# CASE REPORT A Case Report of Belly Dancer Dyskinesia in a 54 Years Old Female: Gastroenterology Meets Neurology

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Abstract: Dyskinesia limited only to the axial musculature is very rare, and if it is localized to the anterior abdominal wall, it is called belly dancer's dyskinesia. Despite reports of variable clinical characteristics, a common feature is a myriad of involuntary, repetitive, sometimes painful, and often

rhythmic movements of the anterior abdominal wall, with the majority being bilateral, resembling that of a belly dancer. As the symptom is mainly localized to the abdomen, patients could visit a gastroenterology service thinking it might be associated with underlying visceral pathology. Since the first report in 1990, only a few cases of belly dancer dyskinesia have been reported over the years. We herein report the case of a 54 years old female who presented to our OPD with a recurrent painless writhing movement of the abdomen, diagnosed as belly dancer dyskinesia and successfully treated with chlordiazepoxide.

Keywords: Africa, Ethiopia, rare, movement disorder, diaphragmatic flutter

#### Introduction

Focal dyskinesia affecting the abdominal wall is a rare phenomenon, first described by Iliceto et al and named "belly dancer's dyskinesia.<sup>1,2</sup> Belly dancer dyskinesia (BDD), also known as diaphragmatic flutter, is characterized by rhythmic, involuntary contractions of the diaphragm resulting in undulating, rhythmic movements of the abdomen resembling a belly dance. Even though both central and peripheral causes have been implicated as etiologic factors, most cases are idiopathic. A preceding history of abdominal procedures like cholecystectomy or appendectomy has been reported.<sup>2</sup> Temporal associations of neuroleptic drugs, like levodopa, galantamine, prochlorperazine, and clebopride, with the onset of symptoms of BDD, have been found in some cases.<sup>3–6</sup> During pregnancy and post-partum periods, its occurrence has been attributed to thoracic cord compression by the gravid uterus.<sup>7</sup> Centrally, the respiratory center of the rostral medulla has also been implicated in its pathophysiology.<sup>4</sup> Even though the diagnosis is always clinical, it can be supported by fluoroscopy and diaphragmatic electromyogram (EMG) showing rhythmic contractions of the diaphragm.<sup>8</sup> There is currently no evidence-based treatment recommendation.<sup>9</sup>

# Case Report

We herein report a fifty-four-year-old female patient who presented to our OPD at Adera Medical Center in Addis Ababa with a history of a painless writhing type of abdominal movement of 6 years duration. The movement presents almost daily and is relieved upon fasting. She has an associated history of indigestion and bloating of the same duration. The onset of symptoms started after cholecystectomy six years back. There is no history of trauma to any part of the body and no prior similar abnormal body movement, gait, or slurred speech. She was not on any form of medication on the index visit. Prior treatments at local health centers with unspecified medications were not effective. She is a mother of eight children, with all her pregnancies being uneventful. On physical examination, her vital signs are stable Neurologic

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examination revealed a continuous, rhythmic, undulating abdominal movement in all positions (Video). The movement did not respond to respiratory maneuvers and distractions. All her investigations, including complete blood count, liver function test, renal function test, random blood sugar, C reactive protein, erythrocyte sedimentation rate, HCVAb, HBsAg, serum albumin, urinalysis, serum vitamin-D level, and abdominal ultrasound, were normal. With the diagnosis of functional dyspepsia and belly dancer's dyskinesia, she was put on amitriptyline 25 mg PO/day (nocturnal) for 2 months, otilonium bromide 40 mg PO/day for 1 month, pantoprazole 40 mg PO/day for 4 weeks, and chlordiazepoxide and clidinium 7.5 mg PO/day for 1 month. Upon the next follow-up after a month, she was doing well, and the symptoms were controlled. The symptoms were better controlled on subsequent visits, and she was functioning well.

### Discussion

BDD is an unusual focal dyskinesia of the abdomen characterized by a repetitive semi-rhythmic multidirectional displacement of the umbilicus with a writhing movement of the anterior abdomen.<sup>1</sup> Antecedent events of local trauma or surgical procedures of the abdomen (appendectomy, laparoscopy, major surgery, etc.) may be present in half of the cases.<sup>2</sup> Various etiologic factors have been reported to cause BDD. Patients on antipsychotic medications have presented with features of BDD. Neuroleptic drugs acting on dopaminergic receptor agonists, and antagonists have also been shown to induce belly dancer's dyskinesia in a number of cases.<sup>3–6</sup>

Even though the pathogenesis of BDD is not clearly understood, central or peripheral nerve lesions that result in hyper-excitability of the respiratory center of the rostral medulla are implicated in the pathophysiology.<sup>4</sup> A report by Roggendorf et al attributes an ongoing but deficient striatal reorganization process to be the culprit in the pathogenesis of BDD, in a case of central pontine myelinolysis.<sup>10</sup> Deficient total body magnesium resulting in an increased acetylcholine release and neuromuscular excitation and hyperactivity of alpha and gamma motor neurons was reported to be central to the pathogenesis.<sup>11</sup> However, in our case, both serum electrolyte and imaging are normal.

BDD diagnosis is solely clinical and often delayed due to its rarity and lack of gold standard diagnostic modality.<sup>9,12</sup> Fluoroscopy and electromyography investigation of the diaphragm has been suggested to be supportive tools in making a diagnosis.<sup>12</sup> However, their use is only supplementary, and a clinical evaluation remains central to the diagnostic algorithm.

Even though different treatment modalities have been suggested, an evidence-based effective and safe management modality has not been identified. The use of benzodiazepines like clonazepam or diazepam has been effective in a few cases.<sup>12</sup> In others, the use of clonazepam has failed to resolve the condition.<sup>13</sup> There is one report of symptomatic improvements upon treatment with magnesium sulfate.<sup>11</sup> Benzodiazepines and magnesium depress the excitability of muscle fiber membrane by modulating the affinity of GABA receptors.<sup>14,15</sup> Thus, producing a symptomatic response in BDD, as witnessed with chlordiazepoxide in our patients. Effective management with invasive techniques like botulinum toxin injection and transcutaneous electrical nerve stimulation (TENS) has also been reported.<sup>6,16</sup>

# Conclusion

BDD is an extremely rare, poorly understood phenomenon that significantly affects a patient's quality of life. Even though the diagnosis is clinical, treatment can be challenging due to a lack of effective evidence-based treatment strategies. Thus, it is crucial for clinicians to be aware of the symptoms and develop a high index of suspicion for prudent evaluation and management of patients.

# **Ethical Statement**

No institutional approval was required to publish the case details. The patient has provided consent to publish the case report and accompanying images.

# **Consent Statement**

The authors have obtained all appropriate patient consent forms. The patient provided informed consent to publish their case details and accompanying video images.

Sincere gratitude to the patient and her family as well as our colleagues who were involved in the care of the patient.

# Disclosure

The authors report no conflicts of interest in this work.

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