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Mediastinal paraganglioma encasing the left main coronary artery

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Abstract

Mediastinal paragangliomas are rare neuroendocrine tumors that can encase major structures in the chest. A 59-year-old male presenting with new chest pain was found to have a 3.5 cm × 5.7 cm × 3.3 cm mass encircling the Left Main (LM) coronary artery within the middle pericardial recess (Figure 1). Intraoperatively, the mass involved the LM, so the LM was ligated and bypassed. Final pathology revealed paraganglioma. Paragangliomas are PET avid benign neoplasms with malignant potential. Preoperative angiography can be helpful for operative planning. Resection with considerations for cardiac reconstruction and/or revascularization is the preferred treatment.

Keywords: Paraganglioma; Mediastinal tumor; Multidisciplinary care.

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Introduction

Mediastinal paraganglioma are rare highly vascular neuroendocrine tumors that can encase major structures in the chest, including great vessels, thereby necessitating Cardiopulmonary Bypass (CPB) to facilitate resection [1]. Complete surgical resection remains the preferred treatment for mediastinal paragangliomas, due to tumor invasiveness and relatively ineffective responsiveness to chemotherapy/radiation [2]. Overall survival following surgical resection is similar to that for the general population, unless the tumor is metastatic, which then lowers the 5-year survival to <50% [3,4]. Analysis of the mediastinal paraganglioma's intimate blood supply should be considered, with plans for possible cardiac reconstruction or revascularization at the time of surgery.

We present a case of a mediastinal paraganglioma encasing the left main (LM) coronary artery requiring Coronary Artery Bypass Grafting (CABG) after tumor excision.

Case report

A 59-year-old male with hypertension presented with new onset chest pain. Nuclear stress test was negative for ischemia. A Transthoracic Echocardiogram (TTE) demonstrated a Left Ventricular Ejection Fraction (LVEF) of 65-70% without wall motion abnormalities or valvular regurgitation. Coronary Computed Tomography Angiography (CCTA) identified a 3.5 cm × 5.7 cm × 3.3 cm enhancing mass encircling a patent LM coronary artery within the middle pericardial recess (Figure 1). The Left Anterior Descending artery (LAD) was encased proximally by the mass. Positron Emission Tomography-Computed Tomography (PET/CT) re-demonstrated an intense hypermetabolic mediastinal mass without other sites of activity (Figure 2). The preoperative diagnosis was paraganglioma. Left Heart Catheterization (LHC) showed 50% narrowing of the proximal LAD, likely from the external compression of the LM with a fractional flow reserve of 0.82 as well as contrast filling into the tumor from the LM.

Intraoperative Transesophageal Echocardiogram (TEE) noted a left ventricular ejection fraction of 50-55% with trace Aortic Insufficiency (AI). Entry to the mediastinum was gained via median sternotomy. Consistent with preoperative workup, the mass was supplied by the LM with some compression to the proximal LAD. The mass was adherent to the Pulmonary Artery (PA). CPB was required. A pericardial patch repair was performed on the PA upon achieving tumor separation. Ultimately, the mass was excised successfully, and the LM was bypassed by using reverse saphenous vein graft x^2 from the aorta to the LAD and the Obtuse Marginal (OM). The patient was extubated on Postoperative Day (POD) 1. TTE performed on POD 3 noted an LVEF of 60% with trace aortic regurgitation. The patient was discharged to cardiac rehabilitation on POD 7. Final pathology confirmed paraganglioma.

On POD 15, the patient was readmitted with complaints of worsening dyspnea and palpitations. TEE showed LVEF of 55% with new onset severe AI. The patient subsequently underwent a redo sternotomy and aortic valve replacement with a 23 mm porcine bioprosthetic valve. The prior bypass grafts were checked with a flow probe and had excellent flow in both grafts. The patient's condition improved, and he was subsequently discharged home on POD 10. On patient's follow up visit, TTE showed LVEF 40-45% with intact bioprosthetic aortic valve function.

Discussion/conclusion

Cardiac paragangliomas are rare neuroendocrine tumors accounting for <1% of all cardiac tumors [5]. These tumors arise from chromaffin cells of the heart and are often highly vascularized. Surgery remains the mainstay treatment, and considerations for cardiac reconstructions and concomitant CABG should be anticipated to achieve complete resection [1-5,6].

The 2004 World Health Organization Classification of Tumors defines malignant cardiac paragangliomas as evidence of metastasis to non-chromaffin tissues [7]. Diagnostic imaging modalities for paragangliomas include ^{131}I-Metaiodoben-zylguanidine (MIBG) scintigraphy and ^{111}In-diethylenetriaminepentaacetic acid-octreotide scintigraphy (OCT) [5]. We employed PET/CT to localize the paraganglioma and exclude metastases.

Cardiac paragangliomas are highly vascularized because of blood supply from coronary vessels. Preoperative coronary angiography is useful in predicting the likelihood of requiring CABG at time of resection. LHC performed on our patient identified the LM as the vascular supply for the paraganglioma allowing us to anticipate the need to perform bypass from the aorta to the LAD and OM. In addition, the LHC revealed extrinsic compression of the LM that was not appreciated on CCTA.

Although imaging can aid in determining tumor respectability, the feasibility of complete resection is often made intraoperatively. Based on preoperative imaging, we anticipated the tumor to be resectable. Intraoperatively, the tumor was densely adherent to the adventitia of the PA. Fortunately, the PA was amenable to a pericardial patch repair after tumor resection. For our patient, the tumor was completely resected without needing significant cardiac reconstruction. Anticipating the need for possible reconstruction should be considered prior to operative intervention. Preoperative imaging may raise suspicion if significant reconstruction is likely.

The patient's postoperative course from the index operation remained otherwise unremarkable, and he was discharged to cardiac rehabilitation in stable condition. Postoperative TTE did not demonstrate any significant concerns aside from the stable trace AI which was also noted on the TEE prior to the cardiac paraganglioma resection and coronary bypass. When the patient represented with heart failure from severe AI, the decision was made to proceed with operative aortic valve replacement instead of transaortic valve replacement due to absence of calcified aortic valves or aortic stenosis. The exact etiology of patient's new significant AI requiring aortic valve replacement is unknown, especially occurring three weeks after the index operation and subsequent TTEs without evidence of severe AI prior to discharge. During the valve replacement operation, we did not identify any specific abnormalities with each leaflet except a foreshortened and slightly downward retraction of the left coronary sinus leaflet that did not coapt with the other leaflets. Therefore, there was a possibility that the presence of the cardiac mass masked the patient's underlying AI by means of a mass-effect mechanism with the symptoms of acute heart failure presenting soon after surgery.

Given the rarity of cardiac paragangliomas, no clear guidelines exist for surveillance after resection. Unless metastatic, it is reported that survival following resection is comparable to that of the general population. If the tumor is metastatic, the "5-year survival is <50% of age-matched controls" [3,8]. Although rare, there is a possibility of recurrence, with rates higher in the metastatic population; therefore, it is recommended that patients undergo periodic imaging studies after resection. We recommend that patients be included in a multidisciplinary team after initial diagnosis including cardiology, surgery, and oncology to monitor their postoperative cardiac function as well as surveillance for recurrence/metastasis.

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