Case Study

Usefulness of continuous actigraph monitoring in the assessment of the effect of corticosteroid treatment for Duchenne muscular dystrophy: a case report

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Abstract. [Purpose] This study evaluated the effect of corticosteroid treatment on the daily activity of a patient with Duchenne muscular dystrophy using an actigraph and examined whether this method produces the same results as the conventional motor-function evaluation methods. [Subject and Methods] A patient with 5 year-old Duchenne muscular dystrophy was recruited. An actigraph was attached to his waist to measure the energy expenditure and the number of steps taken by him during a period of two weeks, 14 days before and 14 days after corticosteroid administration. The outcomes of these measurements were compared with the results of conventional motor-function evaluation methods—the 10-m run test, 6-minute walk test, and North Star Ambulatory Assessment—on his next visit. [Results] The actigraph data for energy expenditure and the number of steps taken correlated well with the results of the above-mentioned conventional motor-function tests, and the value of each data point improved after corticosteroid administration. [Conclusion] An actigraph was effectively used to non-invasively measure consecutive daily activity for four weeks. It was easily done and the results were consistent with conventional motor-function evaluation methods.

Key words: Actigraph, Duchenne muscular dystrophy, Motor function

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INTRODUCTION

The evaluation of motor function for Duchenne muscular dystrophy (DMD) includes the 10-meter run, 6-minute walk test (6MWT), and the North Star Ambulatory Assessment (NSAA). These evaluation methods have also been used to determine the appropriate time to commence corticosteroid therapy for patients with DMD. The reliability and validity of the 6MWT and NSAA have been confirmed in a multi-center longitudinal study¹, and these assessment methods have been employed in clinical studies². The outcomes of these tests can help assess an individual’s motor ability in detail; however, these tests have certain limitations when they are performed at a hospital. They are usually performed at the time of the patient’s visit to the hospital, which makes them less frequent and discontinuous. Moreover, the results are easily influenced by the physical or emotional condition of the patient.

It has been reported that the amount of daily life activity for the motor-functional evaluation of healthy children and pediatric diseases³–⁴ can be monitored using an actigraph. Furthermore, the amount of activity recorded by an actigraph has been used as the main outcome index to reflect the amount of physical activity in the home environment of patients with DMD⁵.

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Although, these reports did not compare the results of actigraph monitoring with conventional motor-function evaluation methods, their results suggest actigraph assessment of daily life activity can be used to evaluate motor function as effectively as the conventional motor-function tests. Moreover, an actigraph can easily and non-invasively record consecutive data.

In this study, whether an actigraph can be used to effectively monitor the changes in the motor function of a patient with DMD before and after corticosteroid administration was examined by comparing the results with conventional motor-function evaluation methods.

**SUBJECT AND METHODS**

The subject was a 5-year-old male patient with Duchenne muscular dystrophy. The subject’s body weight was 17 kg and his body mass index and serum creatine kinase were 15.1 kg/m² and 24,030 U/L, respectively. A nonsense mutation was identified in exon 21 of the patient’s dystrophin gene. The patient’s motor function was evaluated from April 2014 to October 2015.

The actigraph (MTN-210, ESTERA, Saitama, Japan) was constantly attached to the patient’s lower waist, except during bathing, during the evaluation period. Data were collected for 14 days before and 14 days after the administration of corticosteroids, until his next visit to the hospital. The collected data were analyzed using dedicated software (SleepSign Act, Kissei Comtec, Matsumoto, Japan). The evaluation index was calculated by taking the mean energy expenditure and number of steps per day. Statistical analysis was performed using the unpaired Student’s t-test and PASW Statistics software (version 22.0, SPSS, Inc., Chicago, IL, USA). During the 14 days before and after the administration of corticosteroids, the patient did not participate in any high-momentum activities such as athletic events.

Conventional motor-function tests, the 10-meter run, 6MWT, and NSAA, were performed by the patient at the time of hospital visits (once a month). The difference in the results of each test was examined before and after corticosteroid administration. The 6MWT was performed in two ways—the way it is conventionally defined by the American Thoracic Society (conventional 6-minute walk test; C6MWT), and in the way described in our recent report, paced by the sound of a metronome (periodic sound-based 6-minute walk test; PS6MWT). The PS6MWT can determine the maximum potential walking ability of patients with DMD. In this test, the subject initially walked for one minute to a long-interval metronome sound, and the distance walked was measured. The sound interval was then gradually shortened, and the subject walked for one minute at each of the metronome beat intervals. The best periodic sound was determined as that when the subject walked the longest distance in one minute. The subject then walked, paced by the best periodic sound, for six minutes. Since the distance covered in the PS6MWT by both healthy subjects and those with DMD is significantly longer than the distance in the C6MWT, the PS6MWT provides a better indication of ambulatory potential in the evaluation of physical endurance than the C6MWT.

This study was approved by the institutional ethics committee of the Shinshu University School of Medicine, Japan (approval number: 2,340). The study was orally explained to the subject and his parents, and they consented to participation in the experiment.

**RESULTS**

In this study, the conventional motor-function tests were used to assess a patient with DMD before and after corticosteroid administration. The energy expenditure and number of steps taken by the patient increased after corticosteroid administration. Daily activity measured using an actigraph and using conventional motor-function evaluation tests indicated the same tendency.

In this study, the conventional motor-function tests were scheduled once a month, but on one occasion, there was a gap of more than two months between the tests. Thus, the evaluation frequency was low and discontinuous. The main reasons...
for the low and discontinuous frequency of evaluations were the long distance between patient’s residence and the hospital, and the unstable physical and emotional condition of the patient. These factors are not likely to change. On the other hand, the actigraph measured data at any place and time, which made statistical analysis possible using the data collected during a period of just 14 days.

Our results show that an actigraph can be easily and noninvasively used to monitor daily life activity on consecutive days for the evaluation of motor activity before and after corticosteroid administration. Furthermore, the evaluation of motor activity using an actigraph is consistent with the results of conventional motor-function tests. Future studies addressing this topic should include a large number of subjects and evaluate motor functions over a longer period to determine the utility of the actigraph as a primary tool for measuring the outcome of clinical trials and to evaluate the efficacy of therapeutic agencies.

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Table 1. Actigraph results before and after the corticosteroid administration

|                          | Before administration | After administration | *a |
|--------------------------|-----------------------|----------------------|----|
| Number of steps (steps/day) | 10,081.8 ± 2,638.7    | 12,205.0 ± 1,695.4   | *a |
| Energy expenditure (kcal/day) | 1,108.7 ± 137.2       | 1,246.0 ± 127.0      | *a |

*p<0.05, *a unpaired Student’s t-test

Table 2. Conventional motor function test results before and after the corticosteroid administration

|                          | Before administration | After administration | Difference |
|--------------------------|-----------------------|----------------------|------------|
| 10-m run (s)             | 4.4                   | 3.7                  | 0.43       |
| C6MWT (m)                | 366                   | 372                  | 6          |
| PS6MWT (m)               | 375                   | 393                  | 18         |
| NSAA (score)             | 22/34                 | 29/34                | 7          |

C6MWT: conventional 6-minute walk test; PS6MWT: periodic sound based 6-minute walk test; NSAA: North Star Ambulatory Assessment