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Left Atrial Myxoma and Anomalous Coronary Origin: A Case Report

Y. Fihri[1]*, S. El Manir[2], A. Benyass[1], Y. Moutakiallah[2], Z. Lakhal[1]

[1]Cardiology Department, Mohammed V Military Hospital, Rabat, Morocco
[2]Department of Cardiovascular Surgery, Mohammed V Military Hospital, Rabat, Morocco

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***Corresponding author:** Y. Fihri
Cardiology Department, Mohammed V Military Hospital, Rabat, Morocco

Abstract | **Case Report**

Background. Myxoma is the most frequent benign primary tumour of the heart, and most arise in the left atrium. Congenital coronary anomalies are seen in roughly 1% of coronary angiograms. We found no previous report of the two occurring together. ***Case presentation.*** A 66-year-old woman was referred for exertional dyspnoea. Transthoracic echocardiography showed a large, mobile left atrial mass prolapsing into the mitral orifice, with a mean transmitral gradient of 16.8 mmHg. Routine coronary angiography before surgery showed a circumflex artery arising from the right coronary sinus and running behind the aorta. The tumour was excised and she recovered without complications. ***Conclusion.*** As far as we know, this is the first description of a left atrial myxoma combined with an anomalous circumflex origin. In our view, coronary imaging before tumour surgery is worth doing even in patients without angina, mainly to know where the coronary arteries run before the heart is opened.

Keywords: Cardiac myxoma; left atrium; coronary artery anomaly; anomalous circumflex artery; retro-aortic course; cardiac surgery; coronary angiography.

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INTRODUCTION

Cardiac myxoma remains the most common benign primary tumour of the heart, but it is still a rare diagnosis in daily practice [1]. Autopsy series put the frequency of primary cardiac tumours between 0.001% and 0.03%, and myxomas make up around half of the benign ones operated on in large surgical centres [6,7,15]. About three quarters are found in the left atrium, usually attached to the interatrial septum close to the fossa ovalis. Patients present with obstruction, embolism, constitutional symptoms, or a combination of these [1,6,22].

Coronary artery anomalies are usually discovered by chance and most never cause symptoms [2,8,9]. Yamanaka and Hobbs reviewed 126,595 coronary angiograms and found an anomaly in 1.3% of patients. Separate ostia for the left anterior descending and circumflex arteries came first. A circumflex arising from the right sinus or from the right coronary artery was the next most frequent variant [2]. When the circumflex takes this origin, it almost always passes behind the aortic root, very close to the aortic and mitral annuli [3,10].

Myxoma has been reported with acquired coronary artery disease, with coronary embolism, and with tumour vessels fed by the coronary arteries that can produce a steal phenomenon [5,15,16]. We could not find a published case of myxoma associated with a congenital anomaly of circumflex origin. The coronary arteries develop from a vascular plexus that only later connects to the aortic root [4,18], and this happens at about the same time as the atrial septum forms. A shared developmental background is therefore conceivable, although it remains speculative [4,17–19]. We report such a case and discuss why it matters for surgical planning.

CASE SUMMARY

A 66-year-old woman was admitted for dyspnoea on exertion. On transthoracic echocardiography we found a large mobile mass in the left atrium, with the typical appearance of a myxoma. In diastole the mass moved down through the mitral valve into the left ventricle (Figure 1A).

Continuous-wave Doppler of mitral inflow confirmed a significant obstruction (Figure 1B). Peak velocity was 3.02 m/s, peak gradient 36.6 mmHg and mean gradient 16.8 mmHg, with a heart rate of 113 bpm

at the time of recording. The mitral area calculated from the velocity–time integral was 1.2 cm².

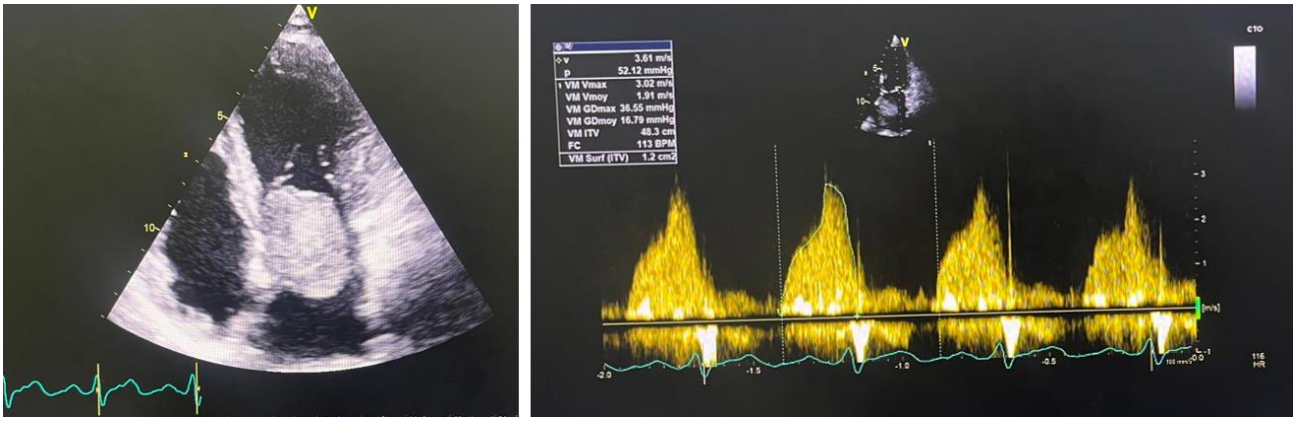


Figure 1: Large left atrial mass prolapsing through the mitral valve with Doppler evidence of significant diastolic inflow obstruction. (A) Apical four-chamber view. (B) Continuous-wave Doppler of mitral inflow

We performed coronary angiography in our catheterisation laboratory as part of the routine preoperative work-up. On the left injection only the anterior descending artery and its branches filled, with no circumflex arising from the left main and no significant stenosis (Figure 2B). This prompted a search for a separate origin. Injection of the right coronary sinus then showed the circumflex arising from it (Figure 2A) and passing behind the aorta to reach the left atrioventricular groove.



Figure 2: Coronary angiography demonstrating anomalous circumflex artery arising from the right coronary sinus with retro-aortic course. (A) Right coronary sinus injection. (B) Left coronary injection

The angiographic findings were discussed with the surgical team before the operation. The tumour was removed under cardiopulmonary bypass, and the course of the circumflex was taken into account when placing the aortic cross-clamp, giving cardioplegia and exposing the left atrium. Excision was complete and the postoperative course was uneventful. Histopathological examination of the resected specimen confirmed the diagnosis of cardiac myxoma. The postoperative course was uneventful, and the patient remained free of symptoms and complications during the three months of follow-up.

DISCUSSION

An exceptional association

Each of these two conditions is uncommon on its own, so finding both in the same patient is unusual [1,2]. In the series of 112 left atrial myxomas reported by Pinede *et al.,* dyspnoea was the most frequent symptom and about two thirds of patients had signs of obstruction [6]. Our patient fits this picture well: her mean transmitral gradient was in the range usually seen in moderate to severe mitral stenosis.

Most reports linking myxoma to the coronary circulation deal with something acquired. Tumour fragments can embolise to a coronary artery and cause infarction, sometimes as the first sign of the disease [16].

Myxomas can also receive feeding branches from the coronary tree, most often from the circumflex or the right coronary artery. On angiography this appears as a "tumour blush", and in some patients it steals enough flow to cause angina without any stenosis [5,15]. Our case is different. The anomaly here is congenital and anatomical, and has nothing to do with the tumour's blood supply or with ischaemia. The two findings coexist; one does not result from the other.

Surgical relevance of the retro-aortic circumflex

From a haemodynamic point of view, a circumflex arising from the right sinus is generally harmless [2,3,8]. In the Coronary Artery Surgery Study registry it was not associated with lower survival [3]. Cheezum *et al.,* also place the retro-aortic course among the low-risk forms of anomalous aortic origin, unlike the inter-arterial course [11].

For the surgeon, however, this course is not trivial. The artery runs at the back of the aortic root, a few millimetres from the non-coronary sinus and the anterior mitral annulus, which is exactly where the surgeon works during clamping and atrial exposure [2,3,10,14]. Roberts and Morrow described anomalous circumflex arteries compressed by the sewing rings of prosthetic valves as early as 1969 [12]. Vaishnava *et al.,*reported a patient who had a myocardial infarction after double valve replacement. The anomalous circumflex had been squeezed between the two prostheses, and the patient needed urgent reoperation [13]. More recently, Portoghese *et al.,*estimated the frequency of this variant at about 0.15%. They advise coronary imaging before every mitral operation, with 3D transoesophageal echocardiography or CT to measure the distance between the artery and the annulus [14].

Even when the artery is perfectly normal in function, its unusual path has to be planned for. It can be compressed or injured, and myocardial protection may be uneven. Cardioplegia given through the left main ostium will not reach a circumflex that comes off the right sinus, so selective ostial or retrograde delivery should be considered. Myxoma excision is usually a straightforward procedure. With this anatomy it needs more care, especially with a biatrial or superior septal approach, a wide septal resection or an associated mitral repair [7,23].

A possible developmental link

We cannot claim any causal link between the two findings. Embryology does, however, offer a plausible frame. The coronary vessels form from a subepicardial plexus that receives cells from the proepicardium, the sinus venosus and the endocardium. This plexus then grows into the aortic root, where the ostia appear [4,18,19]. Remodelling of the plexus and septation of the atria take place during overlapping periods of development [4,18].

Myxoma cells express markers of embryonic endocardial and mesenchymal progenitors. This has led to the idea that they come from multipotent cells left in the septum around the fossa ovalis [17]. A septal tumour and an abnormal coronary origin in the same heart are therefore not incompatible from a developmental point of view, even though one does not explain the other. Carney complex, linked to *PRKAR1A* mutations, should be kept in mind in young patients and in multiple or recurrent tumours [21]. Nothing in our patient's history or presentation suggested it. Our view is that an anatomical variant found together with a septal lesion deserves attention rather than being filed as an incidental finding.

Implications for preoperative assessment

This case argues for routine coronary imaging before tumour surgery, not only to look for coronary disease but to know the anatomy. The ESC/EACTS guidelines already recommend coronary angiography or coronary CT before valve surgery in patients over 40 or with cardiovascular risk factors [20]. Applying the same rule to cardiac tumours would also show feeding vessels that may need to be ligated [15]. A coronary anomaly that causes no symptoms in daily life can become a real problem once the heart is arrested and opened.

From the angiographer's side, a "missing" circumflex on the left injection should never be read as an occlusion before both coronary sinuses have been explored, as Page *et al.,*pointed out in their early angiographic description [10]. More broadly, we think that coronary angiography or coronary CT should be part of the workup before myxoma excision, including in patients without symptoms, and particularly when the septum is going to be opened or resected. CT has the advantage of showing the tumour, its attachment and the 3D course of an abnormal artery in a single examination [11,22]. The aim is not to find disease. It is to avoid an injury during surgery that could have been prevented.

CONCLUSION

We describe a left atrial myxoma associated with a circumflex artery arising from the right coronary sinus. The association may have a developmental basis, although this cannot be proven from a single case. In practice, mapping the coronary anatomy in the catheterisation laboratory allowed the surgical team to adapt clamping, cardioplegia and atrial exposure to this patient. Further reports will be needed to know whether the association is more than a coincidence.

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