Case Report

Coexistence of Pericardial and Hepatic Hemangiomas

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ABSTRACT
Pericardial hemangioma is very rare and their coincidence with other visceral hemangiomas is exceptional. We are reporting a 72-year-old man with an incidentally discovered pericardial hemangioma during coronary artery bypass graft (CABG) and mitral valve replacement surgery and a prior history of multiple hepatic cavernous hemangiomas. This case has been reported due to its extreme rarity. To the best of our knowledge, this is the second case of this kind reported till date.

Keywords: Hemangioma, Liver, Pericardium

Introduction
Primary pericardial neoplasms are uncommon with an incidence of 0.001% to 0.03%, the most common one being mesothelioma. Hemangiomas make up 5 to 10% of primary cardiac tumors (1-3). Cardiac hemangiomas may involve each one of the three layers of the heart. Histologically, they can be of capillary, cavernous or arteriovenous subtypes (1). Pericardial hemangiomas primarily involve visceral pericardium and are most frequently cavernous. They are mostly solitary, well-defined and measure 1 to 13.5 cm in diameter (4). Most patients are asymptomatic and found incidentally at autopsy. The symptomatic patients present with dyspnea, chest pain, pericardial effusion, etc. (3). Computed tomography, angiography and magnetic resonance imaging are very helpful in evaluating their size, location and their relation with great vessels and important adjacent organs (1, 4). Surgical excision is the treatment of choice and a complete recovery is the rule (3). Liver hemangiomas are the most common...
benign tumors of the liver, usually diagnosed using non-invasive methods such as computed tomography, magnetic resonance imaging and ultrasound. They may be solitary or multiple and may be associated with hemangiomas of other body sites, as occurs in syndromes such as Von-Hippel-Lindau. Their size is usually stable, but rapid growth in giant cases and development of clinical symptoms may mandate surgical intervention.

Case Report

A 72 years old male was admitted to the Intensive Care Unit, Loghman Hakim Hospital, Tehran, Iran in 2012, with rising BUN and creatinine levels. His past medical history was remarkable for hypertension, severe ischemic heart disease and gout. The chest X-ray showed mild cardiomegaly. A 2 Dimension Color Doppler Echo revealed severe mitral regurgitation, mild aortic insufficiency and a trace of tricuspid regurgitation. Coronary angiography performed after dialysis showed Bi-vessel coronary artery disease. Abdominal ultrasonography revealed multiple echogenic liver masses, ranging 2-5cm in diameter, followed by magnetic resonance imaging, which showed hypointense masses on T1WI, being hyperintense on dual-echo T2WI consistent with the diagnosis of hemangioma. Then, the patient underwent open heart surgery for mitral valve replacement and coronary artery bypass graft. During surgery, a well-defined mass lesion measuring 2cm in diameter attached to the visceral pericardium and right pulmonary artery was incidentally discovered. Postoperatively, the electrocardiogram showed atrial fibrillation and the patient became oliguric, accompanied with rising BUN and creatinine levels and the patient died in spite of intensive medical therapy. Necropsy of one of the hepatic lesions was performed afterwards. The pericardial mass resection specimen was an irregular fragment of tan soft tissue measuring 2.5×1×1 cm with hemorrhagic spongy cut surface. Microscopically, it was composed of back to back thin walled capillary-sized to cavernous vascular channels separated by mesenchymal, predominantly fibroblastic septae (Fig. 1A, B). The hepatic necropsy specimen was a wedge shaped fragment of reddish brown elastic tissue measuring 3×2.5×2.5 cm with hemorrhagic spongy cut surface. Microscopically, it was consistent with cavernous hemangioma, surrounded by liver tissue (Fig. 2A, B).

Fig. 1: A) Pericardial tissue with mesothelial lining (upper part) involved by a neoplasm composed of large vascular channels (×40 objective); B) Pericardial hemangioma, showing large vascular spaces lined by flat endothelial cells (×100 objective)
Discussion

Primary cardiac neoplasms are distinctly uncommon with a frequency up to 0.28% at autopsies. Primary pericardial neoplasms are mesothelioma, fibroma, lipoma, leiomyoma, teratoma and sarcoma with the former being the most common one (1, 4).

Hemangiomas are rare primary pericardial mass lesions (4, 7). Most of the patients are over 50 years old at the time of diagnosis, but they can occur over wide age range from infancy to late adulthood. They mostly originate from the visceral pericardium and are predominantly of the cavernous type (1, 4). These are often asymptomatic and found at autopsies, but, they may present with chest pain, pericardial effusion, dyspnea and mass effect on adjacent organs such as airways (4). Less common presentations are consumption coagulopathy and constrictive pericarditis (8, 9). Another presentation is spontaneous bleeding and resultant hemopericardium; so in the setting of chronic recurrent episodes of pericardial effusion with unknown etiology, pericardial hemangioma should be considered and pericardial imaging should be performed (4, 10). Pericardial hemangiomas may lead to arrhythmias, tamponade and sudden death. So, their diagnosis and treatment are very important (11).

A pericardial lymphangioma can be considered as a differential of hemangioma. A case of pericardial lymphangioma was reported in 2011 by Shroff et al. (12). Differentiation of the two can be made by noting lack of RBCs in the vascular channels and surrounding perilesional inflammatory lymphatic infiltration as well as lower age at presentation in lymphangioma.

To the best of our knowledge, this is the second case of coincidental pericardial and hepatic hemangioma; the first one reported by Brodwater et al. (4). The previously reported case is a 57-year-old woman, known case of rheumatic fever, with history of chest pain and palpitation. She had no abnormality on physical examination and echocardiogram. Chest X-ray revealed an atrial appendage mass. The pericardial origin of the mass was confirmed in chest MRI. After thoracotomy, histologic findings established hemangioma. Moreover, a 2cm mass was detected in the dome of liver on chest CT scan, confirmed as hemangiomaoncontrast-enhanced MRI (4).

In contrast to this case which was symptomatic, our patient did not show any sign or symptom related to his pericardial hemangioma, and discovered incidentally during an operation performed for other reasons. Like their reported case, our case was also a cavernous hemangioma.

Fig. 2: A, B) Liver mass excisional biopsy, show a well circumscribed vascular lesion composed of large vascular channels containing RBCs (×40 objective)
Conclusion
Pericardial hemangiomas can be asymptomatic and should be in mind in cases of unusual presentations such as dysrhythmia and cardiac tamponade. Additionally, their coincidence with angiomas of other viscera, although rare, should always be clinically suspected.

References