A case report on $^{111}$In chloride bone marrow scintigraphy in management of adrenal myelolipoma

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Abstract
Rationale: Adrenal myelolipoma is a benign hormone-inactive tumor composed of hematopoietic tissue and mature adipose tissue. Because this tumor tends to be rich in fat, in many cases it can be diagnosed based on computed tomography (CT) or magnetic resonance imaging (MRI) findings alone. However, in the presence of much necrosis, calcification and hematopoietic tissue and/or the absence of much fat, and in cases with tumor apoplexy, this tumor becomes difficult to differentiate from other tumors. In such cases, a bone marrow scan may be informative as a non-invasive imaging diagnostic method for preoperative differentiation of the tumor and determination of the method for the surgical treatment. We herein report a case of huge adrenal myelolipoma with the non-adipose portion identified using an $^{111}$Indium chloride ($^{111}$InCl$_3$) bone marrow scan.

Patient concerns: A 69-year-old woman was referred to our hospital because of a left peritoneal mass detected on a medical checkup. Abdominal CT revealed a mass measuring 14.3 cm in diameter located between the left kidney and the left adrenal gland, which showed coexistence of fat and soft tissue densities.

Diagnoses: A bone marrow scan is a nuclear medicine examination to assess hematopoietic activity. To avoid excessive resection of the tumor, we thought that a bone marrow scan could be applied for differentiation between myelolipoma and retroperitoneal liposarcoma by evaluating the hematopoietic activity of the tumor. Tumor enucleation was performed, and pathological examination showed a diagnosis of adrenal myelolipoma.

Intervention: The patient was treated with laparoscopic enucleation.

Outcomes: No metastatic recurrence was found during 8 months of follow-up.

Lessons: Diagnosis of myelolipoma by CT and MRI becomes difficult in the presence of a high volume of hematopoietic tissue. In such cases, a bone marrow scan may be informative as a non-invasive imaging diagnostic method for preoperative diagnosis of the tumor and determination of the method of surgical treatment.

Abbreviations: $^{111}$InCl$_3$ = $^{111}$Indium chloride, CT = computed tomography, MRI = magnetic resonance imaging.

Keywords: adrenal gland, bone marrow scan, myelolipoma

1. Introduction
Adrenal myelolipoma is a benign hormone-inactive tumor composed of hematopoietic tissue and mature adipose tissue.$^{[1,2]}$ This tumor tends to be rich in fat.$^{[3,4]}$ Usually it is not difficult to diagnose adrenal myelolipoma, but differential diagnosis between myelolipoma and retroperitoneal liposarcoma becomes difficult when a huge retroperitoneal tumor is involved with the adrenal gland. Standard treatment of liposarcoma is wide resection, including the organ with which the tumor is in contact, to reduce the risk of local recurrence.$^{[5]}$ However, the standard treatment of myelolipoma is enucleation.$^{[6]}$ Therefore, the preoperative diagnosis is important for determining the method of surgical treatment. These tumors are similar in terms of including fat on computed tomography (CT) or magnetic resonance imaging (MRI), but liposarcoma does not include hematopoietic components. A bone marrow scan is a nuclear medicine examination to assess hematopoietic activity. Myelolipoma is considered more likely to show an uptake in $^{111}$Indium chloride ($^{111}$InCl$_3$)$^{[6]}$ if it includes hematopoietic tissue. Therefore, to avoid the excessive resection of the tumor, we considered that a bone marrow scan could be applied to evaluate its hematopoietic activity.

We experienced a case of huge adrenal myelolipoma detected incidentally, and $^{111}$InCl$_3$ bone marrow scan was informative as a non-invasive diagnostic imaging method for the preoperative diagnosis and determination of the method of surgical treatment.
consent was given by the patient on each occasion of diagnostic examination and therapeutic procedure, as well as for the publication of this case report.

3. Case report
A large tumor was incidentally discovered around the left kidney of a 69-year-old woman by ultrasonography performed during a medical checkup. Abdominal CT showed a mass measuring 14.3 cm in diameter located between the left kidney and the left adrenal gland, along with fat and soft tissue densities (Fig. 1A). The portion of adipose tissue showed a CT attenuation value of $-69.1$ H.U. on non-contrast CT, and the portion of non-adipose tissue showed a CT attenuation value of $4.6$ H.U. on non-contrast CT. No apparent tumor calcification was seen. Retroperitoneal liposarcoma and adrenal myelolipoma were considered as possible diagnoses of the tumor. To exclude a diagnosis of retroperitoneal liposarcoma, $^{111}$InCl$_3$ bone marrow scan was performed as a non-invasive examination using single photon emission computed tomography with CT. Fusion images (color-scale scintigram and gray-scale CT image) revealed faint but definite accumulation (arrow) that corresponded to the non-adipose part of the tumor (Fig. 1B: coronal and Fig. 1C: axial views). The accumulation indicated the presence of hematopoietic tissue in the tumor. Therefore, this tumor was more likely an adrenal myelolipoma. Because the tumor was large despite being asymptomatic, laparoscopic enucleation was performed. Histopathologically, the tumor consisted of mature adipocytes admixed with hematopoietic components, including myeloid, erythroid, and megakaryocytic elements (Fig. 1D), arising from a normal adrenal gland. The pathological diagnosis confirmed the tumor to be adrenal myelolipoma. No metastatic recurrence was found during 8 months of follow-up.

4. Discussions
Adrenal myelolipoma is mainly a non-functioning benign tumor that is mixed with adipose tissue and hematopoietic tissue consisting of erythroblasts and the myeloblasts. Its occurrence is relatively rare and is reported by autopsy with a frequency of 0.08% to 0.8%. However, in recent years, reported cases have increased as a result of the development and widespread use of imaging techniques. Generally, it is not difficult to diagnose myelolipoma, but becomes difficult to differentiate between myelolipoma and retroperitoneal liposarcoma by CT and MRI when the tumor is large. There is 1 report of a diagnosis of myelolipoma after wide excision by preoperative diagnosis of the liposarcoma.

A bone marrow scan is a nuclear medicine examination to assess hematopoietic activity. The radiopharmaceuticals used for bone marrow scan accumulate in the reticuloendothelial system or hematopoietic tissue. The former requires a $^{99m}$Tc-Sn colloid...
using the phagocytic activity of reticuloendothelial cells. About this radionuclide, the high accumulation in the liver and spleen is a problem, and myeloid accumulation is not high. The latter is used for the evaluation of the hematopoietic system based on radionuclide uptake by the bone marrow erythroblasts using $^{111}$InCl$_3$.[6] The latter is often used for bone marrow scans in Japan. Myelolipoma is considered more likely to take in this radionuclide if it includes hematopoietic tissue.

The huge adipose mass that we treated extended to the inferior pole of the kidney from the left adrenal gland, and this tumor appeared to be a liposarcoma. Standard treatment of liposarcoma is wide resection, including the organ with which the tumor is in contact, to reduce the risk of local recurrence.[13] However, the standard treatment of myelolipoma is enucleation of the tumor. Therefore, the preoperative diagnosis is important for determining the method of surgical treatment. Because myelolipoma has a bone marrow component with wide variations, the $^{111}$InCl$_3$ bone marrow scan was performed for the evaluation of the hematopoietic element of the tumor. As far as we know, such a diagnosis using an $^{111}$InCl$_3$ bone marrow scan has not been reported previously. We were able to avoid excessive resection of the tumor by confirming the accumulation in the tumor using the bone marrow scan as a non-invasive diagnostic imaging method.

For a bulky tumor, the presence of much necrosis, calcification, and hematopoietic tissue and/or the absence of much fat, and in cases with tumor apoplexy, adrenal myelolipoma becomes difficult to differentiate from other tumors, such as liposarcoma. The possibility of malignancy increases with the size of the adrenal tumor. Even if the patient is asymptomatic, surgical removal is recommended when an adrenal tumor is ≥4 cm.[8–11] Furthermore, surgical resection would need to be considered in the presence of symptoms such as abdominal or back pain, tumor rupture, or bleeding.[12–13] Our patient reported here was asymptomatic, but because the tumor was large in size (14.3 × 11.5 cm), laparoscopic resection was performed.

Because myelolipoma is usually diagnosed based on CT or MRI findings alone, a bone marrow scan is not necessary, especially in cases where the tumor is low in hematopoietic tissue and high in adipose tissue. However, in the presence of a high volume of hematopoietic tissue, diagnosis by CT and MRI becomes difficult. In such cases, a bone marrow scan may be informative as a non-invasive diagnostic imaging method for preoperative diagnosis of the tumor and determination of the method of surgical treatment.

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