



CASE REPORT

Primary gastric teratoma on the cardiac orifice in an adult

Liu Liu, Wen Zhuang, Zhong Chen, Yong Zhou, Xiao-Ran Huang

Liu Liu, Wen Zhuang, Yong Zhou, Department of Gastrointestinal Surgery, West China Hospital/West China Medical Center, Sichuan University, Chengdu 610041, Sichuan Province, China

Zhong Chen, Department of Pediatrics, The 2nd West China Hospital of Sichuan University, Chengdu 610041, Sichuan Province, China

Xiao-Ran Huang, Department of Pathology, West China Hospital/West China Medical Center, Sichuan University, Chengdu 610041, Sichuan Province, China

Author contributions: Liu L and Chen Z collected the clinical data; Liu L and Huang XR collected the pathological data and wrote the paper; Zhou Y and Zhuang W revised the paper.

Correspondence to: Wen Zhuang, Department of Gastrointestinal Surgery, West China Hospital/West China Medical Center, Sichuan University, Guo Xue Xiang NO. 37, Chengdu 610041, Sichuan Province, China. zhuangwen2008@126.com

Telephone: +86-28-85422872 Fax: +86-28-85164035

Received: January 12, 2009 Revised: March 12, 2009

Accepted: March 19, 2009

Published online: April 14, 2009

Abstract

Gastric teratoma (GT) is a seldom seen congenital abnormality. GT always occurs in children. The greater curvature and posterior wall of the stomach are the most common sites involving GT. We diagnosed a case of GT located on the inferior wall of the cardiac orifice in a 20-year-old man. To the best of our knowledge, this is the first case of GT located on the wall of the cardiac orifice in an adult in the English literature. We report this unusual case as an addition to this rare disease usually found in children. Computed tomography combined with endoscopic ultrasonography can be selected to diagnose GT.

© 2009 The WJG Press and Baishideng. All rights reserved.

Key words: Adult; Cardiac orifice; Endoscopic ultrasonography; Stomach; Teratoma

Peer reviewer: Luis Bujanda, MD, PhD, Avda, Sancho El Sabio, 21-3°C, 20010 San Sebastián, Spain

Liu L, Zhuang W, Chen Z, Zhou Y, Huang XR. Primary gastric teratoma on the cardiac orifice in an adult. *World J Gastroenterol* 2009; 15(14): 1782-1785 Available from: URL: <http://www.wjgnet.com/1007-9327/15/1782.asp> DOI: <http://dx.doi.org/10.3748/wjg.15.1782>

INTRODUCTION

Gastric teratoma (GT) is a seldom seen congenital abnormality. It has been reported in the world literature that GT always occurs in children^[1,2], and an increasing mass in the epigastric region is the main sign of GT^[1,3,4]. Only six cases of GT in adults were reported in Medline^[5-8] between 1922 and 2007. Yoon *et al*^[9] reported the second case of GT in a child in Korea in 2000. GT is usually located on the posterior wall or greater curvature of the stomach^[3,7]. To the best of our knowledge, this is the first report of GT located on the wall of the cardiac orifice in an adult in the English literature. Calcifications in the cystic-solid mass may be features of GT on computed tomography (CT)^[3,4,10]. Endoscopic ultrasonography (EUS) may help to correctly diagnose GT before surgery.

CASE REPORT

The patient was a 20-year-old man. Due to slight pain and distention of the upper abdomen of 3 mo duration, he was admitted to our hospital. Physical examination was carried out and no mass was found in the epigastrium. The level of serum alpha-fetoprotein (AFP) was normal. CT showed a spherical mass on the inferior wall of the cardiac orifice of the stomach. The mass was about 4.8 cm × 5.2 cm, and there was no clear border between the mass and the wall of the stomach. The density of the mass was uneven. High-density and low-density substances were found in the mass (Figure 1). EUS demonstrated a giant mass on the inferior wall of the cardiac orifice. The mucosa of the mass was normal (Figure 2), and there was a lobulated polyp near the mass. The five-layer structure of the stomach was clearly manifested in the mass on ultrasonography (Figure 2). The mass had formed from the outer layer of the stomach. The mass was completely excised with a partial gastrectomy and the digestive tract was rebuilt. Macroscopically, the mass was about 5.0 cm × 5.3 cm × 2.3 cm, and was derived from the inferior wall of the cardiac orifice (Figure 3A). The mass contained small cystic tissue and a large solid area. Microscopically, the tumor was resected completely in one piece with a tumor cell-free margin. Cartilage, squamous cells and respiratory epithelium were observed in the mass (Figure 3B-F). No immature tissue was observed in the mass. The diagnosis of a primary mature GT derived from the cardiac orifice was established. The syndromes

Table 1 Five reported cases of GT in Medline between 1922 and 2007

Author (yr)	Age (yr)	Signs and symptoms	Location	Type	Size
Eustermann <i>et al</i> ^[6] 1922	31	?	Posterior wall	Mature	7 cm × 6 cm × 5 cm
Fadeeva <i>et al</i> ^[12] 1960	25	?	Anterior wall	Mature	4 cm × 3 cm
Gray <i>et al</i> ^[8] 1964	40	?	Lesser curvature	Mature	7 cm × 6 cm
Matsukuma <i>et al</i> ^[5] 1995	83	Tarry stool and anemia	Lesser curvature	Immature	12 cm × 10 cm × 6 cm
Joo <i>et al</i> ^[7] 1999	27	Fever, pain and vomiting	Greater curvature	Mature	9.5 cm × 7.5 cm × 5 cm

Note: Five patients were all male. Complete resection of the tumor was carried out in all patients and all patients recovered.

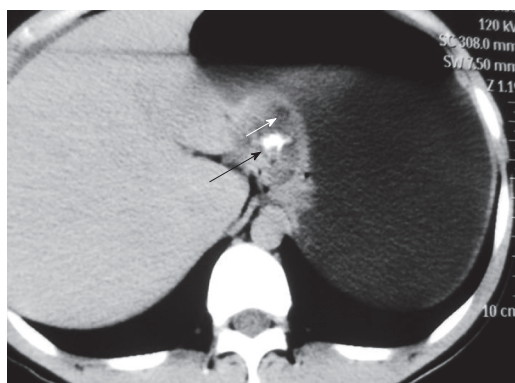


Figure 1 Axial CT shows a large soft tissue mass located on the inferior wall of the cardiac orifice of the stomach. The border of the mass is clear and the mass is from the stomach. The density of the mass is uneven. Low density (white arrow) indicates cystic tissue and the high density (black arrow) indicates calcifications.

in this patient disappeared within 1 wk after surgery. In the 10-mo follow-up period, the patient was well without abdominal discomfort.

DISCUSSION

Teratoma usually occurs in the gonads (mainly between the age of 6 mo and 15 years), the sacrococcygeal region (mainly in infants)^[1,2], and in the retroperitoneum, cranium or mediastinum^[5]. Teratoma derived from the stomach is very rare, and was first reported by Eustermann and Sentry in 1922^[6]. Up to 2002, only 107 cases of GT had been reported in the English literature^[1]. Surprisingly, GT always occurs in male children, especially around the age of 1 year^[1-3,5,11]. GT in an adult is unusual. To the best of our knowledge, there were just six cases of GT in adults reported in MEDLINE between 1922 and 2007, and five of these cases are listed in Table 1^[5-8,12]. GT is commonly located on the greater curvature and the posterior wall of the stomach^[3,7]. In the five cases listed in Table 1, two cases originated from the lesser curvature, and three cases from the posterior wall, anterior wall and the greater curvature, respectively. To the best of our knowledge, we report the seventh case of GT in an adult in the English literature and the first case originating from the cardiac orifice. Partly because of the rarity of GT in adults, the preoperative diagnosis of GT is difficult for surgeons^[7].

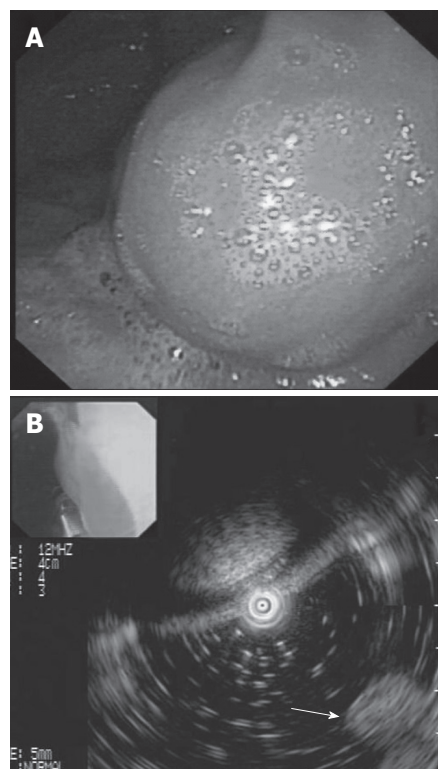


Figure 2 EUS was used to diagnose the gastric teratoma. A: A spherical mass was noted on the inferior wall of the cardiac orifice, and the mass mucosa was normal; B: The heterogeneous mass was formed from the outer layer and the five-layer structure of the stomach in the mass was clearly detected (white arrow).

Typical radiographic findings of GT^[3,4,10], such as calcifications, uneven density of the tumor and an intratumoral solid area with mixed cysts, were predominant in our case (Figure 1). EUS was first used to diagnose GT. A mass from the inferior wall of the cardiac orifice was observed. EUS demonstrated a heterogeneous mass from the outer layer of the stomach, and the five layers of the stomach were clearly detected (Figure 2). The echo pattern in the mass and the mass from the outer layer of the stomach were different from those of other common submucosal tumors, such as lipoma, myogenic tumor and lymphangioma. The use of EUS alone cannot help to diagnose GT qualitatively, but could provide useful information for preoperative diagnosis.

Cartilage, squamous cells and respiratory epithelium were found histologically (Figure 3F). According to pathological criteria, cartilage, squamous cells and respiratory epithelium in the mass are interesting

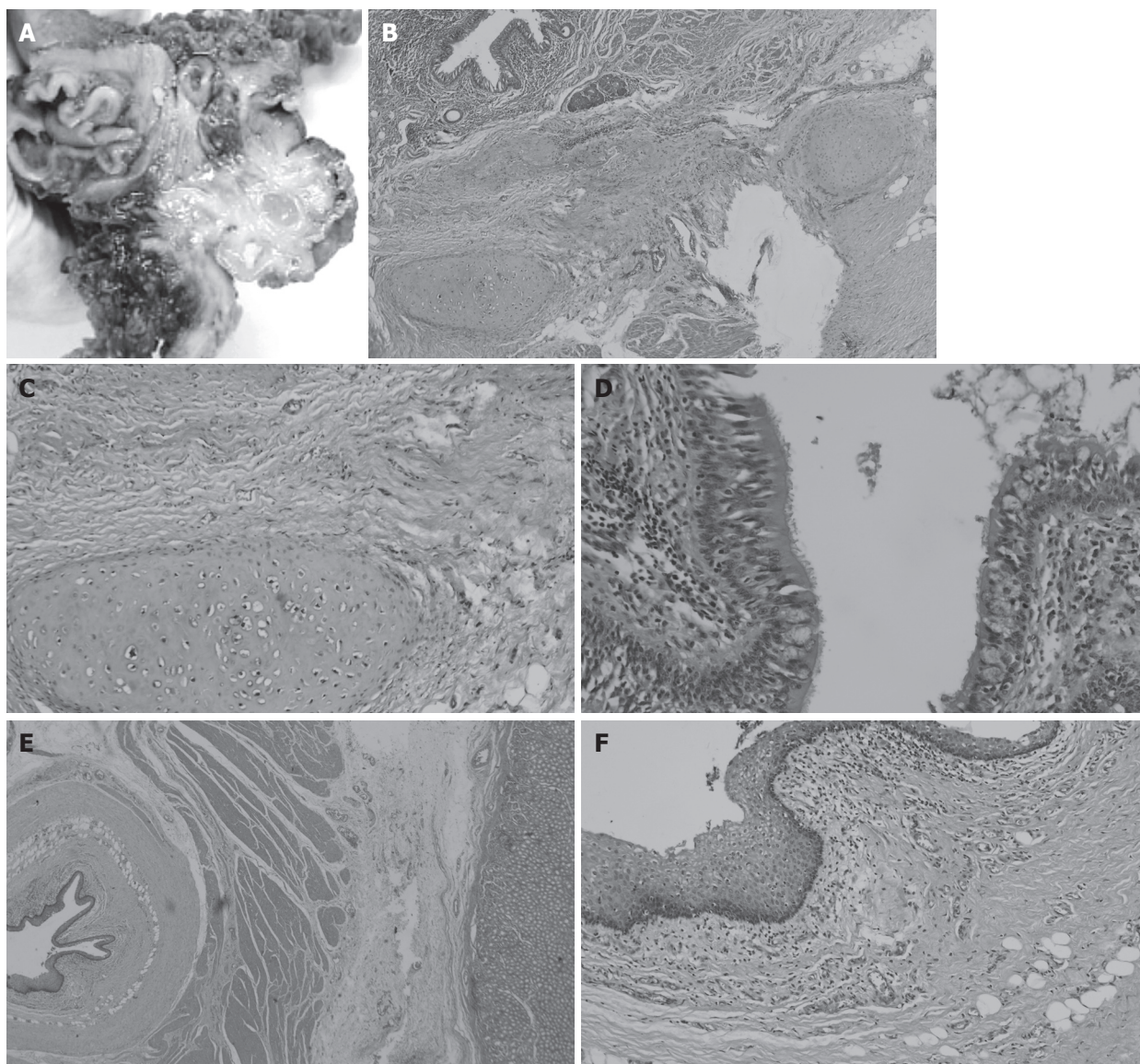


Figure 3 GT was pathologically examined under macroscopy and microscopy. A: The mass was about 5.0 cm × 5.3 cm × 2.3 cm, and it derived from the inferior wall of the cardiac orifice; B: Photomicrograph of an area composed respiratory epithelium and cartilage without immature teratoma components (HE, × 20); C, D: The cartilage (C) and the respiratory epithelium (D) were amplified (HE, × 100); E: The three layers, mucous layer, submucous membrane, and muscular layer, of the wall of the stomach were clear. The tissue of GT was derived from the muscular layer of the stomach (HE, × 20); F: The Squamous cell was amplified (HE, × 100).

histological features of teratoma. The diagnosis of GT from the cardiac orifice was therefore exclusively established. Complete resection is an effective method to treat both benign and malignant GT. Compared to traditional surgery, laparoscopic surgery has many advantages, and has been used to treat teratomas^[13], and will be used to treat GT in the future. AFP is a good indicator of the recurrence of malignant GT^[1,3,11]. Subsequent chemotherapy might be required when the level of AFP is high after surgery.

In conclusion, GT is rare and always occurs in infancy or childhood. The greater curvature and the posterior wall of the stomach are the most common sites involving GT. GT involving the cardiac orifice in an adult is seldom seen. To the best of our knowledge, this is the first report involving GT on the cardiac orifice in the English literature. EUS is helpful in diagnosing GT.

REFERENCES

- 1 Ukiyama E, Endo M, Yoshida F, Tezuka T, Kudo K, Sato S, Akatsuka S, Hata J. Recurrent yolk sac tumor following resection of a neonatal immature gastric teratoma. *Pediatr Surg Int* 2005; **21**: 585-588
- 2 Göbel U, Calaminus G, Engert J, Kaatsch P, Gadner H, Böklerink JP, Hass RJ, Waag K, Blohm ME, Dippert S, Teske C, Harms D. Teratomas in infancy and childhood. *Med Pediatr Oncol* 1998; **31**: 8-15
- 3 Utsch B, Fleischhack G, Knöpfle G, Hasan C, Bode U. Immature gastric teratoma of the lesser curvature in a male infant. *J Pediatr Gastroenterol Nutr* 2001; **32**: 204-206
- 4 Moriuchi A, Nakayama I, Muta H, Taira Y, Takahara O. Gastric teratoma of children--a case report with review of the literature. *Acta Pathol Jpn* 1977; **27**: 749-758
- 5 Matsukuma S, Wada R, Daibou M, Watanabe N, Kuwabara N, Abe H, Suda K. Adenocarcinoma arising from gastric immature teratoma. Report of a case in an adult and a review of the literature. *Cancer* 1995; **75**: 2663-2668

- 6 **Eustermann GB**, Sentry EG. Benign tumours of the stomach: report of 27 cases. *Surg Gynecol Obstet* 1922; **34**: 372-378
- 7 **Joo M**, Kang YK, Lee HK, Lee HS, Yum HK, Bang SW, Cho HJ. Intrapulmonary and gastric teratoma : report of two cases. *J Korean Med Sci* 1999; **14**: 330-334
- 8 **Gray SW**, Johnson HC Jr, Skandalakis JE. Gastric teratoma in an adult: with a review of the literature. *South Med J* 1964; **57**: 1346-1351
- 9 **Yoon SE**, Goo HW, Jun S, Lee IC, Yoon CH. Immature gastric teratoma in an infant: a case report. *Korean J Radiol* 2000; **1**: 226-228
- 10 **Bowen B**, Ros PR, McCarthy MJ, Olmsted WW, Hjermstad BM. Gastrointestinal teratomas: CT and US appearance with pathologic correlation. *Radiology* 1987; **162**: 431-433
- 11 **Corapçioğlu F**, Ekingen G, Sarper N, Güvenç BH. Immature gastric teratoma of childhood: a case report and review of the literature. *J Pediatr Gastroenterol Nutr* 2004; **39**: 292-294
- 12 **Fadeeva VN**, Shafer II. A case of teratoma of the stomach. *Ark Patol* 1960; **22**: 55
- 13 **Takao Y**, Shimamoto C, Hazama K, Itakura H, Sasaki S, Umegaki E, Nakagawa K, Hirata I, Katsu K. Primary rectal teratoma: EUS features and review of the literature. *Gastrointest Endosc* 2000; **51**: 353-355

S- Editor Tian L L- Editor Webster JR E- Editor Zheng XM