Effects of Early Language Deprivation: Mapping between Brain and Behavioral Outcomes

Qi Cheng, Eric Halgren, and Rachel Mayberry

1. Introduction

One core issue regarding language development is the role of early language experience. The literature on child language development has found that the quality and quantity of early language input is associated with children's language performance, but the exact nature of the relation between early language experience and language development is less clear. Because language is ubiquitous in the environment, almost all children are immersed into a linguistically rich environment from birth. This makes it difficult to directly test the effects of a lack of language experience on subsequent language development. Here we present a study of individuals born deaf who experienced sparse language throughout childhood that will help illuminate the question.

Postnatal brain development follows genetically predetermined growth patterns, but is also strongly influenced by learning and environmental factors (Huttenlocher 2002). Nonetheless, little is known about how early linguistic experience may affect the establishment of the brain's dynamic language network. Studies on brain development suggest that the language network matures relatively late (Sowell et al. 2004; Lebel et al. 2012; Pujol et al. 2006), and that its degree of maturation correlates with language performance (Pujol et al. 2006; Mills, Coffe-Corina, and Neville 1997).

However, there are multiple possible explanations for the correlation between late establishment of the neural system and accompanying language development. One possibility is that this protracted development is solely due to late maturation alone, and, as a result, that this constrains language development in early years.

^{*}All authors at the University of California, San Diego. Correspondence should be addressed to Qi Cheng, University of California, San Diego, 9500 Gilman Drive #0108, La Jolla, CA 92093-0108 (email: qcheng@ucsd.edu). The research reported in this publication was supported in part by 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. We thank M. Hatrak and A. Roth for research assistance.

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Another possibility is that early development of the brain language system is gradually shaped by interacting with the linguistic environment. This would suggest that there is a plastic period during which language experience interacts with brain language system development. Again, because language is ubiquitous in the environment of infants, it is extremely difficult to distinguish between biological maturation and learning-dependent plasticity in typically developing children.

Rare cases who suffered from early language deprivation, such as Genie (Fromkin et al. 1974), have shown extra difficulties acquiring a first language (L1) later in life, suggesting the existence of a critical time window for language development. Unknown is whether this critical time window is linked in some way to development of the brain language system. Deaf individuals born into hearing families usually do not have access to either spoken or sign languages early in life, but are nurtured and cared for by their families. This situation of naturally occurring language deprivation offers a unique opportunity to tease apart the effects of early language experience on development of the brain language system. In particular, this study explores the microstructure of language-relevant white matter fibers to examine how anatomical outcomes may be associated with behavioral differences in language outcomes, especially at the morpho-syntactic level.

2. Background

2.1. Effects of early language deprivation

Postnatal brain development is not merely guided by genetics, but also shaped by environmental influences of many kinds. In addition, this early environmental sensitivity is often limited to a critical period. When crucial stimuli are missing during early brain development, this can result in irreversible deficiency of corresponding brain functions, as in vision (Hensch 2005). Studies on infant stroke and typical L1development indirectly suggest that there is a critical, or at least sensitive, period for language as well (Lenneberg, Chomsky, and Marx 1967). The case of Genie, who was isolated from language and social interaction until adolescence until the age of 13, is a rare direct example of how L1 deprivation may affect language development. After more than 5 years of language learning, Genie could use a variety of single words, and combine them to form simple phrases, but she struggled with most aspect of grammar (Fromkin et al. 1974). Because, cases like Genie are fortunately very rare, we cannot use them to discover the possible relation between language experience and brain development.

Examining critical period effects on language development is difficult, because babies are often born into an environment where language is omnipresent, and experimentally depriving babies from language is cruel and impossible. However, an abundant language environment would be inaccessible if it is not perceived. Almost 90% of deaf children are born into hearing families. Due to severe hearing loss, these deaf children usually have limited access to the language spoken by their parents. On the other hand, most hearing parents do not know how to use sign language, and often do not consider it as an alternative way to communicate with their deaf children. As a result, such deaf children can grow up with limited linguistic input, but still receive the nurturing necessary for social and cognitive development. The unique scenario of deaf children acquiring sign language as their first language after early childhood thus offers a rare opportunity to directly test the effects of early language deprivation on later language development.

Research has consistently found late L1 learners of sign languages to show divergent language outcomes compared with native signers for language performance, and to especially have deficits at the morphological and syntactic level (Mayberry and Lock 2003; Boudreault and Mayberry 2006; Newport 1990; Mayberry and Kluender 2017). These language outcomes are similar with other populations who suffer from decreased early language input, such as Genie (Fromkin et al. 1974), internationally adopted children (Scott, Roberts, and Glennen 2014), deaf individuals acquiring spoken language with compensated hearing (Grimshaw et al. 1998), and also late second language (L2) learners (Johnson and Newport 1989; Granena and Long 2013). In addition, difficulties at the morpho-syntactic level are also often observed among younger children with typical language development. This homogeneity in terms of the trajectory of language development across different populations suggests that there may be some uniform mechanisms of language learning dependent on early neural plasticity.

Recent studies on neural correlates of late L1 language outcomes serve as initial attempts to map from language deficits onto underlying neural mechanisms. Late L1 learners of sign languages show decreased activation of the classical language regions and increased activation in alternative regions (Ferjan Ramirez et al., 2016; Mayberry, Chen, Witcher and Klein 2011; Mayberry, Davenport, Roth and Halgren 2017). To date the only study examining at these effects at the anatomical level is Pénicaud et al. (2013). Using voxel-based morphometry (VBM), they found decreased gray matter concentration and increased white matter in occipital visual areas, but no differences with the core language regions. Due to the methodological constraints of VBM, however, certain micro-anatomical differences may not be detected. More research is required to link language and neural outcomes as a function of early language experience and deprivation.

2.2. Language-relevant pathways and behavioral associations

Long-range white matter fiber tracts play an important role in establishing the dynamic and distributed language network. Previous research on spoken

languages has proposed two information streams that are crucial for language processing, namely the ventral and dorsal pathways (Hickok and Poeppel 2004, 2007; Parker and Brorson 2005; Friederici 2009; Saur et al. 2008). The dorsal pathway consists of the superior longitudinal fasciculus (SLF) - arcuate fasciculus (AF) complex, connecting the inferior frontal gyrus (IFG) with the superior temporal gyrus/sulci (STG/STS) and the inferior parietal lobule (IPL), while the ventral stream runs through the extreme capsule (EmC) that links middle-posterior STG to the anterior IFG, the inferior fronto-occipital fasciculus (IFOF) that establishes the occipital-tempo-frontal connection, the inferior longitudinal fasciculus (ILF) connecting the occipital lobe and the temporal lobe, and the uncinate fasciculus (UF) connecting anterior temporal to inferior frontal areas (see Dick and Tremblay, 2012 for a review on the anatomy and functions of each fiber tract). According to the dual-stream model (Hickok and Poeppel 2004, 2007), the dorsal stream is mainly responsible for auditory-motor integration function, carrying acoustic speech signals from the auditory cortex into articulatory representations in the frontal lobe. In contrast, the ventral stream is more responsible for speech recognition, and involves structures in the superior and middle temporal lobe that are crucial for meaning and comprehension.

Compared to other white matter tracts in the brain, these language-relevant pathways, mainly connecting between temporal and frontal regions, often take longer to fully develop. Using structural magnetic resonance imaging (MRI), Pujol et al. (2006) noticed that language-related temporal and frontal regions in the left hemisphere show slower myelination development compared to sensorimotor regions. These findings have been confirmed by studies on white matter development across the lifespan (Lebel and Beaulieu 2011; Lebel et al. 2008). Bilateral frontal-temporal connections develop more slowly than others, especially for the language-relevant ILF, the SLF-AF complex, and the IFOF pathways. Similarly, Brauer, Anwander, Perani and Friederici (2013) found that children at age 7 continue to show immature AF-SLF and IFOF pathways compared with adults.

Studies have also found that the degree of white matter maturation is associated with children's language performance. According to Pujol et al. (2006), accelerated vocabulary development after 18 months is related to a rapid myelination phase in the language-related regions. As for language development beyond vocabulary, Skeide, Brauer and Friederici (2015) found that maturation of the AF-SLF pathways correlates with the ability to comprehend complex sentence structures in children aged 3 to 10 years old. However, as these studies only looked at children with typical language development, we do not know if these observations at both neural and behavioral levels are solely due to late maturation of these specific pathways, or actually reflect gradual development shaped by learning. Previous studies have found that dorsal pathways, especially the left AF, are associated with syntactic processing. Lesions in the AF-SLF complex often result in disturbance in syntactic comprehension as well as verbal working memory, while lexical-semantic knowledge is relatively spared (Caplan, Vanier, and Baker 1986; Wilson et al. 2011; Meyer et al. 2014). By contrast, lesions in relevant fibers in the ventral stream do not show such an effect on syntactic comprehension (Wilson et al., 2011). Fractional anisotropy (FA) of the AF-SLF complex, but not the ventral IFOF, is found to correlate with the behavioral performance (accuracy and reaction time) of comprehending complex sentence structures among children aged 3 to 10 years old (Skeide, Brauer and Friederici 2015). Decreases in FA of the AF-SLF are also found among people with specific language impairment (SLI), which correlates with their behavioral performance in syntactic comprehension tasks (Verhoeven et al. 2011).

It is not clear if deficits at the syntactic level are secondary to deficits in other lower-level functions mediated by the dorsal pathways, such as auditory-motor integration and working memory. Nevertheless, the literature consistently reports a double dissociation between syntactic processing and lexical-semantic processing, and deficits in syntactic processing are often associated with disrupted dorsal pathways.

Our knowledge of language-relevant white matter tracts is mostly based on studies on spoken language. Thus far few studies have explored the language pathways of deaf people who use sign language as their dominant language. Given that sign languages activate very similar brain regions, such as the STG/STS and the IFG, despite obvious modality differences (Neville et al. 1998; Petitto et al. 2000; MacSweeney et al. 2002; Leonard et al. 2012), it is possible that connections between these language regions are similar across signed and spoken languages. Comparing deaf and hearing populations, Kim, Park, Kim, Lee and Park (2009) found deficits in several fiber tracts, including SLF and IFOF tracts in the left hemisphere of deaf people, but other studies (Li et al. 2012; Hribar et al. 2014; Karns et al. 2017) found differences only within the auditory regions. Given that deaf people have very diverse language backgrounds, the inconsistent findings may be due to a confounding factor, namely age of language onset, as discussed in 2.1.

3. Current Study 3.1. Research questions

The current study aims to understand the effects of early language deprivation at the anatomical as well as behavioral level. Our first question is whether early language deprivation affects brain connectivity for language processing. In particular, we are interested in the dorsal AF pathway. Based on findings at the anatomical level, we map behavioral and anatomical outcomes of early language deprivation and propose underlying mechanisms of critical period effects in language development.

There are two main hypotheses regarding our first question. One possibility is that the late establishment of the neural language system is genetically guided and purely biological, and therefore does not require early environmental input to fully develop. If so, we would expect late L1 learners to show fully developed white matter tracts. Another possibility is that the establishment of the language network is not just biological maturation, but also reflects neural plasticity shaped by the interaction of postnatal brain development and early environmental input, thus requiring extended years of language experience within the time window of high-plasticity. If so, a lack of early language input will result in underdeveloped tracts.

Previous research has revealed selective deficits at the morpho-syntactic level as a result of early language deprivation, but its underlying causes remain unclear. If the findings support our first hypothesis, that anatomical development of the language network is purely biological and takes longer to mature, then this would suggest that learning complex syntactic structures later in life after brain maturation is somehow less favorable for reasons other than neural plasticity. For example, maturation of the frontal regions and the temporal-frontal pathway may have result in more top-down cognitive control, which can benefit performance but inhibit learning (Thompson-Schill and Ramscar 2009). On the other hand, if we find that late L1 learners do show differences in the anatomical organization of the brain language system, then this would indicate that the development of complex syntactic structures is contingent on anatomical structures that are shaped by early learning and brain plasticity. Missing the sensitive period would have irreversible effects on later language development.

3.2. Participants

Three deaf individuals participated in the current study. To protect their privacy, we will refer to them using pseudonyms Carlos, Shawna, and Martin. All three individuals were born profoundly deaf, grew up with hearing, non-signing family member(s) during childhood, and were mainly kept at home. As a result, they were all deprived from both spoken and sign language exposure during childhood.

Carlos was born into a hearing and non-signing family in another country and did not receive special services for deaf children, including schooling. He immigrated with some of his family members to the United States at age 11, and was first placed into a classroom for mentally challenged children. At age 13 years and 8 months, he was placed into a group home for deaf teenagers. According to social workers at the group home, Carlos knew very few signs at the time of placement, and mainly used pointing and gestures to communicate. By interacting with deaf fluent signers at the group home, he started acquiring a natural language, ASL, in an immersion setting. At the time of scanning, he was 16 years and 10 months old, with 3 years and 2 months of daily exposure to ASL. At the time of testing, his language skills remained limited. He was able to produce multi-sign utterances in ASL, but the majority were simple structures or fragments, and he seldom produced complex sentences. Similarly, he could comprehend simple sentences such as short sentences with basic word order, but failed to comprehend sentences with complex structures, such as relative clauses.

Shawna was raised by hearing and non-signing guardians. She had been kept at home until the age of 12, and had sporadically attended several schools, both deaf and mainstream, for a total of 16 months. At age 14 years and 7 months, she was placed into the same group home as Carlos. When she first joined the group home, she produced no ASL signs, and relied primarily on behavior and limited use of gestures to communicate. Like Carlos, she also started acquiring ASL by interacting with other deaf fluent signers at the group home. Shawna was 16 years and 9 months old at the time of scanning, with 2 years and 2 months of immersion daily exposure to ASL. Her language skills at the time of testing were also limited and showed characteristics similar to Carlos's language, with basic facility with simple sentence structures but extra difficulties with complex sentence structures.

Martin was born into a hearing and non-signing family in rural Mexico. He did not attend school until the age of 21 when he learned some Mexican Sign Language at a school for deaf children. He immigrated to the United States at age 23, where he started learning ASL by interacting with other deaf fluent signers in the local deaf community on a daily basis. At the time of scanning, he was 51 years old, with about 30 years of daily use of sign language. Despite extensive years of usage, his ASL skills were limited, especially with complex sentence structures such as relative clauses.

Previous studies have reported relevant language development and neural processing of these late learners in detail. Longitudinal vocabulary development of Carlos and Shawna can be found in Ramírez, Lieberman and Mayberry (2013). Ferjan Ramirez et al. (2016) reported longitudinal ASL lexico-semantic processing using aMEG for Carlos and Shawna, while Mayberry et al. (2017) reported Martin's neural activation using the same methods.

In order to compare late L1 learners with early L1 learners who experienced language from birth while controlling for language modality and hearing status, we also included a group of native signers as a control group.

3.3. Methods

3.3.1. Diffusion tensor imaging

MRI scans were performed at the UCSD Radiology Imaging Laboratory on a General Electric 1.5 Tesla EXCITE HD scanner with an eight-channel

phased-array head coil. Diffusion data were acquired using single-shot echo-planar imaging with isotropic 2.5 mm voxels (x pixel spacing 1.875mm, y pixel spacing 1.875mm, slice thickness 2.5 mm). One volume series was acquired with 30 diffusion gradient directions using b-value of 1000 s/mm2 (TE/TR 80.4ms/14300ms). Four pre-processing steps were performed on the acquired diffusion-weighted images using in-house MATLAB packages, including eddy current correction, motion correction, b0 distortion correction, and gradient non-linearity correction.

Diffusion tensors (DTs) were fitted using in-house MATLAB packages. We then calculated the FA value from the eigenvalues (length of the tensor axis), which measures the ratio between parallel and perpendicular diffusion. We used a probabilistic tract atlas to identify tracts of interest. Details of this automated white matter tracking method can be found in Hagler et al. (2009).

Based on previous studies, we selected two fiber tracts from the atlas as relevant long-range pathways for language, namely the temporal section of SLF, which is equivalent to the classical dorsal language pathway AF, and the IFOF, which is considered a crucial pathway for the ventral stream. We looked at these tracts in both right and left hemispheres.

3.3.2. Language testing

We also conducted a sentence-to-picture matching task in order to measure sentence comprehension skills of late L1 learners and the native signers. The signers were asked to first view a signed ASL sentence and then to choose a corresponding picture from three alternatives.

We included 14 ASL structures, ranging from simple to complex based on the number of clauses contained. Simple sentence structures are mono-clausal such as sentences with basic Subject-Verb-Object order, while complex sentence structures are either bi-clausal, such as relative clauses, or inter-sentential, such as wh-questions. Each structure was tested with 6 exemplars using vocabulary known to young children.

The 84 stimuli were randomly presented via computer which recorded accuracy and RT. Prior to the experiment, a screening task ascertained that vocabulary knowledge was not a performance factor.

3.4. Results

Our preliminary data suggests that, compared to native signers, all three late L1 learners showed lower FA in the left dorsal AF pathway, falling outside the native deaf signers' 1.75 interquartile range. Their FA values for this dorsal pathway in the right hemisphere also fall at the lower end of that of the deaf native signers, although not as much as in the left hemisphere. By contrast, the FA values of bilateral ventral IFOF pathways generally fall within the normal

range. As for language outcomes, consistent with the literature, all three late L1 learners could comprehend most simple sentence structures, with accuracy comparable with native signers, but their performance with complex sentence structures were at chance levels.

4. Discussion

In this study, we first asked if early language deprivation affects anatomical connectivity for the language network. Despite the limited number of late L1 learners, we did find effects of early language deprivation on one crucial language pathway, the left AF. Decreased FA indicates less structured directionality, which can be due to underdevelopment of the tract, or less myelination. These findings suggest that growth of the brain language pathways is not solely driven by biological maturation but require language acquisition during childhood. It appears that early language experience is crucial for the brain language system pathways to develop and connect in the expected fashion.

Equally important, these findings also shed light on potential mechanisms of age constraints on language learning. Previous studies have reported selective critical period effects on morpho-syntactically complex structures (Newport 1990; Mayberry and Lock 2003; Boudreault and Mayberry 2006) as well as decreased functional activation in several language regions (Ferjan Ramirez et al., 2016; Mayberry et al., 2011). Unresolved is the question of how language and neural outcomes were influenced by early language experience. Our current preliminary results indicate that late L1 learners who experienced language deprivation during childhood develop a less robust language neural system, especially in the dorsal stream, which is then reflected in their language comprehension performance, especially with complex sentence structures. Mapping late L1 learners' language-relevant anatomical characteristics with their language performance, our present findings confirm the double dissociation of structure and meaning reported in previous studies. In deaf late L1 learners, developmental deficits in the dorsal pathway are associated with syntactic processing difficulties. Our findings suggest that early language experience is crucial to complete development of the dorsal stream for language processing, enabling functional activations across various language regions, thus facilitating learning and processing of complex syntactic structures. A lack of linguistic experience during the critical time window of childhood appears to affect development of the dorsal stream in the left hemisphere, resulting in deficits in language outcomes, especially with morpho-syntactically complex structures.

5. Conclusion

The aim of the current study was to provide initial descriptions of an ongoing project. The aim of this project is to combine observations at both

neural and behavioral level to gain a unique perspective on the underlying mechanisms of age constraints on language learning. We examined white matter microstructures as well as language comprehension performance in two groups of deaf signers of ASL: native signers who had full access to sign language from birth, and the late L1 signers who had little access to any kind of language until puberty. We found that deaf late L1 signers showed decreased connectivity in a language-relevant white matter pathway along with deficits in comprehending complex sentence structures compared to deaf native signers. These findings suggest that early language experience, regardless of its modality, is crucial for the language system to fully develop in the expected fashion. More late L1 learners are required to confirm these preliminary findings, and more aspects of brain and language outcomes need to be explored to expand our preliminary findings.

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