The development of pragmatic skills in children after hemispherotomy: Contribution from left and right hemispheres

CrossMark

Jessica Save-Pédebos [a,b,c,d], Charlotte Pinabiaux [e], Georg Dorfmuller [d], Sarah Ferrand Sorbets [d], Olivier Delalande [d], Isabelle Jambaqué [a,b,c,d], Christine Bulteau [a,b,c,d,*]

[a] INSERM U1129 "Infantile Epilepsies and Brain Plasticity", Paris, France
[b] Université Paris Descartes, Sorbonne Paris Cité, France
[c] CEA, Gif sur Yvette, France
[d] Rothschild Foundation Hospital, Pediatric Neurosurgery Department, Paris, France
[e] CHArt-P10 (EA4004) (Human and Artificial Cognition), Department of Psychology, University Paris Ouest Nanterre La Défense, France

ARTICLE INFO

ABSTRACT

Purpose: Hemispherotomy (H) is the standard treatment used to cure hemispheric epileptic syndromes in childhood. The postoperative linguistic profile involves hemispheric specialization processes and developmental cognitive plasticity. This research concerns pragmatic aspects of language as a tool for communication which involves both linguistic and extralinguistic communication in context. Our aim was to analyze whether any correlation exists with age at surgery and side of surgery on pragmatic skills following H.
Method: Forty children who underwent H (23 females, 16 right H) were evaluated at a mean age of 12.8 years (±2.6) with two receptive tasks (oral comprehension and syntactic judgment), the Children's Communication Checklist (CCC) rating scale, and the Behavioral Rating Inventory of Executive Function (BRIEF) questionnaire in order to evaluate the role of executive functions on pragmatic skills. Children operated on before the age of 18 months were considered the "early" group (5 right H and 9 left H), while those operated on later were called the "late" group (11 right H and 15 left H).
Key findings: The whole group had significant deficits in all three measures. We demonstrated a statistically significant crossed interaction between the side of H and the age at H with pragmatic language impairments ($F(1,36) = 17.48$; $p = .0002$) and disorders in executive function ($F(1,36) = 5.80$; $p = .021$) in left early H and in right late H patients. These findings are consistent with the previous studies of pragmatic language impairments concerning adolescents and adults with right hemisphere damage and emphasize the contribution of structural language in the early stage of verbal communication.
Significance: These results emphasize for the first time that hemispherotomized children have pragmatic language impairments that are independent of receptive language. Our findings are congruent with the recent theory on pragmatic language development in childhood with evidence of a participation of the left hemisphere at the early age followed by right hemispheric specialization and involvement of executive functions, independently of receptive language.

© 2015 Elsevier Inc. All rights reserved.

1. Introduction

In children with medically refractory epilepsy caused by extensive congenital or acquired hemispheric lesions, hemispherotomy (H) (or

hemispherectomy, depending on the surgical procedure) is an effective treatment option leading to seizure freedom in 52 to 80% of cases, associated with an improvement of neurocognitive prognosis, autonomy, and quality of life [1,2]. Many studies have highlighted the remarkable capacity of the brain to recover language after left H, thanks to functional reorganization towards homologous contralateral regions [3–6]. However, some limitations exist concerning phonological skills and syntactic comprehension which specifically depend on the left hemisphere [7,8]. Yet, only a few studies have explored language as a tool for communication after H. Impaired social cognition has been found in some case reports of right hemispherectomized patients [9,10], suggesting that the right hemisphere plays a particularly important role in social cognitive functioning. Pragmatics encompasses the language

abilities required to organize and transmit inner thoughts. Bishop [11] defined the "pragmatic language impairment (PLI)" as a problem in using language appropriately in a given context, which has been reported in pediatric populations such as autism spectrum disorders or epilepsy [12,13]. Bishop [14] developed tools for assessing qualitative aspects of communication impairment in children and proposed the use of Children's Communication Checklist (CCC) in a clinical setting. Pragmatic language impairment could alter social communication and participate in the emergence of behavioral problems [15]. Pragmatic language impairment is, therefore, distinct from the "specific language impairment" (SLI) which refers to a deficit in language structure, especially phonology and syntax. The development of pragmatic skills involves both linguistic and extralinguistic communication in context [16]. Two main theories are still debated regarding the development of pragmatic language abilities: on the one hand, the influence of right hemispheric specialization implicated in visuospatial ability that is required to respond correctly not only in context but also in understanding the conversational implications (drawing correct inferences from the available information) [17] and, on the other hand, the effect of postnatal cognitive developmental processes of language and executive functions (development of lexico-semantic abilities as well as development of inhibitory control during childhood) [18]. A possible implication of executive functions in social communication and pragmatic language is suspected, since 'high-order' abilities are essential to combine knowledge of context and language in communication acts. Furthermore, studies have shown that pragmatic communication is mediated by a widespread neural network involved in a range of specific executive and social cognitive processes [19,20].

The goal of this study was to examine pragmatic language abilities after hemispherotomy during childhood according to the side of the surgery and the age at the surgery. More generally, we sought to determine the relative contribution of left and right hemispheres to the development of pragmatic skills, taking in account the age at H. Indeed, the effect of H could be different whether it had occurred during the prelinguistic period or after the acquisition of language. In the children who had been hemispherotomized after the acquisition of language, we expected PLI to occur after right but not after left H, given the right hemisphere dominance for pragmatic skills that is reported in the literature. On the other hand, the impact of early, prelinguistic H could be different, and findings would argue either for early hemispheric equipotentiality for pragmatic skills or for early right specialization. In the first case, PLI should be observed for neither left nor right early H patients, while in the second case, it should be observed only for right early H patients. Finally, we examined if the pattern of executive performances was the same as that of pragmatic skills, thereby questioning whether a common network is shared by these two components in early stages of development.

2. Patients and methods

2.1. Participants (Table 1)

Forty children (23 females) 7 to 16 years of age (mean = 12.8 ± 2.6) who underwent hemispherotomy [1] for refractory epilepsy and unilateral lesions participated in the study. Experimental sessions were conducted during the longitudinal follow-up at our institution. Informed consent for the neuropsychological study was obtained from patients and their parents. Inclusion criteria comprised the following: (1) successful seizure control after surgery (Engel classification, class I), (2) at least 1-year postoperative follow-up and without antiepileptic drugs, (3) absence of severe mental retardation (verbal comprehension index or perceptive reasoning index >50), and (4) presence of functional language allowing neuropsychological assessment. The patients underwent left hemispherotomy (24 LH) or right hemispherotomy (19 RH) for the treatment of classical hemispheric epileptogenic conditions: hemispheric cortical development

(HCD, N = 14), Sturge–Weber syndrome (SW, N = 4), sequelae of vascular insults (VSeq, N = 9), and Rasmussen encephalitis (RE, N = 13). All the children underwent a presurgical work-up at our institution including scalp video-EEG, MRI, and a careful neurological examination. Preoperatively, neuropsychological assessment was performed in 32/40, and 7/40 were unable to participate since their seizures were too frequent; the patient #26 had mild speech delay with normal sentences and schooling before surgery. The youngest hemispherotomized children (operated on before the age of 18 months) were characterized by the absence of language before surgery. By contrast, the children operated on later had developed language before surgery and experienced normal abilities presurgically or mild speech delay or signs of aphasia (children with Rasmussen encephalitis affecting the dominant hemisphere but with normal language until the onset of seizures).

The medico-surgical features, the preoperative level of language, and the verbal psychometric outcomes are given in Table 1. The socio-economic status was recorded for each child. The total group had subnormal verbal comprehension index evaluated by the WISC-IV (VCI: 77 ± 18) with higher scores obtained for similarities (8 ± 4), whereas vocabulary (6 ± 3) and comprehension (5 ± 3) subscores were weaker on average. We distinguished four groups according to side of (left or right) and age (before or after the age of 18 months) at H. The group of "early H patients" encompassed 14 patients with congenital hemispheric lesion (9 — hemispheric cortical dysplasia, 4 — Sturge–Weber syndrome, and 1 — vascular–ischemic sequelae) with no or very poor verbal communication preoperatively, whereas there were 26 "late H patients" (13 with RE, 8 with VSeq, and 5 with HCD including 4 with polymicrogyria) who had developed normal or subnormal speech abilities before H. Verbal comprehension index in early LH patients was in the normal range, while for the three other groups, it was borderline.

2.2. Procedure

2.2.1. Receptive language

Receptive language assessment comprised two tasks of the BILO (Bilan Informatisé du Langage Oral): oral comprehension task which evaluates receptive vocabulary and syntax (subject listens to a sentence and has to choose the corresponding picture among four) and syntactic judgment task which assesses the subject's ability to judge whether sentences are well formed (requiring metalinguistic and morphosyntactic abilities). All the raw scores were first converted into z-scores based on French age-normative data [21].

2.2.2. Pragmatic language

We used the French version of the Children's Communication Checklist (CCC) rating scale [14,22] to perform an ecological evaluation of pragmatic language abilities. The CCC is a parental report questionnaire providing a general screen for communication disorders and identifying children (age range from 6 to 16 years) with PLI, for example, among children with autism, traumatic brain injury, or epilepsy. The CCC is widely used to measure pragmatic language in research and was developed to distinguish children with primarily pragmatic language problems from those with other language profiles suggestive of SLI or autism spectrum disorder. The CCC has 70 items divided into nine subscales; five of them (Inappropriate Initiations, Coherence, Stereotyped Conversation, Use of Context, and Rapport) are combined to provide a global pragmatic composite score while the four other scales evaluate SLI (Speech and Syntax) or autism spectrum disorder (Social Relation and Interests). The lower the score on the CCC, the more possible it is for the child to have impairment. All the raw scores were first converted into z-scores using published normative data [23].

2.2.3. Executive functions

The assessment of executive functions was carried out using the French version of the Behavioral Rating Inventory of Executive Function