International Surgery Journal
Mahadik AG et al. Int Surg J. 2023 Apr;10(4):786-790
http://www.ijsurgery.com
pISSN 2349-3305 | eISSN 2349-2902

DOI: https://dx.doi.org/10.18203/2349-2902.isj20230999

Case Report

Case report of a 60-year-old male with giant adrenal myelolipoma

Abhishek G. Mahadik*, Nikhil L. Beldar, Meena Kumar, Niket Attarde, Isha Bhatnagar

Department of General Surgery, Dr. D. Y. Patil Medical School, Navi Mumbai, Maharashtra, India

Received: 15 February 2023
Accepted: 16 March 2023

***Correspondence:**
Dr. Abhishek G. Mahadik,
E-mail: Dr.abhishek0706@gmail.com

ABSTRACT

Adrenal myelolipoma is a benign neoplasm of the adrenal gland that is the second most common primary adrenal incidentaloma, following adrenocortical adenomas. It is composed of elements of adipose tissue and with varying amounts of hematopoietic components. In the past, these tumors were discovered at autopsy, with an incidence ranging from 0.08% to 0.4%. Today, with the widespread use of radiological studies such as ultrasonography, computed tomography (CT), and magnetic resonance imaging (MRI), the incidental detection of myelolipoma has become more common, constituting up to 10-15% of incidental adrenal masses. The patient was hospitalized for a 3-month history of discomfort in the right flank, swelling over the right hypochondriac area, constipation, retrosternal burning sensation, and early satiety. His vitals were stable but per abdominal examination revealed a mass in the right hypochondrium. B-mode ultrasound screening revealed an adrenal tumor. A hematological examination revealed thrombocytopenia. The presence of unilateral adrenal mass with the absence of new-onset hypertension with CECT suggestive of mixed tissue density mass with multiple ill-defined heterogeneously hypodense fat attenuation led to the primary diagnosis of an adrenal mass After correction of thrombocytopenia, the mass was excised and subjected to histological evaluation. The histological study results corroborated with the original diagnosis of unilateral adrenal myelolipomas. The patient was followed up for 2 months postoperatively and was found to be relieved from the presenting symptoms with no postoperative complications. Adrenal myelolipoma is an uncommon, benign, and hormonally inactive adrenal gland tumor. It may present with symptoms due to compression of adjacent structures which warrants a surgical excision.

Keywords: Adrenal myelolipoma, Case report, Tumor, Adrenal gland

INTRODUCTION

Adrenal myelolipoma is a rare, benign, non-functional tumor of the adrenal gland made up of mature adipose tissue with varying amounts of hematopoietic components and mature fat that resembles bone marrow.[2] Myelolipoma is most commonly detected unilaterally in the adrenal gland and is rarely bilateral. As the tumors grow in size, they become symptomatic due to pressure on adjacent tissues, producing flank pain and stomach discomfort, and may even manifest with necrosis, rupture, hemorrhage, or hemorrhagic shock. The presence of mature, macroscopic fat deposits, with or without calcification and soft tissue features, may generally be used to confidently identify myelolipoma on CT or MRI scans. Myelolipoma accounts for 7-15% of unintentionally discovered adrenal masses.[3,4] Adrenal myelolipoma treatment is often conservative because these tumors are frequently asymptomatic and seldom show symptoms of spontaneous bleeding if the tumor is small. Some studies recommend surgery if the tumor is symptomatic, expanding, or greater than 6 cm. In the end, the best treatment for myelolipoma is determined by mass's size and symptoms, and requirements of patient. Men and women are equally affected by tumor, which is most usually seen between the fifth and seventh decades of life, with mean age of 62 years.[2,3] They are generally non-secreting in nature and are often smaller than 4 cm in diameter, with the largest reported adrenal myelolipoma measuring 31×24.5×11.5 cm and weighing 6 kg.[5]

CASE REPORT

A 60-year-old male from Assam, with no family history of genetic disorders or personal history of addictions presented in the surgical OPD with a 3-month history of discomfort in the right flank, swelling over the right hypochondriac area, constipation, retrosternal burning sensation and early satiety.

Clinical findings

The patient was stable on general examination. On per abdominal examination, a firm, non-tender lump measuring 5" 5" spherical in shape non mobile was palpable in the right hypochondrium extending to the right lumbar region. Other systemic examinations did not reveal any significant findings.

Table 1: Diagnostic assessment.

Date	Examination	Diagnosis
22/02/2022	CT (Abdo + pelvis) (contrast)	22×18×21 CM sized large well-defined mixed tissue density mass with multiple ill-defined heterogeneously hypodense fat attenuation and hyperdense area of blood attenuation and few non enhancing septae within is noted in the right suprarenal region not seen separately from the right adrenal gland.
8/03/2022	USG (abdo + pelvis)	large heterogeneous fat-containing lesion measuring approximately. 22×17×19 cm is seen in the right suprarenal region. Moderate splenomegaly.
24/03/2022	FNAC	Reveals only hemorrhagic and fibro adipose tissue.
25/03/2022	CBC	HB 7.4, TLC 4.3, PLT 40, PCV 24
26/03/2022	PET scan	Low-grade FDG uptake is noted in large heterogeneous mass lesion with areas of fat content and septae within is noted in the right suprarenal region measuring about 21.2×16.9 cm. The right adrenal gland is not seen separately from this lesion. This lesion displaces right kidney inferiorly and towards midline.
28/03/2022	Peripheral smear	Decreased in number, large platelets are seen

The patient was referred for ultrasound imaging for the abdominal mass. On performing a B-mode ultrasound scanning for right hypochondrium discomfort, large heterogeneous fat-containing lesion measuring approximately 22×17×19 cm is seen in the right suprarenal region with Moderate splenomegaly. For further evaluation and to assess the boundaries of mass CT abdomen done shows an adrenal mass of 18×20 cm was detected. The mass was found to be pushing the hepatic flexure laterally, liver cranially, kidney downward, and IVC medially. Adhesions were seen between the mass and the IVC. All hematological parameters were normal except thrombocytopenia which is diagnosed as asymptomatic constitutional macro-thrombocytopenia or Harris platelet syndrome and this is commonly found in the northeast region of India.

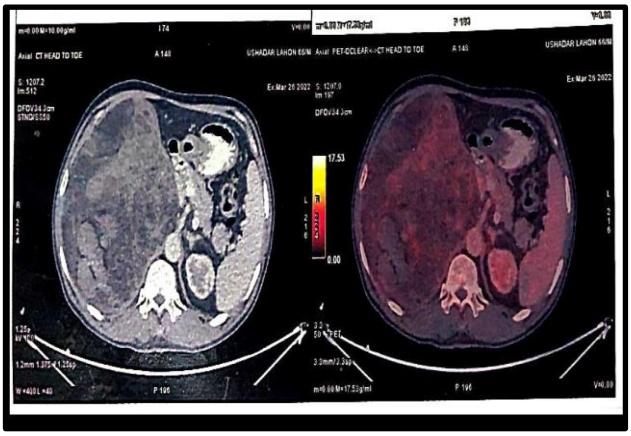

Figure 1: PET scan image of a large mass in the abdomen.

Therapeutic intervention

The diagnosis of an adrenal mass without the new onset of hypertension and causing symptoms due to compression of the adjacent viscera led to a decision of surgical excision of the tumor. As the patient had thrombocytopenia, it was imminent to correct the platelet count before proceeding with the surgery. The patient was administered intravenous dexamethasone for four days and the platelet count was evaluated. One unit of single donor platelets (SDP) was transfused for further correction of the platelet count before the surgery.

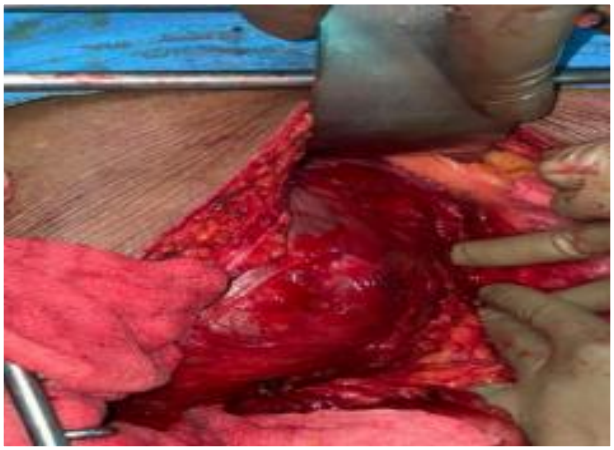

Figure 2: Intraoperative image of adrenal mass excision.

A midline incision was used to open the abdominal cavity and an 18×20 cm mass originating from the upper pole of

the right kidney was identified. The mass was observed to be pushing the hepatic flexure medially, the liver cranially, the kidney downward, and the IVC medially. Adhesions were seen between the inferior vena cava and the adrenal mass. The right adrenal artery and vein were ligated, and the tumor was removed. The excised sample weighing 3 kg was sent for histopathological examination. Following surgery, indwelling urinary catheters and nasogastric tubes were placed in situ until the patient's urinary and gastrointestinal functions returned to normal. Postoperatively thrombocytopenia was persistent without causing any complications related to low platelet and the patient was given symptomatic therapies, and supportive care.

Histopathology (Figure 3)

Pathologists analyzed all resection specimens of the adrenal mass using H and E staining (which was repeated twice on resection specimens). The specimen was made up of a single massive bulk measuring 23×18×9 cm.

The gross examination findings were as follows-

External surface

Capsulated with an area of congestion. The capsule was shiny and smooth with a pale area.

Cut surface

Well-circumscribed firm and solid mass with red and yellow area in the adrenal gland parenchyma could not be appreciated.

Microscopy

Complete effacement of the adrenal gland by mass. Tumour consists of multiple islands of hematopoietic cells and multiple nodules of mature adipose tissue are also seen.

The histopathological examination results confirmed the initial diagnosis of unilateral adrenal myelolipomas in this patient.

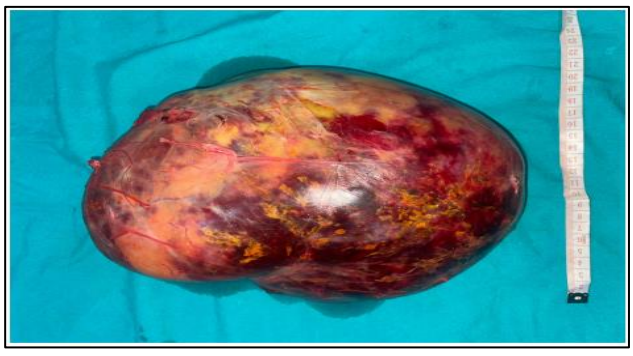

Figure 3: A gross image of the resected specimen.

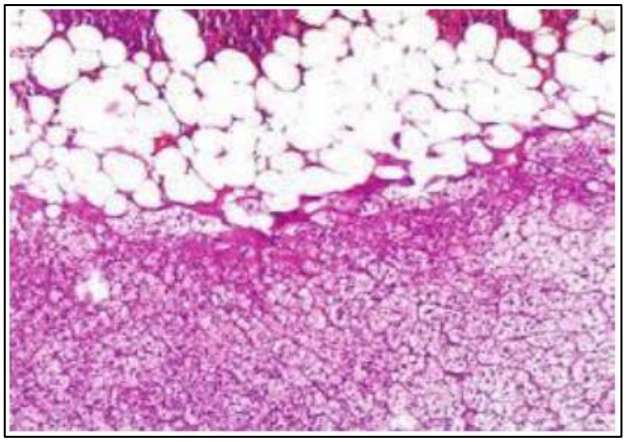

Figure 3: Histopathological image of the resected specimen.

Follow-up and outcomes

The patient was followed up for 2 months postoperatively and was found to be relieved from the presenting symptoms with no postoperative complications.

DISCUSSION

Adrenal myelolipoma is an uncommon, benign; tumor-like lesion composed of mature adipose tissue mixed with hematopoietic cells and hormonally inactive adrenal gland neoplasm. It usually affects one adrenal gland, more commonly to the right. In our case, he is 60 years old, which falls into the mean age group. The mean age at diagnosis is approximately 62 years, and most patients are asymptomatic.[2,3,6] It's difficult to anticipate the occurrence of adrenal myelolipoma and precisely identify possible patients in the community. Because myelolipoma is a benign tumor, it does not spread to other regions of the body. The largest reported AML in the literature was measuring 31×24.5×11.5 cm and weighed 6 kg.[5] In our case its measuring 23×18×9 cm and weighs 3 kg. Myelolipomas remain stable in size or grow slowly. In a large longitudinal follow-up study, overall tumor change ranged from-10 to 115 mm with a growth rate ranging from-6 mm/year to 14 mm/year.[7]

However, the etiology and pathogenesis of adrenal myelolipoma are yet unknown. Adrenal myelolipoma has been defined as the differentiation of adrenal medullary or cortical cells into adipose tissue and extramedullary hematopoietic tissue in the adrenal gland induced by the triggering of several adverse causes such as stress, infection, ischemia, necrosis, and so on. Some investigations showed that adrenal myelolipoma was caused by the ectopic growth of myeloid cells in the adrenal gland during the embryonic phase. Furthermore, some researchers hypothesize that the tumor arose from a non-functioning adrenal cortical adenoma.

Adrenal myelolipoma, which is composed of a considerable quantity of fat and myeloid tissues, is

circular or almost so and has a distinct border as was found in this case. Adrenal myelolipomas are classified into three categories based on their tissue components: adipose tissue, myeloid tissue, and a combination of the two. The myelolipoma in this case falls into the category consisting of adipose tissue components. Adrenal myelolipoma is frequently identified by chance during a physical examination of people who have previously suffered from other conditions. There are practically no visible manifestations of the adrenal gland or endocrine abnormalities in these people.

In general, no particular clinical signs and symptoms are noted in individuals with adrenal myelolipoma. The most common symptoms observed are abdominal discomfort/pain, hypochondriac pain, pressure symptom, and flank pain.[6] In our case, the patient presented with pressure symptoms and flank pain. Most AMLs have mass effect symptoms and spontaneous rupture is observed more in AML >6 cm.[7,11] Spontaneous retroperitoneal bleeding is a well-known complication of adrenal myelolipoma. Most of the AMLs are discovered incidentally and the radiological features are accurate in diagnosing AML in up to 90% of the cases.[8]

The diagnosis of adrenal myelolipoma is mostly based on preoperative diagnostic imaging screening, localization, and qualitative evaluation. Ultrasonography, CT, and MRI are effective in diagnosing adrenal myelolipomas in more than 90% of cases, with CT being the most sensitive diagnostic imaging modality. The presence of fat within these tumors is the key factor for both identification and preoperative diagnosis with imaging methods. However, adrenal myelolipomas may contain different proportions of fat and myeloid tissue, and those with only small quantities of fat may be difficult to differentiate from other adrenal masses. Small lesions are more difficult to diagnose with sonography than large ones.

CT appears to be sensitive to the diagnosis of adrenal myelolipomas. CT explained confusing sonographic findings by showing fat-density tissues within the lesions, AMLs appear well-defined, hypodense, and heterogeneous masses; the presence of fat density is essential for the radiological diagnosis of AML. The fat compound of AMLs is the key factor for preoperative diagnosis on CT or MRI.[11]

MRI of adrenal myelolipoma characteristically demonstrates a bright signal on T1-weighted and T2-weighted sequences, consistent with the presence of fat. When a diagnosis of adrenal myelolipoma is considered, it should be differentiated from other fat-containing retroperitoneal tumors such as retroperitoneal lipoma, liposarcoma, extrarenal Adrenal myelolipoma, primary, or metastatic adrenal malignancy of teratoma.

In cases when the diagnosis can not be made radiologically or in any doubt of malignant potential, the use of fine needle aspiration cytology (FNAC) can be considered.[3] FNAC might give a clue of a lipomatous tumor and be able to detect atypical cells if there is a presence of atypical stromal cells. This will help in the management plan. However, in this case, FNAC is required as this is a fast-growing symptomatic tumor with no suspicious features of malignancy radiologically.

The resected specimen can be histologically evaluated. A thorough gross and microscopic examination are required to confirm the diagnosis and rule out malignancy. To ensure that no malignancy is missed, specimens should be sampled 1 cm apart for microscopic analysis. Microscopically, a typical myelolipoma contained a mixture of mature adipocytes and foci of trilineage hematopoietic elements.[3]

Adrenal myelolipoma management surgical excision is safe, curative, and helpful. However, prior to surgery, the quantitative evaluation and definitive localization of an adrenal myelolipoma may be dependent not only on diagnostic imaging studies, but also on medical histories, clinical symptoms and signs, physical examinations, and laboratory testing. The surgical technique, however, may be determined by the nature, grade, size, and location of the myelolipoma. When patients have tumors greater than 3.5 cm in diameter or tumors that develop quickly, surgical excision is the best option for treating adrenal myelolipoma.[10] Laparoscopic adrenalectomy is often advised for an adrenal tumor less than 6 cm in diameter because it allows patients to recuperate faster and with less postoperative discomfort.[12] However, if an adrenal tumor is larger than 6 cm in diameter or appears to be growing rapidly based on imaging features, a traditional open adrenalectomy is the preferred surgical procedure that was followed in our case.[13]

In the current case report, the adrenal mass was removed, and histology was performed. The reports revealed an encapsulated single huge mass. Multiple hematopoietic cell islands and mature adipose tissue nodules were detected in the tumor.

In this patient, histological study results corroborated the original diagnosis of Unilateral adrenal myelolipomas.

CONCLUSION

Adrenal myelolipomas are clinically silent tumors that are mostly benign. Adrenal myelolipoma patients are typically diagnosed with incidental findings. The "accidental" discovery of an adrenal mass necessitates careful diagnostic investigation in order to plan appropriate treatment. Giant myelolipomas, which are symptomatic, are surgical emergencies and of significant concern. Imaging techniques available today can assist clinicians in making diagnoses. CT can clarify the nature of incidentalomas, as in our patient, and can recommend the best treatment, taking into account tumor size and the

ability to prevent complications associated with a large tumor.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

REFERENCES

1. Olsson CA, Krane RJ, Klugo RC, Selikowitz SM. Adrenal myelolipoma. Surgery. 1973;73:665-70.
2. Wagnerová H, Lazúrová I, Bober J, Sokol L, Zachar M. Adrenal myelolipoma. 6 cases and a review of the literature Neoplasma. Europe PMC. 2004;51:300-5.
3. Daneshmand S, Quek ML. Adrenal myelolipoma: diagnosis and management J Urol. 2006;3:71-4.
4. Kenney PJ, Wagner BJ, Rao P, Heffess CS. Myelolipoma: CT and pathologic features. Radiology. 1998;208:87-95.
5. Nabi J, Rafiq D, Authoy FN, Sofi GN. Incidental detection of adrenal myelolipoma: a case report and review of the literature. Case Rep Urol. 2013;2013:1-3.
6. Decmann A, Perge P, Tóth M, Igaz P. Adrenal myelolipoma: a comprehensive review Endocrine. 2018;59(1):7-15.
7. Hamidi O, Raman R, Lazik N. Clinical course of adrenal myelolipoma: a long-term longitudinal follow-up study. Clin Endocrinol. 2020;93(1):11-8.
8. V.G. Shenoy, A. Thota, R. Shankar, M.G. Desai Adrenal myelolipoma: controversies in its management Indian J Urol. 2015;31(2):94-101.
9. Elsayes KM, Mukundan G, Narra VR. Adrenal masses: MR imaging features with pathologic correlation. Radiographics. 2004;24(1):S73-86.
10. Yang Y, Ye LY, Yu B, Guo JX, Liu Q, Chen Y. Two case reports of bilateral adrenal myelolipomas. World J Clin Cases. 2015;3(9):853.
11. Giant adrenal myelolipoma presenting with spontaneous haemorrhage. CT, MR and pathology correlation - PubMed. 2022;1.
12. Raffaelli M, De Crea C, Bellantone R. Laparoscopic adrenalectomy. Gland Surg. 2019;8(1):S41-52.
13. Gershuni VM, Bittner JG, Moley JF, Brunt LM. Adrenal myelolipoma: operative indications and outcomes. J Laparoendosc Adv Surg Tech A. 2014;24(1):8-12.

Cite this article as: Mahadik AG, Beldar NL, Kumar M, Attarde N, Bhatnagar I. Case report of a 60-year-old male with giant adrenal myelolipoma. Int Surg J 2023;10:786-90.