Endoscopic submucosal dissection in a rare case of carcinoma cuniculatum of the esophagus initially misdiagnosed as benign squamous papilloma

Fig. 1 Esophagogastroduodenoscopy (EGD) in a 65-year-old woman revealed a large, circumferential, partially obstructive esophageal mass. **a** Endoscopic appearance of the large mass in the mid-distal esophagus. **b** Esophageal mass with long, digit-like projections and exophytic growth.

Fig. 2 Endoscopic submucosal dissection (ESD) on the proximal 6 cm of an esophageal mass in a 65-year-old woman. **a** Partial removal by ESD. The remaining portion of the tumor can be seen in the distal esophagus. **b** Dissected part of the lesion with digit-like projections and furrows, mounted for pathologic examination.

Fig. 3 Histopathological analysis of the ESD specimen from an esophageal mass in a 65-year-old woman. Low power view (2×) showing well differentiated squamous epithelium with extensive hyperkeratosis, acanthosis, dyskeratosis, intraepithelial neutrophils, and koilocyte-like cells.

A 65-year-old woman presented for evaluation of a large esophageal mass, diagnosed on biopsy as squamous cell papilloma. Computed tomography (CT) showed no lymphadenopathy or distant metastases. The patient refused surgery. Endoscopic mucosal resection (EMR) was attempted at a tertiary referral center but could not be carried out because of difficulty in lifting the lesion. Thus, she was referred to our center for endoscopic submucosal dissection (ESD).

Esophagogastroduodenoscopy (EGD) revealed a large, circumferential, partially obstructive esophageal mass extending from 22 to 30 cm from the incisors (Fig. 1a,b). Successful circumferential ESD was carried out on the proximal 6 cm of the lesion (Fig. 2a,b), however, the distal 2 cm could not be resected owing to severe submucosal fibrosis.

Histopathological analysis of the ESD specimen revealed an unusual histomorphologic pattern including hyperkeratosis, acanthosis, dyskeratosis, intraepithelial neutrophils, keratin-filled furrows, and koilocyte-like cells consistent with carcinoma cuniculatum (Fig. 3). Invasion in the submucosa was present. Laparoscopy-assisted esophageal resection was performed, and the resected specimen was staged pathologically as T1bN0. The patient had an uneventful recovery. Carcinoma cuniculatum of the esophagus is a very rare and extremely well differentiated variant of squamous cell carcinoma, first described in 2005 [1]. Since then, it has been reported in less than 15 cases [2,3]. The etiology is unknown but there have been reported associations with chronic esophageal irritation, achalasia, diverticulum, and prior caustic injury [1, 4]. Carcinoma cuniculatum of the esophagus has traditionally been managed with esophagectomy [3].

This is the first reported case of carcinoma cuniculatum in which ESD was attempted with partial success. The success was limited owing to submucosal invasion but adequate staging was still achieved: the ESD specimen provided an accurate histopathologic diagnosis and staging to guide further therapy. Thus, ESD could be used not only as a treatment option but also as a staging tool to assess the depth of invasion.

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Competing interests: None

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