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A qualitative motion analysis study of voluntary hand movement induced by music in patients with Rett syndrome

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Abstract: Patients with Rett syndrome are known to respond well to music irrespective of their physical and verbal disabilities. Therefore, the relationship between auditory rhythm and their behavior was investigated employing a two-dimensional motion analysis system. Ten female patients aged from three to 17 years were included. When music with a simple regular rhythm started, body rocking appeared automatically in a back and forth direction in all four patients who showed the same rocking motion as their stereotyped movement. Through this body rocking, voluntary movement of the hand increased gradually, and finally became sufficient to beat a tambourine. However, the induction of body rocking by music was not observed in the other six patients who did not show stereotyped body rocking in a back and forth direction. When the music stopped suddenly, voluntary movement of the hand disappeared. When the music changed from a simple regular rhythm to a continuous tone without an auditory rhythm, the periodic movement of both the hand and body prolonged. Auditory rhythm shows a close relationship with body movement and facilitates synchronized body movement. This mechanism was demonstrated to be preserved in some patients with Rett syndrome, and stimulation with music could be utilized for their rehabilitation.

Keywords: Rett syndrome, music, auditory rhythm, stereotyped movement, body rocking, voluntary movement

Introduction

Rett syndrome is a childhood neurodevelopmental disorder usually caused by a mutation in the gene encoding MECP2 located on the X chromosome (Xq28).[1] Rett syndrome occurs predominately in females although males have been described with MECP-2 mutations.[2] Stereotyped movement of the hands such as wringing, washing, hand clapping, and hand-to-mouth movements following the loss of the functional use of the hands is the most characteristic feature of Rett syndrome.[3] Typically, patients with Rett syndrome have no verbal skills, and about 50% of them are not ambulatory.[3] However, they are reported to respond well to music in comparison with their physical and verbal disabilities.[4–7]

Synchronized movement to music has been observed in all known human cultures, implying that this ability is universal and perhaps unique to human musical behavior.[8] Among various musical elements such as pitch, melody, harmony, rhythm, dynamics, timbre, etc., auditory rhythm is known to exhibit a close and fundamental relationship with body movement from early infancy,[9,10] and facilitates synchronized body movement not only in healthy subjects of all ages but also in patients with various movement disorders such as Parkinson disease,[11] Huntington's disease,[12] stroke,[13] and

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incomplete spinal cord injury.[14] Improvement in walking speed and stride length by auditory rhythm was demonstrated in patients with Parkinson disease.[15,16] These findings suggest that auditory rhythm could also modify motor behavior and induce voluntary movement in patients with Rett syndrome; however, little is known about the basic role of auditory rhythm in these patients.[17]

The motion analysis system is a well established clinical method to examine temporal, spatial, and kinetic movements of various parts of the human body, particularly for the assessment of gait in healthy subjects[18] and intervention evaluation in patients with cerebral palsy.[19] We utilized this method to investigate the relationship between auditory rhythm and behavioral movement in patients with Rett syndrome in order to promote their voluntary hand movement.

Patients and methods

Ten female patients with Rett syndrome, aged from three to 17 years, were included in this study. The diagnosis was made by more than two child neurologists according to the diagnostic criteria for Rett syndrome established by the Rett Syndrome Diagnostic Criteria Work Group.[20] Patient profiles are described in Table 1. All patients showed stereotyped hand movement. In addition, eight of 10 patients had stereotyped body rocking movement: back and forth, three; left and right, four; and both, one (Table 1).

According to the previous study on motion analysis in a patient with Rett syndrome,[21] markers were put on the wrists and shoulders in patients to investigate the movement of their hands and upper bodies, respectively. Color tapes 20-mm wide were employed as the markers, and their movements were captured using a two-dimensional digital video camera at a sampling frequency of 30 Hz. Two-dimensional motion analysis software (Move-Tr/2D ver.7; Library Co., Ltd., Japan) was utilized to record temporal, spatial, and kinetic changes of movements in response to music in a qualitative manner.

First, a tambourine was presented in front of patients for several seconds. When no purposeful movement was observed, music familiar to each patient related to us by caregivers was started with a simple regular rhythm. Sometimes, the music was stopped suddenly or changed from a simple regular rhythm to a continuous tone without any rhythm to assess any behavioral changes in movement. This examination was performed after music therapy for six months. Each session was 40 minutes long and was provided once or twice a month.

This study was approved by the Institutional Ethics and Research Board, and informed written consent was obtained from legally authorized representatives of the patients. Participants were outpatients of Saitama General and Medical Center for the Disabled.

Results

When music with a simple regular rhythm started, body rocking appeared automatically in a back and forth direction in all four patients who had the same body movement as their stereotyped movement (Table 1). Through this body rocking induced by music, voluntary hand movement increased gradually and finally became sufficient to beat a tambourine presented in front of the patient (Table 1). This voluntary hand movement was observed consistently during and after the study when patients were awake and in a good temper. However, the induction of body rocking by music was not

Table 1 Profiles of patients and movement induced by music

Age	Gene	EEG	Sz	Physical ability	Stereotyped body movement	Induced body rocking	Induced hand movement
3	Ab	N	–	Ambulatory	Left and right	–	–
4	Ab	Ab	–	Sitting	Left and right	–	–
4	Ab	N	+	Sitting	Left and right	–	–
5	ND	Ab	+	Ambulatory	None	–	–
6	Ab	Ab	–	Wheelchair	Back and forth	+	+
7	Ab	Ab	+	Ambulatory	Back and forth	+	+
9	ND	Ab	+	Sitting	Left and right	–	–
11	Ab	Ab	+	Ambulatory	Back and forth Left and right	+	+
12	Ab	Ab	+	Crawling	Back and forth	+	+
17	ND	Ab	–	Wheelchair	None	–	–

Abbreviations: EEG, electroencephalogram; Sz, seizure; Ab, abnormal; N, normal; ND, not done; Wheelchair, impossible to maintain any posture without assistance.

observed in the other six patients who did not have stereotyped body rocking in a back and forth direction (Table 1).

The motion analysis system demonstrated that body rocking induced by music consisted of primarily repetitive horizontal back and forth movement and little vertical movement (Figure 1). It persisted with almost the same regular periodic cycle as the auditory rhythm of music and constant amplitude while the music continued (Figure 1). Through this regular and cyclic body movement induced by music, voluntary hand movement gradually increased in a vertical direction with the same periodic cycle as the body, and finally allowed the patient to reach the tambourine presented in front of her (Figure 1). Vertical movement of the hand was much more pronounced than horizontal movement of the body (Figures 1–3).

When the music stopped suddenly, voluntary movement of the hand soon disappeared (Figure 2). Horizontal movement of the body also decreased gradually and stopped after a while (Figure 2). Horizontal movement of the hand merely reflected the accompanying motion of the body in the same direction. When the music changed from a simple regular rhythm to a continuous tone without an auditory rhythm, movements of both the hand and body became slower and the periodic cycle of the motion prolonged (Figure 3).

Discussion

In this study, patients with Rett syndrome showing stereotyped body rocking in a back and forth direction were demonstrated to recognize and respond well to changes in the rhythm of music. A simple regular auditory rhythm induced stereotyped body rocking movement in a back and forth direction at first. Then, the auditory rhythm as well as induced body rocking movement facilitated voluntary movement of the hands.

Purposeful hand movement was performed through this regular and cyclic body movement induced by music. The target object was placed in front of patients and patients watched it. This might explain why only stereotyped body rocking in a back and forth direction was induced in this study.

Stereotyped movement is repetitive, restricted, and nonfunctional motor behavior observed in various neurological and developmental disorders such as Rett syndrome,[3] autistic disorder,[22] and visual or auditory impairment.[23] If it is frequent and severe, it may interfere with normal voluntary movement. Therefore, treatment and rehabilitation usually focuses on how to suppress it in order to facilitate voluntary movement. However, voluntary movement can be induced more easily through using rather than suppressing stereotyped movement in patients whose voluntary movement is very difficult to promote, such as those with Rett syndrome. The reinforcement of rhythm in stereotyped movement by music might be another way of rehabilitation for such patients.[17]

Action induction by music is accompanied by neural impulses in the reticular formation of the brainstem.[24] In addition, listening to a simple regular rhythm without

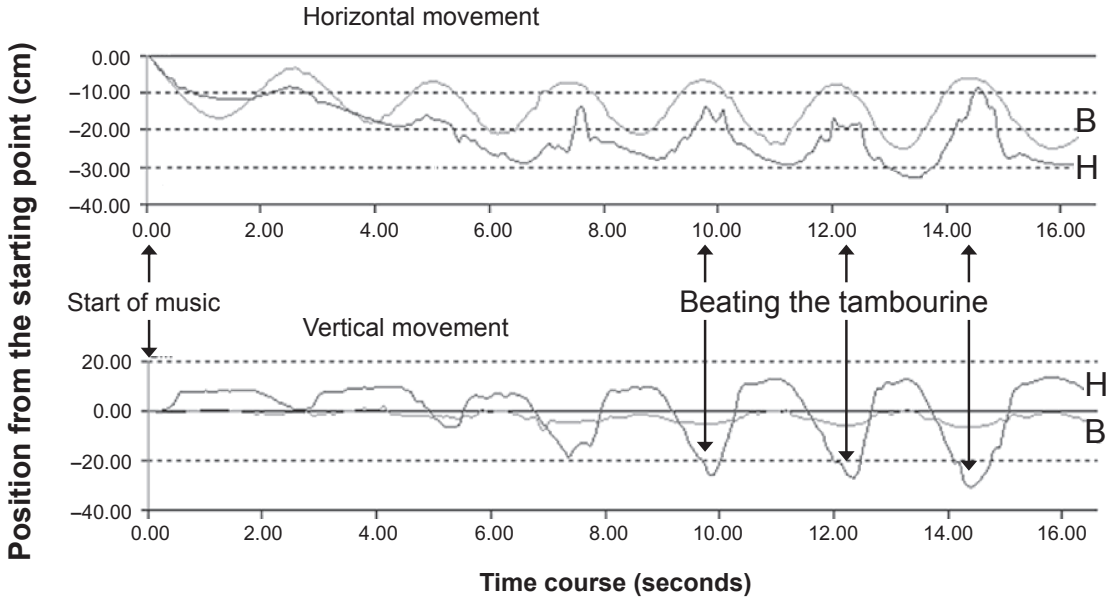

Figure 1 Motion analysis of an 11-year-old patient.
Notes: The horizontal axis is the time course and the vertical axis is the position from the starting point of each body part. Forward and upward positions from the starting point are shown as positive values. Movements of the body and hand are indicated by lines B and H, respectively.

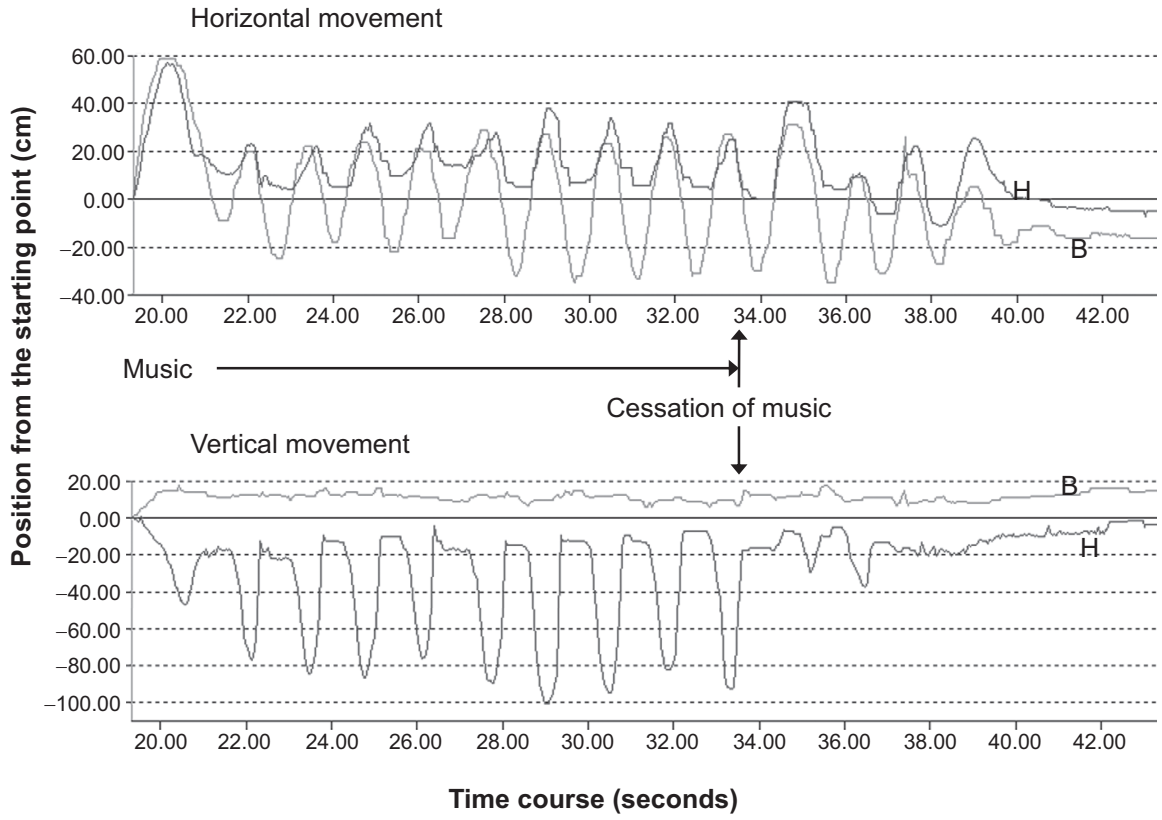

Figure 2 Changes in motion by the abrupt cessation of music in a 7-year-old patient.
Note: Axes and symbols are the same as in Figure 1.

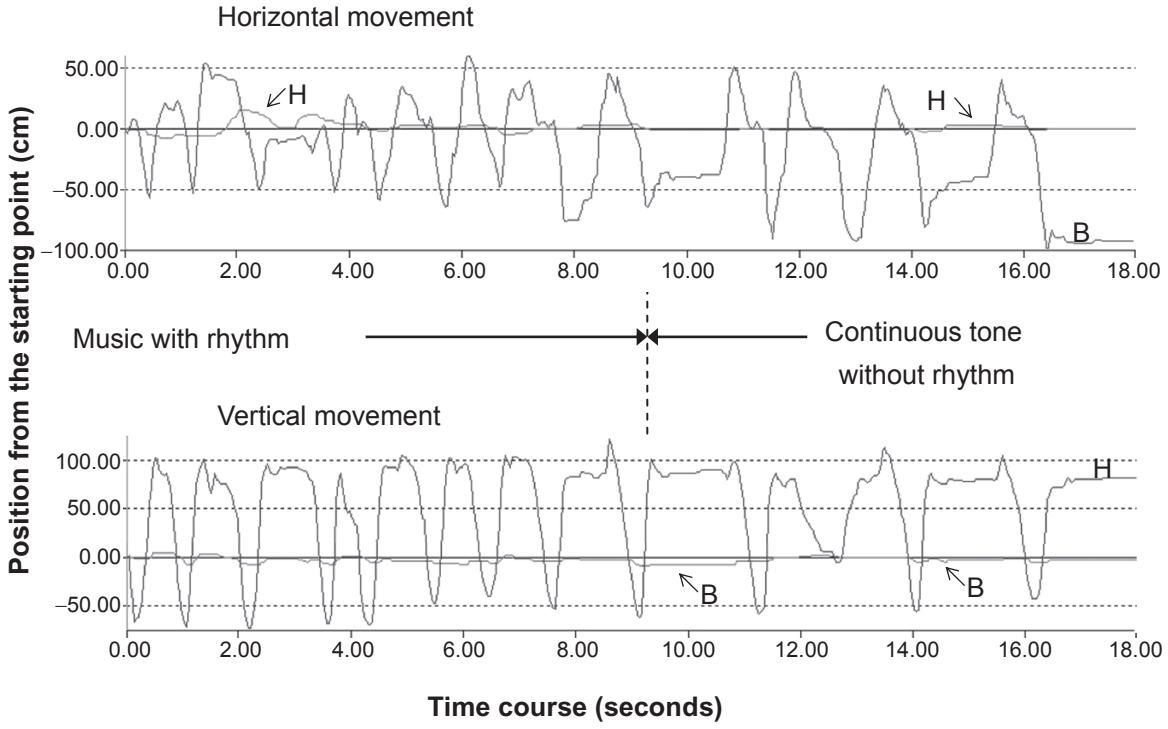

Figure 3 Changes in motion induced by music with and without a rhythm in a 12-year-old patient.
Note: Axes and symbols are the same as in Figure 1.

any actual movement increases the activity of the basal ganglia and supplementary motor area.[9] These brain areas are involved in motor prediction and the timing of future movements.[25,26] Therefore, listening to music with a simple regular rhythm would prime body movement. This mechanism could be preserved in some patients with Rett syndrome, and stimulation with music might be worth attempting for their rehabilitation. Actually, patients who responded to music showed some improvement of their hand use in everyday life such as opening the door or holding an object for a while even though Rett syndrome progresses with gradual deterioration of the motor system.

In this study, the movement of 10 patients was analyzed in a qualitative manner without a control group. Further studies involving more subjects including older patients with a control group are necessary to clarify the role of auditory rhythm in patients with Rett syndrome.

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