Recurrent ureteric obstruction in association with Henoch-Schönlein purpura

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Summary: The most common urological manifestation of Henoch-Schönlein purpura is focal proliferative glomerulonephritis. Ureteric obstruction as a consequence of the disease has been reported in children but is rare.

We report an adult male patient with Henoch-Schönlein purpura causing unilateral, bi-focal, recurrent ureteric obstruction. We speculate that ureteric involvement in Henoch-Schönlein purpura may be more common than hitherto suspected and that even with a demonstrable obstructive episode, consideration must be given for an initial conservative approach.

Introduction

The syndrome of Henoch-Schönlein purpura (HSP) which is characterized by non-thrombocytopenic rash, arthritis, and gastro-intestinal disease is well documented in adults although it is more frequently seen in children. Glomerulonephritis accounts for the majority of deaths in this condition and is the most common renal tract complication. Testicular torsion can also occur, and ureteric stenosis has been described in children.

We present what we believe is the first reported case in an adult of Henoch-Schönlein purpura associated with ureteric stenosis. The stenosis was recurrent and on the second occasion it resolved spontaneously.

Case report

A 21 year old male patient presented with abdominal pain and distension, blood-stained vomitus, and watery diarrhoea. He gave a history of having had a sore throat and lower limb rash three months previously which had resolved spontaneously; more recently, the rash had recurred. On inspection there was a purpuric rash which was most prevalent over the buttocks and the front of the legs. The dorsae of both hands were markedly oedematous and there was a generalized arthralgia and stiffness affecting in particular the knees, ankles, and hands. The abdomen was tympanic, distended, and diffusely tender with increased bowel sounds. He had not experienced loin pain with this episode or previously. His blood pressure was 145/95 mm Hg.

Radiological investigations revealed small bilateral pleural effusions, several small bowel fluid levels, and some thickened small bowel mucosal folds with the typical ‘thumb printing’ pattern of oedematous mucosa. An intravenous urogram was normal.

Urinalysis revealed no protein, red blood cells, or casts, and haematological and serum renal function tests were normal. The serum cholesterol was low (2.7 mmol/l), and the albumin was 20 g/l. Serum DNA binding, complement C3, C2, and C4 levels, antinuclear factor and latex agglutination rheumatoid factor were normal or negative. Immune complexes were intermittently detected by the rheumatoid factor binding assay during the subsequent admission but bore no relationship to the course of his illness. Immunoglobulins were normal on both admissions.

His condition resolved with conservative treatment but 6 weeks later he developed haematuria, malaise, weight loss, and a generalized arthropathy. On this occasion there was no rash but he complained of constant left loin pain, increasing in severity. Urinalysis revealed red blood cells, hyaline casts, and a trace of protein. An intravenous urogram demonstrated a delayed left nephrogram and a mildly distended renal pelvis (Figure 1) with, in successive radiographs, a constant narrowing of the upper part of the left ureter causing an incomplete obstruction and post-stenotic dilatation. Conservative measures failed to resolve the pain which increased in severity, and 3

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deposition of IgA, with occasional flecks of C3, IgG, IgM, and fibrin. These findings were consistent with a diagnosis of Henoch-Schönlein purpura.

Four weeks following operation he developed further left loin pain and haematuria. Intravenous urography demonstrated a patent left upper ureter but another left ureteric stricture, this time at the level of the sacro-iliac joint (Figure 2). A retrograde ureterogram was performed two days later because of increasing loin pain, and complete obstruction was seen at the level of the sacro-iliac joint (Figure 3). The radiological features suggested that the obstruction was due to a mass of oedematous ureteric epithelium, and a conservative approach was therefore adopted with subsequent complete resolution of the pain. The haematuria resolved spontaneously and 4 weeks later an intravenous urogram showed no obstructive features.

Discussion

In the absence of any demonstrable infection, the diagnosis of Henoch-Schönlein purpura (HSP) was confirmed by the clinical features and subsequent histology.

Gastro-intestinal involvement is suggested by hypoproteinaemia in the absence of proteinuria, and a low serum cholesterol level – a known feature of HSP. The arthralgia combined with swelling of the dorsae of both hands is a characteristic, if not well documented, feature in this condition. The presence of mesangial IgA in the renal biopsy is a recognized feature in the pathology of HSP. All of these features were seen in this patient.

The most common genito-urinary manifestation of HSP is focal glomerulonephritis occurring in up to 90% of cases. Urteric involvement is rare and has not previously been reported in an adult. Including our patient, eleven cases have been reported; the other ten cases were in the age range of 5–11 years and were reviewed by Kher et al. The most consistent clinical features were loin pain and haematuria – either macroscopic or microscopic, and the most common site of ureteral obstruction was the uretero-pelvic junction although multiple ureteral segments were involved in three patients and isolated segments in two. Most cases progressed to ureteral stenosis and seven patients required surgical correction of the obstruction.

Histological changes in the ureter varied, but ureteritis was common to most cases and vasculitis, fibrosis, and calcification were also reported. Our patient showed all of these histological features.

It appears that in our patient, the two obstructive episodes were different in nature. The first stricture was caused by dense fibrosis – as seen macroscopically...
Figure 2 Intravenous urogram taken 6 weeks following upper left ureteroplasty. The upper ureter is patent but there is evidence of obstruction at the level of the sacro-iliac joint.

and microscopically, and it is improbable that resolution could have occurred spontaneously. The second stricture appeared to be caused by an oedematous mass of inflammatory tissue which did resolve with conservative treatment.

One wonders if episodes of ureteric obstruction occur more commonly in HSP than is generally thought. In such cases, consideration should be given for an initial conservative approach.

References

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