Chronic perforation in isolated xanthogranulomatous gastritis

Xanthogranulomatous involvement of the gastrointestinal tract is rare, gall bladder is the commonest site affected. Rare involvement of the sigmoid colon, stomach and periampullary region have been reported. Most of these lesions impose preoperative diagnostic difficulties, malignancy being the most common misdiagnosis. Isolated cases of xanthogranulomatous gastritis is very rare and infiltration from an associated xanthogranulomatous cholecystitis is a possibility. We describe a diabetic patient with isolated xanthogranulomatous involvement of the stomach with chronic perforation and communication with the liver parenchyma.

Case report

A 51-year-old diabetic male patient presented with upper abdominal pain of moderate intensity for six months. He had had one discrete episode of severe, dull epigastric pain associated with low grade fever but no vomiting, requiring admission in another medical centre. There was no history of melena, jaundice or weight loss and the rest of his medical history was unremarkable. On examination he was found to have pallor, epigastric tenderness and mild hepatomegaly. Laboratory investigations showed a low hemoglobin level (9.8 gm%). Liver function tests and C-reactive protein level were normal. Screening for HIV, Hepatitis B and Hepatitis C were negative.

Abdominal ultrasonography revealed a heterogenous mass in the left lobe of the liver contiguous with a thickened pylorus. There was another well defined lesion in segment VII of the liver with cystic and necrotic areas. A contrast enhanced CT scan of the abdomen showed a heterogeneously enhancing pyloric growth with perigastric stranding (**Figure 1**). The lesion was infiltrating the liver and there was a cystic focal hepatic lesion, suspicious of metastasis. The gall bladder was normal. The rest of the abdominal and pelvic organs were normal.

Image guided fine needle aspiration cytology (FNAC) from the pyloro-hepatic mass was performed with three passes. The aspirate showed atypical cells, but no definite evidence of malignancy.

Esophago-gastroduodenoscopy showed a nodular friable mucosa in the anterior and superior wall of the antro-pyloric region of the stomach. The first and second parts of the duodenum were normal. Biopsy from antro-pyloric region showed no specific lesion.

At laparotomy, a pus-containing fistulous tract was seen arising from the anterior wall of the stomach, extending onto the inferior margin of segment IV of liver. A small perforation with thickening was noted in the region of the stomach adherent to the liver substance. No definite mass was palpable and there was no significant lymphadenopathy. The gall bladder was normal. The findings were suggestive of an inflammatory etiology rather than malignancy. A frozen section from this region was reported to show xanthomatous and granulomatous inflammation, with no evidence of malignancy. A distal gastrectomy, rather than a subtotal gastrectomy, as planned earlier, was performed. The cystic liver lesion in segment VII was not explored. He had an uneventful postoperative recovery.

Pathological examination of the distal gastrectomy specimen showed diffuse thickening of the gastric wall. The cut surface was soft and gray white with yellowish foci extending into the attached adipose tissue. The adjacent mucosa appeared unremarkable (**Figure 2**). Microscopic examination revealed transmural infiltrates of lymphocytes, plasma cells, many lymphoid follicles, and numerous aggregates of foamy histiocytes (**Figure 3**). A few granulomas composed of epithelioid histiocyte and Langhans' type multinucleate giant cells were also seen (**Figure 4**) but caseation, acid fast bacilli (AFB) or fungal organisms were not present. Gram stain did not show

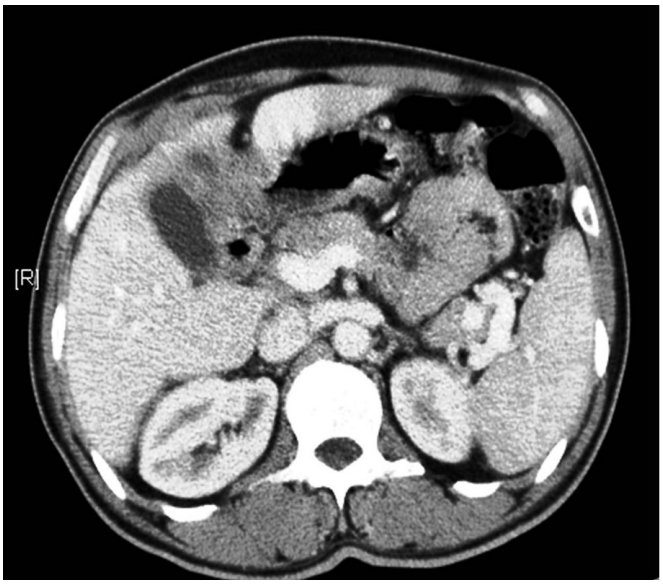

Figure 1: Contrast enhanced CT scan of the abdomen showing heterogeneously enhancing pyloric growth with hepatic infiltration and perigastric stranding.

Figure 2: Gray-white cut surface of the thickened wall of the distal gastrectomy specimen with foci of yellowish discoloration.

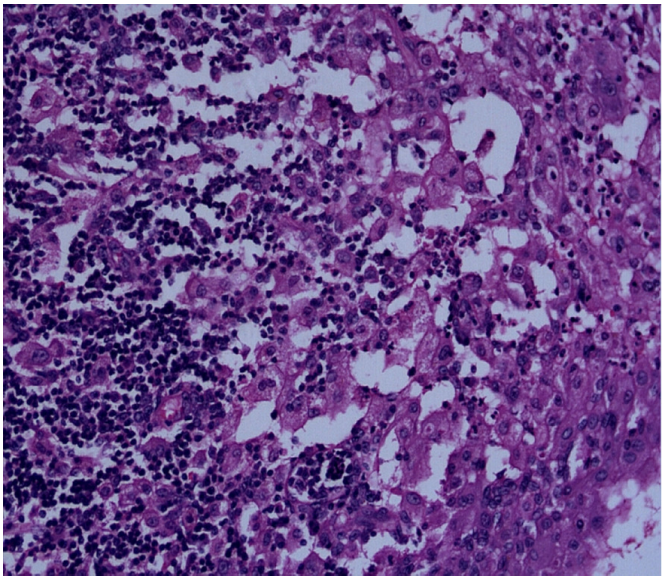

Figure 3: Foamy histiocytes admixed with lymphocytes in the gastric wall (Hematoxylin and Eosin, 200x magnification)

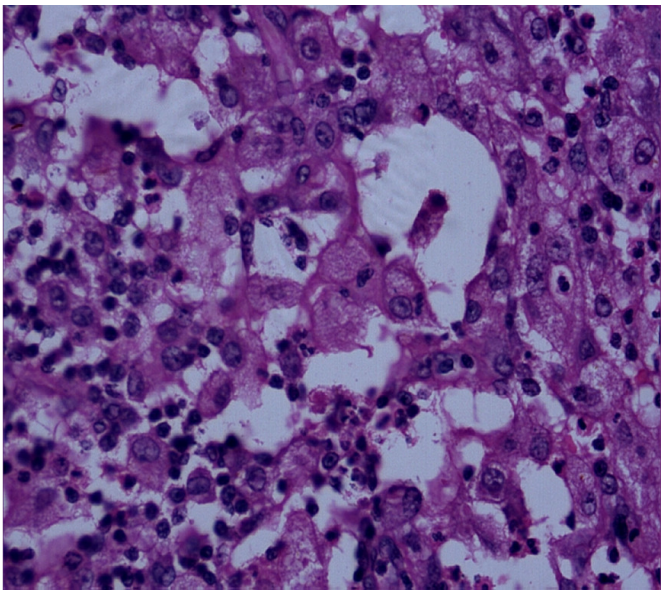

Figure 4: Focal granulomatous inflammation with epithelioid histiocytes in the gastric wall (Hematoxylin and Eosin, 400x magnification)

significant bacteria within the infiltrate. There was submucosal fibrosis and hypertrophy and fibrosis of the muscularis propria with neuronal hyperplasia. There was no evidence of malignancy. On immunohistochemistry, the foamy histiocytes expressed CD68 and were negative for cytokeratin and smooth muscle actin . Based on these findings a diagnosis of xanthogranulomatous gastritis was made.

The patient was well at 20 months' follow up. Gastroscopy showed a jejunal stomal ulcer and biopsy from this site showed chronic active jejunitis with fibrosis. Separate biopsies from the gastrojejunostomy stomal site showed non-specific changes and no granulomas. AFB stain was negative.

Discussion

Xanthogranulomatous gastritis (XGG) is a rare disorder with few case reports in literature[1,2,3]. In the gastrointestinal tract, the gall bladder is the most common site affected by xanthogranulomatous (XG) inflammation and a prevalence of 9% has been reported in a series of cholecystectomies from a large center in India[4,5]. Infiltration of adjacent organs can occur in xanthogranulomatous cholecystitis (XGC), mimicking gall bladder cancer[6]. Gastric involvement by XG inflammation has been reported to be secondary to extension from the gall bladder rather than a primary XGG[2,3], whose existence has even been questioned in literature[3].

Isolated XG inflammation has been reported to present as rapidly enlarging submucosal nodules in the stomach and in the sigmoid colon[1,7]. There is a report of XG inflammation causing an obstructive lesion in the sigmoid[8]. Other organs that can be affected by xanthogranulomatous inflammation include the kidney, bone, ovary, soft tissue, pancreas, lymph nodes and the periampullary region[3,4]. It is difficult to make an accurate preoperative diagnosis of xanthogranulomatous inflammation, an infiltrative malignancy is a common preoperative diagnosis[1,4,6,7]. Acute abdominal pain and fever in the index patient could be due to small, sealed perforation of the stomach, resulted in invasion of liver. Perforation of the gall bladder was observed at operation in XGC[9].

Other radiological differential diagnoses that have been considered with involvement of the sigmoid colon include pelvic inflammatory disease and actinomycosis.[7] Most patients require operative intervention because of symptoms, complications and an inconclusive preoperative diagnosis. Intraoperative frozen section examination was valuable however, to avoid an unnecessary radical resection, as has been emphasized in literature[7].

The precise pathogenesis of isolated XGG is unknown. When XGC and XGG co-exist, it is conceivable that deepening of the XG process in the gall bladder and adhesions may result in transmural involvement of the gastric wall[2]. A bile- reflux induced pathogenesis, like in XGC, has been implicated in XGG, especially when cholesterol crystals have been detected in the inflammatory infiltrate[2]. Bile reflux has also been presumed to have a role in the pathogenesis of gastric xanthoma, a more localized but similar lesion[10]. We did not find any evidence of bile reflux in the stomach. Sinus and fistula formation and the involvement of distant sites, although uncommon, are recognized complications of XG inflammation of the gall bladder, kidney and appendix[11,12]. Although sealed perforation of the stomach can present as a pseudotumor mimicking gastric malignancy, there is only one case report of gastric perforation associated with XG pseudotumor[13]. Rare cases of xanthogranulomatous cholecystitis with liver abscess[14] and XG pyelonephritis with secondary involvement of the liver have been reported[15].

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Tubercular ileal stricture with enterolithiasis presenting as massive lower gastrointestinal bleed

Abdominal tuberculosis is still prevalent in third world nations and has shown resurgence even in developed countries especially amongst immigrants and immunocompromised patients. Intestinal tuberculosis presents chiefly in the form of obstruction or perforation and both enterolithiasis as well as massive lower gastrointestinal bleed have rarely been reported.

Case Report

A 70-year old male presented with persistent bleed per-rectum associated with postural symptoms. There was no prior history of hematemesis, obstructive bowel symptoms or tuberculosis. Physical examination revealed orthostatic hypotension and maroon colored blood per rectum. The nasogastric aspirate yielded bilious clear fluid. Colonoscopy demonstrated maroon colored blood with clots till terminal ileum and no significant pathology. The patient required 6 units of blood transfusion over 24 hours to maintain hematocrit and in view of persistent hemodynamic instability despite adequate volume replacement, the patient was considered for emergent surgical exploration. Laparotomy revealed circumferential stricture of mid-ileum with saccular dilatation of proximal bowel containing multiple enteroliths. The mesenteric lymph nodes were enlarged and matted. Gall bladder and common bile duct were normal. The patient underwent segmental resection of stricture bearing segment of ileum. Cut section of the specimen revealed 69 black colored enteroliths varying in size from 1-2 cm. The mucosa of the saccular portion of the bowel proximal to the stricture was grossly ulcerated (**Figure 1**). Histology was consistent with tubercular ileal stricture. The postoperative course of the patient was uneventful and was discharged on anti-tubercular therapy.

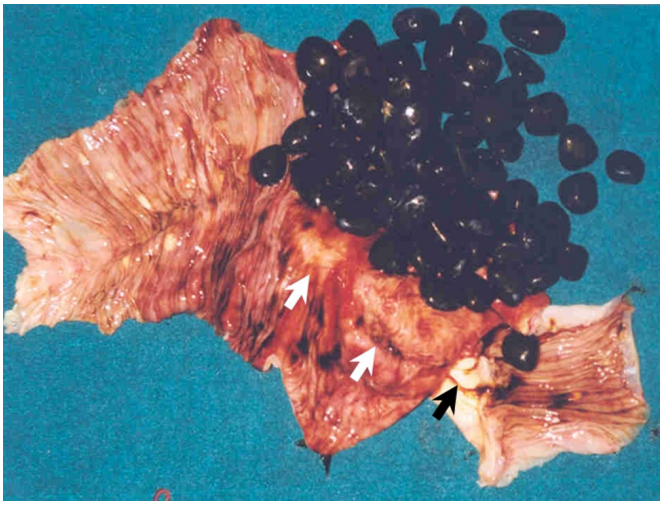

Figure 1. Resected ileal segment with enteroliths showing stricture (black arrow) and dilated proximal bowel with ulcerated areas (white arrows) caused by enteroliths.

Discussion

Enterolithiasis is a rare clinico-pathological entity characterized by formation of concretions within the gastrointestinal tract.[1] Stasis is the crucial factor leading to their formation as occurs in diverticula, proximal to intestinal strictures or in association with blind loop syndrome.[1] Enteroliths may vary in shape and size in the same patient and may be single or multiple.[2] In most of the instances enteroliths are 'innocent bystanders' that merely indicate the presence of underlying disease causing stasis. However, it