[^{18}F]FDG PET/CT in a case of recurrent primary cardiac osteosarcoma

Case Report

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ABSTRACT

Primary cardiac osteosarcoma is a very rare malignancy with a high incidence of local recurrence and systemic metastasis, contributing to the poor prognosis. Radiological modalities are commonly used for the evaluation of cardiac masses. 2-[^{18}F]fluoro-2-deoxy-D-glucose ([^{18}F]FDG) positron emission tomography/computed tomography is a valuable whole-body imaging modality in the evaluation of most subtypes of sarcomas. The value of [^{18}F]FDG PET/CT is not well-established in primary cardiac osteosarcoma, and it has rarely been documented in the literature. Here, we report the findings of [^{18}F]FDG PET/CT in a case of a 38-year-old man with primary cardiac osteosarcoma, which clearly demonstrates the recurrent lesions in the myocardium.

Key words: Primary cardiac osteosarcoma; Cardiac tumor; [^{18}F]FDG; PET/CT

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INTRODUCTION

Cardiac tumors are rare, with an autopsy frequency of 0.001-0.03% [1]. Most of them are secondary malignancies due to direct extension of primary regional tumors originating from breast, lung or mediastinal malignancies, as well as hematogenous or lymphatic metastases originating from other tumors. The remainder of cardiac tumors are mostly primary and benign (myxoma, lipoma, etc.). Only small numbers of cardiac tumors are primary malignancies with sarcomas at the top of the list [1]. Cardiac sarcoma is the cause of over 95% of primary heart malignancies, in order of prevalence including, angiosarcoma, rhabdomyosarcoma, fibrosarcoma, histiosarcoma, and osteosarcoma, respectively [1].

The first case of cardiac osteosarcoma described by McConnel et al. in 1970 [2]. Reviewing the literature in 2016, Wang et al. [3] documented 53 cases of the primary cardiac osteosarcoma. Afterwards, only 5 other cases have been reported ("PubMed" and "GoogleScholar"), so far [4-8].

The differentiation of benign from malignant cardiac tumors is essential [9]. Although conventional imaging modalities, including contrast enhanced computer tomography (ceCT) and magnetic resonance imaging (MRI) are able to differentiate benign from malignant tumors in selected cases, 2-[18F]fluoro-2-deoxy-D-glucose positron emission tomography/CT ([18F]FDG PET/CT) provides complementary diagnostic information, as well as simultaneous staging in a single imaging session [10]. [18F]FDG PET/CT has a significant role in the evaluation of primary bone and soft-tissue sarcomas; however, the primary cardiac osteosarcoma is rare, and the role of [18F]FDG PET/CT in this malignancy is not established.

Here, we report a progressive case of cardiac osteosarcoma underwent [18F]FDG PET/CT for evaluation of recurrence.

CASE PRESENTATION

A 38-year-old man referred to our center for evaluation of the recurrence of cardiac osteosarcoma. The primary tumor was discovered in 2017 following the work-up for the exertional chest pain, and it was reported [5]. At that time, the physical examination and electrocardiogram were normal. The trans-esophageal echo (TEE) showed a 40×30 mm hypovascular lesion in the left atrium. The mass had been originated from the base of the anterior leaflet of the mitral valve with an obstructive effect. The mass was resected in June 2017. The histopathology showed a high-grade telangiectatic type of osteosarcoma. Looking for the primary tumor, post-surgical whole-body bone scan with Technetium-99m methylene diphosphonate ([99mTc]Tc-MDP) showed no possible primary origin or metastasis.

The patient refused to receive any further therapy. Follow-up evaluation was performed in July 2018 with echocardiography and [18F]FDG PET/CT which were unremarkable.

The patient was symptom-free. In July 2020, follow-up TEE showed two lesions located on the mitral valve. Subsequently, he underwent CT angiography, which revealed a polypoid mass measuring about 50×38mm with internal vascularity in the left atrium, involving its medial side and interatrial septum along with another mass (43×32mm) in the lateral side, extending to the left ventricle.

Finally, he referred to our center in August 2020 for precise re-staging. The patient complied with the low-carbohydrate, high protein regimen for 24 hours and fasted for 12 hours before the imaging. He was also recommended to avoided heavy physical activity the day before the scan. Propranolol and alprazolam were administered before [18F]FDG injection to reduce possible brown fat uptake. PET/CT was performed 60 minutes after intravenous injection of 360 MBq [18F]FDG using Biograph 6 PET/CT scanner (Siemens Medical Solutions, Erlangen, Germany). Fasting blood glucose level was 84 mg/dL at the time of injection. PET acquisition was performed from the vertex to mid-thigh in a caudocranial manner, in 3D mode with 3 min per bed position after a non-diagnostic CT (80 mAs, 80-130 keV, pitch of 0.8 and slice thickness of 5 mm) acquisition, which used for attenuation correction and localization. The PET images were reconstructed using CT for attenuation correction and ordered subset expectation maximization algorithm (four iterations and eight subsets).

The PET/CT images demonstrated two masses in the left atrium revealing moderate intensity and inhomogeneous [18F]FDG uptake with calcified foci, indicating disease recurrence (measuring about 32×38mm and 43×48mm). The lesions showed invasion to the lateral wall of the left atrium (Figure 1). Otherwise, no metabolically active lesion was noted through the body. Subsequently, the patient underwent surgical excision (Figure 2), and he accepted to receive chemotherapy.

DISCUSSION

Primary cardiac osteosarcomas are uncommon tumors, accounting for only 3–9% of all cardiac sarcomas. These tumors are diagnosed at a mean age of 43.6 (range 14–77 years) [11, 3], usually late in the disease stage [9]. They almost always arise from the left atrium where is the usual region for myxoma [12, 13].

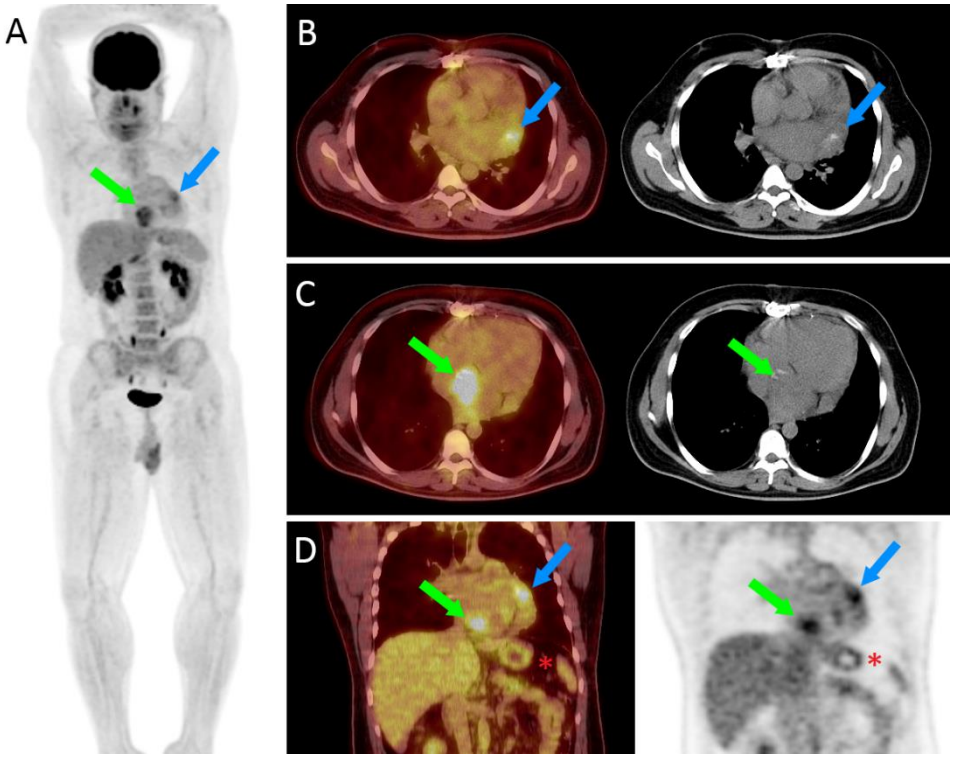

Fig 1. A 38-years-old man with cardiac osteosarcoma. a: Maximum intensity projection (MIP) image shows two intra-cardiac foci of [18F]FDG uptake in the heart. b and c: Trans-axial views (fused and CT) show two hypermetabolic intra-cardiac tumors with areas of calcification revealing inhomogeneous [18F]FDG uptake in the lateral wall (blue arrow) and interatrial septal wall (green arrow) of the left atrium, indicating recurrence. Mild uptake in the sternum is due to the prior sternotomy. d: Coronal views (fused and PET-only) demonstrate the same masses. Moreover, there is physiological [18F]FDG uptake in the stomach (*) and proximal duodenum.

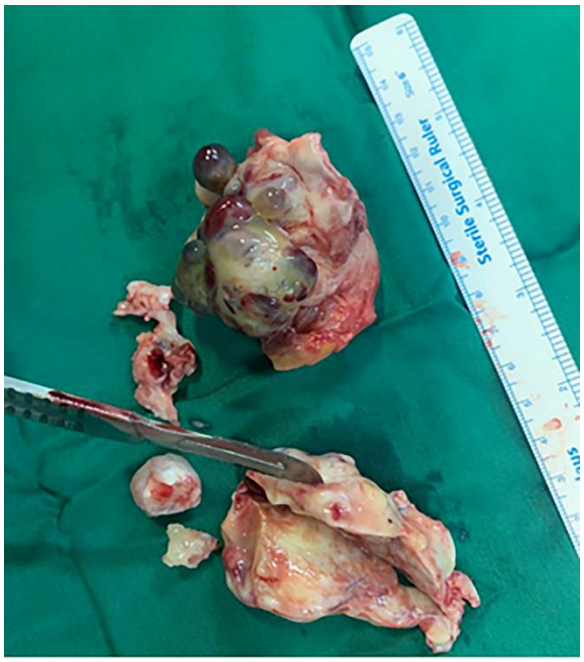

Fig 2. Two tumoral lesions were excised from the left atrium. The histopathology was compatible with osteosarcoma. Also, there was evidence of focal invasion to the myocardial fibers. The cytology of the pericardial fluid was negative for malignancy.

Tumor location, its degree of obstruction and invasion, as well as the presence of intra-cardiac clot, are responsible for the clinical presentation of cardiac tumors [1, 3, 11]. Dyspnea is the most presentation among these patients, which is secondary to the obstruction of the mitral valve [11]. Our patient primarily presented with chest discomfort and dyspnea after exercise.

There are several imaging modalities, employed for diagnosis of cardiac neoplasms [1, 10, 12]. Currently, echocardiography is the first choice, which is more available and has no ionizing radiation, but it cannot reliably differentiate the malignant nature of the lesions [14-16]. Moreover, it is not helpful in the evaluation of the extracardiac extension, provides poor tissue characterization, and usually is non-diagnostic [17]. Certain echo-graphic characteristics of cardiac masses, including large size, immobility, multicentricity, neovascularity, calcification, and invasion into the surrounding structures (septum, valves, or pulmonary veins) may suggest a malignancy; however, these features may not be always present within a malignant lesion [1]. CT and MRI can provide precise information about the anatomical extension and intrinsic characteristics [12].

CT may show dense calcifications within a cardiac mass or aggressive growth pattern; nevertheless, calcification could be minimal, and the tumor might be mistaken as a benign lesion in the early stages [12]. While MRI is useful in the characterization of the soft-tissue components, it cannot show calcification. It also is susceptible to cardiac motion [12]. Moreover, MRI might be contraindicated in some cases [18].

[18F]FDG PET/CT imaging is able to assess metabolic activity. Rahbar et al. evaluated 24 patients with cardiac tumors with [18F]FDG PET/CT [10]. They reported a sensitivity of 100%, specificity of 86%, and an accuracy of 96% for the detection of malignancy (either primary or secondary). Morphologic imaging reached a sensitivity of 82%, a specificity of 86%, and an accuracy of 83% [10]. In another study by Lemasle et al., the transthoracic echocardioghraphy, MRI, CT, and 2-[18F]FDG PET/CT showed the accuracy of 80%, 87%, 86% and 89%, respectively, for the diagnosis of benign cardiac masses. Additionally, the accuracy of these modalities were 82%, 81%, 82% and 94%, respectively, for malignant lesions [19].

Primary cardiac sarcomas have a high incidence of local recurrence and systemic metastasis, contributing to the poor prognosis [20, 11]. Metastasis to the right heart and distant metastasis to skin, brain, thyroid, lung, stomach, liver, kidney, muscle, and bone have been reported [21]; therefore, [18F]FDG PET/CT as a whole-body modality is helpful not only in the assessment of the primary lesion but also in the detection of possible distant involvement [10]. From another perspective, if there is any doubt on whether the left atrial mass is a metastatic lesion, [18F]FDG PET/CT is helpful to find the primary origin [22]. Although, [18F]FDG PET/CT has been rarely used for diagnosis of the primary cardiac osteosarcoma, it has successfully depicted the hypermetabolic primary tumor and its metastases [6, 7, 23].

CONCLUSION

In summary, although cardiac sarcomas are rare, they are the most common primary cardiac malignancies. Myxomas and metastases are most important in differential diagnoses. [18F]FDG PET/CT has a significant role in the evaluation of sarcomas and could be of value in the staging and re-staging of the primary cardiac osteosarcomas. Moreover, since the metastatic lesions to the heart are more common than the primary tumors, [18F]FDG PET/CT is a relevant whole-body imaging modality to demonstrate the possible primary malignancy.

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