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Review Article

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Extra Pulmonary Sarcoidosis Disguised as Nodular Lung & Testicular Lesions

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Abstract

Introduction: *Sarcoidosis can affect multiple systems in the body especially the lungs and the skin. In our case, we highlight an extrapulmonary case of sarcoidosis masquerading as lung & testicular mass in a 30-year-old African-American male.*

Hospital Course: *An African American male without previous medical history presented with coughing and testicular swelling for over 1 month. The patient had adrastic weight loss, exertional dyspnea, and bilateral testicular swelling. Smoked daily one-fourthpack-per-day. On physical exam, patient had clear auscultation of the chest bilaterally and large left testicular mass measuring at least 6 cm. Laboratory tests show relatively unremarkable CBC and CMP. LDH was high at 238. Computed tomography of the chest/abd/pelvis without contrast showed innumerable pulmonary nodules in the bilateral lungs. Ultrasound of testicles found numerous bilateral hypoechoic testicular lesions. A right upper lung endobronchial biopsy revealed granulomatous inflammation without malignant cells related to pulmonary sarcoidosis with testicular involvement. ACE send-out was high at 138 U/L. Patient was started on steroids with 10 mg daily and doxycycline 100 mg twice daily for seven days.*

Discussion: *The "great mimicker" is a well-deserved moniker for sarcoidosis because of its multisystem involvement and broad symptomology it can be a challenging diagnosis. In order to diagnose sarcoidosis, a thorough history and physical exam are required. The genitourinary presentations of sarcoidosis are extremely rare. Extrapulmonary sarcoidosis can regress spontaneously or it can progress in select patients to malignancy which requires tailored treatment with either corticosteroids or orchiectomy.*

Keywords: Sarcoidosis, Testicular Lesions/Masses, Granulomatosis, Extra Pulmonary Sarcoidosis

Introduction

Sarcoidosis is a multisystem granulomatous disease that usually afflicts younger patients, but can span ages between 20 and 60 years without a defined etiology [1,2. The incidence of the disease is about 16.5/100,000 in men and 19/100,000 in women [1]. Sarcoidosis can present similarly to other diseases making it a difficult diagnosis and it is characterized by an abnormal growth of inflammatory noncaseating granulomas that affect multiple systems in the body [2,3]. It has been found in all organ systems and about 30% of cases are extra pulmonary [4]. Most cases of sarcoidosis occur in females and older population (>40 years) with skin, lung and eye involvement [4]. Overall genitourinary presentations of sarcoidosis are extremely rare at less than 0.2% [5].

In our case, we highlight a unique presentation of extrapulmonary sarcoidosis as bilateral testicular and pulmonary lesions with hydrocele in a young African-American male. This represents one of only 60 cases of sarcoidosis in the male reproductive tract that has been reported to date in our literature search [5].

Hospital Course

A 30-year-old African American male without previous medical history presented with coughing and testicular swelling for over 1 month. Patient was seen recently for his cough at a local ED and was prescribed antibiotics without improvement in his symptoms. On review of system, he admitted to loss of appetite with 60lbs weight loss in 9 months, exertional dyspnea, productive cough, and bilateral testicular swelling. Patient denied fevers, night sweats, headaches, visual changes, sinus pressure, hemoptysis, wheezing, chest pain, abdominal pain, nausea/vomiting, dysuria, fatigue or lower extremity edema. No past surgical history. He had no known drug allergies. Family history positive for maternal grandfather passing from cancer, but patient could not provide further history. He does use occasional alcohol and smokes cigarettes daily. One-fourth packs per day x 14 years. The patient works in a warehouse and occupational dust exposure. Lastly, admits to monogamous heterosexual relationship and uses barrier protections with condoms.

No history of any sexually transmitted infections.

Patient has a height of 67 inches and 185 lbs. Temperature 98.5 ˚F, pulse of 68 beats/min; blood pressure 119/75, respiratory rate of 16 breaths/min, and pulse ox of 98% on room air. During physical exam, the patient seemed well and in no distress. Sclerae were anicteric and conjunctivae were pink. Oral cavity did not have lesions. Neck was supple and thyroid was normal without nodules. There was no cervical, supraclavicular, or axillary adenopathy. Chest was clear to auscultation bilaterally and respirations were unlabored. Heart had a regular rate and rhythm without murmurs, rubs or gallops. Abdomen was soft, nontender, and nondistended with positive bowel sounds. There was no lower extremity edema. Strength and sensation were intact in the upper and lower extremities. Skin exam shows no rash or palpable nodules. Genitourinary exam showed overall normal appearance without urethral discharge or lesions Scrotal size normal. Large left testicular mass measuring at least 6 cm. Patient was alert & oriented x 3. Cranial nerves 2-12 grossly intact. No neurological deficits.

Laboratory tests show CBC with WBC of 3.9 k/UL, 13.8 Hgb, and 342 K/UL platelets (Figure 1). CMP was relative unremarkable with exception of total protein of 5.9 and albumin 3.5 which were low (Figure 1). Aspartate aminotransferase was high at 64. Other test of lactate dehydrogenase activity was high at 238. Hepatitis B surface was reactive, which indicated prior immunization. HCV antibody and HIV antigen/antibody were negative.

Computed tomography of the chest/abd/pelvis without contrast showed innumerable pulmonary nodules in the bilateral lungs most concentrated in the right upper lobe compatible with poss. metastatic disease (Figure 2&3). Enlarged mediastinal, hilar, retroperitoneal, and upper abdominal mesenteric lymph nodes compatible with nodal metastatic disease (Figure 4). Heterogeneous lesions of the bilateral testicles with moderate to large left-sided hydrocele (Figure 5). Ultrasound of testicles found numerous bilateral hypoechoic testicular lesions (Figure 6). MRI of the brain with contrast showed no evidence of intracranial metastatic disease.

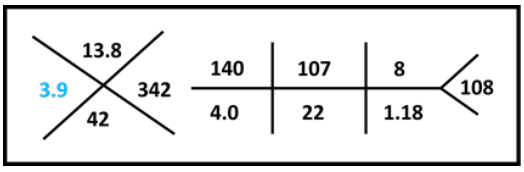
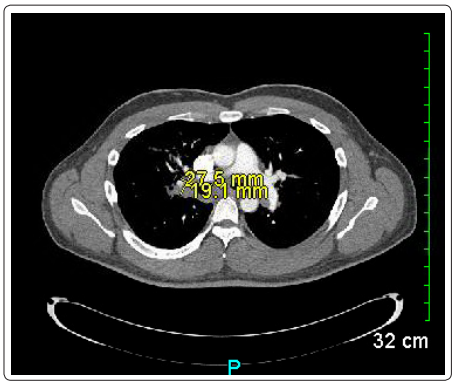


Figure 1: Patient's Labs. CBC and BMP relatively unremarkable



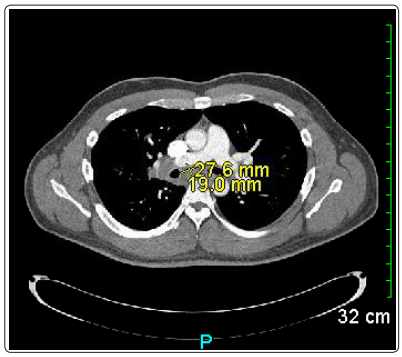


Figure 2: CT of Chest without Contrast

Heart size is within normal limits. No pleural or pericardial effusions. There are bulky mediastinal and hilar lymph nodes. Index lymph nodes are as follows: Subcarinal 2.8 x 1.9 cm. - Right hilum 2.7 x 1.9 cm. - Left hilum 1.5 x 1.9 cm. Aorta is normal in caliber. No dominant thyroid nodules.

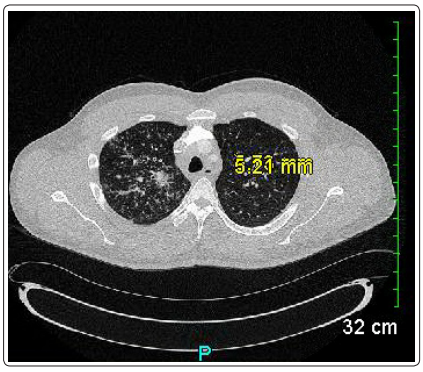


Figure 3: CT of Chest without Contrast

There are innumerable tiny 1 to 2 mm nodules seen throughout the bilateral lungs most pronounced in the right upper lobe. Dominant nodule in the right apex measures 1.2 x 1.1 cm. Dominant nodule in the left upper lobe measures 6 x 5 mm.

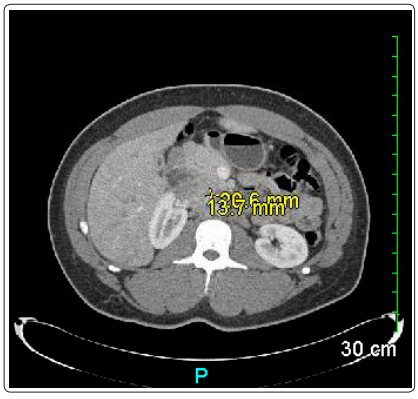


Figure 4: CT of Pelvis/Abdomen without Contrast

Bladder is nondistended. The liver, pancreas, spleen, and bilateral adrenal glands demonstrate no acute abnormalities. No hydronephrosis. No focal renal lesions. No bowel obstruction or free intraperitoneal air. Normal CT appearance of the appendix. Right para-aortic lymph node below the level of the renal vessels measures 2.1 x 1.4 cm. Left para-aortic lymph node below the level of the renal vessels measures 2.7 x 1.3 cm

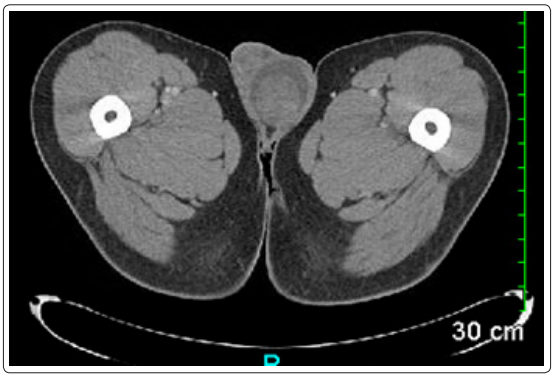


Figure 5: CT of Pelvis/Abdomen without Contrast

Urinary bladder is mildly distended without focal abnormality. Heterogeneous enhancement of the bilateral testicles. Moderate to large left-sided hydrocele. No free pelvic fluid. Prostate gland and seminal vesicles were unremarkable.

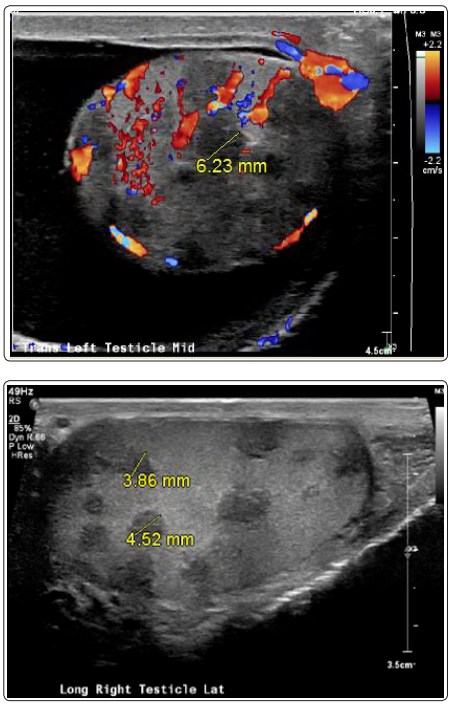


Figure 6: Testicular Ultrasound

A.Numerous bilateral hypoechoic testicular lesions, the majority are subcentimeter. None demonstrate overly increased vascularity. Otherwise, the testicles are normal in size and contour. Subjectively, testicular vascularity appears increased. The right testicle measures 4.1 x 2.6 x 3.6 cm. The left testicle measures 5.0 x 3.1 x 4.2 cm. The epididymides unremarkable. Moderate-sized left hydrocele with low-level echoes.

A CT-guided biopsy was deferred secondary to poor yield and instead radiology recommended bronchoscopy with biopsy. Results of right upper lung endobronchial biopsy showed granulomatous inflammation without malignant cells related to pulmonary sarcoidosis with testicular involvement. ACE send-out was high at 138 U/L. Patient was started on steroids with 10 mg daily and doxycycline 100 mg twice daily for seven days.

Discussion

The "great mimicker" is a well-deserved moniker for sarcoidosis because of its multisystem involvement and broad symptomology it can be a challenging diagnosis. Since its first description by Schaumann in 1936, the exact cause of sarcoidosis is unknown, but manifestations of altered immune systems have been found to be the direct causes affecting its pathological process [6-8]. It presents in the chest in over 80% of cases and thus the symptoms are cough, dyspnea and pleurisy chest pain [9]. In order to diagnose sarcoidosis, a thorough history and physical exam are required. Laboratory testing usually include: a complete blood count with differential, liver function tests, blood urea nitrogen, creatinine, glucose, electrolytes, serum calcium, and urinalysis [10]. The serum marker angiotensin converting enzyme (ACE) can be elevated in over 70% of untreated patients [11]. However, due to poor sensitivity (false negative results) and specificity it holds limited utility as a diagnostic test [11]. Serum soluble interleukin-2 receptor (sIL2R) has emerged as a useful marker for determination of extrapulmonary involvement in sarcoidosis [12]. Imaging studies such as chest x-ray, computerized tomography (CT scan), and positron emission tomography (PET) can be very helpful in aiding the diagnosis [10, 11]. Flexible bronchoscopy with bronchoalveolar lavage (BAL), endobronchial biopsy, and transbronchial biopsyhave been the traditional method to diagnosis pulmonary sarcoidosis [13]. Also, endoscopic ultrasound guided needle aspiration of intrathoracic lymph nodes via esophageal endoscopic ultrasound (EUS) or endobronchial ultrasound (EBUS) and surgical biopsy can be employed [13].

As already noted, the genitourinary presentations of sarcoidosis are extremely rare [5]. It has been noted to be highly prevalent in African-American males [13].When sarcoidosis involves the urogenital system, it can occur in the epididymis, followed by the testes, spermatic cord, prostate, scrotum, and penis [14].The usual presentation is painless testicular swelling, diffuse scrotal mass, nephroncalcinosis and epididymo-orchitis [15]. Testicular sarcoid presents with diffuse, nodular and painless mass of one testis and can be associated with epididymis [15, 16]. It can be seen as either a single hyper/hypoechoic lesion on testicular ultrasound bilaterally or a low signal on T2 weighted images of the MRI [15].

The association between testicular cancer and sarcoidosis has been extensively reported [17]. Currently, it seems that there is a higher incidence of testicular cancer especially seminoma in patients with sarcoidosis, but this can be partly a reaction to the tumor mass then an inciting event [17, 18]. Nevertheless, it is recommended that orchiectomy be performed in cases where a benign process cannot be excluded [19]. Open exploration of bilateral testis with resection or ultrasound-guided biopsy can further risk stratify patients. Also, this method can provide clarity in terms of preservations of long-term fertility as corticosteroids are effective in reducing testicular lesions and manifestations of sarcoidosis [20].

Conclusion

Patients presenting with testicular lesions require rigorous history and physical examinations in order to identify extrapulmonary sarcoidosis and prevent unnecessary procedures or interventions. Imaging studies such as ultrasound sonography of the testicles, lab markers for exclusion of testicular malignancy and pathology of the lesions are all essential. Vigilant monitoring of extrapulmonary sarcoidosis is required as this condition can regress sponteously or it can progress in select patients to malignancy which requires tailored treatment.

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