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Case Report

Acute Phlegmonous Gastritis with Pangastric Necrosis-a Perplexing Presentation in a Young Male with Pyrexia of Unknown Origin

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Abstract

Acute gastric necrosis is an extremely rare condition. Though its pathogenesis is debatable, acute massive gastric dilatation has been attributed as the commonest predisposing condition. The onset of symptoms is generally catastrophic and almost invariably calls for emergent and aggressive surgical therapy as delayed intervention entails increased mortality. We present a case of an 18 year-old male patient with pyrexia of unknown origin and pancytopenia, who presented with pangastric necrosis with anterior gastric perforation communicating with anterior abdominal wall abscess secondary to acute phlegmonous gastritis, an etiology infrequently reported in the literature. Emergency laparotomy with total gastrectomy and drainage of abscess was done. Post op course in the Surgical Intensive Care Unit was complicated with sepsis, acute respiratory distress syndrome, pneumothorax and brachial plexopathy following prone position ventilation. After prolonged intensive care with multidisciplinary consultation, patient condition improved and is in good health after discharge. We stress the importance of early detection of such rare surgical condition to prevent potentially life-threatening outcomes and role of critical care in management of such cases.

Keywords: Acute plegmonous gastritis; Gastric necrosis; Intensive Care Unit; Pyrexia of unknown origin

Introduction

Acute gastric necrosis described first by Duplay in 1833 is an extremely rare condition with only about 60 cases reported in literature [1]. Since the stomach has a rich vascular supply which generally protects it from ischemia, gastric necrosis is an unusal occurrence [2,3]. Various etiologies such as acute gastric dilatation, vascular causes, bulimia, selective vagotomy, abdominal trauma, infectious disease, gastric volvulus, intrathoracic herniation, necrotizing gastritis, ingestion of caustics have been described [3,4] Of the reported cases more than half had preceeding cause as acute massive gastric dilatation with intragastric pressure in excess of 30 cm H2O, leading to impaired intramural blood flow [2-5]. Acute phlegmonous or necrotising gastritis leading to gastric necrosis is thus an absolute rarity. We present the case of a young male with pyrexia of unknown origin and pancytopenia manifesting this uncommon condition, who was operated upon and managed successfully postoperatively in the intensive care unit. We intend to stress the importance of early diagnosis and intervention, and indispensable role of critical care in successful outcome in such cases.

Case Report

A 18 year old male presented to the emergency medical services at our centre with history of high grade fever, non radiating diffuse upper abdominal pain and multiple episodes of non bilious vomiting for one week. He had 2-3 episodes of hematemesis and malena 4 weeks before, without bleeding from other sites. He was admitted at another centre for above complaints with past history of moderate grade fever, anorexia and weight loss of six months duration. Over there he was found to have pancytopenia. Suspecting aplastic anemia on bone marrow studies and haematological investigations, he was administered Granulocyte Monocyte-Col-

ony Stimulating Factor (GM-CSF) injections subcutaneously in anterior abdominal wall with multiple units of blood and blood product transfusion. He was later referred to our centre for further management. There was no history of any medical or surgical illness or addiction in the past.

On examination he was found to be lethargic, febrile, with a radial pulse of 140/ min and tachypnea (40/ min) with respiratory distress. Marked pallor with bipedal pitting edema with mild scleral icterus was noted. Clubbing and lymphadenopathy were absent. Per abdomen examination revealed tender induration of epigastric and left upper quadrant region with erythema. Tender hepatomegaly of 8 cm and splenomegaly of 6 cm below the right and left costal margin respectively was noted. Breath sounds were reduced in bilateral bases without adventitious sounds. With a referral note of diagnosis of aplastic anemia, he was admitted under hematology services in hematology ICU. Suspecting a probable iatrogenic injection abscess of anterior abdominal wall with abdominal sepsis, he was started on intravenous Meropenem and Teicoplanin after drawing blood cultures. He was put on non invasive ventilator for his respiratory distress.

Hemogram showed (Hb-6.6 gm %, RBC-2.67 million/ mm3, MCV-86 fl, MCH-29.5 PG, MCHC-34.3 g/dl, Retic-0.37 %, RDW-16, WBC-4500/mm3%, with marked neutrophilia and shift to left and toxic changes, Platelet count-43,000/mm3). Coagulation profile was normal. Liver function test showed direct hyperbilirubinemia (total bilirubin-2.8 mg % and direct fraction-1.8 mg%) with hypoproteinemia (total protein-4.8 gm% and albumin-1.3 gm%) with normal liver enzymes. A normal renal function test, serum amylase and lipase were noted. Serum procalcitonin was 14. 5. Ultrasound guided pleurocentesis of bilateral moderate pleural effusion showed exudates with WBC-850/ mm3 with lymphocytosis-70% with numerous RBCs, gram stain and acid fast organism negative. Fluid protein was 2.8 gm% and LDH-369 u/l satisfying Lights criteria for exudative pleural effusion. Serum-pleural fluid albumin gradient was 0.6. Infective works up for cause of chronic fever like tuberculosis, malaria, filaria, leishmaniasis, enteric fever, brucella, scrub, retroviral disease were negative. Collagen vascular disorder work up too was inconclusive. Bone marrow examination was in contradiction to the previous diagnosis of aplastic anemia which showed markedly hypercellular marrow with myeloid hyperplasia, increased megakaryocytes and marked increase in reticulin on trephine biopsy.

Surgical consultation was sought for the abdominal pathology and screening ultrasound abdomen done on day of admission showed an evolving collection in the parietal wall at the epigastric region (Figure 1).

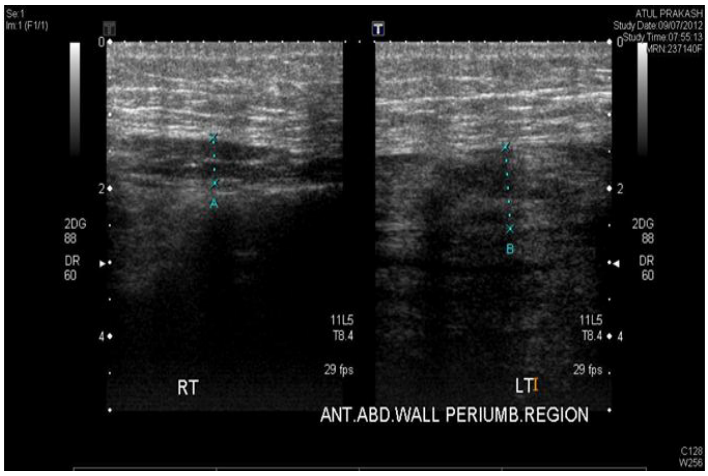

Figure 1: Ultrasound image evolving collection in the parietal wall at the epigastric region

Subsequently a Contrast Enhanced CT abdomen done also showed diffuse subcutaneous edema in the anterior abdominal wall and thickened, hypodense stomach wall. Hepatosplenomegaly with ascites with bilateral pleural effusion was noted (Figure 2).

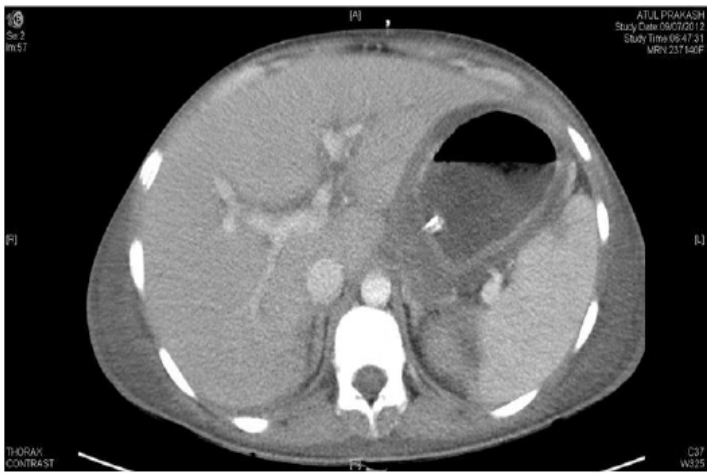

Figure 2: CECT abdomen showed diffuse subcutaneous edema in the anterior abdominal wall and thickened, hypodense stomach wall

Oesopahgo-Gastroscopy though planned, was deferred due to respiratory distress. The next day he underwent anterior abdominal wall abscess drainage of about 50 ml in subcutaneous plane which was tracking between the rectus muscle and posterior rectus sheath under general anaesthesia. Intraoperatively there was no intrabdominal extension and he was electively ventilated post operative in surgical ICU and extubated the next day. Histopapa-

thology specimen was consistent with necrotising fasciitis and the pus culture grew Klebsiella spp. sentitive to Meropenem. Serial blood cultures, pleural fluid culture were negative for any organism growth. Diagnosis of injection associated anterior abdominal abscess was considered and patient was continued on same antibiotics. Inspite of this he continued to be febrile.

Assessment of the abdominal wound on day 4 post op showed purulent discharge with persistent induration surrounding it. Hence he was again posted for redebridement. Before he could be taken up for surgery he had a massive bout of hematemesis with respiratory distress and required emergency intubation. Reevaluation by surgeons showed complete dehiscence of the posterior rectus sheath and visible bowel within the wound. He underwent emergency laparotomy where abscess in the anterior abdominal wall communicating with a perforation in the anterior wall of the stomach was noted.There was full thickness necrosis of the stomach from the GE junction till the first part of duodenum. Extensive adhesions between the liver, spleen, stomach transverse colon and anterior abdominal wall were seen (Figure 3).

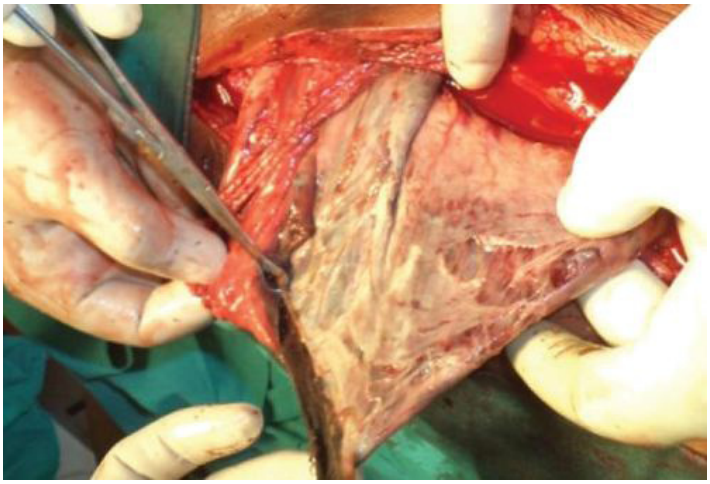

Figure 3: Gross gastrectomy specimen showing full thickness necrosis of stomach

Vascular supply was found to be normal. Total gastrectomy, tube oesophagostomy, lateral duodenostomy and feeding jejunostomy was done by upper GI surgeons. A differential of gastric lymphoma with necrosis was entertained and he was shifted to Surgical ICU for post op care. He required high doses of vasopressor infusion (nor adrenaline, vasopressin) for first 72 hrs due to the sepsis which were later tapered and stopped. He was ventilated on pressure support-synchronised intermittent mandatory ventilation (P-SIMV) with ARDS protocol requiring high Positive End Expiratory Pressures (PEEP) and FiO2 due to the apparent acute respiratory distress syndrome for 48 hrs. Worsening oxygenation even with this led us to try Airway Pressure Release Ventialtion (APRV) for 12 hrs. On this he had a barotraumatic right pneumothorax with extensive subcutaneous emphysema for which urgent intercostal drain with underwater seal was inserted. With no improvement, Prone Position Ventilation (PPV) was done for next 48 hrs which showed acceptable improvement in oxygenation on blood gases. Meanwhile the histopathology specimen showed extensive submucosal, focal mucosal and muscle coat acute necrosis with acute inflammatory response, consistent with severe suppurative infection-Acute Phlegmonus Gastritis. There was no evidence of lymphomatous changes. Pus cultures grew Gram Negative Bacilli-Klebsiella, E. Coli-carbapenemase positive, Vancomycin resistant Enterococci and many yeast like organisms. Injection Colistin and flucanazole were added considering the sensitivity report. Later the antifungal was escalated to liposomal Amphotericin-B due to non remmiting fever. Endotracheal tube aspirate culture grew Non fermenting Gram negative bacteria-Acinetobacter spp. which was panresistant and sensitive to colistin. Hence colistin nebulisation was added to intravenous injections. His pancytopenia had meanwhile resolved and rest other laboratory parameters were stable. Serial blood cultures were negative for any isolates. He was continued on total parenteral nutrition, limb and chest physiotherapy and heparin and sequential compression devices for deep vein thrombosis prophylaxis.

Patient was tracheostomised on day 8 in view of need of prolonged ventilatory support. With decreasing requirement of sedation, neurological assessment on day 4 following discontinuation of prone position ventilation showed left upper limb monoplegia with sensory loss. Suspecting brachial plexus injury due to PPV, Electromyography-Nerve conduction (EMG-NCV) study was done in consultation with Neurology services which showed axillary plexopathy. Physiotherapy and rehabilitative exercises were started by PMR services. Patient was gradually weaned of the ventilator with most antibiotics deescalated by 2nd week and antifungal by 3rd week. Patient was later shifted to surgical High Dependency Unit on day 16 post op with a tracheal mask. His total parenteral nutrition was stopped and started on jejunostomy feeds which he tolerated well. His surgical incision site was healing well and his esophagostomy tube was removed. Later he was shifted to surgical ward where he was taught to take home based jejiunostomy feeds and abdominal wound care and limb physiotherapy exercises. Patient was discharged in health to be followed up with surgical services for salivary diversion and GI reconstructive surgery.

Discussion

Acute gastric necrosis is an extremely rare occurrence in clinical practice [1]. This is explained by the fact that stomach is very resistant to ischemia due to its rich blood supply (right and left gastric, right and left gastroepiploic and short gastric vessels) and its extensive intramural anastomoses. Experimental studies have shown that both arterial and venous circulation must be interrupted before gastric ischemia and necrosis can occur [6-8]. In fact, to be achieved in experimental animals, closure of the right and left gastric and gastroepiploic arteries together with at least 80% of the collaterals is required [9].

Etiologies like Acute Massive Gastric Dilatation (AMGD), vascular causes like Superior Mesentric Artery Syndrome, bulimia, abdominal trauma, infectious disease, gastric volvulus, intrathoracic herniation, necrotizing gastritis, ingestion of caustics leading to massive gastric necrosis have been described in literature [3,4]. Commonest of those is following AMGD defined as intraluminal contents more than 4 litre or intragastric pressures more than 30 cm of H2O. Intragastric pressure exceeding 14 cm of H2O is severe enough to result in intramural venous insufficiency of the stomach with consequent venous ischemia and infarction [8]. Though AMGD may occur in a multitude of medical conditions like anorexia nervosa following binge eating, psychogenic polyphagia and bulimia, diabetes mellitus, trauma, electrolyte disturbances, gastric volvulus, spinal conditions, most of the reported cases are postoperative complications [10]. Rarely AGMD have been described following emergency intubation and chest compression during cardiopulmonary resuscitation and non invasive ventilation. Pathophysiologic theories have been postulated in the past for AMGD like -

a) Upper esophageal sphincter relaxation (due to debilitation or anesthesia) with consequent aerophagia and gastric dilatation

b)Atonic theory (muscular atrophy and atonicity due prolonged starvation does not support rapid refeeding),

c)Superior mesenteric artery syndrome (vascular compression of the third part of duodenum) or

d)Functional disorders due to local inflammation (pancreatitis, cholecystitis, splenectomy, vagotomy). Sequential events postulated are mucosal necrosis, followed by full-thickness involvement of the gastric wall and perforation along greater curvature and fundus [6]. Though our patient had history of anorexia and significant weight loss, there was only thickened hypodense stomach wall without gastric dilatation on CT abdomen done before the first drainage of abscess. There might be a distant possibility of gastric insufflations and dilatation by air during non invasive ventilation, but following the first incision and drainage of abscess stomach was decompressed by dependent nasogastric drainage till he was taken up for total gastrectomy. Hence gastric dilatation as an antecedent cause for gastric necrosis seems an unlikely cause.

Acute phlegmonous gastritis or necrotizing gastritis leading to pangastric necrosis has been infrequently reported. Predisposing factors like immune-compromised state (diabetes, AIDS, hematologic malignancies, neoplasia), chronic gastritis, increased age, alcoholism, hypoacidity, protein-energy malnutrition have been noted [11,12]. Phlegmonous gastritis may arise from a local or disseminated hematogenous infection, and may involve a portion of the stomach (localized type) or the entire stomach (diffuse type). The most frequent causative agents, in order of frequency, are Streptococcus, Staphylococcus, Escherichia coli, Haemophilus influenza, Proteus and Clostridia.

Mixed bacterial infections have also been reported. Even the abdominal esophagus, small intestine and colon can be affected [13,14]. Acute necrotizing gastritis is a variant of phlegmonous gastritis, with organisms producing necrosis and gangrene of the stomach wall rather than just an intramural abscess. Etiologically, Streptococci, fusiform and spirochetal organisms (commonly found in the mouth), or combinations of various organisms have been reported. This can be the possible explanation for the condition in our patient who had immunosupression with hypoproteinemia.

Presentation of gastric necrosis is variable. Following acute necrotising gastritis, severe upper abdominal pain with associated fever, nausea and vomiting is seen. The pain usually increases in severity as the abscess enlarges, does not radiate and is non-colicky in nature. Physical findings include fever, signs of peritoneal irritation and, occasionally, a palpable mass. Diagnosis may be delayed due to the lack of typical signs and this, combined with the rapid progression to peritonitis, often results in a fatal outcome. In case of necrosis following AMGD diffuse abdominal pain may be disproportionately less as compared to distension which may extent to pelvic cavity with worsening of pain following perforation. Emesis is commonly seen but may be absent if there is occlusion of the gastroesophageal junction by the distended fundus angulating the esophagus against the right crus of the diaphragm, producing a one-way valve. On physical examination, a diffuse tympany on palpation, a splash on percussion, and a distended Douglas pouch may be found. Once the stomach perforates, overt peritonitic signs can be elicited. Rapid neurogenic shock (vagal response), hypov-

olemic shock (due to venacaval compression and splanchnic sequestration) and subsequent septic shock may be seen as noted in our patient. Diagnosis of acute necrotizing or gangrenous gastritis is usually made at laparotomy, although endoscopy, endosonography and endoscopic snare biopsy have also been used to arrive at diagnosis. Abdominal CT scan may demonstrate distension, necrosis and pneumo-peritoneum in case of perforation. Nonetheless high index of suspicion is needed for early diagnosis of this condition.

In some cases of dilatation without necrosis and perforation, recovery may occur with emergent nasogastric decompression decreasing the intragastric pressure and reducing the risk of necrosis and perforation thus avoiding surgery [15]. In cases of necrotising pangastritis with or without perforation, urgent total gastrectomy with cervical esophagostomy and feeding jejunostomy or esophago-jejunostomy are the preferred surgical options [3]. At laparotomy, the stomach is usually found to have intact extrinsic blood supply, with thrombosis of the microscopic intrinsic vascular plexus producing the appearance of extensive infarction, dark discoloration and sloughing of the mucosa. Mortality is very high inspite of surgical intervention, ranging from 50 to 80% broad [16] spectrum antibiotic cover guided by organism culture and antibiotic sentivity is desirable due to polymicrobial involvement in majority of cases. Outcomes are worse with sepsis and acute respiratory distress syndrome complicating the postoperative course. This requires a comprehensive critical care and multidisciplinary consultation for a successful outcome. We highlight the significance of same citing the fruitful, though prolonged recovery seen in our patient. Nonetheless intensivists should be aware of this rare and difficult to diagnose condition which would help in early diagnosis and surgical intervention

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