

Recurrent left atrium myxoma: case report and review of literature

Myxome récidivant de l'oreillette gauche : Cas clinique et revue de la littérature

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Résumé

Le myxome cardiaque est la tumeur cardiaque la plus fréquente. La resection chirurgicale est le traitement de référence avec une évolution souvent favorable; Nous rapportons le cas d'un patient âgé de 21ans sans antécédents particuliers, qui a été pris en charge pour myxome récidivant de l'oreillette gauche découvert à l'occasion de dyspnée et d'hémoptysie. Le délai entre la tumeur initiale et la récurrence était de cinq mois. Nous avons aussi révisé les cas de myome récidivant rapportés dans la littérature.

Mots-clés

Tumeur cardiaque, myxome récidivant, complexe de Carney

Summary

Primary cardiac myxoma is the most common primary cardiac tumor. Tumor resection is the treatment of choice and overall longterm prognosis is good and recurrence is rare. Presented here is the case of a 21-year-old man, with no medical history of cardiovascular diseases, familial cardiac neoplasm and endocrinopathy, who presented with a recurrent left atrium myxoma clinically expressing with hemoptysis and dyspnea. The time interval between the first and second recurrence following surgery was five months. We reviewed evidences and summarized reported cases of recurrent myxoma in the literature.

Keywords

Cardiac tumor, Myxoma, Recurrence, Carney complex.

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INTRODUCTION

Cardiac myxoma (CM) is the most common primary heart neoplasm with an estimated incidence around 0.5-1.0 per million cases per year [1]. Most of CM is sporadic and located in the left atrium (LA). It is considered as a benign disease with good long-term survival after operation. But the recurrence of cardiac myxoma is a potential concern because of its biological invasive characteristics [2].

CASE REPORT

In December 2018, 21 years old male came in the emergency room for hemoptysis and dyspnea. He had no personal or familial medical history.

At physical examination, he was afebrile with stable hemodynamic status. The oxygen saturation was at 96%. The lungs were clear to auscultation. Cardiac auscultation revealed a holosystolic murmur at the apex. There were no signs of heart failure.

Chest X-rays showed normal pulmonary parenchyma and no cardiomegaly. Electrocardiogram showed a regular sinus rhythm at 100 cycles per minute. Hemoglobin was at 8 g/100ml.

The patient underwent a bronchoscopy that came back normal and then was referred for thoracic computed tomography revealing a large mass (38*66mm) in the left atrium. (figure1).

Transthoracic echocardiography demonstrated a solid mass in the left atrium obstructing the mitral valve in systole and protruding into the left ventricle. The echocardiographic findings suggested that the mass was a myxoma (figure 2).

The patient was taken to the operating room. The operation was performed under cardio-pulmonary bypass with a median sternotomy. The left atrium incision parallel to the atrial septum exposed a pedunculated white mass of firm consistency and oblong shape, arising from the free wall of the left atrium between the orifices of pulmonary veins. The tumor was obstructing the left superior pulmonary vein's orifice, adherent to the posterior mitral leaflet and its extremity prolapsed in the cleft of the mitral valve (figure3). The tumor was resected completely with the stump attached to the left atrium wall which was repaired with stitches. Surgical specimens were sent for pathologic evaluation and the diagnosis of cardiac myxoma was verified. The patient tolerated the procedure well and had no noteworthy postoperative complications. He was discharged from the hospital one week after the operation.

Five months later (May 2019), he presented again with the same symptoms. We went straight to echocardiography that showed a mass in the left atrium partially obstructing the mitral valve (Figure4). The echocardiogram findings were confirmed by cardiac CT scan (Figure5).

Redo-surgery was performed through second sternotomy. The left atrium was incised parallel to the atrial septum. A large white tumor of solid consistency was occupying the whole left atrium cavity; adherent to the left inferior pulmonary vein and the left atrial appendage (figure6). The mass was released from its adhesions and was totally removed in one block. The left atrium appendage was excluded.

The tumor was sent for pathological examination that confirmed the diagnosis. Postoperative phase was uneventful. The patient was discharged one week later. And the evolution is good at two months.

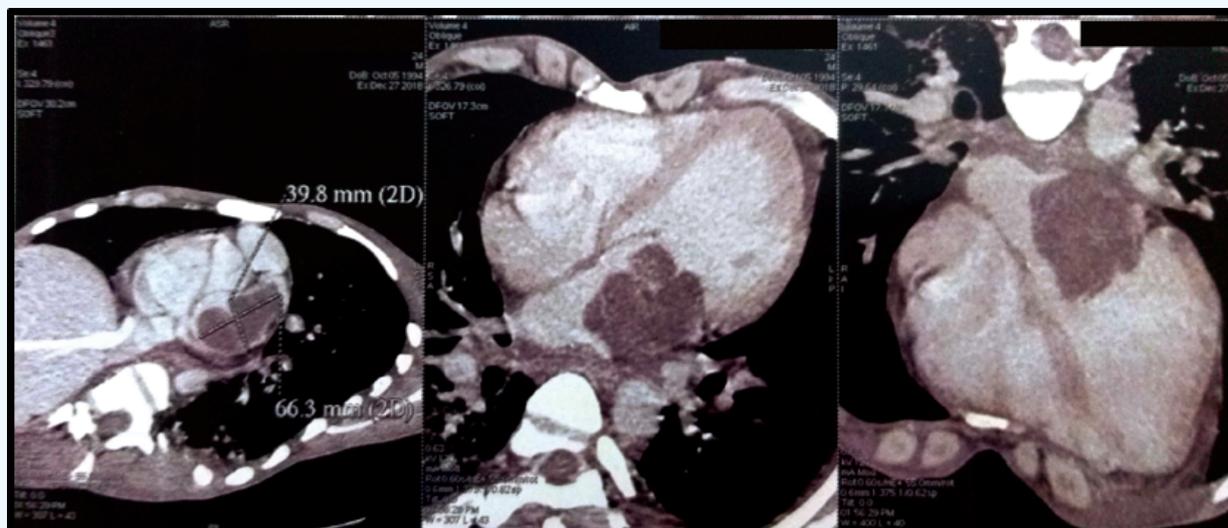


Figure 1: Thoracic CT-scan, December 2018: a large mass of 39*66mm in the left atrium.



Figure 2: Trans-thoracic echocardiography, December 2018: Left atrium myxoma obstructing the mitral valve in systole and protruding into the left ventricle.



Figure 3: The resected mass, December 2018: Pedunculated white mass of firm consistency and oblong shape.



Figure 4: Trans-thoracic echocardiography, May 2019: recurrent left atrium myxoma, partially obstructing the mitral valve.

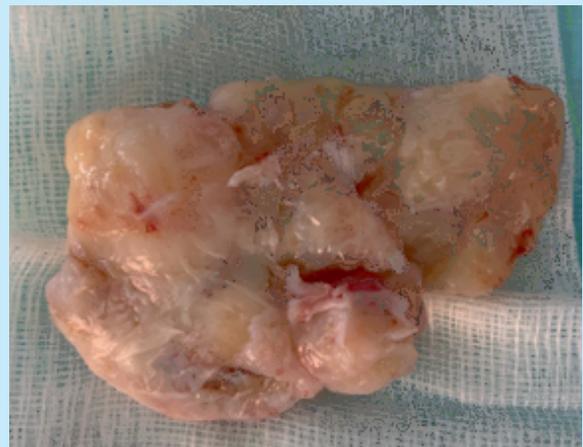


Figure 6: The resected mass, May 2019: white tumor of solid consistency was occupying the whole left atrium cavity.

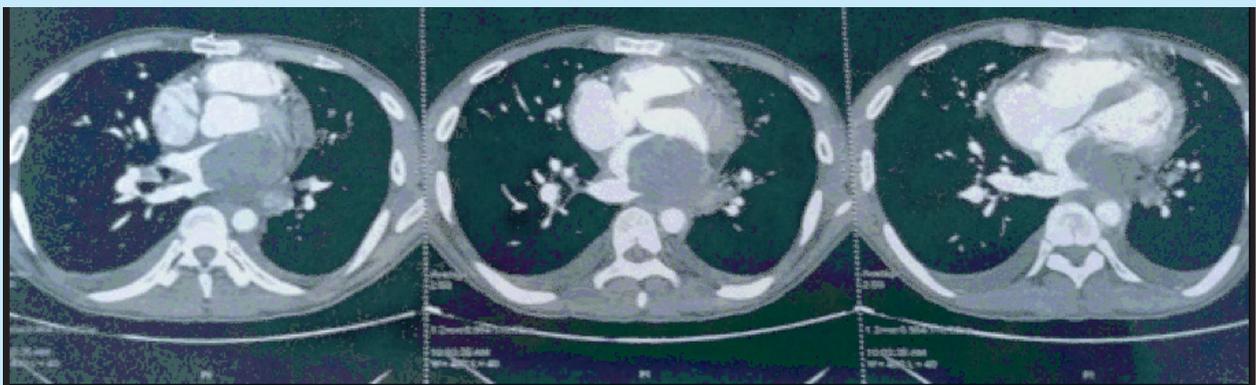


Figure 5: Thoracic CT-scan, May2019: Cardiac myxoma occupying the whole left atrium.

DISCUSSION

Cardiac myxoma is histologically benign, it originates from endocardial tissue and consists of myxoid stroma and variable myxoma cells. Although the tumor usually presented in a sporadic form, a familial form was also rarely found with most patients presented as Carney syndrome [1].

The rate of recurrence of cardiac myxomas is around 2-7% [1,3,4] for sporadic cases and can reach as high as 22% for cases of Carney complex [5]. In familial type, the recurrent rate was found to be as high as 12% [7].

The age range at the time of the first surgical resection was 5 to 63 years, with a mean age of 25.8 years. Most of patients (60%) were of female sex.

The number of recurrences was between 1 to 4 recurrences and the recurrence interval varied from 3 months to 33 years [8-51] [table1].

The symptomatology in recurrent myxomas is the same as in general non-recurrent myxomas. Myxomas usually show signs of obstructed ventricular filling thereby mimicking a mitral or tricuspid valve stenosis [9-14,18,19,23,37,38,41,45,46,48,51] [table1]. Production and release of interleukin 6 by the tumour cells give rise to non-specific symptoms such as fatigue, weight loss, fever and arthralgia [11,12,15,31,44,51] [table1].

Echocardiography is a preferred and efficient modality for the diagnosis of cardiac myxoma. It was the initial modality for the diagnosis in all patients. It shows the tumors, their location, their number, their size as well as their hemodynamic compromise. Myxomas usually appear as well-defined, smooth, oval or lobular lesions that are commonly pedunculated. The most common location of CM was in the left atrium (72%) [8-10,12,13,16-18,20, 31,33,34,36,37,40-45,47-51][table1]. Right atrium was involved in 16% of cases [9,17,21,22,25-17,29,33,36-40,43,45,47][table1] and left ventricle in only 14 cases [15,16,19,21,24,34-36,43-46,48,49] [table1]. CM Size varied from 0.8 cm to 10 cm [9-51][table1] and was multiple in 30 cases [9,15,17,18,21,24-28,33,37,38,43-45,47,48,51] [table1]. Normal intracardiac structures and embryological remnants can sometimes be mistaken for atrial myxomas on TTE.

Transoesophageal echocardiography (TEE) may demonstrate the site of insertion and other surface features of the myxoma as well as hemorrhage, necrosis, cysts and calcification.

TEE has limitations in viewing the right heart and extracardiac structures and requires sedation. [18,21,25-29,34,41] [table1].

Cardiac magnetic resonance imaging has become increasingly valuable for determining cardiac tumors. It is very useful to differentiate myxomas from other cardiac masses, thrombus and anatomical abnormalities. Myxomas typically appear hyperintense compared with

normal myocardium and hypointense compared with the blood pool. Following contrast agent administration, lesions often show more heterogeneous enhancements on late gadolinium enhancement [24,31,35,39,45,46] [table1].

Computed tomographic scan (CT) was useful in only five patients [29,34,40,41,43] [table1], contributing to the diagnosis process, and providing useful perspectives for operative planning and spatial orientation, due to three-dimensional reconstruction, when the position of the myxoma is unusual particularly in posterolateral position [43].

All TEE, cardiac magnetic resonance imaging and cardiac CT scan may be helpful for follow up.

Patients with CM need aggressive intervention. The gold standard treatment of CM was complete surgical resection of the tumor with or without the adjacent endocardium tissue [8-12,15,16,19-31,33-51] [table1]. Excision of the underlying atrial septum with shaving off of part of the myocardium (at least 5-mm margin all around) underlying the stalk is necessary for the prevention of recurrence ; closure with untreated autologous pericardium is dictum in all atrial myxoma excision cases [22,25,26,30,37,41,43,51,52][table1]. In the case of Zehra Bayramoglu [51] the tumors were excised with wide margins and the atrial septal defects were enclosed with a bovine pericardial patch to exclude a third recurrence of the tumor due to incomplete resection.

Orthotopic heart transplantation (OHT) has been considered as a final treatment option in patients with unresectable cardiac tumors. According to the published data, there has been only two cases of CM patient who were treated with OHT [18,48]. Goldstein et al. performed OHT on a patient with recurrent CM due to extensive involvement of the tumor [18].

As for Jakrin Kewcharoen et al [48] the heart transplant was proposed with

the fourth recurrence of CM due to its fast growing, and the complications from the neoplasm including valve regurgitation and obstruction and embolic events [48].

Despite cardiac myxoma being a relatively benign disease, multiple recurrences indicated its malignant biological behavior. Predicting recurrence of cardiac myxomas is still an open question. Recurrent CM was relatively common in patients with multiple lesions [9,15,17,18,21,24-28,33,37,38,43-45,47,48,51][table1], tumor location other than the LA, family history [25,26,29-31,33-40,48,50,51][table1] and younger age [12,15-17,19-21,24,25,28,30,31,34,36,38,41,42,44,46,48-51][table1]. Recurrence is higher in Carney complex than in sporadic myxoma [46,47,50-60,70-72][table1]. Carney complex is an autosomal dominant syndrome characterized by myxomas in the heart and other locations, spotty pigmentation of the skin, endocrine tumors [55]. It comprises 7% of all cardiac

myxomas and can be found in all ages, especially young women.

Possible mechanisms of recurrence were suggested such as incomplete resection, growth from another focus, familial inheritance, or implantation of a fragment from the original tumor [53].

Shah et al. found that ventricular tumors were seven times more likely to recur than atrial tumors. This might be due to the lower likelihood of being able to perform a full-thickness wall resection for ventricular tumors than for atrial tumors. In this study, interestingly, authors reported that smaller myxomas had a higher probability of recurrence, whereas a larger tumor size was associated with a lower risk of recurrence which is different from many tumors elsewhere in the body [54]. In most sporadic myxoma cases, incomplete resection of the tumor attachment area is considered as the common cause of recurrence. Although there was no difference in tumor recurrence based on resection margin, namely, endocardial resection versus base of stalk resection. It seems preferable to perform an endocardial resection when technically feasible. That is almost always the case when the tumor arises from the free wall of the atrium or atrial septum. This is important because the final pathology is often not available in the operating room, and although the typical gross appearance of a myxoma is usually evident to the surgeon, the final pathology report occasionally changes. Consequently, adhering to basic principles of tumor resection with clear margins seems reasonable when there is little risk of excising additional tissue.

Keeling et al. found significant immunological changes in

myxoma patients. In the case of recurrence, serum protein electrophoresis, C-reactive protein, fluorescence-activated cell sorter, interleukin 2 receptor and intracellular adhesion molecule levels may be altered. Immunological and genetic screening of these patients may help to identify patients at risk for additional recurrence [56].

CONCLUSION

There are insufficient data on the recurrence of cardiac myxoma and no guidelines or expert consensus has been established to assess the recurrence. The reported case and the literature review of repeated recurrent cardiac myxomas demonstrate the importance of regular echocardiography after surgical resection in order to detect recurrence and avoid complications. Echocardiographic follow up should be performed each year to detect myxoma recurrences early. Patients with known Carney complex and patients with recurrent CM should have this examination every six months if they have already had a surgical resection. Further studies are required to investigate the reliable predictors of myxoma recurrence.

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