Inflammatory Involvement of Spermatic Vein in a Patient With Immunoglobulin 4–Related Disease

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47-year-old man presented with mild, diffuse, and recurrent A abdominal pain of 30 days of evolution and recent hypertension. Studies were conducted to rule out causes of secondary hypertension. In laboratory studies, renal failure with creatinine 1.57 mg/dL was detected. Blood glucose, erythrosedimentation, and complement were normal. Reactive C protein was 3 mg/dL. Ambulatory blood pressure monitoring revealed uncontrolled arterial hypertension and dipper pattern. The abdominal ultrasound evidenced left hydronephrosis. Doppler ultrasound of kidney arteries was normal. Abdominal computed tomography (CT) scan with intravenous contrast demonstrated an extensive left anterior pararenal and perirenal space soft tissue mass that encased pancreatic tail (Figs. A and C), inferior mesenteric vessels, and splenic and renal veins. In addition, an extensive involvement of the left spermatic vein was observed (Figs. B and D). Laboratory test confirmed polyclonal hypergammaglobulinemia (immunoglobulin G [IgG] 1678 mg/dL, IgE and IgG4 with normal values), carcinoembryonic antigen 2.3 ng/mL, and cancer antigen 19.9 17 U/mL. A pancreatic CT-guided core biopsy showed pancreatic fibrous tissue with storiform pattern. Lymphoplasmacytic inflammatory infiltrate and eosinophils were also observed. Immunohistochemistry showed numerous CD138-positive plasma cells and more than 10 IgG4⁺ plasma cells per high-power field, with more than 50% of all plasma cells staining for IgG4 (Figs. E-H).

Immunoglobulin G4–related disease was diagnosed, and prednisone 40 mg/d with mycophenolate was initiated with favorable clinical and imaging response.

In IgG4–related disease, there may be 2 forms of vascular disease, primary vasculitis and secondary vascular involvement by the compressive effect of tumefactive lesions.¹ In the literature, predominantly arterial lesions (mainly aortitis, periaortitis. and periarteritis) are described,² with some reports of secondary involvement of the iliac vein and cava vein.^{2,3} Secondary involvement is radiologically characterized by homogeneous thickening of the arterial wall, secondary to adventitia sclerosis.² Our patient has obliteration of the splenic and left spermatic veins. The latter finding has not been previously reported.

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FIGURE. A, Contrast abdominal CT scan with white circle demonstrating an extensive left anterior pararenal and perirenal space soft tissue mass that encased the pancreatic tail, inferior mesenteric vessels, and splenic and renal veins. B, In sagittal cut, white arrows show an extensive involvement of the left spermatic vein. C, White circle demonstrating an extensive perirenal space soft tissue mass. D, Homogeneous thickening of left spermatic vein. E, Pancreatic and peripancreatic fibrous tissue with storiform pattern (white arrow), phlebitis (white star) and lymphoplasmacytic infiltrate (enlarged figure in right upper margin). F, Peripancreatic tissue with storiform fibrosis, lymphoplasmacytic infiltrate, and few eosinophils (white circle). G, Immunohistochemistry showed numerous CD138-positive plasma cells. H, Immunohistochemistry of sample of peripancreatic tissue showed more than 10 IgG4⁺ plasma cells per high-power field. Color online-figure is available at http://www.jclinrheum.com.

2 www.jclinrheum.com