AN UNUSUAL NEUROLOGICAL MANIFESTATION OF SHIGELLA FLEXNERI

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Abstract

Shigellosis is an acute infection of the intestine caused by Shigella species, which presents as fever, abdominal cramps and blood and mucous in the stool. The complications of shigellosis include dehydration, rectal prolapse, hemolytic uremic syndrome, septicemia, reactive arthritis, and rarely, myocarditis. The central nervous system manifestations such as convulsions, delirium and coma have been recognized, and are more common in children. We report a case of a 45-year-old male patient with cerebellar symptoms associated with Shigella dysentery, which resolved upon treatment of the Shigellosis. This is an unusual complication of shigellosis, with paucity of documentation in medical literature.

Keywords: Shigellosis, acute infection, intestine
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Introduction

Shigellosis is one of the major causes of diarrhea in developing countries\(^{(1)}\). There are 4 species of Shigella: *Shigella dysenteriae*, *S. flexneri*, *S. boydii*, and *S. sonnei*. The complications of shigellosis may be intestinal or extra intestinal. Central nervous system complications, although rare, can cause significant morbidity, especially among children. Cases of cerebellitis associated with *S. flexneri* infections have not been widely documented as per our extensive search though literature. We present this extremely rare case report of an adult patient with *S. flexneri* associated cerebellitis.

Case report

A 45 year old male patient came to the emergency department of our tertiary care hospital with the complaints of low grade fever of 3 days duration. He also 4 to 5 loose stools per day, associated with blood for 2 days, with diffuse cramping abdominal pain. On day 1 of admission the patient noticed giddiness of sudden onset, generalised weakness and slurring of speech. He also noticed swaying to both sides while walking. He did not complain of headache, vomiting, loss of consciousness, or seizures. He denied the use of illicit drugs, over the counter medications or alternative medications.

At the time of admission, the patient was febrile (Temp - 102º F). The tongue was dry with the skin pinch time of <2 sec. The pulse rate was 100/min, and the blood pressure was 90/70 mm Hg. He had diffuse tenderness on palpation of the abdomen. On examination of the nervous system, he had a scanning type of dysarthria. The examination of cerebellar function revealed bilateral past-pointing, a broad based gait, gait ataxia and inability to do tandem walking. There was no hypotonia, pendular knee jerk, intentional tremors, or nystagmus. The examination of the other systems was unremarkable.

The blood investigations revealed a WBC count of 7300/mm\(^3\) with neutrophil predominance (70%). The renal parameters revealed an elevated blood urea nitrogen level of 21mg/dl and serum creatinine of 1.70 mg/dl, which became normal on the 3\(^{rd}\) day of admission. The microscopic examination of stool showed some red blood cells and plenty of pus cells.
An MRI Brain with contrast was done, which showed age related small vessel ischemic changes, along with mild diffuse cerebellar edema. The cerebrospinal fluid analysis showed a glucose level of 91 mg/dl (with a concurrent blood glucose value of 124 mg/dl), total protein level of 34.5 mg/dl, total count of 1 cell/ mm$^3$. The CSF culture did not show growth of any organisms. The CSF polymerase chain reaction assays for herpes simplex virus 1 and 2, Epstein-Barr virus, Cytomegalovirus, Varicella zoster virus, adenovirus, West Nile Virus, Parvovirus B19, Human herpes virus 6 and Enterovirus were all negative. The blood culture was negative as well. ELISA test done for human immunodeficiency virus I and II were negative.

The stool culture showed growth of *Shigella flexneri* which was susceptible to ceftriaxone and resistant to ciprofloxacin. In view of persistent cerebellar symptoms, in the absence of other identifiable infectious, metabolic and toxic etiologies, Shigella associated cerebellitis was suspected. The patient was started on intravenous ceftriaxone at a dose of 1gm twice daily, in addition to other supportive measures. After 2 doses of antibiotics, there was marked improvement in his symptoms. The disappearance of blood in the stool was followed by an improvement in the consistency and frequency of stools, followed by a significant improvement in his speech, and resolution of ataxia. He got discharged after 5 days of antibiotics. At the time of discharge, the CNS examination was completely normal. On follow up at 6 weeks, 3 months, and one year, the patient did not have any clinical features suggestive of gastrointestinal or residual neurological disease.

**Discussion**

The Complications of shigellosis are more common among children below the age of five. One of the most common extraintestinal manifestations of shigellosis is Shigella encephalopathy, which presents with altered consciousness, seizures and coma, which is frequently encountered in children$^4$. For long, it was postulated that Shiga toxin 1 and 2 were involved in shigellosis associated encephalopathy. The other hypothesized mechanism was direct infection of the central nervous system. Patients with shigellosis and encephalopathy have been found to have cerebral edema$^7$.

The patient had evidence of *S. flexneri* infection, which was confirmed by the stool culture. His gastrointestinal symptoms as well as cerebellar manifestations improved with 2 doses of parenteral antibiotics. Hence a diagnosis of *S. flexneri* dysentery associated acute
cerebellitis is justified in this case. We believe that this is the first such reported case of cerebellitis associated with Shigellosis due to *S. flexneri*.

**Conclusion**

Shigellosis is usually a self-limiting disease in adults, but we should always be vigilant for development of complications. Prompt treatment with appropriate antibiotic plays a major role in treating the complications. We would like to suggest that Shigellosis associated cerebellitis be included in the differential diagnosis of acute neurological dysfunction, if the patient has preceding gastrointestinal symptoms.

**Practical implication**

We would like to suggest that Shigella associated cerebellitis be considered as one of the differential diagnoses in acute neurological dysfunction caused by Shigella infection in adults.

**References**