OPEN ACCESS

Case Report

Adrenal Cavernous Hemangioma Associated with Myelolipoma; a Case Report

Mahsa Ahadi[1], Mohammad Esmaeil Akbari[1], Mitra Abdolahi[1], Mahsa Masoudnezhad[1], Afshin Moradi[1*]

1. Cancer Research Center, Shohada-e Tajrish Hospital, Shahid Beheshti University of Medical Sciences, Tehran, Iran.

Received: October 2017; Accepted: September 2017; Published online: 12 December 2017

Abstract:

Adrenal cavernous hemangioma is a rare benign tumor, most commonly diagnosed incidentally on imaging studies. Hereby we report a case of adrenal cavernous hemangioma with myelolipoma in a 39-year-old female presenting with epigastric pain for six months. CT scan of abdomen and pelvis showed a 38*27 mm left adrenal mass with heterogeneous enhancement and calcification. Laparoscopic left adrenalectomy was performed and pathologic examination grossly showed an encapsulated oval mass with multicystic cut surfaces containing hematoma. Microscopically, blood filled dilated vascular channels lined by endothelial cells with foci of adipose tissue admixed with normal hematopoietic cells were seen. Positive immunostaining for CD31 and CD34 confirmed the diagnosis of cavernous hemangioma. So, cavernous hemangioma associated with myelolipoma of the adrenal gland should always be considered as a differential diagnosis of adrenal masses.

Keywords: Adrenal gland; Cavernous hemangioma; Myelolipoma

Cite this article as: Mahsa Ahadi, Mohammad Esmaeil Akbari, Mitra Abdolahi, Mahsa Masoudnezhad, Afshin Moradi. Adrenal Cavernous Hemangioma Associated with Myelolipoma; a Case Report. Men's Health Journal 2017; 1 (1); e2.

Introduction:

Described by Johnson and Jeppesen for the first time in 1955 (1), adrenal cavernous hemangioma is a rare non-functioning benign tumor (2, 3) with a total of approximately 65 cases reported in the literature. Coincidence of this lesion with myelolipoma is very uncommon and only one case has been documented to date (2).

Patient Information:

A 39-year-old female with epigastric pain for six months was admitted to Shohada Tajrish Hospital on 11 February 2016. The patient had no history of fever or weight loss, her vital signs and the results of all her laboratory tests were within normal limits. She underwent a contrast-enhanced spiral computed tomography (CT) scan of her abdomen and pelvis which revealed a 38*27 mm left adrenal mass with cystic heterogeneous enhancement and calcification, along with a 27*21 mm cystic lesion in the pancreas (Figure 1). There were no signs and symptoms indicative of Cushing's disease. Laparoscopic left adrenalectomy and cystectomy of the pancreatic lesion were performed. Pathologic examination of the adrenal mass showed an encapsulated well delineated oval mass measuring 4*4*1 cm and weighing 80 gr with congested multicystic cut surface (Figure 2). Microscopically, adrenal cortex was found to have been invaded by a neoplastic lesion composed of variable sized dilated vascular channels lined by a single layer of vascular endothelial cells and filled with blood and surrounded by fibrous tissue. Islands of mature adipose tissue were also observed, admixed with bone marrow cells including normal looking hematopoietic cells (Figures 3 and 4). Immunostaining results showed the vascular channels to be positive for CD31 and CD34, which supported the diagnosis. Accordingly, the final diagnosis was established as cavernous hemangioma associated with myelolipoma. Pathologic diagnosis of pancreatic lesion was reported as macrocystic serous cystadenoma. On 25 February 2016, the patient was discharged from the hospital with complete resolution of her symptoms.

Discussion:

Adrenal cavernous hemangioma is a rare tumor with a female preponderance (Female to Male ratio of 1.7) according to the review of 52 cases conducted by Noh et al. in 2014 (4). The mean age of these patients at diagnosis was reported to be 61.8 years. Although the majority of patients were asymptomatic, the most common clinical symptom was abdominal or flank pain. The typical CT scan findings included a soft tissue mass with heterogeneous internal structures with peripheral patchy enhancement, focal or speckled calcification and scarred central region that does not show a significant enhancement (4).

***Corresponding Author:** Afshin Moradi; Pathology Department, Shohada-e Tajrish Hospital, Tajrish Square, Tehran, Iran. Postal code: 198993148. Tel/Fax +982122718049, +989121860059. Email: afshinmo2002@gmail.com.

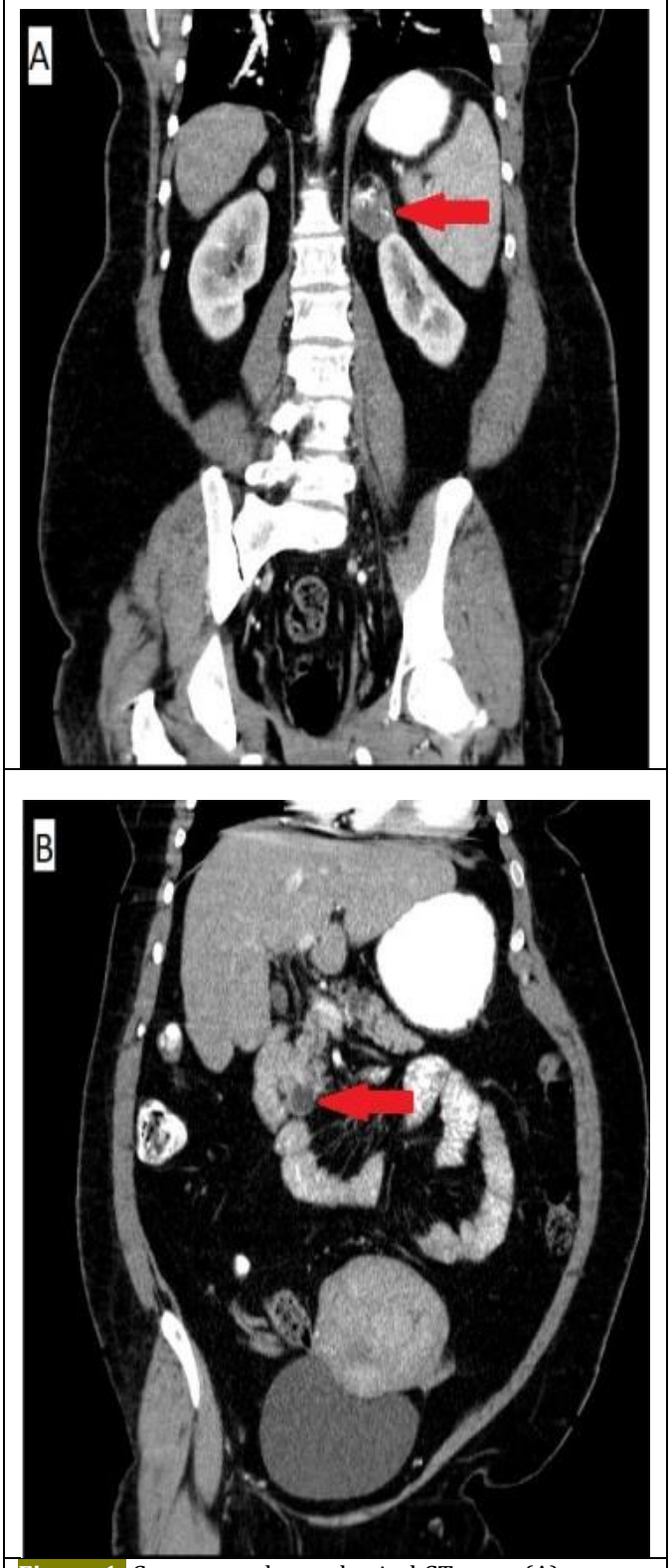

Figure 1: Contrast-enhanced spiral CT scan; (A) arrow points to a 38*27 mm left adrenal mass with cystic heterogeneous enhancement and calcification; (B) arrow points to a 27*21 mm cystic lesion in the pancreas.

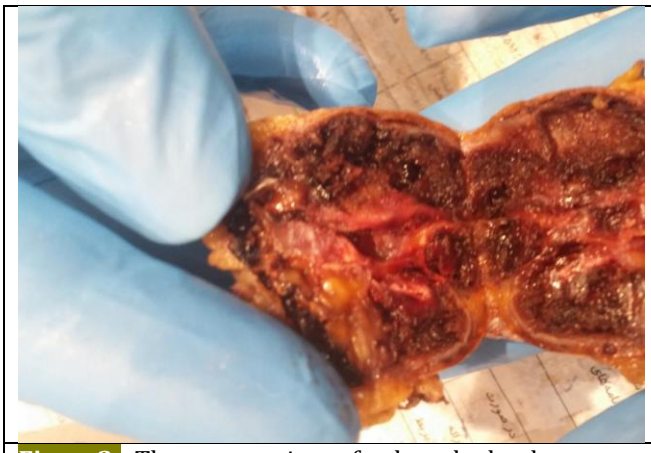

Figure2: The cut section of adrenal gland mass revealed organized hematoma.

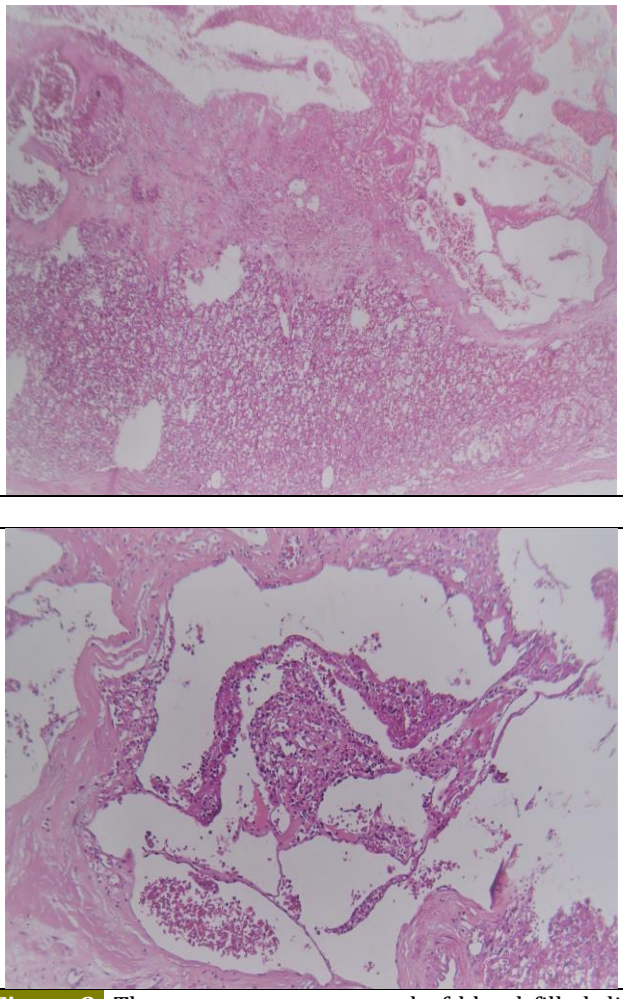

Figure 3: The tumor was composed of blood filled dilated vascular channels lined by a layer of endothelial cells.

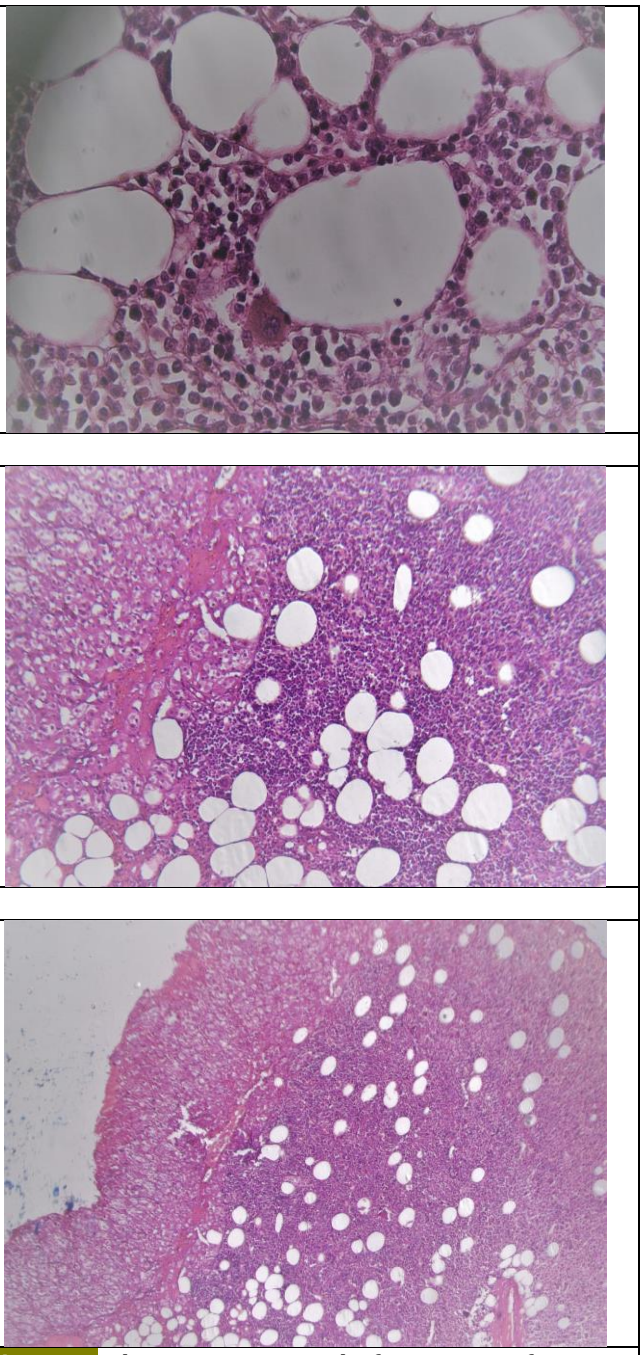

Figure 4: The tumor consisted of a mixture of mature adipose tissue and hematopoietic cells.

Preoperative differential diagnoses for this lesion include adrenal adenoma, adrenal cortical carcinoma, adrenal abscess, adrenal metastases, ganglioneuroma, pheochromocytoma, adrenal myelolipoma and hematoma (5). Most of the reported cases were nonfunctional but three documented cases have had endocrinological function, with mineralocorticoid hypersecretion reported in two of them and glucocorticoid hypersecretion in the other one (6). The majority of adrenal hemangiomas are diagnosed postoperatively based on histological examinations that show variable-sized dilated vascular channels lined by a single layer of endothelium with foci of hemorrhage and thrombosis embedded in the normal looking adrenal tissue. An area of a nodular mature adipose tissue admixed with trilineage hematopoietic cells can also be visualized, along with calcification or fibrosis occasionally reported in the lesions (2). The treatment of choice is laparoscopic adrenalectomy (4).

Conclusion:

Cavernous hemangioma associated with myelolipoma is a rare benign tumor with nonspecific symptoms that is most commonly discovered incidentally, so it should always be considered as one of the differential diagnoses of an adrenal gland lesion.

Acknowledgments:

We would like to express our special thanks to the staff of Surgery department and Pathology department of Shohada-e-Tajrish Hospital.

Conflict of interest:

All authors declare that there is no conflict of interest in this study.

Funding support:

All authors declare that this study was accomplished without any funding or support.

Author's contributions:

All the authors have contributed to drafting/revising the manuscript, study concept, or design, as well as data collection and interpretation.

References:

1. Johnson CC, Jeppesen F. Hemangioma of the adrenal. The Journal of urology. 1955;74(5):573-7.
2. Kinebuchi Y, Daimon H, Kawaguchi K. Adrenal cavernous hemangioma associated with myelolipomatous metaplasia. International Journal of Urology. 2016;23(1):106-8.
3. Quildrian SD, Silberman EA, Vigovich FA, Porto EA. Giant cavernous hemangioma of the adrenal gland. International journal of surgery case reports. 2013;4(2):219-21.
4. Noh JJ, Choi SH, Hwang HK, Kang CM, Lee WJ. Adrenal cavernous hemangioma: a case report with review of the literature. JOP Journal of the Pancreas. 2014;15(3):254-7.
5. Edwards JP, Stuart HC, Urbanski SJ, Pasieka JL. A rare cavernous hemangioma of the adrenal gland. International journal of surgery case reports. 2014;5(2):52-5.

6. Oishi M, Ueda S, Honjo S, Koshiyama H, Yuba Y, Takabayashi A. Adrenal cavernous hemangioma with subclinical Cushing's syndrome: report of a case. Surgery today. 2012;42(10):973-7.