Case Report

BELLY DANCER'S DYSKINESIA: A RARE PHENOMENON

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ABSTRACT

Belly dancer's dyskinesia (BDD), also known as diaphragmatic flutter or diaphragmatic myoclonus is a distinctive and rare movement disorder characterized by involuntary and rhythmic movements of the abdominal muscles resembling belly dance. This phenomenon has a variable clinical presentationthereby making it difficult to diagnose. It is observed in various clinical scenarios, including post-stroke, psychiatric, and chronic neurological disorders, and as adverse effect of certain medications.A high index of suspicion is required in a patient with symptoms including abnormal abdominal wall movements, dyspnoea and respiratory distressfor an early diagnosis of this entity. We report an unusual case of BDD secondary to 5HT3-antagonist use in a patient with multidrug resistant tuberculosis (MDR TB).

Keywords: Belly dancer dyskinesia (BDD), Multidrug Resistant tuberculosis, Ondansetron induced BDD, Diaphragmatic flutter, Diaphragmatic myoclonus

INTRODUCTION

Belly Dancer's Dyskinesia (BDD) also known as diaphragmatic flutter or diaphragmatic myoclonus, is a unique phenomenon which involves focal dyskinesia affecting the abdominal wall and is characterized by involuntary rhythmic undulating abdominal wall movements resembling a belly dance[1]. A few specific anti-emetics, such as domperidone and metoclopramide, were previously associated with this rare condition however the exact cause remains unclear. Previous reports have identified BDD following abdominal surgeries (such as cholecystectomy or appendectomy), in neurodegenerative and metabolic brain diseases, spinal tumors, pregnancy (likely due to thoracic cord compression by the gravid uterus), and secondary to drug (e.g., L-Dopa and antidopaminergics like quetiapine and prochlorperazine, as well as other agents like domperidone, clebopride, and salbutamol)[2]. However, no previous reports have documented the occurrence of BDD in patients with Multi-drug resistant Tuberculosis (MDR-TB).

India accounts for the highest number of drug resistant TB cases[3]. MDR-TB is characterized by resistance to two highly effective first-line drugs, rifampicin and isoniazid. Consequently, MDR-TB treatment involves second-line drugs, which are more toxic and are associated with more severe side effects compared to first-line medications. Drug-related gastritis is a common side effect in first- and second-line anti-tuberculous treatment (ATT). Management of ATT-related gastritis typically involves the administration of anti-emetic

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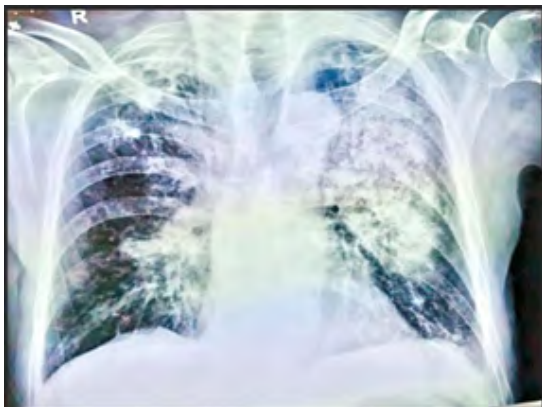
medications and proton pump inhibitors (PPIs). In this report, we present a case of BDD in a patient with MDR-TB who received ondansetron for ATT-induced gastritis. This case highlights the association between ondansetron usage in MDR-TB patients and the development of BDD, adding to the limited understanding of this rare movement disorder in the context of tuberculosis treatment.

CASE REPORT

A 29-year-old male presented with presented with fever with evening rise of temperature associated with night sweats, productive cough with white to yellowish non-foul smelling mucopurulent expectoration associated with occasional hemoptysis and unintentional unquantified weight loss of 3 weeks duration. His chest radiograph revealed non homogenous opacities in bilateral upper zones, left middle zone and right lower zone **(Image 1)** and his sputum was positive for AFB (2+ WHO grading) with molecular testing elucidating drug resistance (MTB GeneXpert – MTB detected medium, rif resistance detected, 1st line LPA – Kat G Mut, rpo B Mut present, 2nd line LPA – No FQ or SLI resistance detected).



On laboratory evaluation, he was found to have anaemia of chronic disease Hb – 11.2 g/dl, erythrocyte sedimentation rate – 40 mm fall/ 1st hr, and rest parameters were within normal limits. He was started on shorter injectable regimen (High dose moxifloxacin, Amikacin, Ethionamide, Clofazimine, pyrazinamide, high dose isoniazid, and ethambutol) and was discharged from the hospital. Six weeks into treatment, he complained of epigastric burning with eight episodes of vomiting, and stomach fullness over the last two days. He was diagnosed with ATT-induced gastritis (other causes were ruled out) and managed with oral ondansetron (8 mg). After three doses of oral ondansetron 8 hours apart patient became symptomatically better. However, he developed rhythmic, episodic involuntary movements of the abdominal muscles, like belly dancing. These movements lasted for several seconds and occurred several times daily (video). The patient did not report any pain, discomfort, or functional impairment associated with the movements. A neurological examination was conducted, which was essentially normal. The patient's serum electrolytes, liver function, and renal function were within the normal range. An electroencephalogram (EEG) was also performed, which showed no evidence of epileptiform activity, and his Magnetic Resonance Imaging (MRI) Brain was also normal. The patient was observed for three days, during which the involuntary movements persisted even during sleep without any change in frequency, severity, or duration. Based on the clinical features of the involuntary movements, a diagnosis of belly dancer's dyskinesia was made, and ondansetron was withheld. He was also started on intravenous injection of lorazepam which is known to be effective in treating such movement disorders. With lorazepam and withdrawal of 5-HT3 antagonist, the involuntary movements completely disappeared, and the patient's clinical condition remained stable during the rest of his treatment for MDR-TB.

DISCUSSION

Iliceto et al[4] were the first to describe belly dancer's dyskinesia (BDD) as a rhythmic, involuntary, repetitive, and slow-writhing movement disorder affecting the anterior abdominal wall, with an unclear underlying cause. Despite the passage of many years, the pathogenesis of BDD remains poorly understood. Various hypotheses have been put forward to explain the condition, including dysfunction of spinal inhibitory interneurons or structural reorganization of local neuronal circuits, deficiency of total body magnesium leading to increased acetylcholine release and neuromuscular excitation, hyperactivity of alpha and gamma motor neurons, as well as hyperexcitability, degeneration, and/or neurotoxicity resulting from altered metabolism of spinal interneurons[4,5]. Etiologically, BDD can be categorized into idiopathic, central, peripheral, and functional types. Most BDD cases

fall under the idiopathic category, while central BDD is associated with brain or spinal cord lesions and may be influenced by medications regulating dopaminergic transmission. Clinically, central BDD, is typically bilateral and can be distinguished from rest by the persistence of symptoms during sleep in the presence of cerebral or spinal pathology[6,7] Among peripheral causes, phrenic nerve irritation is common, often originating from the heart, resulting in diaphragmatic flutter synchronous with systole[8]. Psychogenic factors are also frequently suspected, as symptoms are only present while the patient is awake[9]. Distractibility and breath-holding can help diagnose such cases. The diagnosis of BDD requires a comprehensive evaluation of the patient's medical history, including prior and current medication usage. While brain and spinal cord imaging can help rule out secondary causes, the diagnosis of BDD primarily relies on clinical examination, which may be delayed due to its rarity and the lack of a definitive diagnostic test. Fluoroscopy and electromyography investigations of the diaphragm have been suggested as supplementary diagnostic tools, but they do not replace the importance of clinical evaluation. Regarding treatment, beyond discontinuing substances that may contribute to BDD, various pharmacological therapies, such as benzodiazepines (e.g., clonazepam or diazepam), the sodium channel blocker levetiracetam, haloperidol, aripiprazole, etc., have been attempted with varying degrees of success. When pharmacological approaches prove ineffective, alternative options like botulinum toxin injection, transcutaneous electric nerve stimulation, and deep brain stimulation may be considered for management[10-14]. However, the prognosis of BDD remains highly unpredictable.

Treatment of drug resistant tuberculous can be extremely challenging as it involves numerous medications which have various drug interactions and adverse events. In addition, drugs like ondansetron are required to be added to counteract adverse events like acute gastritis which occurred in our patient. TB being a highly prevalent disease in our country with drug resistant TB contributing to more than 5% of the entire TB burden, the physicians need to be aware of such a disease. Being a drug induced entity as in our case, BDD can be treated easily and early identification can help avoid numerous unwanted investigations and can also help alleviate mental anxiety on an already distressed MDR TB patient.

In contrast to the existing literature, we propose that our patient developed central belly dancer's dyskinesia (BDD) due to ondansetron, as the symptoms persisted even during sleep. Notably, no previous case reports have documented BDD associated with ondansetron usage. Interestingly, our literature search revealed that ondansetron has been found to have a therapeutic role in tardive dyskinesia among patients with schizophrenia[15]. This is attributed to the 5-HT3 antagonistic properties of ondansetron, which counteract the inhibitory effect of dopamine on the firing of medial prefrontal neurons, thereby restoring normal dopamine function in these patients[16].

However, despite the rarity, occasional reports have highlighted the extrapyramidal side effects of ondansetron in post-market surveillance[17,18]. Although ondansetron does not directly affect dopamine, animal studies have suggested that it may inhibit or reduce dopamine activity within the mesolimbic pathway[19]. In our patient, a thorough evaluation did not reveal any other secondary causes for the dyskinetic movements observed. It is also worth mentioning that isoniazid can increased the level or effect of ondansetron through its effect on hepatic or intestinal CYP3A4 enzyme metabolism. However dose adjustment is not recommended[20]. The patient responded favourably to the lorazepam injection and ondansetron was stopped, and there was no recurrence of symptoms was noted after that.

CONCLUSION

In conclusion, belly dancer's dyskinesia (BDD) is a rare and poorly understood movement disorder affecting the anterior abdominal wall. Diagnosing BDD can be challenging due to the lack of a definitive diagnostic test and primarily relies on clinical evaluation. Physicians should maintain high suspicion when encountering patients with abnormal body movements. A thorough review of the patient's medical and drug history is crucial in identifying the potential cause of the dyskinesia. Additionally, our case report emphasizes the possibility of rare extrapyramidal side effects secondary to ondansetron. However,

further research is necessary to comprehend the underlying mechanisms of BDD better and investigate any potential link between ondansetron and the development of this movement disorder. The prognosis of BDD remains unpredictable, and management should be individualized to meet each patient's specific needs.

CONFLICT OF INTEREST

None

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