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Case Report

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The Occurrence and Curing of Stuttering During The Use of Methylphenidate for ADHD in Childhood: An Inconsistent Relationship

Çocukluk Çağında DEHB Tanısı ile Metilfenidat Kullanımı Sırasında Ortaya Çıkan ve Sonlanan Kekemelik: Tutarsız Bir İlişki

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ABSTRACT

Stuttering is a childhood onset fluency disorder in speech. It has been suggested to be associated with brain anatomy, functionality or the dysregulation of the dopaminergic activity. Attention deficit hyperactivity disorder (ADHD) is one of the most common psychiatric disorders in childhood and adolescence. Psychostimulant medications are widely used for the psychopharmacologic treatment of ADHD. In this article, two boys who have ADHD with opposite reactions after methylphenidate (MPH) treatment in terms of fluency in the speech are presented. In the first case stuttering was induced with MPH while in the second case stuttering was resolved after MPH treatment. Although MPH seems to have opposite effects on stuttering in our cases, our case report points that dopaminergic system may play a role for the onset of childhood onset fluency disorder. More comprehensive studies are needed to show the neurodevelopmental process and the mechanisms related with ADHD, stuttering and MPH treatment.

Key Words: Methylphenidate; Attention Deficit/ Hyperactivity Disorder, Stuttering, Neural Dynamics, Dopamine, Child

ÖZ

Kekemelik çocukluk döneminde başlayan konuşmada akıcılık bozukluğudur. Beyin anatomisi, işlevselliği veya dopaminerjik aktivitenin düzensizliği ile ilişkili olduğu öne sürülmüştür. Dikkat eksikliği hiperaktivite bozukluğu (DEHB), çocukluk ve ergenlik döneminde en sık görülen psikiyatrik bozukluklardan biridir. DEHB'nin psikofarmakolojik tedavisinde psikostimülan ilaçlar yaygın olarak kullanılmaktadır. Bu makalede, metilfenidat (MPH) tedavisi sonrası konuşma akıcılığı açısından birbirine zıt tepkiler veren DEHB'li iki erkek çocuk sunulmaktadır. İlk vakada kekemelik MPH ile indüklenirken, ikinci vakada kekemelik MPH tedavisinden sonra çözüldü. Olgularımızda MPH'nin kekemelik üzerinde birbirine zıt etkileri varmış gibi görünmektedir, olgu sunumumuz çocukluk çağı başlangıçlı akıcılık bozukluğunun ortaya çıkmasında dopaminerjik sistemin rol oynayabileceğine işaret etmektedir. DEHB, kekemelik ve MPH tedavisi ile ilgili nörogelişimsel süreci ve mekanizmaları göstermek için daha kapsamlı çalışmalara ihtiyaç vardır.

Anahtar Kelimeler: Metilfenidat; Dikkat Eksikliği/Hiperaktivite Bozukluğu, Kekemelik, Nöral Dinamikler, Dopamin, Çocuk

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Introduction

Stuttering is a speech disorder which may cause deficits in the fluency and timing of the speech. Current data suggest that stuttering may be related with central nervous system dysfunction which effects speaking fluently.[1] Lifetime prevalence of stuttering was reported as 5%.[2] Stuttering has been labelled as "childhood onset fluency disorder" and classified in ''neurodevelopmental disorders" according to the Diagnostic and Statistical Manual of Mental Disorders, 5th edition (DSM-5).[3] Neurodevelopmental disorders emerge in early childhood because of the atypical development of central nervous system.[4] The emergence of stuttering is usually between 25th and 48th months in childhood period. Up to 90% of the cases may recover until adulthood naturally.[1] However, stuttering has been reported to be associated with lower quality of life, higher levels of anxiety, emotional and behavioral problems.[1,2]

Attention deficit hyperactivity disorder (ADHD) is a neurodevelopmental disorder characterized with inattention, hyperactivity and impulsivity. Estimated prevalence of ADHD varies between 6.7% and 12%.[5] Pharmacological agents, psychosocial/behavioral interventions and combined treatments have been widely used for the treatment of ADHD. Imbalance in the dopaminergic and noradrenergic systems in the prefrontal cortex has been suspected for the etiology of ADHD.[5] Psychopharmacologic agents (such as methylphenidate and atomoxetine) that modulate dopaminergic and noradrenergic systems are the first line pharmacologic treatments in ADHD.[6]

Here, we report two cases, one whose stuttering began with MPH therapy and the other whose stuttering resolved with MPH therapy.

Case Presentation

Case 1: A nine year old boy was referred to our clinic by his parents due to hyperactivity, impulsivity, problems engaging with his peers, inattentiveness and failure at school. He had problems to focus on tasks and sustain attention. His psychomotor activity was elevated, impulse control and frustration tolerance was decreased. There was no history of any systemic physical illness, epilepsy or hospitalization in the past. He has achieved psychomotor developmental milestones on time. He was diagnosed with ADHD in our clinic according to DSM-5 criteria.

After diagnosing ADHD, behavioral therapy, psychoeducation and extended release formulation (osmotic controlled-release oral delivery system-OROS) of MPH 18mg/day were initiated. OROS-MPH dose was titrated to 27mg/day and 36mg/day in the second and third interview, respectively and significant recovery was observed. However, after increasing the dose of OROS-MPH to 36mg/day the child's family began to complain about stuttering. There have been no symptoms of stuttering earlier. Moreover, there was no family history of stuttering. They reported that first signs of stuttering have been appeared after increasing the dose to 27mg/day and after taking 36mg daily dose of OROS-MPH stuttering became more severe and frequent. The symptoms were described as not using the breath appropriately. A full medical and neurological work out was done and reported to be normal. These signs have raised the question if there was a relationship between OROS-MPH and symptoms of stuttering. To check this relationship OROS-MPH was stopped. 4 days after cessation the OROS-MPH treatment stuttering resolved while ADHD symptoms were remitted. The patient tolerated the medication well without any side effects that were commonly seen with OROS-MPH.

Case 2: A 7 year old boy who was admitted to our clinic due to behavioral problems at school. His teacher referred him for a psychiatric evaluation for hyperactivity, inattentiveness, difficulty in listening lessons and completing homework. He had had problems with his peers because of his impulsive behaviors and hyperactivity. There was no history of any systemic physical illness, epilepsy or hospitalization in the past. There have been problems in the fluency of the speech for two years before admission. He was diagnosed with ADHD and childhood onset fluency disorder according to the DSM-5. 10mg/day MPH was initiated and the dose was titrated to 20 mg/day in the follow up. Symptoms of ADHD have been improved according to his parents with the dose of 20mg/day in addition to the recovery of stuttering. During the period of COVID-19 pandemics his parents stopped MPH treatment by themselves. After cessation of the oral MPH, symptoms of stuttering appeared again. After the initiation of MPH again, stuttering was disappeared along with the improvement in the symptoms of ADHD.

Discussion

The literature about the association of MPH and stuttering is controversial. A significant relationship was reported between the symptoms of stuttering and MPH treatment.[7,8] In a placebo-controlled trial Rabaeys et al. (2015) found significant decrease in the frequency of stuttering with MPH treatment.[9] Moreover, in a case report it was reported that ongoing stuttering of an 18 year old man from early childhood was resolved after MPH treatment.[10] On the other hand, occurrence of stuttering has been reported under treatment with other dopaminergic agents such as Levodopa and Bupropion.[11,12]

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Wu et al (1997) found increased dopaminergic activity at medial prefrontal cortex, deep orbital cortex, insular cortex and auditory cortex in patients who have stuttering. And they suggested that stuttering has been associated with the brain regions that modulate speech.[13] It has been suggested that stuttering and related motor movements may be due to the dysfunction in the basal ganglia or a general dysfunction of the dopaminergic system.[14] Neuroimaging studies have found differences in the brain anatomy and functionality-especially in motor and auditory regions- in children with stuttering.[15] However, the exact effect of the dopaminergic dysfunction in stuttering is not clear. In some cases neuroleptic agents that have anti-dopaminergic effects have been used for the treatment of stuttering[16,17,12]], while in some cases stuttering was induced by antipsychotic agents.[18,19,20] The pathophysiological mechanisms of ADHD, stuttering and MPH activity have not been fully elucidated, but they are all associated with the dopaminergic system.[7]

In addition, children with stuttering have been reported to have more ADHD symptoms than children without stuttering.[21] However, in our cases, the main complaints for their admission to the clinic were ADHD symptoms. In the first case OROS MPH induced stuttering. However, interestingly, stuttering was recovered after MPH treatment in the second case. In this case, improvement in stuttering was observed under long-acting methylphenidate therapy. However, no explanation can be given as to why/how MPH is responsible for the emergence of stuttering in one case and for the decrease in stuttering in the second case. Furthermore, the exact mechanisms of these individual differences in the effect of MPH treatment are unclear.

In this case report we reported two cases with opposite effects of MPH on stuttering. MPH seems to have opposite effects on stuttering in our cases. Although, the mechanisms underlying the symptoms of stuttering are unclear, these cases may point that dopaminergic system may be related with the childhood onset fluency disorder. More comprehensive studies are needed to show the neurodevelopmental and neurochemical pathways and the mechanisms associated with ADHD, stuttering and MPH treatment.

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