examines the language and cognitive skills of CWE.

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 | 2 | 3 | 4 | 5 | 6 | 7 |
|---|---|---|---|---|---|---|
| Very low quality | | | | | | Very high quality |

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

## VOCABULARY

☹ ☺

| 1 | 2 | 3 | 4 | 5 | 6 | 7 |
|---|---|---|---|---|---|---|
| Very low quality | | | | | | 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 | 2 | 3 | 4 | 5 | 6 | 7 |
|---|---|---|---|---|---|---|
| Very low quality | | | | | | 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 | 2 | 3 | 4 | 5 | 6 | 7 |
|---|---|---|---|---|---|---|
| Very low quality | | | | | | 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           2           3           4           5           6           7

Very low                                                                            Very high

quality                                                                               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           2           3           4           5           6           7

Very low                                                                            Very high

quality                                                                               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           2           3           4           5           6           7

Very low                                                                            Very high

quality                                                                               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)?

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