Duodenal gangliocytic parangangioma treated with endoscopic hemostasis and resection

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Gangliocytic parangangiomas are exceedingly rare tumors that arise in close proximity to the papilla of Vater. There are few reports of the endoscopic resection of duodenal gangliocytic parangangioma. A 61-year-old woman was admitted with a complaint of melena. Endoscopic examination revealed a pedunculated submucosal tumor with erosion in the third portion of the duodenum. Hemostasis, using a gold probe, was performed. Nine days later, we successfully resected the tumor, using endoscopic polypectomy. To determine the depth of tumor invasion, endoscopic ultrasonography was used. The size of the tumor was 3.0 × 2.5 × 1.0 cm. A total of 25 cases of duodenal gangliocytic parangangioma have been reported in Japan. Generally, this tumor is considered benign. However, resection was performed in many patients because preoperative diagnosis was impossible. In Japan, no previous studies have reported using endoscopic hemostasis, to our knowledge. Our patient is the fourth in Japan to be treated by endoscopic resection. We report on our patient, with a review of the literature.

Key words: gangliocytic parangangioma, duodenum, gold probe

Introduction

Gangliocytic parangangioma is an extremely rare benign duodenal tumor. In Japan, 26 patients, including our patient (described below) have been reported. In most patients, resection, including pancreatoduodenectomy, was performed because preoperative diagnosis was impossible. This tumor is generally considered to be benign. Therefore, endoscopic resection may be the best procedure to facilitate diagnosis and treatment. In Japan, no previous studies have reported endoscopic hemostasis for this tumor, to our knowledge. In this study, we report a patient with gangliocytic parangangioma, in whom a detailed examination was performed after endoscopic hemostasis, revealing a tumor that was able to be endoscopically resected. We also review the literature.

Case report

A 61-year-old Japanese woman was admitted because of massive melena. On admission, the patient had no gastrointestinal symptoms and was in good general condition. Laboratory data on admission were as follows: hemoglobin (Hb) and blood urea nitrogen (BUN) levels were 9.3 g/dl and 25.5 mg/dl, respectively. Endoscopy revealed a movable polypoid submucosal tumor, with a smooth surface and a horizontal pedicle, in the papillary duodenal region. In the tumor pedicle region, ulceration was observed. The horizontal duodenal region was full of blood. At the tumor end in the horizontal duodenal region, erosion was observed. Spurting hemorrhage from the site was noted (Fig. 1). It was possible that the bleeding had been induced by endoscopic examination. A hypotonic duodenogram showed a protruding lesion with a smooth surface and a relatively clear protrusion. Partial ulceration was noted (Fig. 2a). Endoscopic ultrasonography (EUS) showed a mass with inhomogeneous echotexture in the third layer of the wall of the duodenum (Fujinon SP-701; 15 MHz) (Fig. 2b). Endoscopic retrograde cholangiopancreatography (ERCP) did not reveal any marked changes in the bile duct or pancreatic duct (Fig. 2c). Neither abdominal computed tomography
Endoscopic hemostasis was performed following emergency endoscopy that had been done on admission. Using a bipolar high-frequency electric coagulation gold probe (output, 30W/s, for 5s; Boston Scientific, Boston, MA, USA), hemostasis was achieved (Fig. 3a,b). After the endoscopic hemostasis, no additional hemorrhage occurred. Nine days after the hemostasis, endoscopic resection was performed. The tumor base was fastened using an indwelling snare (HX20L-1; Olympus, Tokyo, Japan) on the anal side about 1cm from the papillary orifice, and polypectomy was performed. High-frequency output was established with 30W in the coagulation mode (Olympus PSD-30) (Fig. 3c,d).

Macroscopically, the resected specimen was a slightly nodular submucosal tumor with a smooth surface, measuring 30 × 25 × 10mm (Fig. 4). Microscopically, the tumor involved the submucosa to muscular layers, and, partially, the mucosa, but it was well demarcated (Fig. 5a). The tumor consisted of epitheloid cells, ganglion-like cells, and spindle cells. Epitheloid cells with oval or round nuclei showed an alveolar or trabecular arrangement (Fig. 5b), similar to the features of carcinoid tumor or paraganglioma. Furthermore, large ganglion-like cells were observed among numerous spindle cells (Fig. 5c), showing features that were identical to ganglioneuroma.

Table 1 summarizes the results of immunohistochemical studies of this tumor. The epitheloid cells were positive for neuron-specific enolase (NSE) chromogranin A (CGA), pancreatic polypeptide