Adenoma-Hemangioma Adrenal Collision Tumor Associated with Mild Autonomous Cortisol Secretion: A Case Report

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Introduction

Adrenal collision tumors (ACTs) are rare masses of the adrenal gland where two histologically distinct tumors coexist within an adrenal gland. Due to their complex appearance on imaging, accurate preoperative diagnosis can be challenging, often requiring a multimodal imaging approach. Adenoma-hemangioma ACTs have been reported very few times in the literature. To the best of our knowledge, this is one of two hormonally active adenoma-hemangioma ACT reported in the literature.

Presentation

A 65-year-old female was evaluated in the endocrine surgery clinic due to an incidentally found adrenal tumor during chest computed tomography (CT) for cardiac scoring.

Follow-up CT of the abdomen and pelvis:
- Pre-contrast: 4 x 5.5 cm left adrenal tumor without local invasion or calcification but with heterogeneous density.
- Arterial phase (35s): punctate foci of enhancement.
- Portal venous (70s): increasing enhancement compared to the arterial phase.
- Delayed (4 min): Persistent nodular enhancement.

Impression: heterogenous mass concerning for pheochromocytoma vs adrenocortical carcinoma.

Local symptoms: no
Symptoms of excess catecholamines: no
Symptoms of hypercortisolism: osteopenia
Symptoms of hyperaldosteronism: no

Physical exam: unremarkable.

Histories: Thalassemia, osteoarthritis, glaucoma, family history of colorectal cancer.

Workup

Biochemical Profile:

Test	Result	Reference Range
24 hr urine free cortisol	**62.6 mcg/dL**	<45
Dexamethasone suppression test (1g)	**4.4 mcg/dL (10:04 AM)**	<1.8
Adrenocorticotropic Hormone	5.7pg/mL	7-63
24 hr urine catecholamines - Epinephrine - Norepinephrine - Metanephrine - Normetanephrine	 7 mcg/dL 14 mcg/dL 112 mcg/dL 241 mcg/dL	 1-14 14-120 36-229 95-650
Potassium	3.7mmol/L	3.5-5.1
Serum Aldosterone	5.9 ng/dL	3-35
Plasma Renin Activity	0.4 ng/mL/Hr	0.2-1.6 (supine)
DHEA-S	7 mcg/dL	24-244

Imaging:

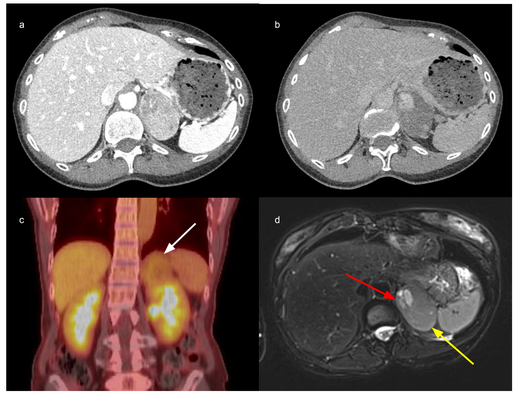

Figure 1: Preoperative imaging of the left adrenal mass. CT axial views in the portal venous (a) and delayed (b) phases showing heterogeneous enhancement and persistent nodular enhancement, respectively. FDG PET coronal view (c) showing mildly hypermetabolic left adrenal activity (SUVmax 2.6, white arrow). MRI without contrast axial view (d) showing medial high T2 signal (red arrow) with and lateral intralesional fat (yellow arrow).

Multidisciplinary Tumor Board Discussion:

- Mass more likely benign than malignant.
- Low concern for metastatic disease.
- Surveillance could be an option.

Given size, hormonally active tumor, and patient desire for definitive pathological evaluation, surgery was elected.

Surgery & Recovery

Laparoscopic Left Adrenalectomy
- R lateral decubitus positioning, OG tube, Foley catheter.
- Veress insufflation at Palmer's point. Ports: three 5mm ports (paramedian supraumbilical, epigastric and left flank), and one 12mm port (left subcostal midclavicular line),
- Splenic flexure mobilization.
- Lateral to medial mobilization of the pancreas, spleen, greater curvature of the stomach.
- Mass identified: no desmoplastic reaction or local tissue invasion.
- The left inferior phrenic vein was traced down to its left adrenal vein junction and renal vein termination.
- The adrenal vein was ligated twice on the stay side and a bipolar energy device used to transect the vein on the specimen side.
- Dissection continued in the cephalad and lateral directions until the specimen was freed & removed from the enlarged 12mm port site.

Post Operative Course
- Hemodynamics were stable during overnight observation.
- AM cortisol was low (12.1 mcg/dL, N >15).
- Follow up cosyntropin stimulation test indicated adrenal insufficiency. (12.1 mcg/dL @ 30 min, 14.7 @ 60 min; N >18).
- A corticosteroid regimen with 10mg of hydrocortisone qAM and 5 mg of hydrocortisone qPM was started.
- Discharge on POD 1.
- No complaints on 2 week post operative evaluation.
- The patient continues on a steroid regimen (approximately 11 months post op).

Pathology

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***SPECIMEN(S) RECEIVED***
A: Adrenal gland, left mass, adrenalectomy

***FINAL PATHOLOGIC DIAGNOSIS***
A.  Adrenal gland, left mass, adrenalectomy:
    - Adrenal cortical adenoma and hemangioma, 7 cm in greatest extent,
completely excised
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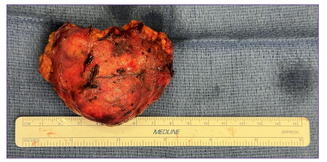

Discussion

- Although rare, a wide gamut of ACTs have been described in the literature with benign, metastatic, and primary adrenal malignant components described.
- To our knowledge, <10 adenoma-hemangioma ACTs have been described in the literature. Their potential biochemical profile has not been well described.
- Due to their heterogeneous appearance, diagnosis of ACTs based on imaging can be challenging
  - Hemangioma components classically have early peripheral nodular enhancement with progressive centripetal filling on contrast enhanced imaging (CT and MRI).
  - Adenoma components will be hypodense lesions with <10 HU on non-contrast CT, or have rapid washout on the 15 min delay phase. MRI will show a signal drop on out-of-phase T2 images.
- While surveillance can be considered for benign appearing adrenal lesions, hormonal activity and evolution should assessed.
  - Functional tumors carry long-term risk of morbidity, even if the patient is otherwise "asymptomatic".
  - Adrenal masses >4-6 cm have higher chance of malignant evolution.

Conclusion

ACTs should undergo thorough workup for signs of malignancy and function. Even in otherwise asymptomatic patients in which malignancy has been ruled-out, consideration for adrenalectomy should be given to hormonally active tumors due to long-term risk of medical morbidity.

Select References

1. Toklu A, Mesa H, Collins K. Incidental adrenal hemangioma clinically suspicious for malignancy: diagnostic considerations and review of the literature. Int J Clin Exp Pathol. 2022 Nov 15;15(11):444-458. PMID: 36507066; PMCID: PMC9729942.
2. Siddiqi AJ, Miller FH, Kasuganti D, Nikolaidis P. Adrenal hemangioma-adenoma: an exceedingly rare adrenal collision tumor. J Magn Reson Imaging. 2009 Apr;29(4):949-52. doi: 10.1002/jmri.21430. PMID: 19306439.
3. Foresti M, Parmiggiani A. Adrenal Adenoma-Hemangioma Collision Tumor: Description of Two Cases. J Radiol Case Rep. 2019 Jun 30;13(6):1-12. doi: 10.3941/jrcr.v13i6.3691. PMID: 31558958; PMCID: PMC6742450.
4. Raveendran S, Babu SP H, Kumar ML A, Iyoob V, Natesh B. Adrenal Adenoma–Hemangioma: A Unique Collision Tumor. World J Endoc Surg 2011; 3 (3):125-127.
5. Yuzhi Zuo, Zhen Liang, Shengmin Yang, Boju Pan, Sihang Cheng, Zhien Zhou, Tianrui Feng, Weigang Yan, Xingcheng Wu, Clinical Characteristics of Adrenal Hemangioma, *Journal of the Endocrine Society*, Volume 8, Issue 5, May 2024, bvae041, https://doi.org/10.1210/jendso/bvae041

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