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Case Report

Osseous metaplasia in adrenal myelolipoma; second documented case in human literature

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ABSTRACT

Adrenal myelolipomas are rare benign tumors composed of mature adipose and hematopoietic tissue. The prevalence of adrenal myelolipoma on autopsy is 0.08-0.4%. Osseous metaplasia within myelolipoma is an extremely rare pathology, with few cases reported in the literature. This study aims to report a case of adrenal myelolipoma with osseous metaplasia, a pathology only reported once in human studies, and to evaluate its clinical and pathological characteristics. A 75-year-old female with a history of hypertension, diabetes, and hyperlipidemia presented with left flank pain. An MRI of the abdomen revealed a left adrenal mass (7x6x5 cm). Preoperative hormonal evaluation, including serum cortisol and plasma metanephrines, were unremarkable and ruled out a functional adrenal tumor. An uneventful robotic assisted adrenalectomy was performed. The resected specimen was an encapsulated adrenal and mass measuring 5.8x5.1x4.8 cm and weighed 78 g. There were areas of hemorrhagic and fatty tissue. Findings were consistent with a left adrenal myelolipoma with osseous metaplasia, calcification, and fibrosis. She had an uncomplicated postoperative course and was discharged on post operative day two. Adrenal myelolipomas are a rare pathology, accounting for only 2-4% of adrenal tumors. Though the etiology remains unclear, the most widely accepted theories are adrenocortical cell metaplasia of the reticuloendothelial blood capillaries or bone marrow cells embolism and hypertrophic reticulum cells. While adrenal myelolipomas are most commonly found incidentally, CT scan is the most sensitive in detecting fat, and therefore detecting these tumors. Most patients are asymptomatic, but larger tumors have been shown to be associated with flank, abdominal and back pain. Most tumors are hormonally inactive, but 10% are hormonally active tumors. Metaplasia can occur due to many conditions including vitamin A deficiency, neoplasia, stress, and chronic inflammation. It was discovered in the adrenal cortex of the two-toed sloth that showed erythroid and myeloid elements. To the best of our knowledge there is only one reported similar case in the human study literature, which documented a patient with hypertension, diabetes, and Cushing syndrome with an adrenal mass. The resected specimen pathology was consistent with mature adipose tissue and areas of osseous metaplasia. Adrenal myelolipoma with osseous metaplasia is an extremely rare pathology, To the best of our knowledge and literature review there is only one case documented in humans. Further evaluation is needed to determine the prognostic factors and sociodemographic associations associated with adrenal myelolipomas exhibiting these specific pathologic characteristics.

Keywords: Adrenal myelolipoma, Osseous metaplasia, Adrenal tumour, Robotic adrenalectomy, Calcification

INTRODUCTION

Adrenal myelolipomas are a rare benign pathology that is composed of a mixture of mature adipose and hematopoietic tissue. The prevalence of adrenal myelolipoma on autopsy is 0.08-0.4%. These tumors account for 10-15% of adrenal masses discovered incidentally, detected now more commonly from

widespread use of imaging modalities such as ultrasonography (US), computed tomography (CT), and magnetic resonance imaging (MRI).[1,2] Both sexes have been shown to be equally affected, with usual occurrence in the fifth and seventh decades of life with a mean age of presentation of 62 years. The majority of adrenal myelolipomas are unilateral and asymptomatic. Myelolipomas are typically less than 4 cm in diameter and are usually hormone-inactive. While these tumours are often asymptomatic, larger myelolipomas can cause symptoms such as abdominal or back pain.[4] Various medical conditions have been shown to be associated with adrenal myelolipomas such as obesity, hypertension, atherosclerosis, diabetes mellitus, cancer, and burn injuries.[5] Due to the typical small nature of these tumours <4 cm, management is usually conservative. Hemorrhage rupture of adrenal myelolipomas is shown to occur in only 4.5% of tumors. Rupture more commonly occurs with larger tumours that are >10 cm in size. Surgical management is recommended when these masses are found to be >6 cm, increase rapidly in size, or become symptomatic.[6] We report a case of adrenal myelolipoma with osseous metaplasia, a pathology to our knowledge only reported once in the literature.

CASE REPORT

A 75-year-old female with a past medical history of HTN, DM, and hyperlipidemia, presented to outpatient clinic with left sided abdominal pain without any other symptoms including palpitation, nausea, vomiting, change of bowel habitus, or urinary symptoms. Upon workup MRI of the abdomen showed left sided adrenal mass of 7x6x5 cm (Figure 1 A, B and C).

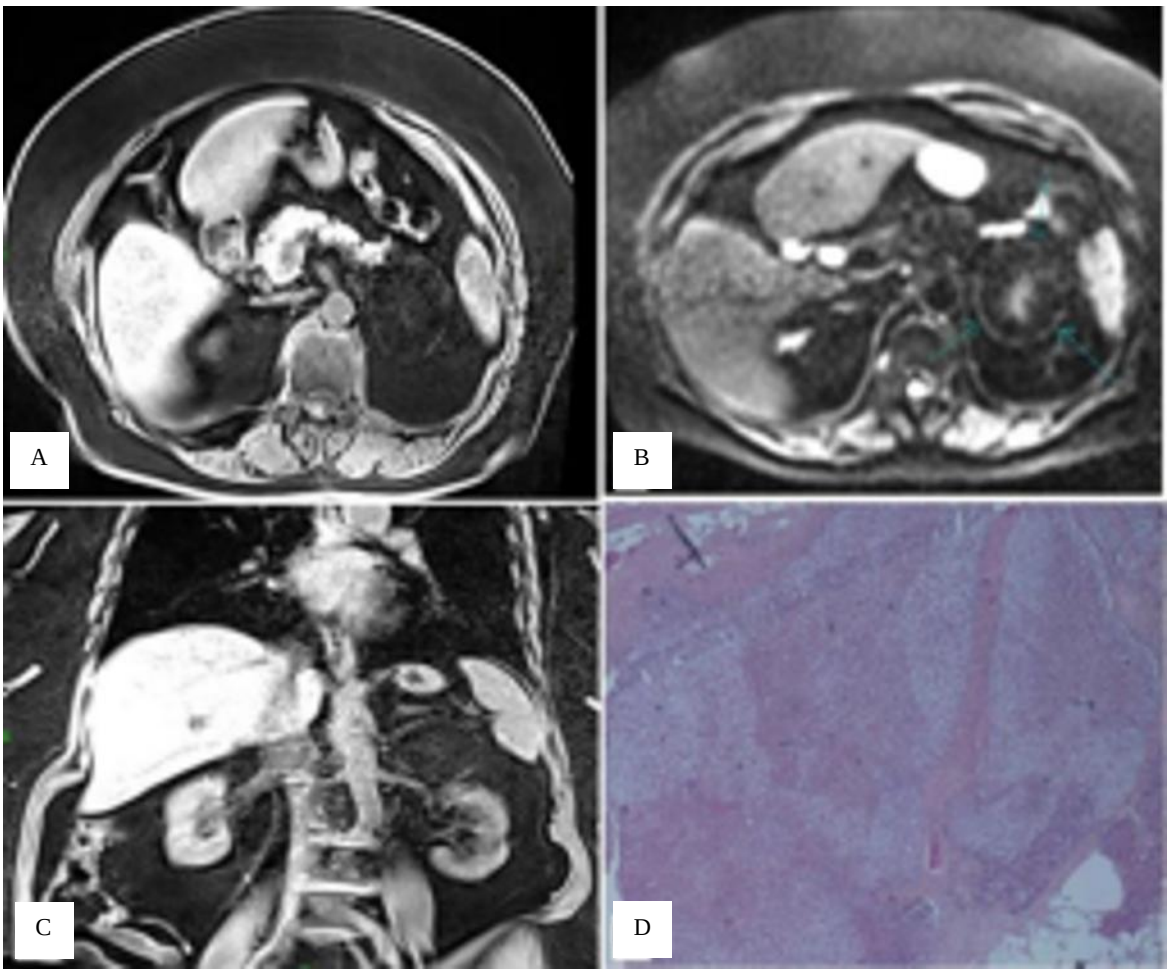

Figure 1: Imaging and histopathologic findings of adrenal myelolipoma with osseous metaplasia: (A and B) axial magnetic resonance imaging demonstrating a left adrenal mass with heterogeneous signal intensity and fatty components, (C) coronal MRI view demonstrating the left adrenal lesion measuring approximately and (D) histopathologic examination showing adrenal myelolipoma with osseous metaplasia, calcification and fibrosis (haematoxylin and eosin stain).

Preoperative hormonal evaluation, including serum cortisol and plasma metanephrines, was unremarkable and ruled out a functional adrenal tumor. Past surgical history consists of three previous C-sections and cholecystectomy. She is a former smoker that achieved smoking cessation 20 years ago. Has a BMI of 33 kg/m^2. Patient underwent robotic assisted adrenalectomy on the left. On gross inspection the specimen was yellow to tan

brown in colour and encapsulated, the tumour was 5.8x5.1x 4.8 cm and weighed 78 g. External surface was smooth with periadrenal fat and membranous tissue. Specimen was sectioned and showed a loose capsule surrounding a yellow, smooth, fatty cut surface with dispersed areas of hemorrhage and partially surrounding yellow-orange adrenal tissue with areas of calcifications.

There were areas of haemorrhagic, fatty tissue that were soft and friable. Findings were consistent with a left adrenal myelolipoma with osseous metaplasia, calcification, and fibrosis with a benign adrenal gland also present (Figure 1 D). Post operative course was uneventful and she was discharged to home on postoperative day two without any complications.

DISCUSSION

Adrenal myelolipomas are a rare pathology, accounting for only 2-4% of adrenal tumors. It remains unclear why this tumor occurs; however, the widely accepted theory is adrenocortical cell metaplasia of the reticuloendothelial blood capillin response to necrosis, infection and stress.[5] Aries Other theories include bone marrow cells embolism and hypertrophic reticulum cells.[7] Specimens of adrenal myelolipoma show microscopic findings of scattered island fat cells mixed with hematopoietic stem cells along with megakaryocytes.[5] In general these masses are well-circumscribed with a pseudocapsulate.[7]

While adrenal myelolipomas are most commonly found incidentally, CT scan is the most sensitive in detecting fat, and therefore detecting these tumors. Most patients are asymptomatic, but larger tumors have been shown to be associated with flank, abdominal and back pain. Most tumors are hormonally inactive, but 10% have been associated with Cushing syndrome, Conn's syndrome, congenital adrenal hyperplasia, pheochromocytoma, etc.[7]

Metaplasia can occur due to many conditions including vitamin A deficiency, neoplasia, stress, and chronic inflammation.[8] Osseous metaplasia is diagnosed when bone tissue is discovered in the neoplastic stroma. It has been reported to occur in the breast, gastrointestinal tract, lung, thyroid, parathyroid and pancreas. It was originally postulated that osseous metaplasia arises from fibroblasts or from cartilage.

Osseous metaplasia found in the adrenal gland is rare, with little report in the current literature of this pathological finding. Osseous metaplasia was discovered in the adrenal cortex of the two-toed sloth that showed erythroid and myeloid elements. This development was elucidated as connective tissue metaplasia due to the bone spicules that developed where bone is absent in the adrenal gland.[9]

One case has been documented to our knowledge of adrenal myelolipoma with osseous metaplasia in humans. This case report documented a patient with hypertension, diabetes mellitus and Cushing syndrome on evaluation. The resected specimen had an outer pseudocapsule and serial cutting showed a cut surface that was fibrofatty with few hemorrhagic, cystic, and calcified areas, similar to the presentation we see in our case. The adrenal gland was shown to have mature adipose tissue and areas of osseous metaplasia were seen. Our case report had similar comorbities of diabetes and hypertension as this case. These possibly related characteristics with this shared rare pathology warrants more research.

CONCLUSION

Adrenal myelolipomas with osseous metaplasia is an extremely rare pathology with little documented in the literature. Further evaluation is needed to determine prognostic factors and sociodemographic associations of these tumors with these specific pathologic characteristics.

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