Subclinical Ovarian Vasculitis Developing in a Patient with Rapidly Progressive Glomerulonephritis Associated with Perinuclear Antineutrophil Cytoplasmic Antibody

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Dear Sir,

The possible relationship between systemic vasculitis and idiopathic crescentic glomerulonephritis has attracted much attention in relation to perinuclear antineutrophil cytoplasmic antibody (P-ANCA) [1, 2]. Herewith we describe a case of ovarian vasculitis incidentally found in a patient with P-ANCA-positive rapidly progressive glomerulonephritis (RPGN).

A 48-year-old woman was referred to Chiba University Hospital in April 1991 because of renal dysfunction and a high fever continuing for 2 weeks. The patient was in good health until January 1991, when she first visited her local physician because of bilateral shoulder pain. She had been under the doctor's care for 3 months, but she gradually became aware of a vague ill feeling and low-grade fever. Laboratory examinations on admission showed anemia (hemoglobin 53 g/l) and renal dysfunction (serum creatinine 424 μ mol/l or 4.8 mg/dl; creatinine clearance 16 ml/min). Urinalysis showed mild protein-uria (0.7 g/day) and hematuria (50-60 RBC/ F). ANCA was positive at a titer of 1:320 with a perinuclear staining pattern by indirect immunofluorescence, but the other autoantibodies including antinuclear and anti-DNA antibody were negative. Chest radiograph was normal. Under the tentative diagnosis of RPGN, treatment was initiated with a large dose of glucocorticoid (methylprednisolone 1 g daily for 3 days) in addition to oral cyclophosphamide

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Fig. 1. Photomicrograph showing fibrinoid necrosis and severe inflammatory infiltrates throughout the wall of a middle-sized artery in the ovarium. HE. × 100.

abdomen, and ultrasound examination of the pelvis revealed a right adnexal cystic lesion in January 1992. The right ovarium was resected with the mass lesion. Histological examination of the mass made the diagnosis of dermoid cyst. In addition, the presence of necrotizing vasculitis affecting medium-sized arteries was demonstrated (fig. 1). Her renal function was stable despite the operation, and she has been in good condition over 1 year.

There have been several reports of ovarian vasculitis [3-5]. Most of these cases were asymptomatic and had no systemic manifestations. Such organ-limited necrotizing vasculitis was treated with prednisolone 50 mg daily and intravenous heparin 5,000 IU daily, and followed by oral prednisolone daily. Her symptoms promptly disappeared, and serum creatinine decreased to 212 $\mu\text{mol/l}$ (2.4 mg/dl) 1 month after admission, when she submitted to open renal biopsy. Seventy percent of the glomeruli were crescentic, but a glomerular immune deposit was absent except fibrinogen. She was discharged 2 months later when serum creatinine decreased to 140 $\mu\text{mol/l}$ (1.6 mg/dl). Over the subsequent 6 months, she has been fairly well despite the reduction of prednisolone doses. However, she gradually became aware of a dull pain in the lower

culitis has been documented in various organs including kidney [6-9]. Several reports showed that systemic involvement became obvious many years after the initial presentation of local arteritis [7, 8]. Ito et al. [9] suggested that the lack of constitutional symptoms or laboratory findings at diagnosis of localized vasculitis does not completely preclude the possibilities of subsequent development of systemic vasculitis. This view seems to be compatible with the present case which might have been interpreted as a renal-limited disease if the ovarian vasculitis had not been incidentally identified. Since active ovarian angitis still existed after intensive treatment with glucocorticoid and cyclophosphamide, subclinical vasculitis might also exist in the other organs. Previous studies showed that a considerable number of patients with P-ANCA-positive RPGN took a form of renal-limited disease [1, 2]. Our case suggested, however, that subclinical vasculitis of extrarenal organs might exist more frequently than expected in patients with P-ANCA-positive RPGN.

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Erratum

In the article by Daya et al. entitled 'Use of Levamisole in Maintaining Remission in Steroid-Sensitive Nephrotic Syndrome in Children' published in vol. 66, no. 4, 1994 (pp. 408-412), table 3, the total under levamisole appearing as (n = 13) should read as (n = 32).

In the article by Wu et al. entitled 'Dialysis-Related Amyloidosis and Acquired Renal Cell Carcinoma in Uremia' published in vol. 67, no. 1, 1994 (p. 129), the first authors name should read as follows: Tsai-Hung Wu.