Successful treatment for gastro-intestinal bleeding of Osler-Weber-Rendu disease by argon plasma coagulation using double-balloon enteroscopy



Figure 1 An endoscopic view of gastric angiodysplasia. **a** Multiple angiodysplasias, each about 3 – 5 mm in diameter, in the stomach between the proximal gastric body and the antrum. **b** Argon plasma coagulation (APC) for gastric angiodysplasia using the APC probe. After treatment, angiodysplasia had completely disappeared with mucosal carbonization.

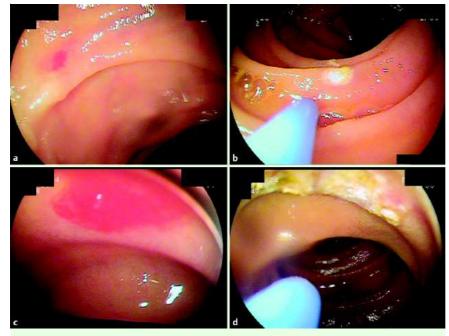


Figure 2 Representative double-balloon enteroscopy (DBE) views of the small intestine. **a** Angiodysplasia of about 3 mm was detected in the jejunum. **b** After argon plasma coagulation (APC) cauterization of **a**. **c** Hemorrhagic angiodysplasia of the jejunum. **d** Application of APC through the DBE to eradicate the angiodysplasia shown in **c**, using the APC probe.

Osler–Weber–Rendu (OWR) disease is an autosomal dominant disorder characterized by aberrant vascular development in the skin, mucous membranes, and visceral organs [1]. In this disease, gastrointestinal bleeding is one of the most critical symptoms. However, no effective therapy is available for this disease. Argon plasma coagulation (APC) is reportedly an effective treatment for the gastrointestinal bleeding, but to date only APC treatment of bleeding from the stomach, duodenum, and colon has been reported. However, patients with OWR usually have angiodysplasia in the small intestine [2], an area that double-balloon enteroscopy (DBE) has been shown to be clinically useful for reaching a diagnosis as well as providing therapy [3]. We report a case of OWR disease in which angiodysplasia of the small intestine was successfully eradicated by APC with DBE.

A 51-year-old woman with OWR disease was admitted due to repeated microcytic anemia and tarry stools. Upper endoscopy disclosed about 10 homogeneous, flat, round angiodysplasias in the stomach, each about 3–5 mm in diameter (> Figure 1 a), and three rounded angiodysplasias in the second position of the duodenum. Colonoscopy revealed no angiodysplasia lesions in the colon or terminal ileum. APC for gastroduodenal lesions was carried out using the ERBE ICC200 and APC300 (ERBE, Tubingen, Germany) (**Figure 1 b**). The argon gas flow was set at 2.0 L/minute and 60 W for 10 gastric lesions and 1.4 L/minute at 40 W for three duodenal lesions. No bleeding or perforation was recorded. After treatment, the angiodysplasia had completely disappeared. As the progression of anemia had halted, and the patient became independent of frequent transfusions, she was soon discharged.

However, after 6 months, the patient presented with a tarry stool without definite epistaxis. Tests again showed iron deficiency anemia. An upper endoscopy disclosed only ulcer scars. Colonoscopy revealed no angiodysplasia lesions in either the colon or terminal ileum. A DBE via antegrade and retrograde approaches (Fujinon-Toshiba, Tokyo, Japan) revealed more than 20 homogeneous, flat, round angiodysplasias of about 3-5 mm each (**Figure 2a, c**) within 1 m of the anal side of the Treitz ligament. APC was carried out (**>** Figure 2b, d) with the argon gas flow set at 1.4 L/minute and 40 W. No complications were recorded. After APC, fecal occult blood test became negative, with no further progression of anemia. The patient has now been stable for over 24 months without transfusions. In conclusion, this case shows that complete gastrointestinal endoscopic examination should be carried out in patients with OWR, and that application of APC through DBE to eradicate angiectasias, especially in the small intestine, is safe and effective for gastrointestinal bleeding of OWR disease.

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