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The Neural Correlates of Childhood Linguistic Isolation

Naja Ferjan Ramírez, Matthew K. Leonard, Eric Halgren, and Rachel I. Mayberry

1. Introduction

Compared to adults, young children have a remarkable ability to rapidly acquire language in all its breadth and complexity. Given adults' superior cognitive skills, the question is why. Is it the case that the neural representation of language is different when acquisition begins at a later age? The idea that the outcome of language acquisition is dependent on the age when the learning begins is known as the critical period for language. The existence of critical period phenomena is well documented within the animal kingdom. An example is learning the species-typical song in songbirds, which is dependent on exposure to conspecific song during a critical temporal window (Marler, 1970). Because almost all typically developing hearing children are deeply immersed in language from birth, however, the critical period hypothesis for language is difficult to test.

Unlike the acquisition of spoken languages, the acquisition of sign languages by deaf children is frequently delayed for a variety of reasons. In the USA, about 90% of deaf children are at risk for language delay because they are born to hearing parents who do not know any sign language (Schein, 1989). In the United States, deaf children typically receive special services by preschool age or younger. In rare cases in the USA and more often in Latin American countries where special services can be non-existent (Senghas & Coppola, 2001), deaf children may not be exposed to any language until adolescence. Such children lack access to spoken or sign language due to variety of life circumstances and a lack of schooling. Sometimes when such cases are discovered, attempts are made to provide them with sign language through immersion. These rare circumstances of language development offer a unique

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[*] All authors at University of California, San Diego. Correspondence should be addressed to Naja Ferjan Ramirez, Department of Linguistics, University of California, San Diego, 9500 Gilman Drive, La Jolla, CA, 92093-0108 (email: naja@ling.ucsd.edu). This research was supported in part by NSF grant BCS-0924539, NIH grant T-32 DC00041, an innovative research award from the Kavli Institute for Mind and Brain, and NIH grant R01DC012797. The content is solely the responsibility of the authors and does not necessarily represent the official views of the National Institutes of Health. An expanded version of the results summarized here is reported in a manuscript currently under review at Cerebral Cortex.

opportunity to study the effects of late language exposure on the neural representation of language.

Here we studied the brain correlates of lexico-semantic processing of American Sign Language (ASL) in two deaf adolescents who experienced no language throughout childhood until becoming fully immersed in ASL at around 14 years. At the onset of the present study, they had two to three years of ASL experience. Cases with such unique developmental histories have never before been neuroimaged. Due to their limited language skills, combined with a lack of previous neuroimaging research with this population, designing a neuroimaging study posed a series of challenges. The aim of this paper is to describe the experimental protocol we developed to study the neural representation of language along with a description of anatomically constrained magnetoencephalography (aMEG), a relatively novel approach to studying the dynamics of language processing. Prior to this work, only one study (Levänen, Uutela, Salenius, & Hari, 2001) had used MEG to study sign language processing. Here we describe the protocol that we developed to study the dynamics of sign language processing (see also Leonard & Ferjan Ramirez, Torres, Travis, Hatrak, Mayberry & Halgren, 2012) using aMEG (Dale, Liu, Fischl, Lewine, Buckner, Belliveau, & Halgren, 2000).

1.1. Previous research

Numerous behavioral studies have shown that delayed age of onset of sign language acquisition has severe and long-term negative effects on language processing in adulthood (Mayberry & Eichen, 1991; Mayberry & Fischer, 1989; Newport, 1990). These age of acquisition (AoA) effects are found in sentence and narrative comprehension, as well as in single word processing (Boudreault & Mayberry, 2006; Mayberry & Eichen, 1991; Mayberry & Fischer, 1989; Newport, 1990). For example, Mayberry and Eichen (1991) used a sentence recall task and found that AoA had significant effects on performance at all levels of linguistic structure. More recently, these psycholinguistic effects have been linked to atypical neural activation patterns in response to language stimuli. One fMRI study showed that older age of L1 acquisition is associated with increased hemodynamic activity in posterior perceptual areas and decreased activity in the classical language areas (Mayberry, Chen, Witcher, & Klein, 2011). Participants in these behavioral and neuroimaging studies were deaf individuals who varied in the age of initial exposure to sign language, but were all adult life-long users of sign language who went to school at the typical age of 5 years.

The neural correlates of language in deaf adolescents without effective language exposure and little schooling have not previously been studied. Behavioral studies show that when such cases become immersed in a sign language, they quickly replace gestures with signs, which indicates that the human sensitivity to linguistic input is not lost even after such extreme delays in language exposure (Berk & Lillo-Martin, 2012; Emmorey, Grant, & Ewan, 1994; Ferjan Ramirez, Lieberman, & Mayberry, 2012; Morford, 2003). What is currently unknown is how they represent their newly acquired signs in the brain.

This question is related to the critical period hypothesis, which posits that the outcome of language acquisition is dependent on the age when the learning begins (Lenneberg, 1967). Today most researchers believe that there is an association between early brain development and language acquisition; however, the exact nature of this relationship is poorly understood due to a lack direct evidence. Previous studies have looked at second language (L2) learners, because the age of L2 acquisition varies widely in the population. These studies typically ask whether L1 and L2 activate overlapping or distinct brain areas and the findings generally indicate that the L2 is processed through neural networks similar to those that support the L1, with some additional activations in the bilateral posterior and right hemisphere frontal areas (for review see Abutalebi, 2008). It is important to note, however, that any L2 learner has, by definition, acquired an L1 well within the critical period. These studies thus do not answer the question of how the human brain responds to a lack of childhood language experience.

To address this question, we studied the neural representation of L1 in Shawna and Carlos (pseudonyms), two adolescent first-language learners who had, at the onset of the study, been learning ASL for a period of 2 and 3 years (for more information on their background, see Section 2.1.). Below we provide a detailed description of the methods we developed to study the neural correlates of language in the two cases.

2. Methods

2.1. Cases

The cases' backgrounds and ASL skills have been described elsewhere (see Ferjan Ramirez, Lieberman, & Mayberry, 2011; Ferjan Ramirez et al, 2012). Briefly, Shawna and Carlos were born deaf and have begun to acquire ASL at age ~14 years through immersion at home and at school. Prior to immersion, Shawna lived with hearing guardians who did not use any sign language. She was reportedly kept at home and not sent to school. Carlos moved to the USA from another country at age 11. The members of his biological family were all hearing. He reportedly enrolled in a deaf school for a brief time of a few months. Upon arriving to the USA he was misplaced in a classroom for mentally retarded children where the use of sign language was limited. Prior to the onset of ASL immersion at age 14, the two cases used behavior and limited gesture to communicate. Carlos knew a few ASL signs, and Shawna did not know any. Neither case had knowledge of spoken language and both were illiterate. For more information regarding the cases' language skills, see section 2.4. and Ferjan Ramirez et al, 2011, 2012.

2.2. Control groups

The technique used in the current study, aMEG, has been used extensively to characterize the spatiotemporal dynamics of spoken and written words (Dale et al, 2000; Halgren, Dhond, Christensen, VanPetten, Marinkovic, Lewine & Dale, 2002; Marinkovic, Dhond, Dale, Glessner, Carr, & Halgren, 2003; Leonard, Brown, Travis, Gharapetian, Hagler, Dale, Elman, & Halgren, 2010; Travis, Leonard, Brown, Hagler, Curran, Dale, Elman, Halgren, 2011; Travis, Leonard, Chan, Torres, Sizemore, Qu, Eskandar, Dale, Elman, Cash, & Halgren, 2012). These studies found an M400 component, corresponding to the N400 component that has been studied with electroencephalography (EEG), and is associated with lexico-semantic encoding. Since it was unknown if signed words also evoke an M400, and if so, the location and latency of its neural generators, our first step was to conduct an aMEG study of ASL sign processing in a group of deaf native signers (see Leonard & Ferjan Ramirez et al., 2012). This participant group was studied in order to establish how single ASL signs are processed in the brains of those deaf individuals who learned ASL from birth. We found that their neural representation of ASL signs was highly similar to the representation of auditory English words in hearing native speakers of English.

The second control group we studied was hearing young adults who learned ASL as a second language in college (L2 learners; Leonard & Ferjan Ramirez, Torres, Hatrak, Mayberry & Halgren, under review). The participants in this second control group, like Shawna and Carlos, were young adult learners with relatively limited ASL experience (i.e. they only experienced between 40 and 50 weeks of classroom ASL instruction). Crucially, and unlike Shawna and Carlos, these controls acquired an L1 (English) from birth. These individuals also showed an M400 with fairly typical latency and location, but with additional areas as is seen in subjects who are learning a second spoken language (e.g., hearing Spanish-English bilinguals- Leonard et al., 2010).

2.3. Anatomically constrained magnetoencephalography

Previous studies using aMEG have demonstrated that it is sensitive enough to characterize the time course of neural activity in the dynamic brain network underlying both written and auditory language processing (Dale et al, 2000; Halgren et al, 2002; Marinkovic et al, 2003; Leonard et al, 2010; Travis et al, 2012). Moreover, aMEG has been used successfully with 12-18 month old infants (Travis et al, 2011), which suggests that it is suitable for use with participants who are just beginning to learn a first language.

aMEG presents a number of important advantages over other methods that have so far been used to study the neural correlates of sign language processing. The MEG signal is generated directly by the electric currents in the apical dendrites of cortical pyramidal cells, which are continuations of the trans-synaptic current flows that are the core mechanism of neuronal information processing (Dale and Halgren, 2001). The transmission of MEG signal within the brain to the sensors is effectively instantaneous (like the transmission of the EEG signal). Hemodynamic methods (like fMRI or PET), on the other hand, reflect an indirect measure of the electrical activity of neurons, as they rely on vascular changes that take place over seconds. Their time scale is thus much longer than the speed of on-line language processing, which takes place at the millisecond scale (see Marinkovic, 2004). Using a method with such high temporal resolution allows us to resolve specific stages of single word processing (for example, early perceptual processing, which takes place in the first 100 ms post stimulus onset, *vs* lexico-semantic encoding, which occurs between 200 and 600 ms post stimulus onset) and presents an important advantage over hemodynamic methods. The slow response measured by fMRI or PET may be insufficient to capture this dynamic, and could thus obscure some important differences between different subject populations, or between the different stages in word processing as they unfold across the brain on a millisecond scale (See Bar, Kassam, Ghuman, Boshyan, Schmidt, Dale, Hamalainen, Marinkovic, Schater, Rosen, & Halgren, 2006; Thesen, McDonald, Carlson, Doyle, Cash, Sherfey, Felsovalyi, Girard, Barr, Devinsky, Kuzniecky, & Halgren, 2012; Travis et al, 2012).

While the hemodynamic methods cannot accurately reflect the timing of language processing, their spatial resolution is excellent. The estimation of the source of MEG (or EEG) activity, on the other hand, is always somewhat uncertain due to the so-called inverse problem: for any distribution of MEG (or EEG) signal recorded on the scalp, there are infinitely many possible configurations of current sources and sinks within the brain that are consistent with the recording. To disambiguate the inverse problem, we analyze the MEG data by imposing constraints based on anatomical and physiological information derived from MRI. The apical dendritic currents generating MEG signals are in the cortex and are oriented perpendicularly to its surface (Nunez, 1981). Once we acquire each subject's MRI and reconstruct the exact shape of their cortical surface, the MEG solution space can be greatly reduced, leading to aMEG (Dale and Sereno, 1993). These solutions have been validated by comparison with direct intracranial recordings (Halgren, Baudena, Heit, Clarke, Marinkovic, 1994a; Halgren, Baudena, Heit, Clarke, Marinkovic, 1994b; Smith, Stapleton & Halgren, 1986; McCarthy, Nobre, Bentin, & Spencer, 1995). For a more detailed description of the aMEG procedure, see Dale & Halgren, 2001.

For the purposes of this study, we developed a protocol that was based on studies of spoken and written language, and pilot work with the two cases and other deaf individuals outside the scanner.

2.4. Stimuli

Because we are studying single cases (not participant groups), our first task was to prepare a task with a large number of trials to decreases the signal-to-noise ratio (SNR) and obtain meaningful data at the level of single subjects. The signal related to the processing of a signs is consistent across trials, while the noise is distributed randomly. If many instances of responses to signs are added together, then the signal will cumulate, and the noise will gradually cancel out.

If the signal is truly invariant and the noise truly random, the SNR improves by the √n, where n is the number of trials. A large number of trials therefore effectively decreases the SNR. Our previous work with MEG sensor-level and dSPM analyses suggest than *n*=150-200 per condition is sufficient to capture clean and reliable single subject responses. Given the uniqueness of the two cases, and the fact that we initially were not certain whether they would be able to provide that many artifact-free trials without breaks, we decided to design a task with 600 trials (300 per condition), which gave us sufficient flexibility.

Given the cases' limited experience with ASL, creating a large number of stimuli was a challenge. To learn as much about the language development of the cases as possible, we first conducted a study of the cases' language (Ferjan Ramirez et al, 2011; Ferjan Ramirez et al, 2012). Briefly, we used the MacArthur-Bates developmental inventory, a picture-naming task, and an analysis of spontaneous language samples. These analyses showed that Shawna and Carlos used almost exclusively ASL signs and almost no gesture to communicate. They were able to produce simple utterances that were generally 3 signs or less in length. The signs that they used were predominantly nouns, followed by predicates, and they used relatively few grammatical signs.

These language analyses yielded an approximation of the ASL signs they had acquired. From these signs, we selected a set of imageable nouns that we used as stimuli. Compound nouns and items that are typically fingerspelled were excluded. Stimuli signs were reviewed by 6 highly skilled ASL signers to ensure familiarity and accurate production. Signs were presented as video clips that were on average 515 ms long (range: 340ms-700ms). Each sign video began when all phonological parameters were in place, and ended when the movement was completed.

2.5. Task

Another requirement was to use a task that the cases could perform with high accuracy in the scanner. To prepare an appropriate experimental task, we conducted several pilot experiments outside the scanner. For example, we showed them line drawings of common objects and asked them to make semantic decisions about them (does the picture depict an animate being?). Further, we tested their ability to make binary decisions about ASL signs and indicate their responses with button presses. Having conducted these pilot studies, we decided to use a paradigm that was used in a previous study of English processing in 12 to 18 month old infants (Travis et al, 2011). In that study, infants saw pictures of objects followed by auditory presentations of English words that were either congruent (dog-dog) or incongruent (dog-cookie) with the picture. In our task, a picture of an object was followed by a short video of an ASL sign that was either congruent or incongruent with the picture (Figure 1). Unlike the infants in the study by Travis and colleagues, the cases were asked to press a button when the word matched the picture. Since little is known regarding language acquisition and processing by adolescent first-language learners, this was important since it allowed us to confirm that the cases were paying attention to the stimuli and performing the task.

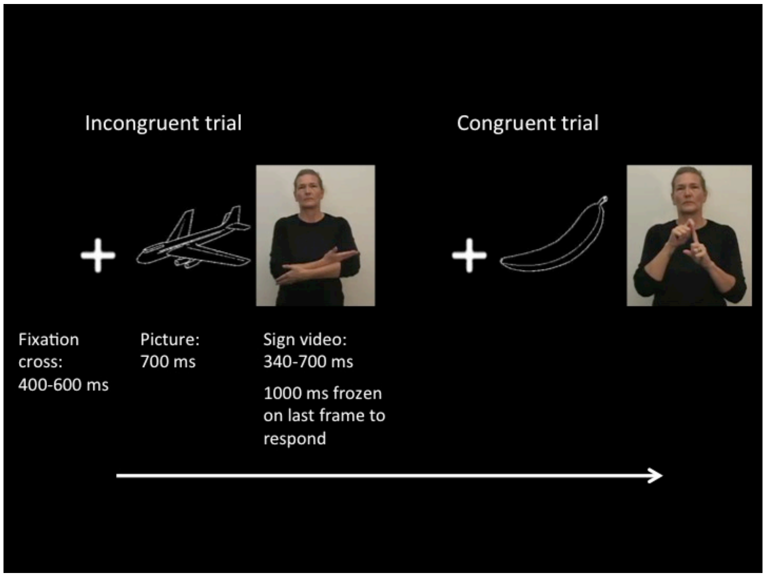

Figure 1: Task design: a picture was followed by a video of an ASL sign. Participants decided whether the sign matched the picture and indicated their response with a button press. Each picture and sign were presented in both the congruent and incongruent conditions.

The above-described task is a classical N400 paradigm based on decades of research on spoken and sign language processing (for a review, see Kutas and Federmeier, 2011). The N400 is a stimulus-evoked potential peaking at ~400 ms post stimulus onset which is modulated by the degree of difficulty of contextual integration, stimulus frequency and repetition, and other factors (Kutas & Hillyard, 1980). In our study, the N400 effect is elicited by presenting the ASL signs immediately following a picture that matches the meaning or not. The N400 to the matched stimuli is attenuated, and we localize this semantic effect in the brain using aMEG (see section 2.3.). Thus, although MEG allows us to visualize neural activity in response to signs as it unfolds over time, this initial study focuses on a narrow time-window and on a very specific stage of word processing (lexico-semantic encoding), i.e., where and when words are linked to their meanings.

2.6. Experimental procedure

In further testing we determined that the cases were able to understand and use all ASL signs that were used as stimuli and that they were able to recognize the pictures that preceded the signs. Our vocabulary selection criteria (section 2.4) yielded over 300 different stimuli signs for Carlos, and nearly as many for Shawna. Each sign was paired one time with a congruent picture, and one time with an incongruent picture (Figure 1), yielding several hundred trials for both Carlos and Shawna. Native signers and L2 signers also saw several hundred trials. Response hand was counterbalanced across blocks within participants. Each block required about 4 minutes of scanning time.

Prior to scanning, Shawna and Carlos participated in a separate acclimation session during which they practiced the task outside the scanner with flashcards and were able to familiarize themselves with the MEG and MRI scanners. On the day of scanning, all participants first performed a practice run with the researchers present in the MEG room. Following the practice block, the researchers left the room and testing began. After each block, the experimenters communicated with the participants and entered the room if any adjustments were necessary. The entire MEG session lasted between 30 and 45 minutes. Structural MRI was acquired immediately after MEG. Participants were allowed to watch a movie, rest, or sleep in the MRI scanner. For a detailed description of the MEG and MRI acquisition parameters, see Leonard & Ferjan Ramirez et al, 2012.

3. Application of the method to studying cases of adolescent L1 acquisition

The current study was designed to look at the processing of word meaning following childhood language deprivation. The specific question that we asked was whether lexico-semantic encoding in two adolescent L1 learners of ASL is different than in deaf native and hearing L2 ASL learners. The above-described experimental protocol turns out to be particularly well suited to study these questions. Both control groups performed the task with high accuracy and fast reaction times; most importantly, the same was true for the two adolescent L1 learners, despite their limited language skills. Shawna and Carlos performed within one standard deviation of the L2 control group in terms of accuracy, and showed fast latencies of discriminative responses. This result is important, as it allows us to conclude that the cases maintained attention throughout the scanning session.

aMEG results with native and L2 signers accord with previous studies on spoken and sign language processing (Abutalebi, 2008; Leonard et al, 2010; Marinkovic et al, 2003; MacSweeney, Capek, Campbell, & Woll, 2008; Mayberry et al, 2011; Petitto, Zatorre, Gauna, Nikelski, Dostie, & Evans, 2000). In native signers, meaning is extracted and integrated from signs using the same classical left hemisphere fronto-temporal network used for speech in hearing individuals (Leonard & Ferjan Ramirez et al, 2012). Furthermore, when ASL is acquired as an L2 in early adulthood, lexico-semantic processing activates the same left fronto-temporal language network, even when proficiency is relatively low (Leonard & Ferjan Ramirez et al, under review). These findings agree with previous sign language research using hemodynamic and lesion methods (Hickok, Bellugi, & Klima, 1996; MacSweeney et al, 2008; Mayberry et al, 2011; Petitto et al, 2000), but add a crucial temporal component to demonstrate that the similar neural substrate for sign and speech is driven by a similar temporal dynamics.

The neural correlates of lexico-semantic processing in Shawna and Carlos differ significantly from those in control groups. Consistent with the fact that they understood the meaning of the stimulus ASL lexical signs, they exhibited semantic modulation effects of comparable magnitude and timing to those observed in the control groups. However, the localization of this activity was different. In both cases, comprehension of lexical meaning localized predominantly to the right hemisphere superior parietal, anterior occipital, and dorsolateral prefrontal cortex, areas that were not activated in either one of the control groups, deaf native signers and hearing L2 signers. Also revealing is the finding that Shawna and Carlos's activity localization patterns did not look identical. For example, Shawna showed semantically modulated activity in the right frontal cortex, which was absent in Carlos. These differences may be a consequence of the variability in their non-linguistic communicative experiences prior to onset of language acquisition. Future longitudinal studies will show how their neural activations change as more language input is received. The current results show that a lack of language experience during childhood significantly affects the neural patterns associated with lexico-semantic processing.

4. Conclusions and perspectives

The goal of the current paper was to provide a description of the methods and procedures that we used to study the neural underpinnings of language in these two unique cases. Given how little is known about language acquisition and processing in very late, first-language learners, designing this study required significant investigation of the best fit of the cases' language skills to traditional neuroimaging techniques.

Our findings with both control groups are consistent with previous neuroimaging research on spoken, written, and signed language (Abutalebi, 2008; Leonard et al, 2010; Marinkovic et al, 2003; MacSweeney et al, 2008; Mayberry et al, 2011; Petitto et al, 2000). When language is acquired from birth, meaning is processed in the classical left-hemisphere fronto-temporal language network. However, when acquisition occurs for the first time at a later age, these activation patterns are significantly altered. The similarities between the cases were significant activations in the right hemisphere superior parietal areas, as well as in the posterior parieto-occipito-temporal areas bilaterally. Right superior parietal activations are known to be used in manual action-recognition in hearing participants (Buccino, Binkovski, Fink, Fadiga, Gallese, Seitz, Zilles, Rizzolatti, & Freund, 2001), but are typically not observed during sign recognition by deaf native signers. The cases' use of these neural action-recognition areas for sign processing might arise from an underdeveloped organization of the phonological and semantic features of signs. That is, the cases may have to overtly consider the visuo-motor forms of signs in order to recognize sign meaning.

Future longitudinal studies with Shawna, Carlos, and other deaf cases with similar backgrounds should allow us to better understand how the neural correlates of a late acquired L1 change as more and more language input is received. Combing behavioral and neuroimaging approaches described above, we hope to arrive at a better understanding of the mechanisms that drive language learning in infancy, as well as the causes behind the age-related decline in the ability to learn language.

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