中文題目：罕見食道鈣質沉積症引起之食道破裂: 一病例報告
英文題目：A Rare Cause of Esophageal Perforation : Esophageal Mucosal Calcinosis
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Introduction

Esophageal mucosal calcinosis (MC) is a rare phenomenon. In the literature, esophageal MC has been reported in four dialysis patients with presentations of dysphagia or upper gastrointestinal bleeding. However, esophageal perforation secondary to esophageal MC has never been reported.

Case Report

A 44-year-old man presented with acute onset of right chest pain and six months history of solid and liquid dysphagia. He has a history of end stage renal disease on hemodialysis for 8 years, type A aortic dissection, status post aortic valve resuspension and ascending aortic reconstruction 10 years ago, diabetes mellitus, and hypertension. Chest X-ray showed right side hydropneumothorax (Figure 1). Chest tube was placed and food remnants were seen from drainage (Figure 2,3). High amylase level (10568 U/L) was detected in pleural effusion analysis. Esophageal perforation was found by computed tomography (CT) scan (Figure 4) and confirmed with panendoscopy (Figure 5). Subsequent thoracoscopic esophagectomy was performed to decortication and drain loculated empyema (Figure 6). Pathology of the perforated lesion demonstrated no evidence of dysplasia or malignancy. Instead, there was an extensive necrosis, transmural acute and chronic inflammatory cell infiltration and foci of submucosal calcification, consistent with a diagnosis of esophageal mucosal calcinosis (Figure 7,8). Pleural effusion culture yielded enterococcus faecium (VRE) and candida albicans. Antibiotics and antifungals were administered for right side empyema. His clinical condition stabilized and he was successfully weaned from mechanical ventilator. Follow up chest x-ray showed resolution of right side empyema (Figure 9). In this case, prompt treatment with preemptive anti-microbial therapy, timely drainage, and surgical intervention contributed to successful outcome.

Discussion

All of the four previously reported cases of esophageal mucosal calcinosis, including our case, were patients with end stage renal disease. The most important risk factors in these cases were hypercalcemia and hyperphosphatemia, which may lead to metastatic calcification. Furthermore, three patients also had calcific uremic arteriolopathy (CUA) or calciphylaxis. Histologically, our case

did not have evidence of calciphylaxis. If esophageal mucosal calcinosis is associated with calciphylaxis, mortality rate would be higher. Currently, there is no effective therapy to reduce the calcium deposits within the esophageal mucosa.

Conclusion

Gastrointestinal tract mucosal calcinosis could be seen in dialysis-dependent patient, while esophageal involvement is rare. We reported a rare case of esophageal perforation secondary to esophageal mucosal calcinosis. Early diagnosis is difficult by its lower incidence and the absence of typical symptoms. Esophageal perforation is a life-threatening disease, which needs to be diagnosed and treated promptly due to high mortality rate (20–50%). Early determination and appropriate treatment of esophageal perforation are life-saving.

References

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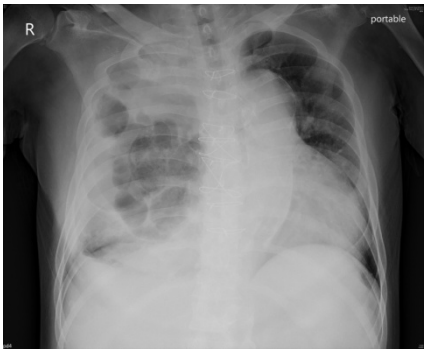


Figure. 1

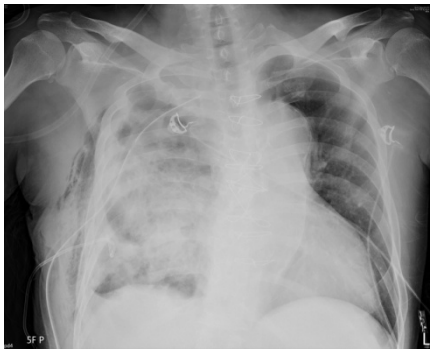
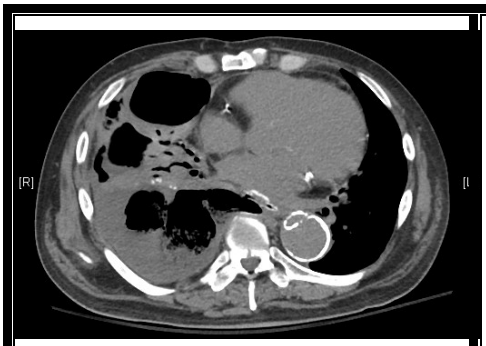


Figure. 2

Figure. 3



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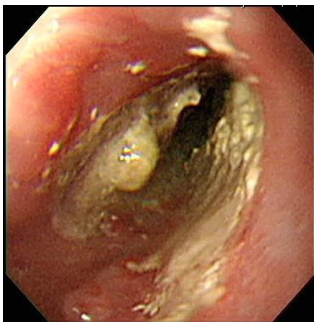


Figure. 5

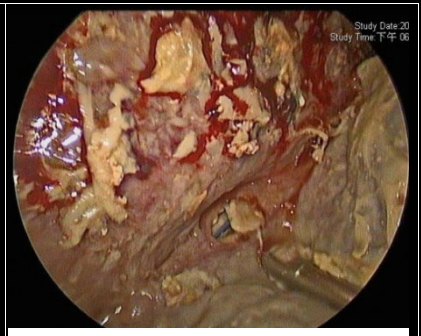
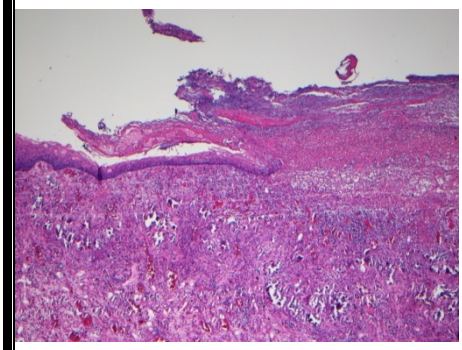


Figure. 6



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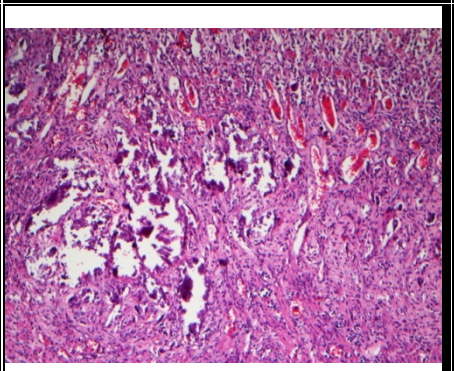

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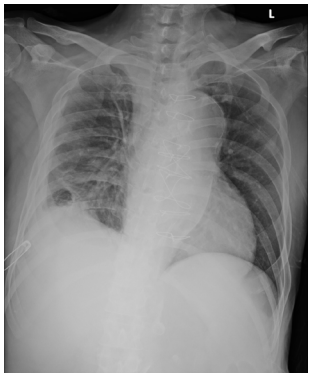

Figure.9