

Gastric teratoma invasion and bulb fistula formation in an adult: report of one case and literature review

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Abstract

Gastric teratoma primarily occurs within 3 months following birth, and is a rare pattern of gastric lesion in adult patients. The present study reports the case of a 60-year-old male patient who was diagnosed with a tumour in the lesser curvature within the gastric cardia area, which grew outside the cavity, invaded into the duodenal bulb and formed a gastroduodenal fistula. Briefly, initial gastroscopy upon hospital admission revealed mucosa bulging into the gastric cavity, gastric ulcer and duodenal bulb mucosal congestion with oedema. Subsequent computed tomography scans showed lesser curvature-occupying hamartoma in the gastric cardia area, and upper gastrointestinal angiography confirmed gastric stromal tumour complicated with cardia duodenal fistula. Total gastrectomy followed by Roux-en-y oesophagojejunostomy was performed, and pathology analysis of the tissue specimen confirmed mature gastric teratoma. The formation of a gastroduodenal bulb fistula with the tumour as a bridge is a rare phenomenon. A notable finding of the present case study was that the final diagnosis of gastric teratoma mainly depended on pathological examination.

Keywords

Gastric teratoma, pathological examination, gastroduodenal fistula

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Introduction

A teratoma is a category of congenital tumour that is derived from three primary germ layers and occurs at a location outside the tissue components. Teratomas

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frequently occur in the sacral, sacrococcygeal and retroperitoneal sites.¹ Gastric teratomas are classified as a distinct sub-group and yield worse clinical prognosis compared with teratomas located at alternative sites.² The most common symptoms associated with teratomas comprise abdominal mass, constipation, anorexia, fever, vomiting and weight loss.¹⁻³

In the present study, the case of a 60-year-old male patient who was diagnosed with gastric teratoma is reported, in which the gastric teratoma invaded into the duodenal bulb and formed a gastroduodenal fistula. To the best of the present authors' knowledge, this is the first published case of gastric teratoma with duodenal bulb invasion and gastroduodenal fistula formation, and aims to add evidence to the clinical diagnosis and treatment of such rare cases.

Case report

A 60-year-old male patient presented with epigastric distension and discomfort for two months and aggravated symptoms for one day. He was admitted to the Department of Gastroenterology, Xingtai People's Hospital, Xingtai, China on 14 March 2017. Written consent to publish the case

was obtained from the ethics committee of Xingtai People's Hospital and written informed consent was obtained from the patient. The patient had a medical history of hypertension for 10 years and cerebral thrombosis for 7 years. He had not previously received surgical interventions. Gastroscopy was performed upon admission to the Department of Gastroenterology, and demonstrated that mucosa bulged into the gastric cavity and apical ulcers appeared on the posterior wall of the lesser curvature in the gastric cardia area, and three pieces of ulcerous tissue were identified by biopsy, as illustrated in Figure 1a. Duodenal bulb mucosal congestion and oedema were noted (Figure 1b). The spherical cavity was so narrow that the endoscope lens could not pass. Endoscopic results provided a diagnosis of suspected cardiac stromal tumours, chronic gastritis and duodenal ulcer. Pathological biopsy revealed signs of acute or chronic inflammation of the cardiac mucosa, papillary hyperplasia of the squamous epithelium, but no abnormal changes in the epithelioid cell mass of the small lesion. The patient was then transferred to the Department of Gastrointestinal Surgery,

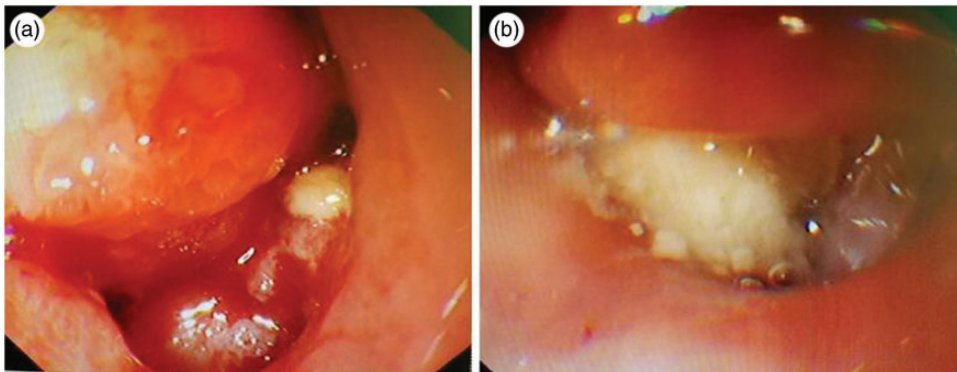


Figure 1. Gastroscopy images showing: (a) mucosa bulged into the gastric cavity and the appearance of apical ulcer on the posterior wall of the lesser curvature of the cardia; and (b) duodenal bulb mucosal congestion and oedema, with a spherical cavity so narrow that the endoscope lens could not pass.

Xingtai People's Hospital on 23 March 2017, and was subsequently diagnosed with gastric stromal tumour, hypertension and the sequelae of cerebral thrombosis. On the second day following admission to the Department of Gastrointestinal Surgery, a coronal computed tomography (CT) scan showed mass shadows that were projected into the abdominal cavity on the lesser curvature side of the gastric cardia, with a clear boundary and unevenly-mixed internal density, visible fat density, soft tissue density, multiple nodular calcium spots and massive calcifications (Figure 2). A biphasic contrast-enhanced CT scan demonstrated no significant contrast enhancement. The CT scan provided the diagnosis of lesser curvature-occupying hamartoma in the gastric cardia area. Upper gastrointestinal angiography was performed on the following day, and demonstrated a circular filling defect, uneven application of barium and irregular niche shadows in the lesser curvature of the gastric cardia (Figure 3). The tumour was found to be growing outside the gastric cavity and adhered to the duodenal bulb. The diameter of the lesion was approximately 5 cm. Some intragastric contrast agents were observed to have entered the duodenal bulb through an 'arch bridge' channel from the cardia along the tumour body, and the duodenal bulb was pulled toward the lesser curvature of the gastric body (Figure 4). Upper gastrointestinal angiography confirmed the final diagnosis of gastric stromal tumour complicated with cardia duodenal fistula. The patient underwent surgery on day 6 following admission to the Department of Gastrointestinal Surgery. Intraoperatively, the mass was found to be approximately 6 × 6 cm in size and located in the lesser curvature of the gastric cardia area. The mass had invaded into the duodenal bulb and had become part of the right cardiac diaphragm. Total gastrectomy was

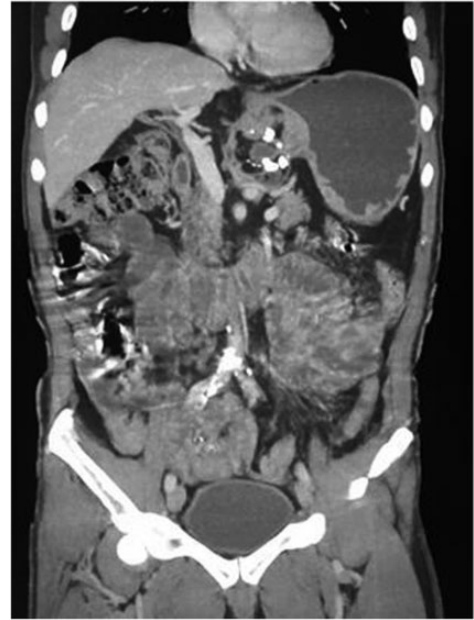


Figure 2. Coronal computed tomography scan image showing mass shadows projecting into the abdominal cavity on the lesser curvature side of the gastric cardia, with clear boundary and uneven mixed internal density, visible fat density, soft tissue density and multiple nodular calcium spots and massive calcifications.

performed, followed by Roux-en-y oesophagojejunostomy, and the gastric specimen was collected and prepared for postoperative pathological examination. Pathological findings revealed that the tumour measured 6 × 5 × 4 cm in size. The mass was cystic and solid with a grey-yellow, soft and tough surface, and the cystic area was full of oil, hair and focal calcification. Microscopically, three layers of embryonic tissues were seen. Haematoxylin and eosin staining of tissue sections demonstrated that the surface of the cyst wall was covered with squamous epithelium, skin appendage, bronchial epithelium, brain tissue and focal calcification (Figure 5). Pathology results provided a diagnosis of mature gastric teratoma in



Figure 3. Upper gastrointestinal angiography image showing a circular filling defect and irregular niche shadows in the lesser curvature of the gastric cardia. The tumour was observed to have grown out of the gastric cavity and adhered to the duodenal bulb.

the lesser curvature of the gastric cardia area.

Discussion

Gastric teratoma is a rare disease that predominantly occurs in male infants within 3 months after birth, accounting for 94% of all cases,¹ whereas gastric teratoma is extremely rare in adult patients.² In most cases, gastric teratoma is located in the posterior wall of the stomach, adjacent to the cardia, and is rarely observed on the greater curvature side.³ In clinical practice, gastric teratoma is generally divided into cystic, solid and mixed categories, whereby mixed and benign gastric teratoma is the dominant type. Like other types of teratomas, gastric teratoma contains three germ-layer components that derive from the outer, middle and inner layers of the original

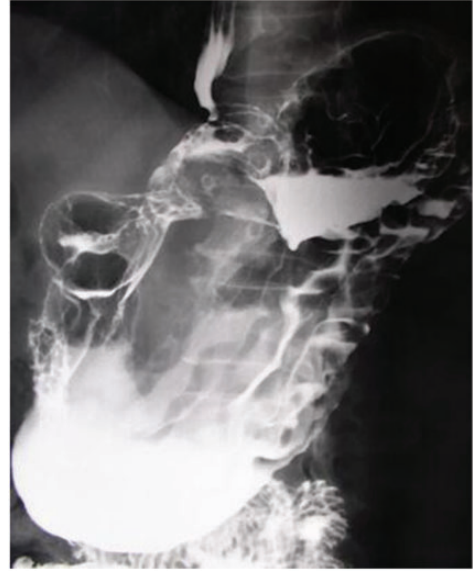


Figure 4. Upper gastrointestinal angiography image showing that some intragastric contrast agents had entered the duodenal bulb through an 'arch bridge' channel from the cardia along the tumour body. The duodenal bulb was observed to have been pulled toward the lesser curvature of the gastric body.

germ layer.² Most of these components are mature tissues, and even if a few immature nerve tissues are visible with the pathological manifestations of immature teratoma, the teratoma is clinically confirmed to be benign.² The use of X-ray, B-scan ultrasound, CT scan and gastroscopy are of significance for clinical diagnosis.¹⁻³ For example, abdominal X-ray film demonstrates irregular calcification in the teeth and in the shadow of the left rib area, which contributes to the clinical diagnosis of gastric teratoma. The barium-meal examination is also significant for clinical diagnosis of gastric teratoma, as defects in filling to the gastric cavity, and irregular or ulcerated mucosa of the posterior wall of the stomach, can be observed. B-scan ultrasound can indicate the solid area of the cyst, and may occasionally detect calcification. A CT scan is

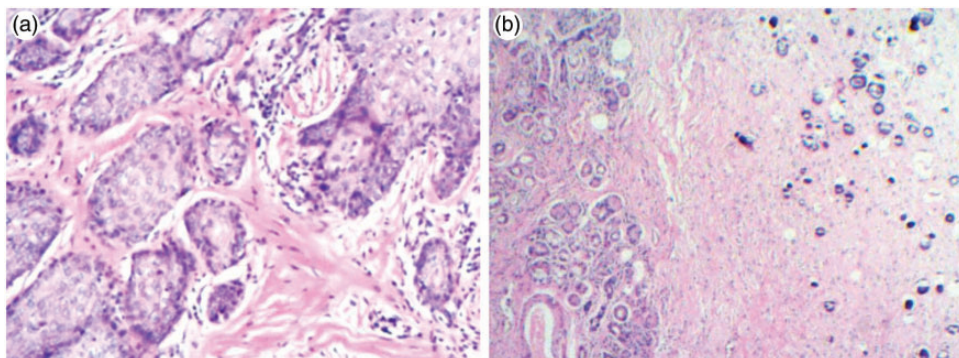


Figure 5. Photomicrographs of haematoxylin and eosin-stained sections of gastric tissue obtained during total gastrectomy, in which three layers of embryonic tissue were observed, showing: (a) The surface of the cyst wall covered with squamous epithelium, skin appendage, bronchial epithelium, brain tissue, and focal calcification; and (b) the teratoma along with residual normal gastric tissues (original magnification $\times 100$).

able to determine the tumour boundaries, distinguish the internal structural components of the tumour and determine the relationship between the tumour and surrounding tissues. Hair is visible on the surface of a typical mature teratoma lesion under gastroscopy, which contributes to the diagnosis.³⁻⁵ In clinical practice, few adult cases with typical features have been reported, and consequently, it is highly likely that gastric teratoma will be ignored and misdiagnosed in suspected adult cases. Indeed, the preoperative misdiagnosis rate for adult cases of gastric teratoma is 100%.⁵ The major reasons are as follows: first, the site of gastric teratoma occurrence is rare; secondly, the size of the tumour is small and it is difficult to identify without pathological biopsy; thirdly, adult gastric teratoma is cystic without bone-like tissues; and fourthly, no clinically specific symptoms can be observed in cases of adult gastric teratoma.⁶

In the present case report, the patient was aged 60 years, and preoperative gastroscopy and CT scan combined with upper gastrointestinal angiography failed to provide a correct diagnosis. Gastroscopy showed that mucosa bulged into the gastric cavity and apical ulcer

appeared on the posterior wall of the lesser curvature in the gastric cardia area, both of which are common symptoms of gastric stromal tumours, and thus, likely to be misdiagnosed as gastric stromal tumour. The finding of hair growing on the tumour surface is more likely to result in correct identification. The patient was diagnosed with duodenal bulb ulcer due to the findings of duodenal bulb mucosal congestion and oedema, and the narrow spherical cavity. The pathological results seemed reasonable because biopsy failed to sample deeply enough or the sampling location was incorrect. Retrospective CT scan showed no significant enhancement, hinting at the possibility of benign tumour. Multiple calcifications were detected in the mixed density mass, which is a typical manifestation of gastric teratoma, whereas the CT scan gave the signs of suspected gastric hamartoma. A review of the literature revealed that gastric hamartoma is also extremely rare and mainly occurs in the greater curvature of the gastric antrum and gastric fundus, and the gastric pedicle or subpedicle of multiple small polyps with more than 6 pieces with a diameter <0.5 cm, complicated with congestion without erosion or ulcer.^{7,8} However, the location, size,

structure and density of the lesion in the present case were not consistent with the symptoms of gastric hamartoma. Upper gastrointestinal angiography indicated a diagnosis of gastric stromal tumour due to the following: First, gastric stromal tumour is the most common tumour of the digestive tract; Secondly, the age of onset was just appropriate, occurring primarily in the middle-aged and elderly population; Thirdly, the location of onset was similar to the sites of stromal tumours, as gastric stromal tumours are more likely to occur in the gastric fundus, cardia and gastric body, but less common in the gastric antrum; and Fourthly, in cases of gastric stromal tumour, large lesions have often grown outside the cavity with necrosis inside the lesion, which was similar to the present case.⁸ Nevertheless, calcification is rarely observed in gastric stromal tumours, and gastric teratoma often has calcification or ossification, which significantly contributes to the final diagnosis. In the present case, this specific symptom was neglected during the upper gastrointestinal angiography.

In conclusion, gastric teratoma is extremely rare in adults, and gastric teratoma complicated with duodenal bulb invasion and cardioduodenal fistula formation is even less frequently encountered in clinical practice. Upper gastrointestinal tract angiography, and CT scan combined with endoscopy were found to yield no specific identification, which may likely result in a high misdiagnosis rate. Consequently, the final diagnosis of gastric teratomas still relies upon pathological examination and biopsy.

Declaration of conflicting interest

The authors declare that there is no conflict of interest.

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