Lymphangioma of the anal canal: first description of the endoscopic features

A 42-year-old woman was referred to our hospital for the work-up of iron deficiency anemia. Esophagogastroduodenoscopy (EGD) did not reveal a bleeding source. The appearances on total colonoscopy were normal except for a 20-mm submucosal tumor in the left lateral anal canal (Fig. 1). The tumor could be compressed with the endoscope, indicating that it was soft. It was also translucent and lustrous. The tumor and the mucosa surrounding its base were lined with stratified squamous epithelium.

Narrow-band imaging (NBI) revealed scattered bullous-like transparent areas on the tumor surface (Fig. 2a). Magnifying endoscopy with narrow-band imaging (ME-NBI) of these areas demonstrated elongated, dark-brown, looping capillary vessels, without dilatation or caliber change, in the superficial layers (Fig. 2b). In addition, small blood vessels were observed in the deeper subepithelial layers (Fig. 2c). The tumor was totally removed using endoscopic submucosal dissection (ESD) and was diagnosed histologically as a lymphangioma (Fig. 3).

Lymphangiomas of the anal canal are extremely rare. To the best of our knowledge, there are only two previous cases reported, and neither of these included detailed endoscopic findings [1,2]. Here, we present the notable epithelial and microvascular findings on ME-NBI of an anal lymphangioma for the first time. By comparing the endoscopic and histological findings, we presume that the continuous stretching by distended lymphatic channels resulted in the thinning of the underlying squamous epithelium. This probably caused mucosal transparency and capillary elongation, resulting in the visualization of deeper stromal microvessels.

In a case report of esophageal lymphangioma [3], very similar ME-NBI findings were described as being characteristic features. Because the anal canal and esophagus are both covered with stratified squamous epithelium, the ME-NBI diagnostic criteria used for esophageal lymphangioma may also be applicable to anal lymphangioma, as is the case with squamous cell carcinoma [4,5]. We consider that these unique endoscopic findings might be characteristic of lymphangioma of the anal canal and could be useful in making this diagnosis.

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References
Fig. 3  Histopathology of an area of the resected lesion corresponding to the endoscopically transparent epithelium showing dilated lymphatic channels of different sizes beneath the thin squamous epithelium (hematoxylin and eosin [H&E] stained, original magnification × 10).  
Inset  Immunohistochemical staining for CD34 showing positive staining of the small blood vessels as they run in the stroma (red arrows) and just beneath local regions of thinned and slightly protruding squamous epithelium (green arrows; original magnification × 40).