



Case report

Primary gastric adenocarcinoma in a 2.5-year-old girl

ALI IHSAN DOKUCU¹, HAYRETTIN ÖZTÜRK¹, NIHAL KILINÇ², ABDURRAHMAN ÖNEN¹, YASAR BUKTE³,
and MURAT SOKER⁴

¹Dicle University, Medical School, Department of Pediatric Surgery, 21280 Diyarbakır, Turkey

²Dicle University, Faculty of Medicine, Department of Pathology, Diyarbakır, Turkey

³Dicle University, Faculty of Medicine, Department of Radiology, Diyarbakır, Turkey

⁴Dicle University, Faculty of Medicine, Department of Pediatrics, Diyarbakır, Turkey

Abstract

Primary gastric adenocarcinoma is extremely rare in children. Here, we report an additional case of primary adenocarcinoma, located at the lesser curvature in a girl at the age of 2.5 years. She had no family history and no apparent underlying cause for the tumor. She died 4 months after admission despite complete resection of the mass and chemotherapy.

Key words Gastric carcinoma · Children adenocarcinoma

Introduction

Primary gastric carcinomas are extremely rare in childhood, although gastrointestinal malignancies represent approximately 5% of all pediatric neoplasms [1]. A brief literature review showed around ten cases reported in patients under the age of 5 years since 1978 [2–5]. Here, we report a patient suffering from primary adenocarcinoma of the stomach at age 30 months.

Case report

A 2.5-year-old girl from a rural area presented at our institution with complaints of abdominal distension, weight loss, paleness, and weakness during the previous few weeks. Her parents had discovered an abdominal mass and blood in the stool a week before her admission. The mass, which was about 8 × 6 cm in diameter, was palpable in the upper left abdominal region. Evaluation of the abdomen by both ultrasonography and computerized tomography (Fig. 1) showed a heterogeneous, lobulated mass (which was 8 × 8 cm in diameter) in the stomach, with direct extension into surrounding tissues. Before an exploratory laparotomy, the severe

anemia (hemoglobin [Hb], 2.6 g/l; hematocrit [Htc], 7) of the child was treated. Routine laboratory test results were nonspecific. Serum tumor markers, such as carcinoembryonic antigen alpha-fetoprotein, antigen (CEA), and human chorionic gonadotropin (HCG) were negative. Her blood group was B rh (+). She came from a family of six children living in a rural mountain area. The family history of the child was not informative. There was no history of childhood tumor or gastrointestinal malignancies in the family and close relatives. However, several gastrointestinal infections had frequently occurred in the family.

At surgery, we found a large, irregularly circumferential solid mass, extending from the corpus to 1.5 cm below the pylorus, originating from the lesser curvature. There was a firm extension to the omentum and the abdominal wall. The stomach outside the mass invasion had a normal appearance. The liver and other intraperitoneal organs appeared free of gross disease. Complete resection of the mass and a subtotal gastrectomy (80%) was performed. A retrocolic gastrojejunostomy and omentectomy, removing all gross disease, completed the procedure. The resected specimen was a dark brown mass, 12 × 7 × 7 cm in diameter, which had almost obliterated the gastric lumen. The intraluminal part of the mass had a soft, grayish, highly fragile nodular form in which gross mucous areas were remarkable. Lymph nodes in the area appeared normal, and representative biopsies were free of tumor.

Microscopically, tumoral infiltration through the gastric wall was seen. We observed glandular structures presenting atypical epithelial cells, atypical mitosis, abundant eosinophilic cytoplasm, and highly hyperchromatic cells, and some large extracellular mucin-containing areas among these structures. Immunohistochemical staining of the specimen for epithelial membrane antigen (EMA) was positive, and staining for vimentin, smooth muscle actin, and alpha-fetoprotein was negative. The histological diagnosis, according

Offprint requests to: H. Öztürk

Received: January 10, 2002 / Accepted: August 6, 2002

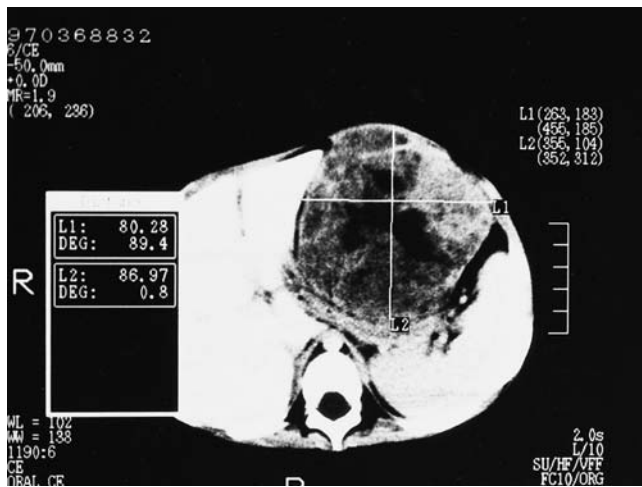


Fig. 1. Computerized tomography (CT) scan of abdomen shows a large, heterogeneous mass, 80 × 85mm in diameter, located between liver, spleen, and abdominal wall

Table 1. World Health Organization (WHO 2000) histological classification of gastric epithelial tumors [6]

	ICD-O Codes
Intraepithelial neoplasia-adenoma	8140/0
Carcinoma	
Adenocarcinoma	8140/3
Intestinal type	8144/3
Diffuse type	8145/3
Papillary adenocarcinoma	8260/3
Mucinous adenocarcinoma	8480/3
Signet-ring cell carcinoma	8490/3
Adenosquamous carcinoma	8560/3
Squamous cell carcinoma	8070/3
Small cell carcinoma	8041/3
Undifferentiated carcinoma	8020/3
Others	
Carcinoid (well-differentiated endocrine neoplasm)	8240/3

ICD-O, International Classification of Diseases for Oncology

to the International Classification of Diseases for Oncology (ICD-O), WHO classification of tumor [6], was mucin-containing gastric adenocarcinoma, intestinal type (ICD-O code, 8144/3; Table 1) (Fig. 2). The margin of the resected specimen was free of the tumor. Histology of the gastric mucosa outside the carcinoma showed no abnormality and was evaluated as normal.

The patient tolerated oral feeding on the fifth postoperative day and was transferred to the Oncology Department for chemotherapy. Combination chemotherapy containing 5-fluorouracil (FU), adriamycin, and mitomicin C was given in weekly cycles. Three weeks after the initial surgery, the patient was re-admitted for an intestinal obstruction unresponsive to medical treatment. During re-exploration and enterolysis, we found two metastatic nodules on the liver surface. She was discharged 6 days after this admission. The patient was

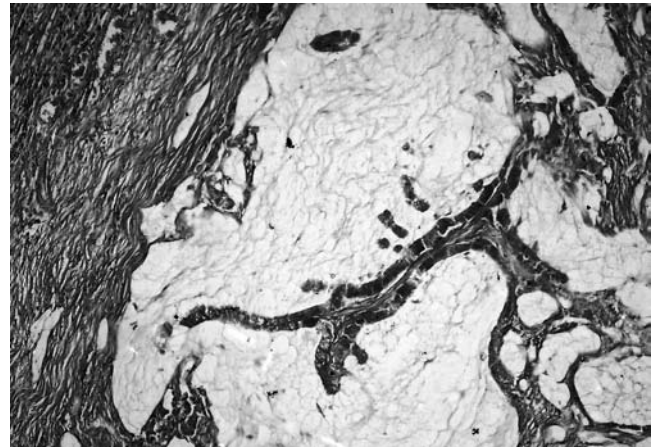


Fig. 2. Adenocarcinoma with prominent glandular formation and abundant mucin deposition. H&E, ×100

doing well for 3 months, postoperatively but then began to lose weight and to present with anemia just before the occurrence of supraclavicular lymph node enlargement. The patient died at home 1 month later.

Discussion

In most parts of the world, adenocarcinoma of the stomach is primarily a disease of older individuals, and is rare under the age of 30 years [1]. Although nearly 10% of gastric cancer cases are found in patients younger than age 41 years [7], gastric cancer is exceedingly rare in children under the age of 5 years. Our case was in a 2.5 year-old girl and our literature review showed five cases in patients under the age of 5 years [2,3,8].

The family history of the child was not enlightening in indicating the possible cause of the disease. No oncological event was reported in this large, poor rural family, who came from a socioeconomically underdeveloped area. On the other hand, frequent gastrointestinal infection in the rural areas in our region is a common problem, originating from insufficient infrastructure, that may not be related to gastric carcinoma.

Gastric adenocarcinomas are not common among gastric malignancies [1] and they may develop in three ways in the pediatric age group: de novo, as part of a polyposis syndrome, and following treatment of a gastric lymphoma [1]. Our patient is a new case of de-novo occurrence such as those that have been sporadically reported in the first and second decades of the life [1,6,9]. The lack of any kind of causative information and the normal histological findings of gastric mucosa outside the tumor suggest that our case was a new primary gastric carcinoma, which may be classified as de-novo occurrence, and which ended fatally 4 months

after diagnosis in spite of complete resection and administered chemotherapy.

References

1. Goldthorn JF, Canizaro PC. Gastrointestinal malignancies in infancy, childhood, and adolescence. *Surg Clin North Am* 1986;66: 845–61.
2. Siegel SE, Hays DM, Romansky S, Isaacs H. Carcinoma of stomach in childhood. *Cancer* 1976;38:1781–4.
3. Bethel CA, Bhattacharyya N, Hutchinson C, Ruymann F, Cooney DR. Alimentary tract malignancies in children. *J Pediatr Surg* 1997;32:1004–9.
4. McGill TW, Downey EC, Westbrook J, Wade D, de la Garza J. Gastric carcinoma in children. *J Pediatr Surg* 1993;28:1620–1.
5. Vos A. Rare tumors. In: Carachi R, Azmy A, Grosfeld JL, editors. *The surgery of childhood tumors*. New York: Arnold 1999. pp. 381–402.
6. Hamilton SR, Aaltonen LA. Gastric carcinoma. In: Hamilton SR, Aaltonen LA, editors. *Pathology and genetics of tumours of the digestive system*. 1st Ed. Oxford: Oxford University Press; 2001. pp. 38–52.
7. Kokkola A, Sipponen P. Gastric carcinoma in young adults. *Hepatogastroenterology* 2001;48:1552–5.
8. Murphy S, Shaw K, Blanchard H. Report of three gastric tumors in children. *J Pediatr Surg* 1994;29:1202–4.
9. Michalek J, Kopecna L, Tuma J, Hrstkova H, Feit J. Gastric carcinoma in a 9-year-old boy. *Pediatr Hematol Oncol* 2000;17:511–5.