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## **Idiopathic Systemic Capillary leak syndrome – A Case Report**

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**Case Study:** Idiopathic systemic capillary leak syndrome (ISCLS) is a rare disease characterized by recurrent episodes of acute life-threatening attacks of shock, hemoconcentration, and hypoalbuminemia. Increase of capillary permeability results in reversible plasma extravasation and subsequent vascular collapse accompanied by related symptoms. Here, we report a case of well controlling ISCLS, which can be difficult to meet in clinical field.

A 47-year-old man was admitted to our institution with general weakness and abdominal pain on the day of admission. The patient had no underlying diseases or medications. He was never-smoker, and social drinker. The initial vital sign was stable, but the blood pressure (BP) decreased to 74/60 mmHg at 3 hours in the emergency room. Initial laboratory tests revealed several abnormal findings, including an elevated leukocyte count (29,180 cells/mm<sup>3</sup>), hemoglobin (Hgb, 22.6 g/dL), and hematocrit (66.1%), hypoalbuminemia (2.6 g/dL), and acute kidney injury (blood urea nitrogen [BUN] 28 mg/dL, Creatinine [Cr] 1.95 mg/dL). In bone marrow biopsy performed on polycythemia, reactive marrow was suspected without any abnormal findings. After treatment with vigorous fluid therapy and empirical antibiotics, he was recovered and discharged with Hgb 14.1 g/dL and BUN/Cr 13/0.96 mg/dL.

There was a gradual upward trend in Hgb from 14.1 g/dL to 16.6 g/dL for 3 months after discharge, but there was no significant symptoms. However, he visited our hospital again on the 3 months and 2 weeks after the discharge with symptoms similar to the first episode. The BP was 70/50 mmHg, WBC 30,150 cells/mm<sup>3</sup>, Hb 19.7 g/dL, hematocrit 59.3%, BUN/Cr 39/2.93 mg/dL and albumin 3.0 g/dL, respectively. Based on these repeated clinical features, he was diagnosed with ISCLS. Based on the case series reported previously, bambuterol 10 mg and theophylline 400 mg once-a-day were prescribed, he has been following by 8 months without recurrence.