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Navigating risk factors and interventions on early-onset attention deficit hyperactivity disorder: A case report

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Case Report

ABSTRACT

Attention deficit hyperactivity disorder (ADHD) is a neurodevelopmental illness primarily affecting children. The peak onset age for ADHD is around 7.5 years with early-onset ADHD linked with worse clinical outcomes if not properly managed. We reported a 3-year-old girl presented with symptoms including selfharm, aggression toward her sibling and peers, hyperactivity, inability to focus, and distractibility. The patient had a history of post-term birth and oligohydramnios, and her symptoms were exacerbated by reduced maternal care, limited parental care, and inconsistent-lacked firmness parenting style. A multiaxial evaluation confirmed the diagnosis of ADHD, supported by Skala Penilaian Perilaku Anak Hiperaktif Indonesia (SPPAHI) score. The patient received a comprehensive non-pharmacological intervention, including applied behavior analysis (ABA) therapy with a discrete trial training (DTT) approach, family therapy to address parenting dynamics, and occupational and speech therapy to target fine motor and language delay. At 6-month follow-up, significant improvements were observed in behavior, attention, and total quality of life. This case highlights the complex interplay of genetic disorders, maternal stress, postterm birth, pregnancy-related complications, and problematic family dynamics in early-onset ADHD. Therapeutic management in early-onset ADHD must emphasize behavioral and family-focused therapies with DTT and family therapy proving effectiveness, while pharmacological treatment is considered as a last resort.

INTRODUCTION

Attention deficit hyperactivity disorder (ADHD) is a neurodevelopmental illness primarily affecting children. According to the American Physiatric Association, ADHD has an average prevalence of approximately 7.2% in children.¹ The prevalence of such condition varies across regions due to differences in age demographics and social characteristics.^{2–5} This condition is most commonly diagnosed at ages 3 to 12,⁶ peaking at around 7.5 year old.⁷ Early-onset ADHD is linked with worsened clinical onset due to its potential to interfere critical periods of brain development, disrupt the early school experiences, more likely to coexist with other neurodevelopmental disorders, and may persist into adulthood ADHD if not properly managed.⁸

Diagnosing ADHD in early childhood, especially before the age of 5 can be challenging since children in this age group often have more active and impulsive behavior, which is considered normal for their developmental stage.⁹ The diagnostic and statistical manual of mental disorder, fifth edition, text revision (DSM-V-TR) criteria continue to be the recommended diagnostic instrument despite these difficulties.6 Early diagnosis and intervention for ADHD are crucial as



Copyright @2024 Christopher Daniel Tristan, Awalil Rifqi Kurnia Rahman, Aventus Immanuel Gifta Kurniawan, Fatkan Hidayat Anggriawan, Susi Rutmalem Bangun Licensee Universitas Islam Indonesia they can prevent complications and improve behavioral outcomes, enhancing total quality of life.¹⁰ Therefore, early screening using various behavior rating scales is essential for assessing potential ADHD cases as soon as possible.⁹

To treat ADHD in children under 5 years old, a specialized method is required. Pharmacological therapy, while available, is considered risky and is typically reserved as a second-line treatment for this age group. Rather, behavior training and other non-pharmacological interventions are the recommended first line treatments for children under 5 years old with ADHD.⁹⁻¹³ This case report will contribute significantly to pediatric psychiatry in Indonesia by providing clinicians with insight into early-onset ADHD. The aim of this current case report is to highlight the risk factors and challenges in managing early-onset ADHD, focusing on the role of early intervention through a non-pharmacological approach.

CASE DESCRIPTION

A 3-year-old girl, accompanied by her mother, visited a psychiatric clinic due to focus difficulties and frequent tantrums. Her mother described persistent behavioral challenges starting at 18 months, with the patient screaming and throwing objects when her desires went unmet. By age 2, she showed signs of self-harm by pulling her hair and hitting herself when frustrated. Recent observations showed that the patient had increased irritability, particularly when she was unable to complete activities. She had aggression toward family members, including her younger siblings. Additionally, her early childhood schoolteacher reported issues with focus, emotional outbursts, and disruptive behavior toward colleagues.

From history taking, the patient originated from a nuclear family in a suburban area with a socioeconomic background characterized by modest means. The patient was born post-term through cesarean section due to indications related to being post-term and oligohydramnios. Throughout infancy (0-18 months), she experienced reduced maternal care and limited paternal care due to both parents working. During this period, the main care throughout the day was provided only by a babysitter who could not deliver appropriate discipline when the patient displayed inappropriate behaviors. During the toddler period (18-36 months), the patient's mother stopped working to care for her and her siblings. In this phase, direct interaction with the father was still rare because the father worked far from the family. During the first 36-month period, the parenting care was inconsistent and lacked firmness. The mother's response was inconsistent, alternating between permissive and restrictive action, with the father unable to establish clear boundaries due to his limited presence.

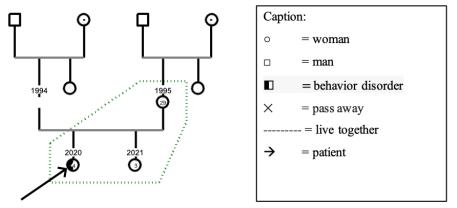


Figure 1. A family genogram highlights the patient (noted by the arrow) diagnosed with ADHD. The patient is the only individual in the family who is diagnosed with the condition. The dashed green outline indicates the current family configuration, with the father living separately and only occasionally coming home.

During the hospital examination, the patient's mother reported a significant decrease in her daughter's self-harming behavior. However, she observed that the patient still had ongoing challenges with focus, hyperactivity, and disruptive behavior toward siblings and peers at her early childhood school. A physical examination showed that she was in totally good health, with normal vital signs and neurological status. In the mental status assessment, the patient was reported to have a healthy body weight appropriate for her age but had impulsive behavior, hyperactivity, and difficulty maintaining eye contact. It was evidenced that she could follow instructions, but her concentration and attention were poor. The patient was presented with a euthymic mood and appropriate affect without any psychotic signs. While she was able to respond to questions, her speech included imperfect syllable pronunciation and persistent babbling.

Based on the absence of social communication deficit, restricted repetitive behavior or fixation on routines and further evaluation constituted an assessment of hyperactivity using the SPPAHI, with a score of 68 (reference < 30), the patient indicated a strong positive diagnosis for ADHD. The assessment of developmental milestones showed a delay in fine motor skills, equivalent to those of a 30-month-old child. Based on the results of the mental and follow-up examinations, a multiaxial diagnosis was established. On Axis I, the patient was diagnosed with ADHD combined presentation (F90.2). Meanwhile on Axis II, there were no signs of mental retardation or personality disorder due to her young age of 3 years. Axis III showed no clinical or organic disorders, and Axis IV diagnosis indicated issues related to the patient's lack of direct care from both parents during the ages of 3 to 18 months. The assessment on Axis V indicated a children's global assessment scale (CGAS) score of 70-61, suggesting some difficulties in one area, but generally functioning well.

Considering the patient's young age of 3 years, non-pharmacological therapy was chosen over pharmacological intervention due to safety concerns. The main therapy was ABA therapy with a DTT approach. When the patient exhibits appropriate behavior, she receives a reward, whereas her inappropriate behavior is disregarded. For instance, when the patient is given an instruction, like "put the ball under the table" and responds correctly, the clinician gives a food treat or a positive affirmation such as "good job". The therapy was then taught to the parents daily with weekly follow-up appointments. Following the weekly appointment, a family therapy session is conducted involving communication between the mother and on occasion, both parents, the patient, as well as the clinician as moderator. The parents were also instructed to change their parenting style, to set boundaries and consistent decisions regarding patient behavior as well as to build father-child communication at least once a day although through online sessions. In addition, the patient also undergoes occupational therapy to help with the delay in fine motor skills and speech therapy to improve the difficulties in speech. After a 6-month period, positive results were observed, including a decrease in dangerous behavior, reduced hyperactivity, and improved focus. Additionally, communication within the family improved, leading to increased attention and support for the patient.

DISCUSSION

Attention deficit hyperactivity disorder is a complex neurodevelopmental disorder characterized by abnormalities in the brain, specifically affecting the prefrontal-striatal and frontal-cerebellar circuits. While dysfunction in these circuits correlated with ADHD symptoms, not all patients had abnormalities in the same areas.^{14–16} The diagnostic and statistical manual of mental disorder, fifth edition (DSM-V) provided the most effective diagnostic criteria for such condition, showing a higher prevalence compared to diagnostic and statistical manual of mental disorder, fourth edition (DSM-IV).⁶ The DSM-IV had set the onset age at 7 years, similar to the typical start of formal schooling, while DSM-V allowed for the detection of onset as late as age 12 years.¹⁷ Therefore, early-onset ADHD before age 7 years was considered rare and associated with specific factors.

Several investigations suggested that genetic factors contributed to early-onset ADHD symptoms. The genetic factors were associated with the cumulative effects of minor gene

variations within an individual.^{18,19} However, certain rare gene mutations, such as DiGeorge syndrome (22q11.2 deletion),¹⁹ Down syndrome (trisomy of chromosome 21)²⁰ and Fragile X syndrome did not adhere to the theory of polygenic minor effects.²¹ These genetic conditions covers specific mutations that contributes to neurodevelopmental disorder such as ADHD.

Apart from genetic factors, prenatal factors were suspected to be risk factors for earlyonset ADHD. Maternal psychological stress during the prenatal period was expressed to increase the risk of such condition in children.²² Several pregnancy-related conditions that were highly associated with ADHD included preeclampsia and hypertension,²³ maternal urinary tract infections,²⁴ gestational diabetes, and hyperemesis gravidarum.²⁵ Additionally, post-term birth served as an important factor in children's cognitive developmental disorder. According to Cohort research in the Netherlands (2012), post-term birth could increase the risk of ADHD by 2.5 times, with an odds ratio of 2.10. Individuals born late post-term was at higher risk of mental disorder,²⁶ attributed to children born after 41 weeks of gestation being more vulnerable to perinatal asphyxia. This mental condition could possibly affect the supply of oxygen to the developing brain of children.²⁷

Oligohydramnios, amniotic fluid loss, and abnormal uterine bleeding during pregnancy have been hypostasized to be associated with ADHD. However, their roles as definitive risk factors remain inconclusive and require further investigations due to the limitation of vigorous evidence.²⁸ Several reviews suggested that oligohydramnios may impair proper nutrition and oxygen supply during neurodevelopment, potentially affecting brain development. Additionally, insufficient amniotic fluid may fail to adequately neutralize oxidative stress experienced by children.²⁷ In cases where patients had a history of post-term birth with oligohydramnios, such factors could potentially contribute to the risk of developing ADHD. Further investigations were thereby needed to conclusively establish these associations.

The care and attention given by both parents, in particular, was a significant factor in the family dynamics that may have a major contribution to the early emergence of ADHD in children.²⁹ For instance, when a child primarily receives reduced care from the mother with limited interaction with the father during the critical developmental period between ages 3 to 18 months, the child's development could be significantly impaired. During the preschool years, children had a high need for attention, and when a child did not receive consistent and attentive care, symptoms of hyperactivity might manifest.³⁰ This showed the importance of consistent and balanced parental participation in a child's early development, particularly during critical periods, to support healthy emotional and behavioral growth and potentially reduce the risk of ADHD symptoms. Parenting styles and family socioeconomic status were also important factors influencing ADHD in children, with those from lower socioeconomic backgrounds facing higher risks.³¹ Authoritarian parenting, characterized by constant demands and criticism, could contribute to ADHD symptoms. In contrast, the lack firmness in parenting also potentially contribute to hyperactivity manifestation. Family disharmony further worsens the risk, as children request balanced care from both parents to thrive.³²⁻³⁴

Treating early-onset ADHD requires special care, with a focus on non-pharmacological therapy as recommended by guidelines.^{35,36} The National Institute for Health and Care Excellence (NICE) reccomends group-based parent programs to be the primary treatment approachfor children in this age group.³⁷ While detailed guidelines in Indonesia are not publicly available, similar recommendations by guidelines in several Asian countries such as Malaysia, Singapore, and India, prioritize non-pharmacological therapies before considering pharmacological interventions. In particular, Malaysia's guidelines recommends that pharmacological therapy be administered with a minimum six year old age with significant impairment in at least one domain after being given behavioral and environmental therapies.³⁸

The group-parent programs and other family therapy are effective treatments for children with behavioral disorders,^{39,40} suggesting intensive communication and mutual understanding within the family. These programs helped parents recognize their children's special needs and provide necessary attention, while children learned to understand their parents' work obligations. However, for early-onset ADHD before age 5 years, family therapy needs to

predominantly require the participation of parents.^{41,42} In this case, the child was 3 years old and did not understand that parents needed to work. Therefore, it is necessary for parents to take the lead in providing attention and understanding their children's needs. This is evidenced by the findings of an improvement in the child's behavior after the mother resign from her job to focus on caring for her.

Behavioral therapy has been well-known and widely used to treat ADHD. The main goal of behavioral therapy is to understand and modify behavior to reduce negative actions and promote positive ones.⁴³ In this case, problematic behaviors such as self-harm, aggression toward siblings, and aggression toward colleagues, were required to be eliminated. A structured approach like ABA with DTT approach was employed, which is originally effective in managing autism spectrum disorder but has shown significant efficacy in ADHD.^{38,39} The DTT involves a structured repetitive teaching method to improve adaptive behavior. This mechanism by DTT is effective in managing ADHD symptoms, particularly in improving attention and reducing hyperactivity as demonstrated by our case.^{11,12}

Crucially, behavior therapy is used in combination with family therapy. Family therapy was integrated to address inadequate parenting styles that had failed to understand the child's behavior.⁴² This combination of DTT and family therapy provided a dual benefit: modifying the child's behavior while equipping the parents with strategies to better respond to the child. By consistency with long-term follow-up, DTT and family therapy could effectively manage early-onset ADHD. In addition, occupational therapy and speech therapy have also proven effective in enhancing fine motor skills and speech, which is particularly beneficial for children with ADHD who may experience delay in this area.^{44,45}

According to NICE guidelines, the recommended primary pharmacological therapy for ADHD covers psychostimulants such as methylphenidates and amphetamines.³⁷ Furthermore, in several Southeast Asian countries, atomoxetine is another psychostimulant that can be used for ADHD.³⁸ Psychostimulants worked by increasing dopamine and norepinephrine levels, prefrontal cortex activity, executive function, and attention in ADHD patients.⁴⁶ Although psychostimulants have demonstrated effectiveness in improving ADHD symptoms,^{47,48} their use in preschool-age children, comprising those under 5 years old, should be strictly limited due to two primary reasons. The first is the potential long-term risks, including substance abuse in adulthood.⁴⁹ Second, drugs-therapy that is especially related to dopamine and norepinephrine in child-ADHD had been observed followed by significant side effects, such as sleep disturbances, cardiovascular effects, and growth retardation.⁵⁰ Therefore, when an early-onset ADHD responded well to non-pharmacological therapy, initiating pharmacological treatment was not recommended according to NICE guidelines.

CONCLUSION

Early-onset ADHD was a multifaceted neurodevelopmental disorder influenced by genetic factors, prenatal history (maternal stress and preterm/post-term birth), and family dynamics (parenting style and socioeconomic status). The non-pharmacological therapy, particularly ABA with DTT approach combined with family therapy is effectively managed ADHD and is preferred for children under 5 years old, with pharmacological options such as psychostimulants reserved as the last option for cases where non-pharmacological intervention is inadequate. Additional occupational and speech therapy are also needed to regain the normal function of an ADHD child. Consistency, long-term, and comprehensive management by the parents is essential to optimize cognitive function and mitigate future ADHD symptoms.

CONFLICT OF INTEREST

There is no conflict of interest declared by all authors. Written consent for publication had been obtained from the patient's parents.

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AUTHOR CONTRIBUTION

CDT made a major contribution in writing the manuscript. CDT is a provider of concepts for these ideas. ARKR and FHA contributed to the form of a literature search. AIGK contributed by writing the case description. SRB contributed to provide treatment, to review the manuscript, and to write ideas. All authors read and approved the final manuscript.

LIST OF ABBREVIATIONS

ABA: applied behavior analysis; ADHD: Attention deficit hyperactivity disorder; CGAS: children's global assessment scale; DSM-IV: Diagnostic and Statistical Manual of Mental Disorder, Fourth Edition; DSM-V: Diagnostic and Statistical Manual of Mental Disorder, Fifth Edition; DSM-V-TR: Diagnostic and Statistical Manual of Mental Disorder, Fifth Edition; DTT: Discrete trial training; NICE: The National Institute for Health and Care Excellence; SPPAHI: Skala Penilaian Perilaku Anak Hiperaktif Indonesia

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