Case report

Spontaneous fistulization of a caseous calcification of the mitral annulus: an exceptional cause of stroke

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Abstract

We present the case of a caseous calcification of the mitral annulus, responsible for two strokes, in a 72-year-old female patient. The brain computed tomography (CT) scan confirmed the presence of a calcific embolus. The echocardiography showed a liquidy, pseudotumoral mass combined with numerous calcifications located in the posterior part of the mitral annulus and extending toward the inferior surface of the left ventricle. During surgery, we found a direct communication between the caseous necrosis and the lumen of the left ventricle at the level of its inferior wall. We performed a valve repair procedure and excision of the caseous necrosis, combined with injection of bioglue into the cavity, to avoid recurrence. Six months after the procedure, the patient was in good health, and had no recurrence of stroke with a satisfactory echocardiography. This is the first description of spontaneous fistulization of a caseous necrosis in the lumen of the left ventricle, explaining a new mechanism for cerebral embolism during the course of calcifying diseases of the mitral annulus.

Keywords: Stroke; Intracardiac masses; Mitral annulus; Calcification; Echocardiography

1. Introduction

Caseous calcifications of the mitral annulus (CCMA) are rare and the prevalence is approximately 0.067% in the general population [1]. Generally asymptomatic, they can, however, be responsible for hemodynamic and embolic complications [2]. We would like to report the case of a female patient having presented two strokes, where the cause was finally attributed to CCMA.

2. Case report

A female patient aged 72 years, was admitted for two strokes. A brain computed tomography (CT) scan provided images of multiple calcific emboli. Transthoracic followed by transesophageal echocardiography revealed a pseudotumor 34 mm × 16 mm, resembling a cyst, calcified in the left atrial wall and developed on the posterior mitral annulus with lateral extension toward the left ventricle (Fig. 1(A) and (B)). Mobile images were found in this tumor. No mitral insufficiency or other cardiac anomalies were found. A cardiac CT scan showed calcifications on the posterior part of the mitral annulus, with an extension toward the inferior wall of the left ventricle (Fig. 1(C)). A diagnosis of CCMA was established. During the surgery, the posterior mitral valve was clean without calcification protuberance. After disinsertion of the posterior valve, the calcifications as well as a white milky liquid were visible. An extension of the detachment of approximately 5 cm toward the inferior wall of the left ventricle confirmed a small direct communication into the lumen of the left ventricle. Excision with removal of calcifications from the entire posterior mitral annulus [3] and its extension toward the left ventricle was carried out (Fig. 1(D)). The communication was too small to be close. After an abundant lavage, the detached area was filled with an injection of bioglue®. The detachment of the posterior valve was directly closed without any biological or synthetic patch. To avoid any destabilization of the mitral annulus, an external annuloplasty was performed (ring CE® Physio Ring II 26 mm). Peroperative monitoring by transesophageal echography showed good results for the surgical procedure, with disappearance of the pseudotumoral image. Histological analysis of the removed specimen found calcified lesions with fibroadipose tissue. A substantial inflammatory reaction was found with neutrophils, histiocytes, plasmocytes and some lymphocytes (Fig. 1(E)). Grocott and Giemsa staining did not
Among the complications described, clinical manifestations [1,2]. Cases of retransformation of treatment seems to be a good option in the absence of symptomatic cases only? According to some authors, absence preventively then arises, or should surgery be limited to CCMA. The question of operating asymptomatic patients has been reached with regard to the therapeutic management of (stroke) events can be noted. Consensus is far from having been reached with regard to the therapeutic management of CCMA. The formation of a thrombus and its embolization has been described [9]. Another hypothesis might be that of the migration of a calcification from the mitral annulus [10]. In this respect, our case offers an additional hypothesis as to etiology: the embolic mechanism might be caused by a spontaneous fistulization of the caseous cavity in the lumen of the left ventricle. The valvular tissue was clean without calcification protuberance. The only possibility to explain the embolization was the spontaneous fistulization, as confirmed by the direct communication between the mitral annulus and the left-ventricle lumen. Embolization is, in fact, possibly a rare mechanism and fistulization is probably more frequent as imagined.

In conclusion, we demonstrated, for the first time, a new possible mechanism of embolism in CCMA caused by a spontaneous fistulization in the left ventricle.

find any bacteria; Zielh–Neelsen staining, cultures and polymerase chain reaction (PCR) found no Koch’s bacillus. Recovery was rapid and, 6 months later, the patient was in good health with a satisfactory echography and no recurrence of stroke.

3. Discussion

CCMA represents less than 1% of all mitral calcifications (calcification of the mitral annulus, CMA) with a prevalence of 0.067% in the general population [1,2]. The diagnosis is echographic, either fortuitous, or on the occasion of complications. Typical presentation will be in the shape of a large, round, echodense, inconsistent mass with a liquid central region, which can be confused with an intracardiac tumor [4]. Typical location is at the posterior part of the mitral annulus [5]. Among the complications described, hemodynamic (mitral insufficiency and syncope) and embolic (stroke) events can be noted. Consensus is far from having been reached with regard to the therapeutic management of CCMA. The question of operating asymptomatic patients preventively then arises, or should surgery be limited to symptomatic cases only? According to some authors, absence of treatment seems to be a good option in the absence of clinical manifestations [1,2]. Cases of retransformation of CCMA into CMA have been reported [1,2] and even cases of spontaneous resolution [6]. However, hemodynamic problems and strokes are an indication for curative surgery or for preventing recurrence. The link between CMA and stroke was emphasized by Benjamin et al. [7] and Malaterre et al. [8], and this can be logically extrapolated to cover CCMA. In the Harpaz et al. series [1], 26% of patients included had a prior history of cardiovascular events but with no established link to cardiac embolism.

In our case, the causal link between the repeated strokes and the CCMA was therefore very strong, and the indication to operate absolute. However, opinion is divided as to the type of surgical treatment suitable. Excision of the caseous zone seems to be admitted by all, and the techniques of repair [5] or of valve replacement [1] are preferred, depending on local conditions. The pathophysiological mechanism underlying emboli is still the subject of debate. The formation of a thrombus and its embolization has been described [9]. Another hypothesis might be that of the migration of a calcification from the mitral annulus [10]. In this respect, our case offers an additional hypothesis as to etiology: the embolic mechanism might be caused by a spontaneous fistulization of the caseous cavity in the lumen of the left ventricle. The valvular tissue was clean without calcification protuberance. The only possibility to explain the embolization was the spontaneous fistulization, as confirmed by the direct communication between the mitral annulus and the left-ventricle lumen. Embolization is, in fact, possibly a rare mechanism and fistulization is probably more frequent as imagined.

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