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Clinical usefulness of In-111 chloride and Tc-99m Sn colloid scintigraphy in the diagnosis of intrathoracic extramedullary hematopoiesis

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We report a case of intrathoracic extramedullary hematopoiesis associated with hemolytic anemia. While the paravertebral localization of the lesions demonstrated on CT and MRI was suspicious of intrathoracic extramedullary hematopoiesis, In-111 chloride and Tc-99m Sn colloid bone marrow scintigraphies showing a bone marrow element of the lesion were useful to confirm the diagnosis.

Key words: intrathoracic extramedullary hematopoiesis, In-111 chloride, Tc-99m Sn colloid

INTRODUCTION

EXTRAMEDULLARY HEMATOPOIESIS is the formation and development of blood cells in tissues other than bone marrow. It occurs in chronic hemolytic states, ineffective erythropoiesis, and myeloproliferative disorders as a compensatory process of anemia. The most common sites of involvement include the liver, spleen, and lymph nodes.[1,2] We report a case of intrathoracic extramedullary hematopoiesis associated with hemolytic anemia, in which In-111 chloride and Tc-99m Sn colloid bone marrow scintigraphies were instrumental for the diagnosis.

CASE REPORT

A 45-year-old woman with a 13-year history of hemolytic anemia was admitted for further examination of paravertebral masses on a chest radiograph performed as part of an annual checkup. She looked anemic but complained of no chest symptoms. Hepatosplenomegaly was noted on physical examination. The red blood cell count was $289 \times 10^4/\mu l$, the hemoglobin level was 9.0 g/d*l*, the serum ferritin was 220.8 ng/m*l* and the haptoglobin was less than 6.2 mg/d*l*. The Ham test was positive. The white blood cell count and platelet count were normal.

Lower thoracic CT sections showed bilateral smoothly marginated paravertebral masses that did not erode the adjacent bone. The larger mass was located on the right side and was $8 \times 5 \times 3$ cm in size. The mass on the left side was $5 \times 4 \times 2$ cm in size. The CT density of the masses was homogeneous and similar to that of muscles (Fig. 1). MRI depicted the masses intermediate signal intensity on T_1- and T_2-weighted images, and the contrast enhancement of the masses was moderate (Fig. 2).

As she had been suffering from hemolytic anemia and the masses were formed at the bilateral paravertebral regions, we suspected the masses to be extramedullary hematopoietic foci. To confirm the diagnosis, we performed bone marrow scintigraphy using In-111 chloride and Tc-99m Sn colloid, which revealed uptake in the paravertebral masses, confirming the diagnosis of intrathoracic extramedullary hematopoiesis (Fig. 3). The scans also showed expansion of the bone marrow into the distal extremities, compatible with marrow hyperplasia secondary to chronic hemolysis.

DISCUSSION

Intrathoracic extramedullary hematopoiesis is an unusual condition occurring predominantly in the posterior mediastinum.[1,2] As most of intrathoracic extramedullary hematopoiesis occurs in patients with thalassemia or hereditary spherocytosis[3] and these diseases are rarely seen in Japanese, intrathoracic extramedullary hematopoiesis is rarely encountered in Japan.[4] Lawson et al. theorized that extramedullary hematopoiesis arises from the extrusion of proliferating marrow through the cortex

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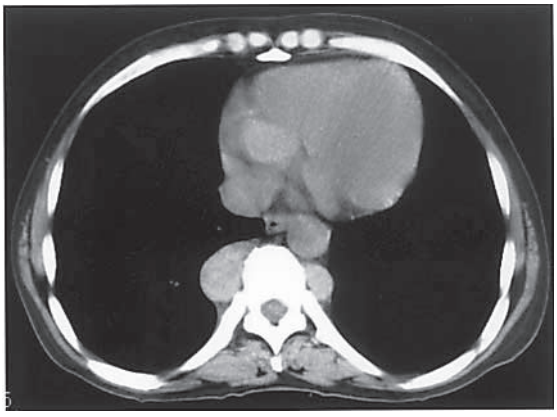
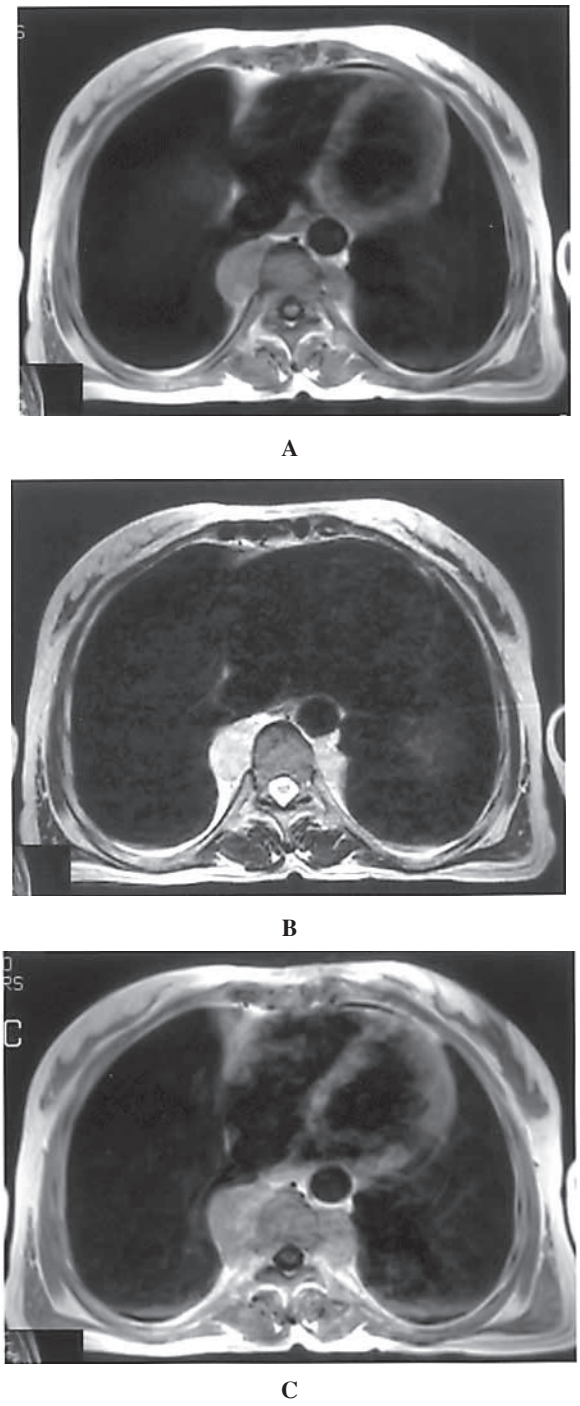


Fig. 1 Thoracic CT showed bilateral paravertebral masses with smooth margin and isodensity to muscles.

into a subperiosteal location, which explains its presence in a paravertebral or presacral location.[5] Although intrathoracic extramedullary hematopoiesis may induce spinal cord compression, pleural effusion, hemothorax or dyspnea, secondary to involvement of lung interstitium,[3,6,7] it is generally asymptomatic and requires no therapy. In addition, thinking of the risk of hazards of biopsy due to its highly vascular property, it is desirable to establish the diagnosis noninvasively.[2] For these reasons, various imaging procedures are advocated to establish the diagnosis of intrathoracic extramedullary hematopoiesis, including CT, MRI, or radionuclide bone marrow scanning.

Intrathoracic extramedullary hematopoietic foci are typically seen as smoothly marginated masses at the paravertebral region on CT and MRI.[6] Active recent hematopoietic lesions show soft tissue CT density and intermediate signal intensity on both T_1- and T_2-weighted MR images. Contrast enhancement study shows some enhancement in the active hematopoietic extramedullary lesion. Inactive lesions show increased CT density due to iron deposit or low density due to fatty infiltration, and high signal intensity on both T_1- and T_2-weighted MR images due to fatty infiltration or low signal intensity on both T_1- and T_2-weighted MR images due to iron deposition.[8] As the lesions in our patient showed soft tissue density on CT and intermediate signal intensity on T_1- and T_2-weighted MRI, they were considered to be consistent with actively hematopoietic foci.

However, although the density on CT or signal intensity on MRI is helpful to speculate as to the pathologic features of the mass, they do not offer definitive evidence that the lesion consists of bone marrow. In addition, the variability of the appearance in extramedullary hematopoietic foci on CT or MRI itself may confuse the diagnosis. For the differential diagnosis of intrathoracic extramedullary hematopoiesis, various paravertebral masses have been included such as neurogenic or mesenchymal tumors, lymphomas, aneurysms, meningoceles,



A

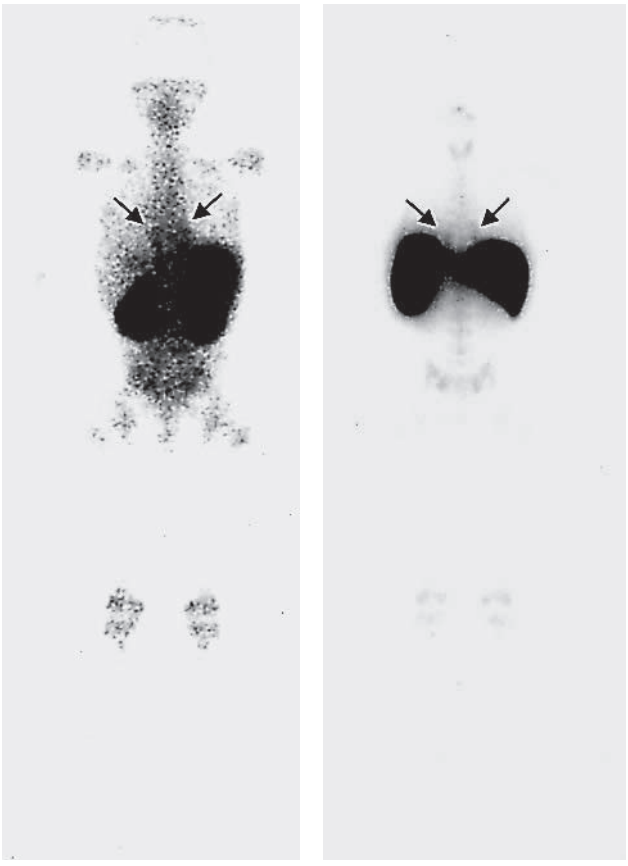
B

C

Fig. 2 Axial T_1- and T_2-weighted images (A and B, respectively) revealed that the masses had intermediate signal intensity on both scans and were moderately enhanced (C).

and hemangiomas.[1,2]

Bone marrow scintigraphy has been used to know the physiologic function of bone marrow and also employed for the diagnosis of extramedullary hematopoiesis. There have been a number of reports indicating the usefulness of In-111 chloride and Tc-99m-labeled colloid in extramedullary hematopoiesis.[1,2,9,10] In-111 chloride originally



A B

Fig. 3 In-111 chloride (A) and Tc-99m Sn colloid (B) scintigraphies (posterior views) showed radiotracer uptake in the masses (*arrows*) demonstrated on CT and MRI, and the diagnosis of intrathoracic extramedullary hematopoiesis was confirmed. Expansion of the bone marrow into the distal extremities was also revealed.

was believed to localize in red cell precursors. However, it is now known that it may localize in both reticuloendothelial and hematopoietic elements in the bone marrow.[10] The distribution of erythropoietic and reticuloendothelial cell lines is similar in the bone marrow, with the exception of aplastic anemia or after irradiation.[11] Tc-99m-labeled colloid, such as Tc-99m Sn colloid, we employed for the present patient, is taken up by reticuloendothelial system and thus traces the erythropoietic marrow. Since the full bone marrow components of erythroid, reticuloendothelial, and megakaryocytic cells are present in the areas of extramedullary hematopoiesis, both reticuloendothelial system and erythropoietic agents can be used.[9]

It must be kept in mind that In-111 chloride and Tc-99m-labeled colloid may accumulate in tumors.[12,13] In particular, In-111 chloride concentrates in various tumors, as it initially drew attention as a tumor seeking agent.[14] However, to our knowledge, there has been no report concerning tumors taking up both In-111 chloride and Tc-99m Sn colloid.

In our patient the typical paravertebral localization of the lesions demonstrated on CT and MRI suggested intrathoracic extramedullary hematopoiesis, which was confirmed based on the accumulation both of In-111 chloride and Tc-99m Sn colloid in the masses. Also bone marrow scintigraphies using these tracers, showing bone marrow expansion into the distal extremities, provided other useful information about the status of erythropoietic marrow.

CONCLUSION

We reported a case of intrathoracic extramedullary hematopoiesis associated with hemolytic anemia. Bone marrow scintigraphies performed with In-111 chloride and Tc-99m Sn colloid were useful to determine the diagnosis.

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