A 53-year-old man, with known Klippel–Trenaunay syndrome (KTS), presented with massive life-threatening gastrointestinal blood loss. After hemodynamic resuscitation, upper endoscopy and ileo-colonoscopy were performed. These did not reveal the source of bleeding. Video capsule endoscopy was subsequently performed using the Given (Yoqneam, Israel) system. This revealed a large number of small-bowel hemangiomas (Figure 1). However, none of the lesions showed signs of active or recent bleeding. The clinical course was favorable, with spontaneous resolution of the blood loss. It remains unclear what may have precipitated the bleeding episode.

KTS is a rare congenital vascular anomaly characterized by limb hypertrophy, cutaneous hemangiomas, and venous malformations. Gastrointestinal bleeding is a potentially serious complication of diffuse hemangiomatous involvement of the gut in KTS. Several cases have been described in the literature, mostly concerning bleeding hemangiomas in the colorectum [1]. Reports of involvement of the small intestine are extremely rare [2]. However, it must be noted that most cases published date from the era before the introduction of wireless video capsule endoscopy. Our case illustrates the diagnostic value of capsule endoscopy in establishing suspected small-bowel hemangiomas in KTS syndrome.

Endoscopy_UCTN_Code_CCL_1AC_2AB

Figure 1 Capsule endoscopy findings in a patient with Klippel–Trenaunay syndrome.

**R. K. Weersma¹, W. J. Thijs²,
G. D. C. Vosmaer², J. J. Koornstra¹**

¹ Department of Gastroenterology and Hepatology, University Medical Center Groningen, University of Groningen, The Netherlands

² Department of Gastroenterology, Scheper Hospital, Emmen, The Netherlands.

Corresponding author

J. J. Koornstra, MD, PhD

University Medical Center Groningen
PO Box 30001

9700 RB Groningen

The Netherlands

Fax: +31-50-3619306

Email: j.j.koornstra@int.umcg.nl

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