rule out motility disorder which was completely normal. Colonoscopy was normal with no evidence of any mechanical obstruction and mucosa was normal.

Full thickness rectal biopsy taken to rule out any visceral myopathy which showed expanded myentericplexus, increased nerve trunks and mature ganglion cells within myenteric plexus. Muscular layer didn't show any vacuoles. All these histological features were suggestive of megacystis microcolon intestinal hypoperistalsis syndrome.

On retrospective examination we also found bilateral mydriasis which failed to respond to pilocarpine, which was also reported in previous case reports[4]**.** Both parents underwent renal ultrasounds to rule out possibility of an autosomal dominantly inherited condition with reduced penetrance. No renal abnormalities were identified in parents. We have planned for proximal ileostomy to defunction distal colon. Total parenteral nutrition was started.

Discussion

Our patient presented with megacystis and proximally dilated small bowel loops, clinical features of Berdon syndrome. Berdon et al[1], first described what he called megacystis microcolon intestinal hypoperistalsis syndrome (MMIHS) in five infant girls, two of whom were sisters. MMIHS is a rare, severe form of functional intestinal obstruction in the newborn, and only more than 200 MMIHS cases have been reported in the literature[5]. The frequency of the disease is observed three or four times more in girls than in boys. The clinical course of MMIHS is complicated, and patients usually become symptomatic early in life[5]. A consensus on the theory of the pathogenesis of the disease does not exist yet. Some of the theories are the lack of nicotinic acid receptor subunits, a defect in fiber synthesis, an inflammatory process of the gastrointestinal and urinary tract, generalized axonal dystrophy in central, peripheral and autonomic nervous system[6]. Treatment is supportive and involves an ileostomy to defunction the colon, with TPN. Because of the gastrointestinal dysmotility, attempts to give enteral feeding may cause fatal pneumonia; on the other hand, to prevent malnutrition, most patients with MMIHS are maintained by parenteral nutrition, which may lead to the loss of venous access, thrombosis of vascular access, catheter sepsis and chronic liver failure[5]. Long term survival is sometimes noted in patients with MMIHS receiving TPN; however, most patients develop complications from prolonged TPN[5]. Therefore, the prognosis of MMIHS is poor, and most patients die early in life. Major causes of death are sepsis, malnutrition, renal insufficiency, and multiple organ failure[1].

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Uncommon presentation of Strongyloidiasis: chronic malabsorption, multiple small bowel strictures and appendicitis in HTLV-1 positive patient

Introduction

Strongyloidiasis is a parasitic disease, caused by a nematode helminth, Strongyloidesstercoralis. Strongyloidiasis may present with indigestion, abdominal pain, vomiting, chronic diarrhea, and weight loss. Intestinal obstruction is a very rare complication of strongyloidiasis. Protein losing enteropathy and appendicitis are also rare complications of strongyloidiasis.

Case report

A 24 year old male presented with recurrent episodes of abdominal pain with vomiting and loose motions lasting for 1-2 days for last 8 years with frequency of one episode a year. He presented to us aggravation of symptoms and weight loss of 20 kgs for 2 months. Before two and half months he had an episode of severe abdominal pain and vomiting. He was diagnosed as acute appendicitis with small perforation and underwent appendicectomy. On physical examination the patient was malnourished with BMI of 12, afebrile, had blood pressureof 100/60 mmHg, pulse rate of 110 beats per minute and a respiratory rate of 24 breaths per minute. He had pallor and bilateral pedal edema. He did not have lymphadenophaty, cyanosis or clubbing. The spleen and liver were not palpated. There was no evidence of free fluid in abdomen. Laboratory investigations showed Hb -7 gm%, TLC-14,000 cumm and platelet count of 2,76,000 cumm. He had total bilirubin 1 mg/dl, AST 47 IU/L, ALT 26 IU/L, ALP 604 IU/L. Serum proteins were decreased substantially with total protein 3.8 gm%, serum albumin 1.2 gm% and serum globulin 2.6 gm%. He was HIV negative and immunoglobulin levels were within normal limit. But he was detected to be positive for HTLV-1.

Fresh stool sample was sent for examination suggestive of presence of mucus, occult blood with 8-10 pus cells/HPF, 30-40 Red blood cells/HPF and multiple larvae of *Strongyloidesstercoralis* (**Figure 1**).

Upper GI endoscopy was performed in view of persistent vomiting. It was suggestive of diffusely erythmatous, edematous and friable mucosa with multiple hemorrhagic lesions in antrum and pylorus. Duodenal mucosa also had similar features with large ulcer in 2nd part of duodenum and narrowing of the lumen in the 3rd part of the duodenum (**Figure 2a,b**).

Biopsies of the antrum and duodenum revealed presence of multiple adult and larval forms of *Strongyloidesstercoralis* on the mucosal surface as well as in the submucosa (**Figure 3**).

Barium small bowel series also showed large ulcer in 2nd

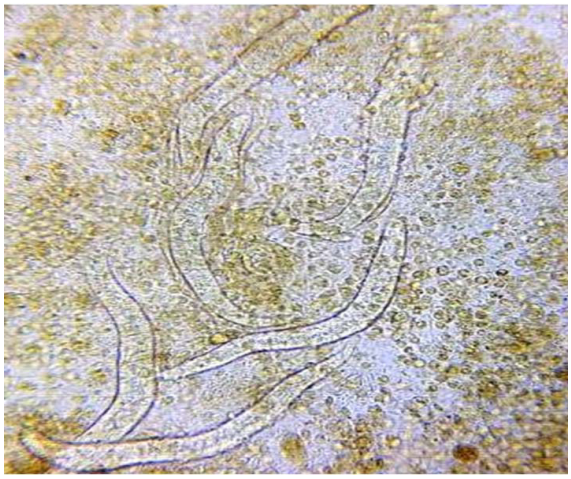

Figure 1: Stool sample showing multiple larvae of strongyloides stercoralis

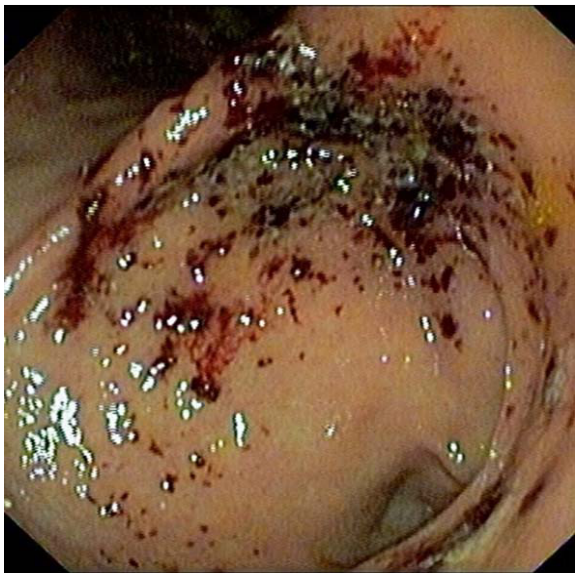

Figure 2a: UGI endoscopy showing multiple haemorrhagic stops in antrum

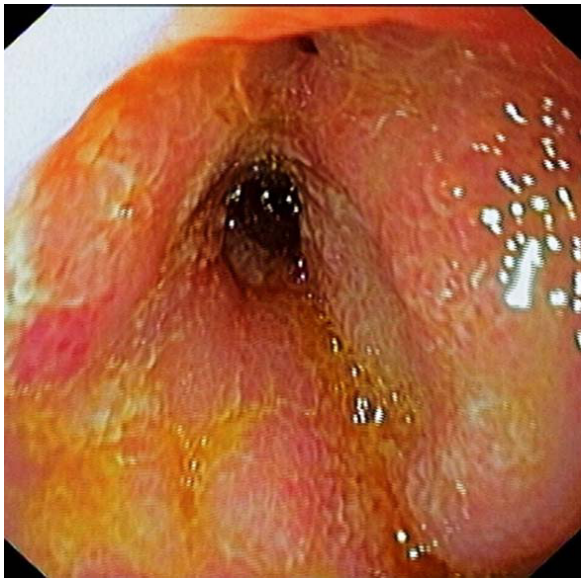

Figure 2b: Duodenal ulcer with narrowing of second part of duodenum

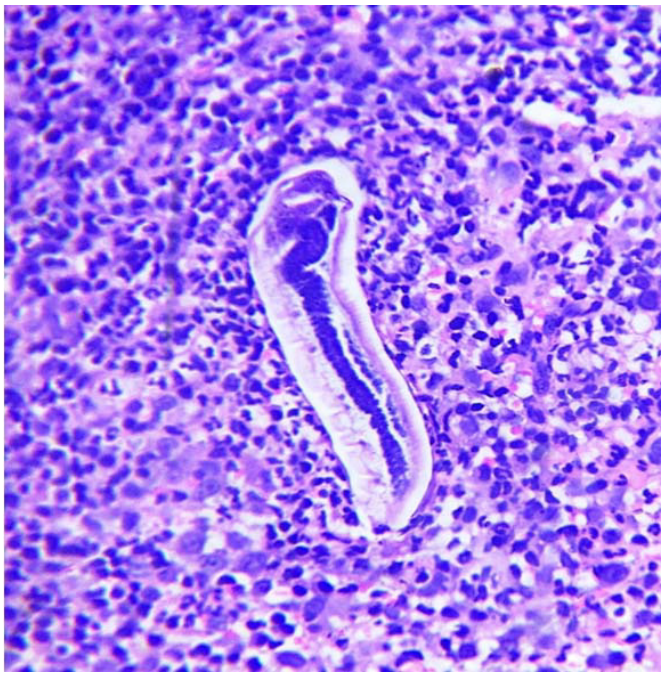

Figure 3: Antral showing adult and larval form of strongyloides stercoralis

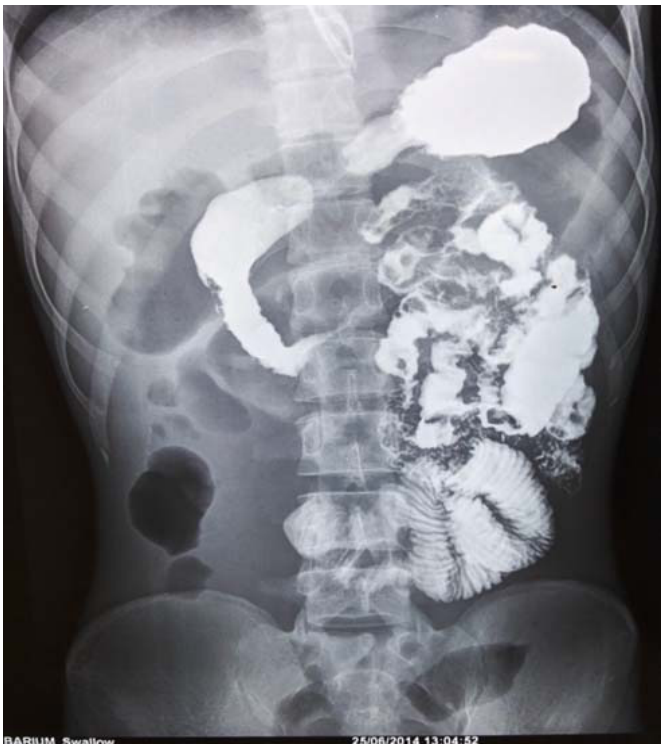

Figure 4: Barium meal follow through study showing multiple strictures in jejunum and ileum

part of duodenum, stricture at duodenojejunal flexure, multiple levels of strictures in jejunum and ileum with dilated bowel loops and sluggish peristalsis (**Figure 4**).

Patient was started on IV fluids, IV antibiotics and enteral nutrition. He was treated aggressively with oral Ivermectin (6 mg) OD and albendazole (400 mg) BD. Treatment was continued 2 weeks after stool became negative for *Strongyloides*. Patient responded in form of decrease in abdominal pain, vomiting and diarrhea and weight gain.

Discussion

Risk factors for such severe disseminated strongyloidasis are immune deficiency, hematologic malignancy, steroids administration, HTLV-1 infection, chronic alcoholism, renal failure and transplantation.[1] Most likely precipitating factor for hyper infection in present case was HTLV-1 infection.

Gastrointestinal involvement causes symptoms like anorexia, nausea, vomiting, weight loss, abdominal pain, flatulence, and diarrhea. Less commonly, in severe disease intestinal obstruction, gastrointestinal bleeding, paralytic ileus, protein losing enteropathy and malabsorption syndrome can occur.[2]

Duodenal obstruction is a rare complication of Strongyloides infection.[3]

Eosinophilia is an inconsistent finding, present in up to 35% during the acute phase, and less frequent in patients with chronic or disseminated disease[4]. Most patients with duodenal obstruction presented with normal or low eosinophil count indicating a chronic infection. Our patient has normal eosinophil count suggesting chronic infection.

Diagnosis of Strongyloides is routinely done by stool examination which has only 30% sensitivity. Duodenal aspirate is the most sensitive method for diagnosis. In our case larvae were found in stool as well as in duodenal biopsy. Recently, Kishimoto et al. showed that the S. stercoralis larvae identification in duodenal biopsies is feasible in 71% of cases.[5]

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