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"Double imaging" for the diagnostic work-up of alveolar soft part sarcoma with Tc-99m MIBI

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The authors report a case of alveolar soft-part sarcoma with lung metastases demonstrated by "double imaging" with Tc-99m HDP and Tc-99m MIBI. The tumor originated in the soft tissue with direct invasion to the right scapula, which was hypoactive on bone scan and hyperactive on Tc-99m MIBI images. A focus of dense accumulation of Tc-99m MIBI in the lungs, suggesting metastasis was also demonstrated.

Key words: sarcoma, bone scan, Tc-99m MIBI, Tc-99m HDP, double imaging

INTRODUCTION

ALVEOLAR SOFT PART SARCOMA (ASPS) was described as a distinct entity in 1952. ASPS mainly affects young adults with a slightly higher prevalence in female patients. It arises in the extremities, trunk and head and neck region. ASPS usually presents as a slow-growing, painless mass nearly without functional impairment. Metastases are frequent, and are mainly to the lungs, brain and bones.[1] In rare instances, there is erosion or destruction of the underlying bone.[2] Tc-99m HMDP, Tc-99m MDP, Tc-99m(V) DMSA and Ga-67 were used for imaging of alveolar soft-part sarcoma and its metastases.[3,4] Tc-99m MIBI is now commonly used for tumor imaging.[5–8] In this article, we report a "cold" bone lesion of ASPS in the right scapula demonstrated by a new technique, double imaging with two different radiopharmaceuticals.

CASE REPORT

A 46-year-old man had a painful mass ($14 \times 12 \times 10$ cm) in the right scapular region for a year. The patient was assessed by radiological methods including computerized tomography (CT) of the thorax, which revealed a solid mass of $16 \times 13 \times 11$ cm showing destruction of the right scapula. In addition, a few nodules in the left lung and one nodule in the right lung were shown (Fig. 1). Magnetic resonance imaging (MRI) was carried out by using a 0.5 Tesla MR scanner (GE Medical Systems, VECTRA, Milwaukee, USA). It showed a multiseptated, multilobulated mass in the right axillary fat space with slight contrast enhancement invading the infraspinatus muscle (Fig. 2). The patient was then evaluated scintigraphically for the primary tumor, bone invasion and distant metastasis. Whole body and static spot images after three hours of injection of 740 MBq Tc-99m HDP were acquired with an Elscint SPX-6 gamma camera (Elscint Ltd., Haifa, Israel). A hypoactive area with an irregular hyperactive rim was observed in the inferolateral portion of the right scapula. No other pathologic uptake was detected (Fig. 3).

Two days later the patient was injected with 740 MBq Tc-99m MIBI. Imaging started as early as 10 minutes and as late as 2.5 hours after injection. Early and delayed static spot images of posterior the thorax and whole body in anterior and posterior projections and SPECT images of the thorax were obtained with the same gamma camera. There was a prominent Tc-99m MIBI uptake in the whole mass in the right scapula, strongly suggesting a malignant nature. The multiple nodular appearance detected on CT scan (suggesting metastases in both lungs) was not seen

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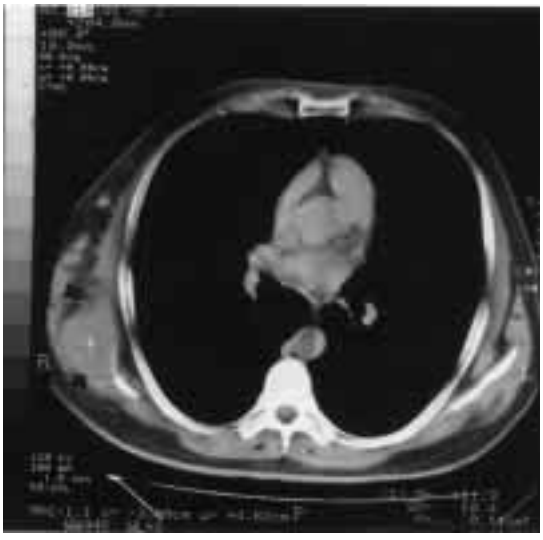
A



B

Fig. 1 CT of thorax showed a parenchymal nodule (*arrows*) in the left lung (metastasis) (A). A solid mass (*arrows*) of 16 × 13 × 11 cm in right scapular region was revealed, destructing right scapula antero-medially (*arrows*) (B).

A

B

Fig. 2 MRI of right scapular region. T1-weighted pre-contrast coronal spin-echo (A) and T2-weighted axial gradient echo (B) images. In the right axillary fat space there was a multiseptated, multilobulated mass (*large arrows*) of 14 × 12 × 10 cm, invading infraspinatus muscle (*arrows*), displacing the right subscapularis muscle antero-superiorly (*arrows*).

on either Tc-99m MIBI images or Tc-99m HDP bone scan, and a new focus of dense uptake was detected in the left lung on Tc-99m MIBI images (Fig. 4).

After radiological and scintigraphic studies, fine-needle aspiration biopsy of the mass was carried out, and ASPS was histologically diagnosed. The mass was surgically removed and alveolar soft part sarcoma was diagnosed in the specimen. Macroscopically, a poorly circumscribed, soft, nodular, white-gray tumor measuring 19 × 8.5 × 11 cm was located within the soft tissue. The tumor was composed of cells forming alveolar structures separated from each other by sinusoidal vascular channels lined by flattened epithelium. The tumor cells were large and polygonal with distinct cell borders, and contained abundant granular eosinophilic cytoplasm (Fig. 5). There were large scattered areas of necrosis. Vascular invasion was detected most prominently in the peripheral large veins on PAS staining. Cytoplasmic crystalloids were detected in tumor cells. Immunohistochemically, tumor cells were consistently negative for vimentin, desmin, actin, S-100 and cytokeratin. The focal, dense uptake of Tc-99m MIBI in the left lung did not correspond to the location of the nodular appearance detected on CT scan but it was not further investigated for confirmation because it would not significantly change the treatment strategy for this patient.

A

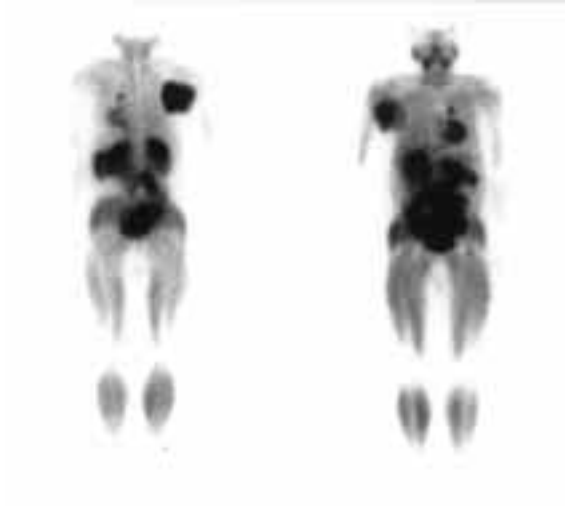
B

Fig. 3 Bone scan performed 3 hours after injection of Tc-99m HDP. Whole-body images in anterior and posterior projections (A), and spot image of thorax in posterior projection (B). No uptake was seen in the soft tissue mass. In the inferolateral portion of right scapula, there was a hypoactive region (*arrows*) with an irregular hyperactive rim. No other distant skeletal metastasis was detected.

DISCUSSION

Alveolar soft part sarcoma is a rare neoplasm with an incidence of 0.5% to 1% of all soft tissue sarcomas.[2] The tumor occurs principally in adolescents and young adults (most frequently in patients between 15 and 35 years of age). Female patients outnumber males. It usually occurs in the lower extremities in adult patients, and the head and neck region in children. It generally presents as a slow-growing painless mass almost always without functional impairment. Because of the relative lack of symptoms, it is easily overlooked, and metastasis to the lungs or brain

A



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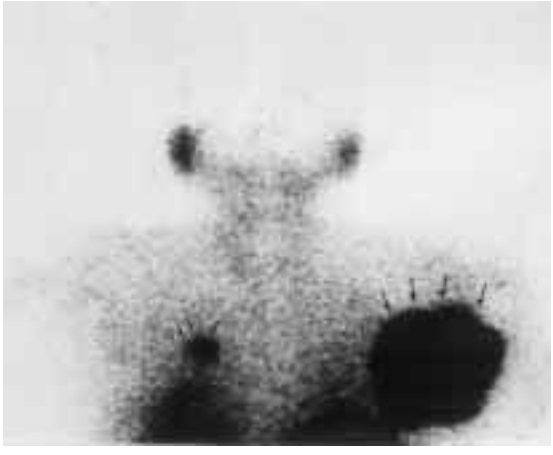

C

Fig. 4 Early whole body (A), taken 10 minutes following injection of Tc-99m MIBI, delayed whole body (B) taken 2.5 hours after injection and spot image of thorax in posterior projection taken after 40 minutes after injection (C). Marked Tc-99m MIBI uptake in primary ASPS originating from the right scapular region (*arrows*), focal uptake in left lung, near to myocardium (*arrows*).

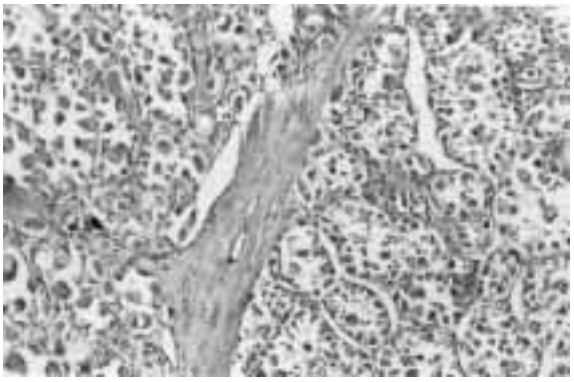

Fig. 5 Photomicrograph of the pathologic specimen showing typical nest-like pattern of tumor with large polygonal cells having abundant cytoplasm. Cellular aggregates were separated by thin walled sinusoidal vessels (Hematoxylin Eosin ×400).

is the first clinical manifestation.[1,2] Therefore the extent of the tumor should be evaluated by reliable methods. In rare instances, there is erosion or destruction of the underlying bone.

In medical literature, 3 cases of ASPS were studied scintigraphically.[3,4] Ohta et al. evaluated 2 cases of ASPS with Ga-67, Tc-99m MDP and Tc-99m(V) DMSA. According to this study, all primaries accumulated tumor-seeking agents, and images with Tc-99m(V) DMSA were the best. Skull metastasis could also be detected by all agents, but lung metastases did not take up any of the agents.[3] Hirano et al. used Tc-99m HMDP for imaging an ASPS case. Primary lesion in the right gluteal region showed intense uptake but, no metastasis accumulated the bone seeking agent Tc-99m HMDP.[4]

In this case, the primary lesion in the right scapular region eroded the underlying scapula, which was hypoactive on bone scan with an irregular hyperactive rim. Tc-99m MIBI is used for clinical trials in various tumors.[5–9] Considering its advantages over gallium-67 and thallium-201, and the cumulative experience with this agent in several types of tumors, we used Tc-99m MIBI for the evaluation of this tumor in the right scapular region. There was prominent uptake in the mass on Tc-99m MIBI images. This mismatch in two sessions of imaging using bone-seeking agent and tumor-seeking agent (double imaging) was exploited for diagnosis.

This case is of special clinical interest because, to the best of our knowledge, in previously reported cases, primary lesions of ASPS accumulated bone-seeking agents (hyperactive).[3,4] This case, the tumor consisted of several areas of necrosis, and as expected the bone-seeking agent was not taken up by the tumor tissue, causing hypoactive appearance with a hot rim (cold spot). And this is the first report describing the uptake of Tc-99m MIBI in ASPS causing a hot spot. The mismatch on bone scan (hot) and Tc-99m MIBI images (cold) in this tumor has not previously been reported as far as we know. In addition, the tumor occurs only rarely, and this tumor eroded the underlying bony structure (the scapula). It was also interesting to observe the increased uptake of Tc-99m MIBI in the tumor despite extensive necrosis, the mechanism of which cannot be explained with the previously mentioned Tc-99m MIBI tumor uptake mechanisms, all of which are based on the availability of viable tumor cells as a prerequisite of uptake: i.e. previous reports pointed out that Tc-99m MIBI was not taken up by necrotic tumor tissue or by a tumor which was previously exposed to radiotherapy.[6,10] And, from the technical point of view, one-to-one comparison of hypoactive lesion on bone scan with a hyperactive spot on Tc-99m MIBI tumor images (double imaging) can be a new clinical method for the diagnostic work-up of hypoactive bone lesions, which significantly decreases the sensitivity of bone scan. Previously published reports describing the filling-in of the cold lesions on Tc-99m MIBI images also suggest the possible use of this technique in other patients with tumors (e.g. renal cell carcinoma and larynx carcinoma) possibly causing cold bone lesion, which is still a clinical challenge for the imaging physician, but the use of "double imaging" as a clinical tool in routine practice requires further confirmation in a larger group of patients.[11]

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