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Case Report

Infective calcified amorphous tumor on mitral valve and critical course of left ventricular rupture

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ABSTRACT

Calcified amorphous tumor is a rare intracavitary cardiac lesion and an accompanying infection is extremely rare. A 76-year-old woman was transferred to our hospital because of cerebral infarction. Echocardiography and chest computed tomography showed a calcified large mobile mass on the posterior mitral valve that was diagnosed with a calcified amorphous tumor. Moderate aortic regurgitation and severe mitral regurgitation were also confirmed. Her blood culture detected *Gamella* sp. We surgically dissected this infective calcified amorphous tumor. The border between this infective tumor and the mitral annulus was unclear because of severe infection and necrotic tissue. After careful complete resection, the healthy ventricular muscle was exposed and we performed annular reconstruction with bovine pericardial patches. And we replaced the aortic and mitral valves using bioprosthesis. While weaning from cardiopulmonary bypass, however, left ventricular rupture occurred twice. Despite successful repair of left ventricular rupture, which controlled bleeding, she died from multi-organ failure on postoperative day 6. An infective calcified amorphous tumor in such a critical case has not been reported previously. The calcified amorphous tumor probably become serious when the infection occurred. In this situation, the utmost caution should be paid to the patient.

<**Learning objective:** Calcified amorphous tumor (CAT) is a rare non-neoplastic intracavitary cardiac lesion. There have been some reports of CATs but they are extremely rare with accompanying infection or critical situations. Our patient was a 76-year-old female with infective CAT who suffered from cerebral infarction, and she died from multi-organ failure despite best surgical treatment. CAT probably become serious when the infection occurred. In this situation, the utmost caution should be paid to the patient.>

Introduction

Calcified amorphous tumor (CAT) is a rare non-neoplastic intracavitary cardiac lesion characterized by nodular calcium on the background of amorphous degenerating fibrinous material [1]. There have been some reports of CATs [2] but they are extremely rare with accompanying infection [3] or critical situation. Here we report the rare and severe case of infective CAT on the mitral valve (MV).

Case report

A 76-year-old woman was transferred to our hospital because of left hemiplegia and dysarthria due to right middle cerebral artery infarction revealed by head computed tomography (CT) and magnetic resonance imaging. Her neurological symptoms were alleviated by endovascular thrombectomy. Her past medical history included hypertension, dyslipidemia, and osteoporosis. But she had no medical history of hemodialysis. She was febrile (body temperature 37.4 °C). Her laboratory data revealed the presence of a severe inflammatory reaction (white blood cell count, 18,700/mm^3; C-reactive protein, 6.88 mg/dL) and coagulopathy (platelet count, 47,000/mm^3; prothrombin time, 80%; activated partial thromboplastin time, 68.8 second). Her renal function was normal (serum

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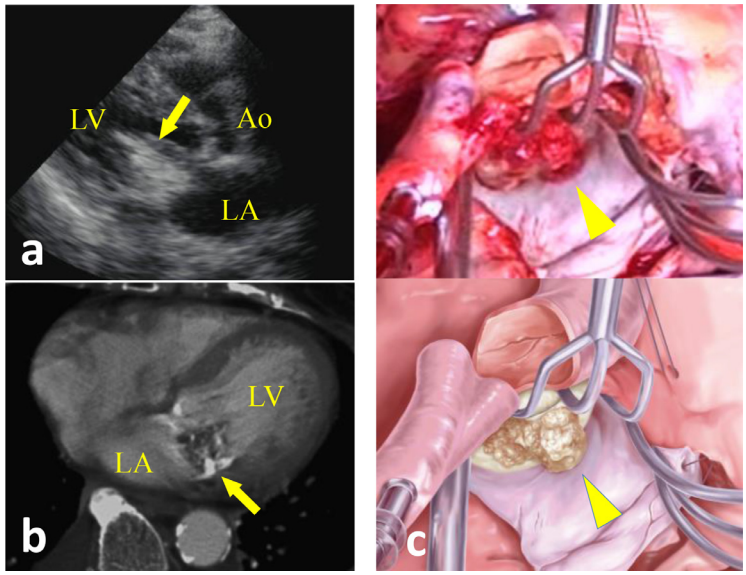

Fig. 1. Preoperative transthoracic echocardiography and contrast enhanced chest computed tomography, and intraoperative findings of posterior mitral leaflet formed with a calcified tumor. (a) Echocardiography, long-axis view, shows a calcified tumor with a mosaic echoic lesion (arrow) (37 × 24 mm) at the annulus of the posterior mitral leaflet. (b) Chest computed tomography shows the calcified tumor (arrow) at the posterior mitral annulus. (c) Posterior mitral leaflet formed a calcified tumor (arrowhead) with vegetation. Ao, aorta; LA, left atrium; LV, left ventricle.

creatinine 0.51 mg/dL). Blood culture detected *Gamella* sp. Echocardiography showed a large calcified mass with a mosaic echoic lesion (37 × 24 mm) on the posterior MV (Fig. 1a). Moderate aortic and severe mitral regurgitation were confirmed. Chest CT showed a large calcified tumor on the posterior MV (Fig. 1b). The symptoms and preoperative cardiac imaging were characteristic of CAT, so the tumor was strongly suspected to be CAT. We decided that the infective CAT on the MV and the impaired aortic valve (AV) should be addressed surgically. We scheduled AV and MV replacement 6 days after admission, at which time head CT confirmed that there was no hemorrhage.

Following median full sternotomy, cardiopulmonary bypass (CPB) was routinely established by placing an atrial cannula in the ascending aorta and direct bi-caval cannulation, and we approached the MV via a superior transseptal incision. The posterior mitral leaflet formed a large calcified tumor, which was consistent with the findings of a CAT (Fig. 1c). The border between this infective CAT and the mitral annulus (MA) was unclear because of severe infection and necrotic tissue. We resected the infective CAT, crushing the calcified tissue. After the careful complete resection, the healthy left ventricular (LV) muscle was exposed. We performed mitral annular reconstruction using bovine pericardial patches on the exposed LV muscle to avoid LV rupture. We also performed MV replacement using a 23 mm Carpentier–Edwards Perimount (CEP) Magna Mitral Ease (MAGNA) (Edwards Lifesciences, Irvine, CA, USA) using the half-and-half technique [4] everting mattress sutures at the posterior MA and non-everting mattress sutures in the anterior MA (Fig. 2a). The AV was tricuspid. Vegetation was present on the LV side of the left coronary cusp, without an aortic annulus abscess. AV replacement was then performed using a 19-mm CEP MAGNA aortic bioprosthesis. After cross-clamp release, type III LV rupture of the Treasure's classification occurred during CPB weaning with ejection of the stent post of the LV prosthetic valve (Fig. 2b). The LV rupture site was the outside area of the mitral annular reconstruction with the bovine pericardial patches. Under cardioplegic arrest, we sandwiched the rupture site between the outside and inside of the LV using bovine pericardial patches by interrupted mattress sutures, filling the area between the patches with BioGlue® (CryoLIfe, Inc., Kennesaw, GA, USA) (Fig. 2c). After cross-clamp re-release and during CPB weaning, a second LV rupture occurred at the cranial part of the repaired site with ejection of the stent post of the LV prosthetic valve (Fig. 2d). We repaired the LV rupture site using the same method with the large bovine pericardial patches and well controlled any bleeding (Fig. 2e).

Massive alveolar hemorrhage occurred 3 days after the surgery and she died from multi-organ failure on postoperative day 6. Histological examination of this crushed tissue revealed nodular calcified necrotic tissues and many inflammatory cells and Gram-positive cocci (Fig. 3).

Discussion

Reynolds and colleagues first reported a CAT in 1997 as a non-neoplastic cardiac mass composed of calcified nodules on the background of amorphous fibrous material [1]. Patients with a CAT often suffer from dyspnea followed by syncope or pulmonary or systemic embolization [2]. Some features of clinical characteristics of the patients and current cardiac imaging techniques may contribute to establishing the diagnosis [2]. On echocardiography, CAT appears as a mosaic echoic lesion with a calcified mass and irregularly shaped edges. It may be found in any chamber, on any valve, or on an annulus [2,5]. In our case, the symptoms, preoperative characteristic cardiac imaging, and operative findings led to the diagnosis of CAT. It is true that infected mitral annular calcification (MAC) might have similar findings to our case, but our patient had no findings of MAC on transthoracic echocardiography, transesophageal echocardiography, CT-scan, or intraoperative findings. When we debrided the CAT and MA, we also could not find MAC.

Many cases regarding CAT on MV have been reported with good postoperative course [1,2]. However, there were no cases with infection or in such critical situations as in our case. CAT probably becomes serious when bacterial infection occurs.

After this case, we engaged in some discussions and suggestions regarding the strategies for treating CAT with associated infection.

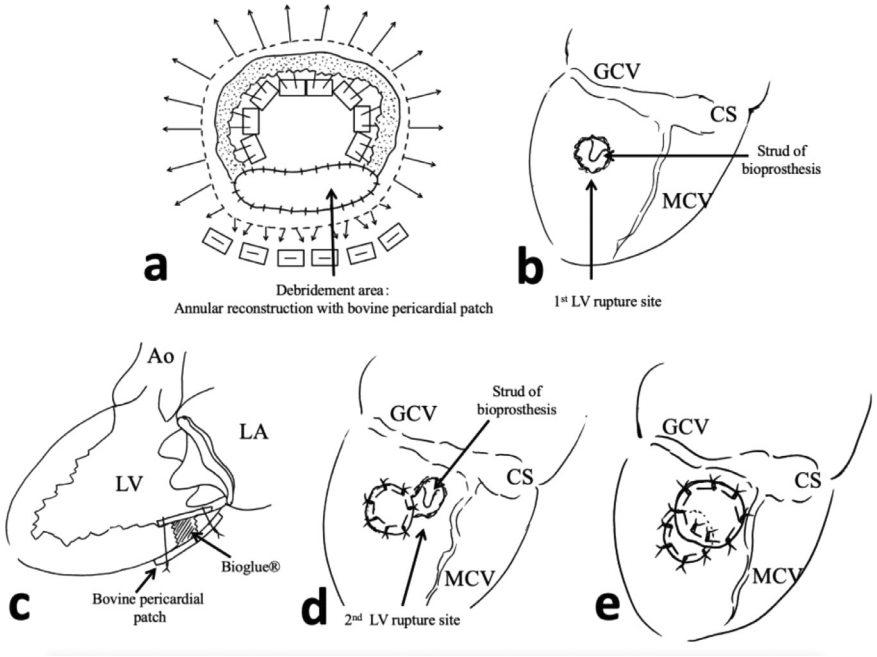

Fig. 2. Operative procedure of the range of debridement, the mitral valve replacement, and the repair of the LV rupture. (a) After the complete debridement of the CAT, mitral annular reconstruction was performed using bovine pericardial patch on the exposed LV muscle. Mitral valve replacement was performed by the half-and-half technique, everting mattress sutures at the posterior mitral annulus and non-everting mattress sutures in the anterior mitral annulus. (b) The first type III LV rupture occurred with ejection of the stent post of the prosthetic valve. (c) The rupture site was sandwiched using bovine pericardial patches by interrupted mattress sutures, filling with BioGlue® (CryoLIfe, Inc., Kennesaw, GA, USA). (d) The second LV rupture occurred at the cranial part of the repaired site with ejection of the stent post. (e) We repaired the LV rupture using the same method with the large bovine pericardial patches. CAT, calcified amorphous tumor; LV, left ventricular; CPB, cardiopulmonary bypass; GCV, great cardiac vein; MCV, middle cardiac vein; CS, coronary sinus; Ao, aorta; LA, left atrium. .

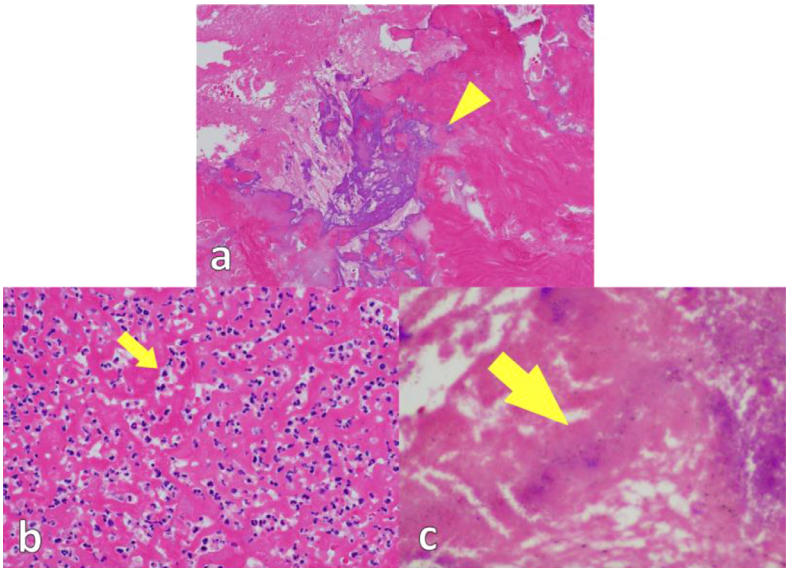

Fig. 3. Pathological examination of the infected calcified tumor. (a) Histopathological examination revealed multiple nodular calcification (arrow head) and fibrous connective tissue. (b) The small arrow showed accumulation of neutrophils in the tissue. (c) The abscess was formed by Gram-positive cocci (large arrow).

The complete resection of the CAT had been needed because recurrence had been reported [6], which we had undertaken, performing radical resection of the infected CAT and debridement even though there was a risk of LV rupture. We had covered the posterior MA and exposed LV muscle using a bovine pericardial patch that was slightly larger than usual to protect the LV muscle from the stent post on the bioprosthesis. This LV rupture site was the outside area of the mitral annular reconstruction with the bovine pericardial patches. The size of bovine pericardial patch might not be enough to protect the LV muscle from the stent post. Thus, our discussion led us to agree that other bioprostheses, with lower stent posts, should be used to avoid LV rupture [7]. In fact, using mechanical valves with no stent post might be a good choice if the patient could tolerate anticoagulant therapy postoperatively. Me-

chanical valves could not be used in our patient because she was at risk of cerebral bleeding after cerebral infarction, thereby contraindicating the use of a postoperative anticoagulant. We therefore concluded that, if the patient has an infective CAT and does not suffer from central nervous system complications, it may be necessary to completely resect the infected CAT, place a large bovine pericardial patch enough to protect the LV muscle from the stent post of prosthetic valve on the LV endocardium, and insert a prosthesis with a low stent post, or if needed, mechanical valve.

Conclusion

Despite numerous case reports and reviews of CAT, a CAT associated with infection or in critical situations has been rarely reported. CAT probably became serious when the infection occurred. In this situation, the utmost caution should be paid to the patient. Although we could not save this patient, this rare case report of a CAT with infection should be of great interest to many medical staff and should be discussed carefully to save future patients with an infected CAT.

Declaration of Competing Interest

The authors declare that there is no conflict of interest.

CRediT authorship contribution statement

Kazuma Handa: Writing - review & editing. **Shinya Fukui:** Writing - review & editing. **Yukitoshi Shirakawa:** Writing - review & editing. **Tomohiko Sakamoto:** Writing - review & editing. **Mutsunori Kitahara:** Writing - review & editing. **Yumi Kakizawa:** Writing - review & editing. **Hiroyuki Nishi:** Writing - review & editing.

Availability of data and materials

The authors declare that all data in this article are available within this published article.

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Ethics approval and consent to participate

Informed consent was obtained from the patient and her family.

Consent to publication

Written informed consent for publication of this case report was obtained from the patient.

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References

[1] Reynolds C, Tazelaar HD, Edwards WD. Calcified amorphous tumor of the heart (cardiac CAT). Hum Pathol 1997;28:601–6.
[2] de Hemptinne Q, de Cannière D, Vandenbossche JL, Unger P. Cardiac calcified amorphous tumor: a systematic review of the literature. Int J Cardiol Heart Vasc 2015;7:1–5.
[3] Okazaki A, Oyama Y, Hosokawa N, Ban H, Miyaji Y, Moody S. The first report of calcified amorphous tumor associated with infective endocarditis: a case report and review of literature. Am J Case Rep 2020;21:e922960.
[4] Bito Y, Shibata T, Yasuoka T, Inoue K, Ikuta T. Mitral valve replacement for extensive calcification: half and half technique. Gen Thorac Cardiovasc Surg 2008;56:526–8.
[5] Kimura N, Haruta H, Tamaki T, Migita S, Saito Y, Aizawa Y, Kato M, Hirayama A. A case of calcified amorphous tumor found with cerebral infarction. Shinzo 2017;49:502–7.
[6] Fealey ME, Edwards WD, Reynolds CA, Pellikka PA, Dearani JA. Recurrent cardiac calcific amorphous tumor: the CAT had a kitten. Cardiovasc Pathol 2007;16:115–18.
[7] Sersar SI, Jamjoom AA. Left ventricular rupture post mitral valve replacement. Clin Med Cardiol 2009;3:101–13.