Title

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Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
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Conflict of interest: None declared

Patient: Male, 67-year-old
Final Diagnosis: Infectious endocarditis
Symptoms: Diarrhea • fatigue • oral ulcer
Medication: —
Clinical Procedure: Surgery • antibiotics
Specialty: Infectious Diseases

Objective: Rare disease

Background: Calcified amorphous tumor (CAT) of the heart is a rare non-neoplastic intracardiac mass, which is composed of calcium deposition surrounded by amorphous fibrous tissue. The clinical presentation of cardiac CAT resembles that of other cardiac tumors or vegetation, though there is no previous report of a CAT complicated with infective endocarditis.

Case Report: A 67-year-old male with a history of end stage renal failure and gastric cancer who was on adjuvant chemotherapy presented with a cardiac mass. The mass was resected and diagnosed as CAT pathologically. Two separate sets of blood cultures were positive for *Enterococcus faecalis*, thus, the patient was diagnosed with infective endocarditis. Antibiotic treatment was continued for 6 weeks after surgery, and the patient recovered uneventfully. However, he died from a complication of his gastric cancer 5 months later.

Conclusions: This is the first report of CAT associated with infective endocarditis. Blood cultures should be obtained to differentiate infective endocarditis or CAT with infectious endocarditis from CAT alone, because CAT with infective endocarditis may present atypically and may be more likely to require antibiotic treatment along with surgery.

MeSH Keywords: Cardiac Imaging Techniques • Endocarditis, Bacterial • *Enterococcus faecalis* • Heart Neoplasms

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Background

Calcified amorphous tumor (CAT) of the heart is a rare non-neoplastic intracardiac mass, which is composed of calcium deposition surrounded by amorphous fibrous tissue [1]. The clinical presentation of cardiac CAT resembles that of other cardiac tumors or vegetation. The diagnosis of CAT requires surgical resection and pathological study. Although there are a number of reported cases of CAT, CAT complicated by infective endocarditis (IE) has never been reported.

Case Report

A 67-year-old male with a history of end-stage renal disease (ESRD) who was on hemodialysis due to chronic glomerulonephritis, and who had congestive heart failure and hypertension was admitted to our hospital with fatigue, stomatitis, and diarrhea. Three months prior to admission, he was diagnosed with both sigmoid colon and gastric cancer. He underwent total laparoscopic gastrectomy and sigmoid colon resection. The pathological examination revealed lymph node metastasis from the gastric cancer, and he was started on a reduced dose of tegafur/uracil (UFT) as adjuvant chemotherapy. Two weeks after starting the chemotherapy, he self-discontinued the UFT because of fatigue, loss of appetite, stomatitis, and diarrhea. His performance status worsened over the week, and he was admitted to the hospital. On admission, he was afebrile, his blood pressure was 119/75 mmHg and his heart rate was 92 beats per minute and regular. A systolic ejection murmur was heard at the left fourth intercostal space and a splinter hemorrhage of the nail bed of the left index finger was noted. His metabolic panel revealed a sodium of 134 mEq/L, blood urea nitrogen 28 mg/dL, serum creatinine 8.57 mg/dL, calcium 8.6 mg/dL, phosphorus 4.8 mg/dL, aspartate aminotransferase 18 U/L, alanine aminotransferase 10 U/L, total bilirubin 0.9 mg/dL, and brain natriuretic peptide 2340 pg/dL. His complete blood count (CBC) showed a hemoglobin of 10.6 mg/dL, white blood cell count of 8300/μL, and platelet count of 113 000/μL, C-reactive protein was 2.14 mg/dL, intact parathyroid hormone was 148 pg/mL, parathyroid hormone-related peptide was <1.0 pmol/L, and 25-OH vitamin D3 was 5.9 ng/mL. All other laboratory tests were normal including potassium, bicarbonate, and glucose.

Transthoracic echocardiogram (TTE) revealed a mobile 29×18 mm cystic lesion with a 10×8 mm mass inside the cyst attached to the apex of the left ventricle wall (Figure 1) and a turbulent blood flow around the cyst. A previous echocardiogram performed 5 months before this admission suggested a high echoic lesion of the apical wall. The ejection fraction declined from 47% to 34% over 5 months. Two separate sets of blood cultures were positive for *Enterococcus faecalis* (*E. faecalis*), thus, *E. faecalis* IE was diagnosed, and ampicillin plus gentamycin were initiated. Cardiac computed tomography (CT) demonstrated a calcified apical wall lesion, a low-density mass in the apex and bilateral pleural effusions (Figure 2). Magnetic resonance imaging (MRI) of the brain revealed several small subacute infarctions. Transesophageal echocardiogram (TEE) was performed 3 days after TTE and revealed a single 8×11 mm vegetation of the aortic valve. Additionally, the cystic lesion previously noted on TTE had disappeared, but there were several small lesions ranging in size from 5×10 mm to 9×25 mm as well as punctate lesions in the apex (pictures not available). We believe these smaller lesions represent the disintegration of the cyst containing a mass, causing embolization. Acute decompensated heart failure developed, possibly because of increased volume flow after disintegration of the cyst, however, we cannot be certain of the exact mechanism. Nevertheless, he underwent emergent cardiac surgery to replace the aortic valve with a biologic valve because of the heart failure. Intraoperatively, there was a vegetation on the aortic valve (Figure 3A). There was no mass lesion in the left ventricle, but a small pit in the apex was noted (Figure 3B). The surgeon excised the pit, and pathology revealed a calcified lesion with an abscess inside the myocardial wall (Figure 4A). The surgeon resected the lesion and replaced the aortic valve with a Carpentier Edwards Perimount valve. *E. faecalis* was cultured from both the resected aortic valve and the calcified lesion. Antibiotics were continued for 6 weeks after surgery. The pathology report further revealed lymphocytic infiltration of the myocardium and multiple nodular calcifications in both the endocardium and myocardium surrounded by amorphous fibrous tissue(Figure 4A, 4B). There was also an accumulation of neutrophils in the endocardium with formation of multiple small abscesses. These findings confirmed the diagnosis of calcified amorphous tumor (CAT) complicated by infective endocarditis. The patient recovered uneventfully, and he was scheduled to be discharged to home after completion of treatment. However, he had a sudden decline in his clinical course and died from a complication of his gastric cancer 5 months later.

Discussion

This is the first report of CAT with IE. Cardiac CAT was first reported by Reynolds et al. in 1997 as a non-neoplastic intracardiac mass, which is composed of calcified deposition and amorphous fibrous tissue [1]. The potential roles of phosphocalcific metabolic abnormalities [2] and hypercoagulability [1] are presumed necessary for their formation. In a literature review by de Hemptinne and colleagues, valvular disease (31%) concomitant with mitral annular calcification (MAC) (14%), ESRD (21%), diabetes mellitus (14%), and coronary artery disease (12%) were the most frequently associated conditions. MAC and ESRD were the most common, with approximately

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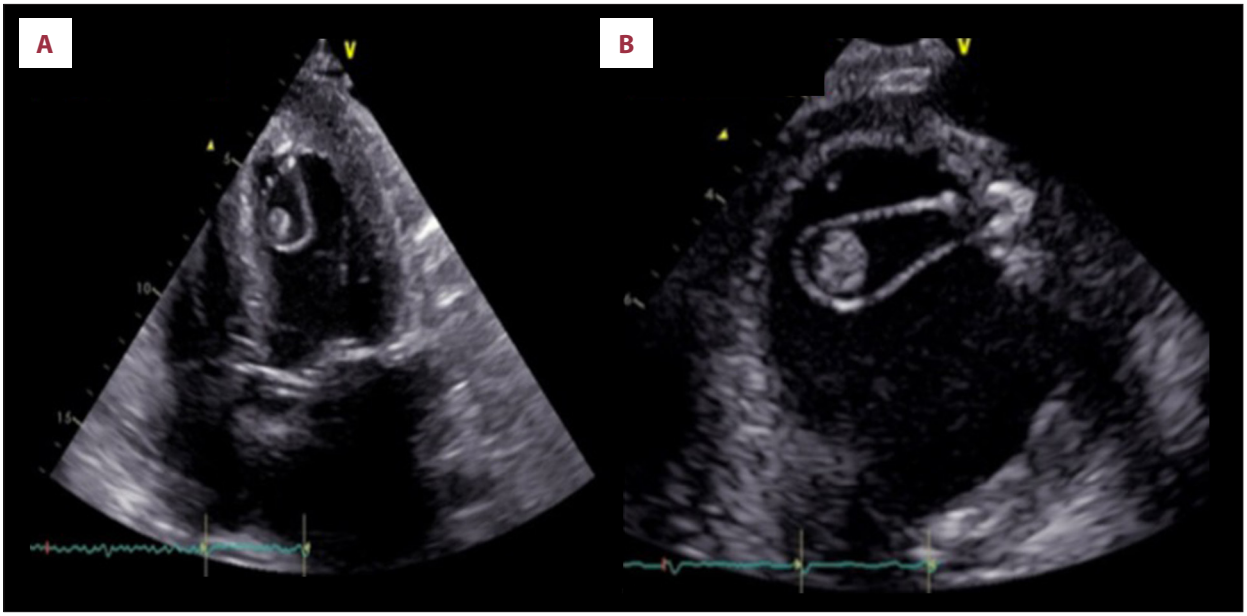

Figure 1. (**A, B**) TTE demonstrating a mobile 29×18 mm cystic lesion with a 10×8 mm mass inside the cyst attached to the apex of the left ventricle wall.

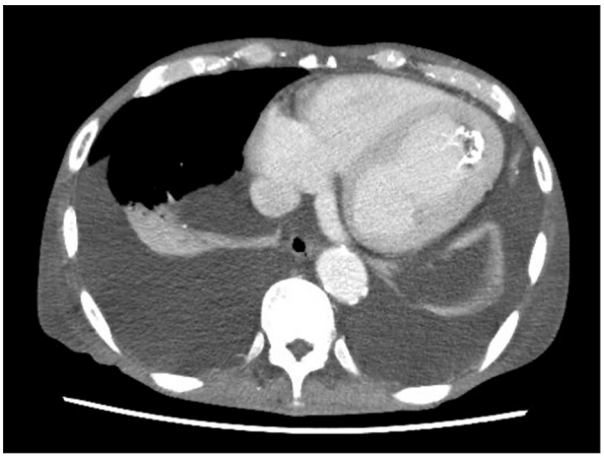

Figure 2. Cardiac computed tomography demonstrating a calcified apical wall lesion, a low-density mass in the apex and bilateral pleural effusions.

20% of patients with CAT had ESRD [3-6]. Some patients with CAT are asymptomatic, but the masses can cause obstructive or embolic symptoms like syncope and dyspnea. Systemic or pulmonary embolic events were reported in 31% of the cases [3]. Mobile lesions, which were described as "swinging CAT" by Kubota et al. [7], indicate a higher embolic risk and rapid growth. The speed of growth of a CAT is unknown, but MAC-related CATs and swinging lesions have been reported to grow rapidly, from 6 weeks to 1 year [2,7]. The mean size of a CAT is reported to be 29×17 mm [3], and it is usually a single lesion. The presence of multiple CAT, similar to this case, is rare (only 2 reported cases) [7,8]. CAT is usually detected on echocardiography. Since CAT appears as calcified intracardiac masses in any heart chambers or valve, it is difficult to differentiate from fibroblastoma, cardiac myxoma, osteoblastoma, vegetations, teratoma, or thrombus [3,6]. Surgical resection with pathological study remains the mainstay of CAT diagnosis and treatment. A recent report by Yılmaz et al. described the radiographical characteristics of CAT as large foci in a partially calcified mass or diffuse calcification of a mass on CT, and low signal intensity on T1- and T2-weighted images with no contrast enhancement on MRI [10]. Vegetation with infective endocarditis could be an important differential diagnosis of CAT, as this report suggests. In our review of the literature concerning cardiac CAT using PubMed, there were 53 full-text English language case reports of 68 cases [1–9,11–54]. Thirty-four reports (48 cases) of cardiac CAT considered the vegetation of IE in their differential diagnosis, though only 11 reports (11 cases) mentioned negative results of blood or valvular culture. Because inflammatory markers were negative in some reports, the authors concluded that there was no evidence of IE. In this case, our patient did not present with typical signs of IE, such as fever. Thus, blood cultures were first obtained after TTE revealed the intracardiac cystic lesion. Of importance, blood cultures should be obtained to differentiate IE or CAT with IE from CAT alone, because CAT with IE may present atypically and may be more likely to require antibiotic treatment along with surgery. Given that the organism was enterococcus, and that the patient had gastric cancer, there is a possibility that he had subacute IE first, which led to a systemic inflammatory response resulting in a complication of CAT. It remains uncertain if the patient had CAT first and then IE, or whether the vegetations of subacute IE became a complication of CAT. Further investigation is needed concerning CAT

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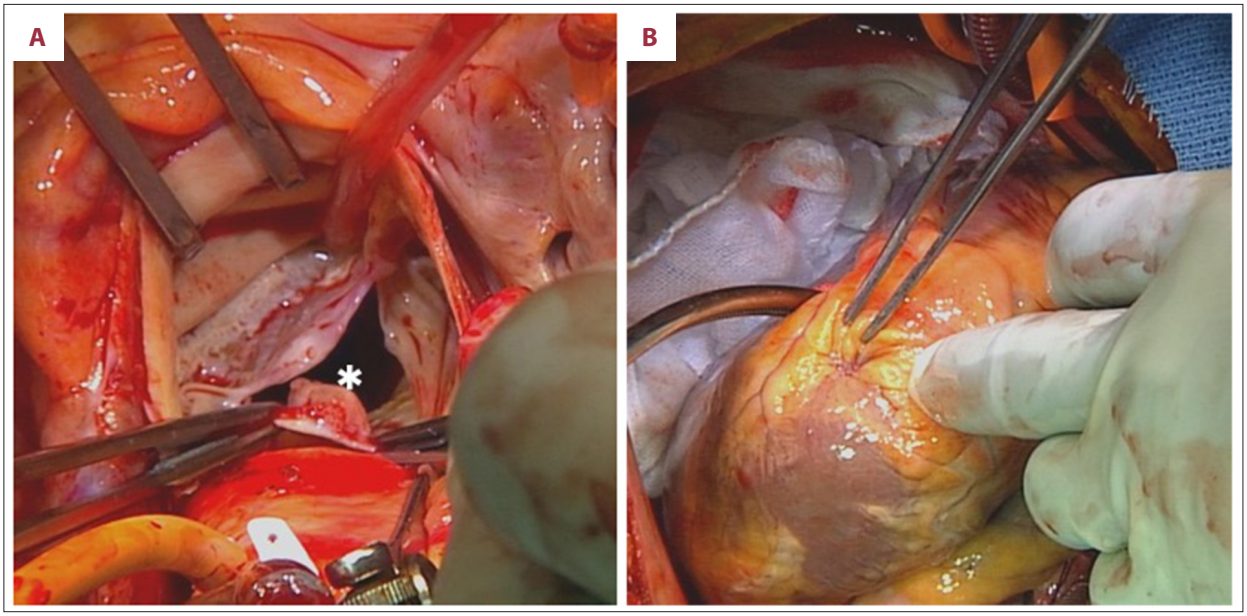

Figure 3. (**A, B**) Surgical photographs. (**A**) The asterisk shows a vegetation on the aortic valve; (**B**) A small pit in the apex where there had been a mass legion.

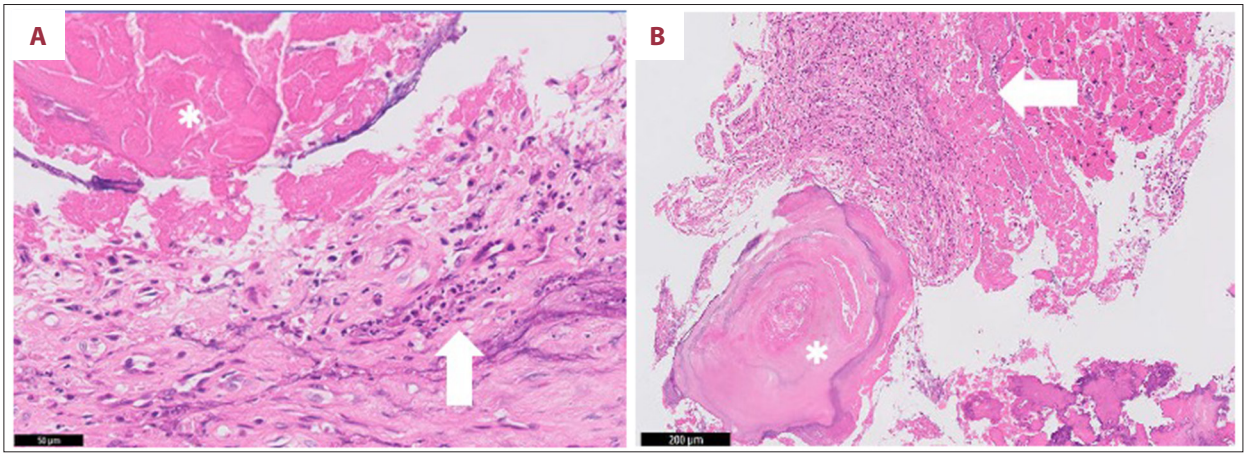

Figure 4. (**A, B**) Pathological examination of the excised lesion in the apex. The asterisk of (**A**) and (**B**) shows multiple nodular calcifications in both the endocardium and myocardium surrounded by amorphous fibrous tissue. (**A**) The arrow shows accumulation of neutrophils in the endocardium with formation of multiple small abscesses. (**B**) The arrow shows lymphocytic infiltration of the myocardium.

as a nidus of IE, as well as the contribution of systemic infection in the development of cardiac CAT.

Conclusions

This is the first report of cardiac amorphous tumor associated with IE. CAT had been thought to be a sterile non-neoplastic mass, but this case suggests that CAT can also be complicated by IE. Blood culture should be obtained to differentiate CAT from IE or CAT with IE.

Department and Institution where work was done

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Conflicts of interest

None.

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