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Case of a young girl with hemichorea induced by oral contraceptive pills

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INTRODUCTION:

Chorea is an involuntary movement disorder derived from the Greek word “dance”. It is characterized by abrupt, irregular, non-repetitive, non-rhythmic movements with dance-like appearance, moving from one body part to another. When chorea involves half of the body then the term hemi-chorea is used, which usually signify structural pathology in the contralateral Basal ganglia. Use of Oral contraceptive pills, is a well-established but a rare cause of hemi-chorea. Mechanism includes either re-activation of Sydenham’s chorea or drug induced lupus erythematosus. Although our case does not have any systemic manifestation of lupus but ANA positivity likely favors later mechanism. We described a case of a young girl who developed right sided hemi-chorea after 2 months of oral contraceptive pills which were prescribed for delayed menarche. Other causes of hemi-chorea were excluded on the basis of extensive blood workup and normal MRI brain. Her pills were stopped immediately and she received dopamine receptor antagonists. Follow up in clinic showed significant clinical improvement after 4 weeks.

KEYWORDS:

Hemi chorea, Oral contraceptive pills, Young.

CASE REPORT:

A case of 14 year old girl presented in Emergency department with the insidious onset of involuntary movements of right upper and lower limbs for 2 days. From one part of the limb to another. Her mother added that these movements were absent during sleep. There was no family history of abnormal movements. There was no history of any antecedent streptococcal infection, joints pain, oral ulcers or rash. She was taking oral contraceptive pills(combination of estrogen and progesterone) for the past 2 months, for the management of delayed menarche. Other causes of hemi-chorea were excluded on the basis of extensive blood workup and normal MRI brain. Her pills were stopped immediately and she received dopamine receptor antagonists. Follow up in clinic showed significant clinical improvement after 4 weeks.

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Her MRI Brain was unremarkable. ASO titers and thyroid profile were within normal limits. Autoimmune workup showed ANA (antinuclear antibody) positive while antidsDNA, anti-cardiolipin antibodies, antiphospholipid antibodies, ENA profile, anti-thyroglobulin, anti-thyroid peroxidase were negative.

Her oral contraceptive pills were stopped. She was started on dopamine receptor antagonists (Olanzapine 5 mg daily) and benzodiazepine (Clonazepam 0.5 mg at night). Her symptoms improved and she followed up in clinic with the complete resolution of symptoms after 3 weeks.

**DISCUSSION:**

Chorea is an involuntary movement disorder derived from the Greek word “dance”. It is characterized by abrupt, irregular, brief, non-stereotyped movements. Chorea can affect various body parts, and interfere with speech, swallowing, posture and gait. It may worsen with anxiety or voluntary movements, and subsides during sleep. Chorea may be focal or generalized. When involves one side of the body then term hemichorea is used. Causes of hemichorea includes focal vascular lesions in the basal ganglia, infection or immune-related disorders such as Sydenham's chorea and systemic lupus erythematosus, Various metabolic and endocrinological disorders such as hyperthyroidism, hypo/hyperparathyroidism and hypo/hyperglycemia, drugs such as levodopa, neuroleptics and oral contraceptive pills.

In 1966, Fernando reported the first description of chorea associated with use of OCPs, till now total of 24 cases have been described with chorea and use of oral contraceptive pills. The mechanism of oral contraceptive-induced chorea is unknown, but clinical and experimental data suggest that it involves altered central dopaminergic activity. GABA is the neurotransmitter responsible for the inhibitory pathway especially in the indirect and direct pathway in basal ganglion. In the indirect pathway, the interruption of GABAergic transmission from the striatum to the external segments of the globus pallidus (GPe) would cause abnormally increased GPe neuron inhibitory activity on the subthalamic nucleus (STN) (1). Increased inhibition on STN would decrease its excitatory action on the internal segments of the globus pallidus (GPI), which would lead to decreased GPI neuron inhibitory action on thalamus. The decreased inhibition on thalamus would lead to increased excitatory action on cortex. Besides excitatory STN inputs the GPI neurons also receive inhibitory afferent inputs directly from the pathway. The imbalance between the indirect excitatory and direct inhibitory pathways ultimately leads to a disinhibition of the motor thalamus and caused the motor cortex over excited (2,3)

Review of five cases of chorea in patients receiving estrogen containing contraceptives showed that two patients had an episode of Sydenham Chorea, one had Henoch-Schönlein purpura and two had congenital heart disease. Chorea resolved in all patients upon discontinuing the medication. Patients with pre-existing striatal abnormalities appear more susceptible to oral contraceptive induced chorea (4,5). In our case we didn't find any striatal abnormalities on neuro-imaging. Literature search also revealed that underlying mechanism in oral contraceptive pills induced chorea includes either re-activation of Sydenham's chorea or drug induced lupus erythematosus. Although our case do not have any systemic manifestations of lupus but ANA positivity likely favors later mechanism. Four cases have been reported in Japan with OCPs related lupus erythematosus (6). Drug induced lupus erythematosus (DILE) has been recognized as a side effect of therapy with 80 drugs since its first description in association with sulfadiazine in 1945. It is defined as the development of lupus-like symptoms (commonly fever, musculoskeletal involvement and serositis) that is temporally related to continuous drug exposure (1 month) which resolves with cessation of the offending drug. It is usually accompanied by serologic findings of a positive antinuclear antibody (ANA). The clinical manifestations may be limited as in our case or systemic.

**REFERENCES:**


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Saba Zaidi; data collection, data analysis, manuscript writing, manuscript review