Perception and comprehension of linguistic & affective prosody in children with Landau-Kleffner syndrome

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Introduction

Landau-Kleffner syndrome, aka acquired epileptic aphasia in children, is a type of epilepsy in which the most characteristic symptom is word deafness (aphasia) evolving into auditory agnosia and/or mutism. Since Landau-Kleffner syndrome was discovered by Dr. Landau and Dr. Kleffner in 1957 [1], there have been some case studies examining language outcomes in children suffering from this disease [2], [3], [4], [5], [6]. But most studies tested vocabulary in the context of short-term phonological memory. To our knowledge, there have been no investigations of the use of prosodic cues in children with Landau-Kleffner syndrome. We thus tested oral language perception/comprehension with linguistic and affective prosodic cues: rhythmic *vs* arrhythmic sentences and neutral *vs* anger sentences. With regard to linguistic prosody, we chose not to use the intonation pattern that distinguishes statements from questions because a) rhythm – the temporal organization of the prominences [7] – is also a very important prosodic element, b) most tests of prosody use this pattern [8], [9], [10], and c) more subtle prosodic elements need to be tested than statement *vs* question pattern in order to better examine language capacity in children who have already undergone speech-language therapies. With regard to affective prosody, the emotion anger was chosen for its precocity (even a baby gets angry when he doesn't get what he wants immediately whereas we cannot be sure of sadness, for example, in a baby) and salient prosodic features compared with neutral. Given the clinical manifestation of LKS (word deafness), it is hypothesized that children with LKS would have fewer difficulties than healthy subjects in perceiving delexicalized sentences with affective prosody, i.e. sentences without semantic information.

Materials and methods

1. Subjects

Twenty-two healthy children (M.A.: 7;7) in CE1 (2nd year of elementary school), twelve healthy adults (M.A.: 26;3), two children (9;9 and 8;4) and an adolescent (16;3) with Landau-Kleffner syndrome participated in this study. All participants were native French speakers and had normal hearing. None of the healthy subjects had any known history of audiological or neurological impairments. None of LKS participants had autism (one third of epileptics have autism) [11].

2. Stimuli

We used auditory visuo-motor tasks that consisted of auditory stimuli and visuo-motor responses for our protocol.

Our battery of tests consists of two sets of tests of prosody: the first set focuses on linguistic prosody and the second on affective prosody. The first set of tests includes two groups of sentences, one delexicalized (with unintelligible lexical information) and the other non-delexicalized (with intelligible lexical information). Each of these sentence groups is subdivided into two subgroups, one with rhythm and the other without rhythm (no prominences). The second set of tests also comprises two groups of sentences, one delexicalized and the other non-delexicalized. Each of these two groups is subdivided into two subgroups, one with sentences of neutral emotion prosody and the other with sentences containing prosodic cues for anger.

2.1. Auditory stimuli

The sentences were constructed using Novlex, a French lexical database for 7 to 8 year-old children. Novlex provides estimates of the lexical extent and frequency of the written vocabulary for primary students. Proper nouns, numbers, country names and onomatopoeias are not included in the database. The words used in our protocol have a frequency greater than 3000 per 100 million and each word has fewer than three syllables. The syntactic structures are either S-V-O or S-V-Adv without relative or conjunction clauses, and the sentences have from 6 up to 13 syllables. The tenses used in our stimuli are the present, the compound past and the imperfect. To evaluate their suitability for the target age group, three 7 and 8 year-old children were asked to judge the sentences by choosing one of the following five adjectives: comprehensible, difficile (difficult), rigolote (funny), bizarre and mal dite (incorrect). For the last choice, they were asked "Comment on le dit? (How would you say it)?" for a possible correction. The 140 selected sentences (90 neutral sentences and 50 anger sentences) were read by a 30 year-old actress and recorded with Apple Soundtrack Pro.

Signal processing was carried out with Praat version 5.1.03. To remove lexical (and thus semantic and syntactic) information, the fundamental frequency of the original signal was estimated and delexicalized in the form of sinusoids. As a result, the final stimulus was a pure sound which follows the structure of the original fundamental frequency. Certain artefacts resulting from delexicalization were eliminated by applying a low-pass filter at 400 Hz. To remove prosodic rhythm (the temporal organization of the prominences of each sentence), the duration of each syllable in all stimuli was modified so that each had the same duration (mean duration of syllables). Finally, all sentences were normalized in intensity to an equivalent RMS value.

2.2. Visuo-motor response mode

Responses involved multiple-choice among drawings format (visual) and a with a button-press responses (motor). This type of response mode does not involve a heavy cognitive load thanks to the absence of written language, and this absence allows us to evaluate uniquely oral language perception/comprehension.

Four images were proposed to subjects for each stimulus of linguistic prosody with semantic information. Each participant had to find the image depicting the sentence he/she had just listened to. The target words (subject & object) of the sentence were alternated with others (cf. Figure 1).

Figure 1. 'L'ours goûte le miel.'
(The bear tastes honey.)

For the tests of linguistic prosody tasks without semantic information in which the delexicalized sentences were either prosodic/rhythmic or aprosodic/arrhythmic, the children were asked to judge the sentences as being either flat (plates) or irregular (avec des pics vers le haut ou vers le bas)[1] in their sound. To respond, subjects were asked to choose one of two drawings of line: one with peaks, the other was dotted and flat representing the patterns of each type of sentences. The position of the two lines was alternated (cf. Figure 2).

For the tests of affective prosody either with or without semantic information, we used two smilies of which one depicted a neutral expression, the other an angry one. These two emotions are expressed by the form of the mouth and the eyebrows. The position of the two smilies was alternated (cf. Figure 2).

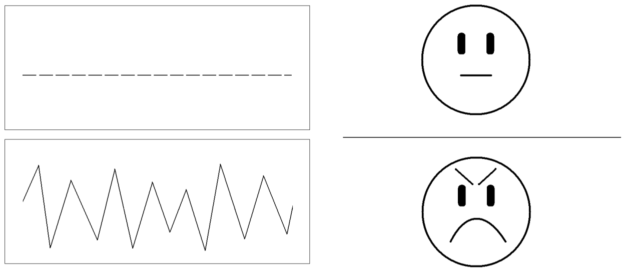

Figure 2. Lines for linguistic prosody (Left) & Smilies for affective prosody (Right)

Subjects responded by pressing buttons that had been placed over the four keys on a USB numeric pad (7, 9, 1 & 3 for the tests with 4 images, and 9 & 3 for the tests with 2 images). The other keys were covered with a board. The location of each button corresponded to that of each image.

3. Procedure

Children were screened with a pure tone audiometry in order to identify any auditory disorder. Only children without auditory deficits participated in this study. The stimuli were delivered using Presentation® on a Windows® XP interface. We administered randomly four series of tests (LP SEM+, LP SEM-, AP SEM+ & AP SEM-)[2]. Each series had thirty stimuli which included two sub-tests (cf. Table 1).

Linguistic prosody				Affective prosody			
with semantics		without semantics		with semantics		without semantics	
R+	R-	R+	R-	N	A	N	A

R+ : with rhythm N : neutral
R- : without rhythm A : anger

Table 1. Structure of the battery of tests

4. Analysis

The raw response data recorded using the Presentation® platform was statistically analyzed using SPSS and Excel. Independent two-sample t-test (Levene's test) was used to compare typically developing children (TDC) group with healthy adults. Paired sample t-tests were used in order to verify any discrepancy between two conditions within the same group. For each LKS child we calculated averages in different conditions and then compared them with the means of TDC +/- 2 standard deviations ($p<0.05$). A group comparison of TDC and LKS was not possible given the small number of subjects (n=3). We also used hierarchical clustering analysis (dendrogram) with 4 different types of distance (Ward, barycentre,

[1] Explained with example sounds & hand motions

[2] LP = linguistic prosody, AP = affective prosody

complete & median) to evaluate the distance between participants.

Results

1. Healthy children vs Adults

Levene's test demonstrated no significant difference in LP SEM+R+, LP SEM+R- and AP SEM+N. The rest of the tests had significant differences between the two groups (cf. Table 2). When there is no semantic information, children's performance is at chance level.

Linguistic Prosody (LP)			
SEM+R+	SEM+R-	SEM-R+	SEM-R-
N.S.	N.S.	0.001	0.004
Affective Prosody (AP)			
SEM+N	SEM+A	SEM-N	SEM-A
N.S.	0.004	0.006	0.026

Table 2. TDC *vs* Adults (Levene's test)

In the TDC group no discrepancy was found in the paired sample t-tests. As for adults, a difference appeared between AP SEM-N and AP SEM-A (p=0.009) but no discrepancy between LP SEM+R+ and LP SEM+R-, LP SEM-R+ and LP SEM-R-, and AP SEM+N and AP SEM+A.

2. LKS group

J (9;9) and A(8;4) were compared with TDC, and T (16;3) was compared with adults. Participant J's performance fell within 2 SD of the one of TDC group. Participant A showed better performance than the TDC in AP SEM-A with over 2 SD distance and a trend in AP SEM-N. It was notable that for T scores were much lower than that of adults' one with over 2 SD distance in LP SEM-R-, AP SEM+A, AP SEM-N & AP SEM-A. In fact, his performance was comparable with TDC rather than adults' one. The dendrogram shows that T was far away from adults whatever the clustering method we used (cf. Figure 3).

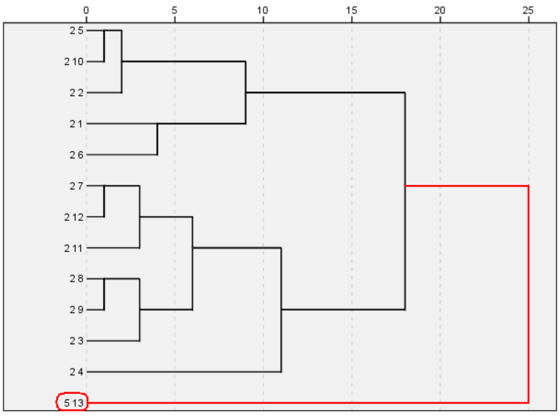

Figure 3. Dendrogram of adults using Ward's distance, T. is encircled in red

All participants were better at SEM+ tasks with varying degrees in both prosody tests (cf. Figure 4). Adults (1) maintained high scores with very little difference in both LP and AP tests whereas other participants' score (2, 3 & 5) dramatically dropped in SEM- tasks except A. (4) whose high score was maintained in AP tests.

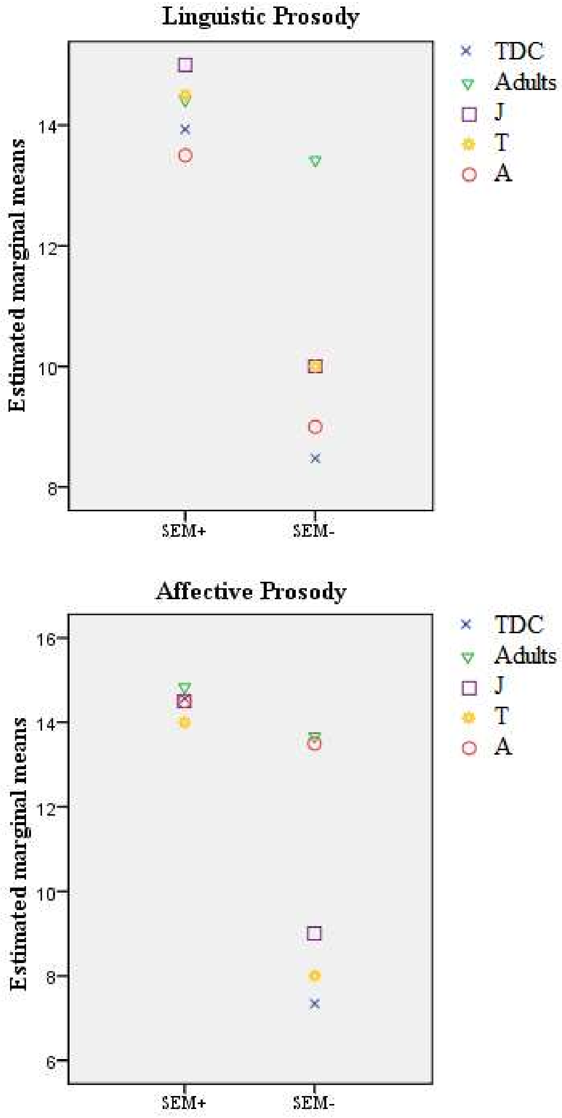

Figure 4. Estimated marginal means of scores
SEM+ = with semantic information
SEM- = without semantic information

Discussion

Healthy children at the age of 7 to 8 showed chance level performance in SEM- tasks. This may be due to specificity of language development in children [12]: The function of language for children is to indicate an intention to 'make do things with words'. They emphasize the use of language rather than its form, in other words, it is rather the communicative function of language that interests children than language itself. In the absence of communicative function, children cannot identify emotions. It could be argued that children acquire lexicon, semantics, syntax, pragmatics, etc. at the expense of primary elements of language such as prosodic features that they had used before they started speaking. These primary elements of language will resurface when children will be able to manipulate correctly lexicon, semantics, syntax, pragmatics, etc. Once one has mastered arbitrarily coded semantics and syntax, he has no trouble in understanding the relational aspects of language and its paralinguistic forms [13]. Adults can distinguish emotions without semantic information because they can apply elements of other situations to the context of tests. Besides, it is no longer only the communicative function of language that they are interested in, but they can 'manipulate' its form. That is why we observe good performance in adults regardless of the task.

Our study did not show any significant difference between conditions in the healthy children group. Children at the age of 7 to 8 use contents (semantic/lexical information) rather than paralinguistic cues in oral comprehension [14]. This fact can explain the reason of paired sample t-test results in the typically developing children group. In the adults group, only between AP SEM-N and AP SEM-A was found a significant difference (p=0.009). When some anger sentences were not strongly enough imbued with anger characteristics, adults judged them neutral for lack of semantic information. The fact that no discrepancy was found between AP SEM+N and AP SEM+A may be due to the ceiling effect observed in this group (14.67/15 for N *vs* 15/15 for A).
Participant J has performance as good as TDC (M.A. 7 years 7 months), but given his age, 9 years 9 months, we cannot rule out slightly delayed language development. Since he is still getting speech-language therapy weekly, this possible delay could get remediated.
As for the participant T, in spite of his age (16;3) he had quite poor performance comparable with that of 7-8 year-old children. He had obvious difficulties in perceiving prosodic cues. This may be due to the premature termination of speech-language therapy: T stopped all therapies at the age of 11. This case may demonstrate the importance of long-term speech-language therapies for children with LKS.
The results from A confirm our hypothesis: her performance was as good as that of adults in AP SEM-tasks. In contrast she was comparable with typically developing children in the rest of our tests. Participant A has recovered from LKS for 2 years, her recovery is more recent than J's and T's. Her recent recovery may explain the persistence of affective prosody. We hypothesized that children with LKS would use prosodic cues to understand others in their everyday life while they are aphasic. These primary elements of language would be the only way that they rely on to try to be in oral communication.

Future research

Although one participant's results could confirm our hypothesis, it doesn't allow generalization or deeper understanding on language outcomes in children with LKS. In order to verify our hypothesis – the primary elements of language (prosody) would persist in children with Landau-Kleffner syndrome and they would use the prosodic cues for communication, it will be interesting to test children with LKS either in their aphasic phase or shortly after their recovery. The results obtained from our test battery could allow us to better understand language development in children and it could be helpful for speech-language therapists to have another type of check-up of language capacity to better frame therapies for patients.
We initially included a group of children with ASD with an opposite hypothesis to the one of this study considering their clinical manifestation (problems in understanding others' emotions): children with ASD would be better at linguistic prosody tasks with semantic information. Data analysis of this group is ongoing.

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