Belly Dancer’s Dyskinesia: 3 Cases of a Rare Entity

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Abstract
Involuntary, undulating, or circular movements of abdominal wall muscles are a rare condition descriptively termed belly dancer syndrome or belly dancer’s dyskinesia. Differential diagnoses range from cerebral, spinal, peripheral, hormonal/drug-induced effects to local or even unknown/idiopathic and functional/psychogenic causes. Thorough diagnostic work-up is mandatory and therapeutic options depend on etiology and include surgical approaches and symptomatic treatment. We present 3 cases of belly dancer’s dyskinesia, in which differential diagnostic work-up did not uncover known etiologies for this movement disorder. Strikingly, 2 patients manifested vitamin B12 deficiency while 1 case showed values close to the lower limit of normal. While relevance of vitamin B12 levels remains elusive, all patients improved substantially following a combined therapeutic approach of clonazepam treatment and vitamin B12 supplementation.

Introduction
Belly dancer’s sign was first published in 1976 as a result of phrenic palsy in a newborn [1]. The condition was termed belly dancer’s dyskinesia (BDD) and differentiated from diaphragmatic flutter in 1990, when Iliceto et al. [2] reported four adult patients with involuntary, writhing, partly painful movements of the abdominal wall without establishing a cause.
Since then, few case reports or small case series have been published describing involuntary, mostly bilateral movements of the abdominal wall reminiscent of the undulating movements of a belly dancer. BDD etiology appears to be diverse since cerebral, spinal, peripheral as well as local causes have been suggested. Some cases, however, were classified as idiopathic or functional [3]. There are reports of BDD in neurodegenerative [4] as well as metabolic [5] brain diseases. Moreover, it has been described as adverse event of deep brain stimulation [6]. Spinal tumors [7] induced BDD and it also occurred during pregnancy as well as after child delivery and abdominal surgery indicative of local neural irritation [8]. Interestingly, drug-induced BDD seems to be the most common form of this movement disorder with L-Dopa and antidopaminergics like quetiapine and prochlorperazine as well as other agents like domperidone, clebopride, and salbutamol having been identified [9–13]. Symptomatic therapy beyond withdrawal of accountable substances includes benzodiazepines like clonazepam or diazepam, the sodium channel blocker levetiracetam, botulinum toxin injection as well as transcutaneous electric nerve stimulation and deep brain stimulation [2–4, 8, 13–15]. Here, we present 3 cases of BDD in which differential diagnostic work-up did not uncover known etiologies for this movement disorder. However, all individuals exhibited abnormalities in their vitamin B12 levels. Following symptomatic treatment with clonazepam combined with vitamin B12 supplementation symptoms could be controlled effectively.

Case Presentation

Case 1

Upon admission to our neurological department in 2010 the 84-year-old male patient complained about increasing, involuntary contractions of the abdominal muscles for the last 2 months (Fig. 1; see online suppl. Video 1; for all online suppl. material, see www.karger.com/doi/10.1159/000521813). Initially, these contractions had a preference during rest and while falling asleep. Sleep was massively disturbed because muscle movements did not stop while sleeping. Meanwhile, contractions were also present throughout the day. Patient’s medication comprised of a betablocker, diuretic, statin, and aspirin plus dipyridamole. Clinically, besides the involuntary movements of the abdominal muscles on the right side, patient showed some cognitive decline and gait disorder resembling discrete signs of Parkinson's disease. Tendon reflexes of the legs were alleviated. MRI scans of the brain revealed a global atrophy and pronounced confluent vascular lesions in line with advanced vascular encephalopathy (Fig. 2). EEG ruled out ictal etiology. CT scans of thorax and abdomen did not show any signs of tumor or aortal dissection but substantial vasosclerosis (not shown).
Laboratory examinations confirmed vitamin B12 deficiency (204 pg/mL; normal range 208–964 pg/mL). Symptomatic treatment was initiated with clonazepam and vitamin B12 supplementation. BDD resolved instantly and did not reappear after termination of benzodiazepine treatment while signs of vascular Parkinson syndrome persisted.

**Case 2**  
This male patient was first examined in our neurological walk-in clinic in 2005. At that time, he was 52 years of age and complained of involuntary contractions of abdominal muscles when lying on his back or one side for at least 4–5 years duration. He related his complaints to a herniated lumbar disc L4/5 that had been diagnosed by that time. Medication comprised of an ACE inhibitor and β-blocker. Neurological examination revealed symmetric, myoclonic contractions of the rectus abdominis and adjacent abdominal wall muscles seconds after lying on his back (Fig. 3; see online suppl. Video 2). Cervical MRI scans showed only mild degeneration of discs without affection of the spinal cord. Thoracical MRI scans revealed...
widening of the central canal from vertebra D4 to D8 without signal abnormalities of the cord. One year later, the patient showed up again and reported complete disappearance of abdominal muscular movements following spinal anesthesia due to arthroscopy of the right knee. Six months thereafter, contractions gradually reappeared especially while falling asleep, but without sleep disturbances and less intense as before. Repeated spinal MRI scans did not show any new pathologies. CT scans of thorax and abdomen did not result in findings explaining reoccurrence of muscle contractions. Temporal clonazepam medication at that time could not resolve the problem to the patient's satisfaction and contractions gradually vanished without further intervention for several years. In 2016 and 2017, we saw the patient again, who reported deterioration of the known muscle contractions for about a year. Sleep was disturbed but during the day he was free of complaints. Medication had been unchanged, EEG remained normal. Extensive blood tests including electrolytes, micronutrients, thyroid-/parathyroid hormones, and NT-pro BNP as well as MRI scans of the brain remained without relevant findings. However, in retrospect, mean corpuscular volume of erythrocytes was substantially enlarged (102.5 fL; normal range 80.0–96.0 fL) at that time. Re-evaluation in June 2021 revealed sufficient dyskinesia control under continuous intake of clonazepam 1 mg once daily. Mean corpuscular volume of 101 fL still was high, while liver enzymes were not elevated. Consistent with latent vitamin B12 deficiency blood levels were close to the lower limit of normal (265 pg/mL; normal range 208–964 pg/mL), while methylmalonic acid was close to the upper limit of normal (28.0 μg/L; significant >30 μg/L). Substitution was recommended to the general practitioner with follow-up visit upon request.

**Case 3**

The 56-year-old male was admitted to our department in autumn 2020 due to unwillingly, writhing, undulating contractions of the abdominal wall muscles. The condition persisted for 2 years already, occurred especially while sitting or lying with no respiratory distress (Fig. 4; see online suppl. Video 3) but accentuation in strenuous situations. However, the patient described the contractions as uncomfortable and sometimes painful. They subsided during sleep. Patient's history included adiposity, hyperuricemia, hypercholesterinemia, type 2 diabetes as well as arterial hypertension. Neuroleptics or other psychotropic medication had never been taken. Besides BDD physical as well as neurological examination revealed no relevant findings. MRI scan of the neurocranium was within normal limits. Spine MRI demonstrated multiple degenerative changes like bulging discs and discs herniations. CT thorax displayed no mediastinal lesion especially none in relation to the phrenic nerve. Ultrasound examination of the abdomen showed no abnormalities except for...
a fatty liver. EEG as well as central motor latency indicated no abnormalities. Somatosensory evoked potentials from tibial nerve showed prolonged latency (47.0 ms; calculated upper limit with body height of 160 cm: 41.4 ms), while those from medial nerve were normal. Electromyogram of abdominal muscles was not done. Blood exams, however, revealed a significant deficit in vitamin B12 (201 pg/mL; normal range 208–964 pg/mL). Serum values for copper, vitamin B1, B6, E as well as folic acid were within normal range. Following symptomatic treatment with clonazepam in combination with vitamin B12 substitution subcutaneously resulted in complete symptom recovery within 1 week.

**Discussion/Conclusion**

BDD has been linked to numerous different etiologies like cerebral, spinal, peripheral, and local causes as well as hormonal/drug-induced effects. Though it is still a rare movement disorder presenting with undulating, writhing contractions of abdominal muscles, BDD requires extensive evaluation not only of the current status but also of the patient’s medical history including previous as well as current medication (compare diagnostic work-up in our cases summarized in Table 1). In particular, medications that regulate dopaminergic transmission have been reported to be associated with this dyskinesia [9–13]. However, in our cases, a history of dopamine-regulating medication could be ruled out. The same holds true for local trauma in our small cohort, rendering the debate, whether trauma can cause such a movement disorder or is a trigger for a functional disorder, unnecessary [3]. Of interest, in Case 2 of our series, dyskinetic movements had a timely association with falling asleep and awakening concomitant with an enlarged central canal in the spinal cord and improvement.
of the condition following spinal anesthesia. This is in line with the lack of BDD disappearance during sleep in case of cerebral or spinal pathology [3, 9]. On the other hand, BDD has also been related to metabolic alterations of the nervous system [5]. Involuntary hyperkinetic movements associated with vitamin B12 deficiency as well as supply have not only been described in children but also adults [16, 17], with positive therapeutic effects of vitamin B12 supplementation within days [17]. In this regard, two of the cases presented here showed relevant vitamin B12 deficiency and substantially improved following a combined therapeutic approach of clonazepam treatment and vitamin B12 supplementation. The pathogenesis associated with vitamin B12 deficiency is unknown. Possible mechanisms include hyperexcitability, degeneration, and/or neurotoxicity resulting from altered metabolism of spinal interneurons [17].

We conclude that belly dancer’s dyskinesia often shows amelioration following clonazepam treatment. Up to date, low vitamin B12 levels in this movement disorder remain a clinical observation of uncertain significance.

Table 1. Summary of diagnostic work-up

|                    | Case 1          | Case 2          | Case 3          |
|--------------------|-----------------|-----------------|-----------------|
| Vitamin B12        | Deficiency      | Lower limit of normal | Deficiency      |
| EEG                | Normal          | Normal          | Normal          |
| CT thorax/abdomen  | Vasosclerosis   | Normal          | Normal          |
| MRI spine          | Not done        | Mild cervical disc degeneration, widening of central canal D4–D8 | Disc degeneration |
| MRI head           | Atrophy, vascular encephalopathy | Not done | Normal |

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Statement of Ethics

The present research complies with the guidelines for human studies, and the research was conducted ethically in accordance with the World Medical Association Declaration of Helsinki. This retrospective review of patient data did not require ethical approval in accordance with local/national guidelines. For the publication of this case report and the accompanying pictures/video documentation, written consent was obtained from the patients (case 2 and 3) or from their next of kin (case 1 already deceased). They are part of the patient records archived in the hospital.

Conflict of Interest Statement

The authors declare that there are no conflicts of interest relevant to this work. During the last 3 years, P.K. speaker’s honoraria from Biogen, Boehringer Ingelheim, Daiichi Sankyo, Pfizer, and Roche.
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Author Contributions

K.R.: data collection and manuscript review. J.H.: data collection and manuscript review. N.M.: data collection and manuscript review. R.R.: project outline, data collection, and manuscript review. P.K.: project outline, data collection, and writing of the first draft.

Data Availability Statement

All data generated or analyzed during this study are included in this article and online supplementary material files. Further enquiries can be directed to the corresponding author.

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