OMCR

Oxford Medical Case Reports, 2015;4, 269–271

doi: 10.1093/omcr/omv032
Case Report

CASE REPORT

Yersinia pseudotuberculosis aortitis in a patient with diverticulosis and polycystic kidney disease

Sarah McCloskey[1], Philip Haslam[2], David A. Price[3], and John A. Sayer[1,4,*]

[1]Renal Unit, Freeman Hospital, Newcastle upon Tyne Hospitals NHS Foundation Trust, Newcastle upon Tyne, UK, [2]Departmarnt of Radiology, Freeman Hospital, Newcastle upon Tyne Hospitals NHS Foundation Trust, Newcastle upon Tyne, UK, [3]Department of Infectious Disease, Royal Victoria Infirmary, Newcastle upon Tyne Hospitals NHS Foundation Trust, Newcastle upon Tyne, UK, and [4]Institute of Genetic Medicine, Newcastle University, Newcastle upon Tyne, UK

*Correspondence address. Institute of Genetic Medicine, Newcastle University, Central Parkway, Newcastle upon Tyne NE1 3BZ, UK. Tel: +44-191-2418608; Fax: +44-191-2418666; E-mail: john.sayer@ncl.ac.uk

Abstract

An 81-year-old gentleman with chronic kidney disease presented with pyrexia and a new systolic cardiac murmur. Investigations revealed infective aortitis of a pre-existing aortic aneurysm graft repair. Peripheral blood cultures were positive for *Yersinia pseudotuberculosis* and the patient was successfully treated with an extended course of antibiotics. Abdominal imaging also revealed progressive bilateral polycystic kidney disease with associated diverticular disease, which was postulated as the source of the *Y. pseudotuberculosis*. An autosomal dominant polycystic kidney disease may present late in life and extra-renal manifestations of this disease are an important cause of morbidity.

INTRODUCTION

Autosomal dominant polycystic kidney disease (ADPKD) is a disorder whereby the normal kidney architecture is replaced by numerous renal cysts. Clinical features are usually evident by the third or fourth decade with the most common presenting symptoms being loin pain or episodic haematuria associated with cyst rupture [1]. However, patients can have indolent disease with asymptomatic progression of renal dysfunction with the disease being discovered only after investigations [such as renal ultrasound scan (USS)] are undertaken for hypertension or chronic kidney disease (CKD). Often asymptomatic patients are diagnosed by a screening USS, following identification in an affected relative. Some patients present with symptoms secondary to the associated extra-renal manifestations of ADPKD rather than polycystic kidneys. Extra-renal manifestations are common and include sub-arachnoid haemorrhage due to intracranial aneurysm (which has a prevalence of ~12%), extra-renal cysts [involving the liver disease (94%), seminal vesicle (40%) and pancreas (9%)] and connective tissue abnormalities including thoracic aortic aneurysms, mitral valve prolapse (25%), abdominal hernia (10%) and diverticulosis [2, 3].

CASE REPORT

An 81-year-old gentleman presented with a 4-month history of progressive lethargy, intermittent pyrexia and a new systolic cardiac murmur. He had no urinary or gastrointestinal symptoms. His past medical history included systemic hypertension, diagnosed at 50 years of age, a graft repair of an infra-renal abdominal aortic aneurysm (AAA) at 74 years of age (at which time cystic change in both kidneys was noted incidentally but not followed up) and bilateral total hip replacements aged 78 years of age.

Received: December 9, 2014. **Revised:** March 19, 2015. **Accepted:** March 25, 2015

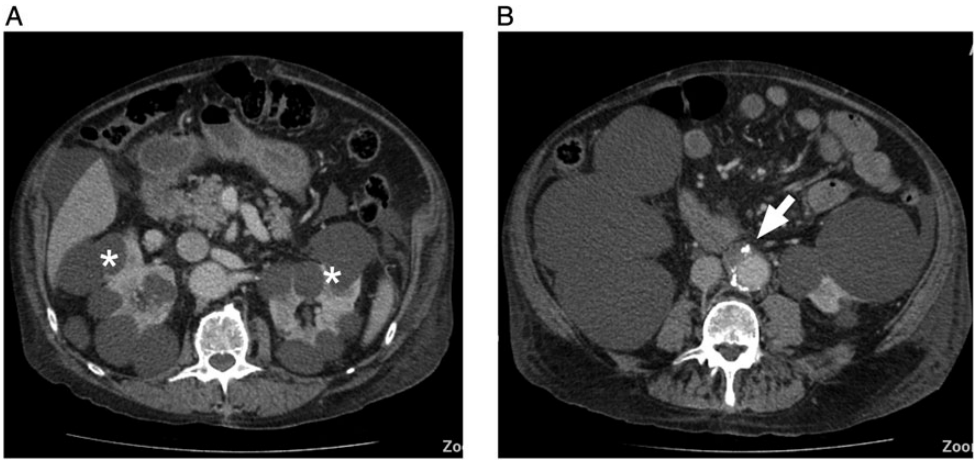

Figure 1: Abdominal CT scan. (**A**) Cystic change bilaterally within kidneys suggestive of ADPKD. Cystic kidneys are marked with asterisks. (**B**) Soft tissue mass (arrowed) anterior to the aortic graft suggestive of graft aortitis.

His primary care records had documented CKD stage 3 since 75 years of age. He was otherwise fit and active, an ex-smoker of 40 years and drank occasional alcohol. A paternal uncle had died of end stage renal disease (ESRD) at 48 years of age. There was no family history of sub-arachnoid haemorrhage.

On admission, he was pale, breathless on minimal exertion and lethargic. Initial blood tests revealed a haemoglobin of 7.4 g/dl, raised inflammatory markers (CRP peaking at 47 mg/l) and acute kidney injury (stage 1) with a serum creatinine of 169 µmol/l and a serum bicarbonate of 24 µmol/l (compared with a pre-admission baseline of 113 µmol/l). A chest X-ray was normal. Peripheral blood cultures grew *Yersinia pseudotuberculosis* after 24 h. The source of the sepsis was investigated using transthoracic and transoesophageal echocardiography, but these showed no evidence of cardiac valvular vegetations. A dual-phase bone scan and radiolabelled white cell scan showed no cause for symptoms, and a CT scan of the abdomen excluded any occult collections but revealed that the patient had bilaterally polycystic kidneys and several small liver cysts, consistent with a clinical diagnosis of ADPKD (Fig. 1A). A comparison with imaging performed prior to his AAA repair showed an increase in total kidney volume (measured using an ellipsoid equation) over the course of these 8 years, with right renal volume increasing from 954 to 1900 m and left renal volume increasing from 499 to 1360 ml. He was treated with intravenous Tazocin (piperacillin and tazobactam) for 7 days and then switched to oral amoxicillin. The patient completed 14 days treatment and was discharged home 41 days after his admission but represented after 14 days with on-going fever and further deterioration. Blood cultures taken on re-admission again grew *Y. pseudotuberculosis*, and the patient was commenced on doxycycline and ciprofloxacin. Repeat CT abdomen imaging showed evidence of mild graft aortitis and extensive sigmoid diverticulosis, which was postulated as the source of the infective organism (Fig. 1B). Further immunological screens and screens for infections for blood-borne viruses were negative. He was treated with a 6-month course of oral ciprofloxacin and doxycycline antibiotics and made a full recovery. His renal function improved to a new baseline serum creatinine of 150 µmol/l (eGFR 40 ml/min/1.73 m^2).

DISCUSSION

In patients with ADPKD, the kidneys are typically normal at birth and multiple cysts develop slowly over time. Around 68% of patients have evidence of renal cysts on imaging by age 30 and 50% of patients will have ESRD by age 60 [4]. However, the disease may also be clinically silent and may present in the later decades of life.

ADPKD has a number of extra-renal manifestations and it has been debated whether patients are at an increased risk of diverticular disease [3]. Several studies have shown no increased rate of diverticular disease in ADPKD patients with CKD when compared with patients in the general population with non-ADPKD CKD [5]. Any association is confounded by the high incidence of diverticulosis in the general populations, which may be as high as 45% [3]. However, a number of small studies have shown an increased incidence in patients with ESRD with ADPKD when compared with those with ESRD due to other aetiologies [6]. These studies also demonstrate more severe diverticular disease in patients with ADPKD requiring a higher rate of surgical intervention. There are also conflicting reports regarding the association of AAA with ADPKD, but the latest reports conclude that there is no definite association [7]. Similarly, there are also discussions around the apparent association of dissection of the thoracic aorta with ADPKD [8].

Yersinia pseudotuberculosis is a pathological organism that causes infections in mammals and birds and is contracted in humans through consumption of contaminated food products. It often presents with abdominal pain and fever, and has been documented to be causative in a number of bowel pathologies including appendicitis, terminal ileitis, mesenteric lymphadenitis and diverticulitis [9]. There are very few documented cases of overt bacteraemia in the literature and those identified occurred in patients with underlying disorders such as hepatic dysfunction with iron overload or immunosuppression [10]. There are a small number of cases associated with renal transplant patients and no documented cases of *Y. pseudotuberculosis* infection occurring in patients with ADPKD. There is one reported case of AAA infected by *Y. pseudotuberculosis* in an elderly patient with underlying coronary heart disease [11] and a rare documented case of infectious aortitis [11].

Infection of endografts is rare (0.2–0.7% in a major series [12]) and requires a high index of suspicion given its non-specific presentation. A number of causative organisms are recognized, but there are no documented cases associated with *Y. pseudotuberculosis* infection. Graft infection is notoriously difficult to treat and often requires graft explantation; however, conservative management can be considered in patients with multiple co-morbidities [13]. The choice of antibiotics should be based on culture sensitivities and the ability for good tissue penetration.

In conclusion, our patient appears to have suffered a rare bacterial infective aortitis, presumed secondary to diverticular disease and associated with a recent clinical diagnosis of ADPKD.

CONFLICT OF INTEREST STATEMENT

None declared.

REFERENCES

1. Grantham JJ. Clinical practice. Autosomal dominant polycystic kidney disease. *N Engl J Med* 2008;**359**:1477–85.
2. Pirson Y. Extrarenal manifestations of autosomal dominant polycystic kidney disease. *Adv Chronic Kidney Dis* 2010;**17**: 173–80.
3. Luciano RL, Dahl NK. Extra-renal manifestations of autosomal dominant polycystic kidney disease (ADPKD): considerations for routine screening and management. *Nephrol Dial Transplant* 2014;**29**:247–54.
4. Nahm AM, Henriquez DE, Ritz E. Renal cystic disease (ADPKD and ARPKD). *Nephrol Dial Transplant* 2002;**17**:311–4.
5. Sharp CK, Zeligman BE, Johnson AM, Duley I, Gabow PA. Evaluation of colonic diverticular disease in autosomal dominant polycystic kidney disease without end-stage renal disease. *Am J Kidney Dis* 1999;**34**:863–8.
6. Lederman ED, McCoy G, Conti DJ, Lee EC. Diverticulitis and polycystic kidney disease. *Am Surg* 2000;**66**:200–3.
7. Bailey MA, Griffin KJ, Windle AL, Lines SW, Scott DJ. Cysts and swellings: a systematic review of the association between polycystic kidney disease and abdominal aortic aneurysm. *Ann Vasc Surg* 2013;**27**:123–8.
8. Nacasch N, Werner M, Golan E, Korzets Z. Arterial dissections in autosomal dominant polycystic kidney disease—chance association or part of the disease spectrum? *Clin Nephrol* 2010;**73**:478–81.
9. Leino R, Granfors K, Havia T, Heinonen R, Lampinen M, Toivanen A. Yersiniosis as a gastrointestinal disease. *Scand J Infect Dis* 1987;**19**:63–8.
10. Ljungberg P, Valtonen M, Harjola VP, Kaukoranta-Tolvanen SS, Vaara M. Report of four cases of *Yersinia pseudotuberculosis* septicemia and a literature review. *Eur J Clin Microbiol Infect Dis* 1995;**14**:804–10.
11. Hadou T, Elfarra M, Alauzet C, Guinet F, Lozniewski A, Lion C. Abdominal aortic aneurysm infected by *Yersinia pseudotuberculosis*. *J Clin Microbiol* 2006;**44**:3457–58.
12. Capoccia L, Mestres G, Riambau V. Current technology for the treatment of infection following abdominal aortic aneurysm (AAA) fixation by endovascular repair (EVAR). *J Cardiovasc Surg (Torino)* 2014;**55**:381–9.
13. Murphy EH, Szeto WY, Herdrich BJ, Jackson BM, Wang GJ, Bavaria JE, *et al*. The management of endograft infections following endovascular thoracic and abdominal aneurysm repair. *J Vasc Surg* 2013;**58**:1179–85.