CASE REPORT / OLGU SUNUMU

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Is Occupational Therapy Beneficial for Attention-deficit/ Hyperactivity Disorder Accompanying Agenesis of the Corpus Callosum?

Korpus Kallosum Agenezisine Eşlik Eden Dikkat Eksikliği ve Hiperaktivite Bozukluğu Olan Bir Hastada Ergoterapi Faydalı mıdır?

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ABSTRACT

Attention-deficit/hyperactivity disorder (ADHD) is a disorder characterized by inattention, hyperactivity, and impulsivity, causing social, academic, emotional, and cognitive difficulties. ADHD symptoms start before age 12, and in some children, they're noticeable as early as 4 years of age. This case documents an ADHD case accompanying agenesis of the corpus callosum (ACC) who showed clinically significant improvement after 12 occupational therapy sessions. Sensory integration therapy, which is a branch of occupational therapy, was applied to the patient by an occupational therapist for approximately 12 sessions. After 3 months of occupational therapy sessions, there was six-month progress on the Denver Development Test 2. The ACC, an organic pathology, may lead to academic, psychiatric, and social problems. Patients can benefit from pharmacological treatment, physical therapy, speech therapy, physiotherapy as well as occupational therapy. Treatment is symptomatic according to the patient's current problem, and early intervention and supportive treatment are critical. Most children need individual rehabilitation, even if their intelligence quotient remains within the normal range.

Keywords: Agenesis of the corpus callosum, attention-deficit/hyperactivity disorder, occupational therapy

ÖZ

Dikkat eksikliği/hiperaktivite bozukluğu (DEHB), dikkat eksikliği, hiperaktivite ve dürtüsellik ile karakterize, sosyal, akademik, duygusal ve bilişsel zorluklara neden olan bir bozukluktur. DEHB belirtileri 12 yaşından önce başlar ve bazı çocuklarda 4 yaşında fark edilir. Bu olgu sunumu, on iki ergoterapi seansından sonra, klinik olarak anlamlı iyileşme gösteren korpus kallozum agenezisine (KKA) eşlik eden bir DEHB olgusunu belgelemektedir. Ergoterapinin bir dalı olan duyu bütünleme terapisi hastaya bir ergoterapist tarafından yaklaşık 12 seans uygulandı. Hastaya uygulanan 3 aylık ergoterapi seanslarından sonra, Denver Gelişim Testi 2'de altı aylık ilerleme kaydedilmiştir. KKA akademik, psikiyatrik ve sosyal sorunlara yol açabilen organik bir patolojidir. Hastalar ergoterapinin yanı sıra farmakolojik tedavi, fizik tedavi, konuşma terapisi ve fizyoterapiden de faydalanabilmektedir. Tedavi hastanın mevcut sorunlarına göre semptomatik olup erken müdahale ve destek tedavisi çok önemlidir. Çoğu çocuk, zeka kapasitesi normal aralıkta kalsa bile bireysel rehabilitasyona ihtiyaç duymaktadır.

Anahtar Kelimeler: Korpus kallozum agenezisi, dikkat eksikliği hiperaktivite bozukluğu, ergoterapi

Introduction

Attention-deficit/hyperactivity disorder (ADHD) is characterized by inattention, hyperactivity, and impulsivity, a common neuropsychiatric disorder in which symptoms usually begin around the age of four years. This disorder can cause social, academic, emotional, and cognitive turmoil. The prevalence of ADHD in school-age children varies between 5.0 and 10.0%.[1] Although the cause of ADHD has not been yet been clarified, the deterioration in the early developmental stage is caused by various genetic and environmental factors[2]. This article documents a case of ADHD accompanying corpus callosum agenesis showing clinically significant improvement after twelve occupational therapy sessions. The development of the Corpus Callosum begins with the formation of primitive lamina terminals in the sixth week of fetal life[3]. Structural formation continues between the 8th and 20th weeks of pregnancy. Meanwhile, radiation, infectious and chemical agents, chromosomal defects, maternal hormones, and nutritional disorders may cause complete or partial defects of the corpus callosum[4]. Agenesis of the corpus callosum (ACC) is a white matter anomaly characterized by the absence of a main interhemispheric commissure. The prevalence of ACC, which is the most common brain malformation, is 3-7/1000[5].

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The corpus callosum is a neuroanatomical region that connects stimuli from the cortex to the contralateral hemisphere, aiding sensory and cognitive processes. Communication and perception problems are observed in the anatomical problems of this structure. Although its characteristics in adults are well known, its mechanism(s) remain to be elucidated in the prenatal, early childhood, and childhood stages. A large body of literature examining the relationship between callosal dysgenesis/agenesis and clinical reflections has produced inconsistent findings. Most of the clinical findings in patients with ACC are mental retardation (60.0%), vision problems (33.0%), speech delay (29.0%), seizures (25.0%), abnormal muscle tone (25.0%), and nutritional problems (20.0%)[6]. If there is no developmental delay or mental problem, clinical symptoms such as mild behavioral or social problems, cognitive dysfunction, and ADHD may be seen[7]. Cognitive dysfunction includes learning difficulties, intellectual impairment, and mental retardation and ranges from mild (2/3 of cases) to moderate and severe (1/3 of cases). Behavioral or psychiatric disorders include ADHD, movement disorder, balance problems, speech disorders, learning difficulties, emotional disturbances, and autistic symptoms[8]. Despite these problems, functioning may be nearly intact.[9,10]

Case Report

A 4-year-old male patient was admitted to the outpatient clinic with complaints of sudden anger attacks, cognitive abnormalities, poor motor coordination, hyperactivity, restlessness, excessive talkativeness and difficulties in problem solving and planning. Physical examination findings were normal (no dysmorphic features, no hypotonia, ataxia, autistic behavior, seizures, or speech problems). On clinical examination, distractibility, hyperactivity, disinhibition, and inability to maintain attention were evident. In the developmental history of the patient, it was learned that milestones such as ambulation and speech were delayed. The teacher form stated that the patient was constantly on the move, had difficulty paying attention to the given tasks, and frequently quarreled with his friends. No features were identified in his family history. The Denver Developmental Screening Test 2 was applied to the patient. The Denver test revealed a 1-year delay in fine motor-adaptive and personal-social areas according to the age of the patient.

Routine blood tests and electrocardiogram were normal. Since neuro-genetic syndromes may also accompany hyperactivity, a neurology consultation was requested. While the electroencephalography was normal in neurological examination, magnetic resonance imaging showed asymmetry in the cerebral hemisphere, agenesis of the carpus callosum, and colpocephaly. The patient was started on 0.25 mg risperidone treatment for complaints of hyperactivity, restlessness, and excessive speech. During the follow-up, the dose was rearranged to 0.50 mg and the patient's complaints were controlled. After obtaining informed consent from the child's parents, occupational therapy sessions were started. Sensory integration therapy, which is a branch of occupational therapy, was applied to the patient once a week (approximately 12 sessions) for 3 months.

After the sensory integration therapy sessions, six-month progress was achieved in fine motor-adaptive and personal-social areas on the Denver Developmental Test 2. According to the renewed Denver Developmental Test 2, personal social development was determined as 3 years-6 months, gross motor development 4 years-3 months, fine motor-adaptive 3 years-6 months, and language 4 years-3 months. The final exam was better than before in perceiving instructions, cognitive functioning, and cooperating with others.

Discussion

Agenesis of the corpus callosum is one of the most common brain malformations in children with developmental delays. It is a heterogeneous condition (an isolated or congenital condition) and is more common in males than in females (2: 1)[11]. These children may present with mental retardation, vision problems, microcephaly, epilepsy, and behavioral, social, and attention-related symptoms. This case did not have epilepsy, but febrile seizures are more common in the ACC than in the normal population. Additionally, problems related to the corpus callosum may cause deterioration in areas such as cognitive, attention, language, learning skills, and impulse control mechanisms[7]. Because of these symptoms, the probability of being diagnosed with some psychiatric disease such as ADHD may increase. The impact of the structural change in the brain throughout the patient's clinic was not fully understood in ADHD, but it was clear that there was a neurodevelopmental impairment. Moreover, microstructural changes in the corpus callosum were also seen in ADHD[12]. We could not be sure whether ADHD might be related to ACC in this patient, or whether these two conditions were independent of each other. More case-control studies are needed to understand the relationship between ADHD and ACC. Therefore, neuro-radiological tools are of great importance to exclude corpus callosum agenesis, especially in cases with developmental delay. Here, we focused on occupational therapy, which has a beneficial effect on the patient's developmental delay, rather than the etiological underlying causes of this comorbidity. Additionally, we observed that sensory integration therapy improved the current clinical situation in this patient with developmental delay and hyperactivity complaints. Within the case of sensory integration therapy; the Wilbarger approach, deep pressure assessment, vestibular studies in a platform swing, proprioceptive applications in the ball pool, and selective visual stimulus therapy applications were applied to the patient[13].

Occupational therapy, which increases the participation of patients in daily activities and to support areas where they have difficulties, is gaining increasing value today. When determining the interventions to be applied to the patient, the patient's limitations and sensory and motor skills should also be taken into account[13]. Sensory integration interventions, especially applied to children with developmental delay, can be considered an early support program while focusing on increasing the

patient's participation in activity areas. Organizing an individual early support program is essential for children to reach their full potential.

Agenesis of corpus callosum is an organic pathology and may lead to academic, psychiatric, and social problems. Most children need an individual rehabilitation program, even if their IQ remains within normal limits. These patients may benefit from many treatments, including pharmacological and neurological treatment, auditory rehabilitation, physical therapy, occupational therapy, and speech therapies. Treatment is symptomatic, and early intervention and supportive treatment are critical for the child to reach the normal developmental stage

Ethics

Informed Consent: Informed consent was obtained from the parents.

Peer-review: Internally peer-reviewed.

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References

1. Polanczyk G, Jensen P. Epidemiologic considerations in attention deficit hyperactivity disorder: a review and update. Child Adolesc Psychiatr Clin N Am. 2008:17:245-260.
2. Sharp SI, McQuillin A, Gurling HM. Genetics of attention-deficit hyperactivity disorder (ADHD). Neuropharmacology. 2009;57:590-600.
3. Hatten ME. Central nervous system neuronal migration. Ann Rev Neurosci. 1999;22:511-539.
4. Pisani F, Bianchi ME, Scarano A, Viola P, Volante E, Faienza C. Clinical features in subjects with congenital anomalies of the corpus callosum. Acta Biomed Ateneo Parmense. 2000;71:497-502.
5. Glass HC, Shaw GM, Ma C, Sherr EH. Agenesis of the corpus callosum in California 1983-2003: a population-based study. Am J Med Genet A. 2008;146A:2495-2500.
6. Schilmoeller G, Schilmoeller K. Filling a void: Facilitating family support through networking for children with a rare disorder. Family Sci Rev. 2000;13:224-233.
7. Brown WS, Paul LK. Cognitive and psychosocial deficits in agenesis of the corpus callosum with normal intelligence. Cogn Neuropsychiatry. 2000;5:135-157.
8. Moutard ML, Kieffer V, Feingold J, Kieffer F, Lewin F, Adamsbaum C, Gélot A, Campistol I Plana J, van Bogaert P, André M, Ponsot G. Agenesis of corpus callosum: prenatal diagnosis and prognosis. Childs Nerv Syst. 2003;19:471-476.
9. Blum A, André M, Droulle P, Husson S, Leheup B. Prenatal diagnosis of the corpus callosum agenesis. The Nancy experience, 1982-1989. Genet Couns. 1990;38:115-126.
10. Pilu G, Sandri F, Perolo A, Pittalis MC, Grisolia G, Cocchi G, Foschini MP, Salvioli GP, Bovicelli L. Sonography of fetal agenesis of the corpus callosum: a survey of 35 cases. Ultrasound Obstet. Gynecol. 1993;3:318-329.
11. Barbara KB. Agenesis of the corpus callosum. In: Kumar P, Burton B, eds. Congenital Malformations: Evidence Based Evaluation and Management. New York: McGraw-Hill Companies, Inc.; 2007;77-82.
12. Seidman LJ, Valera EM, Makris N. Structural brain imaging of attention-deficit/hyperactivity disorder. Biol Psychiatry. 2005;57:1263-1272.
13. Schaaf RC, Dumont RL, Arbesman M, May-Benson, TA. Efficacy of occu-pational therapy using Ayres Sensory Integra-tion: A systematic review. Am J Occup Ther. 2018;72:7201190010p1-7201190010p10.