SAGE Open Medical Case Reports
Volume 11: 1–3

DOI: 10.1177/2050313X231177751
journals.sagepub.com/home/sco
Sage

A case report and a brief literature review of belly dancer's dyskinesia in a pregnant patient

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Abstract

Belly dancer's dyskinesia is a rare movement disorder that can be classified under hyperkinetic movement disorders. It is characterized by rhythmic or semi-rhythmic contractions of the diaphragm and other abdominal muscles that are brief and involuntary that cannot be voluntarily suppressed but could be influenced by respiratory maneuvers. Belly dancer's dyskinesia in pregnancy even rarer, there have only been five reported cases. Here, we reported 19-year-old Ethiopian pregnant women who presented with oscillating movements of the abdomen that occurred at her ninth month of pregnancy. The general medical and neurological examinations were unremarkable. Complete blood count, basic metabolic panels, and biochemistry tests were all within the normal range. The patient responded to the trial of valproate with complete resolution of the abdominal dyskinesia after delivery.

Keywords

Belly dancer's dyskinesia, pregnant, Ethiopia

Date received: 1 February 2023; accepted: 8 May 2023

Introduction

"Belly dancer's dyskinesia" (BDD) is one of the peripherally induced hyperkinetic movement disorders.[1] It is one of the rarely encountered movement disorders with few reported cases in the literature. It is characterized by a waving or oscillating movement of the abdomen brought by a rhythmic or semi-rhythmic, involuntary contractions of the diaphragm, and other abdominal wall muscles. It resembles a belly dance, hence the name "belly dancer's dyskinesia."

It was described for the first time in 1716 by Antony van Leeuwenhoek, who himself has suffered from the disorder.[1] It was described as a "violent movement" of the diaphragm. The BDD nomenclature was coined for the first time by Iliceto et al.[2] on his article that was published in 1990, where he presented a summary of five similar dyskinesia cases. In 2011, a compilation of previous studies done amounting to 48 reported cases spanning from 1920 to 2011 was presented by Patterson.[3] Only a few cases have been reported in the literature ever since.

BDD in pregnancy is even rarer, there have only been five reported cases.[4–7]

Currently, there is limited study regarding this condition, and our patient would be the second reported case from Ethiopia.

Case summary

A 19-year-old primigravid lady on her 38th week of gestation from a reliable last normal menstrual period (LNMP) presented with 8-h history of intermittent semi-rhythmic repetitive waving/oscillating involuntary movements of the

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abdominal wall, which are rapid with brief interruptions. There is no migration to or involvement of the other parts of the body. There was no associated pain, but some discomfort was accompanying the movements; she reported that it sometimes hurt around the area below the subcostal margin of the right abdomen after prolonged movement episode. Otherwise, she had no associated dyspnea or fatigue and has no burning or tingling sensation over the abdomen. There was no associated difficulty breathing or shortness of breath. There were no specific precipitating or alleviating factors, and the patient was unable to voluntarily suppress the contractions. It is not relieved on sleeping and reported sleep disturbance.

She denied any history of trauma, spinal or abdominal surgeries, or personal or family history of similar episodes. Drug history was unremarkable. Careful psychiatric assessment showed no evidence of depression, anxiety, or psychosocial stressors. General exam was normal. There were no signs of fetal distress. Neurological examination revealed normal higher mental functions, cranial nerves, as well as motor and sensory functions except for the abdominal movements. She has an involuntary undulating movements of the abdominal wall muscles, in a wave-like fashion that rolled across the abdomen, predominantly involving the upper part of the abdomen (https://youtu.be/1EoQ2yrzu64). The movements did not show any variation in intensity or frequency in different positions nor during respiration (Supplemental material).

Basic laboratory tests, including complete blood count (CBC), serum electrolytes (Na, Ca, K, and Mg), liver enzymes (alanine transaminase (ALT), aspartate transaminase (AST), and alkaline phosphatase (ALP)), and serum bilirubin level, serum creatinine level, blood urea nitrogen (BUN) level and thyroid function tests (thyroid stimulating hormone (TSH), free thyroxine (FT4), free tri-iodothyronine (FT3)) were normal.

Thereafter, a clinical diagnosis of BDD was made. The patient was initially started on diazepam 10 mg intravenous (IV) PRN (as needed) and subsequently valproic acid 200 mg PO BID is orally, twice a day was added. With this, she had significant improvement in terms of frequency and degree of the abnormal movement. After 2 days from the onset of these complaints, she gave birth via spontaneous vaginal delivery with good outcome of both the baby and the mother. Subsequently, the abnormal abdominal movement subsided completely after delivery. Valproic acid was discontinued 4 days after delivery and the patient was then discharged home with an appointment. After 2 weeks, she was seen at our neurology follow-up clinic and there was no recurrence of the abdominal movement or any other complaint and hence was given a longer follow-up re-appointment.

Written informed consent was obtained from the patient for publication of this case report and any accompanying images/videos.

Discussion

BDD is one of the peripherally induced hyperkinetic movement disorder that is rarely encountered in clinical practice and hence less studied with paucity of evidences describing the exact etiologies, pathophysiology and effective treatment. It is characterized by brief, involuntary repetitive rhythmic or semi-rhythmic contractions of the diaphragm, and abdominal muscles resulting in a waving or oscillating movement of the abdomen resembling a belly dance. It cannot be voluntarily suppressed although influencing could be possible by breath holding, counting routines or other distraction techniques; resulting in variation in intensity or frequency diagnosis is mainly made by clinical examination, but at times can be challenging.

Several etiologies have been implicated in the literatures. A common cause is tardive dyskinesia commonly in those patients who have history of exposure to dopamine antagonists or neuroleptics. Dyskinesia also may be due to spinal segmental myoclonus.[8] Structural lesions like syringomyelia, myelitis, spinal cord trauma, vascular lesion, or malignancy with a common resultant segmental abdominal myoclonus may result in dyskinesia.[9] A lesion disrupting inhibitory spinal interneurons functioning or a structural reorganization of local neuronal circuits has been suggested as a possible underlying pathophysiology of BDD.[2] Even though their pathophysiology has not been understood, metabolic causes, such as hyponatremia and its consequence of myelinolysis, have also been postulated as etiologies. Some cases, however, were classified as idiopathic or functional.[4] Local compressive or hemodynamic changes in the thoracic cord or roots from a gravid uterus have been suggested as a possible cause in BDD in pregnancy.[8]

In our case, clinical and laboratory examinations failed to identify a definitive etiology. The lack of further neurophysiological tests, including multi-channel electromyography (EMG), is among the limitations. These studies could help to identify the pattern and specific muscle groups involved. Electroencephalogram (EEG) as well was not done as seizure is an important differential diagnosis in this particular patient;[10] the longer duration and stopping of the abnormal movement after delivery, absence of other body part involvement or associated change in mentation, might make a seizure less likely.

The management of BDD could at times be challenging. The treatment strategies or options should depend on the underlying etiologies and include surgical approaches and symptomatic treatment. As described above in short of understanding the etiologies and pathophysiology current treatment paradigms remain based on anecdotal evidence.[4] Among the list of medications, which has been used for treatment of BDD is included benzodiazepines, trihexyphenidyl, valproate,[2] and carbamazepine.[11] Both clonazepam and levetiracetam as well have been reported to be effective.[10] Botulinum toxin can also be helpful. Bilateral deep brain

stimulation of the globus pallidus interna has also been reported in one patient and the response was very good.[12]

Our patient achieved significant improvement with sodium valproate.

Conclusion

A pregnant woman on her third trimester presented with an abnormal body movement involving the abdomen and was diagnosed with BDD without any attributing underlying pathological finding. This is a rare type of movement disorder with no clearly understood etiologic agent, pathogenesis, and effective treatment. To understand the etiology and possible mechanism or pathogenesis as well as to determine the best effective treatment of similar cases a large-scale study and thorough investigation is important.

Acknowledgements

The authors would like to extend their gratitude to the patient for her cooperation. They would also like to acknowledge all the nursing staff, treating physicians, and supporting staff involved in the management of their patient.

Author contributions

All the authors were involved in concept design for the manuscript, manuscript preparation, critical analysis, and revision and involved in the management of the patient.

Availability of data and materials

All data sets on which the conclusions of the case report based, to be available as a medical record document and available from the corresponding author on reasonable request from the editors.

Consent to publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images/videos.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The author(s) received no financial support for the research, authorship, and/or publication of this article.

Ethical approval and consent to participate

The authors' institution does not require ethical approval for the publication of a single case report.

Informed consent

Written informed consent was obtained from the patient for their anonymized information to be published in this article.

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Supplemental material

Supplemental material for this article is available online.

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