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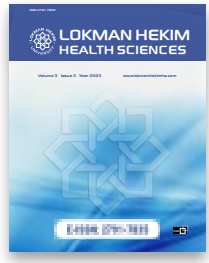

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CASE REPORT

6-week Results of Physiotherapy and Cognitive Behavioral Therapy in a Patient with Bilateral Lower Extremity Dystonia

Bilateral Alt Ekstremite Distonisi Olan Bir Hastada Fizyoterapi ve Bilişsel Davranışçı Terapinin 6 Haftalık Sonuçları

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Abstract

The aim of this study was to investigate the effectiveness of physiotherapy and rehabilitation applications for balance and coordination problems caused by segmental bilateral lower extremity dystonia and simultaneously applied cognitive behavioral therapy (CBT). There are few studies in the literature about physiotherapy in lower extremity dystonia. In our study, the application of CBT in addition to physiotherapy in lower extremity dystonia makes our study unique and we aim to guide future studies. The patient, who was referred to the clinic due to balance and coordination disorders accompanied by lower extremity pathologies, underwent a 90-min rehabilitation program for 6 weeks, 3 sessions per week, with 60 min of exercise and 30 min of hydrotherapy in each session. Simultaneously with physiotherapy, the patient received psychotherapy with CBT by a specialist psychologist for 3 sessions per week. Static and dynamic balance exercises, lower limb coordination, gait training, relaxation training, breathing exercises, and Whirlpool were applied to the patient. The patient's balance was evaluated with Berg Balance Scale and one-leg stand tests, quality of life with SF-36, mobility with Timed Stand and Sit Test, frequency and severity of dystonic spasms with Modified penn spasm frequency scale, pain with VAS Pain Scale, and anxiety with Beck Anxiety Scale. In this patient with lower extremity dystonia, it was concluded that the patient's balance improved, quality of life and mobility increased, dystonic spasms and pain decreased with a 6-week physiotherapy and rehabilitation program, and anxiety decreased with concurrent CBT. As a result, physiotherapy and CBT can increase the functional capacity and quality of life of the person.

Keywords: Case report; Cognitive behavioral therapy; Lower extremity dystonia; Physiotherapy

Dystonia is characterized by continuous or intermittent muscle contractions, leading to abnormal movements and/or postures. Segmental dystonia consists of dystonic contractions of neighboring muscle groups. It affects not only the extremities but also the proximal junction muscles of the same extremity. The prototypic phenotype of adult-onset lower limb dystonia includes foot torsion with inversion at the ankle joint, often accompanied by flexion

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of the forefoot and toes.[1] The literature frequently emphasizes psychological factors in adult-onset lower limb dystonia.[2] Therefore, dystonia is considered a complex movement disorder with a poorly understood pathogenesis. In the literature review, no publications on physiotherapy and rehabilitation of lower extremity dystonia were found. In most of the studies, the usability of medical treatments was investigated.[3] This situation makes our study unique. This study aims to investigate the effectiveness of physiotherapy and concomitant cognitive behavioral therapy (CBT) in a woman with lower extremity dystonia.

Case Report

A 28-year-old patient with balance, coordination, and gait problems presented to the neurology clinic. As a result of genetic tests, EEG, EMG, and Spinal Cord MRI examinations, no pathology was found and no diagnosis could be made. Although a definite diagnosis could not be made for the patient whose lower extremity deformity started to progress, a single session of botulinum toxin (BoNT) was applied to the hamstring, quadriceps femoris, and gluteal muscles at the age of 30, but the patient reported that he did not benefit from this treatment. At the age of 32, the patient was admitted to the physiotherapy clinic and physiotherapy was applied for 30 sessions each year for 4 years until the age of 36. The patient, whose prognosis worsened every year, presented to our clinic at the age of 36. The patient stated that he felt that he lost his walking balance and felt that he could not walk and locked up, especially when environmental stimuli were high, when he was anxious, excited, and when his motivation decreased. Psychologist consultation was requested to control the anxiety level and for CBT to support physiotherapy applications.

Evaluation Methods

The patient's balance was evaluated with Berg Balance Scale and one-leg stand tests, quality of life with SF-36 (Short Form), mobility with Timed Stand and Sit Test, frequency and severity of dystonic spasms with Modified Penn Spasm Frequency Scale, pain with VAS Pain Scale, and anxiety with Beck Anxiety Scale. The patient underwent a rehabilitation program for 6 weeks, 3 sessions per week, each session consisting of 60 min of exercise and 30 min of hydrotherapy for 90 min. Simultaneously, the patient was admitted to psychotherapy for 3 sessions per week by a specialist psychologist for CBT. The results of the evaluations performed at week 1 and week 6 of the 6-week rehabilitation program are shown in Table 1.

Table 1. Pre- and post-treatment values

	1. week	6. week
Berg balance scale	49 Score	56 Score
One-leg stand tests		
Right		
Eyes open	9.73 s	13.49 s
Eyes closed	2.03 s	3.31 s
Left		
Eyes open	4.47 s	4.60 s
Eyes olosed	1.57 s	1.76 s
Timed get-up-and-go test	18.7 s	13.25 s
SF-36 (short form)		
Total score	87 score	93 score
Modified penn spasm frequency scale		
Spasm frequency	3	1
Spasm violence	3	1
VAS pain scale		
Total score	3	1
Beck anxiety scale		
Total score	40	18

Berg Balance Scale is a test that evaluates changes in forward bending and standing position, transfers between positions, and the ability to maintain sitting and standing balance.[4]

Standing on one leg test; the duration of the subject's standing on one leg on the right and left legs, with the eyes open and closed, with the arms next to the trunk, was recorded in seconds.[5]

Timed get up and go test; the subject was asked to sit on a chair without arm support, with his/her feet touching the floor, and with the "get up" command given, to get up, walk a marked 3-m area, return and sit back on the chair. The duration of the activity was recorded. The measurement was repeated 3 times and the mean value was calculated.[5]

SF-36 (Short Form) is a scale that has reliability in evaluating the quality of life of individuals and is used quite frequently. It consists of 36 items. It has two parameters related to physical health (physical function, physical role, pain, and general health) and mental health (energy, social function, emotional role difficulty, and mental health) and eight sub-parameters under these sections.[6]

The modified Penn spasm frequency scale is a 5-point scale developed by Penn et al.[7] to evaluate the effectiveness of intrathecal baclofen treatment.

VAS pain scale: The patient was evaluated for extremity pain during movement with a VAS scale between 0 and 10 (0 points – no pain and 10 points – severe pain).[8]

The Beck anxiety test, which has been proven to be reliable in our country, includes four questions, each consisting of four questions and ranging from 0 to 3 points (none, 0; mild, 1; moderate, 2; and severe, 3). It is evaluated as "score between 8 and 15=mild anxiety" "score between 16 and 25=moderate anxiety" "score between 26 and 63=severe anxiety."[9]

Physiotherapy Protocol

After the necessary evaluations, the patient was enrolled in a physiotherapy program simultaneously with psychotherapy. Static and dynamic balance exercises, lower limb coordination, gait training, relaxation training, breathing exercises, and Whirlpool were applied to the patient. Whirlpool application was performed at the same temperature as body temperature for 30 min in each session.

CBT Protocol

CBT focuses on the problems in the cognitive system of the person and aims to change behaviors by solving these problems. CBT is the therapy system with the most evidence about its effectiveness. CBT's wide range of effects has been shown by meta-analyses[10] and it is still the subject of research and continues to expand.

Especially in anxious situations where dystonic spasms occurred, the negative thoughts of the person were identified and these were tried to be changed with the affirmation technique. Again, to determine the situations where the level of anxiety increased during the day and the basic emotions underlying these situations, the negative emotions and anxieties experienced by the subject during the day were recorded and these negative emotions were tried to be changed with the affirmation technique in the therapy. The environments where the anxiety was the highest were determined and necessary observations were recorded in real environments. The methods taught to the patient to control this situation in a real environment that increases dystonic spasms were practiced with an expert. Breathing and relaxation training was given to the patient to control these spasms in situations where anxiety increased and dystonic spasms occurred.

Discussion

Lower limb dystonia affects gait parameters and disrupts the normal gait pattern. In our study, it was concluded that a 6-week physiotherapy and rehabilitation program in a patient with segmental lower extremity dystonia improved the patient's balance parameters, increased quality of life and mobility, decreased dystonic spasms and pain, and decreased the anxiety level of the concurrent CBT. In the literature, there are limited studies on physiotherapy programs for individuals with lower extremity dystonia.

Adatepe[3] stated that botulinum toxin applications are a preferred treatment option in patients with dystonia, while physiotherapy applications are a factor that helps to reduce symptoms in patients with dystonia.

In a randomized and controlled study in which Vizcarra et al.[11] compared BoNT-A applied before CBT with placebo BoNT-A applied before CBT in individuals with functional dystonia; they concluded that CBT applied with placebo BoNT-A showed positive effects.

According to both the patient's statements and the results of the evaluation parameters, psychological factors had a great effect on the severity of dystonia. For this reason, the contribution of CBT to the rehabilitation process was too strong to be ignored. In our study, it was concluded that CBT applied for 6 weeks and 3 sessions per week decreased the level of anxiety and showed positive effects on dystonia.

There are some limitations in our study. Our limitations were that there was only one case in our study and the study could not be continued for a longer period of time because the patient learnt that she was pregnant at the end of the 6th week. There is a need for more comprehensive studies in which the effectiveness of CBT applied concomitantly with physiotherapy is investigated and this subject, which has great deficiencies in the literature, is applied on more cases.

Physiotherapy and CBT can be used as an alternative treatment method other than medical treatments in lower extremity dystonia. These applications can improve many physical and psychological parameters of patients. There is a need for more comprehensive studies on this subject, which has great deficiencies in the literature.

Peer-review: Externally peer-reviewed.

Informed Consent: Written informed consent was obtained from patients who participated in this study.

Authorship Contributions: Concept: MB, AAK; Design: MB, AAK; Supervision: AAK; Materials: AAK; Data Collection or Processing: MB; Analysis or Interpretation: MB; Literature Search: MB?; Writing: MB; Critical Review: AAK.

Conflict of Interest: None declared.

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