Pathology Section

Adrenal Myelolipoma: A Series of Three Cases

KUMARI SUNITA BHARATI¹, ASHISH RANJAN SINGH², RASHMI RANI BHARTI³, REECHA SINGH⁴



ABSTRACT

Adrenal myelolipomas are rare benign adrenal tumour composed of mature adipocytes intermixed with myeloid tissue. They are second most common cause after adrenal adenomas and comprise of 6-16% of all the adrenal incidentalomas. They are often asymptomatic and therefore, rarely detected but in recent times due to radiological advancements, these cases are increasingly being detected and therefore, treated. Extra-adrenal myelolipoma are the myelolipomas, detected outside the adrenal gland but occur rarely. Three cases (53 years old male patient, 60 years old male patient and 46 years old female patient) of clinically and radiologically suspected cases of symptomatic adrenal myelolipoma are discussed here. The first case described, presented with right flank pain radiating to back which was suspected as adrenal mass by Computed Tomography (CT) evaluation. The second case presented with right flank pain, haematuria and weight loss which was suspected on Ultrasonography (USG) and Contrast Enhanced Computed Tomography (CECT) as an adrenal mass. The third patient presented with bilateral pedal oedema and suspected adrenal mass was found on USG. All three cases were histopathologically confirmed as adrenal myelolipoma and managed by surgical excision. The uniqueness of the case lies in its early detection due to technological advancements and hence, proper management of the patients.

Keywords: Adenomas, Adipocytes, Adrenal gland, Incidentalomas, Myeloid tissue

INTRODUCTION

Myelolipoma is a rare benign neoplasm that predominantly occurs in the adrenal gland [1] and hence is usually diagnosed incidentally on advanced diagnostic modalities hence often called "incidentalomas". The recent increased prevalence may be attributed to wider accessibility of computed tomography and magnetic resonance imaging scans leading to diagnosis and hence, contributing to the novelty of the cases [2]. It is composed of mature adipose tissue and scattered islands of haematopoietic elements [3]. It is usually small and asymptomatic but some can cause symptoms such as abdominal pain, nausea or vomiting [4]. Here, the authors are reporting three cases of myelolipoma of adrenal gland.



[Table/Fig-1]: Computed Tomography showing a well-circumscribed adrenal gland mass measuring 10.2×10×9.2 cm

CASE SERIES

Case 1

A 53-year-old male patient who was a known case of diabetes mellitus and hypertension from ten years presented in surgery Outpatient Department (OPD) with complaint of right flank pain radiating to back. Patient was apparently asymptomatic one year back, when, he developed pain in right lower abdomen which gradually radiated to back from past one month.

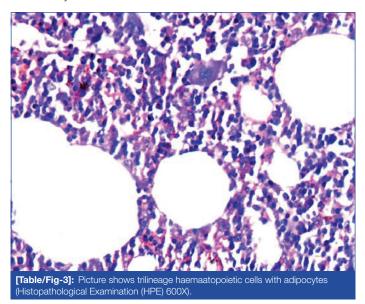
On clinical examination, there was no palpable and tender organomegaly and rest of the systemic examination was found to be within normal limits. On further investigation, his cortisol levels were decreased (4.79 µg/dL) while metanephrines and normetanephrines were 147.94 mg/24 hours and 505.77 mg/24 hours, respectively which seemed apparently normal. On radiological examination, CT scan showed evidence of well-defined large lobulated mass measuring 10.2x10x9.2 cm in size apparently originating from the right adrenal gland. Few small areas of calcifications were also seen on radiological examination [Table/Fig-1]. Gall bladder showed few small calculi while spleen and pancreas were normal. A provisional diagnosis of abdominal lump was made. As the tumour was large, it was managed via surgical excision in the Department of Surgery and the sample was sent for grossing [Table/Fig-2].



[Table/Fig-2]: Globular mass measuring 10.2×10×9.2 cm. Cut section showed yellowish, greasy with areas of haemorrhage.

The cut gross histological specimen showed yellowish, greasy with areas of haemorrhage. Histopathological examination (HPE) 600X

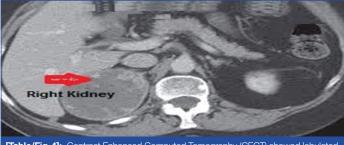
showed trilineage haematopoietic cells with adipocytes [Table/Fig-3]. A confirmed diagnosis of adrenal myelolipoma by radiological and histopathological examination was made. On routine followups, patient was asymptomatic and general condition was found satisfactory.



Case 2

A 60-year-old male patient, who was a known case of diabetes mellitus, hypertension from seven years presented with chief complaints of right flank pain from six months in surgery OPD. He was apparently asymptomatic six months back when he developed right lower abdomen pain, haematuria and progressive weight loss from six month.

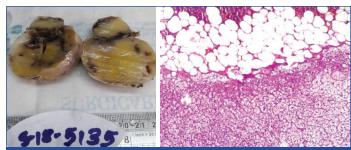
On clinical examination per abdomen was soft and non tender. No other significant finding was noted during systemic examination. On laboratory investigation, his metanephrines and nor metanephrines were 0.30 nmol/L and 0.13 nmol/L, respectively which seemed significantly decreased. Radiological examination CECT showed lobulated, capsulated hypodense lesion in right adrenal gland [Table/Fig-4] while USG showed well-defined hyper echoic mass in right adrenal gland. Patient was managed via surgical excision of tumour in Department of Surgery and tissue specimen sent for examination in the Department of Surgery. On gross section, wellcircumscribed soft tissue mass measuring 5.5×4×1.5 cm with pale yellow, soft, glistening surface were seen [Table/Fig-5]. On HPE, 600X showed trilineage haematopoietic cells with adipocytes were noted [Table/Fig-6]. Provisional diagnosis of abdominal lump was made, which was finally confirmed as adrenal myelolipoma by radiological and histopathological examination. Routine followups patient were asymptomatic and general condition was found satisfactory.



[Table/Fig-4]: Contrast Enhanced Computed Tomography (CECT) showed lobulated, capsulated hypotense lesion in right adrenal gland.

Case 3

A 46-year-old female patient who was a known case of hypertension from past five years presented with complaints of swelling in both legs from six months in the Department of Medicine. She was



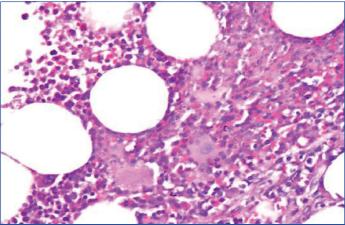
[Table/Fig-5]: Well-circumscribed soft tissue mass measuring 5.5×4×1.5 cm. Cut section showed pale yellow, soft, glistening surface. **[Table/Fig-6]:** Picture shows adrenal tissue with part of tumour composed of lobules of mature adipocytes admixed foci of trilineage haematopoiesis (HPE 400×). (Images from left to right)

apparently asymptomatic six months back when she developed progressive bilateral pedal oedema for which she consulted local quack without any improvement and was finally referred to this centre.

On investigation, she had hypercortisolism (cortisol 22.3 µg/dL) and decreased ACTH level (0.3 pmol/L). On radiological examination USG showed well-circumscribed right adrenal mass [Table/Fig-7]. Linear core biopsy was advised based on clinical and radiological parameters by Department of Surgery which showed mature adipocytes admixed trilineage haematopoiesis [Table/Fig-8]. After histopathological confirmation, the mass was completely excised and histopathologically reconfirmed as adrenal myelolipoma. In routine follow-ups, patient was asymptomatic and general condition was found satisfactory.



[Table/Fig-7]: Ultrasonography showing well-circumscribed right adrenal mass.



[Table/Fig-8]: Linear core biopsy shows mature adipocytes admixed trilineage haematopoiesis (HPE 400X).

DISCUSSION

Adrenal myelolipoma constitutes a rare incidental finding usually diagnosed on autopsy or radio-imaging generally for reasons unrelated to adrenal diseases hence, are also sometimes termed

incidentalomas entity and are also called "incidentalomas". Adrenal myelolipoma comprise of 6 to 16% of total adrenal incidentalomas and were approximately 0.08 to 0.2% of total autopsied individuals. They are usually recognised in late adulthood, with a median age of around 51 years at the time of diagnosis. There is no gender predilection; however, there may be a slight right-sided predilection [4]. There are several theories for the aetiology and the natural history of adrenal myelolipoma. The most widely accepted theory is adrenocortical cell metaplasia which occurs in response to stimuli like necrosis, inflammation, infection, or stress. Many co-morbid conditions such as Cushing's disease, obesity, hypertension, and diabetes may be considered as major adrenal stimuli [5]. Among ultrasonography, computed tomography, and MRI; CT scan is considered the most sensitive; however, all imaging modalities are more than 90% effective in diagnosing adrenal myelolipoma [6]. Most of the cases are non functional tumours and are hormonally inactive. In the three case series studies, the authors found one hormonally active and two inactive myelolipoma [7].

Histological study showed mature adipocytes admixed trilineage haematopoiesis and a diagnosis of adrenal myelolipoma was made. Dense fatty tissue is the diagnostic key in both typeshormonally active and inactive adrenal myelolipomas [8]. Management and treatment of adrenal myelolipoma should be individualised. Small asymptomatic lesions less than 5 cm should be regularly monitored over a period of 1-2 years with the help of radio-imaging controls. Any symptomatic tumours or myelolipomas larger than 7 cm should be surgically excised and appropriately followed-up [9,10].

CONCLUSION(S)

The management of adrenal myelolipoma should be individualised. The first step in diagnostic work-up of this disease is to assess hormonally active cases from other cases. Surgery is appropriate for hormonally active myelolipoma, growing tumours and tumours size is larger than 7 cm.

REFERENCES

- [1] Wang F, Liu J, Zhang R, Bai Y, Li C, Li B, et al. CT and MRI of adrenal gland pathologies. Quant Imaging Med Surg. 2018;8(8):853-75.
- Haro A, Fujishita T, Nishikawa H, Taguchi Y, Kouda T, Kajiwara K, et al. A rare case of gradual enlargement of a multifocal myelolipoma of the posterior mediastinum for 12 years after surgical resection of an adrenal myelolipoma. Int J Surg Case Rep. 2018;51:400-03. Doi: 10.1016/j.ijscr.2018.09.027.
- Wadood DQ, Qureshi DSA, Singh DP, Freedman DJ. A rare case of co-existing adrenal and pelvic myelolipomas, Radiol Case Rep. 2018;13(5):999-1002.
- Decmann Á, Perge P, Tóth M, Igaz P. Adrenal myelolipoma: A comprehensive review. Endocrine. 2018;59(1):07-15.
- Liu W, Chen W, He X. An unusual cause of cushing's syndrome and virilization. Gastroenterology, 2015;149(4):e05-06.
- Lam AK. Lipomatous tumours in adrenal gland: WHO updates and clinical implications. Endocr Relat Cancer. 2017;24(3):R65-79.
- Littrell LA, Carter JM, Broski SM, Wenger DE. Extra-adrenal myelolipoma and extramedullary haematopoiesis: Imaging features of two similar benign fat-containing presacral masses that may mimic liposarcoma. Eur J Radiol. 2017;93:185-94. Doi: 10.1016/j.ejrad.2017.05.039. Epub 2017 May 29.
- Wilson B, Becker A, Estes T, Keshavamurthy J, Pucar D. Adrenal haemangioma definite diagnosis on CT, MRI, and FDG PET in a patient with primary lung cancer. Clin Nucl Med. 2018;43(6):e192-94.
- Campbell MJ, Obasi M, Wu B, Corwin MT, Fananapazir G. The radiographically diagnosed adrenal myelolipoma: What do we really know? Endocrine. 2017;58(2):289-94.
- Shenoy VG, Thota A, Shankar R, Desai MG. Adrenal myelolipoma: Controversies in its management. Indian J Urol. 2015;31(2):94-101.

PARTICULARS OF CONTRIBUTORS:

- Senior Resident, Department of Pathology, Indira Gandhi Institute of Medical Science, Patna, Bihar, India.
- Senior Resident, Department of Pathology, Indira Gandhi Institute of Medical Science, Patna, Bihar, India.
- Assistant Professor, Department of Pathology, Indira Gandhi Institute of Medical Science, Patna, Bihar, India.
- Professor, Department of Pathology, Indira Gandhi Institute of Medical Science, Patna, Bihar, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:

Kumari Sunita Bharati,

Senior Resident, Department of Pathology, IGIMS, Patna, Bihar, India. E-mail: simmy9955885777@gmail.com

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