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Inflammation and infection

Unilateral adrenal histoplasmosis

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Introduction

Histoplasmosis capsulatum is a ubiquitous 2- to 4-μm yeast with narrow based-buds that is endemic to certain regions of the southern United States as well as regions of Africa, Central and South America, Europe, and Asia. Histoplasmosis is acquired via inhalation of the yeast from soil or caves that contain bird or bat droppings. People affected by histoplasmosis may have a spectrum of symptoms from completely asymptomatic to acute or chronic pulmonary disease. Some may even have disseminated infection. Common sites of dissemination may include bone marrow, the liver or spleen, skin, or in the GI tract. Less commonly the central nervous system can be affected.[1] Adrenal involvement is usually a sequela of previous infection or is seen bilaterally with disseminated histoplasmosis. It is not uncommon to have focal organ involvement with minimal symptoms. Disseminated histoplasmosis is uncommon but when present, frequently affects the adrenals bilaterally. Bilateral adrenal involvement can present as Addison's disease.[2] The presentation of a unilateral adrenal lesion in an asymptomatic immunocompetent host is extremely rare based on extensive literature searches.

Case report

A 69 year old asymptomatic male from Amite City, Louisiana with past medical history of Hepatitis B on no medications was found to have an incidental right adrenal mass on a non-contrasted computed tomography (CT) scan of the abdomen performed for evaluation of chronic low back pain. Subsequent CT scan of the abdomen with and without contrast as well as with delayed images showed a 3 cm × 3 cm x 2.8 cm adrenal mass, with peripheral contrast enhancement (Fig. 1). Delayed images showed an approximately 20% contrast washout concerning for a possible malignancy. Magnetic Resonance Imaging (MRI) showed a right 3.1 cm × 2.5 cm isointense T1 and isointense T2 adrenal mass. On T1, out of phase images did not show signal dropout which could be consistent with a lipid poor adenoma. The adrenal mass did not have a high T2 signal like a pheochromocytoma (Fig. 2). The lesion was found to be non-functioning based on plasma free metanephrine level of 28 pg/ml (normal < or = 57), total metanephrine level of 85 pg/ml (normal < or = 205), free normetanephrine level of 57 ng/dl (normal < or = 148), normal cortisol, and normal plasma aldosterone to renin levels. His chest x-ray showed no abnormalities, specifically no masses or scarring. His liver function tests and CBC with differential revealed no abnormalities. He had a history of hepatitis B per patient report but was not tested for hepatitis or HIV at our facility. He had no hypertension, weight loss, or other complaints upon initial evaluation at a clinic visit. After options were discussed, the patient elected to undergo a right robotic assisted laparoscopic adrenalectomy. Final pathology revealed adrenal glandular tissue with necrotizing granulomatous inflammation (Fig. 3) which stained negative for acid fast bacilli and positive for fungal organisms. The fungal organisms are in yeast form without capsule, and are morphologically consistent with histoplasmosis. Special stains were GMS and PAS positive and AFB negative which is also consistent with histoplasmosis.[1] He returned for a post-operative follow up visit and was doing well. He was referred to infectious disease but elected to follow up locally. On phone interview he has not received any treatment with antifungal medication or developed any new symptoms.

Discussion

The presentation of unilateral focal adrenal histoplasmosis is extremely rare. In addition to cancer, the differential diagnosis for adrenal

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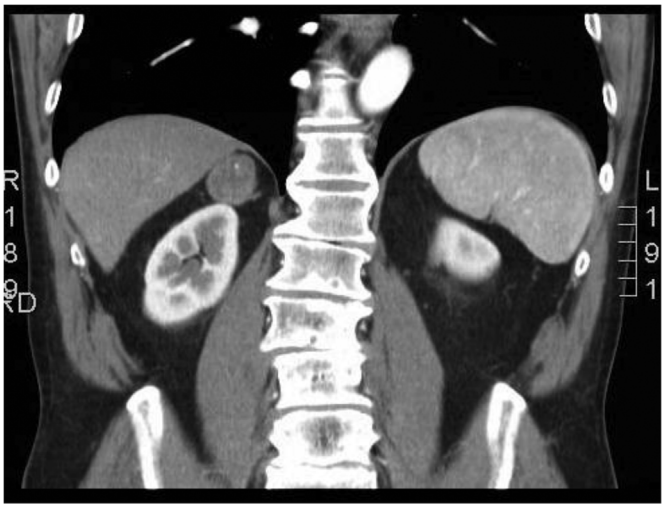

Fig. 1. Subsequent CT scan of the abdomen with and without contrast as well as with delayed images showed a 3 cm × 3 cm x 2.8 cm adrenal mass, with peripheral contrast enhancement.

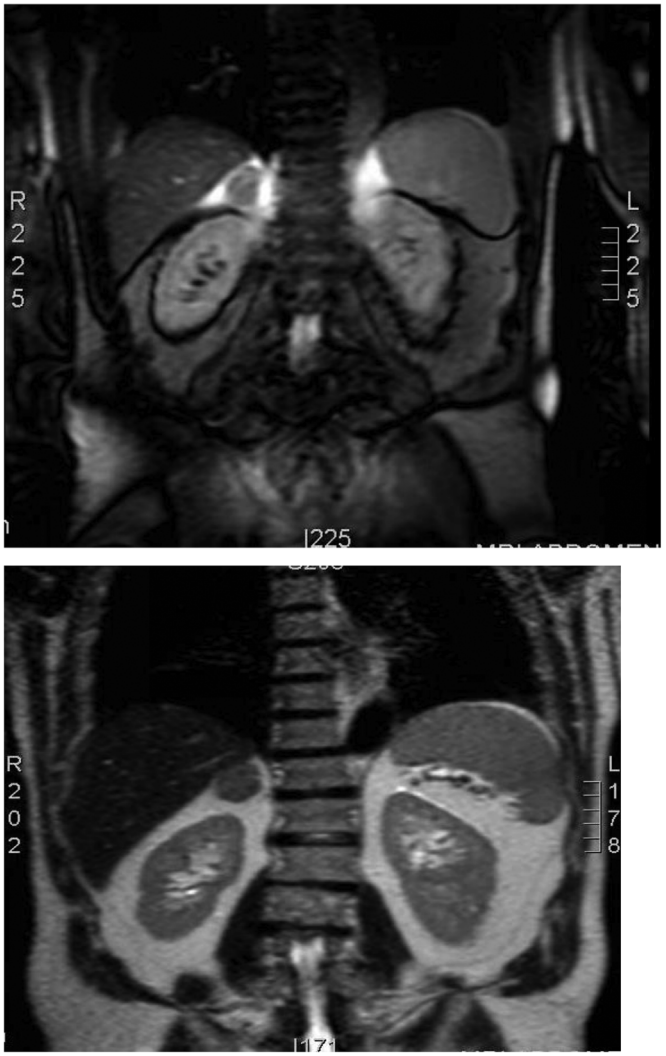

Fig. 2. Magnetic Resonance Imaging (MRI) showed a right 3.1 cm × 2.5 cm isointense T1 and isointense T2 adrenal mass. On T1, out of phase images did not show signal dropout which could be consistent with a lipid poor adenoma. The adrenal mass did not have a high T2 signal like a pheochromocytoma.

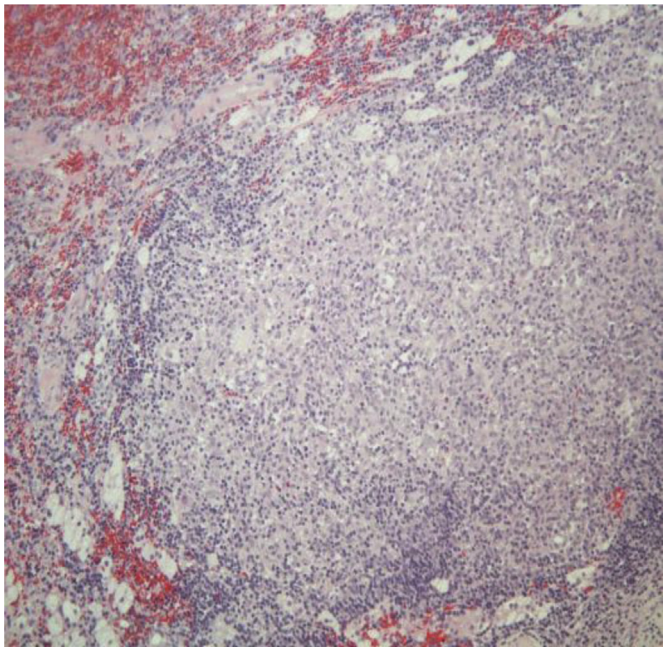

Fig. 3. Final pathology revealed adrenal glandular tissue with necrotizing granulomatous inflammation (Fig. 3) which stained negative for acid fast bacilli and positive for fungal organisms.

incidentalomas should include infectious etiology with consideration of tuberculosis, histoplasmosis, and blastomycosis in endemic areas. Our patient was born and lived in Louisiana which falls within the known exposure area of the Southern United States. The other main identified risk factor for histoplasmosis is immunocompromise including use of steroids, malignancy, and organ or stem cell transplantation.[3] Our patient did have a history of hepatitis B but normal liver function tests and CBC with differential indicating likely normal immune function. Patients with no symptoms of adrenal mass or systemic infection may be identified with radiologic tests. Common tests include MRI or CT scan. On contrast enhanced CT scan with delayed images, an absolute washout of more than 50% of the contrast medium was reported to be 100% sensitive and specific for adenoma in a comparison between patients with adenomas and those with carcinomas, pheochromocytomas, or metastatic disease. Our patient had 20% washout on his delayed image contrast enhanced CT which is more in line with possible carcinoma rather than a benign adenoma. MRI findings typical of a benign lesion include normal signal on in phase imaging with loss of signal on out of phase images.[4] Once again, our patient had no dropout of signal on out of phase T1 images. This could be consistent with a lipid poor adenoma but not a typical lipid rich adenoma. Other etiologies could include metastases. Based on our patients imaging, there was concern that his adrenal lesion was not a simple adenoma. This prompted his surgical resection. The data is lacking for how to manage solitary organ histoplasmosis especially when it is managed surgically as the general course for known histoplasmosis would be antifungal treatment. The mortality of untreated histoplasmosis can be as high as 80–100% but if treated with antifungals can be reduced to 25%. Treatment usually consists of amphotericin followed by itraconazole for 1–2 years.[2]

Conclusion

Our patient had a nonfunctioning adrenal lesion smaller than 4 cm suggesting a non-malignant etiology. However his abnormal washout of contrast on CT scan and abnormal signal intensity on MRI was worrisome for etiology other than benign adrenal lesions prompting the patient to undergo surgical removal. He was found to have adrenal

histoplasmosis despite being immunocompetent. In addition to cancer, the differential diagnosis for unilateral adrenal lesion in an immunocompetent patient should include histoplasmosis. Further research should be carried out to help elucidate ideal management of the patient with solitary solid organ involvement of histoplasmosis following surgical resection.

Appendix A. Supplementary data

Supplementary data related to this article can be found at http://dx.doi.org/10.1016/j.eucr.2018.03.010.

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