Language Development in Children with Simple-Partial Left-Hemisphere Epilepsy

Henri Cohen*† and Marie-Thérèse Le Normand†

*Laboratoire de neuropsychologie de l'enfant, Hôpital de la Salpêtrière and †Laboratoire de neuroscience de la cognition, Université du Québec à Montréal

The nature of cerebral involvement in the acquisition of language was addressed in this longitudinal study of children with an early diagnosis of epilepsy with simple-partial seizures (SPE) and with epileptogenic foci localized in the left frontal (LF) lobe. Yearly evaluations of six SPE-LF children on tests of linguistic comprehension (pointing, understanding of narrative, and understanding of prepositions) and production (repetition, lexical diversity, and grammatical production) were carried out between the ages of 3 and 8 years and compared to those of large samples of control children on the same tasks and at each age level. Linguistic production of all children were transcribed, coded, and analyzed using the Child Language Data Exchange System (MacWhinney & Snow, 1991). Individual evolution trajectories revealed that SPE-LF children showed a clear dissociation in linguistic performance between comprehension and production. Linguistic comprehension gradually improved to reach normal performance levels by age 7 while linguistic production, even at later stages, remained quite poor. This dissociation in the development of linguistic performance in SPE-LF children suggests a complex interplay between brain maturation dynamics and dysfunction modulating the succession of stages in language development. The observed persistent deficits in specific aspects of linguistic performance argue for an early involvement of the anterior areas of the left cerebral hemisphere in the production of language.

INTRODUCTION

There have been, these past 20 years, an increasing number of studies on the cognitive consequences of early brain damage on language performance and competence. In general, the effects of unilateral brain injury at different ages have shown that the functions of the hemispheres become progressively...
distinct over a period of years (Hecaen, 1976; Dennis & Whitaker, 1977). Although it was believed that perinatal brain damage rarely produces sustained language deficits in later life, a few studies have clearly revealed that there are long-term consequences to early left-sided brain injury such as subtle deficits in syntactic abilities (Dennis & Kohn, 1975; Dennis & Whitaker, 1976; Woods & Carey, 1979; Aram, Ekelman, Rose, & Whitaker, 1985; Dennis, 1987; Varga-Khadem, O’Gorman, & Watters, 1985; Aram & Ekelman, 1986). Some studies, however, have not found greater linguistic or syntactic deficits with left- than with right-hemisphere damage (e.g., Feldman, Holland, Kemp, & Janosky, 1992; Levy, Amir, & Shalev, 1992), probably because they included children whose lesions were smaller or in different locations than those of the children described in studies where such differences were found. Moreover, the linguistic tests used varied from study to study, making it hard to compare their results in regards to greater linguistic or syntactic deficits with left- than with right-hemisphere damage.

More recently, Thal and colleagues (Thal, Marchman, Stiles, Aram, Trauner, Nass, & Bates, 1991) have observed that children between 12 and 35 months of age with early acquired focal brain lesions showed delays in lexical comprehension and production (see also Marchman, Miller, & Bates, 1991). Moreover, side of lesion was associated with selective effects of language outcomes in that study. Right hemisphere (RH)–damaged children were slightly poorer in comprehension than children with left-hemisphere (LH) lesions, and lesions extending in the left posterior regions were associated with more protracted delays in expressive, but not receptive, language. In a study with 53 children with unilateral brain damage, Bates and colleagues (Bates, Thal, Trauner, Fenson, Aram, Eisele, & Nass, in press) have also found selective deficits in word comprehension and gesture from 8 to 16 months following RH damage as well as selective effects of left temporal injuries on expressive aspects of language across the 10- to 44-month period. The injuries suffered by more than half of the children in the Bates et al. study were multifocal and usually involved more than one area or lobe within the damaged hemisphere. Although these expressive deficits associated with left temporal damage had subsided after 5 years of age, older brain-injured children (between 6 and 9 years) in general performed significantly below normal controls in syntactic diversity (Reilly, Bates, & Marchman, 1998). In contrast to the results in the Thal’s et al. (1991) study, lesion side was not found to be a neurological predictor in a longitudinal investigation of the cognitive correlates of unilateral single brain injury in six Italian children (Dall’Oglio, Bates, Volterra, Di Capua, & Pezzini, 1994). In that study, however, subjects varied as to lesion site and severity with lesions involving multiple lobes in two subjects.

Investigations with children afflicted with epilepsy have also been conducted to study the long-term impact of transient, abnormal clinical events in the brain on cognitive development. Because severe epilepsy may contrib-
due to abnormal patterns of functional organization, it has been difficult to
draw specific conclusions as to the effect of ictal episodes on cognitive de-
velopment. Partly for this reason, little is known about the impact of epileptic
seizures on language acquisition and development, and case studies generally
constitute the bulk of observations. Temporary selective regression of speech
(Deonna, Fletcher, & Voumard, 1982), childhood acquired aphasia, or Lan-
dau-Kleffner syndrome (Landau & Kleffner, 1957; Deonna, 1991), delayed
language and cognitive development (Green & Hartlage, 1971; Dalby, 1977;
Allen & Rapin, 1980), and verbal auditory agnosia (Cooper & Ferry, 1978;
Tuchman, Rapin, & Shinnar, 1991) are examples of linguistic dysfunction
associated with epilepsy. Long-term consequences of such disruption are not
well known but follow-ups of several Landau-Kleffner cases suggest a poor
prognosis (e.g., Soprano, Garcia, Caraballo, & Fejerman, 1994) or moderate
recovery at best (e.g., Zardini, Molteni, Nardocci, Sarti, Avanzini, & Granata
1994). Thus, the diverse etiologies of epilepsy, the often diffuse localization
of epileptic focus, the numerous methods of language assessment, the ab-
sence of careful long-term follow-up, and the lack of appropriate normative
standards of linguistic performance certainly contribute to the paucity of
knowledge about the interaction between epilepsy and language and the dif-
ficulty in determining the extent to which the language outcomes in epilepsy
differ from those in unilateral focal brain damage caused by brain insult other
than epilepsy.

The investigation of language development in children with simple-partial
onset epilepsy (SPE) may help elucidate aspects of the consequences of epi-
leptogenic brain damage on language development. The seizures in SPE are
characterized by focal onset, brief ictal episodes localized in one hemisphere,
without alterations of consciousness and include only motor or sensory
symptoms; there is no mental retardation. These episodes usually occur in
childhood and have generally disappeared by the early teens. In most cases,
the epileptic discharges are restricted to the motor strip, primary sensory
area, and parts of the frontal and parietal lobes (Aicardi, 1986; Njokiktiijien,
1988; Masur & Shinnar, 1992). The child affected with SPE is usually able
to meaningfully interact with his or her environment and, as such, the investi-
gation of language development in these children may contribute to central
issues in developmental neurolinguistics.

In the present study, we collected clinical and experimental observations
of receptive and expressive linguistic performance in six children with SPE
using a normative language protocol and comparing performance to a large
population of age-matched control subjects. The evaluation of linguistic abil-
ities spanned the period from oral language acquisition to the first years of
primary education. It was believed that long-term follow-up of a few, well-
chosen subjects would better reveal the evolution and maturation of language
abilities than the observation of a larger number of individual subjects, dif-
fering in age, in side and site of lesion, and observed only once. Further, a
longitudinal approach would also reveal whether language acquisition proceeds uniformly or in steps and whether specific aspects of language evolve at different rates than others. These concerns are rather difficult to address in one-time observation studies of subjects with neurogenic language disorders. Two questions were addressed in this research: (1) to what extent does language development in children with SPE differ from that of normal age-matched populations and (2) is the nature of deficits, if any, observed in this SPE-LF disorder different from that observed in comparative cases of focal brain injury?

METHOD

Subjects

Six right-handed children were selected from the Department of pediatrics at the Necker and La Salpêtrière hospitals. Inclusion criteria were a language disorder associated with an early diagnosis of SPE, at birth or a few months after birth, and an epileptic focus localized in the left frontal lobe. In all cases, diagnosis of SPE and localization of focus was established by Dr. Jean Aicardi and members of his team, based on EEG analyses showing evidence of slow and epileptogenic abnormalities localized to the anterior region of the left hemisphere. Based on these findings, anticonvulsive therapy was initiated but, by 3 years of age, all six children were off anticonvulsive medication. For all SPE-LF subjects, exclusion criteria were an auditory impairment as revealed by an audiometric examination, a psychopathologic disorder, or a development quotient under 90, as evaluated with the Brunet-Lézine scales (1965). The scales provide a single general development index together with individual scores in language, sociability, oculomotor coordination, and motor development; scores above 90 are considered normal on this scale.

The samples of control subjects in the study came from urban settings and were balanced for sex and family sociocultural level. Exclusion criteria were a sensory or neurological impairment and a mother tongue other than French. This reference group was made up of 576 children equally distributed over six age groups (45, 54, 66, 75, 84, and 96 months) for all three comprehension tasks and the repetition task and of 200 children equally distributed over five age groups (33, 38, 44, 55, and 65 months) in the two remaining language production tasks.

Tasks

Subjects were given three tests of linguistic comprehension and three of linguistic production. Language comprehension tasks included

1. Pointing: The subject was asked to choose which items in a set of pictures were named by the experimenter; there were 29 target and 35 distractor items in the task and the stimuli were chosen from words commonly found in the vocabulary of these children at each age group.
2. Understanding of narrative: The subject was required to answer 14 “what, where, who, does, how, and why” types of questions in the context of a story in pictures, “The fall in the mud.”
3. Understanding of prepositions: The subject was asked to position play figures, ducks in a pond, following instructions from the experimenter; there were 18 such requests to assess representation of space, quantity, partition, and localization.

The linguistic production tasks included (1) Repetition of 33 simple and easy bisyllabic words spoken by the experimenter, (2) lexical diversity as measured by the number of types
TABLE 1
Distribution of Linguistic Evaluations of the Six SPE-LF Children

<table>
<thead>
<tr>
<th>Subjects</th>
<th>33</th>
<th>38</th>
<th>44–45</th>
<th>55–56</th>
<th>65–66</th>
<th>75</th>
<th>84</th>
<th>96</th>
</tr>
</thead>
<tbody>
<tr>
<td>S1</td>
<td>All</td>
<td>All</td>
<td>All</td>
<td>All</td>
<td>C1, P1</td>
<td>C1, P1</td>
<td></td>
<td></td>
</tr>
<tr>
<td>S2</td>
<td>All</td>
<td>All</td>
<td>All</td>
<td>All</td>
<td>C1, P1</td>
<td>C2</td>
<td></td>
<td></td>
</tr>
<tr>
<td>S3</td>
<td>All</td>
<td>All</td>
<td>All</td>
<td>All</td>
<td>C1, P1</td>
<td>C2</td>
<td>C3</td>
<td></td>
</tr>
<tr>
<td>S4</td>
<td>All</td>
<td>All</td>
<td>All</td>
<td>All</td>
<td>C1, P1</td>
<td>C2</td>
<td></td>
<td></td>
</tr>
<tr>
<td>S5</td>
<td>All</td>
<td>All</td>
<td>All</td>
<td>All</td>
<td>C1, P1</td>
<td>C2</td>
<td></td>
<td></td>
</tr>
<tr>
<td>S6</td>
<td>All</td>
<td>All</td>
<td>All</td>
<td>All</td>
<td>C1, P1</td>
<td>C3</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Note. Task legend: All = all six linguistic tasks; C1 = pointing task; C2 = understanding of prepositions; C3 = understanding of narrative; P1 = repetition task.

or different words uttered in a 20-min controlled play session; this is a form of narrative where the subject is asked to verbalize all manipulations and actions with dolls and objects in and around a doll house, and (3) grammatical production as measured by the mean length of the utterances (MLU)—an index of grammatical maturity—derived from the language produced in the preceding task.

Normative data on the comprehension tasks and repetition task are found in Chevrie-Muller (1981) for ages between 3 years, 9 months and 8 years and in Le Normand (1996) on the lexical diversity and grammatical production tasks for ages between 2 years, 9 months and 5 years, 4 months.

Procedure

Epileptic subjects were tested a minimum of two and a maximum of six times on each of the language tasks, between the ages of 3 and 9 years. Table 1 shows the distribution of test evaluations over this period of 6 years. Subjects were tested individually in a quiet room, with one parent present for younger subjects. Testing lasted about 2 hr, including rest periods. The controlled play session was recorded on videotape and all other language productions and interactions were recorded on audio tape. Order of test presentation was the same for all subjects.

RESULTS

Verbal interactions and productions were transcribed directly from the audiotape or videotape to a computer by two speech and language therapists. Transcription, coding, and analysis were done following the Child Language Data Exchange System (CHILDES; MacWhinney & Snow, 1985, 1991) convention for automated analysis of language transcripts. CHILDES includes precise guidelines for orthographic transcription and coding (Codes for Human Analysis of Transcripts; CHAT) and programs for analysis (CLAN). In
this procedure, the sample is segmented into utterances based on terminal intonational contours and pauses. Interrater agreement of transcribed material was above 90%.

The MLU measure was computed as the ratio of the total number of words over the number of utterances produced in 20 min of controlled play, and lexical diversity was the number of types, or different words, produced in this 20-min play context. Scores in all other tasks were transformed into T-scores following conversion tables (Chevrie-Muller, 1981). Performance scores of the six children with SPE were then plotted in reference to the mean performance of the control groups, at each age level, for each particular task. The evolution of the SPE-LF subjects’ linguistic performance in comprehension is presented in Figs. 1, 2, and 3 and in production in Figs. 4, 5, and 6. Observations showed that children with SPE demonstrate marked and sustained deficits in aspects of linguistic production and gradual improvement, over a period of 4–5 years, in aspects of linguistic comprehension. Table 2 shows instances of utterances of one SPE-LF child and one control subject.

**DISCUSSION**

This study attempted to determine whether the developmental outcome in linguistic performance in children with SPE was markedly different from
that of normally developing children and whether the observed patterns of deficits were concordant with those found in children with unilateral focal brain lesions. The data revealed three important results. First, all children with SPE, despite individual differences in linguistic achievement, showed similar patterns of performance, attesting to the apparently uniform effect of left frontal lobe focal seizures on language development. Second, there was a dissociation between the development of linguistic comprehension and linguistic production confirming, at least in this group of children, a preferential involvement of the left cerebral hemisphere in the processing of specific production aspects of language. Third, contrary to recent observations with children with unilateral focal brain lesions, the impact of epileptic seizures on language development appears to last longer and serious deficits in specific language domains may still be observed by 9 years of age.

For all SPE-LF children, there were marked initial deficits in both comprehension and production performance. Delays in comprehension persisted for a period of years before reaching comparably normal levels by age 7. This delay in comprehension performance suggests two compensatory hypotheses. One is that the initial impact of the epileptic seizures on the recruitment of classic language areas in the LH is gradually overcome and, with time, alternative areas are recruited in the same hemisphere. The other hypothesis suggests that the involvement of the RH, in addition to LH participation, is
a necessary condition for normal development in language comprehension. Indirect evidence for this latter alternative comes from a Bates et al. (in press) study where comprehension deficits in children with unilateral focal brain damage were associated with RH but not with LH lesions. It may also be that the RH is more involved and compensates for the loss or diminished LH function.

This dissociation between broad linguistic domains suggests that these aspects of linguistic competence and performance are differentially affected by neurogenic perturbation. Language acquisition in this particular group of children with SPE is apparently characterized by a gradually compensated delay in comprehension and a sustained deficit in production. To the extent that this dissociation can be taken as an index of cortical areas involvement, it seems reasonable to speculate that the integrity of left frontal brain structures is an important condition for the normal development of language. It is tempting to speculate that the contrast between unilateral epileptic disturbances—whether focal or more generalized—and unilateral focal brain lesions is responsible for the sustained nature of the delays and deficits. The dynamic and continuous action of the seizures, as opposed to a probably more static and time-limited action of the brain lesions, may explain why some children recover from brain damage while others show sustained defi-
Evolution of linguistic production: repetition.

Language development in childhood—epileptic seizures constantly impacting on brain tissue while early focal lesions may allow forms of compensatory mechanisms or processes to become more firmly established at or before critical periods. Thus, the so-called window of recovery between 1 and 5–6 years of age, a period in which children with focal brain injury find alternative ways to solve the problem of language acquisition (e.g., Isaacson, 1975), is not readily available. It may be that compensatory mechanisms may be activated only in the case of loss of function; if there is residual function, there may not be (or there may be less) compensation.

The assessment of linguistic abilities of SPE-LF children revealed, in the pointing task, normal levels of comprehension suggesting an absence of encoding problems and a good passive representation of common words. These subjects thus appear to have a good labeling of items in their environment, adequate for their age. At the level of prepositions, there is also evidence of sentence processing beyond simple vocabulary or lexical knowledge. Subjects in this task are required to focus on and recognize a specific element in the sentence which helps with the processing of the whole sentence. These subject–verb–object+nominal expansion sentences (e.g., “Fais nager le canard autour du bassin” “Move the duck around the pond”) contain words which are close to each other in a prepositional space (e.g., around, along,
before, after, behind, etc.; e.g., Clark, 1971). Performance in this task is an index of good perception of the salient elements in a sentence. Although a delay is observed for a few subjects, all children are performing at comparatively normal levels by age 7. Understanding of narrative is another area where these children demonstrate, with time, adequate abilities. The task reflects a good understanding of the story itself, an appropriate encoding of intonational contours, suggesting adequate processing of linguistic prosody. In normally developing children, this is very well attained by 4 to 5 years of age. In the SPE-LF children, there is a lag of a few years but, again, by 7 years of age, all children show relatively normal levels of comprehension. Although this may not be a purely linguistic task—because of the play and picture support in this situation—this treatment of a particular morphological form (‘‘wh-’’ questions; De Villiers, 1996) suggests that these children with SPE-LF possess adequate abilities in receptive aspects of linguistic communication. Temporal and causal forms are included in a narrative to help constitute a story. These explicit relations, previously obscured or partially understood, are generally understood and produced by age 2 in normal children (Nelson, 1996; p. 351). An adequate, normal level of performance is reached, again gradually, after a few years delay. In summary, SPE-LF children always reach a comprehension performance level adequate for their chronological age, after a gradual recovery period of a few years.
In contrast, performance in the three production tasks lagged well behind and below normal levels. Protracted deficits in the repetition task, a simple test of auditory–phonetic aptitude, may suggest encoding problems with phoneme sequences as well as an inadequate motor control of speech. Words in this test were chosen to sample the whole spectrum of French consonants and the poor level of performance of SPE-LF children may reveal an immaturity in articulatory phonetics. Normal children have generally mastered consonant production by 4–5 years of age. Except for one or two subjects, the other SPE children still show a serious deficit in repetition of simple bisyllabic words.

Two bursts of lexical diversity, at about 30 and 42 months of age, characterize the development of language in normal children and correspond to the emergence of simple and more complex sentence production, respectively (Le Normand, 1996). At around 5 years, children produce complex scripts and the number of words produced can reach 1500 token words in a controlled play setting, suggesting high levels of verbal fluency and richness of vocabulary, including the entire paradigm of function and closed-class words (e.g., personal pronouns, articles, tense markers, etc.). It is quite evident that none of the SPE-LF children reaches normal levels by 7 years of age. They probably do not make good use of the elements or component parts of syntax and may also have lexical access problems.
<table>
<thead>
<tr>
<th>MLU/Lexical diversity</th>
<th>SPE-LF subject 1</th>
<th>Control subject</th>
</tr>
</thead>
<tbody>
<tr>
<td>33 months</td>
<td></td>
<td></td>
</tr>
<tr>
<td>1.7/59</td>
<td>3.2/105</td>
<td></td>
</tr>
<tr>
<td>Un peigne (A comb)</td>
<td>C’est un renard (It’s a fox)</td>
<td></td>
</tr>
<tr>
<td>Voilà fermé (There closed)</td>
<td>Trouvé une assiette (Found a plate)</td>
<td></td>
</tr>
<tr>
<td>45 months</td>
<td></td>
<td></td>
</tr>
<tr>
<td>1.9/79</td>
<td>4.1/174</td>
<td></td>
</tr>
<tr>
<td>Escalier cassé (Staircase broken)</td>
<td>Je mets là pour l’instant (I put there for now)</td>
<td></td>
</tr>
<tr>
<td>Encore un (One more)</td>
<td>Il aura chaud aux pieds (He’ll be warm in the feet)</td>
<td></td>
</tr>
<tr>
<td>J’sais pas (I don’t know)</td>
<td>C’est l’histoire d’un marchand (This is the tale of a trader)</td>
<td></td>
</tr>
<tr>
<td>Mettre un comme ça (Place one like that)</td>
<td>T’appuies sur les jambes et sur la tête et tu vois ça ouvre ici (You press on the legs and on the head and see it opens here)</td>
<td></td>
</tr>
<tr>
<td>Je crois que j’ai trouvé (I think I’ve found)</td>
<td>Elle approche et elle entre dans la maison (She comes near and she enters the house)</td>
<td></td>
</tr>
<tr>
<td>Le singe est tombé (The monkey fell)</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Mean length of utterance is an index of syntactic maturity. In normal children, the classical MLU burst at 45 months signals that the child is now producing sentences longer than three words long, is now able to set the morphological parameters constitutive of grammar and is also able to share the common features of its social linguistic community. SPE-LF children, at a late age, are still frozen at the one- to two-word utterances.

In summary, this study represents one of the first exhaustive, longitudinal attempts to document language acquisition following cerebral dysfunction in early childhood. The data first revealed that the impact of unilateral focal epileptic disturbance produces effects comparable to those with focal brain injury. In this case, early epileptic damage to the anterior area of the left hemisphere, a brain region already noted for its importance in speech and language, is associated with severe and lasting deficits in linguistic production only. There is a point of contrast, however, between the consequences of epileptic and focal injury on the development of language. If children
with focal posterior left lesions are most delayed in production of speech and onset of single words, there is apparently no consistent mapping of lesion site onto production breakdowns at later stages. Second, language acquisition in SPE-LF is not uniform. If marked deficits in all aspects of language are initially characteristic of this neurogenic disorder, there is gradual recovery in linguistic comprehension only. Production levels, even at later stages, remain well below those of very young normal children. Whether this pattern of deficits is still observed at later stages in the development of these children remains to be determined.

REFERENCES

Deonna, T., Fletcher, P., & Voumard., C. 1982. Temporary regression during language acquisi-


