

**Table 1.** Case series

Series	Years	Cases	Incidence	Mortality (%)	Age	Mean time on HD (months)
Dahlberg <sup>(2)</sup>	1985	3*	1,5%	66,6	60,6	¿
Diamond <sup>(3)</sup>	1979-85	12	¿	75	60,3	40
Dumazer <sup>(7)</sup>	1989	10	¿	80	52	56
Bender <sup>(8)</sup>	1988-93	11	1,94%	50	56,9	79
Hung <sup>(9)</sup>	1986-97	5	0,2%	25	62,4	¿
Flobert <sup>(10)</sup>	1985-97	20**	¿	5	65	¿
John <sup>(11)</sup>	1992-97	29	2,11%	56 ID 31 IG	59,6	50
Bassilios <sup>(1)</sup>	1988-99	15	0,3/ patients/year	27	65,8	79,5
Ori <sup>(12)</sup>	1988-2002	19	¿	60 after 1 week, 85 after 1 year	70,6	67
Yu <sup>(13)</sup>	2007-2008	9***	0,95/ patients/year	33	71,5	19,9
Picazo <sup>(14)</sup>	1988-2007	11	0,87/ patients/year	73	71	¿
Quiroga <sup>(4)</sup>	2003-11	57	2,29/ patients/year	59	72,6	56
Gutiérrez	2008-13	5	6%	80	81	120

\*6 cases, 3 with nonocclusive ischaemic colitis. Non-homogeneous data.

\*\*Study with 60 patients, 20 on HD.

\*\*\*Study with 12 patients, 9 on HD.

HD: haemodialysis.

- Bassilios N, Menoyo V, Berger A, Mamzer MF, Daniel F, Cluzel P, et al. Mesenteric ischemia in haemodialysis patients: a case/control study. *Nephrol Dial Transplant* 2003;18(5):911-7.
- Quiroga B, Verde E, Abad S, Vega A, Goicoechea M, Reque J, et al. Detection of patients at high risk for non-occlusive mesenteric ischemia in hemodialysis. *J Surg Res* 2013;180:51-5.
- Ergün T, Lakadamyal H. The CT frequencies of various non-traumatic acute abdominal emergencies in hemodialysis, peritoneal dialysis patients and the general population. *Eur J Radiol* 2012;81(1):13-20.
- Vaziri ND, Dure-Smith B, Miller R, Mirahmadi MK. Pathology of gastrointestinal tract in chronic hemodialysis patients: an autopsy study of 78 cases. *Am J Gastroenterol* 1985;80:608-11.
- Dahlberg PhJ, Kiskan WA, Newcomer KL, Yutuc WR. Mesenteric ischemia in chronic dialysis patients. *Am J Nephrol* 1985;5:327-32.
- Diamond SM, Emmett M, Henrich WL. Bowel infarction as a cause of death in dialysis patients. *JAMA* 1986;256:2545-7.
- Dumazer PH, Dueymes JM, Vernier I, Thierry FX, Conté JJ. Ischémie mesentérique non-occlusive chez l'hémodialysé périodique. *Presse Med* 1989;18:471-4.
- Bender JS, Ratner LE, Magnuson TH, Zenilman ME. Acute abdomen in the hemodialysis patient population. *Surgery* 1995;117(5):494.
- Hung KH, Lee CT, Lam KK, Chuang FR, Hsiu KT, Chen JB, et al. Ischemic bowel disease in chronic dialysis patients. *Changeng Yi Xue Za Zhi* 1999;22(1):82.
- Flobert C, Cellier C, Berger A, Ngo A, Cuillierier E, Landi B, et al. Right colonic involvement is associated with severe forms of ischemic colitis and occurs frequently in patients with chronic renal failure requiring hemodialysis. *Am J Gastroenterol* 2000;95(1):195-8.
- John AS, Tuerff SD, Kerstein MD. Nonocclusive mesenteric infarction in hemodialysis patients. *J Am Coll Surg* 2000;190(1):84.

- Ori Y, Chagnac A, Schwartz A, Herman M, Weinstein T, Zevin D, et al. Non-occlusive mesenteric ischemia in chronically dialyzed patients: a disease with multiple risk factors. *Nephron Clin Pract* 2005;101(2):c87-93.
- Yu CC, Hsu HJ, Wu IW, Lee CC, Tsai CJ, Chou CC, et al. Factors associated with mortality from non-occlusive mesenteric ischemia in dialysis patients. *Ren Fail* 2009;31(9):802-6.
- Picazo M, Cuxart M, Sans R, Sardá C, Expósito E. Mesenteric ischemia in hemodialysis patients. *Nefrologia* 2008;28:198-202.

**María J. Gutiérrez-Sánchez<sup>1</sup>,**

**Vladimir Petkov-Stoyanov<sup>1</sup>,**

**Juan A. Martín-Navarro<sup>1</sup>,**

**Miguel López-Quiñones-Llamas<sup>2</sup>**

<sup>1</sup> Sección de Nefrología. Hospital del Tajo. Aranjuez, Madrid. (Spain)

<sup>2</sup> Servicio de Radiología. Hospital del Tajo. Aranjuez, Madrid. (Spain)

**Correspondence:** Juan A. Martín-Navarro  
Sección de Nefrología.

Hospital del Tajo. Avda. Amazonas Central, sn. 28300 Aranjuez, Madrid. (Spain)

juanmartinnav@hotmail.com

## Treatment-resistant Churg-Strauss syndrome: Progression after five years using rituximab

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### To the Editor:

Churg-Strauss syndrome (CSS) is a necrotising vasculitis of medium and small vessels that affects the respiratory tract with asthma and eosinophilia.<sup>1</sup> Renal involvement is rare, occurring in the form of pauci-immune necrotising glomerulonephritis.<sup>2</sup> Prognosis without treatment is poor, but with immunosuppressants, the survival reaches 80%. In recent years the use of rituximab has spread to ANCA positive vasculitis resistant to

conventional treatment, with encouraging results.<sup>3</sup>

## CLINICAL OBSERVATION

A 70-year-old male with a history of bronchial asthma was admitted due to pulmonary infiltrates, eosinophilia, and MPO-p-ANCA 1:160. The patient was diagnosed with CSS and he was started on 1mg/kg/day prednisone. After 20 days he showed deterioration in renal function, microhaematuria without proteinuria, high blood pressure and elevated p-ANCA 1:1320. A percutaneous renal biopsy was performed, and we observed glomeruli without significant abnormalities and a medium-sized artery with fibrinoid necrosis and transmural infiltration of neutrophils along with intense interstitial lymphoplasmacytic inflammatory infiltrate with abundant eosinophils (Figure 1). Using direct immunofluorescence (DIF) we detected IgG, IgM, C3, C4 and C1q vascular deposits.

In view of the active vasculitis data, treatment was started with three intravenous boluses of 1g of 6-methylprednisolone followed by 1mg/kg/day of oral prednisone and 2mg/kg/day of oral cyclophosphamide and began haemodialysis. After 15 days, we conducted a second renal biopsy, which displayed new vasculitic lesions (Figure 2). We carried out plasmapheresis, with four sessions being performed due to coagulation abnormalities. Cyclophosphamide was suspended due to thrombocytopenia and leukopenia. After the failure of previous therapeutic procedures, we decided to administer rituximab with an intravenous dose of 375mg/m<sup>2</sup> per week for four weeks and a tapering steroid dosage. In the following three months, renal function improved, the p-ANCA became negative and eosinophils decreased, and dialysis was discontinued. Due to pulmonary involvement, 5mg/day of prednisone was maintained, and no further doses of rituximab were administered (Figure 3). After five years, there was remission of the disease. No infectious complications were present.

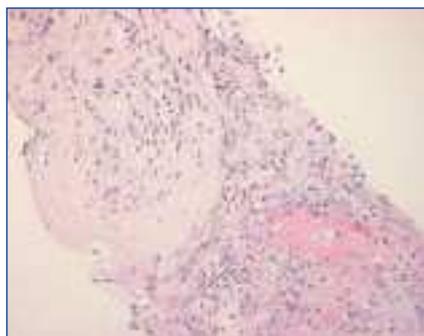


Figure 1. First renal biopsy (H-E).

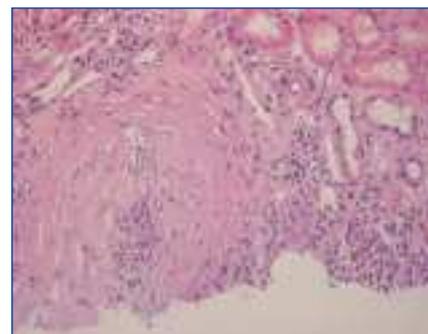


Figure 2. Second renal biopsy (H-E).

## DISCUSSION

CSS is a rare disease included in the group of positive ANCA vasculitis characterised by peripheral eosinophilia, asthma, migratory pulmonary infiltrates, mono-polyneuropathy and necrotising vasculitis of small and medium-sized vessels.<sup>4</sup> Renal involvement is rare and it is an unfavourable prognosis factor, and as such, immunosuppression should be assessed carefully.<sup>5</sup> Before the use of steroids and cyclophosphamide, the mortality rate was 50%, with infections being the main cause of death.<sup>6</sup> In vasculitis, we observed B lymphocytes involvement in the immune response, multiplying the expression of antigen-pre-

senting and cytokine-producing cells.<sup>7</sup> Rituximab is a chimeric murine monoclonal antibody that binds specifically to the CD20 membrane antigen expressed by B lymphocytes and its use results in their sustained depletion. For some years now, it has been used in non-lymphoma diseases with B lymphocytes abnormalities; it seems safe in the long term in positive ANCA vasculitis<sup>8-10</sup> as a treatment for relapses with or without other immunosuppressants and has been shown to be as effective as cyclophosphamide and with far fewer side effects.<sup>11,12</sup> The B lymphocytes depletion period is not well established and the doses used are 375mg/m<sup>2</sup>/week intravenously for four

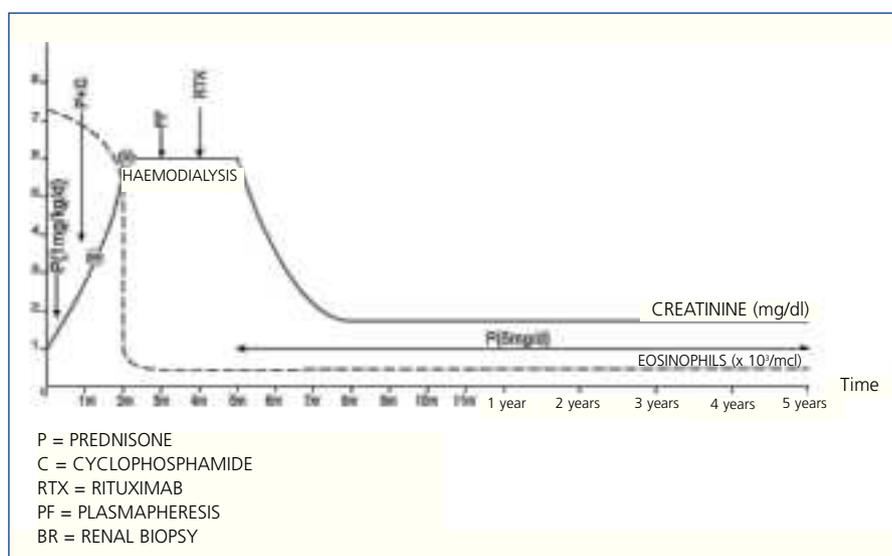


Figure 3. Treatment and progression.

RB: renal biopsy; C: cyclophosphamide; P: prednisone; PF: plasmapheresis; RTX: rituximab.

weeks, although some authors recommend an additional two doses per month. Prednisone is started at a dose of 1mg/kg/day (maximum 80mg daily) for a month with a progressive decrease in the next six months.<sup>13</sup>

### CONCLUSIONS

We present this case due to the excellent response and because rituximab had no adverse effects in treatment-resistant CSS with renal involvement. There are increasingly more publications that indicate rituximab in cases of treatment-resistant ANCA-associated vasculitis, but more studies will be needed in order to find drugs with safer profiles and with less risk to the patient.

### Conflicts of interest

The authors declare that they have no conflicts of interest related to the contents of this article.

- Noth I, Strek ME, Leff AR. Churg-Strauss syndrome. *Lancet* 2003;361:587-94.
- Clutterbuck EJ, Evans DJ, Pusey CD. Renal involvement in Churg-Strauss syndrome. *Nephrol Dial Transplant* 1990;5:161-7.
- Stone JH, Merkel PA, Spiera R, Seo P, Langford CA, Hoffman GS, et al. Rituximab versus cyclophosphamide for ANCA-associated vasculitis. *N Engl J Med* 2010;363:221-32.
- Jennette JC, Falk RJ, Andrassy K, Bacon PA, Churg J, Goss WL, et al. Nomenclature of systemic systemic vasculitides. Proposal of an international consensus conference. *Arthritis Rheum* 1994;37:187-92.
- Merino JL, Galeano C, Espejo B, Rivera M, Fernandez-Lucas M, Caldés S, et al. A retrospective study on outcome of microscopic polyangiitis in chronic renal replacement therapy. *Nephrol Dial Transplant* 2011;26:1360-6.
- Gayraud M, Guillevin L, Le Toumelin P, Cohen P, Lhote F, Casassus P, et al.; French Vasculitis Study Group. Long-term follow-up of polyarteritis nodosa, microscopic polyangiitis and Churg Strauss syndrome: analysis of four prospective trials including 278 patients. *Arthritis Rheum* 2001;44(3):666-75.
- Mitchison NA. T-cell-B-cell cooperation. *Nat Rev Immunol* 2004;4:308-12.
- Stasi R, Stipa E, Del Poeta G, Amadori S, Newland AC, Provan D. Long-term observation of patients with anti-neutrophil cytoplasmic antibody-associated vasculitis treated with rituximab. *Rheumatology (Oxford)* 2006;45:1432-6.
- Rocatello D, Baldovino S, Alpa M, Rossi D, Napoli F, Naretto C, et al. Effects of anti-CD20 monoclonal antibody as a rescue treatment for ANCA-associated idiopathic systemic vasculitis with or without overt renal involvement. *Clin Exp Rheumatol* 2008;26(3 Suppl 49):S67-71.
- Keogh KA, Wylliam ME, Stone JH, Specks U. Induction of remission by B lymphocyte depletion in eleven patients with refractory antineutrophil cytoplasmic antibody associated vasculitis. *Arthritis Rheum* 2005;52:262-8.
- Gottenberg JE, Gullevin L, Lambotte O, Combe B, Allanore Y, Cantagrel A, et al. Tolerance and short term efficacy of rituximab in 43 patients with systemic autoimmune diseases. *Ann Rheum Dis* 2005;64:913-20.
- Saech J, Owczarczyk K, Rösigen S, Peterleit H, Hallek M, Rubbert-Roth A. Successful use of rituximab in a patient with Churg-Strauss syndrome and refractory central nervous system involvement. *Ann Rheum Dis* 2010;69(6):1254-5.
- Cartin-Ceba R, Keogh KA, Specks U, Sethi S, Fervenza FC. Rituximab for the treatment of Churg-Strauss syndrome with renal involvement. *Nephrol Dial Transplant* 2011;26:2865-71.

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**María Martínez-Villaescusa<sup>1</sup>,  
Aurora López-Montes<sup>1</sup>,  
Esperanza López-Rubio<sup>1</sup>,  
Lourdes de la Vara-Iniesta<sup>1</sup>,  
Marina Méndez-Molina<sup>1</sup>,  
Dionisio Donate-Ortiz<sup>1</sup>,  
Hyun Nam-Cha<sup>2</sup>, Carmen Gómez-Roldán<sup>1</sup>**

<sup>1</sup> Servicio de Nefrología. Hospital General Universitario de Albacete. (Spain)

<sup>2</sup> Anatomía Patológica. Hospital General Universitario de Albacete. (Spain)

**Correspondencia:** María Martínez-Villaescusa  
Servicio de Nefrología, Hospital General Universitario de Albacete. (Spain)  
mariamvillaescusa@yahoo.es

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## Transovarian venous access in a patient with exhausted vascular access for haemodialysis

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### To the Editor:

The availability of a vascular access is essential for adequate haemodialysis. Vascular access is therefore the Achilles' heel of this treatment. When patients present venous access depletion, either for the creation of an arteriovenous fistula, a vascular prosthesis or the placement of a tunnelled catheter, we must resort to a change of dialysis modality if possible, otherwise unconventional venous access routes must be considered.<sup>1,2</sup>

We report the case of a patient with stage 5D chronic kidney disease (CKD) with exhausted accesses in whom we placed a tunnelled catheter transovarially.

### CASE REPORT

Our patient is a 27-year-old female with CKD secondary to renal hypoplasia diagnosed at 2 years of age. At 9 years of age she had stage 5D CKD and received a living related donor kidney transplant; eight years later, she lost transplant function due to chronic rejection and had to begin a tri-weekly haemodialysis treatment. There were multiple complications due to obstructions and infections of the vascular access points for haemodialysis. Ten years later, she was transferred to our institution due to exhaustion of vascular accesses. An angiography of the upper and lower limbs and vena cava was performed, which showed thrombosis in both jugular and subclavian veins and the brachiocephalic artery, as well as in both femoral and iliac veins. Without the possibility of a living related donor transplant, it was decided to start peritoneal dialy-