Aplastic anemia and Crohn’s disease – coincidence or association?

A 41-year-old man presented with history of swelling over the right lower abdomen, post-meal abdominal discomfort, and abdominal pain lasting for 1-2 hours, relieved with defecation and passing flatus, for the last 10 years. He had anorexia and had lost 23 kg during this period. There was no history of bleeding manifestations. He was evaluated elsewhere and received antitubercular treatment for 9 months. He also had pancytopenia at that time, for which he was subjected to bone marrow examination and was diagnosed as having aplastic anemia, which improved partially with stanazolol.

At admission with us, his hemoglobin was 4.4 g/dl, total leukocyte count 1900/µL, platelet count 24000/µL and reticulocyte count 0.4%. Repeat bone marrow examination showed hypocellular marrow with depression of erythrocyte, myeloid and megakaryocyte lineages. Bone marrow biopsy revealed predominant fat spaces with overall cellularity <5%, consistent with diagnosis of aplastic anemia, which improved partially with stanazolol.

Ultrasoundography showed fatty liver and thickening of terminal ileal and cecal wall. Colonoscopy showed cecum to be grossly deformed and ulcerated with friability and nodularity. Ileocecal valve was deformed and ulcerated. The rest of the colon showed loss of vascularity and few haustiations. There were no ulcers, polyps or cobblestone appearance. Colonic biopsy revealed chronic colitis with ulceration in the cecum. Barium meal follow-through study showed normal stomach, duodenum, jejunum and proximal ileum. Cecum and proximal ascending colon were grossly deformed. On reviewing his earlier colonic biopsies, his clinical picture and no response to antitubercular drugs, a diagnosis of Crohn’s disease was established.

An association between aplastic anemia and inflammatory bowel disease, both Crohn’s disease and ulcerative colitis, has been noted. Bone marrow aplasia has also been reported after drug therapy for Crohn’s disease.

Whether underlying immunologic irregularities account for the development of both aplastic anemia and inflammatory bowel disease or whether immunosuppression caused by one disease predisposes to development of the other is unknown. The dysfunctional neutrophils characteristically found in patients with aplastic anemia could predispose to chronic gut infections, which can play an important pathogenetic role in inflammatory bowel disease. Some investigators recently reported an increased susceptibility to Crohn’s disease in patients with missense and frameshift mutations in the NOD-2 gene, which is important in the regulation of the immune system. We speculate that mutations in NOD-2 gene could lead to development of both Crohn’s disease and aplastic anemia through altered NF-kB regulation of hematopoietic progenitor cell development.

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References


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of large joints. Clubbing has been associated with various gastrointestinal disorders like inflammatory bowel disease, sprue, and bowel neoplasms. It has also been rarely associated with esophageal carcinoma, Plummer-Vinson syndrome and achalasia cardia. There is only one earlier report of its association with corrosive stricture of esophagus.

An 18-year-old girl presented with history of dysphagia three months following ingestion of formic acid. She had noticed pain and progressive enlargement of all her digits about one month after ingestion of the corrosive. Clinical examination revealed grade IV clubbing of all digits of both hands and feet (Fig). The patient did not have cyanosis and had no evidence of cardiac disease, lung disease, inflammatory bowel disease, or any connective tissue disorder. No other family member had this finding. Cardiac and respiratory systems were normal.

Investigations: hemoglobin 12.6 g/dL, WBC 6400/mm³ and ESR 14 mm in 1st hour. Liver and renal function tests were normal. Echocardiography and radiography of the chest revealed no abnormalities. X-rays of hand and foot showed periosteal elevation characteristic of HOA (Fig, inset). Barium swallow revealed stricture in the mid esophagus with proximal dilatation. Upper GI endoscopy showed a stricture in the middle third of esophagus.

She underwent five sessions of dilatation using Savary-Gilliard dilators. The patient had relief of dysphagia and gradual regression of clubbing after 9 months.

The exact mechanism of clubbing is unknown. The theory that stimulation of vagal neural arc is an etiological factor is supported by reversal of the syndrome after vagotomy. Recently various growth factors like fibroblast growth factor, hepatocyte growth factor, platelet-derived growth factor and vascular endothelial growth factor have been proposed to play a role in the formation of digital clubbing.

Development of clubbing in this case appears to be related to esophageal stricture, as there was no other cause, and the clubbing regressed following stricture dilatation.

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References

Endoscopic management of esophageal bezoar in a child

Bezoars are very rare in the esophagus. We report a young child with esophageal bezoar who was managed endoscopically.

A 15-month-old male child was referred with complaints of vomiting and cough after intake of solid or semisolid food, for four months. The symptoms were progressive and for the last one month, the child had vomiting immediately after each meal. He had lost one Kg of weight. The child was delivered full term, normally, and weaning was started at the age of 6 months. On examination he was mildly malnourished; his weight was 7.5 Kg, height 78.7 cm. Milestones were normal and systemic examination was unremarkable.

Hemogram, renal and liver function tests were within normal limits. Chest X-ray was normal. Barium swallow showed smooth narrowing at the lower third of the esophagus, and above this the esophagus was grossly dilated with multiple filling defects of varying size.

Upper GI endoscopy with forward-viewing video endoscope (GIF-V; Olympus, Tokyo, Japan) under conscious sedation showed dilated esophagus, filled with multiple brownish black semisolid material of size 0.5-2 cm. This consisted of multiple food particles including seeds and betel nut (Fig). These were removed by using a Roth retrieval basket (indigenously made; Endotech, Jaipur). After clearing the esophagus, a web was seen at 18 cm from the incisors, which was dilated with balloon dilator (MaxForce TTS; Boston Scientific, USA). The