Explosive *Campylobacter jejuni* Diarrhea in Immunoproliferative Small Intestinal Disease

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**Abstract**

*Campylobacter jejuni* is an infrequent cause of self-limiting acute diarrheal disease in adults in the Indian subcontinent. We report the occurrence of a life threatening diarrhea due to *C. jejuni* infection in a patient with immunoproliferative small intestinal disease. We postulate that immunosuppression due to malignancy, malnutrition and cancer chemotherapy was responsible for the unusually severe diarrhea. (*Indian J Gastroenterol* 1992; 11: 141-143)

**Introduction**

Although immunoproliferative small intestinal disease (IPSID) has been reported from several parts of the world, it is a major health problem only in the Middle East and North Africa. A few cases have been reported from India. These patients have a high incidence of gastrointestinal infections like giardiasis. We report a case of IPSID associated with a life threatening diarrheal disease caused by *Campylobacter jejuni*.

**Case Report**

A 43 year old man presented in March 1991 with history of abrupt onset of diarrhea, periumbilical colicky pain, vomiting and weight loss since October, 1989. He used to pass 4 to 5 watery stools per day which were free of mucus and blood. He lost about 10 Kg of weight in the ensuing three months. For these complaints, he was treated elsewhere empirically with tetracycline, ethambutol and rifampicin starting in December 1989. Over the next 3 months, he experienced marked relief of pain, partial improvement in diarrhea and weight gain. He continued to take the antitubercular drugs till January 1991, when while still on this treatment, he developed recurrence of diarrhea (6-8 stools/day) along with weakness, weight loss and pedal edema. There was no history of fever, cough, night blindness, bleeding from gums, bone pains or paraesthesia.

Physical examination revealed marked muscle wasting, mild pallor, pedal edema and clubbing of fingers and toes. There was no hepatosplenomegaly, ascites or palpable mass in the abdomen. Examination of chest and cardiovascular system was normal.

Laboratory data were as follows: hemoglobin 10 g/dL, total leucocyte count 17,400/µL, neutrophils 72%, lymphocytes 24%, platelets 500,000/µL, ESR 4 mm fall in 1st hour, serum creatinine 1.1 mg/dL, sodium 134 mEq/L, potassium 3.5 mEq/L, bilirubin 0.7 mg/dL, albumin 3.4 g/dL, AST 24 U/L and ALT 28 U/L. Serum electrophoresis showed a normal pattern. Serum immunoglobulins levels were as follows: IgG 1500 mg/dL, IgA 261 mg/dL and IgM 53 mg/dL (normal: IgG 1331±202, IgA 229±66, IgM 160±43). Turbidimetric method, Behring turbidimetric, Germany). Immunoelectrophoresis (Fig 1) using polyacrylamide purified monoclonal antibodies against α, λ, γ, μ and κ chains revealed normal patterns with α, κ and γ antisera and a very faint staining of normal pattern with κ antisera. The pattern for κ chain was sharp but showed an anodic extension that was not seen with the normal serum suggesting the presence of abnormal κ-chains. Stool smear examination was normal and culture for aerobic bacteria was negative. Chest X-ray showed bilateral pleural effusion. Small bowel bari um meal series
showed thickened and irregular folds in the duodenum, jejunum and ileum. Areas of narrowing and small intraluminal nodular filling defects were seen in the jejunum. Compression of small bowel loops by extramural masses was also seen. Fecal fat excretion on 50 g/day fat diet was 19.8 g/24h (average of three 24 h collections; normal <7 g/24h). Urinary D-xylose excretion was 0.5 g/5g/5h; normal 1.0 g/5g/5h). Jejunal fluid immunoelectrophoresis was not done.

At endoscopy, multiple small yellowish blebs were seen in the second part of the duodenum giving a cobblestone appearance. Biopsy from these blebs showed complete loss of villous pattern, paucity of crypts of Lieberkuhn and intense infiltration of lamina propria by lymphocytes, plasma cells, immunoblasts and eosinophils (Fig 2). Liver and bone marrow aspiration biopsies were normal and did not show any evidence of lymphomatous involvement. A diagnosis of IPSID stage B3 was made on the basis of clinical, biochemical, radiological and histological data. Systemic chemotherapy with cyclo-phosphamide, adriamycin, vincristine and prednisolone was started on an outpatient basis.

One day after the completion of the first cycle, he was readmitted in a moribund condition with severe dehydration and shock following an abrupt worsening of his diarrhea (25 to 30 watery stools/day, very large volume). Examination also revealed a left sided hemiplegia with an upper motor neuron type left facial nerve palsy. He was reeuculated with 9 L of intravenous fluids in the first 24 hours. Stool specimen obtained at admission was cultured on Brucella Agar Plate medium and showed growth of C jejuni. Diarrhea was controlled within 72 h with norfloxacin (400 mg twice a day) therapy. Cerebrospinal fluid examination did not show pleocytosis or abnormal cells. Computed tomography showed infarction in the right posterior and middle cerebral arterial territories along with compression of the ipsilateral lateral ventricle and effacement of ipsilateral sulci due to associated edema. There was no neurological improvement over the next two weeks. He refused further therapy and left the hospital after stabilization of general and neurological status.

Discussion

This case highlights the morbidity of a severe bacterial diarrhea occurring in a patient suffering from IPSID, by itself an uncommon disease in India. Published Indian experience of IPSID is limited to a single report by Tandon et al who described the clinicopathological features in three patients.

Clues to the diagnosis of IPSID in this case were a history of prolonged large volume intermittent diarrhea, clubbing and abnormal fecal fat and D-xylose tests. Intestinal tuberculosis may have co-existed in the initial part of the illness as suggested by an abnormal chest x-ray and initial favorable response to antitubercular therapy. Alternatively, the initial remission may well be explained by the natural history of IPSID.2-3 Relapse of symptoms in January 1991 while receiving anti-tubercular treatment is unlikely to be due to reactivation of tuberculosis. The correct diagnosis was suggested by the predominant duodenal involvement on small bowel barium meal series and was confirmed by histological examination of endoscopic duodenal biopsy.

C jejuni has been recognized as an etiological agent
of acute diarrheal disease only in the last two decades. In developed countries, *C. jejuni* affects both children and young adults and produces a self-limiting diarrheal illness. In one study, nearly two thirds of patients with *C. jejuni* diarrheas from Britain were adults. On the other hand, in developing countries, it causes acute diarrhoea predominantly in preschool children with the maximum incidence at the age of 12-17 months. Most children develop immunity by the age of 5 years. In contrast, *C. jejuni* has been isolated from only 1.9%-2.9% of adult patients with acute diarrhoea from the Indian subcontinent, in most in association with another enteropathogen. In adults, the disease is mild and no death has been reported.

To our knowledge, this is the first report of *C. jejuni* infection causing a life threatening diarrhoea in a patient with IPSID. *C. jejuni* has recently been described as an etiological agent of diarrhoea in immunosuppressed patients, viz those with human immunodeficiency virus infection. The unusually severe diarrhoeal disease in our patient may have been due to a depressed immune response as a result of malignancy, malabsorption, malnutrition and cancer chemotherapy.

References

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**JOURNAL NEWS**

*Journal membership*

The Treasurer, Indian Society of Gastroenterology has informed the Journal office that a large number of ordinary ISG members have not paid their subscription to the Society and hence to the Journal. As announced in the April 1992 issue, the Journal will not be mailed to these members w.e.f. October 1992 issue. Members are advised to check with the Treasurer, Indian Society of Gastroenterology (Dr M P Sharma, Department of Gastroenterology, All India Institute of Medical Sciences, New Delhi 110 029) the status of their membership dues with advice to the Journal office.

**C JEJUNI DIARRHOEA IN IPSID – PURI ET AL**

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