Sporadic gastric neurofibroma underneath early cancer: MDCT gastrography and histological findings

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Abstract We report the case of a sporadic gastric submucosal neurofibroma underneath a T1 stage cancer. A 61-year-old man underwent gastroscopy because of epigastralgia and was diagnosed as having T1 stage gastric cancer by an experienced gastroenterologist. Subsequently performed computed tomography (CT) showed poorly circumscribed wall thickening underneath the converged folds on three-dimensional images. On a dynamic enhancement study, the thickened wall was seen to be enhanced gradually from the arterial phase to the equilibrium phase. Based on these findings, we diagnosed stage T2 cancer. Total gastrectomy was performed, and the surgically removed specimen revealed that the wall thickening was caused by a submucosal neurofibroma and that cancer existed in this neurofibroma, invading the submucosa. This patient had no family history of neurofibromatosis, and so the lesion was diagnosed as early gastric cancer with a sporadic submucosal neurofibroma. Coexistence of gastric cancer and a submucosal tumor is rare, but such a case is one of the pitfalls of a CT diagnosis of T stage gastric cancer.

Key words Stomach · Submucosal tumor · Neurofibroma · CT gastrography

Introduction

Recent advancements of multidetector-row computed tomography (MDCT) have made it possible to detect and assess gastrointestinal lesions both two- and three-dimensionally.1–4 We describe a case of poorly circumscribed submucosal neurofibroma located underneath an early gastric cancer, which led to misdiagnosis of advanced gastric cancer on CT gastrography. Sporadic neurofibroma is a rare submucosal tumor of the gastrointestinal tract, and to our knowledge this is the first case of one located underneath a gastric cancer, with its characteristics demonstrated by CT.

Case report

A 61-year-old man came to our hospital complaining of epigastralgia and subsequently underwent gastric endoscopy, which detected gastric cancer in the posterior wall of the upper gastric corpus (Fig. 1A). The histological diagnosis of a biopsy specimen was well-differentiated tubular adenocarcinoma. An experienced gastroenterologist staged the lesion as T1 based on the mucosal feature of converging folds. On subsequently performed upper gastrointestinal X-ray barium series (UGIs), faint fold convergence on a slight elevation at the upper gas-
tric corpus was noted (Fig. 1B). Radiologists and gastroenterologists diagnosed the T stage of this lesion as T1 because prominent thickening of the fold was not noted, and the slight elevation was thought to be caused by edema, lymphoid hyperplasia, or something else.

The patient underwent MDCT gastrography before surgery to determine the clinical stage of the cancer. On three-dimensional images, subtle mucosal changes with converging folds was noted in the posterior wall of the upper gastric corpus. Although this mucosal lesion appeared to be located above an ill-defined low elevation, no fold thickening was noted (Fig. 2A). Two-dimensional images revealed poorly circumscribed localized gastric wall thickening in the thin gastric wall stretched by air manually pushed with a plastic syringe via a nasoesophageal tube. On a dynamic enhancement study, this thickened segment of the wall was gradually enhanced from the arterial phase to the equilibrium phase (Fig. 2B). Neither perigastric strands nor regional lymphadenopathy was noted around the cancer. Based on these findings, we diagnosed the lesion as T2 advanced cancer.

Total gastrectomy was performed, and the removed specimen revealed that a poorly circumscribed submucosal lesion had caused the gastric wall thickening (Fig. 2C). Furthermore, well-differentiated tubular adenocarcinoma had mainly invaded the mucosa propria of this submucosal lesion, although a small number of cancer cells had invaded the submucosa. Immunohistochemical study revealed that this submucosal lesion consisted of S-100 protein antibody-positive spindle cells. The patient had neither signs nor a family history of neurofibromatosis type 1 (NF1). Thus, the pathological diagnosis was T1 stage gastric cancer with a sporadic submucosal neurofibroma (Fig. 2D).

Discussion

Neurofibroma is a rare gastric submucosal tumor that is commonly associated with NF1. It may cause gastric bleeding and is sometimes located in the gastric antrum, where it can cause gastric outlet obstruction. Sporadic or solitary neurofibroma of the stomach is rare and has been reported in only a few cases. The clinical features of sporadic neurofibroma of the stomach are the same as those associated with NF1, and CT findings have been reported for only one such case, with no reports having discussed its imaging characteristics. To the best of our knowledge, there have been no cases reported in the English-language literature of a gastric cancer coexisting with a submucosal tumor.

Most gastric submucosal tumors are well circumscribed, as in the case of gastrointestinal stromal tumors (GISTs), and thus the diagnosis of a submucosal tumor is easily done with UGIs or endoscopy or in some cases with CT or magnetic resonance imaging (MRI). In our case of neurofibroma, the tumor was poorly circumscribed and had no fibrous capsule with little mucosal elevation. Because well-differentiated tubular adenocarcinoma was proved in the mucosal lesion in the biopsy specimen, we performed CT gastrography to determine the T stage of the cancer. This mucosal lesion was located just above the locally thickened gastric wall. A dynamic enhancement study showed that this submuco-

![Fig. 1. A Conventional endoscopy shows converging folds to slight depression in the posterior wall of the upper gastric corpus (surrounded by arrows). B Double-contrast upper gastrointestinal barium enhancement study shows faint folds convergence on a minimally elevated mucosa (arrows)](image)