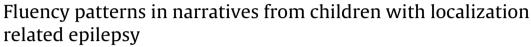
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ABSTRACT

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 psychoeducational 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 wellstructured 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

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