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## CASE REPORT

# A Waving Horn on the Big Mitral Annulus Calcification: Caseous Calcification of the Mitral Annulus with Abscess Formation



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## KEY WORDS

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Caseous calcification of the mitral annulus (CCMA) is a rare variant of mitral annular calcification. It comprises a combination of calcium, fatty acids, and cholesterol, and is characterized by heterogeneity in echocardiographic images, with peripheral areas of calcification surrounding a central area of echolucency, resembling a periannular mass. Here, we describe a case of CCMA combined with a mitral annulus abscess, manifesting as a waving, horn-like structure. Although the image characteristics of the posterior mitral annulus suggested CCMA, additional findings warranted further work-up and studies.

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## Introduction

Mitral annular calcification (MAC) is a common echocardiographic finding, notably in elderly and hemodialysis patients [1]. A rare variant, caseous calcification of the mitral annulus (CCMA), appears as a round, echo-dense mass with central echolucency on transthoracic echocardiography (TTE) images. CCMA can be misinterpreted as an intracardiac tumor, an abscess, or a thrombus [2]. With the lack of a general consensus on the optimal management, a conservative strategy is usually adopted. However, surgical

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intervention should be considered in symptomatic patients or when the diagnosis is uncertain [3]. Here, we present a case of CCMA complicated with ischemic stroke and additional TTE findings, which were found to be abscess formation in postoperative culture results.

## Case report

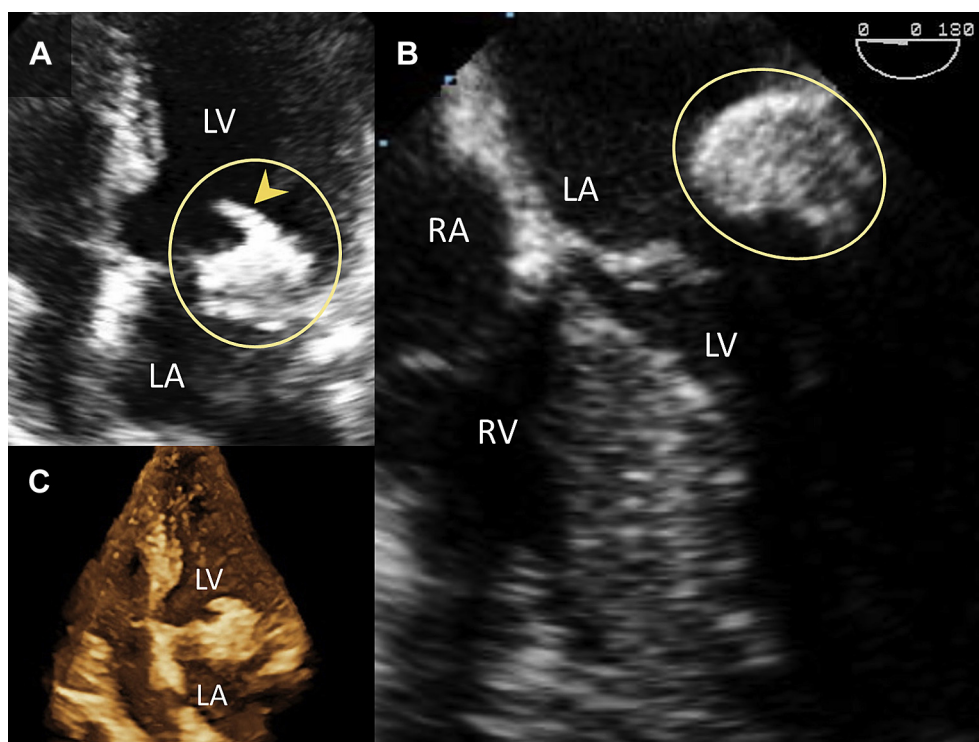
A 68-year-old woman with a history of diabetes mellitus, hypertension, and end-stage renal disease under regular hemodialysis was referred to our hospital for a suspected cardiac tumor that was found during admission to a suburban hospital for a left-hemispheric ischemic stroke. She denied fever, chills, or other constitutional symptoms. A physical examination and laboratory data were unremarkable except for mild anemia (hemoglobin, 10.6 g/dL). Electrocardiography showed sinus rhythm with left ventricular hypertrophy, and chest X-ray revealed cardiomegaly. In addition to adequate global left ventricular systolic performance and MAC, TTE demonstrated a semi-lunar, large, fixed echo-dense mass (1.6 cm × 2.3 cm), which occupied the posterior mitral annulus and extended to the base of the posterior mitral leaflet (PML) (Figs. 1A and 2A, circles and Fig. 2C, arrows). The mass was characterized by a calcified outer shell with inner echolucency. Without disrupting the mitral leaflet, the mass resulted in mild limitation of the motion of the PML leading to mild anterior eccentric mitral regurgitation (Fig. 2B). On top of the mass, a soft, mobile, horn-like structure was noted, swinging rhythmically with each heartbeat, resembling a waving gesture (Fig. 1A, arrowhead). The three-

dimensional TTE image also clearly showed this mass (Fig. 1C). The heterogeneous mass, which had a smooth border with central echolucency simulating liquefaction and with no acoustic shadowing, was further clearly delineated on the transesophageal echocardiography image (Fig. 1C, circle). She underwent surgical treatment owing to an uncertain diagnosis. Partial rupture of the chordae tendineae of the PML (P1 and P2) was noted intraoperatively. One bulging lesion was palpated at the mitral annulus (near P2), and whitish–yellowish mucus content of the lesion was drained after incision. Debridement was performed, and no cardiac tumors were found. The surgical specimen showed calcified materials, fibrins, and deposition with inflammation (Fig. 3). The culture yielded *Escherichia coli*. Antibiotics were then given for 6 weeks with negative results in serial blood culture examinations. The patient was discharged smoothly after the operation.

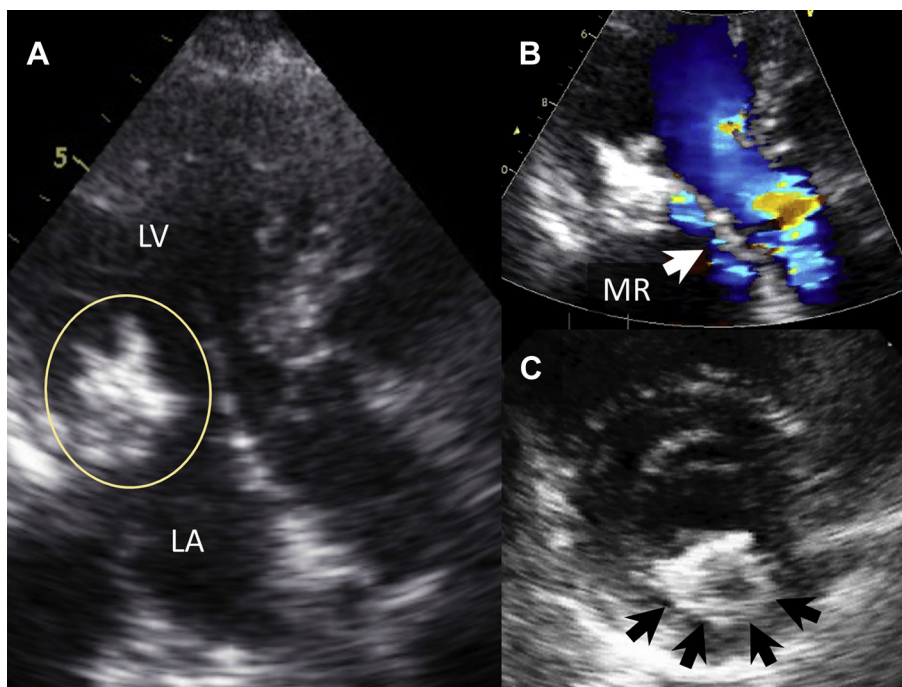
Based on the echocardiographic, operative, and culture findings, the diagnosis of CCMA combined with a mitral annulus abscess was made, which may have been attributable to a prior episode of subacute infective endocarditis, leading to destruction of the chordae tendineae.

## Discussion

CCMA, or liquefaction necrosis, is regarded as a rare variant of the more common MAC. This disease entity has been overlooked until recently, and hence the exact incidence may be underestimated [4]. Two studies reported that the prevalence is 0.06–0.07% of the population [5,6]. The most common clinical presentation is incidental findings during



**Fig. 1** Caseous calcification of the mitral annulus (A, B, circles) with an additional horn-like structure (A, arrowhead) is shown in both transthoracic and transesophageal echocardiography, and is demonstrated by three-dimensional transthoracic echocardiographic images (C). LA = left atrium; LV = left ventricle; RA = right atrium; RV = right ventricle.



**Fig. 2** Transthoracic echocardiography shows caseous calcification of the mitral annulus via both the parasternal long axis view (A, circle) and short axis view (C, arrows). Mild anterior eccentric mitral regurgitation is also noted (B, arrow). LA = left atrium; LV = left ventricle; MR = mitral regurgitation.

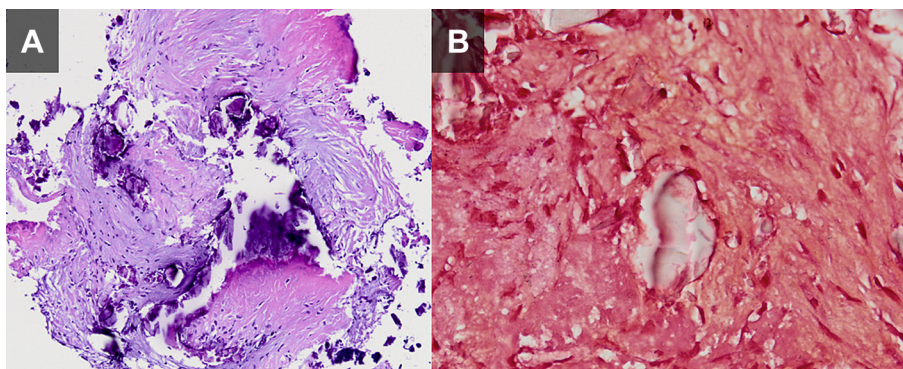
cardiac imaging, and this underscores the importance of cardiologists being familiar with this diagnosis. Failure to identify CCMA may lead to unnecessary work-up or invasive treatments because it is often misinterpreted as a calcific periannular tumor or abscess [7,8].

Echocardiography provides valuable information for the diagnosis. CCMA appears as a round or semilunar mass with central echolucency encompassed by a calcified envelope without acoustic shadowing. It is usually located in the posterior region on the left atrioventricular groove, bulging inwardly. By contrast, MAC winds in a C-shaped pattern and can create an acoustic shadow. Grossly, the interior material of CCMA is toothpaste-like, milky, and caseous [6].

Although it is believed to be benign intrinsically, CCMA may be complicated by systemic embolization.

Nevertheless, a conservative management strategy is suggested initially. Surgical indications include malfunction of the mitral valve, embolic events, or an uncertain diagnosis [3]. Our case had prior ischemic stroke, and the waving, horn-like structure made the disease *per se* more sophisticated. Therefore, surgical intervention was performed. Interestingly, the culture from the tissue sample dissected intraoperatively showed *E. coli*, which implied a possible prior infective endocarditis event and could also potentially explain the underlying mechanism. We speculate that the presence of CCMA may create an environment prone to bacterial infection, thereby leading to subsequent abscess formation.

In conclusion, CCMA can be misinterpreted as an intracardiac tumor, vegetation or abscess in echocardiography.



**Fig. 3** (A) The surgical specimen shows dense calcification, inflammatory cell infiltration, granulation tissue formation, and focal necrosis (hematoxylin–eosin stain, magnification 100 $\times$ ). (B) No specific bacteria could be identified by the special stain (Gram stain, magnification 400 $\times$ ).

Being familiar with the imaging characteristics helps to make a correct diagnosis and avoid unnecessary treatment. Additional findings or symptoms should prompt further work-up and studies on a case-by-case basis.

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