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A CASE OF CHILDHOOD SHIGELLOSIS WITH MUTISM*

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SUMMARY: Selimoğlu M, Akdağ R, Kırpınar İ. (Department of Pediatrics, Atatürk University Faculty of Medicine, Erzurum, Turkey). A case of childhood shigellosis with mutism. Turk J Pediatr 1995; 37: 431-434.

Bacillary dysentery, an acute infection caused by various strains of Shigella, is characterized by abdominal pain, tenesmus, and diarrhea with mucus, pus and blood. Neurologic manifestations including meningismus, delirium and convulsions may accompany the infection. We describe a thirteen-year-old girl who presented with headache, convulsion and loss of consciousness at the onset and developed diarrhea with blood and pus after hospitalization. The diagnosis of shigellosis was based on clinical data and isolation of the microorganism in the stool specimen. After improved physical functions, the patient developed mutism that continued for two days in the course of her illness, despite having no history of neurologic or psychological problems. She was diagnosed by a psychiatrist with organic mental syndrome NOS (Not Otherwise Specified) according to DSM-III-R criteria. None of the conditions that may cause mutism could be confirmed. This is the first reported case of mutism accompanying shigellosis.
Key words: childhood shigellosis, mutism.

Bacillary dysentery is an acute infection caused by various strains of Shigella. The classical clinical picture is one characterized by severe abdominal pain, tenesmus and constitutional symptoms with frequent stools containing mucus, pus and blood[1]. Neurologic manifestations including meningismus, delirium and convulsions may accompany Shigella dysentery. Convulsions are the most frequent and prominent symptom among them[1,2].

Some rare neurologic symptoms such as hallucinations have been reported in childhood shigellosis. In this report we describe a child with shigellosis who had convulsions, postictal coma and organic mental syndrome with mutism, the inability or unwillingness to speak resulting in absence or marked paucity of verbal output, in the course of her illness[3,4].

Case Report

A 13-year-old girl was admitted to the hospital because of severe headache, convulsions and loss of consciousness. The child had no history of neurologic or psychological problems. On the day of admission, she had had a febrile,

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generalized convulsion of five minutes' duration and lost consciousness. She had been taken to the hospital within a few hours after her seizure.

Physical examination revealed a critically ill girl in a coma. Her body temperature was 36.5 °C, pulse rate 100 beats/min and blood pressure 110/60 mm Hg. She had no signs of meningeal irritation except nuchal rigidity. No pathological reflex was detected. The EEG revealed a normal pattern.

The white blood cell count was 19.7 x 10^9/L (24% neutrophils, 2% band forms, 2% monocytes, 72% lymphocytes), and hemoglobin level 15.5 g/dl. Serum electrolyte, glucose, calcium and liver function values were within normal limits. Serum creatinine was 0.8 mg/dl and blood urea nitrogen was 22 mg/dl. Urine analysis was normal. Lumbar puncture revealed a nonpathologic screen with no organism on Gram stain and a negative culture.

Approximately 12 hours later she began to respond to mechanical stimulation. During this period she developed diarrhea containing blood and mucus. Polymorphonuclear leukocytes were detected in stool, and the culture specimen showed Shigella flexneri; for this reason ampicillin administration was initiated. Approximately 24 hours later she regained consciousness and was fed by her mother within four hours. Approximately 12 hours later the watery diarrhea disappeared. Despite improved physical functions, she demonstrated mutism during the waking state. She was not confused or disoriented and she was indicating what she wanted with her hands. For two days she had no hallucinations, agitation, anxiety or mood disturbances. She ate her meals, slept well and obeyed other suggestions. However, she did not speak, and she refused to write and read. General physical and neurological examination, EEG and other laboratory assessments including parathyroid hormone (PTH), thyroid stimulating hormone (TSH), thyroxine (T_4), serum glucose, calcium, liver function tests and blood pH were repeated to rule out the conditions which might cause mutism, and none revealed an abnormality. After two days she began to speak, and despite of all our efforts she did not tell us why she had refused to speak.

A psychiatric evaluation was performed by an experienced psychiatrist. The patient was diagnosed with organic mental syndrome NOS (Not Otherwise Specified) according to DSM-III-R criteria[5]. No psychotropic medication was started. On the sixth day of her hospitalization, she was discharged with complete recovery.

Discussion

Neurologic symptoms are among the most frequent extra-intestinal manifestations of shigellosis. Convulsions have been described in 12-45 percent of hospitalized patients with culture-proven shigellosis[2]. Other neurologic symptoms, including lethargy, confusion and severe headache, are usually considered manifestations

of encephalopathy and occur in approximately 49 percent of patients with shigellosis[6]. Both seizures and transient encephalopathy are considered benign and rarely recur or have permanent sequelae, although fatal cases have been reported[3, 7].

Our case exhibited mutism accompanying shigellosis. The literature reveals that a myriad of psychiatric and non-psychiatric illnesses can present with mutism, and that patients with non-psychiatric disorders who are mute are frequently misdiagnosed. Of the cases reported in the literature, only three gave sufficient information to assess the prevalence of various disease states that may present with mutism. According to these reports, 39 percent of patients presenting with mutism are likely to have an affective disorder, 29 percent to have some form of schizophrenia, 17 percent to have organic brain syndrome, nine percent to have character disorders, and six percent to have an uncertain diagnosis[4].

Mutism in childhood is usually of the non-catatonic type. Mutism without catatonia may also be a symptom of conversion hysteria. The features considered helpful in making this diagnosis include an abrupt onset, a recent stressful event, an indifferent attitude (la bella indifference) and the absence of identifiable organic disease[4]. In our case the child had culture-proven shigellosis, and the syndrome did not meet the criteria for any other organic mental syndromes. She also had no history of a psychiatric problem.

The known toxic and metabolic conditions that cause mutism are hypoparathyroidism, Addison's disease, myxedema, diabetic ketoacidosis, hypercalcemia, acute intermittent prophyria, hepatic encephalopathy, glutethamide withdrawal and organic fluoride toxicity[4]. Our clinical and laboratory assessment confirmed none of these conditions. Petit mal status epilepticus closely resembles elective mutism and must be considered in the differential diagnosis[4], however the EEG of the patient was normal.

The mechanism involved in the development of convulsions and encephalopathy in shigellosis is poorly understood. There is a little evidence to support the shiga toxin hypothesis. The mechanism of the toxin and its relationship to other cytotoxins are still unclear if one or more toxins are involved[3].

Other pathologic mechanisms may contribute to the development of neurologic manifestations in shigellosis. Direct invasion of the central nervous system by shigella strains is rare and does not explain the frequent neurologic findings. Fever has been implicated as the cause of the neurologic manifestations; however, children may develop seizures during shigellosis even if they do not experience seizures during other febrile infections, and shigella-associated convulsions occur in children older than the usual age for febrile seizures. It has been suggested that the rate of temperature increase is especially rapid in shigellosis and accounts for the high incidence of seizures and other neurologic symptoms, but the clinical

assessment is difficult and this assumption has not been proven. In addition, host factors may play a role in defining individuals who are most susceptible to developing neurologic disturbances during shigellosis[2, 3, 6].

This case of shigellois with mutism should alert physicians that mutism can be a neurologic symptom of shigellosis, and that shigellosis should be considered in the differential diagnosis of mutism.

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