



CASE REPORT/CASO CLÍNICO

Very Early Onset of Schizophrenia Spectrum Disorders: A Case Report of a Challenging Diagnosis and Treatment

Início Muito Precoce de Perturbações do Espectro da Esquizofrenia: Um Relato de Caso de um Diagnóstico e Tratamento Desafiadores

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ABSTRACT

Disorders within the schizophrenia spectrum are severe illnesses characterized by disturbances in thinking, perception and behavior that are associated with deterioration in emotional, cognitive, and social functioning. Symptoms typically emerge in early adulthood, but in extremely rare and severe cases they can begin in childhood.

The primary objective of this paper is to provide a concise overview of the scientific evidence regarding very early-onset schizophrenia based on a clinical case description. Data were sourced from clinical records and meticulously scrutinized within electronic databases, including PubMed and Embase.

We present a case study involving a 16-year-old male adolescent with psychotic symptoms, previously diagnosed with obsessive-compulsive disorder and autism spectrum disorder. This case epitomizes the challenges associated with the diagnosis of an uncommon and markedly severe manifestation of one of the most debilitating psychiatric conditions, thereby shedding light on the intricate challenges inherent in managing a treatment-resistant disease.

RESUMO

As perturbações do espectro da esquizofrenia são doenças graves caracterizadas por alterações do pensamento, percepção e comportamento, associadas à deterioração do funcionamento emocional, cognitivo e social. Os sintomas surgem geralmente o início da idade adulta, mas em casos extremamente raros e graves podem ter início na infância. O principal objetivo deste artigo é rever a principal evidência científica disponível relativa à esquizofrenia de início muito precoce, com base na descrição de um caso clínico. Os dados foram obtidos a partir de registos clínicos e através da pesquisa criteriosa nas bases de dados eletrónicas PubMed e Embase.

Apresentamos o caso de um adolescente de 16 anos com sintomas psicóticos previamente diagnosticado com perturbação obsessivo-compulsiva e perturbação do espectro do autismo. Este caso demonstra os desafios associados ao diagnóstico de uma manifestação incomum e particularmente grave de uma das perturbações psiquiátricas mais debilitantes, salientando as complexas dificuldades inerentes ao tratamento de uma doença resistente ao tratamento em idade pediátrica.

Keywords: Adolescent; Child; Psychotic Disorders/diagnosis; Schizophrenia/diagnosis

Palavras-chave: Adolescente; Criança; Esquizofrenia/diagnóstico; Perturbações Psicóticas/diagnóstico

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INTRODUCTION

Schizophrenia spectrum disorders (SSD) are severe illnesses characterized by disturbances in thinking, perception, and behavior, associated with deterioration in emotional, cognitive, and social functioning, leading to long-term disability with a significant impact on the everyday life of the individual and their family.^{1,2} These conditions typically manifest in early adulthood, but in the case of early-onset schizophrenia, they manifest before the age of 18 – early-onset SSD (EOSSD). Very early-onset SD (VEOSSD) begins in childhood before the age of 13 representing a more severe and extremely rare presentation, with a prevalence of less than 1 in 40 000.¹⁻³ The majority of children exhibiting psychotic symptoms do not go on to develop a SSD. Psychotic symptoms can occur in other psychiatric and non-psychiatric conditions, in the presence of head trauma, and with use of certain medications and psychoactive substances, particularly in children.² Simultaneously, SSD are often marked by the presence of a prodromal phase, which may encompass not only attenuated psychotic phenomena, but also anxiety, obsessive symptoms, motor and language impairments, and socialization difficulties; approximately 70% of the children with VEOSSD exhibit a pre-morbid phenotype featuring this type of symptoms.^{2,3} Certain psychiatric conditions even appear to increase the risk of developing SSD, such as obsessive-compulsive disorder (OCD) and autism spectrum disorder (ASD).^{4,5} Considering this complexity, the diagnosis of VEOSSD requires a longitudinal assessment for differential diagnosis.² Although rare, EOSSD and VEOSSD are associated with a poorer prognosis when compared to adult-onset SSD. The occurrence and length of psychotic episodes adversely impact neuropsychological, neurophysiological, and neurostructural health.¹ In the recovery phase, typically occurring in adulthood, patients often remain impaired. Adults who experienced symptoms that began in childhood tend to have more significant social deficits, higher unemployment rates and a lower likelihood of living independently.² Early and expert treatment is crucial for optimizing outcomes. Treatment with antipsychotic medications is effective when combined with psychotherapeutic, psychosocial, and psychoeducational interventions, although some patients may not respond or respond only partially to treatment.¹⁻³

CASE REPORT

We present a case study of a 16-year-old male adolescent, previously diagnosed with OCD and ASD, who presented to the emergency department (ER) with thought and behavioral disturbances that had worsened over the previous six months. The condition initially manifested as persecutory delusional ideas related to school, coupled with compulsive checking behaviors. Over time, it progressed to feelings of guilt and self-reference ideas related to the COVID-19 pandemic – the boy claimed responsibility for causing the pandemic and had, in this context, turned himself into the police station twice in the last month. Two weeks before coming to the ER, he developed the belief that he had cured the pandemic, along with other

megalomaniac and mystical ideas, such as the belief that he was God and Jesus's brother. His parents also observed him in the past week engaging in soliloquies, experiencing unmotivated laughter, displaying disorganized speech, and exhibiting some bizarre behaviors like strange movements of his arms and hands; he was also insisting that his mother was pregnant, despite of his family's denials. Three days before initial observation he had experienced almost total insomnia. He had an initial diagnosis of OCD and was treated with increasing doses of aripiprazole, sertraline, and risperidone but did not show any symptomatic improvement. The boy had no personal history of medical or surgical conditions. Substance use, head trauma, other physical symptoms or recent episodes of any infectious disease were denied. There was a family history of recurrent depressive disorder in the father and the grandfather and reports of disorganized behavior in an uncle and aunt on the father's side of the family. In clinical observation, the patient was disoriented in time and space and presented with psychomotor retardation, circumstantial and tangential speech, neologisms, concretism, thought blocking, fragmentation and derailment, along with auditory-verbal hallucinations, thought broadcasting, low affect reactivity, and insomnia. There were clear alterations in thought content, including mystical, grandiose, and Capgras delusions. He had little insight into his condition. Physical and neurological examinations in the ER showed no changes. An analytical routine screening was carried out (blood count, renal function, ionogram, liver profile and drug testing), as well as an electrocardiogram and a computed tomography (CT) scan of the brain, all without alterations. The patient and his family accepted the treatment plan and the boy was admitted to the inpatient Child and Adolescent Psychiatry unit, for clinical stabilization and extended etiological study.

A further detailed assessment of the patient's personal history was conducted during hospitalization. A slight delay in language development was identified, without any other abnormalities in early development. Parents denied any specific behaviors or interests during childhood. He played with other children, although he was generally shy. At the age of 5, the kindergarten teacher noted some of the boy's behaviors as unusual and recommended specialized observation. Despite a psychological evaluation indicating only anxiety symptoms, he received a diagnosis of autism spectrum disorder (ASD) from his pediatric neurologist. The father believed that the boy has always had difficulty reading social cues, despite maintaining interest and ease in interactions. A more thorough evaluation of the patient's symptoms revealed an insidious development of thought and behavioral alterations with episodic progression over the past 5 years, accompanied by decreased functionality and academic performance. At age 11, the patient reported hearing neighbors talking about him at home, exhibited hypervigilance on the streets, and believed motorcycles were following him; after this period, there was a marked decline in academic performance. At that time, he began consultations with a child psychiatrist, who prescribed aripiprazole, leading to complete remission of symptoms.

One year later, at age 12, he experienced a decrease in food intake lasting about a week, accompanied by total insomnia, behavioral disorganization, and incoherent speech for one day. This episode rapidly remitted with the prescription of risperidone. At age 14, a new period of symptomatic worsening occurred, characterized by insomnia and excessive concerns about family security, with several compulsive checking behaviors, such as repeatedly calling his grandparents during the day and inspecting the family's car at night. Symptomatic stabilization was achieved through therapeutic adjustments with risperidone and titrated sertraline and doses were progressively reduced. Six months before hospitalization, when the current symptoms began, a new titration of risperidone was performed, with no symptomatic relief.

During hospitalization, an initial comprehensive complementary assessment was carried out. The patient was again evaluated by neurology team, with neurological examination showing no alterations. An electroencephalogram (EEG), a brain magnetic resonance imaging (MRI) and multiple analytical evaluations were performed, including hemogram, renal and thyroid function, ionogram, liver profile, viral serologies, VDRL test, folate, vitamin B12 and vitamin D dosages, ceruloplasmin, Antinuclear Antibodies (ANA), all without alterations. A psychological assessment was also carried out, which confirmed the presence of psychotic symptoms severely impacting cognitive and executive brain functions.

During hospitalization, after the outpatient administration of risperidone at adequate doses, pharmacological treatment with olanzapine was first tried, without any improvement for several weeks. Following the initiation of clozapine, there was complete remission of sensory-perceptual disturbances, increased affective reactivity, improved thought

and speech organization, and a reduction in psychomotor slowing. The patient also showed significantly increased willingness to engage in other therapeutic and psychosocial interventions. Complete remission of delusional activity was not achieved during hospitalization, although it presented with reduced dynamism and less impact on daily functioning.

DISCUSSION

This case epitomizes the challenges associated with diagnosing an uncommon and markedly severe manifestation of a debilitating condition, thereby shedding light on the intricate challenges inherent in managing a treatment-resistant disease at such an early age. While sharing neurobiological and phenomenological features with adult-onset schizophrenia, VEOSSD manifests as a more severe variant of the disorder.¹ Diagnosis should be approached with caution in childhood and adolescence, but early recognition is essential as delayed treatment is associated with a poorer prognosis.^{1,6} Conducting a thorough history and physical examination during the patient assessment is essential. Prompt treatment is crucial for alleviating both positive and negative symptoms and for reducing deficits in social, cognitive, and motor functioning.² Clozapine is an atypical antipsychotic that is extremely effective in treating schizophrenia. It should be used if there is a poor response to two other antipsychotic agents, each administered at adequate doses for at least four weeks. Epidemiological studies indicate that its administration often happens much later than what clinical guidelines suggest.^{1,2} Efforts should be directed towards identifying risk factors for the development of these disorders in childhood and adolescence, as well as addressing treatment-refractory cases.

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NAB e MM: Desenho do trabalho, interpretação dos dados, redação e revisão final.
Os autores aprovaram a versão final a ser publicada.

CONTRIBUTORSHIP STATEMENT

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