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Alveolar hemorrhage, rectus muscle hematomata, and gastrointestinal bleeding accompanied by crescentic glomerulonephritis in a patient with PR3-ANCA disease

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Introduction.– This is the first report of a case of GPA accompanied by alveolar hemorrhage, rectus muscle hematoma, and gastrointestinal bleeding.

Patients.– A 67-year-old female was admitted to our hospital because of general fatigue, fever, and edema for one month, and hemoptysis, dyspnea for 1 week. Scr was rapidly increased to 4.0 mg/dL with hematuria and proteinuria. CT revealed large areas of ground glass opacity in the right lung; hence the hemoptysis was due to alveolar hemorrhage. Sputum culture revealed the presence of Staphylococcus. The titer of PR3-ANCA was 340 U/L, and he was diagnosed with ANCA disease (BVAS 48) associated with pneumonias. Thus, she was treated with antibiotics and plasma exchange (PE). However, after the second PE, she suddenly complained of abdominal pain and swelling in the right rectus muscle region with hypotension. Her Hb dropped from 8.8 to 4.5 g/dL in the subsequent hours. Abdominal CT showed an irregular low-density mass in the right muscle, so he was diagnosed as rectus muscle hematoma. Her condition was improved after receiving a transfusion and supportive therapy. Then, she was started on methylprednisolone pulse therapy followed by the oral prednisolone and cyclophosphamide result in improvement of hemoptysis with gradually decreased Scr. However, she developed tinnitus and bloody nasal discharge and diagnosed as frontal sinuses by CT. Therefore, we gave her the third PE. Unfortunately, she developed tarry stools and her Hb dropped soon. Endoscopy showed two duodenal ulcers, and the biopsy specimen revealed angitis of the duodenum. Another methylprednisolone pulse therapy, inhibition of gastric acid and transfusion were given. Renal biopsy showed the presence of crescent-shaped glomerulonephritis.

Results.– In the GPA group LTB4, PGD2, and 11-dehydro-TXB2 were elevated when compared to HC, whereas: 5-HETE, LTC4, trans LTC4, eoxins (C4, D4, E4), 9 α, 11β PGE2α and tetranor-PGE-M were decreased (figure S1).

Conclusion.– Significant differences in the EBC eicosanoid profiles between the GPA and HC groups were observed. This finding may help us to elucidate the pathogenesis of the respiratory tract inflammation in GPA patients.

Supplementary data associated with this article can be found on the website of La Presse Médicale (http://www.em-consul-lete.com/revue/lpm).

Figure 1. Differences between eicosanoid concentrations in exhaled breath condensate from patients with granulomatosis with polyangiitis (GPA) and healthy controls (HC). Abbreviations: M: metabolite; *: < 0.05; **: < 0.01.

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