

Case Report

Very Early-onset Schizophrenia with Secondary Onset Tic Disorder

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ABSTRACT

Very early-onset schizophrenia (defined as an onset of psychosis before 13 years of age) is a rare and severe form of the disorder which is clinically and neurobiologically continuous with the adult-onset disorder. It is rarely reported <12 years of age in Indian literature. Here, we present a 15-year-old boy who developed psychosis at 9 years of age and during illness developed tic disorder.

Key words: *Haloperidol, tic disorder, Tourette syndrome, very early-onset schizophrenia*

INTRODUCTION

Early-onset schizophrenia (EOS) is a term used to indicate the onset of psychotic symptoms before the age of 18 years. It is also frequently unrecognized, notably because its clinical aspect varies with age. Onset of the disorder rarely occurs before the age of 13 years. If the onset is earlier than 13 years, it is termed Very Early Onset Schizophrenia (VEOS).^[1,2]

Genetic factors are believed to play an important role in the pathogenesis of schizophrenia. An increased family history of schizophrenia and schizophrenia spectrum disorders (e.g., schizotypal or paranoid personality disorders) has been found in patients with VEOS.^[2,3] Communication deficits are also often found in the families of children with VEOS.^[3] The great majority of patients with VEOS have significant premorbid abnormalities in language, motor, and social

development.^[3] Hallucinations, thought disorder, disorganized behavior, and flattened affect, all have been consistently found in EOS while systematic delusions and catatonic symptoms may be less frequent.^[2,3] Developmental differences in language and cognition may affect the range and quality of symptom presentation.^[1] Symptom presentation in EOS or VEOS generally has prodromal, acute, recovery, and residual phases. The diagnosis of EOS or VEOS in children and adolescents is made using the same Diagnostic and Statistical Manual of Mental Disorder (DSM-5) criteria as in adults.^[4] However, to make the diagnosis of VEOS, a comprehensive psychiatric and physical assessment is needed. General medical causes of psychotic symptoms should also be ruled out, and diagnosis should be confirmed in a longitudinal follow-up.

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VERY EARLY ONSET SCHIZOPHRENIA (VEOS) WITH SECONDARY TIC DISORDER

A 15-year-old male child presented with his mother with complaints of odd behavior, senseless laughing, odd eccentric speech, unprovoked aggressive, abusive assaultive behavior toward family members and strangers, wandering away from home without informing family members, not eating or dressing himself properly, loss of interest in playing, schooling, and all age appropriate activities, and social withdrawal for approximately last 6 years.

The first symptoms were to be recognized at 9 years of age when he was studying in 4th standard with good social interactions with family and peers at the time of onset of his symptoms. Although regular in school, the patient was termed as “Slow learner” by his teachers and was academically poor compared to same aged 4th standard children.

During this time, he started remaining aloof along with features such as smiling and muttering to self, purposeless starting in any particular direction, suspiciousness toward strangers on the street and accusing them of teasing him with resultant irritability, throwing objects, hitting walls, or injuring himself. These symptoms were observed several times throughout the day without any provocation or fluctuation in intensity.

Over last 4 years, he had trouble relating to and interacting with family members, being quiet, and withdrawn at other times. Any attempt to engage him in household work, study, or play was met with irritability, abusive assaultive behavior. He also stopped going to school since as reported by teachers he would often run away from school without informing teachers or would become irritable and aggressive toward other kids and teachers. The onset was gradual with all aforementioned symptoms developing over few months with no apparent preceding medical illness, head injury, stressors, etc., The course of the illness was fluctuating with the patient achieving partial remission with olanzapine 15 mg/day, Sodium Valproate 400mg/day in divided doses, but never reaching baseline functioning in the illness course.

He was admitted to Government Medical College, Akola, Psychiatry ward in February 2016 when there was an exacerbation of assaultive, abusive behavior despite taking regular outpatient department treatment. In addition, it was noticed that patient had developed repetitive, stereotyped grunting, throat clearing, tongue protrusions, and lip smacking appearing at regular intervals. This feature was not present in earlier follow-ups. On requests, the patient was able to voluntarily suppress these for few seconds. These

vocalizations and acts would disappear with distraction and during sleep. On interview and questioning, the mother reported that these acts and vocalizations had developed over preceding 1 month, and the patient would continue doing these repetitions despite admonishment by parents.

During interview, the patient remained aloof with remarkably flattened affect, inappropriate giggling, and poor eye contact. He could not give any explanation about these acts and vocalizations. There was no history of preceding fever or throat infection.

The early developmental history of the patient revealed full-term normal vaginal delivery with no history of prenatal, natal, or early postnatal complications. Mother reported the occurrence of febrile seizures at 1 year age during a bout of high-grade fever for which he received some treatment thereafter (record not available). There is no history of seizures or seizure-like activities reported thereafter.

He had delayed motor developmental milestones with walking at 2 years of age and language milestone with first meaningful words spoken at 2 years and short-sentenced meaningful speech occurring around 4 years age. Mother reported age-appropriate social interactions with the patient actively engaging with toys, pretend play with all family members as per age. There is no history suggestive of autism or pervasive developmental disorder (PDD) considering attainment of aforementioned milestones. There was no history suggestive of sexual or physical trauma.

There was no family history suggestive of any psychiatric illness.

After admission, provisional diagnosis of VEOS with Tourette syndrome (TS) as per DSM-5 diagnostic criteria was kept.^[4] Positive and negative syndrome scale (PANSS) rating scale^[5] was used for symptom monitoring. His PANSS ratings at admission were as follows: Positive symptom score: 36, negative symptom score: 32, and general psychopathology score: 67.

A full medical work-up consisting of complete and differential blood count, serum electrolytes, kidney function tests, electrocardiogram was ordered and was found to be within normal limits. In addition, noncontrast computed tomography brain (NCCT brain) was ordered in lieu of history of febrile seizures at 1 year age. NCCT brain also revealed no abnormality.

Patient's dose of olanzapine was increased to 20 mg/day along with lorazepam 2 mg BD for controlling aggressive, assaultive abusive behavior. In addition, for repetitive,

stereotyped grunting, throat clearing, and related vocalizations along with repetitive tongue protrusions and lip smacking, he was started on tablet haloperidol 5 mg HS + tablet trihexyphenidyl 4 mg/day divided dose.

Over 2 weeks of ward stay, the patient showed significant improvement in aggressive, assaultive abusive behavior, and became cooperative and amenable to suggestions and orders. His clinical improvement was also evidenced by decreased PANSS rating^[5] score over 2 weeks compared to admission. His repetitive, stereotyped grunting, vocalizations, and tongue protrusions were reduced in intensity over 2 weeks of ward stay.

Patient's mother also reported more than 50% improvement in symptom severity compared to the time at admission. However, deficits in language, reciprocal interactions, play activities, cognitive functioning along with flattened affect continued at some level. His PANSS rating at the time of discharge was as follows. Positive symptom score: 19, negative symptom score: 20, and general psychopathology score: 36.

DISCUSSION

We would like to highlight some important points in comparison with existing literature in this case. This is a rare case with an onset of VEOS before 12 years of age. Since the average age of onset in Indian studies in childhood-onset schizophrenia is reported as 12.25 years.^[6] Any case of VEOS should be carefully questioned, and diagnosis should be confirmed in a longitudinal follow-up. It is evident that the first issue with this case is the diagnosis of schizophrenia with TS. Differential diagnosis of VEOS should include organic disorders (such as delirium, seizure disorders, central nervous system lesions, metabolic disorders, substance abuse, and toxic encephalopathies), mood disorders with psychotic features, PDD, sexual or physical traumas, nonpsychotic behavioral, or emotional disorders (including dissociative disorder).^[7] We excluded any possible organic disorders with appropriate investigations. There was no history of substance abuse or intoxication.

A relatively early deterioration of social and language skills may prompt one to think about childhood disintegrative disorder (CDD). However, there was no loss of motor skills or bladder and bowel control in favor of CDD. The presence of overt hallucinations and delusional thinking are not expected in CDD.^[4] In addition, the onset and course of the symptoms (including hallucinations and possible delusional thinking), partial improvement with olanzapine treatment suggest VEOS rather than

CDD. The presence of delay in premorbid motor and language development along with subtle academic difficulties may also suggest borderline intelligence and a possibility of autism spectrum disorder (ASD) as described in DSM 5.^[4] However, his premorbid language and social skills were sufficient to communicate easily. Furthermore, there had been no stereotypic behavior typical of ASD before onset of symptoms. Furthermore, repetitive stereotyped grunting, throat clearing vocalizations, and acts occurred later in the course of his illness (we interpreted these new onset vocalizations and acts to be tics and independent of psychotic symptoms).

Gilles de la Tourette's syndrome and related tic disorders have been reported with comorbid psychological disorders such as obsessive-compulsive disorder and attention-deficit hyperactivity disorder.^[8,9] Dopaminergic hyperactivity in tic disorders has been suggested since dopaminergic (D2) receptor blockers such as haloperidol are effective in suppressing tics^[10] At the same time, dopaminergic hyperactivity in schizophrenia is well documented.

Despite this shared neurobiology, onset of tics during symptoms of schizophrenia has been reported to be quite rare.^[11] Thus, in our understanding, this case is a rare presentation due to Very early onset schizophrenia (VEOS) and occurrence of tics during illness without any preceding change in drug regimen, infection, or development of other comorbidities. On extensive literature search, we found a case report of late onset tics in adult patient of schizophrenia after 12 years of good symptom control with haloperidol followed by abrupt discontinuation of antipsychotic. The patient showed a good response to olanzapine monotherapy.^[12] Although atypical antipsychotics are known to carry lesser risk of extrapyramidal side effects, TD, and tics; in our patient, tics developed despite being on olanzapine and showed reduced intensity on addition of haloperidol.

CONCLUSION

VEOS can present at a much earlier age in the Indian population.^[6] The development of TS along with symptom exacerbation despite regular treatment with atypical antipsychotic olanzapine and a significant reduction in intensity of tics after addition of haloperidol are other points of intrigue in this case. Thus, the treatment of such cases should be tailored to meet the needs of the patient and aim at maximum symptom reduction. Further, systematic studies of these patients may help better understanding the etiologic processes and the varied clinical presentations involved in schizophrenia.

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Conflicts of interest

There are no conflicts of interest.

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