Abstract

Objective: This study aimed to investigate the relationship between the change of language symptoms and the change of regional cerebral blood flow (rCBF) in the recovery process of two children with acquired aphasia caused by infarctions from Moyamoya disease with an onset age of 8 years.

Methods: We compared the results for the Standard Language Test of Aphasia (SLTA) with rCBF changes in 7 language regions in the left hemisphere and their homologous regions in the right hemisphere at 4 time points from 3 weeks for up to 5 years after the onset of aphasia, while controlling for the effect of age.

Results: In both cases, strong correlations were seen within a hemisphere between adjacent regions or regions that are connected by neuronal fibers, and between some language regions in the left hemisphere and their homologous regions in the right hemisphere. Conversely, there were differences between the two cases in the time course of rCBF changes during their recovery process.

Conclusion: Consistent with previous studies, the current study suggested that both hemispheres were involved in the long-term recovery of language symptoms in children with acquired aphasia. We suggest that the differences between both cases during their recovery process might be influenced by the brain states before aphasia, by which hemisphere was affected, and by the timing of the surgical revascularization procedure. However, the changes were observed in the data obtained for rCBF with strong correlations with the changes in language performance, so it is possible that rCBF could be used as a biomarker for language symptom changes.

Keywords: Children with acquired aphasia; Recovery process; Language symptoms; rCBF

1. Introduction

Acquired childhood aphasia is a language disorder caused by a cerebral lesion, even if a patient grows normally until pathogenesis [1]. Cerebral lesions causing...
aphasia include morphological and functional changes [2]. Morphological changes are said to be the seat of organic, structural, and anatomical lesions. Morphological changes include necrosis, atrophy, cicatrix, and slough, while functional changes include hypoperfusion and metabolic decline. One of the methods used to evaluate changes to cerebral function is single photon emission computed tomography (SPECT), which is an imaging test, that easily detects cerebral circulatory failure. This method provides information about focal cerebral ischemia that is consistent with the clinical symptoms, even immediately after pathogenesis, because it can detect regions with a reversible decline in cerebral blood flow (functional lesions) without organic changes, which cannot be discovered using morphological methods [3]. To date, functional neuroimaging diagnosis has depended on the experience of those interpreting the images, including some problems such as individual differences and reproducibility. However, the development of statistical imaging analysis, e.g., statistical parametric mapping (SPM), has resolved these problems and made it possible to examine whole brain circulation or metabolic changes with high accuracy [4–8].

Change in the distribution of cerebral blood circulation during the growth of children [9,10] has made it difficult to utilize SPECT in the field of acquired childhood aphasia for the purpose of pursuing the relationship between language symptoms and local reduction in cerebral blood flow. Ohnishi et al. [11] and Fukushima et al. [12] established a normal database (NDB) of blood flow that consisted of 3 age groups: 1–5, 6–10, and 11–15 years of age. This database was applied to the easy z-score imaging system, developed by Matsuda et al. [13,14], and statistical imaging analysis has since become widely used for the evaluation of cerebral blood flow SPECT in children.

Kojima et al. [15,16] reported case studies of acquired childhood aphasia, in which they used cerebral blood flow SPECT. Kojima et al. investigated changes in the results of the Standard Language Test of Aphasia (SLTA) [17] and regional cerebral blood flow (rCBF) after pathogenesis. They suggested that the left hemisphere is more important for early stage recovery of acquired childhood aphasia, and both hemispheres contribute equally to long-term recovery. However, rCBF changes during the process of acquired childhood aphasia are possibly affected by both age and the restoration of cerebral lesions. Therefore, we think that cerebral lesions should be observed after controlling for age-related rCBF changes.

In this study, we investigated the relationship between the change of language symptoms and the change of rCBF during the recovery process of 2 children with acquired aphasia with an onset age of 8 years. They underwent cerebral blood flow SPECT several times due to clinical necessity. Their language symptoms were evaluated using the SLTA, which was conducted at almost the same time as rCBF. We used the z-score to control for the effect of age.

2. Methods

2.1. Cases and assessment time points

Case A was a right-handed female child with an onset of aphasia at age 8 years and 4 months. At approximately 7 years and 9 months of age, she started to present with mild hemiplegia on the right side. A computed tomography examination found evidence of stroke in the left parietal watershed area, and she was diagnosed with Moyamoya disease. At the age of 8 years and 4 months, she had a stroke in the left frontal and temporal lobes following a surgical revascularization procedure of the left hemisphere, and she started showing symptoms of aphasia. After that, she underwent a surgical revascularization procedure of the right hemisphere at the age of 8 years and 10 months. Case B was a right-handed female child with an onset of aphasia at the age of 8 years and 5 months. She started experiencing numbness in all limbs and cataplexy at approximately 6 years of age. At the age of 8 years and 5 months, she sought medical attention after experiencing numbness in all limbs, headache, and a brief loss of consciousness. A head magnetic resonance imaging (MRI) exam at the time demonstrated flow voids in the basal ganglia, and small infarcts in the right frontal, right parietal, and left parietal lobes, and she was diagnosed with Moyamoya disease. At the age of 8 years and 5 months, she had a stroke in the tempo-parietal region including the left angular and supramarginal gyri following a surgical revascularization procedure of the right hemisphere, and developed symptoms of aphasia. There were no changes in her language symptoms following a subsequent surgical revascularization procedure of the left hemisphere at the age 8 years and 10 months. Both cases had surgery using encephalo-du-ro-arterio-syngiosis and encephalo-myo-syngiosis on both hemispheres. In Cases A and B, speech therapy was started at 3 and 6 days after onset, respectively. Since then, they have received speech therapy 1–2 times a week when they were in the hospital, and 1–2 times a month after being discharged.

For rCBF measurements, we used data from 99mTc-ethyl cysteinate dimer (99mTc-ECD) SPECT examinations at awake state that were conducted 4 times during the acute and chronic stages as part of the treatment process in both cases.

For language symptom assessment, we used data from the SLTA, which was administered at approximately the same time as the SPECT examinations. The largest interval between a SPECT examination and...
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