Endoscopic submucosal dissection for an early-stage neuroendocrine carcinoma composited with squamous cell dysplasia

A 51-year-old man complained of dysphagia for 2 months. A reddish and rough lesion was found 25 cm from the incisor under esophagogastroduodenoscopy (EGD) (► **Fig. 1 a**). Pathological assessment of the biopsy indicated neuroendocrine carcinoma (► **Fig. 1 b**). The patient was an alcohol addict. Pre-operative examination revealed pancytopenia and portal hypertension with collateral development (► **Fig. 1 c**). Positron emission tomography/computed tomography (PET/CT) excluded distal metastasis. Considering the high risk of bleeding and leakage for esophagectomy, endoscopic treatment was suggested after multi-disciplinary discussion. The lesion was resected with endoscopic submucosal dissection (ESD) (► **Fig. 1 d, e**). Iodine staining indicated an unstained area in part of the gross specimen. Final diagnosis was collision carcinoma, neuroendocrine carcinoma combined with squamous cell high grade intraepithelial neoplasm (HGIN) (► **Fig. 1 f, g**). The tumor was confined to the mucosal muscularis layer, without lymphovascular invasion. The lateral and horizontal margins were both negative. The patient declined further chemotherapy and received active surveillance. No recurrence or metastasis was detected 18 months after ESD (► **Fig. 1 h, i**) (► **Video 1**).

Esophageal neuroendocrine carcinoma is a rare disease characterized by high malignancy. Patients usually present with distant metastasis at diagnosis [1–2]. Histological diagnosis of neuroendocrine carcinoma by biopsy is difficult for it generally presents as a submucosal tumor covered by non-neoplastic epithelium [2]. On the other hand, neuroendocrine carcinomas sometimes have mixed components of squamous and/or glandular cells, which often lead to misdiagnosis

[3]. The standard management of esophageal neuroendocrine carcinomas has not yet been established owing to the paucity of available data. Radical esophagectomy should be considered as the primary treatment for stage I/IIA patients [4]. Whether radiochemotherapy is necessary for early-stage esophageal neuroendocrine carcinoma remains controversial. Until now, only few cases of esophageal neuroendocrine carcinoma treated by ESD instead of surgery have been reported [2,5]. ESD provides an alternative for patients at high risk of surgical complications. However, whether ESD is equally effective compared with traditional radical surgery for superficial esophageal neuroendocrine carcinoma is still uncertain. Further evidence with more cases and longer follow-up are warranted for this specific problem.

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Competing interests

The authors declare that they have no conflict of interest.

► **Fig. 1** Endoscopic submucosal dissection (ESD) for an early-stage neuroendocrine carcinoma composited with squamous cell dysplasia. **a** Esophagogastroduodenoscopy (EGD) examination found a reddish and rough lesion 25 cm from the incisor. **b** The small round cells formed a cluster in the submucosal layer. **c** Portal vein thrombosis under contrast computed tomography. **d** ESD for the lesion. **e** Specimen of ESD. **f** Both neuroendocrine carcinoma (yellow circle) and high grade intraepithelial neoplasia (yellow arrow) component could be seen in the same view ($\times 20$). **g** The neuroendocrine carcinoma component was immunohistochemically positive for synaptophysin. **h** Scar detected after 18 months under EGD. No local recurrence was detected. **i** Gallium-68 DOTA-TATE positron emission tomography/computed tomography scan 6 months after ESD excluded tumor residue or distant metastasis.

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