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SHORT REPORT

Xanthogranulomatous inflammation of ascending colon with mucosal involvement: Report of a first case

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Abstract

Xanthogranulomatous inflammation (XGI) is a rare phenomenon and can involve any organ system. The involvement of colon is rarely described in the literature. We herein report a case of XGI in a 60 year-old male who presented with friable ulceroinfiltrative mass in colon. Right hemicolectomy was performed with clinical and radiological suspicion of malignancy. This is the first reported case of XGI in ascending colon with mucosal involvement.

1. Introduction

Xanthogranulomatous inflammation (XGI) is a rare but well-described disease process first reported in the genitourinary tract.[1] It can involve any organ, but the most common sites are kidney and gallbladder. Other possible locations include endometrium, ovary, fallopian tubes, vagina, testis, epididymis, gall bladder, stomach, bone, skin, appendix, urinary bladder, thyroid and adrenal glands. Involvement of the colon is very rare. There are so far only three reported cases, two involving sigmoid colon and one involving cecum.[2–4] It presents as a mass-like lesion, with predominant submucosal involvement. Infiltration into the surrounding tissues often mimics advanced cancer clinically, radiologically as well as on macroscopic examination of the specimens. Mucosa was also affected in our case while it was normal in all the previously reported cases. We herein report a case of XGI that presented as a friable ulceroinfiltrative mass in the ascending colon. With the best of our knowledge this is the first reported case of XGI in ascending colon with mucosal involvement.

2. Case report

A 60 year-old male presented to the emergency department with pain in the abdomen. He had history of recurrent episodes of vomiting with fullness in the abdomen and constipation for the last one month. There was no history of hematemesis, melena or weight loss. He had no past history of diabetes, hypertension or tuberculosis. He was a chronic smoker and alcoholic. His past history was significant for hernia repair surgery two years back. General physical

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and systemic examination including respiratory, cardiovascular and central nervous system showed no significant abnormality. Per abdomen examination revealed a lump palpable in the right hypochondrium measuring about 6×6 cm. The lump was firm, non-tender and moving with respiration. No free fluid was present in the abdomen. Per rectal examination was normal. X-ray chest and electrocardiography (ECG) were within normal limits.

Contrast enhanced computed tomography (CECT) of abdomen showed irregular, circumferential and contrast enhancing thickening involving approximately 6 cm of the ascending colon (Fig. 1A). Multiple small peri-cecal lymph nodes were also seen. Colonoscopy revealed large friable mass present within the ascending colon (Fig. 1B).

Diagnosis of neoplastic mass lesion, with surrounding haziness, stranding and nodularity, suggestive of pericolonic spread was entertained. Multiple biopsies were taken.

Histopathologic examination revealed multiple fragments of colonic mucosa, which showed mild focal distortion of crypt architecture with adequate goblet cell population. Lamina propria showed moderate lympho-plasmacytic infiltrate including many eosinophils. There were scattered large cells having hyperchromatic nuclei with irregular nuclear margin and moderate finely vacualated cytoplasm recognized. The large cells were negative for cytokeratin (CK) and leukocyte common antigen (LCA). CD68 immunostain was positive. No granulomatous or neoplastic pathology was seen. A diagnosis of XGI was suggested. However, in view of strong clinical and radiological suspicion of malignancy, right hemicolectomy was performed by laparoscopic minimal access surgery.

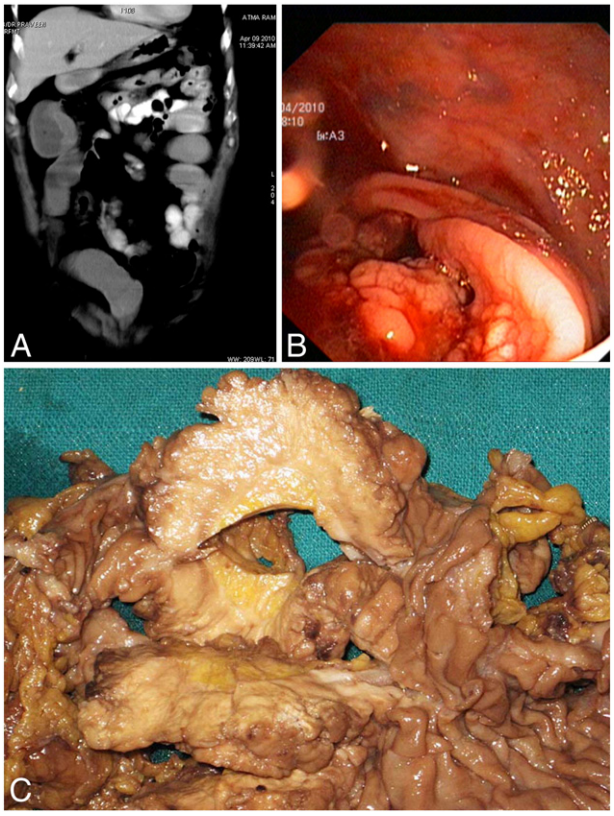

Figure 1 Contrast enhanced computerized tomography abdomen shows irregular circumferential enhancing thickening involving ascending colon (A), large friable mass in ascending colon on colonoscopy (B), and gross photograph to show markedly thickened and yellowish colonic wall (C).

The specimen on gross examination showed an ulceroproliferative growth situated 4 cm from the distal resected end (Fig. 1C). The growth involved the entire circumference. On opening it was greyish yellow, firm and infiltrating the full thickness of the wall. Ileum, rest of the colon and appendix were within normal limits. Serial sections of mesocolon revealed six lymph nodes varying in size from 0.2 to 0.5 cm.

Microscopic examination from the mass lesion showed diffuse infiltration of colonic wall by large cells (Fig. 2A–C). These cells possessed irregular nuclei with occasional prominent nucleoli and abundant foamy cytoplasm; morphologically resembled macrophages or histiocytes. Many multinucleated foreign body type of giant cells and admixed inflammatory cells rich in lymphocytes and eosinophils were also seen. Many cells also had signet ring like appearance. The cells were seen throughout the wall from mucosa to subserosa, though the infiltrates were more in submucosal region than in the mucosa. The colonic glands were focally displaced while muscularis propria showed marked destruction by these cells. At places collagenised stroma was also noted. No increase in mitosis was seen. The resection margins (proximal, distal and radial) and all the six lymph nodes isolated from pericolic fat showed no such cells. Random sections from the ileal and colonic tissue as well as from the appendix showed no significant abnormality. There were no features to suggest inflammatory bowel disease, diverticulosis or tuberculosis. The cells were negative for mucin stains (mucicarmine and alcian blue-PAS). The large cells including ones with signet ring configuration were positive for CD68 immunostain, proving that they were of histiocytic in origin. Cytokeratin was negative while LCA stain was positive only in background lymphoid cells (Fig. 2D–F). The morphology was that of xanthogranulomatous colitis.

The patient was discharged on the 4th day of surgery and is doing well in 5 months follow up period.

3. Discussion

XGI is a rare chronic inflammatory condition that is characterized by aggregation of lipid-laden foamy macrophages or xanthoma cells. This disease entity is well recognized in the kidney and gallbladder, yet involvement of the gastrointestinal tract is extremely rare. To the best of our knowledge, there are only three cases so far reported in the colon. (Table 1) Clinically, it can be difficult to differentiate from infiltrative cancer because XGI usually presents as an irregular mass-like lesion with an extension of fibrosis and inflammation into the surrounding tissues, which often mimics infiltrative cancer.

There are many case reports of secondary involvement of colon in cases of xanthogranulomatous cholecystitis, masquerading as stage IV cancer of gallbladder, including two cases of our own experience.[5] Secondary anorectal involvement by primary mullerian duct remnant xanthogranulomatous abscess was first described by Devis et al.[6] XGI involving the large bowel as primary site is extremely rare. All three