



Case History Study

A case of expressive-vocal amusia in a right-handed patient with left hemispheric cerebral infarction



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ABSTRACT

A 53-year-old right-handed woman had an extensive lesion in the left hemisphere due to an infarction caused by vasospasm secondary to subarachnoid bleeding. She exhibited persistent expressive-vocal amusia with no symptoms of aphasia. Evaluation of the patient's musical competence using the Montreal Battery for Evaluation of Amusia, rhythm reproduction tests, acoustic analysis of pitch upon singing familiar music, Japanese standard language tests, and other detailed clinical examinations revealed that her amusia was more dominantly related to pitch production. The intactness of her speech provided strong evidence that the right hemisphere played a major role in her linguistic processing. Data from functional magnetic resonance imaging while she was singing a familiar song, a scale, and reciting lyrics indicated that perilesional residual activation in the left hemisphere was associated with poor pitch production, while right hemispheric activation was involved in linguistic processing. The localization of infarction more anterior to the left Sylvian fissure might be related to the dominant deficits in expressive aspects of the singing of the patient. Compromised motor programming producing a single tone may have made a major contribution to her poor singing. Imperfect auditory feedback due to borderline perceptual ability or improper audio-motor associations might also have played a role.

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1. Introduction

Amusia is a collective term denoting loss or impairment of musical capacity that may occur as a consequence of brain disease (Benton, 1977), or a neurodevelopmental disorder (Peretz & Hyde, 2003; Tillmann, Albouy, & Caclin, 2015; Williamson & Stewart, 2013). Clinical and neuroimaging studies in humans have shown that different hemispheric specializations underlie singing and speaking (Albert, 1998; Confavreux, Croisile, Garassus, Aimard, & Trillet, 1992; Jeffries, Fritz, & Braun, 2003; Murayama, Kashiwagi, Kashiwagi, & Mimura, 2004; Patel, 2003; Riecker, Ackermann, Wildgruber, Dogil, & Grodd, 2000; Schön, Lorber, Spacal, & Semenza, 2004; Straube, Schulz, Geipel, Mentzel, & Miltner,

2008). In most right-handed people, production and recognition of speech predominantly depend on functions of the left hemisphere. Thus, damage or anomaly to this hemisphere often leads to impaired ability to speak or to understand spoken and written language.

In contrast, the capacity for music processing seems to be less specific to either hemisphere. However, production of songs in right-handers may be a function of the right hemisphere because there are severely non-fluent aphasics with a relatively well-preserved ability to sing among dextral patients with extended left hemisphere lesions (e.g. Amaducci, Grassi, & Boller, 2002; Yamadori, Osumi, Masuhara, & Okubo, 1977). Gordon and Bogen (1974) created an experimental model of a temporary state of expressive vocal amusia with preserved speech by injecting amobarbital into the right carotid artery to produce pharmacological disruption of right hemisphere function. Regarding receptive aspects of music, the left hemisphere may play an essential role since its damage commonly results in inability of song recognition

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(Brust, 1980; Hofman, Klein, & Arlanzoroff, 1993; Lechevalier, Eustache, & Rossa, 1985; Piccirilli, Sciarma, & Luzzi, 2000).

In this paper, we report a rare case of a dextral patient with stroke who developed expressive-vocal amusia with intact speech after formation of a large left hemisphere lesion. The intact speech in this patient provides strong evidence that the right hemisphere played a major role in her linguistic processing. Detailed evaluation of her abilities concerning music perception and acoustic analysis of pitch showed that her disturbed singing performance was largely related to pitch production. Functional magnetic resonance imaging (fMRI) confirmed perilesional residual activation in the left hemisphere associated with poor pitch production and right hemispheric activation associated with linguistic processing.

2. Case report

A 53-year-old woman was admitted to a local municipal hospital because of severe headache with acute onset. She was diagnosed with subarachnoid bleeding from a ruptured aneurysm in the left middle cerebral artery. Craniotomy was performed for clipping of the artery on the first day of admission. Vasospasm developed ten days after the surgery and led to cerebral infarction in the territory of the left middle cerebral artery. MRI one month post-onset showed a large lesion in the left hemisphere, which included the whole left inferior frontal gyrus, adjacent parts of the precentral gyrus, the anterior insula, and the posterior superior temporal gyrus. The damage extended to the white matter of the left frontal and inferior parietal lobule. A white matter lesion was also found subjacent to the posterior parts of the middle and inferior temporal gyri. In contrast, the right hemisphere was intact (Fig. 1).

2.1. Neurological and neuropsychological findings

The patient was referred to our hospital for rehabilitation two months post-onset because of gait disturbance. She was alert and attentive, and could engage in conversation with no hearing disturbance. Her speech was well articulated with normal prosody, and she was fluent with no paraphasia or apparent difficulty in word retrieval. A visuoperceptual examination revealed mild visual inattention on the right side with slight constructional disabilities, both of which vanished shortly thereafter. Right-sided mild visual

inattention at admission had resolved one month later in a follow-up test, with the initial score of 3 out of 9 changing to zero on the line bisection task of the Behavioral Inattention Test.

Hemiparesis and superficial sensory deficits including the trunk and face were present on the right side. The patient also walked unsteadily in a hemiplegic posture. Her handedness was assessed using the Edinburgh Inventory (Oldfield, 1971). Her laterality quotient value was +88 right handedness with a decile rank of 7th right. She also reported no familial history of left handedness.

The Mini-Mental Examination score was 27 out of 30. The Japanese Standard Language Test for Aphasia (SLTA) was used to evaluate language functions, and scores for abilities for listening (40/40 = 100%), speaking (86/91 = 95%), reading (40/40 = 100%), writing (38/46 = 83%), and calculation (18/20 = 90%) were recorded. These values indicated language proficiency within normal limits. She had some difficulties in writing kana characters (phonograms) and recalling kanji characters (ideograms), but these abilities recovered within a few weeks. Regarding declarative memory function, she scored 6 forward and 4 backward on the digit span test. The frontal assessment battery (FAB) and Wisconsin card sorting test (WCST) were administered as frontal function tests. The scores were 15 out of 18 points on the FAB, and 4 categories were achieved on the WCST, indicating frontal lobe function similar to that in normal adults (Kugo et al., 2007).

2.2. Musical assessment

At the start of in-hospital physical and occupational training, the patient reported that her singing ability had notably declined, especially with high-pitch notes, and thus she was referred to the music therapy department. She reported that she had received music education as a compulsory subject at elementary and junior high school, and as an elective subject at high school. She had no experience of playing musical instruments. She worked as a school clerk after finishing high school and she enjoyed listening to music of various kinds during post-adolescence. She also learned and memorized songs by ear, which was one of her hobbies. She occasionally went to a karaoke club with her family or friends, and on each occasion she spent several hours singing her favorite songs prior to her illness. Her family members all reported that she was a good singer and that she sang songs well in tune, including the national anthem of Japan (“Kimigayo”).

3. Material and methods

The purpose and methods of the study were carefully described to the patient before the start of the study, after which she gave written informed consent. The study was approved by the Ethical Review Board of Nishi-harima Rehabilitation Hospital. The following tests for musical ability were performed.

3.1. Receptive and expressive functions with regard to music

Tests were completed during the first week of music therapy. Receptive function was tested using simple tone pitch discrimination, the Montreal Battery for Evaluation of Amusia (MBEA), and familiar song recognition tests. For simple tone pitch discrimination, the patient listened to a standard sound of 440 Hz lasting two seconds, followed by a comparison test sound of the same duration. The comparison test sound was from a set of tones having 10 different levels of pitch between 400 Hz and 490 Hz with a 10-Hz interval. Each sound from the set was tested in a random order and the test was performed twice. All sounds were created using digital sampling with Artemiy Pavlov Oscillator software. After presentation of the second sound, the patient was asked if

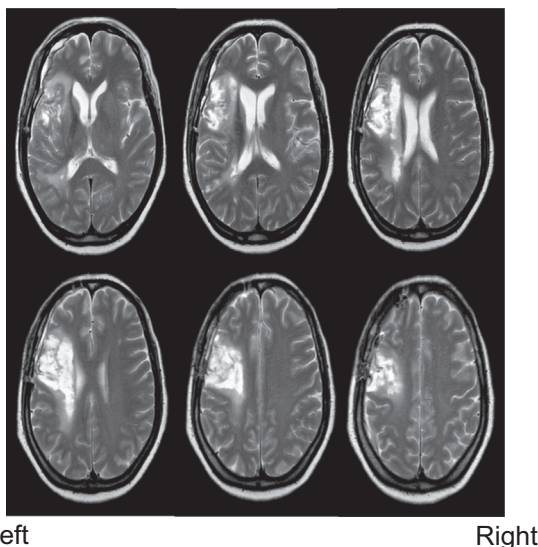


Fig. 1. T2-weighted images in a brain MRI scan at one month after craniotomy for clipping of the aneurysm.

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