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Case report

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A rare case of adrenocortical oncocytic neoplasm presenting with abdominal lump: a case report

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Introduction

Oncocytic neoplasms are composed of oncocytic tumor cells, which are characterized by having large, eosinophilic, granular cytoplasm owing to the aberrant accumulation of mitochondria [1]. The most commonly reported sites for oncocytic neoplasms are the Thyroid, kidney and Salivary gland. Oncocytic neoplasms of the adrenal cortex are extremely rare. Most adrenocortical oncocytic neoplasms are benign and non-functioning and are detected incidentally. We report the case of a patient with a non functioning adrenal oncocytic neoplasm who presented with abdominal lump.

Case Presentation

A 40 years old normotensive female presented to our centre with complaints of left sided abdominal discomfort for last 4 months. There were no associated symptoms of fever, pain, anorexia, weight loss or complaints related to bowel or bladder disturbances. She had no features suggestive of any endocrine dysfunction. There was no significant past or personal or family history.

On physical examination a well defined mobile lump could be palpated in left lumbar region.

Routine blood investigations were within normal limits.

She underwent contrast enhanced CT scan of abdomen , which showed a 15 x 11 square cm mass lesion in the retroperitoneum with areas of focal non enhancing necrotic region and calcification. Mass was located in the left adrenal region, left adrenal gland could not be seen separate from the mass. Mass was pushing left kidney medially and inferiorly and pancreatic tail superiorly. There was no vascular encasement of renal vessels. There was no significant lymphadenopathy (Figure - 1).

A functional adrenal tumor was ruled out by, serum catecholamines and 24 h Urinary VMA and serum cortisol, all were within normal range.

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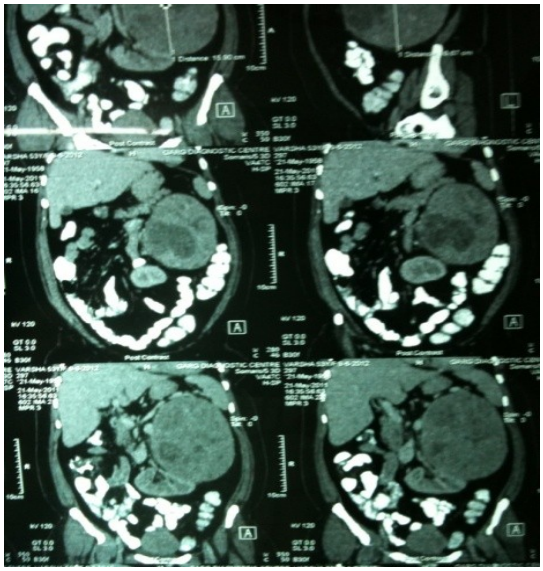

Figure 1: Contrast enhanced CT scan of abdomen showing left adrenal mass

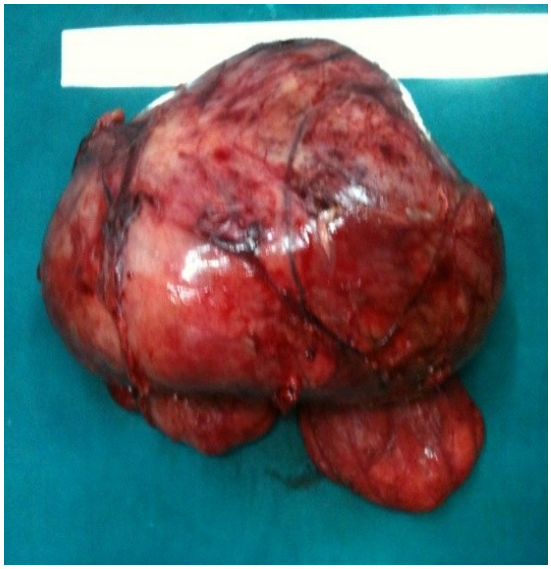

Figure 2: Resected specimen of left adrenal mass

She underwent exploratory laparotomy and excision of mass,there was no fluctuation of blood pressure intra-operatively (Figure – 2).

Postoperative period was uneventful. Histopathological examination revealed oncocytic arenocortical neoplasm. There was focal necrosis and infarction seen. There was no mitosis or capsular or vascular invasion seen.

Discussion

Oncocytic tumors originating from the adrenal cortex are extremely rare. To date, only 47 cases have been reported, including 24 oncocytomas, 7 oncocytic neoplasms of uncertain malignant potential and 16 oncocytic carcinomas. The patients had a wide age range (27 to 74 years) with a significant female to male predominance. The neoplasms varied in size from 2.2 cm to 15cm. All arises from the adrenal gland with the exception of two cases, which occurred in heterotopic retroperitoneal adrenal tissue [2, 3] and all but one case, oncocytic neoplasms were diagnosed incidentally or during investigation for symptoms that were not attributable to the tumor, such as abdominal pain, hematuria, essential hypertension, episodic vomiting, ascites and edema of the lower extremities [4, 5]. El- Naggar *et al.* [6] reported the case of one patient who suffered from abdominal pain caused by tumor infiltration to the adjacent vena cava and liver.

Oncocytic neoplasms of the adrenal cortex were non-functioning with the exception of three cases who were presented with features of cushing's syndrome [7, 8] and virilizing syndrome [9].

Our patient had a non functioning adrenocortical oncocytic neoplasm who presented with abdominal discomfort.

The biologic behavior of adrenocortical neoplasms is usually indolent. A combination of clinical, biochemical and histological features can differentiate benign and malignant adrenocortical tumors. Clinical features such as distant metastases, surgical unresectability and/or invasion of adjacent organs suggest malignant nature of lesion. The Weiss system is the most widely used and accepted histological scheme to distinguish benign from malignant adrenal tumors [10]. According to this system, the presence of four or more of the nine criteria (high mitotic rate, atypical mitoses, high nuclear grade,low percentage of clear cells, necrosis, diffuse tumor architecture, capsular invasion, sinusoidal invasion and venous invasion) indicates a malignant neoplasm.

However, the Weiss criteria have limitations. First, Weiss studied only 43 adrenocortical tumors of which 25 were benign and 18 were malignant tumors according to his classification. Second, patients with benign tumors in his series had a longer follow-up period than those with malignant tumors. Third, tumor tissue could be heterogeneous within the same lesion. Therefore, the Weiss score, even if established by experienced pathologists, cannot be completely reliable. Pohlink *et al.,* [11] reported a patient with an adrenal incidentaloma, which was initially diagnosed as benign but on follow-up was reclassified as malignant because of local recurrence and pulmonary metastases.

Bisceglia *et al.*, [12] proposed new criteria that modified the Weiss system. According to this system, if the tumor exhibits any of the major criteria (high mitotic activity, atypical mitoses or venous invasion), it is considered malignant; if the tumor exhibits any of the minor criteria (large size, necrosis, capsular or sinusoidal invasion), it is considered to have uncertain malignant potential; and none of these features indicates a benign tumor [12]. Therefore, this case was diagnosed as oncocytic neoplasm with uncertain malignant potential according to the system proposed by Bisceglia *et al.*

Adrenocortical oncocytomas are generally considered as benign neoplasms. In 22 of the 25 reported patients for whom follow-up information was available, no recurrence or metastases were observed within a follow-up period ranging from 1 to 99 months.

Borderline adrenal oncocytomas also seem to have a benign clinical behavior. Bisceglia *et al.*, [12] reported four patients with a mean follow-up of 38.75 months (10 to 61 months) with no evidence of the disease. Lin *et al.,* [5] reported two patients with uncertain malignant potential with a mean follow-up of 15.5 months (12 to 19 months) who had not experienced recurrence or metastases.

Recurrence and metastases have been described in patients with an adrenal oncocytic carcinoma. Kurek *et al.,* [13] described a patient who exhibited widespread retroperitoneal infiltration 7 years after the removal of an adrenal tumor. Local invasion into the inferior vena cava and extension to the right atrium was observed in one case and to the liver in another.

There was no evidence of metastases in our patient. The tumor was compatible with the diagnosis of an oncocytic neoplasm with uncertain malignant potential. The mass was surgically removed and no other therapy was given. After 6 months of surgery patient is still alive without evidence of recurrence and metastases. The patient will be followed-up to check for any recurrence or metastases every 4 months.

Conclusion

Although rare, adrenocortical oncocytic neoplasms must be considered among the differential diagnosis of both functional and non-functional adrenal masses. Clinical, biochemical and histological features must be evaluated together to assess the biologic behavior of these tumours.

Abbreviations

VMA: Vanillyl Mandellic Acid.

Authors' Contribution

SS, SJV: Concept, editing and review of manuscript.
SJV: Design and Literature search of manuscript.
SS, SJV, RS: Definition of intellectual content of manuscript.
SJV, RS: Manuscript preparation.
SS: Guarantor of manuscript.

Conflict of Interests

The authors declare that they have no competing interests.

Ethical Considerations

Written consent was obtained from the patient for publication of this case report.

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