



EXCEPTIONAL CASE

Atypical anti-glomerular basement membrane disease presenting as macroscopic haematuria, loin pain and acute kidney injury after intensive exercise

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ABSTRACT

We report a 35-year-old man who suffered from recurrent macroscopic haematuria after intensive exercise. One episode was associated with bilateral loin (flank) pain and severe acute kidney injury. His kidney biopsy revealed an atypical anti-glomerular basement membrane (GBM) disease typified by bright linear GBM staining for monotypic immunoglobulin G but without a diffuse crescentic phenotype and no circulating anti-GBM antibody. Outcome was spontaneously favourable. The patient had no recurrence or urine abnormality without running. The original presentation emphasized that exercise could reveal an underlying glomerulopathy.

Keywords: acute kidney injury, atypical anti-glomerular basement membrane disease, exercise, haematuria, loin pain

BACKGROUND

Post-exercise haematuria and post-exercise acute kidney injury (AKI) can reveal an underlying disease, for example, drepanocytosis, uropathy or renal hypo-uricaemia. Rarely Alport disease, immunoglobulin A (IgA) nephropathy or focal segmental glomerulosclerosis has been observed [1]. We report the case of a patient with an atypical anti-glomerular basement membrane (GBM) disease revealed by post-exercise AKI associated with macroscopic haematuria and loin pain.

CASE REPORT

A 35-year-old man without a medical history complained of recurrent macroscopic haematuria since he began training for a marathon. After each training session he noticed isolated haematuria in the first urination. His physical examination was normal. Serum creatinine (S_{Cr}) was $93 \mu\text{mol/L}$ and the estimated glomerular filtration rate was $95 \text{ mL/min/1.73 m}^2$. He had no proteinuria or haematuria. He was gradually increasing his training sessions.

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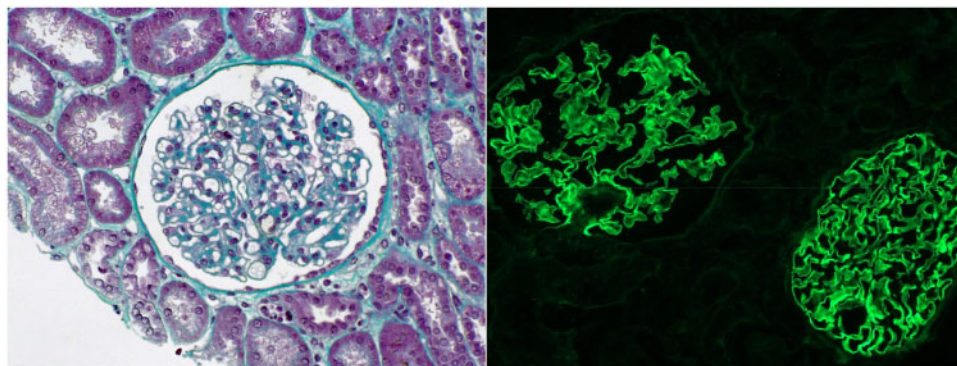


FIGURE 1: Renal biopsy. (A) Optic microscopy, Masson's trichrome stain: normal glomerulus. The tubulointerstitial compartment showed only minor alterations. (B) Immunofluorescence microscopy showed linear capillary loop staining for IgG.

One hour after running 22 km, he had several gross haematuria, which stopped by the end of the day. The next day he was admitted to the hospital because of bilateral loin pain radiating down the pelvis. His blood pressure was 137/75 mmHg and he was oliguric, but the rest of his physical examination was normal. He reported no medication or toxics use.

A computed tomography scan was normal. Laboratory findings were S_{Cr} 425 $\mu\text{mol/L}$, proteinuria/creatininuria 550 mg/mmol, haematuria 37/mm³ and leucocyturia 27/mm³. Autoimmune tests were normal, including anti-GBM (Bio-Flash, Werfen, Brussels, Belgium). Hepatitis C and B and human immunodeficiency virus serologies were negative. There was no evidence of rhabdomyolysis or haemolysis.

Two days after admission, the pain partially improved, but despite intravenous hydration, S_{Cr} increased to 657 $\mu\text{mol/L}$. A kidney biopsy was performed. The biopsy (Figure 1) showed 17 glomeruli, all normal in optic microscopy. The tubulointerstitial compartment showed only minor alterations, with rare foci of acute tubular necrosis and microvacuolizations of the proximal tubules. Red cell casts were absent and vessels were normal. Direct immunofluorescence showed linear capillary loop staining for IgG, with a κ light chain restriction, and IgG1 subclass predominance.

One day later, the flank pain totally disappeared and S_{Cr} decreased slowly to baseline in a month. Proteinuria and haematuria disappeared in a few days. Eight months later, urinalysis, renal function, uricaemia and serum protein electrophoresis were normal. The patient had no recurrence without running.

DISCUSSION

Recurrent post-exercise haematuria is reported in healthy subjects probably because of an impaired GBM selectivity during exercise. Rarely the persistence of haematuria leads to a glomerulopathy diagnosis [1].

Our patient also complained of loin pain and AKI. The loin pain haematuria syndrome was described in a cohort of 43 patients, half induced by exercise. IgA nephropathy was diagnosed in nine of them. More than 50% of the others had abnormal GBM width [2]. Exercise-induced AKI with loin pain is also reported in renal hypouricaemia, but usually without haematuria [3].

In our case, AKI might be related to macroscopic haematuria with red cell tubular obstruction or cytotoxic effects of oxidative stress induced by haemoglobin, as proposed by Moreno et al. [4].

The kidney biopsy revealed a linear GBM staining without crescent glomerulonephritis. This can be observed in diabetic nephropathy, monoclonal Ig deposition disease or in atypical anti-GBM disease [5]. We compared our case with the reported characteristics of atypical anti-GBM nephritis (see Supplementary data, Table S1).

This original presentation emphasizes that exercise can reveal an underlying glomerulopathy. Macroscopic haematuria can also induce AKI and loin pain regardless of the aetiology. We suggest that kidney biopsy should be considered in cases of post-exercise AKI associated with haematuria.

SUPPLEMENTARY DATA

Supplementary data are available at ckj online.

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The authors confirm that written consent for submission and publication of this case report, including images and associated text, has been obtained from the patient.

CONFLICT OF INTEREST STATEMENT

None declared.

REFERENCES

1. Varma PP, Sengupta P, Nair RK. Post exertional hematuria. *Ren Fail* 2014; 36: 701–703
2. Spetie DN, Nadasdy T, Nadasdy G et al. Proposed pathogenesis of idiopathic loin pain-hematuria syndrome. *Am J Kidney Dis* 2006; 47: 419–427
3. Ohta T, Sakano T, Igarashi T et al. Exercise-induced acute renal failure associated with renal hypouricaemia: results of a questionnaire-based survey in Japan. *Nephrol Dial Transplant* 2004; 19: 1447–1453
4. Moreno JA, Martin-Cleary C, Gutierrez E et al. AKI associated with macroscopic glomerular hematuria: clinical and pathophysiologic consequences. *Clin J Am Soc Nephrol* 2012; 7: 175–184
5. Nasr SH, Collins AB, Alexander MP et al. The clinicopathologic characteristics and outcome of atypical anti-glomerular basement membrane nephritis. *Kidney Int* 2016; 89: 897–908