**Case Report**

**Adrenal myelolipoma with abdominal pain: A rare presentation**

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**ABSTRACT**

Adrenal myelolipomas are rare benign tumors. Most of the cases are asymptomatic and discovered incidentally. We are reporting a case of myelolipoma involving right adrenal cortex of a 40-year-old woman who presented with abdominal pain. A short review of etiology, clinical features, and differential diagnoses of this neoplasm are also discussed. Radiologic features are often helpful in diagnosis but histology must be done to exclude other fat-containing lesions. Although uncommon, myelolipomas should be considered in differential diagnosis of retroperitoneal lesions.

**Key words:** Adrenal myelolipoma, histology, radiology, abdominal pain

**INTRODUCTION**

Gierke, in 1905, described a lesion of adrenal gland containing fat and myeloid elements.[1] The nomenclature myelolipoma was used for this tumor by Oberling in 1929.[2] Myelolipomas are uncommon, small, asymptomatic benign lesions of adrenal cortex.[3] Incidence at autopsy has been reported from 0.08% to 0.4%.[4] Similar lesions are also reported from extra-adrenal sites. Most of the lesions are discovered incidentally and are called incidentolomas. With improved radiologic techniques, the rate of detection of lesions is gradually increasing.[3,5,6]

**CASE REPORT**

A 40-year-old woman presented with pain in the right upper abdomen for last 6 months. Ultrasonography of upper abdomen failed to show suspected biliary pathology. But a possible adrenal mass was noted close to the upper pole of right kidney. The clinician suspected of an adrenal mass favoring adrenal adenoma. But the routine blood and urine examination along with estimation of 24 h urinary cortisol and vanillyl mandelic acid excretion did not show any abnormality. Computed tomography (CT) scan of the abdomen was advised, which showed a lesion of right adrenal gland (7 × 5.5 cm) containing fat and specs of calcification [Figure 1]. A possible radiologic diagnosis of adrenal myelolipoma was suggested and histopathologic examination was recommended. The tumor was dissected out. Grossly, it was soft, well circumscribed, 6.5 × 4.5 cm in size. Multiple sections from different parts of the tumor were processed and examined under microscope. The tumor was seen to be composed of mature adipose tissue and trilineage hematopoietic elements (erythroid, myeloid, and megakaryocytic) without any evidence of dysplasia [Figure 2]. A thin rim of normal adrenal tissue was also seen adjacent to the tumor [Figure 3]. A diagnosis of adrenal myelolipoma was confirmed. The patient was followed-up for 1 year, which was uneventful.

**DISCUSSION**

Myelolipomas are uncommon benign neoplasms composed of mature fatty tissue and myeloid elements. These are not related to anemia or hematopoietic diseases.[5] Commonly the tumor involves adrenal cortex with possible origin

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from zona fasciculata layer. Only about 50 cases of extra-adrenal myelolipomas have been described in the literature. Presacral soft tissue was the commonest involved site followed by retroperitoneum, pelvis, stomach, and perirenal tissue.

These tumors are seldom found before puberty and predominantly involve older persons. There is no sex predilection. Most of the lesions are asymptomatic. Endocrine dysfunction is occasionally reported and usually occurs due to underlying adrenocortical pathology.

Myelolipomas are smaller lesions varying from microscopic foci to 8 cm in diameter. However, larger giant lesions (as large as 30 cm in diameter) are rarely reported, often presenting as a palpable abdominal mass producing compressive features and flank pain due to hemorrhage.

In our case, no hemorrhage was noted in the tumor and the pain might be due to its large size (>6 cm). Rare cases of bilateral adrenal involvement have also been documented.

Majority of these tumors being asymptomatic, are discovered incidentally during radiologic evaluation of abdomen. Although small adrenal myelolipoma does not require surgical intervention, but in this case the surgeon decided to operate the case not only for confirmatory diagnosis (CT diagnosis was suggestive of myelolipoma only) but also to relieve the patient from abdominal pain because of its large size. The surgeon also wanted to exclude the possibility of adrenal adenoma.

CT and magnetic resonance imaging can differentiate myelolipomas from other adrenal tumors by demonstration of abundant fatty tissue. But reduction of fat component due to extensive hemorrhage, calcification, or abundance of myeloid component may complicate detection. Radiologic distinction of extra-adrenal myelolipomas from other fat-containing tumors is quite difficult.

Histologically, myelolipomas are well circumscribed, separated from the main mass of adrenal gland, and consist of myeloid and lipoid elements in variable proportions. We observed a thin rim of normal adrenal tissue adjacent to the mass in our case as well. In some lesions, fatty elements predominate consisting of mostly mature fatty tissue. Myeloid elements include normoblasts, myelocytes, megakaryocytes, and mature leukocytes. These elements can predominate in some lesions. A fibrous stroma is present in the tumor, which rarely contains osseous components. Areas of hemorrhage and necrosis with calcification are often present, particularly in case of larger lesions.
Microscopical diagnosis of adrenal myelolipoma is straightforward. If the tissue of origin is not ascertained, confusion can arise with renal angiomyolipoma, lipoma, or low-grade liposarcoma of surrounding tissues. Extra-adrenal myelolipomas must be differentiated from foci of extramedullary hematopoiesis by their increased proportion of fat content and circumscription. Lipomas consist of only mature fat. Low-grade liposarcomas are infiltrative with cellular atypia and lipoblasts. Sometimes mesenchymal tumors or teratomas may cause confusion with myelolipomas. But these tumors usually contain other tissue elements.

Prognosis of myelolipoma is excellent. Malignant change is still not documented. As lesions larger than 10 cm have a risk of hemorrhagic complications, these should be removed. But smaller lesions, less than 6 cm in diameter, should be followed-up for 6–12 months without any intervention. In this case also the patient was followed-up for 1 year and no complication was observed.

Pathogenesis of this unusual tumor is yet to be confirmed. Theories put forward by different workers include retention of embryonic rests, extramedullary hematopoiesis, bone marrow embolization, and others. But the most accepted hypothesis of tumorigenesis is metaplasia of adrenal mesenchymal cells or migrated hemopoietic stem cells.

Adrenal or extra-adrenal myelolipomas should come into differential diagnosis of fat containing tumors of retroperitoneum. Uncommon presentations of this benign tumor sometimes cause confusion with other aggressive neoplasms. Care must be taken for accurate diagnosis to avoid hazardous therapy over this innocuous lesion.

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