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Successful treatment of belly dancer's dyskinesia with levetiracetam: A case report

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CASE STUDY

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ABSTRACT

Belly Dancer's Dyskinesia is an extremely rare hyperkinetic movement disorder. It is a form of axial myoclonus which affects the diaphragm and abdominal muscle, causing pain and discomfort. It is characterized by brief, repetitive contractions of the diaphragm and abdominal muscles, which can be suppressed with deep respiration. Diagnosis is mainly made by clinical examination, but can be challenging, and treatment can be difficult. Here we report a case of Belly Dancer's Dyskinesia which was successfully treated with Levetiracetam.

Key Words
Belly dancer's dyskinesia, myoclonus, diaphragmatic flutter, abdominal wall dyskinesia

Implications for Practice:

1. What is known about this subject?
Belly Dancer's Dyskinesia affects less than 8.6 people per 100.000.

2. What new information is offered in this case study?
Belly Dancer's Dyskinesia can be successfully treated with levetiracetam.

3. What are the implications for research, policy, or practice?
Levetiracetam may become an option in treatment of patients with Belly Dancer's Dyskinesia.

Background

Movement disorders can be categorized as either hypokinetic or hyperkinetic disorders. Hyperkinetic movement disorders include tremors, tics, ballism, myoclonus, chorea, athetosis and dystonia.[1] Myoclonus is a characterized by sudden, brief, involuntary jerks of a single muscle or group of muscles, and can be induced by activation of cortical, subcortical, spinal cord or peripheral nerves. Propriospinal myoclonus is characterized by myoclonus of the trunk and abdominal muscles, with a fixed up-and-down pattern of muscle activation.[2] Diagnosis of this condition can be challenging due to its rarity[3] and the lack of laboratory or imaging diagnostic methods to confirm diagnosis. Diagnosis relies, therefore, on clinical assessment.[4]

Belly Dancer's Dyskinesia (also called diaphragmatic flutter or belly dancer syndrome) is an extremely rare phenomena characterized by rapid myoclonus of diaphragm in which the abdominal muscles may be involved.[5] Here, we described the case of young lady who was referred for psychogenic movements. Thorough examination led to the diagnosis of Belly Dancer's Dyskinesia, and we initiated a short course of clonazepam followed by levetiracetam which resulted in a marked improvement in symptoms.

Case details

A 34-year-old right-hand dominant female with a medical history of meningitis at the age of two weeks with sequalae

of mutism presented to the emergency department on 4/12/2019 with jerky abdominal movements lasting for one year prior to presentation. The movements had worsened over the last two days and had started to interfere with her eating but disappear when she sleep. The movement was irregular, almost continuous, and affected both sides of the abdomen. She was unable to suppress the movement, but it ceased when she took deep breaths. She did not report any myoclonic jerks of the arms or the legs, nor generalized tonic-clonic seizures. There was no history of dysphagia, facial weakness, or limb weakness or numbness. There was no gait abnormality.

Clinical findings

Clinical examination revealed vital signs to be stable and normal mental status. We were unable to assess fluency and repletion because the patient was mute. The cranial nerve was found to be normal apart from impaired abduction of the right eye (which was long standing). Motor examination revealed normal muscle tone, bulk, and power. Deep tendon reflexes were 2^+ symmetric in all four limbs, abdominal reflexes were normal. Gait and cerebellar examination revealed normal findings. Regarding the abdomen, irregular contraction of the abdominal wall muscle was observed on both sides, which was almost constant but stopped when the patient took a deep breath (Video 1). The abdomen was soft and no organomegaly was noted.

Diagnostic assessments

Head magnetic resonance imaging (MRI) showed features of a sequalae of previous brain injury (significant volume loss and blooming artifacts related to calcification within the bilateral inferior frontal gyrus) Spine MRI, electroencephalogram (EEG), chest X-ray, echocardiography, and abdominal ultrasound were all normal.

Therapeutic intervention

Clonazepam (0.5mg po bid) was initiated on 5/12/2019 and the movement improved within 48 hours by about 50 per cent–70 per cent according to the patient. After two days, on 7/12/2019, the patient was started on levetiracetam (250mg po bid), and on 10/12/2019 clonazepam was increased to 1mg po bid.

Follow-up and outcomes

The patient was followed up in the clinic on 23/12/2019, at which time her mother reported that the movement had almost completely resolved. The patient had only experienced two short episodes of abdominal myoclonic movement.

The dose of levetiracetam was increased to 500mg po bid, and clonazepam was reduced by 0.5mg every three days to taper until cessation of administration. The patient visited the clinic on 2/1/2020 and reported that the movement recurred again after the cessation of clonazepam administration. We increased the dose of levetiracetam to 1000mg po bid. At the follow-up visit on 26/1/2020, the patient reported that the movement had almost resolved after increasing the dose of levetiracetam on 2/1/2020.

Discussion

Belly Dancer's Dyskinesia is a complex hyperkinetic movement disorder characterized by abnormal and irregular movements of the diaphragm. The movements are usually bilateral, as in the present case, but can be restricted to one side,[5] and can also involve the abdominal muscle. This movement cannot be supressed voluntarily or attenuated with distraction, which is a distinguishing feature from other functional movement disorders. It disappears when the diaphragm is activated during respiration and is attenuated during sleep. The incidence and prevalence of this disorder has been estimated to be less than 8.5 cases per 100,000 people.[3]

The aetiology is idiopathic in most cases, but the condition has also been reported to occur secondary aetiology to abdominal surgery, osmotic demyelinating syndrome, spinal-cord tumour, tardive syndromes, compressive cervical radiculopathy, levo-dopa-induced movements and basal ganglia lesions.[6] The diagnosis is made clinically and work-up to confirm the diagnosis with diaphragmatic electromyography or ultrasound fluoroscopy to observe the irregular diaphragmatic contractions can be necessary in certain cases.[5] Other diagnostic tests aim to rule out secondary causes and include brain and spinal MRI to rule out basal ganglia or spinal cord lesions and echocardiography to rule our phrenic nerve irritation. The treatment is symptomatic and includes the administration of benzodiazepines. Clonazepam was used in the present case following the results of a prior case report.[6] This resulted in initial improvements, but when the drug was tapered, movement recurred. We decided to administer levetiracetam because it is effective for cortical and subcortical myoclonus and it has fewer side effect compared to benzodiazepine;[7] our patient responded well to levetiracetam at a dose of 1000mg po bid. Other treatment options that have been reported in the literature include diazepam and diphenhydramine,[8] phenytoin,

carbamazepine, haloperidol, phrenic nerve block in resistant cases,[9] valproic acid, pimozide, clonidine and fluoxetine.[5] Invasive therapeutic options may be considered in extreme cases that do not respond to oral medication; for example, Alshubaili described four patients with serious cases of the condition, who received treatment with ultrasound-guided administration of botulinum toxin A.[10]

Conclusion

Belly Dancer's Dyskinesia is an extremely rare form of axial myoclonus which can be a challenge to diagnose. Generally, examinations aim to rule out other secondary aetiologies. Multiple therapeutic options have been described previously, and here we want to highlight a case of Belly Dancer's Dyskinesia improved with Levetiracetam.

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CONFLICTS OF INTEREST

The authors declare that they have no competing interests.

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PATIENT CONSENT

The author, *Mohammed A*, declares that:

1. They have obtained written, informed consent for the publication of the details relating to the patient(s) in this report.
2. All possible steps have been taken to safeguard the identity of the patient(s).
3. This submission is compliant with the requirements of local research ethics committees.

Figures and Tables

Video 1:

Video 1.mov