17 Years of Treatment-Resistant Mutism in Non-Catatonic, Childhood-Onset Schizophrenia: A Rare Case Report

To the Editor: Mutism is one of the commonest psychopathological symptom manifestations seen especially in catatonic schizophrenia and may appear in several different clinical settings. Important organic causes of mutism include head injury; posterior fossa surgery; encephalitis; frontal lobe lesions; the post-ictal phase of epilepsy; laryngeal tumors; and endocrine disorders, including hyperparathyroidism, myxoedema, diabetic ketoacidosis, and Addison's disease. Medications capable of inducing mutism include tacrolimus and cyclosporine.[1,2] There are reports of mutism lasting for years in catatonic schizophrenia. The largest report on mutism associated with non-catatonic schizophrenia is from the Kosraean population, where it is described as a specific cultural variant of psychopathology. Mutism has lasted from a few days to 20 years in this community and is noted in the initial part of the illness, predicting relapse.[3] To our knowledge, only two cases of longstanding mutism associated with paranoid schizophrenia had been reported in the literature, but not in the pediatric age-group as yet.[4,5] We report the first case of longstanding, treatment-resistant mutism in childhood-onset, non-catatonic schizophrenia, which was repeatedly suspected to be of neurological nature and lasted for about 17 years from its onset without remission, observed and treated in a rural, tertiary-care psychiatric hospital in Central India.

Case Report

"MR. AS," 28-year-old, right-handed, single, unemployed man, presented to us in November 2008 with a 17-year history of childhood-onset schizophrenia characterized by delusions; auditory hallucinations, commenting type; aggressiveness; social withdrawal; insomnia; poor self-care; and marked socio-occupational dysfunction. About 1 year after the initial symptoms, at the age of 12, he had developed mutism. His speech had become restricted to terse responses and monosyllables, and he would indicate his needs by gestures, overtures, and writing. Upon persuasion from parents, he had written a note implying that, from now on, he would not like to speak with anybody, and it was on his own accord that he had decided to remain mute. Apathy, asociality, anhedonia, and affective flattening had evolved over the last few years and persisted. There was no history of head injury, seizures, substance use or abuse, dementia, confusion, or a mood disorder. Findings of his baseline investigations, including electroencephalography, were within normal limits. His IQ was 110. The only abnormal finding in his investigation was on his mri brain scan, which showed mild enlargement of the frontal horn of left and right lateral ventricles. No other structural abnormality was evident, even with contrast imaging. He had received adequate trials of haloperidol, chlorpromazine, risperidone, and olanzapine without any significant improvement in the initial 6 years. He subsequently had received a course of 20 modified ect treatments; thereafter, his hearing of voices, aggressiveness, and personal care were improved, but his mutism continued. We started him on clozapine 450 mg/day (gradually built-up with monitoring of his blood levels). In November 2008, his PANSS score was Positive: 34, Negative: 13, and General Psychopathology: 38. After several months on clozapine, his mutism continued in the same manner, despite augmentation with risperidone up to 6 mg per day. Later, levosulpiride 50 mg per day was added to the clozapine 450 mg per day, and about 4 months after the augmentation, he began showing improvement; his eye contact improved; he was less defensive, hugged his father, was willing to communicate passively with everyone around him after almost 17 years, and has not show any further episodes of mutism since then. His current PANSS score is Positive: 7; Negative: 9; and General Psychopathology: 21, and he is still pursuing the cognitive retraining and assertiveness training sessions monthly, along with regular compliance with medications from our psychiatric services.

Discussion

Mutism is an important sign as well as symptom in psychiatric disorders with an extensive differential diagnosis. Chronic, longstanding mutism with neurological disorders is known especially in Alzheimer's disease, stroke, fronto-temporal dementia, and Creutzfeldt-Jakob's disease.[6] The critical brain areas involved are the frontal lobe (cingulate gyrus, supplementary motor area, and the dorsolateral border zones), basal ganglia (caudate and putamen), and the mesencephalus and thalamus.[7,8] Important clues pointing to neuro-

logical cause include irregular respiration, abnormal pupil responses, roving eye movements, facial weakness, and exaggerated jaw jerk. On the contrary, patients with primary psychiatric disorders may be induced to whisper or communicate in writing, although the latter may also occur with infarction, leading to pure word-dumbness (an apraxia restricted to the movements required for speech). In the index patient, it is difficult to explain the psychopathological presentation of mutism with the other accompanying positive and negative symptoms, but the possibility of its occurrence, either as an extreme form of alogia or secondary to delusional beliefs, is very likely. Interestingly, all three cases of long-lasting mutism, including ours, had been proved to be treatment-resistant cases, not responding to clozapine alone. Maybe combining clozapine with low-dose levosulpiride, both proven to be effective and efficacious in alleviating negative symptoms in clinical trials, could make an interesting combination against such psychopathological presentations.

Finally, the field of Child Psychiatry has undergone significant transformation with the advent of newer medications, along with growing recognition that children suffer from major mental illness and could benefit from psychotropic medications for these conditions. With limits, precision, predictability, and nurturance, children could be healed from their psychic wound.

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