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Case Report

Small direct right ventricular cardiac metastasis of osteosarcoma in a 10-year-old boy affirmed by cardiac MRI

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ABSTRACT

Background: Metastatic osteosarcoma with direct cardiac involvement is an exceptionally rare finding. Reliable detection of cardiac metastases is known to be crucial for patients therapy and prognosis.

Case Summary: In a 10-year-old boy affected by osteosarcoma of the left femur, a baseline Fluorine-18-fluorodeoxy-glucose positron emission tomography/computed tomography (^{18}F-FDG PET/CT) was performed to assess the full extent of disease. Whole-body scan detected numerous bone metastases together with a single pulmonary metastasis. Moreover, increased tracer uptake was observed in the intracavitary right cardiac ventricle in the position of a subtle spot of calcification. Because of nondetectability of a cavitary lesion on echocardiography, cardiac magnetic resonance imaging (CMRI) examination was performed to evaluate cardiac ^{18}F-FDG PET/CT finding. CMRI revealed a small nodule in the right ventricle attached to the trabeculae, highly suspicious of a direct cardiac metastasis. After 4 cycles of chemotherapy, complete regression of tracer uptake of the lesion was observed on a follow-up ^{18}F-FDG PET/CT scan.

Conclusion: CMRI is able to detect even small, clinically asymptomatic cardiac metastases in young patients affected by osteosarcoma.

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Introduction

Osteosarcoma is the most common primary malignant bone tumor in children, adolescents and young adults. Osteosarcomas are mostly located in the metaphyseal regions of long bones [1,2]. According to location, degree of differentiation and histological variants they can be divided into intramedullary (IMOS), parosteal (POS), periosteal (perOS) and high grade surface (HGSO) subtypes [3]. Most common metastatic sites are lung, bone and bone marrow, respectively [1,2]. Although osteosarcoma is known to spread hematogenously, cardiovascular structures are rarely affected [4,5].

Cardiovascular involvement can be subtle and therefore remains a challenge even to modern cross-sectional imaging techniques such as Computed Tomography (CT), Positron Emission Tomography/Computed Tomography (PET/CT) or Magnetic Resonance Imaging (MRI). On the other hand, early recognition of these findings can improve staging accuracy, allowing better determination of appro-priate cancer therapy and prognosis [4].

In the following case, we describe PET/CT and MRI findings of a small intracavitary cardiac lesion in a young male patient affected by metastatic osteosarcoma.

Case presentation

A ten-year-old boy, the only child of healthy Asian parents, was admitted for evaluation of increasing pain in the left knee for the past months. Main clinical finding was a swollen left knee, slightly painful to touch and unable to flex and bear full weight, when walking. Clinical examination of the heart, abdomen and lung was free of pathological findings. He was on the 50th percentile for his weight and height. His previous medical history was unremarkable, and the family had no history of cancer. Laboratory analysis showed a normal blood count and normal serum electrolytes. Lactate dehydro-genase was elevated to 532 U/l and uric acid slightly elevated to 5.2 mg/dl.

MRI of the left knee demonstrated an expansive tumorous lesion in the distal femoral metaphysis together with skip lesions in the adjacent diaphyseal and epiphyseal regions, suspicious of a malignant bone tumor. Subsequent open tissue biopsy enabled histopathological diagnosis of a highly malignant intramedullary osteosarcoma (IMOS).

For evaluation of metastatic disease, a baseline Fluorine-18-fluorodeoxyglucose (^{18}F-FDG) PET/CT was conducted using a hybrid scanner (Biograph 40 PET/CT, Siemens Healthcare, Erlangen, Germany). The patient was studied after fasting for 6 hours. 89 MBq ^{18}F18-FDG was injected intravenously 60 minutes prior to the aquisition. Blood glucose level at the time of application was 66 mg/dl. Examination was conducted as a whole-body scan, using a low dose mode for the co-registered unenhanced CT (100 kV, 84 ref. mAs). Pathologically increased glucose metabolism was seen in the region of primary tumor (left distal femur, SUV_{max} 8.7). Additional focal uptake indicating metastatic lesions was detected in the left proximal femur (SUV_{max} 4.2), right distal femur (SUV_{max} 6.1), left tibia (SUV_{max} 5.5), head of the left humerus (SUV_{max} 7.8) and in the lower lobe of the left lung (SUV_{max} 1.9). Moreover ^{18}F-FDG PET/CT demonstrated increased tracer uptake (SUV_{max} 5.3) in the right cardiac ventricle in the position of a subtle spot of calcification, visible on the co-registered CT (Figs. 1A and B).

In order to confirm the high suspicion of direct intracardiac metastasis, echocardiography was conducted independently by two experienced sonographers (paediatrician >15 years of expertise, paediatric cardiologist >20 years of expertise). Both examinations did not reveal any pathological findings. In particular, there was no evidence of a circumscribed lesion inside the right ventricular cavity.

As a consequence, decision was made to perform a Cardiac MRI (CMRI). The examination was conduc-ted using a 1.5 Tesla MR scanner equipped with high-performance gradients (Magnetom AERA, Siemens, Erlangen, Germany). For functional analysis, the imaging protocol included retrospectively gated, electrocardio-graphically triggered balanced steady-

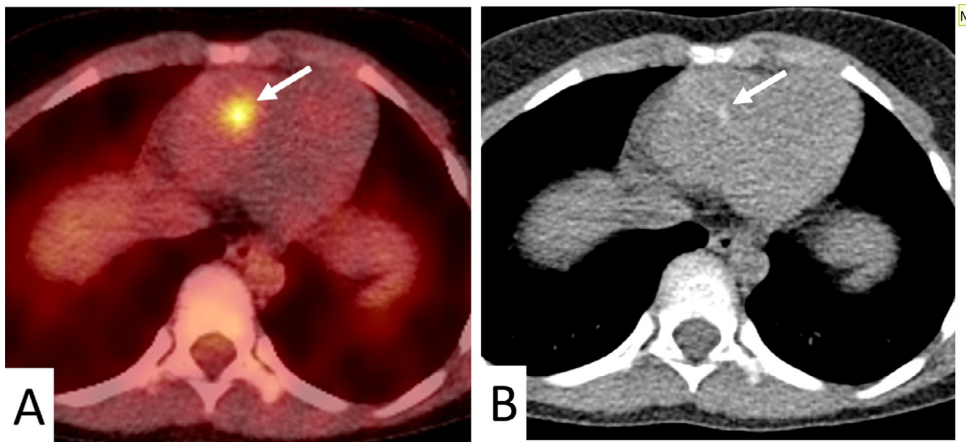

Fig. 1 – Baseline ^{18}F-FDG PET/CT. Fused axial PET/CT image (A) shows focally increased glucose metabolism in the right cardiac cavity (arrow). In the same position, a subtle spot of calcification is visible in the co-registered low dose CT (B).
18F-FDG PET/CT, Fluorine-18-fluorodeoxy-glucose positron emission tomography/computed tomography

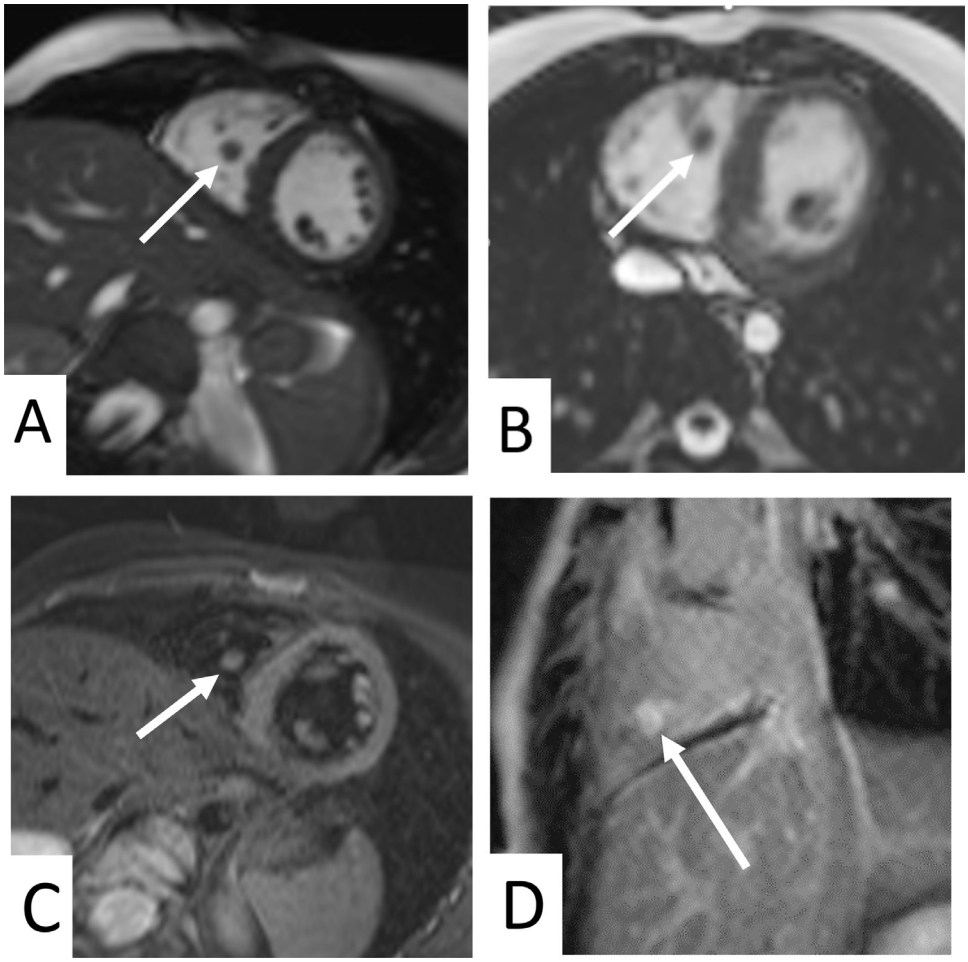

Fig. 2 – CMRI. Balanced steady-state free-precession (BSSFP) cine images in SAX (A) und axial (B) plane confirm the presence of a right intracavitary nodular lesion with a diameter of 6 mm (arrow). T2w TSE image in SAX plane (C) reveals isointense signal of the lesion in comparison to ventricular wall. Intense contrast agent uptake can be seen in sagittal postgadolinium MDE image (D). CMRI, cardiac magnetic resonance imaging; MDE, myocardial delayed enhancement; SAX, short-axis; TSE, turbo spin echo.

state free precession (bSSFP) cine se-quences in axial as well as short-axis (SAX) views. Moreover T1- and T2-weighted turbo spin echo (TSE) sequences with and without fat saturation (FS) were performed at the region of the pathologic right ventricular finding. Finally, first pass myocardial perfusion imaging (FPP) and myocardial delayed enhancement imaging (MDE) was conducted after intravenous administration of 4 ml Gadobutrol (Gadovist 1.0 mmol/ml, Bayer, Leverkusen, Germany). Sequences were acquired during breath holding with a slice thickness of 4-6 mm.

BSSFP cine sequences demonstrated a nodular lesion with a diameter of 6 mm in the middle cavitary section of the right ventricle next to the anterior papillary muscle (Fig. 2A, B). Probably attached to the right ventricular trabeculae, movement of the lesion coincided with right ventricular contraction. T1- and T2-weighted TSE images revealed nearly isointense signal of the nodule when compared to adjacent myocardium (Fig. 2C). There was no evidence of fatty components when FS was used. Postgadolinium FFP images did not show considerable contrast agent uptake of the nodule, whereas there was intense enhancement in MDE images (Fig. 2D).

Because of corresponding localisation of a circumscribed lesion on CMRI and tracer uptake on ^{18}F-FDG PET/CT, the diagnosis of a direct right ventricular metastasis was affirmed.

We implanted a double lumen Hickman-catheter and started the patient on chemotherapy according to European and American Osteosarcoma Study-1/Cooperative Osteosarcoma Study Group (EURAMOS-1/COSS) with Adriamycin, Cisplatin, and Methotrexate.

After 4 cycles of chemotherapy, complete regression of intracardiac tracer uptake was observed on a follow-up ^{18}F-FDG PET/CT scan.

Discussion

In our patient, ^{18}F-FDG PET/CT demonstrated pathological tracer uptake in the right cardiac ventricle. Because of con-

fusing nondetectabilty on echocardiography, CMRI was conducted, affirming diagnosis of a direct cardiac metastasis. To our knowledge, imaging of such a small intracavitary metastasis in a child using cMRI has not yet been described in the literature.

Yedururi et al. retrospectively reviewed imaging findings of patients with osteosarcoma. At their institution, 20 patients with osteosarcoma and cardiovascular involvement were identified on CT or MRI by 2 radiologists during their routine practice within a time period of 6 years The locations of metastases were systemic veins draining the primary tumor ($n = 6$) or extrapulmonary metastases ($n = 3$). Moreover, 16 of 20 patients showed metastases in the pulmonary arteries, including patients whose tumor further extended into pulmonary veins and the left atrium ($n = 3$). Only 3 of the 20 patients showed noncontiguous involvement of the heart [4]. After exclusion of all cases with tumor thrombus extending into the cardiac chambers, only very few cases with isolated ventricular metastases of osteosarcoma remain. In their extensive review of pediatric literature, Hartemayer et al. only found 63 reported cases of so called direct cardiac metastases over the past 123 years, mostly significantly larger in diameter than reported in our case [5]. For instance, Maleki et al. reported a 5 cm × 4 cm × 2 cm large lobulated metastasis in the left atrium of a 38-year-old man. He had a history of osteosarcoma in the left femur 5 years prior to his re-hospitalization. Diagnosis was made using spiral CT [6].

Interestingly, metastatic osteosarcoma to the heart was previously much more common. In the earliest autopsy review of osteosarcoma by Dorfman and Michaels in 1966, cardiac metastases were seen in nearly 20% of cases [7]. The difference between the incidence in older studies and contemporary data can be explained by earlier detectability of disease by means of modern imaging modalities as well as the use of continuously refined, multiagent chemotherapy in current treatment protocols [5].

Nowadays, PET is of major importance for diagnostic evaluation and staging of different malignant tumors. ^{18}F-FDG PET has been shown to detect metastatic lesions in various anatomic locations with high sensitivity and specificity [8]. Moreover, it is well known that PET/CT is significantly more accurate than PET alone for the detection and localization of malignant lesions, reducing false-positive and false-negative results, thus improving the staging of patients [9]. In our patient, ^{18}F-FDG PET/CT demonstrated increased intracardiac tracer uptake together with a subtle spot of calcification.

CMRI findings of direct cardiac metastases in patients affected by osteosarcoma are rarely described in the literature. Yamagishi et al. reported CMRI appearance of a huge metastasis in the right ventric-ular outflow tract with extensive invasion of the ventricular free wall in a 30-year-old woman [10]. In 2011, Ahn et al. decribed the extremely rare case of a primary cardiac osteosarcoma in the left atrium of a 47-year-old woman [11]. In our patient, we were able to identify the subtle intracavitary metastasis using a dedicated CMRI examination protocol. The latter should include bSSFP cine imaging as well as T1- and T2-weighted sequences. Moreover, FPP and MDE should be performed.

Conclusion

This report suggets that CMRI is able to detect even small, clinically asymptomatic cardiac metastases in young patients affected by osteosarcoma when a dedicated examination protocol is used.

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