

Review

Language skills in children with benign childhood epilepsy with centrotemporal spikes: A systematic review

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ARTICLE INFO

Article history:

Received 13 February 2018

Revised 6 April 2018

Accepted 6 April 2018

Available online xxxx

Keywords:

Benign childhood epilepsy with centrotemporal spikes

Rolandic epilepsy

Language

Cognition

ABSTRACT

Benign childhood epilepsy with centrotemporal spikes (BECTS) is the epileptic syndrome that most affects preschool and school-age children. Despite being usually considered a benign condition, several studies have shown that this epileptic syndrome is responsible for cognitive morbidities in children, namely at the level of attention and memory, as well as language. However, language disorders are still superficially addressed by the literature. This review aimed to compile and synthesize recent literature in this area. This systematic bibliographic research comprises studies published between 2005 and 2016 in PubMed, Science Direct, and PsycInfo computer databases that included the keywords “language”, “cognition”, “benign childhood epilepsy with centrotemporal spikes”, “rolandic epilepsy”, and “children”. Studies were selected according to the following criteria: (i) published in scientific peer reviewed journals; (ii) performed with children between the ages of 3 and 16 years; and (iii) performed in children with BECTS in the active phase or after remission. Eighteen studies met these criteria. Nine studies mentioned language skill disorders in children with BECTS in the receptive and productive domains of semantics and seven studies in morphosyntax. Regarding phonological awareness, six studies have found limitations in the intrasyllabic, syllabic, and phonemic levels. These studies have also detected deficits in verbal fluency (semantic and phonemic) and in verbal memory. Because of the heterogeneity of the study samples and the use of innumerable distinct tasks in the evaluation of language skills, the results obtained still show some lack of consensus regarding the affected areas. Despite this, it was possible to synthesize and define more precisely the oral language variations presented by this population. We have concluded that the changes in semantic skills are the most frequently mentioned. The studies have also showed morphosyntactic and phonological disorders, in spite of showing some variability among them.

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1. Introduction

Epidemiological studies have showed that epilepsy affects about 1 to 2% of preschool and school-age children [1–3]. This neuropathology encompasses several syndromes, differing in clinical manifestations, etiology, age at onset, and neurophysiological characteristics. It does not present a static feature, existing several factors – genetic, physiological, and developmental – that influence its appearance and manifestations [4,5]. Benign childhood epilepsy with centrotemporal spikes (BECTS) is an idiopathic focal epilepsy, usually starting at ages 3 to 13 years [6,7]. It is considered the most common epilepsy syndrome among children with this neuropathology, corresponding approximately to 24% of the total [8–11].

Although BECTS has been reported as a benign condition in terms of seizure control, several evidences had shown cognitive comorbidities and behavioral disorders [6,10,12]. The various neurocognitive

comorbidities associated with this epilepsy arise from the fact that epilepsy seizures may inhibit, alter, or distort brain development, as well as several underlying functions [5,13–15]. The brain functions necessary for the use of language may be diminished, either indirectly, as a result of associated cognitive limitations, or directly, as a consequence of epileptic seizures [3,4]. Since epileptic discharges in this type of epilepsy occur in central or medial regions of the temporal lobe, we consider this a good model to understand the relationship between epileptic activity and language functions.

In recent years, because of the development of knowledge in the field of epileptology, there has been an increased interest in the study of language in people with this epileptic syndrome [3]. Several investigations prove the presence of language and speech disorders in preschool and school-age children with epilepsy. However, the study of the relationship between epilepsy and language is sometimes complex, because of the existence of several factors that are difficult to study separately, such as neurobiological, pharmacological, and psychosocial factors, which may be the cause of language disorders [3,6,16]. This complexity leads some authors to highlight the lack of consensus regarding the language changes observed in these children [3,5,8–10,14,17–19].

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Therefore, besides assembling and synthesizing studies that report results in this area, this review aimed to extract accurate information about language alterations in children with BECTS and to highlight the consequences of this epileptic syndrome regarding oral language skills.

2. Material and methods

We have conducted a systematic bibliographic research in PubMed, Science Direct, and PsycInfo computer databases, by using the keywords “language” and/or “cognition”, combined with “benign childhood epilepsy with centrottemporal spikes” or “rolandic epilepsy”. A period for the research was established, comprising papers published from 2005 to 2016, written in English, French, Spanish, and Portuguese. We have made an initial analysis and classification of the articles, followed by a more detailed selection according to inclusion and exclusion criteria, in order to select only the relevant studies. We have included studies (i) published in scientific peer-reviewed journals, (ii) performed with children between the ages of 3 and 16 years, and (iii) performed in children with BECTS in the active phase or after remission. We have excluded the following: (i) dissertations or theses not published in scientific peer-reviewed journals, (ii) studies with adults, and (iii) studies including only children with another epilepsy syndrome or other neurological/cognitive deficits. After this selection, we have made a more detailed review and evaluation of the designated studies.

2.1. Study selection

After conducting the research in the three databases, 397 papers were found. The duplicate results were automatically eliminated, and the number of articles identified was reduced to 374. We have analyzed titles and/or abstracts of these articles, excluding articles that did not specifically evaluate language components, as well as studies performed in children with other neurological/cognitive disorders or in children aged above the defined age range or studies that only presented neuroimaging data. From this initial selection, we have selected 70 papers for full reading. At the end, 18 studies that fully complied with the previously established criteria (Fig. 1 and Table 1) were selected. The

most frequent causes for excluding studies were the use of unreliable instruments of language evaluation and the lack of information about children’s language skills.

3. Results

The 18 selected studies (Table 1) were conducted in 13 countries and published in eight scientific peer-reviewed journals. A total of 496 children with BECTS, aged between 3:11 and 16 years, were evaluated in the active phase of epilepsy and/or after remission. Most of the authors differentiate gender, which led us to observe that, in the total sample, there is a greater percentage of male children (56%, $n = 278$) than female children.

All studies were empirical, two of which were retrospective, and the remaining were prospective. In 14 studies, the results obtained from children with BECTS were compared with the results from children in control groups, paired by several variables. In the remaining four studies, the results were compared with the normative data of the applied tests. The main features of each of the included studies are presented in Table 2.

From the review, we could verify that nine of the studies mention changes in the area of semantics. Several tasks were identified in which children with BECTS had deficient results in this language domain: verbal re-elaboration of semantic knowledge, lexical field recovery [9,20,21], correction of semantically incorrect sentences, analysis of the emotional content of sentences, recognition and expression of interpersonal relations [11], vocabulary [22–24], naming attributes [25], word definition, and auditory comprehension [21]. In terms of morphosyntax, seven investigations identified perturbations in both morphological and syntactic knowledge: sentence completion [6], understanding of ambiguous sentences [8], association of sentences with pictures, use of clitic pronouns [26], derivation of adjectives [11], formulation of sentences [27], word production [23], and comprehension of oral instructions of varying syntactic complexity [25]. Some tasks that were presented as altered in the morphosyntax domain appear as preserved in other studies, and there was no consensus regarding performance in these tasks. The phonological metalinguistic skills were also affected at various levels. Six studies have identified changes in

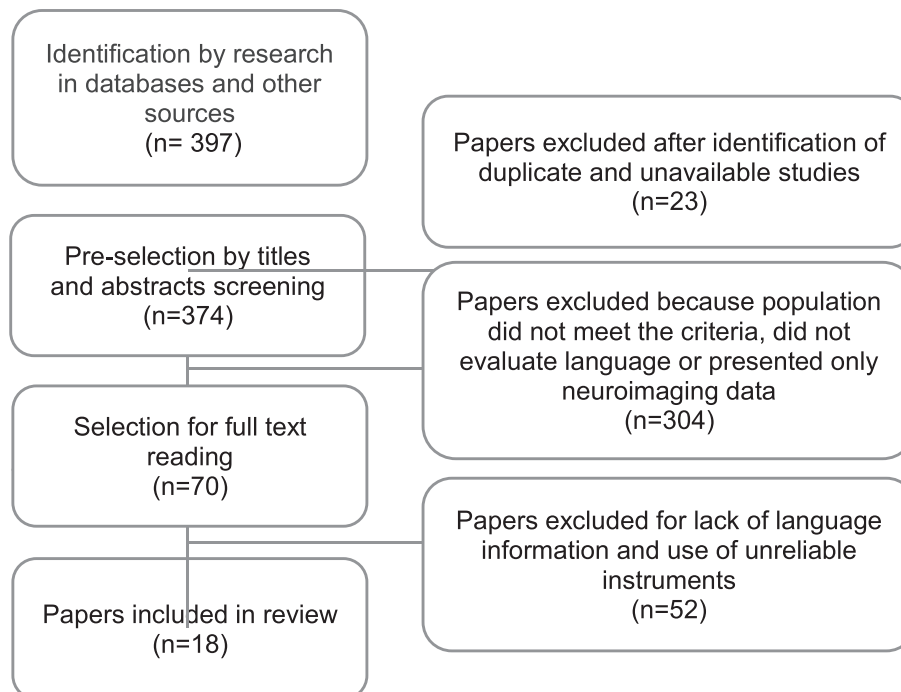


Fig. 1. Flow of studies through the review: selection method.

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