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Case Report

Primary and Isolated Renal Hydatid Cyst in Systemic Lupus Erythematosus

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Keywords

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Abstract

The primary renal localization of the hydatid cyst (HC) is exceptional and unusual. It is one of the so-called "aberrant" or "ectopic" localizations of human echinococcosis with an overall prevalence estimated at 1.5-5% but is exceptionally isolated and primitive.

The association of echinococcosis with Systemic Lupus Erythematosus (SLE) remains exceptional and suggests a hypothetical causal link.

We report the original observation of a primary and isolated renal HC diagnosed in a 59-year-old Tunisian woman followed for SLE.

Our observation is, to our knowledge, the fourth to report this association. It is characterized by its exceptional location (kidney) and its isolated and primitive character.

Thus a screening for hydatidosis seems to be beneficial for subjects monitored for lupus disease in the endemic areas of this parasitosis.

ABBREVIATIONS

ALAT: Alanin-Aminotransferase; ASAT: Aspartat-Aminotransferase; CB: Conjugated bilirubin; PT: Prothrombin Time; TB: Total Bilirubin

INTRODUCTION

Systemic lupus erythematosus (SLE) is a multi-system autoimmune disease whose pathogenesis involves an often inducing infectious factor [1]. Infections are also a common complication and a leading cause of death during lupus disease [2,3]. Among the infections mentioned, parasitic ones are not usual [1].

The primary renal localization of the Hydatid Cyst (HC) is exceptional and unusual. It is one of the so-called "aberrant" or "ectopic" localizations of human echinococcosis [4-7]. Its overall prevalence is estimated at 1.5-5% [8,9] but it is exceptionally isolated and primitive [8,9]; it is most often secondary (associated with a pulmonary and/or hepatic localization) or else integrates into a diffuse form of echinococcosis with multi-organ involvement [8,9].

The association of echinococcosis with SLE remains exceptional and, to our knowledge, only three cases have already been reported in the world medical literature [10-12].

As exceptional as it is, this association suggest a hypothetical causal link. Indeed human echinococcosis can be the cause of several dysimmune disturbances and even real autoimmune diseases. For SLE, the induction of anti-nuclear autoantibodies and lupus anticoagulant (specific autoantibodies of SLE) has been proven during human hydatidosis [10-12].

We report the original observation of a primary and isolated renal HC diagnosed in a patient followed for SLE.

OBSERVATION

A 59-year-old Tunisian woman, with a history of autoimmune thyroiditis (Hashimoto thyroiditis) for 10 years on 75µg/d of thyroxine, and of SLE for 12 years with cutaneous, articular and hematological involvement treated with hydroxychloroquine 400mg/d and salicylated acid 100 mg/d, was explored for isolated left low back pain evolving for three months without fever or associated urinary signs.

The somatic examination found an apyretic patient with a well preserved hemodynamic and general condition. There were no active skin lesions or other specific clinical signs of lupus disease. Likewise, his lumbar spine was flexible and painless when mobilized, and there was no pain in the shaking of the two sides.

His basic biological assessment was without abnormalities (total blood count: leukocytes at 7700/mm^3, hemoglobin at 13g/dl, and platelets at 248000/mm^3), erythrocyte sedimentation rate (35mm/H1), C-reactive protein (6 mg/l), creatinine (70 µmol/l),

liver tests (ASAT at 11 IU/l, ALAT at 19 IU/l, TB at 9.9 µmol/l, CB at 8 µmol/l, and PT at 100%), fasting blood sugar (4.16 mmol/l), Addis count (red cells = 1/min and Leucocytes = 1/min), urine direct bacteriological examination, and culture.

The standard radiographs (lumbar spine, pelvis, and urinary tree without preparation) were without significant abnormalities.

The abdominal ultrasound showed a cystic left renal mass, measuring 15 mm in diameter, and with a calcified wall (Figure 1). The abdominal computed tomography with the uro-scanner showed a left renal cystic and homogeneous mass, measuring 18mm in diameter, with a regular and calcified wall (Figure 2), not taking the contrast product (Figure 3), and having no mass effect (Figure 4).

The hydatid serology was positive confirming the diagnosis of calcified renal HC (type V of Gharbi). The thoraco-abdomino-pelvic computed tomography did not objectify other hydatic localizations confirming the isolated and primitive character of the renal HC.

The patient had symptomatic treatment for her low back pain. Surgery was not indicated given the calcified nature of the cyst and the absence of repercussions on the renal function and the excretory tracts.

DISCUSSION

Echinococcosis is an anthropozoonosis still endemic in several countries of the world and represents a real public health problem [8,9]. In humans, the main localizations for this parasitosis are the liver (65-70%) and the lungs (25%) [4-7].

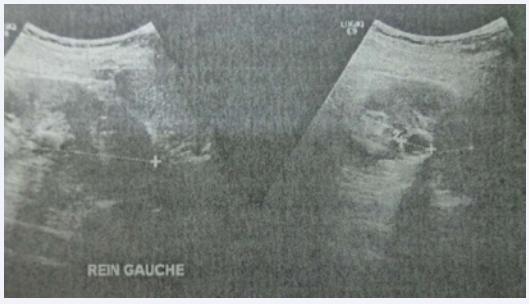

Figure 1 Renal ultrasound: left medio-renal cystic mass with a completely calcified wall.

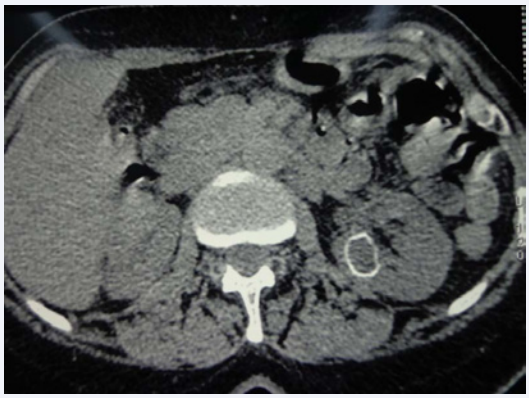

Figure 2 Abdominal CT in axial section without contrast: left renal cystic mass with calcified wall, and without mass effect.

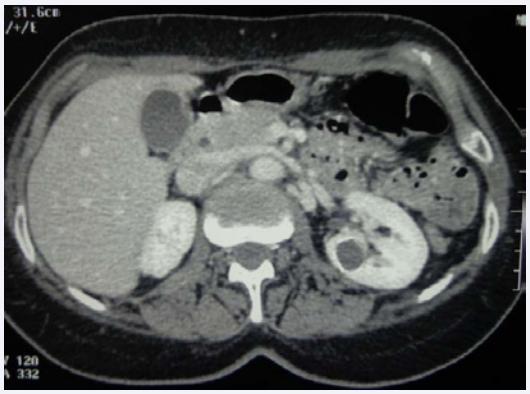

Figure 3 Abdominal CT in axial section with contrast injection: left renal cystic mass, with calcified wall, without mass effect, and not taking the contrast product.

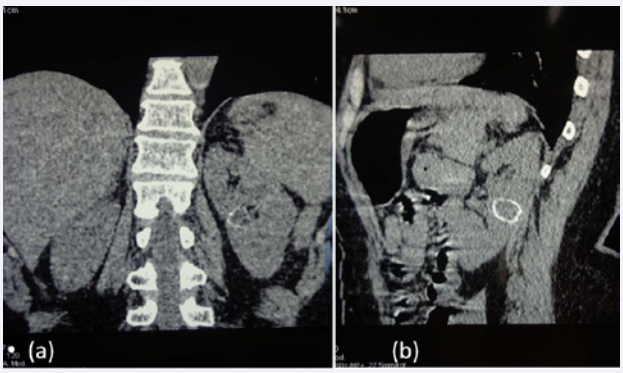

Figure 4 Abdominal CT in three-dimensional reconstruction in coronal (a) and sagittal (b) section without contrast: left renal cystic mass with calcified wall, and without mass effect.

The other organs and viscera, including the kidneys, are only rarely affected [4-9]. The primitive forms of these locations are exceptional: only 0.9% in the series of Lianos GD et al, of 233 human HC of unusual locations collected over 33 years of experience [13].

Only a few sporadic observations of HC have been reported during SLE [10-12]. Jallouli M et al, found only one case of pulmonary hydatid cyst in their series of 185 patients followed for SLE [11]. These observations were characterized by their invasive, bulky, disseminated, difficult to diagnose, and sometimes even fatal character [10-12].

These observations suggest a hypothetical causal link [14]. Probably by its excretory/secretory products, *Echinococcus granulosus* (*E. granulosus*), actively influences both innate and adaptive human immunity. It was demonstrated that several parasite molecules/antigens (particularly antigen B, AgB) are highly immunogenic and the surface of *E. granulosus* is able to activate the alternative pathway of the complement system [14-16].

All these immune modulatory properties of *E. granulosus* can lead to autoimmune reactions during this parasitic infection, particularly in genetically predisposed person [14-16]. Parasite-induced impairment of human immune reactions could explain the appearance of many autoimmune diseases: autoimmune hemolytic anemia, autoimmune polyendocrine syndrome, autoimmune cytopenia, psoriasis [14-16].

Additionally, it was demonstrated that many autoantibodies may be induced by human echinococcosis such us: anti-nuclear, anti-phospholipid, anti-cardiolipin, anti-smooth muscle, anti-lactopherin, anti-neutophil, lupus anticoagulant, and anti-myelo-proteinase [14-16]. Some of these autoantibodies (anti-nuclear, lupus anticoagulant, anti-phospholipid, and anti-cardiolipin) are specific of SLE [1-3].

The disappearance of these immune abnormalities after radical surgical treatment of hydatidosis is an argument comforting this direct causal link [16].

CONCLUSION

Our observation is, to our knowledge, the fourth to report the association of human echinococcosis with lupus disease. It is characterized by its exceptional location and its isolated and primitive character.

Thus a screening for hydatidosis seems to be beneficial for subjects monitored for lupus disease in the endemic areas of this parasitosis.

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