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Transradial Embolization Prior to Surgical Resection of a Cardiac Paraganglioma Presenting as an Acute Coronary Syndrome With Normal Coronaries

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Manuscript Preparation E
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Patient: Female, 67-year-old
Final Diagnosis: Cardiac paraganglioma
Symptoms: Chest pain
Clinical Procedure: Surgical resection
Specialty: Cardiac Surgery • Cardiology

Objective: Unusual clinical course

Background: Cardiac paragangliomas are extremely rare neuroendocrine tumors arising from chromaffin cells of neural crest origin. These tumors are typically hypervascular and can derive part of their arterial supply from the coronary circulation. Because of their rarity and heterogeneous clinical presentation, diagnosis can be challenging. Symptoms can result from catecholamine secretion, local mass effect on adjacent cardiac structures, or incidental discovery during imaging studies. In rare cases, cardiac paragangliomas may mimic an acute coronary syndrome despite the absence of obstructive coronary artery disease.

Case Report: A 67-year-old woman presented with non–ST-segment elevation myocardial infarction in the context of rapid atrial fibrillation. Coronary angiography demonstrated normal epicardial coronary arteries but revealed an intense vascularization of a right retroatrial mass supplied by branches of both the right and left coronary arteries. Subsequent multimodality imaging identified a well-defined hypervascular mediastinal mass highly suggestive of a cardiac paraganglioma. Elevated urinary normetanephrine levels confirmed the secretory nature of the tumor. Given the extensive vascular supply visualized on angiography, preoperative transradial transcatheter embolization of the coronary feeding vessels was performed to reduce the risk of intraoperative bleeding and possibly reduce the tumor size. The patient subsequently underwent complete surgical resection with histopathologic confirmation of cardiac paraganglioma.

Conclusions: This case highlights the importance of recognizing a coronary tumor blush, which should prompt comprehensive diagnostic evaluation. In this patient, preoperative embolization combined with complete surgical resection was feasible and associated with a favorable long-term outcome, and may be considered in selected cases.

Keywords: coronary artery disease • cardiac surgical procedures • radial artery • paraganglioma

Full-text PDF: https://www.amjcaserep.com/abstract/index/idArt/953581

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Introduction

Cardiac paragangliomas are rare neuroendocrine tumors originating from neural crest–derived chromaffin cells [1]. These highly vascular lesions can receive part of their blood supply from the coronary circulation, rendering their anatomical and functional assessment particularly complex. Owing to their rarity and heterogeneous clinical presentation, diagnosis is often challenging and typically relies on multimodal imaging. We report a rare case of cardiac paraganglioma presenting as a pseudo–acute coronary syndrome, emphasizing the diagnostic value of coronary tumor blush on angiography, which guided transradial embolization of coronary feeding vessels prior to surgical resection. The case also illustrates the role of multimodality imaging and the favorable long-term outcome.

Case Report

A 67-year-old woman with a history of active smoking and essential tremor was referred to our institution for a non–ST-segment elevation myocardial infarction, which occurred in the context of rapid atrial fibrillation. There was no prior history of similar episodes or known coronary artery disease, structural heart disease, endocrine disorders, or prior malignancy. She had no documented history of hypertension or previously diagnosed catecholamine-related symptoms, such as episodic headaches or paroxysmal hypertension. However, she described recurrent episodes of diaphoresis over the preceding year, which had not been previously investigated. On admission, she was in hemodynamically stable condition. Electrocardiography showed atrial fibrillation with a rapid ventricular response and nonspecific ST segment changes. Cardiac biomarkers were elevated, consistent with myocardial injury.

The patient subsequently underwent coronary angiography, which revealed normal epicardial coronary arteries with no evidence of obstructive disease. However, it revealed a prominent tumoral blush supplied by branches of both the right and left coronary arteries, suggesting a marked vascularization of an extracoronary mass (**Figure 1A, 1B**).

Transthoracic echocardiography identified a mass in the right retroatrial region associated with a moderate pericardial effusion, initially raising suspicion for a cardiac myxoma. Pericardial drainage was performed because of the effusion and showed no malignant cells on cytologic examination. Subsequent contrast-enhanced computed tomography of the chest revealed a 57 × 52 × 55-mm well-defined, hypervascular mediastinal mass with mild compression of the right atrium and superior vena cava (**Figure 2A**). Further functional imaging with metaiodobenzylguanidine (MIBG) positron emission tomography demonstrated intense tracer uptake with marked hypermetabolic activity with no evidence of metastatic disease, findings highly suggestive of a paraganglioma (**Figure 2B**). Urinary normetanephrine levels were elevated, confirming the secretory nature of the tumor.

Given the extensive vascular supply visualized on coronary angiography, preoperative embolization was performed to reduce the risk of intraoperative bleeding (**Figure 3A, 3B**). A JR4 7-F guiding catheter was advanced to the proximal right coronary artery via radial access. An HT Balance Middleweight Universal 0.014" guidewire (Abbott) was positioned distal to the right marginal branch, followed by advancement of an ASAHI SION

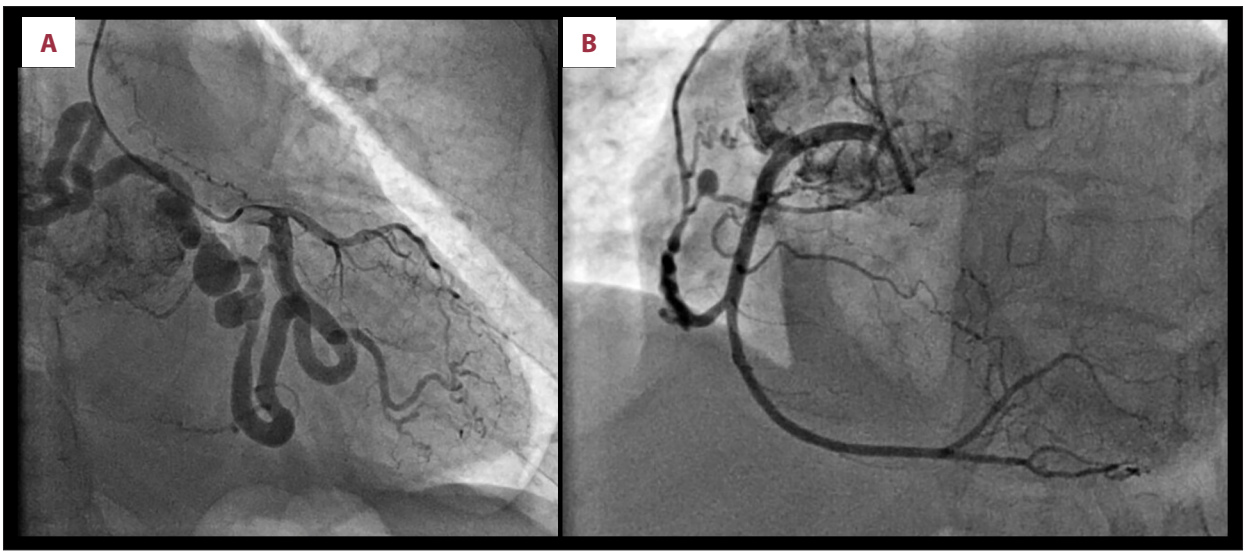

Figure 1. (**A**) Coronary angiography of the left coronary artery demonstrating a marked tumoral blush consistent with a highly vascularized cardiac mass with no obstructive coronary disease. (**B**) Coronary angiography of the right coronary artery showing additional arterial supply to the mass.

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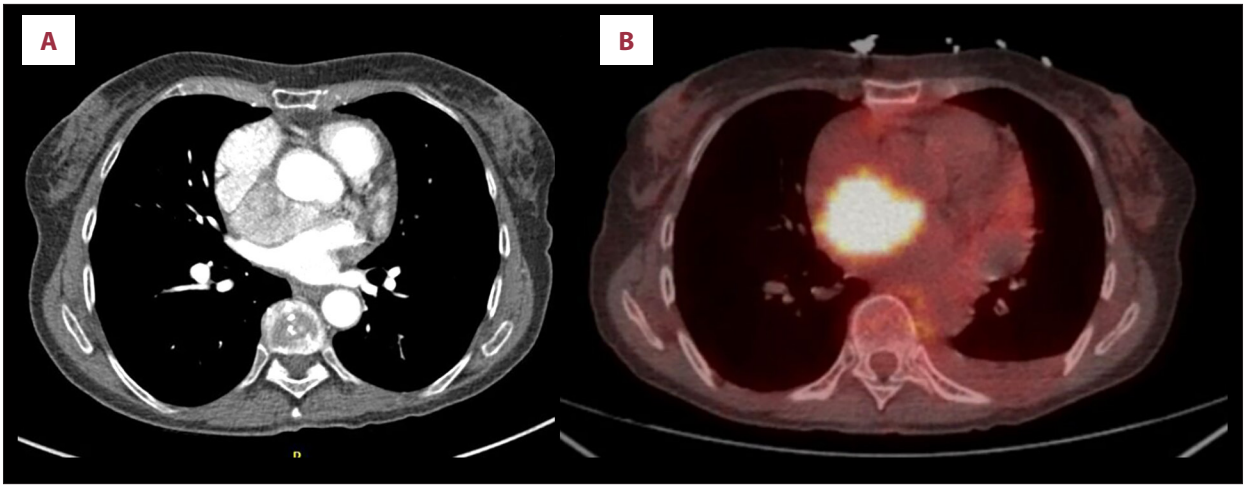

Figure 2. (**A**) Contrast-enhanced computed tomography of the chest revealing a well-defined hypervascular right retroatrial mediastinal mass. (**B**) Metaiodobenzylguanidine (MIBG) positron emission tomography showing intense tracer uptake within the mass.

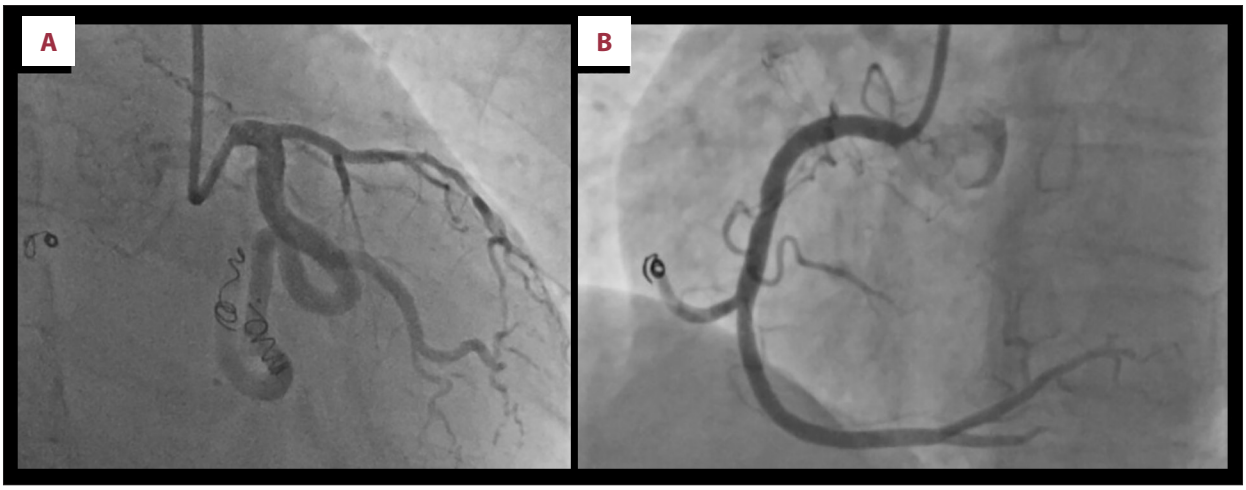

Figure 3. (**A, B**) Preoperative embolization demonstrating a marked reduction in tumoral blush from branches of the left and right coronary artery.

guidewire into the same branch. Coil embolization was performed at the ostium of the right marginal artery, achieving complete vessel occlusion. Additional coils were deployed to ensure secure occlusion. Subsequently, an XB 3.0 7-F guiding catheter was engaged in the left main coronary artery. Coil embolization was then performed at the level of the mid circumflex artery, resulting in successful vessel occlusion. Following the embolization procedure, the patient developed elevated blood pressure, which was successfully managed with medical therapy consisting of doxazosin 4 mg twice daily and labetalol 50 mg twice daily, resulting in good blood pressure control.

Three months later, a complex surgical resection of the paraganglioma was performed, involving an en bloc excision of the left atrial dome, interatrial septum, two-thirds of the right atrium, and the right superior vena cava. Reconstruction was subsequently achieved using bovine pericardial patches to restore cardiac and caval continuity. Because the sinus node had been resected with the involved tissue, the patient developed a very slow escape ventricular rhythm. A temporary pacing lead was placed, followed by implantation of permanent atrial and ventricular leads connected to a right subcostal pacemaker. Gross, histological, and immunohistochemical analyses confirmed the diagnosis of cardiac paraganglioma. Macroscopically, the tumor appeared as a well-defined, lobulated mass with a central scar (**Figure 4A, 4B**). Histological evaluation revealed a neuroendocrine proliferation arranged in a characteristic “zellballen” pattern. Immunohistochemistry showed strong expression of neuroendocrine markers (chromogranin, synaptophysin, CD56) in tumor cells, while sustentacular cells demonstrated positivity for S-100 (**Figure 5**). The postoperative course was uneventful.

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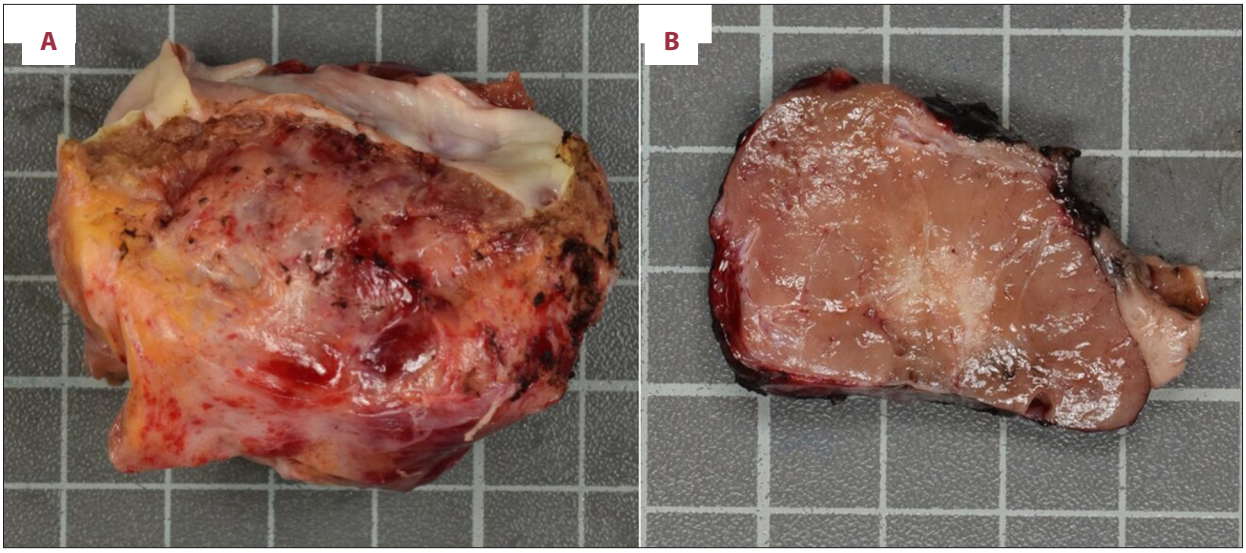

Figure 4. (**A**) Gross specimen of the resected cardiac paraganglioma showing a well-circumscribed, lobulated mass. (**B**) Representative cross-section highlighting the lobulated architecture with a central fibrous scar.

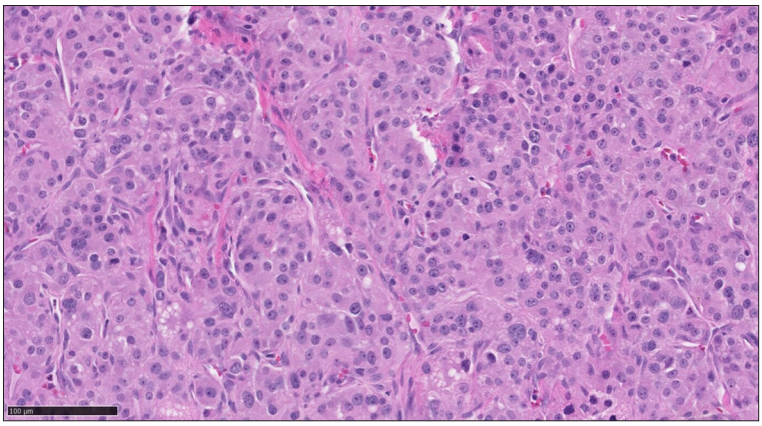

Figure 5. Histological examination (hematoxylin and eosin, 25×) demonstrating the classic organoid "zellballen" pattern, characterized by nests of neuroendocrine cells surrounded by sustentacular cells and separated by delicate fibrous septa.

Genetic testing identified a pathogenic variant in the SDHC gene (c.397C>T, p.Arg133Ter), resulting in a premature stop codon and predicted loss of normal protein function due to truncation. This finding is consistent with hereditary paraganglioma pheochromocytoma syndrome, an autosomal dominant condition associated predominantly with head and neck paragangliomas.

At 10-year follow-up, the patient remained clinically asymptomatic, with no evidence of tumor recurrence on transthoracic echocardiography or positron emission tomography imaging.

Discussion

Primary cardiac tumors remain a rare finding with an incidence of 0.001% to 0.03% in reported autopsy series [1], and cardiac paragangliomas are extremely rare neuroendocrine tumors, with fewer than 150 cases reported in the literature [2]. Paragangliomas arise from chromaffin cells derived from the neural crest. When these tumors originate in the adrenal medulla, they are referred to as pheochromocytomas. In contrast, chromaffin cell tumors arising outside the adrenal gland are termed paragangliomas, approximately 2% of which occur within the thoracic cavity [3], and 10% are malignant [4]. However, malignancy can only be definitively established by the presence of metastatic disease, for which the 5-year survival rate remains below 50% [5]. Their clinical presentation is highly variable and depends on tumor location, secretory activity, and degree of mass effect. Only approximately 18% of patients present with chest pain or distress [6]. These tumors are often hypervascular and may receive blood supply from the coronary arteries. Although uncommon, they may present with acute coronary syndrome, either due to catecholamine secretion, coronary steal phenomena, or incidental discovery during angiographic evaluation [7]. In our case, the patient presented in the setting of an acute coronary syndrome that may be explained by several pathophysiological mechanisms, including

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catecholamine-induced coronary vasospasm and microvascular dysfunction leading to myocardial ischemia, as well as a coronary steal phenomenon due to the highly vascularized tumor diverting blood flow away from the myocardium. Coronary angiography, performed in this context, revealed a prominent tumoral blush suggestive of a hypervascular cardiac mass. Subsequent multimodality imaging, together with elevated urinary catecholamine levels, raised strong suspicion for a paraganglioma. In light of the tumor's pronounced hypervascularity, a transradial transcatheter embolization was performed in our patient. Preoperative embolization of the tumor-feeding arteries is a key strategy to reduce intraoperative bleeding [8]. Only a limited number of paraganglioma cases have been managed with transcatheter arterial embolization. The patient in a previously reported case underwent embolization followed by successful surgical resection [9], and another case required embolization of both coronary and bronchial feeding vessels prior to definitive surgery, which was also successfully completed [10]. Covered stents have previously been used in the coronary circulation to occlude branch vessels prior to paraganglioma surgery [11,12]. However, their use has been associated with relatively high rates of in-stent restenosis and stent thrombosis at mid- and long-term follow-up [12]. Surgical resection is the only curative strategy but is associated with a significant risk of intraoperative hemorrhage [13] and can require complex reconstruction of adjacent structures, as illustrated by the atrial and superior vena cava reconstruction performed in our case, with subsequent permanent pacemaker implantation.

Long-term follow-up is essential, as recurrence can occur years after resection [14]. The absence of recurrence at 10 years in our patient supports the favorable prognosis.

This case underscores several important clinical messages. First, the presence of a tumoral blush during coronary angiography in the absence of obstructive coronary disease should alert clinicians to the possibility of a hypervascular cardiac or mediastinal mass. Second, multimodal imaging and biochemical testing are indispensable for accurate diagnosis. Third, recognition of coronary arterial supply to the tumor has critical therapeutic implications, particularly regarding preoperative embolization and surgical planning. Ultimately, coronary angiography in this setting served not only to exclude obstructive coronary disease but also as a key diagnostic and therapeutic step guiding definitive management.

Conclusions

This case illustrates how coronary angiography can reveal unexpected extracoronary pathology through the identification of a tumoral blush. Recognition of this finding should prompt further multimodality imaging and biochemical evaluation for hypervascular cardiac tumors, such as paraganglioma. In selected cases, preoperative transcatheter embolization of coronary feeding vessels can be an effective strategy to reduce intraoperative bleeding and facilitate definitive surgical resection.

Institution Where Work Was Done

Quebec Heart-Lung Institute, Quebec, QC, Canada.

Patient Consent

Informed consent was obtained from the patient for publication of this case report and any accompanying images.

Declaration of Figures' Authenticity

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.

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