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Evaluation of the language profile in children with rolandic epilepsy and developmental dysphasia: Evidence for distinct strengths and weaknesses

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ABSTRACT

Although benign, rolandic epilepsy (RE) or benign childhood epilepsy with centro-temporal spikes is often associated with language impairment. Recently, fronto-rolandic EEG abnormalities have been described in children with developmental dysphasia (DD), suggesting an interaction between language impairment and interictal epileptiform discharges. To investigate if a behavioral-linguistic continuum between RE and DD exists, a clinical prospective study was carried out to evaluate the language profile of 15 children with RE and 22 children with DD. Language skills were assessed using an extensive, standardized test battery. Language was found to be impaired in both study groups, however RE and DD were associated with distinct language impairment profiles. Children with RE had difficulties with sentence comprehension, semantic verbal fluency and auditory short-term memory, which are unrelated to age of epilepsy onset and laterality of epileptic focus. In children with DD, sentence comprehension and verbal fluency were among their relative strengths, whereas sentence and lexical production constituted relative weaknesses.

1. Introduction

Rolandic epilepsy (RE), or benign epilepsy with centro-temporal spikes, is among the most common types of idiopathic focal childhood epilepsy, accounting for 14–20% of all pediatric epilepsy cases (Camfield & Camfield, 2002; Cavazzuti, 1980; Fejerman, 2009; Holmes, 1993). The electroencephalography (EEG) pattern of RE is characteristic, showing frequent interictal sharp waves in centro-temporal or sylvian brain regions (Wolff et al., 2005). Typical seizure onset ranges from 4;00 to 10;00 years (Shields & Snead, 2009) and seizures usually remit during adolescence, even when no treatment is provided (Goldberg-stern et al., 2010). These seizures occur infrequently, are of limited duration (30–120 s) and commonly take place nocturnally during sleep (Chan & Lee, 2011; Fejerman, 2009; Hughes, 2010). The symptoms of these seizures are diverse and can include brachial and orofacial tonic or clonic contractions, hypersalivation, loss of consciousness and speech arrest if the seizure occurs in the language dominant hemisphere (Shields & Snead, 2009). Although RE is considered a benign disorder, several recent studies provide evidence for mild to

Abbreviations: CELF-4NL, Clinical Evaluation of Language Fundamentals, fourth edition, Dutch version; CFD, Concepts and Following Directions; CLS, Core Language Score; DD, developmental dysphasia; DSM-V, Diagnostic and Statistical Manual of Mental Disorders, fifth edition; EEG, electroencephalography; ELI, Expressive Language Index; EV, Expressive Vocabulary; FS, Formulating Sentences; ICD-10, International Classification of Diseases and Related Health Problems, tenth edition; ILAE, International League Against Epilepsy; IQ, intelligence quotient; LCI, Language Content Index; LSI, Language Structure Index; MUCLA, Multidisciplinary University Centre for speech, Language pathology and Audiology; N, Number; NR-B, Number Repetition Backward; NR-F, Number Repetition Forward; PA, Phonological Awareness; Pc, percentile rank; PPVT-III-NL, Peabody Picture Vocabulary Test, third edition, Dutch version; RE, rolandic epilepsy; RLI, Receptive Language Index; RS, Recalling Sentences; SA, Sentence Assembly; SC, Sentence Comprehension; SR, Semantic Relationships; USP, Understanding Spoken Paragraphs; WA, Word Associations; WC-E, Word Classes – Expressive; WC-R, Word Classes – Receptive; WD, Word Definitions; WHO, World Health Organization; WS, Word Structure.

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moderate impairments in a broad range of cognitive functions, including psychomotor speed and dexterity, executive functions, attention, visuoperceptual skills, memory and, most prominently, language (Garcia-Ramos et al., 2015; Giordani et al., 2006; Goldberg-stern et al., 2010; Kwon, Seo, & Hwang, 2012; Neri et al., 2012; Overvliet et al., 2013; Pinton et al., 2006; Riva et al., 2007; Smith, Bajomo, & Pal, 2015; Vannest, Tenney, Gelineau-Morel, Maloney, & Glauser, 2015; Verrotti, Filippini, Matricardi, Flavia, & Gobbi, 2014). These language impairments can vary over a wide range of language skills including phonological processing, lexical and semantic knowledge, verbal memory, written language skills, verbal fluency and grammar skills (Clarke et al., 2007; Goldberg-stern et al., 2010; Monjauze, Tuller, Hommet, Barthez, & Khomsi, 2005; Northcott et al., 2005; Overvliet et al., 2013; Riva et al., 2007; Vannest et al., 2015).

Though seemingly unrelated at first glance, RE shares several features with developmental dysphasia (DD), a developmental disorder characterized by persistent and severe deficits in receptive and/or expressive language skills in the absence of sensory deficits, a major neurologic or psychiatric condition, or an environmental disabling state (Billard, Fluss, & Pinton, 2009; De Guibert et al., 2011; van Weerdenburg, Verhoeven, & van Balkom, 2006). Whereas language impairment is likely to be subtle in children with RE (Riva et al., 2007; Vannest et al., 2015), severe language impairment is one of the clinical diagnostic criteria of DD (e.g.: ICD-10: WHO, 1993; DSM-V: American Psychiatric Association, 2013). Receptive and expressive components of language form (phonology, morphology and syntax), language content (vocabulary and semantics) and language use (pragmatics) can be impaired to different degrees. Although DD refers to children with a resistant language deficit, in the international literature, DD (Parisse & Maillart, 2009), developmental language disorder (Bishop et al., 2016; Rapin & Dunn, 2003) and specific language impairment (Bishop, 1991, 2006) are often used interchangeably (De Guibert et al., 2011). The etiology of DD remains largely unknown. However, whereas epileptiform activity is a recognized and essential diagnostic criterion of RE (Smith, 2005), several studies reported a higher incidence of nocturnal interictal encephalographic discharges in the fronto-rolandic region in children with DD compared to typically developing children (Billard et al., 2009; Nasr, Gabis, Savatic, & Andriola, 2001; Neuschlová, Štěrbová, Žáčková, & Komárek, 2007; Parry-Fielder et al., 2009). The question then arises whether DD can be perceived as an entity distinct from epilepsy syndromes associated with language dysfunctions, or should be given a position on the continuum alongside RE.

Identifying and comparing the extent of cognitive deficit profiles is a major challenge in the research field of childhood neurological disorders. The presence of similar cognitive disabilities may reflect similar underlying cognitive, neural, molecular and/or genetic mechanisms between two or more neurological disorders (Archibald & Alloway, 2008; Ypsilanti & Grouios, 2008). This may provide indications where to search for the neurobiological substrate underlying the impaired cognitive systems (Rice, Warren, & Betz, 2005). The goal of the present study is to evaluate a broad range of language functions in children with RE and DD using the comprehensive language test battery 'Clinical Evaluation of Language Fundamentals 4', Dutch version (CELF-4NL) (Kort, Schittekatte, & Compaan, 2008) and the 'Peabody Picture Vocabulary Test III', Dutch version (PPVT-III-NL) (Dunn & Dunn, 2005). Additionally, this study aims to compare the language deficit profiles of children with RE and DD to investigate if a behavioral-linguistic continuum between the two disorders might exist, i.e. whether both patient populations present with similar or distinct patterns of linguistic strengths and weaknesses.

2. Methods

2.1. Participants

Fifteen children with RE and 22 children with DD, matched for age and gender, were included in this study. All children were aged between 7;00 and 14;11 years. They were native monolingual Dutch speaking children and had normal hearing skills. Participants with RE were recruited through the pediatric Neurology outpatient clinic and diagnosed on the basis of all available clinical and EEG data by a pediatric epileptologist (L.L.) and according to the diagnostic criteria formulated by the International League Against Epilepsy (ILAE, 1989). Patients with DD were recruited through the Multidisciplinary University Centre for Speech, Language Pathology and Audiology (MUCLA), University Hospital Leuven and diagnosed by a multidisciplinary team based on neuropediatric, neuropsychological and language examinations. To ensure the persistent character of their language problems, children with DD had to perform below percentile 10 on at least one of the subtests of the language test battery used, after receiving intensive speech language therapy for at least 1 year. Children were excluded if they had a history of chronic medical, neurological or psychiatric conditions other than the disease of investigation.

This study protocol was approved by the Ethical board of the University Hospitals Leuven, Belgium (ML7889). Parents and children were informed about the experiment; informed consent was obtained from all parents/guardians according to the Declaration of Helsinki, with additional assent from all participating children.

2.2. Language and IQ assessment

The Clinical Evaluation of Language Fundamentals, fourth edition, Dutch version (CELF-4NL) was used to assess language performance in an expressive and receptive way (Kort et al., 2008). The following subtests of the CELF-4NL were used (Table 1): Concepts and Following Directions (CFD), Word Structure (WS), Recalling Sentences (RS), Formulating Sentences (FS), Word Classes-Receptive (WC-R), Word Classes-Expressive (WC-E), Sentence Comprehension (SC), Expressive Vocabulary (EV), Word Definitions (WD), Understanding Spoken Paragraphs (USP), Sentence Assembly (SA), Semantic Relationships (SR), Number Repetition Forward (NR-F), Number Repetition Backward (NR-B), Word Associations (WA) and Phonological Awareness (PA).

After administrating this CELF-4NL battery, five indices were derived: the Core Language Score (CLS), a measure of general language ability that quantifies a child's overall language performance, and four other specific language index scores. First, the Receptive Language Index (RLI) provides a measure of auditory comprehension and listening skills. Second, the Expressive Language Index (ELI) gives an indication of the ability to express oneself verbally. Third, the Language Content Index (LCI) is a measure of various aspects of semantic development, including vocabulary, word definitions, comprehension of directions and spoken paragraphs and comprehension of associations and relationships between words. Finally, the Language Structure Index (LSI) measures skills related to the interpretation and production of structural aspects of language, including word structure and formulating and recalling sentences (Semel, Wiig, & Secord, 1998). Each composite index score consists of 2–5 subtests depending on the age category of the child (Table 1).

All subtest scales and composite index scores can be converted to percentile ranks and test-age equivalents, based on an extensive normative study which included 1280 Dutch speaking children