Primary bilateral adrenal lymphomas are extremely rare. To the best of our knowledge, there are only 26 cases reported in the literature. We present an additional case and review the computed tomography (CT) findings of the reported cases. There is no pathognomonic appearance of primary bilateral adrenal lymphoma on CT.

Key words: Adrenal glands, Neoplasm; Lymphoma, Computed tomography (CT)

Secondary involvement of the adrenal gland is found in 25%-35% of patients with non-Hodgkin's lymphoma [1-4]. However, bilateral primary adrenal non-Hodgkin's lymphomas are extremely rare [1-25]. There are only 26 patients with bilateral primary adrenal non-Hodgkin's lymphoma reported in the literature, with computed tomography (CT) studies in 21 cases. We herein report an additional case and review the CT features of this disease.

CASE REPORT

A 37-year-old female was sent to the emergency department because of persistent dull abdominal pain, low-grade fever, anorexia, fatigue, and body weight loss for 1 month. The abdominal pain was aggravated by inspiration and relieved by hip flexion; no radiation to the back was noted. Her past medical history was not contributory.

Physical examination revealed pale conjunctivas, mild hyperactivity of bowel sound, and abdominal tenderness. A complete blood count disclosed microcytic anemia. The serum biochemistry profile, ACTH stimulation test, and urine analysis were normal. Abdomino-pelvic CT revealed inhomogeneous tumors involving bilateral adrenal glands, with the right gland measuring about 6.4 × 5.4 cm and the left 5.5 × 3.8 cm (Fig. 1). A percutaneous ultrasound-guided biopsy of the right adrenal gland was performed. Diffuse large B-cell non-Hodgkin's lymphoma was diagnosed according to histopathological examination. Bone marrow biopsy and routine chest radiography were normal.

Primary bilateral adrenal non-Hodgkin's lymphoma without adrenal insufficiency was our impression. The patient received 2 cycles of...
intravenous chemotherapy using endoxan, adriamycin, and oncovin, and prednisolone. The general condition of this patient was good except for being neutropenic during the follow-up period of 6 months.

**DISCUSSION**

Primary bilateral adrenal lymphomas are extremely rare [1, 5]. To the best of our knowledge, only 26 cases have been reported in the literature. Since lymphomas arise from hematopoietic tissue which may be present in the adrenal glands, a primary lymphoma arising in the adrenal glands is not unexpected. Lymphomas affecting the adrenal glands are usually associated with disease at other sites, most commonly the retroperitoneal lymph nodes and ipsilateral kidney [1]. There is a male predominance in the 27 cases of bilateral primary adrenal lymphomas (M:F = 16:11). The median age was 61.9 years (range, 37 to 81 years). The present case, a 37-year-old woman, is the youngest one. Primary adrenal insufficiency was found in 17 cases (62.9%) of primary bilateral adrenal lymphoma. Vita et al. reported that destruction of more than 90% of the adrenal glands resulted in primary adrenal insufficiency [6]. Bilateral adrenal glands were entirely involved by the tumor cells which produced necrosis in our patient, but her adrenal function was still normal. The presenting symptoms and signs are non-specific, such as abdominal pain, body weight loss, fever, anemia, nausea, and vomiting.

Ultrasoundography, CT, and magnetic resonance imaging can all detect the existence of an adrenal mass, but a confirmatory diagnosis of primary adrenal lymphoma is based on pathological examinations. We summarize the CT findings of 16 cases with primary bilateral adrenal lymphoma, including our case, in Table 1. The suprarenal soft tissue masses are always homogeneous and slightly hypodense compared to muscles on pre-contrast abdominal CT, and mildly enhanced and less hyperdense than the kidneys after intravenous contrast administration. There is no calcification, fat content, or hemorrhage in the adrenal masses. Additionally, no lymphadenopathy can be detected during CT studies. The contour of the adrenal gland is deformed in most primary lymphomas, except one case in which the adreniform shape was maintained [9]. Four cases (25%) had low attenuation foci inside the masses which may have been due to necrosis [1, 11, 16, 17]. Similarly, CT-revealed low-attenuation areas in our case were confirmed pathologically to be necrotic tissue. However, it has been noted that necrosis in lymphomas without treatment is uncommon [7]. Since there are no specific CT findings for primary bilateral lymphomas of the adrenal glands, differentiation of an adrenal lymphoma from other primary or metastatic adrenal lesions using imaging modalities is not possible. However, if there is simultaneous enlargement of bilateral adrenal glands or the

**Figure 1.** Bilateral adrenal non-Hodgkin’s lymphoma. a. Non-enhanced CT scan shows bilateral adrenal masses of homogeneous density in this patient presenting with normal adrenal function. b. Postcontrast CT scan shows bilateral large adrenal tumors of soft tissue density and with low attenuation foci. Multiple splenic cysts are an incidental finding.
presence of bilateral suprarenal masses on an abdominal CT scan, as shown in our case, primary bilateral adrenal lymphoma should be considered as a potential underlying disease. Paling et al. revealed that the chance of involvement of the adrenal gland during the course of non-Hodgkin lymphoma is only 4% [8]. Therefore, coexistence of bilateral adrenal soft tissue masses with homogeneous density and mild enhancement might be an important clue for a diagnosis of bilateral primary adrenal lymphoma.

Differential diagnosis of bilateral adrenal masses include cortical adenomas, tuberculosis, fungal infections, pheochromocytomas, metastatic tumors, and bilateral adrenal hemorrhage. Cortical adenomas are usually smaller in size than primary adrenal lymphomas. Tuberculosis and fungal infections may show necrosis and enlargement of adrenal glands which contain calcifications. Most large-sized pheochromocytomas show foci of inhomogeneous density caused by necrosis, hemorrhage, and cyst formation [7]. A metastatic malignancy is usually solid. Adrenal hemorrhage always has a typical clinical history and shows symmetrical enlargement of bilateral adrenal glands and hyperdense foci. Opposed-phase MR imaging has been advocated to differentiate benign adrenal adenomas or adrenal malignancies based on their fat content [26]. Adrenal carcinomas or metastases contain less fat and will not lose signal intensity when imaged out of phase. Opposed-phase MR imaging enables the differentiation of adrenal adenomas from other adrenal malignancies, but a specific diagnosis might be not achieved during the study.

In conclusion, there are no pathognomonic CT findings for diagnosis of primary bilateral adrenal lymphomas. Patients with bilateral primary adrenal lymphoma have a wide age range and

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**Table 1. Clinical presentations and CT findings of primary bilateral adrenal lymphoma in 16 patients**

<table>
<thead>
<tr>
<th>Case No/Age/Sex</th>
<th>Symptoms</th>
<th>Diagnostic procedure</th>
<th>Pathology</th>
<th>Adrenal function</th>
<th>CT findings</th>
</tr>
</thead>
<tbody>
<tr>
<td>1/74/F</td>
<td>Lethargy, weight loss</td>
<td>CT ⊕, bone marrow biopsy</td>
<td>Diffuse B-cell NHL*</td>
<td>↓</td>
<td>Bil adrenal mass, partial necrosis</td>
</tr>
<tr>
<td>2/64/F</td>
<td>Fever, electrolyte imbalance</td>
<td>CT, US</td>
<td>Diffuse large B-cell NHL enlargement</td>
<td>↓</td>
<td>Bil adrenal enlargement</td>
</tr>
<tr>
<td>5/69/M</td>
<td>Abdominal pain, fever, weight loss</td>
<td>CT, US</td>
<td>Angiotrophic[?] large-cell lymphoma</td>
<td>↓</td>
<td>Bil adrenal enlargement</td>
</tr>
<tr>
<td>9/71/M</td>
<td>Fever, weakness, deterioration</td>
<td>CT, US</td>
<td>Small cleaved cell NHL</td>
<td>↓</td>
<td>Bil adrenal enlargement</td>
</tr>
<tr>
<td>4/68/F</td>
<td>Abdominal pain</td>
<td>CT</td>
<td>NHL, CD45+</td>
<td>↓</td>
<td>Bil adrenal enlargement</td>
</tr>
<tr>
<td>7/59/M</td>
<td>Fever, anemia</td>
<td>CT, US</td>
<td>Lymphoma, hepatosplenomegaly</td>
<td>↓</td>
<td>Bil adrenal enlargement, Mass. hom.</td>
</tr>
<tr>
<td>12/43/M</td>
<td>Abdominal pain, fever, hypertension</td>
<td>CT, US</td>
<td>NHL</td>
<td></td>
<td>Mass. hom.</td>
</tr>
<tr>
<td>3/42/F</td>
<td>Elevated serum lactate dehydrogenase</td>
<td>CT, angiography</td>
<td>Adrenal mass</td>
<td>↓</td>
<td>Enlargement of adrenal</td>
</tr>
<tr>
<td>8/71/M</td>
<td>Diarrhea, weight loss</td>
<td>CT, MRI ⊙</td>
<td>Small non-cleaved autopsy cell NHL with foci of low attenuation</td>
<td>↓</td>
<td>Bil adrenal enlargement</td>
</tr>
<tr>
<td>10/74/M</td>
<td>Abdominal pain, fever, weight loss</td>
<td>CT</td>
<td>Lymphoma</td>
<td>↓</td>
<td>Bil adrenal mass</td>
</tr>
<tr>
<td>11/81/M</td>
<td>Back pain</td>
<td>CT</td>
<td>Large-cell NHL</td>
<td>↓</td>
<td>Bil adrenal enlargement</td>
</tr>
<tr>
<td>13/64/F</td>
<td>Fever, weight loss, night sweating</td>
<td>CT, MRI</td>
<td>Large-cell immunoblastic lymphoma</td>
<td></td>
<td>Mass. hom.</td>
</tr>
<tr>
<td>14/59/F</td>
<td>Abdominal pain, weight loss, weakness</td>
<td>CT</td>
<td>Large-cell NHL</td>
<td></td>
<td>Bil adrenal mass</td>
</tr>
<tr>
<td>15/69/M</td>
<td>Epigastric pain, weight loss, weakness</td>
<td>CT</td>
<td>Large-cell NHL</td>
<td></td>
<td>Bil adrenal mass with foci of low attenuation</td>
</tr>
<tr>
<td>16/37/F</td>
<td>Abdominal pain, fever, weight loss</td>
<td>CT, US</td>
<td>Diffuse large B-cell NHL</td>
<td></td>
<td>Bil adrenal mass with foci of low attenuation</td>
</tr>
</tbody>
</table>

* Correlated to the sequence in “References”.
+ Non-Hodgkin’s lymphoma.
⊕ Bilateral.
∆ Ultrasonography.

# Magnetic resonance imaging
¬ Normal adrenal function.
usually present nonspecific symptoms. Most of them have presentations related to primary adrenal insufficiency, but some do not. Therefore, the possibility of primary bilateral adrenal lymphoma should be considered in a patient with bilateral adrenal enlargement, even if initial primary adrenal insufficiency is not found [3].

**REFERENCES**

原發性雙側腎上腺淋巴瘤：一病例報告

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雙側原發性腎上腺淋巴瘤是極為罕見的腫瘤，文獻記載的僅有二十六例。本病例是一位三十
七歲女性，因腹痛就醫，由電腦斷層攝影發現雙側腎上腺腫塊，經超音波導引生檢後，病理組
織證實為B細胞非何傑金氏淋巴瘤，回顧文獻記載對雙側原發性腎上腺淋巴瘤的電腦斷層影像
分析，大多只有發現雙側腎上腺腫大，少數有壞死，但這些表現不具特異性，也就是說電腦
斷層攝影並無法提供病徵學特徵。

關鍵詞：腎上腺，腫瘤；淋巴瘤，電腦斷層攝影