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EXCEPTIONAL CASE

Infected renal cyst presented with pleural effusion in a woman with autosomal dominant polycystic kidney disease

Ester Casillas-Sagrado¹, Victor Burguera¹, Maria Eugenia Rioja-Martín² and Maite Rivera-Gorrín¹

¹Department of Nephrology, Hospital Universitario Ramon y Cajal, Madrid, Spain and ²Department of Nuclear Medicine, Hospital Universitario Ramon y Cajal, Madrid, Spain

Correspondence and offprint requests to: Ester Casillas-Sagrado; E-mail: estercsag@gmail.com

Abstract

We present a 64-year-old woman with autosomal dominant polycystic kidney disease and hepatic cysts admitted to our hospital for high fever, intense coughing and right abdominal pain. The chest X-ray showed right pleural effusion suggestive of pneumonia. An abdominal ultrasound and computed tomography (CT) were done but did not show evidence of cyst infection or other abdominal complications. A gallium-67-citrate single-photon emission CT/CT, a relatively cheaper technique than positron emission tomography/CT was performed. This revealed an infected kidney cyst that was the cause of the right pleural effusion and fever.

Key words: autosomal dominant polycystic kidney disease, cyst infection, gallium-67-citrate, pleural effusion, SPECT/CT

Introduction

Renal cyst infection is a serious complication that affects autosomal dominant polycystic kidney disease (ADPKD) patients and causes >15% of the hospital admissions of these patients [1].

Among the possible contributing factors are age, female sex and recent instrumentation of the genito-urinary tract and it is difficult to determine the causal microorganism. In fact, <50% of cases have a positive culture and Escherichia coli is the most frequent pathogen isolated [2].

Diagnosis of renal cyst infection is determined by drainage of the cyst fluid showing bacteria and leucocytes [3]. However, when this is not accessible, diagnosis can be considered 'probable' based on two of the three following clinical criteria:

high fever ($>38^{\circ}$ C) for at least 3 days, abdominal pain and C-reactive protein plasma level >5 mg/dL [1].

Imaging examinations are recommended for cases without improvement of the symptoms after 48 h of antibiotic treatment or those with unusual clinical presentation [1]. Imaging tests showing either the presence or absence of cyst infection can be critical to decide the need for hospitalization and the duration of antibiotics [3]. Abdominal ultrasound and contrast-enhanced computed tomography (CT) are most commonly performed, although 18-fluorodeoxyglucose positron emission tomography/CT (18F-FDG-PET/CT) is more sensitive for localizing the infected cyst [3].

In recent years, gallium-67-citrate scintigraphy has been improved by the incorporation of single-photon emission CT/CT

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(SPECT/CT) technology and can be useful for diagnosing ADPKD patients.

Clinical case

We present the case of a 64-year-old woman diagnosed of ADPKD without a history of cyst complications. She had previously consulted her health centre for dysuria and fever and was treated with ciprofloxacin with moderate improvement of her

She was admitted to our hospital one week later with coughing attacks, right back pain and persistent high fever (>39°C). On physical examination she only presented hypoventilation at the right lung base. Blood analysis showed elevated inflammatory markers (C-reactive protein 232 mg/dL, procalcitonin 0.29 mg/dL, total leucocyte count 9410/mm³ with 8230/mm³ neutrophils). The urinalysis showed mild bacteriuria and pyuria. Blood, urine and sputum cultures were sterile. The abdominal ultrasound and abdominal-pelvic CT did not show evidence of cyst infection or renal lithiasis and the chest X-ray showed right pleural effusion (Figure 1).

Right basal pneumonia in addition to a urinary tract infection was suspected and treatment was intensified with meropenem. One week later high fever persisted and a second chest Xray showed no improvement of the pleural effusion. The respiratory study was then broadened with a cardiothoracic scan, thoracocentesis and a fibrobronchoscopy. The scan showed the presence of moderate pleural effusion with a collapsed basal lung lobe but no infiltrates compatible with pneumonia. The pleural fluid had inflammatory characteristics and imperceptible levels of adenosine deaminase and the bronchoscopy was normal. Sputum cultures and lung cytologies were performed but no pathogen or cellular abnormalities were found.

Since no evidence of an infection source had been found so far, a ⁶⁷Ga-SPECT/CT was performed. It revealed the presence of an infected renal cyst in the right kidney (Figure 1) that was considered the cause of her pleural effusion. It could be confirmed by periodic $^{67}\text{Ga-SPECT/CT}$ performed every 2 weeks until the patient had successfully completed 6 weeks of treatment with meropenem: lower gallium uptake of the cyst was coincidental with progressive improvement of the pleural effusion.

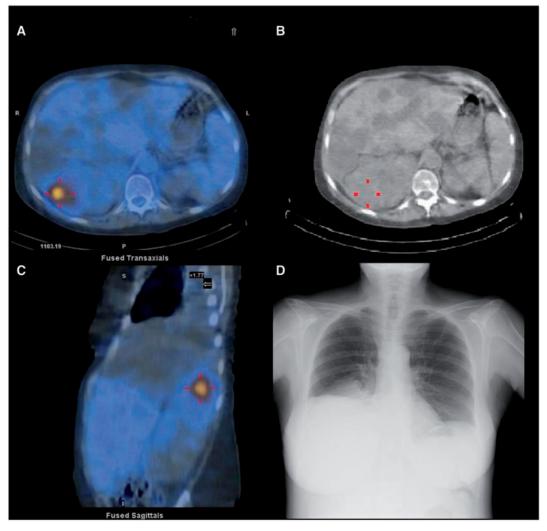


Fig. 1. SPECT/CT sections showing gallium uptake of an infected cyst in the upper pole of the right kidney (A,C). CT image of the same abdominal section (B). Chest radiography with right pleural effusion (D)

Discussion

The symptoms of persistent coughing attacks and pleural effusion have not previously been associated with kidney cyst infection in ADPKD patients even though they are known to be caused by hepatic cyst infection [4] in these patients and can also be provoked by single-cyst infection and pyelonephritis in the general population [5].

In our case, the patient's kidney had such a large growth that it had pushed the liver forward and it stayed in direct contact with the diaphragm. The prolonged inflammation of the cyst may have spread to the diaphragm, reaching the visceral pleura and forming the pleural effusion.

The 'probable' diagnosis of renal cyst infection is based on clinical criteria, but these are not homogeneous and vary in the different clinical guidelines [3]. Abdominal ultrasound is considered useful to exclude other urinary complications, like lithiasis or obstruction, but has little sensitivity for cyst infection (15-50%). Abdominal CT has better sensitivity, but this is only improved by using iodinated contrast, which is not recommended in ADPKD patients with advanced kidney disease. The alternative use of indium 111-marked leucocyte scintigraphy to localize the inflammatory focus has been replaced by ¹⁸F-FDG-PET/CT. However, PET/CT is an expensive technology and at the time of our patient's admission was unavailable in our hospital. For this reason we performed a ⁶⁷Ga-SPECT/CT, a cheaper imaging technology not included in the current guidelines, but which allowed the correct diagnosis of the patient. This technique could be an appropriate tool for the diagnosis and management of infected renal cysts.

Conflict of interest statement

None declared.

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