Correlation between language impairment and problems in motor development in children with rolandic epilepsy

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Abstract

Objective: An association between impaired school performance and rolandic epilepsy is frequently reported. Language outcome, in particular, seems to be affected, although rolandic epilepsy originates from the motor–sensory cortex. In this study we tried to find a correlation between locomotion problems and language impairment.

Methods: In this noncontrolled, open, clinical cohort study of 48 children with rolandic epilepsy, a 24-hour EEG and a neuropsychological assessment were obtained for all children.

Results: Children with rolandic epilepsy had a significant delay in reading skills (reading words: mean = 6 months, SD = 11.9, P = 0.002; reading sentences: mean = 8.6 months, SD = 12.7, P = 0.001), compared with the healthy population. There was a significant correlation between problems in motor development and delays in reading skills (reading words: r = −0.426, P = 0.006; reading sentences: r = −0.343, P = 0.03).

Conclusion: Reading performance is impaired in children with rolandic epilepsy. Reading of sentences is more impaired than reading of words. There is a significant correlation between problems in motor development and language, suggesting their interaction at the level of the cortex.

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1. Introduction

Rolandic epilepsy is one of the most common epilepsy syndromes in childhood [1]. Most children with this syndrome have nocturnal seizures with or without secondary generalization, with (1) unilateral paresthesia of the tongue, lips, gum, and cheek; (2) unilateral clonic or tonic activity in the face, lips, and tongue; (3) dysarthria; and (4) drooling [2]. The seizures last from seconds to minutes; frequency is variable, but seizures may occur in clusters [3,4].

The interictal EEGs of these children reveal centrotemporal spikes, which may be unifocal (in 60%) or multifocal and increase during drowsiness and all stages of sleep [5,6]. EEG characteristics that appear to correlate with educational impairments are the presence of an intermittent slow wave focus during wakefulness, a large number of spikes in the first hour of sleep (and during the whole night of sleep), and multiple asynchronous bilateral spike–wave foci in the first hour of sleep [7].

Rolandic epilepsy is generally considered to be a relative benign disorder without cognitive, neurological, or intellectual abnormalities [8–10]. However, recently, the benign character of rolandic epilepsy has become a matter of vigorous debate as outcomes of neuropsychological test results, especially tests of language, in children with rolandic epilepsy differ from those for healthy children [11–15]. Performance on both phonological and semantic language tests is impaired in children with rolandic epilepsy, especially in those children with atypical seizures. However, the Full Scale IQ is often in the normal range in these children [13]. It is unknown at the moment where this language impairment originates from in children with rolandic epilepsy, as the rolandic spikes originate not from the brain’s language areas, but from the motor–sensory cortex. For that reason, we would expect more problems in locomotion than in language.

Currently, there is no consensus on the necessity for drug treatment in children with rolandic epilepsy [18,19]. The low seizure frequency and relatively mild character of the seizure symptoms are arguments against the use of antiepileptic drugs. However, if rolandic epilepsy can induce cognitive and specifically language impairments and consequent problems in school, this could be an argument to start treatment [20–22].

The goal of this study was to explore the nature of education impairment in children with rolandic epilepsy as reported by parents.
and assessed with neuropsychological tests to find a possible explanation for the language impairment frequently reported in the literature. We also wanted to determine if this language impairment correlates with problems in motor development.

2. Methods

2.1. Participants

In total, 48 children consecutively referred to the tertiary Epilepsy Centre Kempenhaeghe, Heeze, The Netherlands, in the period July 2001 to December 2009 were included. Electroencephalographic recordings and neuropsychological assessments were obtained within the same week. Clinical and seizure characteristics were critically analyzed blindly by two pediatric neurologists (S.K. and J.N.) to confirm the diagnosis of rolandic epilepsy in these children. Agreement between the pediatric neurologists and EEG confirmation were required to confirm the diagnosis.

2.2. Measures

2.2.1. Semistructured interview

Parents were administered a semistructured questionnaire on the child’s developmental history. For the purpose of this study we focused on complaints or problems in the fields of attention and concentration, perception of language, language expression, visuospatial skills, memory, planning, motor development, reading, writing, and calculating. Achievement of developmental milestones (such as walking) was systematically assessed during history taking. The milestones achieved by the children were compared with those of their siblings. In addition, parents were asked whether their children had undergone physical therapy in the past.

2.2.2. Academics: Learning efficacy quotient

To measure possible educational delay the learning efficacy quotient, education level/months of education (EL/ME) × 100, was calculated by using the standardized Dutch instrument Tempo Testen. The Tempo Testen comprised three subtests: reading words, reading sentences, and calculating. In the subtest on reading words, subjects have to read aloud as many words as possible in a specific time. In the subtest on calculating, they had to read aloud as many sentences as possible in a specific time. In the subtest on calculating, the children had to do as many calculations (sums) as possible in a specific time. Only correct words/sentences/calculations were counted. Subsequently performance was compared with age- and school-level norms to establish an estimate of educational attainment.

The learning quotient was calculated by dividing the determined educational level (EL) by the actual months of education (ME) the child had received in primary school (1 school year consists of 10 educational months) and multiplying by 100. A child with a learning quotient <100 was considered to have an educational delay. It is generally assumed that in children with normal intelligence, a reading efficacy quotient <50 indicates serious reading or mathematical problems. Word and sentence reading and mathematical performance were assessed [23–25].

2.3. Inclusion and exclusion criteria

The diagnosis of rolandic epilepsy had to be confirmed independently by the two pediatric neurologists. Their decision was based on EEG findings and seizure semiology.

2.3.1. Electroencephalography

Rolandic spikes are located in the centrotemporal region and in only one hemisphere or independently on both sides. Spikes tend to spread to adjacent regions, and are broad, diphasic high-voltage (100–300 μV) sharp waves with a transverse dipole and often followed by a slow wave. They tend to occur in isolation or in clusters (rhythm of about 1.5–3 Hz). The EEGs were critically analyzed by two clinical neurophysiologists at our institute.

2.3.2. Seizure characteristics

Seizure characteristics include orofacial motor signs, speech arrest, and somatosensory symptoms. Usually there is a hemifacial seizure, which may spread to the arm (and leg) and may become secondarily generalized, mainly during sleep. Seizure duration is generally a few minutes although a status epilepticus may also occur. Atypical seizure characteristics are daytime-only seizures, screaming as part of the seizure, postictal palsy, and aura.

2.3.3. Other criteria

Children had to be between 6.5 and 13 years of age. Those with an IQ <70, an inability to speak or understand the Dutch language, a neurological comorbidity, proven structural abnormalities on MRI, or a lack of MRI were excluded.

2.4. Statistical analysis

The one-sample t test was used in comparisons with normative values. Spearman correlations and Fisher exact tests were analyzed with SPSS Version 17.

3. Results

3.1. Patient characteristics

A total of 48 children were included in this study. Their characteristics are summarized in Table 1. Twenty-six (54.2%) children were boys, and 22 (45.8%) were girls. The mean age of the children at testing was 115.0 months (9 years 7 months) with a SD of 19.7 months. The mean age at diagnosis of was 84.2 months (7 years 0 months) with a SD of 28.9 months. Thirty-nine (81.2%) children were right-handed, 7 (14.6%) left-handed, and 2 (4.2%) ambidextrous.

3.2. Semistructured interview

Reading problems were most commonly reported, occurring in 23 (47.9%) children, followed by attention and concentration problems in 22 (45.8%) children (Table 2). Parents reported problems with language expression in 18 (37.5%), problems with perception of language in 17 (35.4%), problems with mathematics in 14 (29.2%), problems with motor development in 11 (22.9%), and problems with visuospatial skills in 7 (14.6%) children. Three (6.3%) children had problems with both memory and executive function (planning). All these outcomes are subjective reports by the parents. Fisher exact tests revealed P values of 0.202 for problems in motor development and perception of language, 0.165 for problems in motor

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<td><strong>Patient characteristics.</strong></td>
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Note. Values are means (SD) or n (%).
found a significant difference between these two groups with respect to learning efficiency in reading performance: for words $P = 0.06$, and for sentences, $P = 0.03$. This significant effect was not observed for learning efficiency in mathematical skills.

4. Discussion

4.1. Language impairment

Our results confirm that language is an important domain of potential impairment in children with rolandic epilepsy. In more than 30% of the cases, parents reported problems in reading, language expression, and perception of language. Our findings suggest that the more complex the language skill, the stronger the impairment. Interestingly, mathematics capacity did not appear to be impaired, confirming that rolandic epilepsy is a language-related learning disorder and not a general learning disorder.

4.2. Impairment of morphological and semantic skills

In analysis of the reading disorders in greater depth with the EL/ME quotient, reading of sentences was more impaired than reading of words, suggesting greater impairment of semantic skills of language. This is in line with the findings of Monjaize et al. in their study of 16 children with rolandic epilepsy, either active or in remission. They found moderate or severe language impairment of mainly expressive grammar and literacy skills. Language skills obtained later were more vulnerable, and treatment did not have an effect on language disorders [17]. Riva et al. tested language functions in 24 children with rolandic epilepsy with an active focus and 16 control children. They found that the children with rolandic epilepsy had mild language defects on verbal re-elaboration of semantic knowledge and lexical comprehension [16].

4.3. Reading disorders

According to their parents, the children with rolandic epilepsy had more frequently problems with reading (47.9%). This was confirmed by our neuropsychological assessment of reading performance, which indicated that 45% of the children had a word reading quotient <70 and 55% of the children had a sentence reading quotient <70, in line with other studies. Clarke et al. performed a study in which they tested 55 children with rolandic epilepsy, 150 children in a control group, and their siblings and parents. Among the rolandic epilepsy group, 55% had reading disorders (odds ratio = 5.78) [26]. Papavasiliou et al. compared 32 children with rolandic epilepsy with 36 healthy control children. The rolandic epilepsy group had significantly lower scores than controls in spelling, reading aloud, and reading comprehension. Increased epilepsy duration and increased seizure frequency were less common in patients with no or slight written language problems [27]. Carlsson et al. compared 15 adolescents and young adults with dyslexia and rolandic epilepsy with 15 adolescents and young adults with dyslexia but without rolandic epilepsy. The rolandic epilepsy group scored significantly lower than the control group on reading ability of nonrelated words. There were more reading errors and a tendency toward attention impairment in the rolandic epilepsy group [28].

4.4. Correlation between problems reported by parents and delay in learning efficiency

Correlation analysis showed an association between problems perceived by parents in motor development and language impairment (reading sentences and words), as assessed by neuropsychological testing, in children with rolandic epilepsy. Rolandic epilepsy is a syndrome characterized by epileptiform discharges originating
from the rolandic strip, also known as the motor–sensory cortex. Language impairment is typically observed in children with rolandic epilepsy [13,16,17]. It has been suggested that the arcuate fasciculus, which connects and transports information between Wernicke’s area and Broca’s area, has an endpoint in the precentral gyrus (premotor and primary motor areas) [29]. It was shown previously that during overt reading, the excitability of the primary motor hand area is increased in the language-dominant hemisphere in children [30]. The children who were tested in this study on reading sentences and words had to read aloud the sentences and words. This result confirms our own hypothesis that epileptiform activity originating from the rolandic strip might spread to the perisylvian cortex, where language function is located [31]. Our results show an association on both levels—a delay in motor and language development. This hypothesis could explain the language impairment frequently observed in children with rolandic epilepsy. Past research has shown a functional connection between the inferior frontal gyrus (which plays a critical role in speech production) and the orofacial motor cortex in patients with intractable temporal lobe epilepsy. During surgical treatment of these patients, stimulation of the former resulted in evoked responses in the latter, and vice versa. The other two significant correlations suggest that parent-reported complaints have high validity. Children with parent-reported mathematical problems indeed had a delay in learning efficiency in mathematics, and children with parent-reported problems with language expression did have a longer delay in learning efficiency in reading words.

4.5. Implications for the future

Children with an age at onset <8 years and children with epileptic discharges more than 50% of the time during sleep are at risk for language impairment [33]. Failure in language development at a specific age influences further cognitive development [34], for example, school achievement [35]. This can be an additional motivation to treat rolandic epilepsy [21,22,23]. This issue was addressed by Kossoff et al., who described a pilot study in six children with rolandic epilepsy. They replaced the AED taken by the children with levetiracetam 40 mg/kg/day and retested the children after 6 months. Subjective parent-reported improvement was noted in reading and writing. A trend toward improvement was found for auditory comprehension and auditory verbal memory when the children were assessed neuropsychologically [37]. Ay et al. found that reading disorders could persist in children with rolandic epilepsy. In their study, they divided the children with rolandic epilepsy into three groups: children with a rolandic focus who had not yet received AED treatment, children with a rolandic focus who were receiving AED treatment, and children with an improving rolandic focus in whom AED treatment was withdrawn. In the latter two groups reading ability was impaired, and the authors concluded that reading problems could persist when rolandic epilepsy was already in remission [38]. Von Stülpnagel et al. tested levetiracetam in 32 children with rolandic epilepsy. Improvement in cognition, including language and behavior, was reported by 31.3% of the parents of these children [39].

Another option in treatment of language impairment is speech therapy and remedial teaching [40,41]. An appropriate early intervention program could have long-lasting positive effects, as was shown in both children with developmental dysphasia and in children who were at risk for reading disability because of a family history of reading disabilities [42], as well as in children who were at risk for dyslexia [43]. In a randomized controlled trial, Gillam et al. tested four language intervention programs in children with language impairment and found that all forms of therapy improved language and temporal auditory processing [44]. To our knowledge, no such studies exist for rolandic epilepsy.

4.6. Limitations of the study

First, this study was performed in a tertiary referral center for epilepsy. The population of children with rolandic epilepsy may not reflect the general population of children with rolandic epilepsy. Second, we did not take into account the use of AEDs, which could also have cognitive side effects. Third, developmental motor skills were assessed by questioning parents on achievement of milestones and not objectively determined by a physical therapist.

5. Conclusions

Our results confirm that there exists a high prevalence of language impairment in children with rolandic epilepsy as previously reported [13,16,17]. Reading disorders are frequently observed by parents of children with language impairment as was confirmed by our assessments. Reading sentences (semantic language skills) is more impaired than reading words (morphological language skills). Interestingly, mathematics capacity was not impaired, confirming that children with rolandic epilepsy have a language-related learning disorder and not a general learning disorder. We found a correlation between problems with motor development and language impairment in children with rolandic epilepsy, which agrees with the manifestation of epileptiform activity originating from the rolandic strip and language impairment.

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References


