# A Potential Protective Effect in Multilingual Patients with Semantic Dementia: Two Case Reports of Patients Speaking Taiwanese and Japanese

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**Abstract-**

***Purpose:*** Several reports have suggested that multilingualism has a protective effect against semantic dementia. Here, we provide further evidence for this effect.

***Case Reports***

***First:*** The patient was a 75-year-old right-handed Taiwanese woman who had retired after working as a tailor. She was able to speak Taiwanese, Japanese and Mandarin Chinese fluently until 5 years ago. She gradually developed symptoms of profound anomia and difficulty with word-finding. Her mother tongue was Taiwanese and she had learned Japanese as her first symbolized language. She had used Mandarin Chinese for most of her life, but depended on Japanese to read and write (such as reading a newspaper and keeping accounts). However, she could now speak only very simple Taiwanese and Japanese, and could recognize only simple Japanese characters.

***Second:*** The patient was a 62-year-old right-handed man who had worked as an ironworker. He could speak Taiwanese and Mandarin Chinese fluently until 5 years ago. His mother tongue was Taiwanese. After 5 years of language deterioration, he was unable to communicate with his family members or recognize any characters, including numbers.

***SPECT results:*** Brain perfusion ECD SPECT (Tc-99m-ethyl cysteinate dimer single-photon emission computed tomography) showed less perfusion in the multilingual patient (Case #1) than in the bilingual patient (Case #2). Neuropsychological tests also demonstrated a slower rate of degeneration in the multilingual patient.

***Conclusion:*** We speculate that reading and writing in Japanese had a greater impact on the semantic system in Case #1. Thus, this patient showed relatively less degeneration or functional inactivity, as shown by perfusion in the frontal lobe, and this might be due to the persistent activation involved in multilingualism.

**Key words:** Multilingualism, bilingualism, semantic dementia, primary progressive dementia, neurodegenerative disease, cognitive reserve

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## INTRODUCTION

A multilingual or bilingual person is defined as someone who has spent the majority of their life regularly using at least 2 or 3 languages, at least from early childhood. More than half of the population in the world is considered to be bilingual. In the Netherlands, for example, about 80% of the population speaks English as a second language [1]. An association between bilingualism and language disorders or aphasia has been reported in previous studies, especially in patients with stroke or focal brain lesions [2], or Alzheimer's disease (AD) [3,4].

Disorders in a second language compared to the mother language in bilingualism can be viewed in terms of the semantic system in relation to the two languages. In this regard, assessing patients with semantic dementia (SD), in which semantic systems are deteriorated, is a good approach for understanding languages and the semantic system in bilingualism. Two studies reported that languages acquired after the mother tongue were more vulnerable to degenerative brain disease due to less intact semantic systems in SD [5] or primary progressive aphasia (PPA) [6]. Another case report of a Chinese-English bilingual woman with PPA showed that degenerations of the primary and secondary language were identical or may even be worse for the primary language [7]. However, she had used her second language (English) more frequently in her life and this language may have been better preserved for this reason.

In this preliminary report of two cases with a clinical diagnosis of SD, degeneration of the second language was more severe in both cases. However, one of the patients was multilingual while the other was only bilingual, which gave us the opportunity to observe the relative effects of multilingualism on semantic dementia.

## CASE REPORTS

### Case #1

The patient was a 75-year-old right-handed Taiwanese woman who had worked as a tailor before retirement. She was able to speak Taiwanese, Japanese and Mandarin Chinese fluently until 5 years ago. She gradually developed symptoms of profound anomia and difficulty with finding words. Her mother tongue was Taiwanese and she had learned Japanese as her first symbolized language at school, starting 5 years before WWII. She had used Mandarin Chinese throughout most of her life, but she depended on Japanese for reading and writing, such as reading a newspaper and keeping accounts. Currently, she was able to speak only very simple Taiwanese and Japanese, and she could only recognize simple Japanese characters. Her Mini-Mental State Examination (MMSE) score was 15 and CDR was 0.5. We diagnosed SD based on clinical features, a structural study with brain MRI, and cerebral perfusion SPECT (single-photon emission computed tomography)[8]. The clinical course is shown in Figure 1.

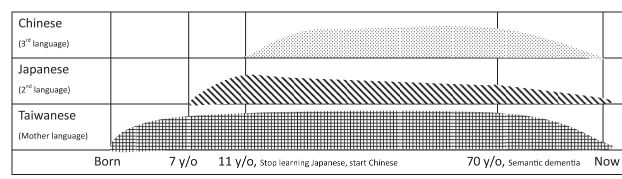


**Figure 1.** Clinical course of Case #1 (C.T)

### Case #2

The patient was a 62-year-old right-handed man who had been employed as an ironworker. He was able to speak Taiwanese and Mandarin Chinese fluently until 5 years ago. His mother tongue was Taiwanese. After 5 years of language deterioration, he was unable to communicate with his family members and could not recognize any characters, including numbers. His MMSE score was 0 and CDR was 3. This case was also diagnosed as semantic dementia based on clinical features, structural brain imaging, and cerebral perfusion SPECT (single-photon emission computed tomography) [8]. The clinical course is shown in Figure 2. Detailed clinical and diagnostic information for this case has also been described elsewhere [9].

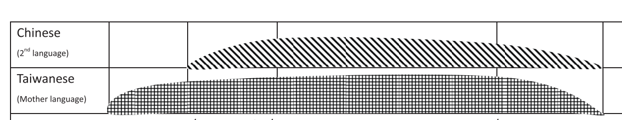


**Figure 2.** Clinical course of Case #2 (L.W)

## MRI AND SPECT

Brain MRI (GE Signa EXCITE 1.5T MRI, GE Taiwan) was performed for Case #1 and computed tomography (CT) was performed for Case #2. Tc-99m-ethyl cysteinate dimer (ECD, Neurolite, Dupont Company, USA) cerebral perfusion single-photon emission computed tomography (SPECT) scans were performed in both cases. Data were analyzed using statistical parametric mapping (SPM2; Wellcome Department of Cognitive Neurology, Institute of Neurology, London, UK) implemented in MATLAB (MATLAB6.5; MathWorks, Inc., Natick, MA). Automated coregistration of brain images to a standard stereotactic space was performed, followed by voxelwise statistical analysis. The SPECT template in SPM2 with a 12-parameter linear transformation to Talairach coordinate space was used to eliminate inter-subject anatomical variance. We also performed scaling and shearing to account for differences in individual brain shape (i.e., stereotactic anatomical standardization). All spatially normalized scans were smoothed using a 12-mm full width at half maximum isotropic Gaussian spatial filter before the voxelwise statistical analysis. Global voxel intensity normalization was achieved using proportional scaling, so that differences in global CBF (gCBF) were removed by scaling all voxels within a scan to the mean gCBF of that scan. Each voxel value in patient images was converted to standard equivalents (i.e., z scores) using the mean and standard deviation of the voxel level calculated from a normal control group.

Independent t tests (one-tailed) were performed to examine differences between 5 age- and sex-controlled normal control scans and the scans of the patients with semantic dementia. The height threshold was set to p<0.005 (uncorrected p value) and the extent threshold was set to 30 voxels. The resulting t-maps revealed areas of significantly decreased rCBF in the patients in comparison with the normal controls.

Since SD is a very unique neurodegenerative disease restricted to frontal and temporal lobe [10], we chose SPM analysis to demonstrate the local difference of these 2 patients. We have added a description of these in the revised manuscript.

## RESULTS

SPECT images of Case #1 are shown in Figure 3. SPM2 window images and Talairach coordinate areas showed markedly decreased uptake in the left inferior temporal gyrus (cluster size: 247 voxels, uncorrected p<0.005) compared to normal controls.

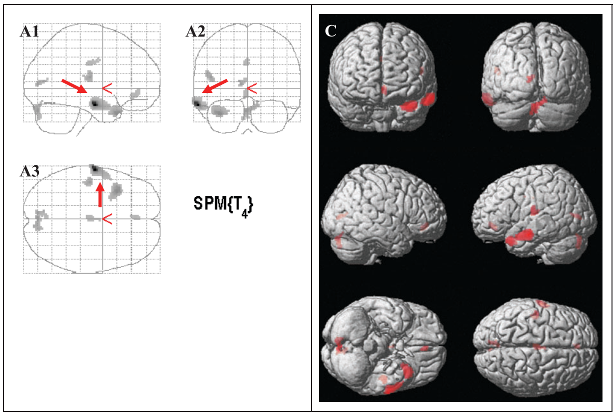


**Figure 3.** SPM2 window images and Talairach coordinate areas showed a marked decreased in Tc-99 ECD uptake in the left inferior temporal gyrus in Case #1 (A1, A2, and A3; red arrow indicates Brodmann area 20). 3D rendering views (C) show the correlated abnormal areas of absent localization (red areas).

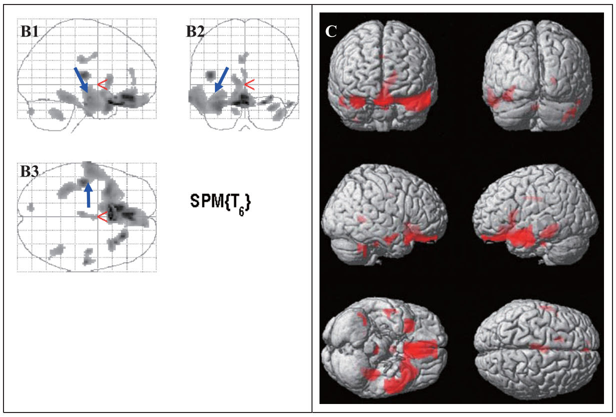


**Figure 4.** Significant decreases in Tc-99m ECD uptake were apparent in the left inferior temporal gyrus, left parahippocampal gyrus and left fusiform gyrus in Case #2 (B1, B2, and B3; blue arrow indicates Brodmann areas 20, 36 and 37). 3D rendering views (C) show the correlated abnormal areas of regional hypoperfusion (red areas).

Images for Case #2 are shown in Figure 4. Lower rCBF was apparent in the left inferior temporal gyrus, left parahippocampal gyrus and left fusiform gyrus (cluster size: 536 voxels, uncorrected p<0.005) compared to normal controls.

In comparison of the two cases, SPECT images showed less hypoperfusion in the multilingual patient (Case #1) than in the bilingual patient (Case #2). A slower rate of degeneration in the multilingual patient was also found in neuropsychological tests.

## DISCUSSION

Both cases in this report were diagnosed with semantic dementia based on research criteria for this condition. Clinically, patients with semantic dementia are known to show hypoperfusion in the frontal and temporal lobes. However, Case #1 showed relatively less hypoperfusion in the frontal lobe compared to Case #2. The patient in Case #1 had used Japanese for writing and reading throughout her life, but used Mandarin Chinese for business communication and spoke Taiwanese at home. Thus, we speculate that reading and writing in Japanese may have had a greater impact on the semantic system in Case #1. The lesser degeneration and functional inactivity in this case relative to Case #2, as shown by perfusion in the frontal lobe, might have been due to persistent activation associated with multilingualism. There is some evidence showing that separate semantic systems are present for different languages [5]. Thus, patients with more semantic systems should show better preservation of language function after development of semantic dementia, as observed in Case #1.

The protective effects of education and multilingualism may result from the cognitive reserve theory. That is, subjects with higher cognitive reserve would present clinical significant demented symptoms when pathological change of AD is severe. Recently Schweizer et al.[11] demonstrated bilingual AD patients have more severe brain atrophy than monolingual AD patients at the mild stage. Our SPECT findings, multilingual semantic dementia patient (Case #1) had less hypoperfusion than bilingual patient (Case #2), is seemingly contrary to the theory. However, the SPECT examinations of the two cases were not performed at the same clinical severity. Both cases had symptoms since recent five years and Case #1 showed slower deterioration course than Case #2, and Case #1 showed less hypoperfusion at the present time. Suppose the two cases received SPECT examinations at the same clinical severity, the SPECT of Case #1 is considered to be more severe. The important thing is that despite the same onset of years (five years), the actual clinical deterioration course different, multilingual patient showed slower course. These findings support the cognitive reserve hypothesis.

The theory of cognitive reserve suggests that education, occupation and active lifestyle could help people to maintain normal cognitive function after developing a neurodegenerative disease such as AD [12,13]. However, it remains uncertain whether languages used in everyday life play a role in cognitive reserve. Recent neuroimaging studies using fMRI revealed a wider activated cortical area in multilingual speakers compared to monolingual speakers when speaking a language other than their native language [14]. Another study also found that bilingualism has a positive impact on cognitive control through recruitment of brain regions involved in language control when performing non-linguistic cognitive tasks [15]. These fMRI studies show that multilingualism or bilingualism involves more cortical areas. A study of healthy bilingual and monolingual subjects [16] showed that lifelong bilingualism can enhance cognitive status through maintenance of the integrity of white matter. Such a finding is consistent with animal research [17] showing that an enriched environment enhances adult neurogenesis.

Multilingualism or bilingualism has recently been suggested to have protective effects against dementia [18,19]. Bialystok et al. studied 184 patients with dementia [19], including 91 judged to be monolingual and 93 bilingual. The average age of dementia onset was 71.4 years old in the monolingual group and 75.5 years old in the bilingual group, which suggested that protective effects of bilingualism could delay onset of dementia for 4.1 years on average. A more recent study [18] conducted by the same group in a cohort with a narrower diagnosis of probable Alzheimer's disease also provided evidence that bilingualism protected cognitive function. Besides protective effects in people with dementia, multilingualism has also been shown to have cognitive benefits in older persons [20]. However, such a cognitive advantage from lifelong multilingualism or bilingualism seems to be more effective in relatively poorly educated people in different studies [20,21].

In contrast, a study performed in Hawaii [22] failed to reveal similar effects in Japanese-Americans, and a subsequent study did not find that learning another language could protect people from cognitive decline [23]. Another study conducted in Canada [4] showed that the protective effects were restricted to a specific population. Among 632 patients (379 monolingual and 253 multilingual), protective effects were only found in immigrants and in non-immigrants whose first language was French. In this study, there was also a small but significant protective effect found in people who spoke more than two languages.

The potential cognitive effects of multilingualism on neurodegenerative diseases require further study in places with a multilingual environment, such as Taiwan, where Mandarin Chinese remains the official language but most people speak Taiwanese at home. Mandarin Chinese and Japanese share some common characters (Kanji) but have huge differences in grammar and oral communication. On the other hand, Taiwanese is not a symbolized language, and the effect of being multilingual in symbolized and non-symbolized languages, such as dialect in a local area, is also an important issue for future research.

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