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Case report

A case of paraneoplastic vasculitic neuropathy associated with gastric cancer

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1. Introduction

Paraneoplastic neurologic syndromes (PNS) refer to any neurologic disorders of the peripheral nervous system in the setting of cancer that is not explained by any of these mechanisms, which are divided into classical and non-classical forms. The definition of PNS is broad, and confirmation of this diagnosis is accordingly difficult. In 2004, an expert panel proposed diagnostic criteria for PNS including definite or possible PNS [1]. Among the possible PNS, paraneoplastic vasculitic neuropathy (PVN) is neurological syndrome not associated with known onconeuronal antibodies [1]. In 1997, Oh highlighted the unique characteristics of PVN and recommended its separate classification [2]. Although PVN was caused by many kinds of cancer, there was a case report on PVN associated with gastric cancer. However, it was asymptomatic and was confirmed via post-mortem biopsy of the sciatic nerve [3]. In this report, we describe a case of confirmed PVN associated with gastric cancer.

2. Case report

A 71-year-old man presented to a local hospital with tingling in his left leg. He had a history of tuberculosis which was cured 35years ago, and was on medication for hypertension and benign prostate hypertrophy. Several days after presentation, he developed the right-sided foot drop. Magnetic resonance imaging of the brain and lumbar spine demonstrated no abnormalities. He was transferred to our hospital 2 weeks after symptom onset. At the time of admission, medical research council (MRC) grade of ankle dorsiflexor, great toe extensor and ankle plantar flexor was 0 on the right side and 2 on the left. His MRC sum score was 55 (normal total score, 60), and his modified Rankin scale score was 2. Hypesthesia was noted in the right lower extremity and deep tendon reflexes were mildly decreased in both lower extremities. Two weeks after admission, new neurologic symptoms including distal weakness and paresthesias of both the left lower leg and the right upper arm were developed and consequently, his MRC sum score decreased to 45, and his modified Rankin scale score was 3.

The initial laboratory evaluation revealed increases in white blood cell count (WBC: 16,430/μL), erythrocyte sedimentation rate (ESR: >120 mm/h), and C-reactive protein (CRP: 14.969 mg/dL). These results suggested a systemic infection and systemic vasculitis, prompting further evaluation. Follow-up laboratory results demonstrated a positive antinuclear cytoplasmic antibody (ANCA) test with an elevated p-ANCA but not c-ANCA titer. Additionally, the patient had elevated rheumatoid factor (RF) and anti-double stranded (ds) DNA antibody levels (RF: 121 IU/mL; anti-ds DNA: 5.95 IU/mL). All other laboratory evaluations were within normal limits, including cerebrospinal fluid analysis.

The patient had history of smoking for 50 years without any respiratory symptoms. Routine initial chest X-ray revealed bilateral reticular opacities most prominent in the peripheral and basal lung fields, suggestive of interstitial lung disease. Subsequent

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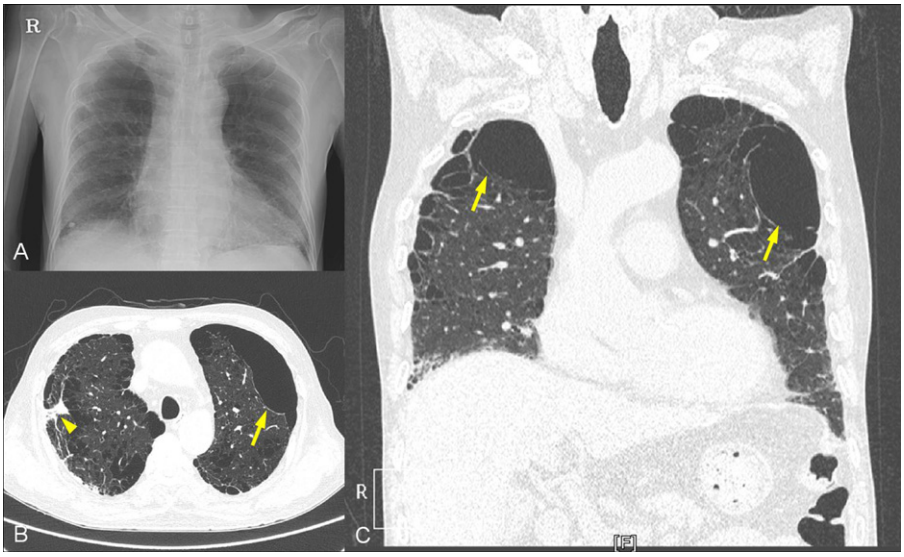

Fig. 1. Initial chest X-ray demonstrating bilateral reticular opacities most prominent in the peripheral and basal lung fields (A). High-resolution chest CT showing severe bullous changes (arrows) with diffuse emphysema and focal fibrotic changes (arrowhead) with honeycombing, suggestive of interstitial lung disease.

high-resolution computed tomography (HRCT) of the chest showed severe bullous changes, diffuse emphysema, and fibrotic interlobular septal thickening with honeycombing, confirming the presence of interstitial lung disease (Fig. 1). With these HRCT findings, idiopathic pulmonary fibrosis was diagnosed without undergoing biopsy this aged smoker with small lung reserve.

One week after admission, electrodiagnostic examination was performed, revealing an asymmetric motor and sensory polyneuropathy involving the bilateral lower extremities and the right upper extremity. The right median motor response demonstrated prolonged latency, low amplitude, and mildly decreased conduction velocity, and the bilateral peroneal and tibial motor responses were unobtainable. The right median and ulnar responses were of low amplitudes bilateral sural responses were unobtainable. The right median and ulnar F-waves were prolonged at initial examination, and the bilateral H-reflexes were

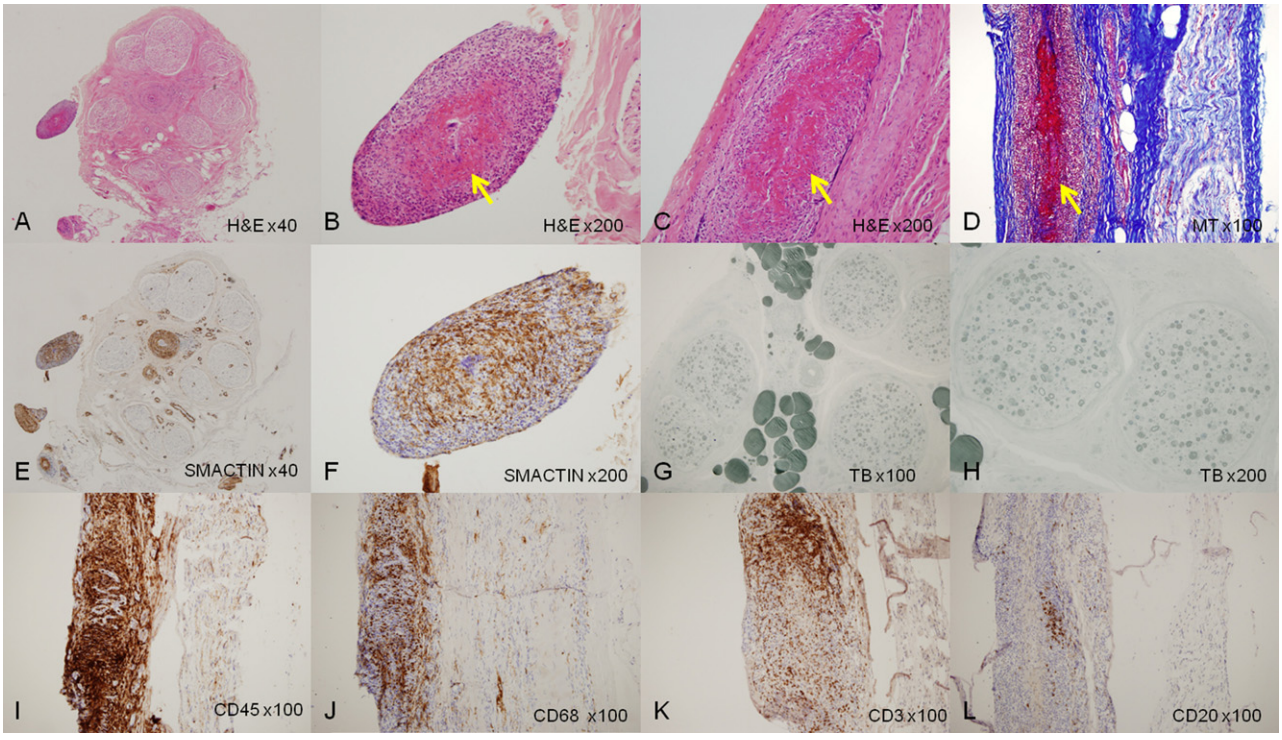

Fig. 2. Paraffin sections (A–F) demonstrating the large arterioles of a peripheral nerve with necrotizing vasculitis and fibrinoid necrosis (arrows), epineurial mononuclear inflammatory infiltrates (small to moderate collections), individual endoneurial inflammatory cells, inflammation within the walls of large epineurial arterioles, and neovascularization. The density of myelinated fibers is moderately decreased in a multifocal pattern. An increased rate of axonal degeneration with frequent degenerating profiles on the semi-thin sections is demonstrated (G and H). Immunohistochemical studies (I–L) show strong reactivity within the wall of a large epineurial arteriole with a leukocyte (CD-45) preparation, as well as moderate reactivity within the wall of a large epineurial arteriole with macrophage (CD-60) and T-cell (CD3) preparations. H&E, hematoxylin & eosin staining; MT, methylene blue staining; SMACTIN, smooth muscle actin staining; TB, toluidine blue staining.