

Gastric Pneumatosis from Isolated Gastric Calciphylaxis

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ABSTRACT

A 58-year-old man with end-stage renal disease presented with hypotension and emesis, pale conjunctivae, and a distended abdomen. Labs revealed hypercalcemia and leukocytosis. Abdominal imaging showed gastric pneumatosis. Endoscopy demonstrated significant hemorrhage and necrosis in the gastric cardia and fundus. Biopsies revealed acute ulcerative gastritis and focal intravascular calcium phosphate crystals. The patient remained nil per os and was placed on omeprazole and sucralfate. Repeat endoscopy demonstrated mucosal healing. Gastric calciphylaxis in the setting of gastric pneumatosis is an uncommon finding, especially in patients without cutaneous findings.

INTRODUCTION

Calciphylaxis, otherwise known as calcific uremic arteriopathy, is a syndrome of inappropriate calcification of small arterioles and subcutaneous capillaries. This phenomenon most commonly occurs in the skin and causes ulceration, necrosis, and often, significant pain. Calciphylaxis is a rare and sometimes fatal complication of end-stage renal disease. Selye first described the pathophysiology in rodents as a hypersensitivity condition.¹ While the definite pathogenesis of calciphylaxis remains unclear, it appears to be an acute hypersensitivity reaction of the capillaries.

CASE REPORT

A 58-year-old man with end-stage renal disease on regular hemodialysis for the past 10 years presented with hypotension and non-bloody, non-bilious emesis. His surgical history was significant for the creation of an arteriovenous fistula without any prior history of abdominal surgeries. He had been compliant with hemodialysis prior to his presentation. There had been no prior discussion regarding kidney transplantation. Upon presentation, he was afebrile but tachycardic with a heart rate 134 beats per minute, blood pressure 84/52 mm Hg, breathing 17 times per minute without requiring supplemental oxygen. Physical examination was notable for pale conjunctivae and a distended abdomen, but was otherwise unremarkable. He had no skin lesions, ulceration, scars, or rashes. Laboratory analysis revealed leukocytosis to 27.7 mg/dL, hemoglobin of 17.4 g/dL, blood urea nitrogen of 37 mg/dL, creatinine of 9.62 mg/dL, calcium of 10.6 mg/dL, phosphorus of 3.0 mg/dL, and albumin of 3.1 g/dL. Initial lactate was 3.4 mmol/L, and initial blood gas was within normal limits. There were no known recent fluctuations in the patient's calcium and phosphorus levels prior to presentation. Prior to admission, he was taking cinacalcet 60 mg twice per day, calcium acetate 667 mg three times per day with meals, and vitamin D2 50,000 units every 30 days. He reported no recent changes in his medication. His chest radiograph was concerning for a left lower lobar consolidation. He was therefore treated with intravenous fluids and empiric antibiotic therapy for sepsis secondary to healthcare-associated pneumonia.

Given his initially high hemoglobin, abdominal computed tomography (CT) ordered to assess for renal masses showed gastric pneumatosis (Figure 1). Shortly after his CT scan, the patient developed sudden-onset epigastric

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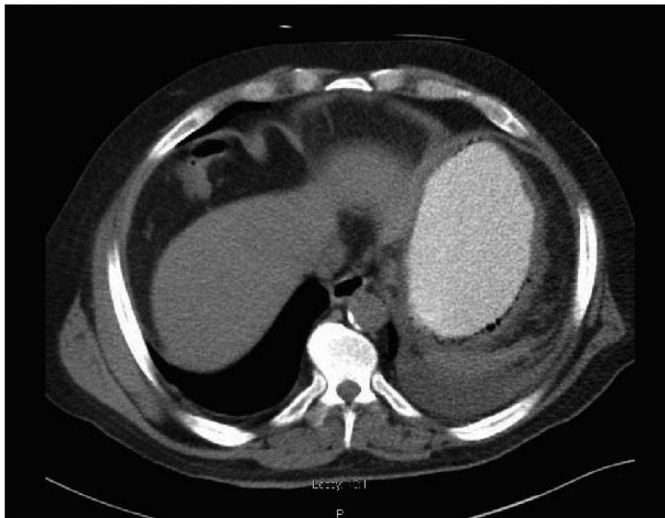


Figure 1. Computed tomography showing gastric pneumatosis.

abdominal pain and coffee-ground emesis. These CT findings, in addition to the patient's clinical presentation, called for an upper endoscopy, which demonstrated significant hemorrhage and ulceration involving a large portion of the gastric cardia, fundus, and body (Figure 2). Multiple biopsies revealed acute ulcerative gastritis and the presence of patchy intravascular calcium phosphate crystals (Figure 3). Findings were consistent with gastric calciphylaxis.

A conservative, non-surgical approach was taken. The patient was made nil per os (NPO) with a nasojejunal (NJ) tube placed to allow for gastric mucosal healing along with the use of a proton pump inhibitor. He was started on broad-spectrum antibiotics, which were ultimately discontinued, as cultures remained negative with no demonstration of ongoing

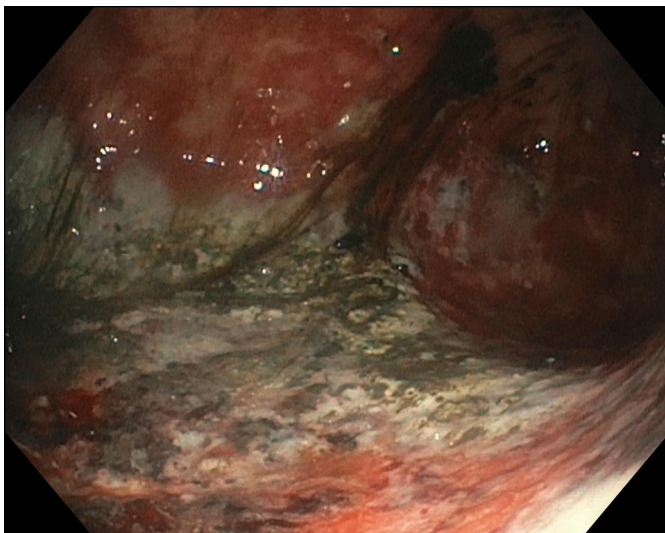


Figure 2. Initial endoscopy with significant gastric hemorrhage and necrosis.

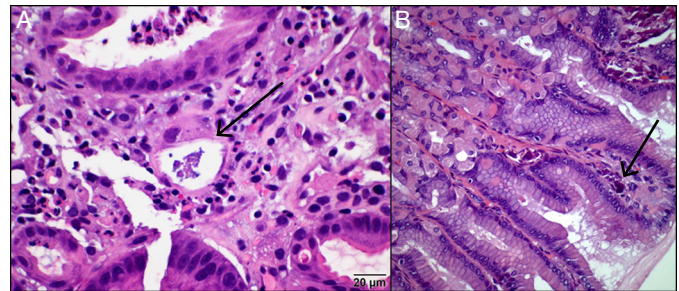


Figure 3. Biopsy pathology showing (A) calcium phosphate deposits within the gastric vasculature (arrow, 600x), and (B) calcium phosphate deposits within the gastric vasculature (arrow, 400x).

infection. Due to multiple spontaneous displacements of his NJ tube, within 10 days of his initial endoscopy he was put on a clear liquid diet. He was converted from calcium acetate to sevelamer (non-calcium-based renal supplements), omeprazole, and sucralfate, and he was maintained on a strict clear liquid diet with nutritional supplements. After these medication changes, the patient's calcium and phosphorus began to normalize. Repeat endoscopies due to nasojejunal tube replacement showed serial improvement of gastric mucosal changes. Hence, sodium thiosulfate was not considered. Repeat endoscopy 4 weeks after initial presentation demonstrated significant ulcer and gastric mucosal healing (Figure 4). At this time, his calcium was 9.7 mg/dL and phosphorus was 4.0 mg/dL. Biopsies taken from this repeat endoscopy showed the absence of calcium deposits from his initial endoscopy (Figure 5).

DISCUSSION

Calciphylaxis is a syndrome of diffuse vascular calcification of the capillaries and skin necrosis, with few cases reported thus far.²⁻⁴ Proposed mechanisms include the metastatic

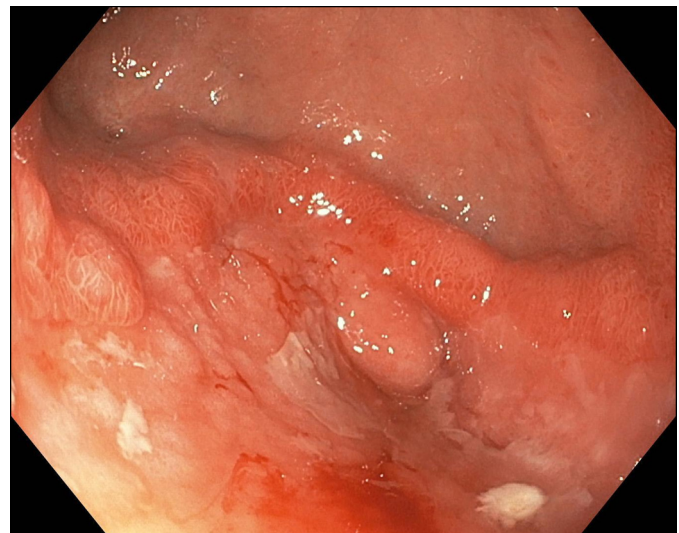


Figure 4. Follow-up endoscopy with significant improvement in gastric mucosal injury approximately 4–6 weeks after initial presentation.

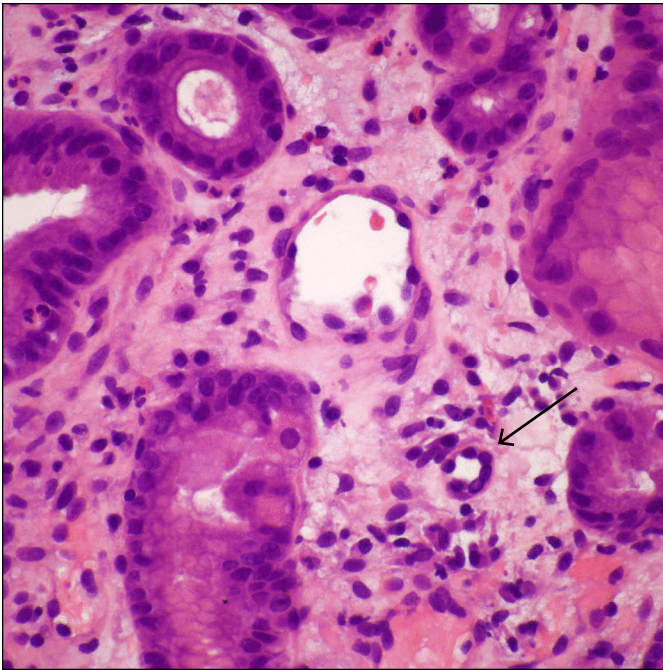


Figure 5. Biopsy pathology from follow-up endoscopy demonstrating gastric antral tissue and a lack of intravascular calcium phosphate deposits with newly formed capillaries (arrow, 600X).

calcification and deposition of calcium salts in an abnormal biochemical environment of calcium phosphate metabolism, with the skin, lungs, and kidneys as the preferential sites of deposition.^{2,5} The formation of these lesions is likely mediated through 2 steps—the medial calcification and intimal fibrosis of arterioles and the thrombotic occlusion after calcification coupled with endothelial dysfunction.¹

Our patient is a unique case of isolated gastric calciphylaxis without any involvement of the peripheral extremities or skin. This phenomenon can occur in the setting of sepsis or disequilibrium, as evidenced in our patient. We propose that calciphylaxis caused capillary wall obstruction, leading to ischemia and pneumatosis of the gastric wall. Isolated gastric calciphylaxis in the setting of gastric pneumatosis is a rare condition. Furthermore, given its rarity, there is little guidance with regard to management. Proposed methods of preventing calciphylaxis include managing comorbid conditions, such as diabetes mellitus, chronic kidney disease, and obesity, as well as decreasing calcium supplements and using non-calcium-based renal supplements.^{6,7} Previous case reports cite the resolution of cutaneous calciphylaxis after kidney transplantation.^{8,9} On

the basis of the existing literature, this unfortunate complication of hemodialysis may have been prevented by a kidney transplant. Our method of treating the underlying healthcare-associated pneumonia while maintaining NPO status with jejunal feeds, switching to non-calcium-based supplements, and stopping synthetic vitamin D supplements and analogs is a viable therapeutic strategy in the management of this rare condition. The patient improved after several weeks of bowel rest, and repeat endoscopy revealed significant improvement.

DISCLOSURES

Author contributions: A. Mulgund wrote the manuscript and is the article guarantor. S. Razeghi and S. Poreddy wrote and edited the manuscript.

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Informed consent was obtained for this case report.

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