



# Immature Gastric Teratoma with Rhabdomyosarcomatous and Primitive Neuroectodermal Tumor Components in an Adult Patient: A Case Report



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**Submission:** February 03, 2017; **Published:** May 15, 2017

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## Abstract

Gastric teratomas are extremely rare; the majority of cases are mature teratomas with less than two dozen immature teratomas reported. Most teratoma cases have been reported in the pediatric population with less than 10 cases reported in adults. We report a case of immature teratoma of the stomach in a 55-year-old man who presented with melena and epigastric abdominal pain. Under the clinical impression of gastric cancer, a subtotal gastrectomy was performed. Grossly, a fungating, friable, pedunculated mass (8cm in size) was identified in the gastric fundus. Histologically, an immature teratoma with rhabdomyosarcomatous and peripheral primitive neuroectodermal tumor components was identified. There was no associated gastric carcinoma or mature teratoma component.

**Keywords:** Stomach; Immature teratoma; rhabdomyosarcoma; Peripheral primitive neuroectodermal tumor; Adult

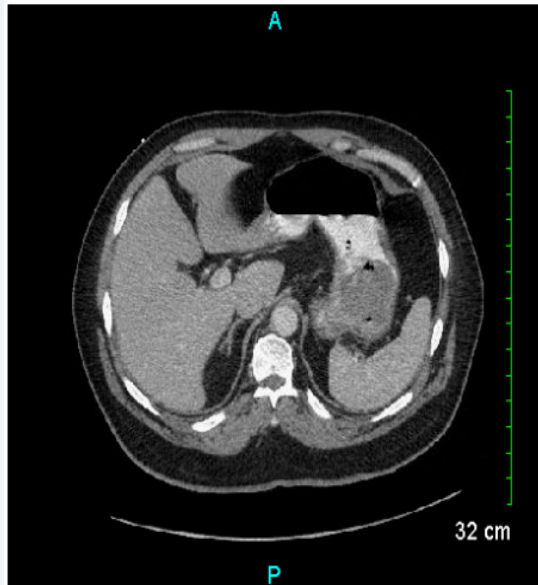
## Introduction

Teratomas of the stomach are extremely rare, comprising only about 1% of all teratomas in the pediatric population [1,2]. The majority of cases are mature teratomas with less than two dozen reported cases of immature gastric teratoma [2-4]. Most reported gastric teratomas occur in the pediatric population and less commonly in adults. The most common location is the posterior wall or greater curvature of the stomach [5,6]. Rare cases of gastric teratoma have been reported on the wall of the cardiac orifice of an adult [7] and the lesser curvature [5,8,9]. Clinically, patients can present with an abdominal mass, vomiting, and gastrointestinal bleeding [10,11]. Computed tomography (CT) findings of gastric teratomas may include calcifications in solid cystic masses [5]. Here, we report a case of an immature teratoma in a 55-year-old man who presented with melena and epigastric abdominal pain. To the best of our knowledge, this is the first case of immature teratoma with no associated gastric carcinoma or mature teratoma components in an adult.

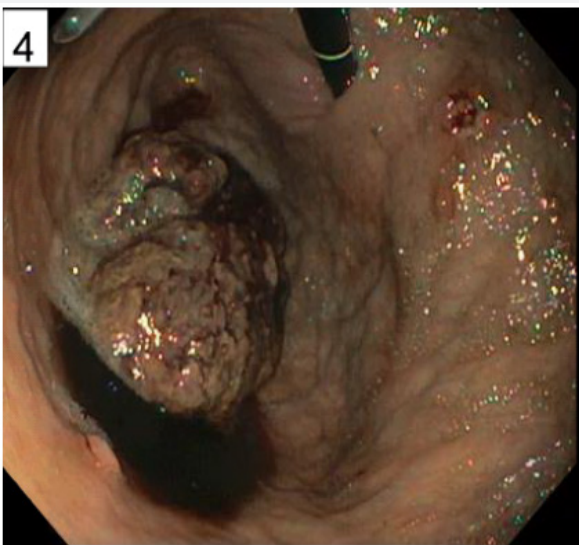
## Case Report

The patient was a 55-year-old Hispanic man who presented with anemia (hemoglobin (Hg) 7.6g/dL, hematocrit (Hct) 26%), epigastric abdominal pain, and melena. The patient had these symptoms for the past couple of months and they worsened 5 days prior to admission. His past medical history included hyperlipidemia and type 2 diabetes mellitus. On physical exam, he had a soft abdomen that was tender in the epigastric area, and had normal bowel sounds. On CT scan, an increased soft tissue density was identified in the proximal portion of the stomach (Figure 1). No other abnormality within the abdomen was found. Laboratory tests were unremarkable except for low hemoglobin and hematocrit. Preoperative serum tumor markers, such as alpha-fetoprotein (AFP), beta human chorionic gonadotropin ( $\beta$ -hCG), carcinoembryonic antigen (CEA), or lactic dehydrogenase (LDH) were not evaluated. Upper gastrointestinal endoscopy showed a polypoid mass (Figure 2). Due to continued bleeding, surgery was considered necessary

and a was performed. H&E and immunohistochemical (IHC) stainings, including desmin (Ventana, Tucson, Arizona), MyoD1 (Cell Marque, Rocklin, California), neurofilament (Ventana, Tucson, Arizona), SALL-4 (Biocare Medical, Concord, California), and synaptophysin (Ventana, Tucson, Arizona), were performed.



**Figure 1:** CT findings of an increased soft tissue density in the proximal portion of the stomach. No other abnormality was found within the abdomen.



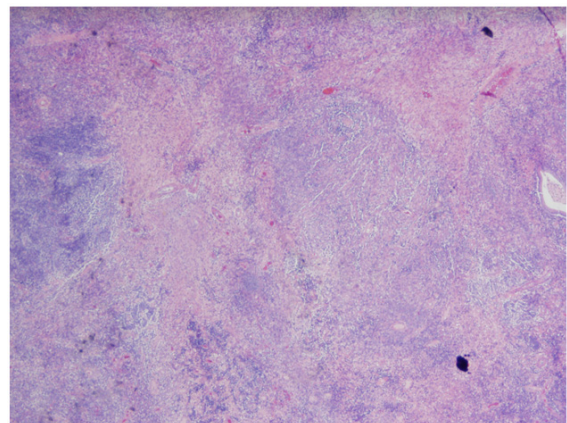
**Figure 2:** Upper GI endoscopy findings of a single polypoid mass

On gross examination, the fungating, partially fragmented, friable mass (8x6.5x2.6cm) was located in the fundus along the greater curvature, 2cm from the greater curvature margin (Figure 3). The cut surface was grayish-tan, focally necrotic, and hemorrhagic. The uninvolved gastric mucosa displayed normal rugal folds. No other

solid mass or ulcer was identified. The entire mass was submitted for histologic evaluation. Histologic examination showed immature teratoma with primitive spindle and oval-round mesenchymal sarcomatous elements, which included a rhabdomyosarcomatous (RS) component, confirmed by positivity of desmin and MyoD1 (Figure 4A-D) and a peripheral primitive neuroectodermal tumor (PNET) component confirmed by SALL-4, neurofilament, and synaptophysin (Figure 5A-E). No mature teratoma component or gastric carcinoma was seen. The tumor involved the entire wall and invaded into perigastric fibro-adipose tissue. The postoperative course was uneventful; the patient remained well, with no evidence of residual disease or metastasis. Postoperative tumor markers (AFP and  $\beta$ -hCG) were within normal limits.

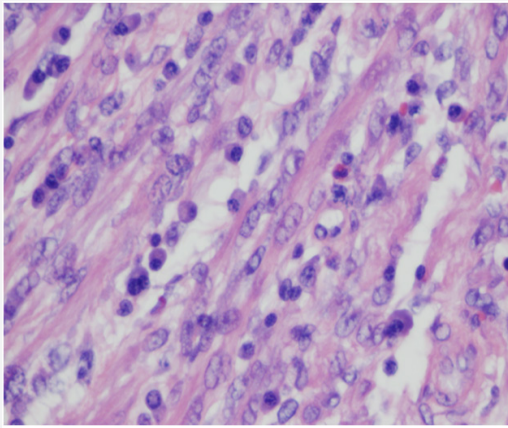


**Figure 3:** Gross findings of the fungating, friable mass found along the greater curvature of the stomach. No other solid mass or ulcer was found.

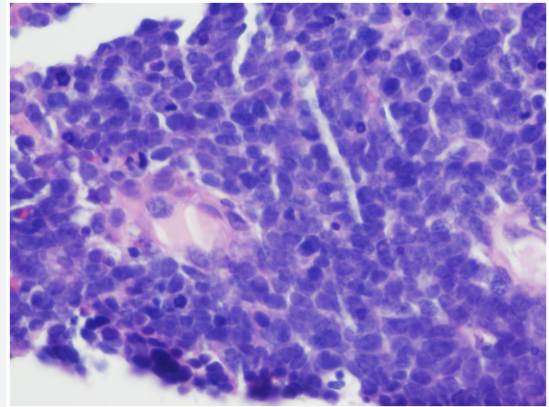


**Figure 4A:** Photomicrographs of primitive spindle mesenchymal sarcomatous component. Stains, original magnifications. H&E, X20

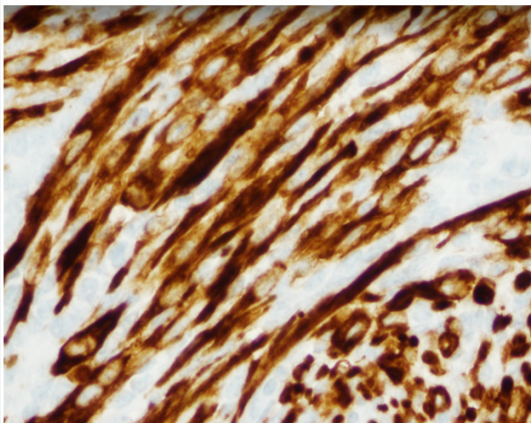




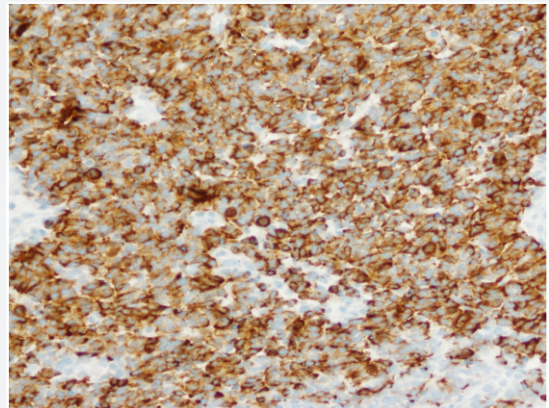
**Figure 4B:** Photomicrographs of primitive spindle mesenchymal sarcomatous component. Stains, original magnifications. H&E, X400



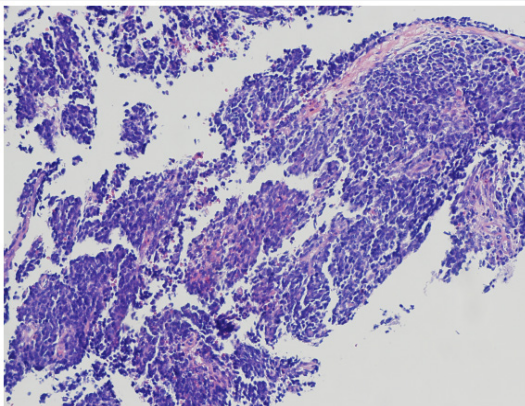
**Figure 5B:** Photomicrographs of primitive neuroectodermal tumor component. Stains, original magnification. H&E, X400



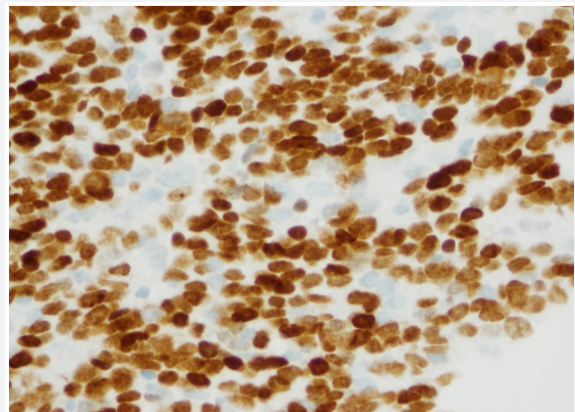
**Figure 4C:** Photomicrographs of primitive spindle mesenchymal sarcomatous component. Stains, original magnifications. Desmin, X400



**Figure 5C:** Photomicrographs of primitive neuroectodermal tumor component. Stains, original magnification. Neurofilament, X200



**Figure 5A:** Photomicrographs of primitive neuroectodermal tumor component. Stains, original magnification. H&E, X40



**Figure 5D:** Photomicrographs of primitive neuroectodermal tumor component. Stains, original magnification. SALL-4, X400

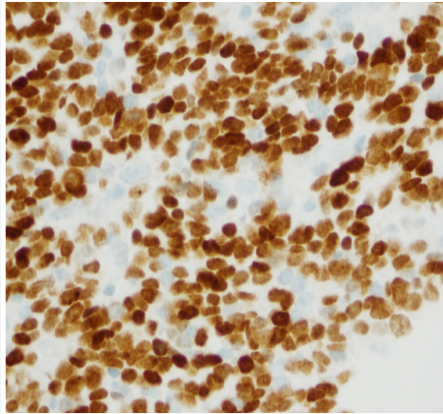


Figure 5E: Photomicrographs of primitive neuroectodermal tumor component. Stains, original magnification. Synaptophysin, X400

Discussion

Teratomas are neoplasms comprising all three germinal cell layers (ectoderm, endoderm, and mesoderm). Mature teratomas are composed of well-differentiated tissue whereas immature teratomas contain various immature fetal tissues. In the pediatric population, teratomas most commonly occur in gonads, the sacrococcygeal region, and mediastinum, and rarely in the retroperitoneum, cranium, and cervix [3,8,12]. Gastric teratomas, mostly mature, are exceedingly rare with just over 100 cases reported in the literature since 1922 [4,12]. Most of these cases were reported in infants and children, and the majority was male infants with less than 10 cases in females [5,8,12,13,14]. In adults, however, fewer than 10 cases have been reported for ages ranging from 20 to 83 years [7,8,11,16,17].

Table 1: Gastric Teratomas in Adults\*

Reference	Age (years)	Sex	Location	Tumor Size (cm)	Components	Follow-up
Matsukuma [8]	83	Male	Upper to middle portion on the lesser curvature	12x10 <sup>6</sup>	Fibroblastic mesenchymal cells, striated muscle cells, cartilaginous islands, neuroepithelial components, gliatissues, squamous cell nests, glandular components, and foci of adenocarcinoma; (composed of immature or mature elements from the mesoderm, endoderm and ectoderm)	Well after 1 year
Joo [14]	27	Male	Entire body along the greater curvature	9.5x7.5x5	Well-differentiated tissue: Fat, smooth muscle, cartilage, lymphoreticular, bone with marrow component, foregut mucosa, lung parenchyma, neuroglia; no immature tissue	Not available
Liu [7]	20	Male	Inferior wall of cardiac orifice	5.3x5x2.3	Cartilage, squamous cells, and respiratory epithelium; no immature tissue	Well after 10 months
Eustermann [13]	31	Not available	Posterior wall	7x6 x5	Not available	Not available
Fadeeva [25]	25	Not available	Anterior wall	4x3	Not available	Not available
Gray [11]	40	Not available	Lesser curvature	7x6	Not available	Not available
Gray [11]	23	Not available	Not available	Not available	Not available	Not available
Gray [11]	37	Not available	Not available	Not available	Not available	Not available

\*Modified from Liu et al. [7] "Primary gastric teratoma on the cardiac orifice in an adult"

The most common presentation is a palpable abdominal mass or abdominal distension during the first year of life [18]. Few cases can present with hemorrhage [19], melena [20], or rarely, perforation [22]. Most cases are benign, but malignant transformation (invasive adenocarcinoma), arising from an immature gastric teratoma, has been reported in an 83-year-old man [8]; however, sarcomatous transformation with RS and PNET, which was found in our case, has not been previously reported. The exact cause of gastric teratoma is not known; currently, it is hypothesized that these masses originate from migrated totipotential germ cells [4,14].

As in the present case, most gastric teratomas arise in the greater curvature or the posterior wall of the antrum and fundus [3]. Extra gastric growth occurs in the majority of cases whereas mixed extra gastric and endogastric growth is rare [3,12].

In this case, the immature teratoma consists of RS and PNET components without mature teratoma component or associated gastric carcinoma. Due to their rarity and various possible histologic

patterns, gastric teratomas may be misdiagnosed in adults. Differential diagnosis includes rhabdomyosarcoma, malignant small round cell tumor, or other spindle cell sarcomas.

CT and ultrasound findings typically show a heterogeneous mass with cystic and solid components as well as fat and calcifications [4,19]. The present mass showed soft tissue density without any fat or calcifications. Lack of the typical teratoma findings on CT led to different differential diagnosis including gastric carcinoma and gastrointestinal stromal tumor. The definitive diagnosis is only possible after taking into consideration the results from surgical excision with histological examination.

Complete surgical excision with tumor free margins and long-term follow-up are the standard management for teratomas [23]. Close follow-up with adjuvant chemotherapy has been recommended for patients with elevated AFP after surgery [6], whereas others recommend aggressive postoperative chemotherapy if there is histologic evidence of grade 3 immature teratoma or malignancy



[24,25]. Others suggest chemotherapy only after recurrence or metastasis is evident [15]. Monitoring serum AFP levels to detect recurrence has also been recommended [19]. The previous recommendations have been made for the pediatric population; however, no definitive recommendations are available for adults. In this case, serum biomarkers were not evaluated because teratoma was not suspected. After the diagnosis of immature teratoma was made, postoperative serum biomarkers (AFP and  $\beta$ -hCG) were measured and showed within normal limits. The serum biomarker evaluation during follow up may be potentially important to check tumor recurrence.

In summary, we report an unusual case of immature teratoma with RS and PNET components, which has not been previously reported in the literature.

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DOI: [10.19080/ARGH.2017.05.555663](https://doi.org/10.19080/ARGH.2017.05.555663)

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