Pediatric Gastric Teratoma

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ABSTRACT

Neoplasms from germ cell origin are a heterogeneous group of tumors rarely seen in the pediatric population, teratoma is the most frequent among them. They can occur in either gonadal or extragonadal locations. Extragonadal teratoma arising from abdominal viscer is very unusual. There are less than a hundred reported cases of gastric teratoma in the worldwide literature. Since the occurrence of this pathology in the pediatric age group is quite rare, we describe a case of a teratoma located in the lesser curvature of the stomach in an infant with an emphasis in radiologic-pathologic correlation.

CASE REPORT

This is a 6-month old boy, first born to healthy parents, uncomplicated pregnancy, born at term, vaginal delivery. An abdominal mass was discovered during a routine visit to pediatrician, and was sent to our hospital. The mass was located in both right upper and lower abdominal quadrants, it was firm, mobile, non tender and apparently not attached to deep structures. Remaining physical examination was normal. Patient was otherwise asymptomatic, with no vomiting and no bloody stools referred. Laboratory studies showed hemoglobin 9.7 g/dl (normal values 10.5-14 g/dl), hematocrit 30.7% (normal values 33-42%), mean corpuscular volume 69.3 fL (normal values: 70-74 fL), mean corpuscular hemoglobin 23 pg/cell (normal value >30 pg/cell), red cell distribution width 14% (Normal values: 11.5%-14.5%) and platelet count 625,000/mm³ (normal values: 150-350/mm³).

Abdominal plain film showed a large mass in the right hemiabdomen, extending to the left inferior quadrant with subtle calcifications with displacement of small bowel downwards and to the left (Figure 1). Upper gastrointestinal series demonstrated a fixed, large, rounded, well defined filling defect of approximately 5 cm of diameter at the antrum and confirmed the small bowel displacement (Figures 2 and 3). Abdominal computed tomography (CT) with intravenous contrast (iopromide, 300 mg of iodine/ml, 2 ml/kg) revealed a heterogeneous mass with fluid, soft tissue and fat attenuation mixed with gross calcifications arising from the stomach with a prominent exogastric component and displacing aside the small bowel and liver. Thick and regular septa with moderate post-contrast enhancement were present. There were no adenomegalies or apparent invasion to other structures (Figures 4 and 5).

Alpha-fetoprotein levels were reported within normal range for age. Endoscopy study showed a 4 cm-diameter exophytic lesion right next to the antrum in stomach lesser curvature. Exploratory laparotomy was performed and a pedunculated mass arising from the stomach anterior wall in the lesser curvature was completely resected with primary closure of the gastric defect (Figure 6). Pathologic examination of the tumor confirmed the diagnosis of a mature teratoma depending from gastric wall with extension to the mucosa, with less than 10% of immature neuroectodermal component (Figures 7 and 8). During the next month after the surgical procedure was done, patient had gastro-esophageal reflux symptoms which responded very well to
pharmacological treatment and this has been withdrawn 4 months after surgery with no symptoms recurrence. Patient has been followed for 36 months and he is asymptomatic with normal alpha-fetoprotein levels.

DISCUSSION

Germ cell neoplasms are a heterogeneous group of diverse histology: dysgerminoma, yolk sac tumours, embryonic carcinoma, polyembryoma, choriocarcinoma, teratoma, and mixed tumours. The incidence of germ cell tumours is 2.4 per million in children under 15 years of age, and they account for approximately 1% of childhood malignancies [1].

Teratoma is the most frequent tumor among germ cell neoplasms in children. It may be gonadal or extra-gonadal. Extragonadal teratomas are more frequently seen in newborns, infants and toddlers, whereas gonadal tumours occur more often in older children [2].

Most cases of extragonadal teratomas occur in the sacro-coccygeal region and mediastinum as well. Less than 1% are found in abdominal organs such as liver, kidney, vagina and stomach [3]. A case of gastric teratoma was first reported in the literature in 1922 by Eustermann and Sentry [1], and no more than 100 cases have been reported ever since [2]. Male predominance is notorious in children [3]; however, there are 6 reported cases in girls [4]. Most of the cases occur in children before one year of age, but older children are not infrequently seen [5]. In a report published by Curtis et.al. 21 patients were diagnosed with primary gastric tumor in a 54-year study performed in a single pediatric health institution. Teratoma was the second most frequent of this neoplasms, preceded by stromal tumors and with no deaths reported in a mean follow-up period of 22.3 months [6].

A variety of symptoms has been reported, such as upper digestive tract bleeding and gastric perforation, but the most frequent ones are vomiting, abdominal distention and abdominal mass [5]. Although these tumors occur more often in the greater curvature, as it was observed in 90% of the 44 patient series reported by Moriuchi [3], gastric teratomas can arise from any site of the gastric wall [4]. They can be pedunculated and have exogastric growths in up to 70% of cases [7].

Because of the infrequent occurrence of these tumors, clinical diagnosis becomes very difficult. Combination of abdominal mass with upper digestive tract bleeding should suggest the clinician the probability of this kind of tumor. Complementary studies must be made to confirm diagnosis, such studies include abdominal X-rays, upper gastrointestinal series, abdominal sonogram, and CT or magnetic resonance (MRI) scans. Calcifications inside the mass could be found in abdominal X-rays in up to 50% of patients with gastric teratomas, as it happened with our patient [8]. Sonogram will hardly demonstrate the origin of great size masses; however in most of the patients CT scans will be very useful in this matter. The presence of a well margined mass at the gastrointestinal tract which demonstrates ultrasound (US) and CT features related to cysts, associated with fat and calcifications should arise the suspicion of teratoma [9].

Differential diagnosis of cystic abdominal masses include teratoma, Wilms tumor, neuroblastoma, angiomylipoma, mesenteric lymphangioma, stromal tumors and congenital cysts, with the former three frequently associated with amorphous calcifications and only teratoma and angiomylipoma presenting with fatty components. Stromal tumors, the most frequent gastric neoplasms in children, are predominantly exogastric masses with a small intraluminal polyloid component, with heterogeneous and predominantly peripheral enhancement on CT, with cystic or hemorrhagic transformation, they are rarely associated with calcifications and, sometimes, present adjacent organ invasion, liver metastasis, ascites or omental spread [10]. Fat is not present in these tumors (table 1).

In the case we are reporting, teratoma arose from the stomach anterior wall in the lesser curvature with mainly exogastric growth. Clinical manifestations were abdominal mass and iron deficiency anemia attributed to upper tract digestive chronic blood loss.

Most of the gastric teratomas are benign in nature, as it happened with our patient, nevertheless there are reported cases of malignancy [11]. In these particular cases, surgery has been enough treatment, with no adjuvant chemotherapy or radiation therapy needed [12]. In most of reported cases, performing a partial gastrectomy seems to be enough treatment, however, in few patients total gastric resections were performed with good results [13]. The recommended treatment for both mature and immature teratomas is surgery and observation, with chemotherapy reserved for extragonadal malignant germ cell tumors. It is considered that incomplete resection is the main feature associated with recurrence [14].

Serial plasmatic alpha-fetoprotein levels are the main follow up parameter, but there is no standardized schedule, specially for extragonadal non-malignant germ cell tumors, whereas follow-up with this test is recommended for malignant lesions for at least three years [14]. Almost four years after surgery, this patient is asymptomatic with normal alpha-fetoprotein levels for his age (normal values: 27-397 ng/ml) [6].

Even though there are other reports of gastric teratomas in the literature, many of them present few images or lack surgical or histopathological correlation which we think is an interesting feature of this case.

TEACHING POINT

Abdominal cystic masses associated with fat or calcifications involve a wide differential diagnosis list including teratoma, Wilms tumor, neuroblastoma, angiomylipoma and stromal tumors, among others. Of these, teratoma is the only commonly presenting all of those three features; although rarely, it can arise primarily from gastrointestinal tract and it is usually associated with a good prognosis.
REFERENCES


Figure 1: 6-month old male with gastric teratoma. Abdominal plain film in anteroposterior upright position showing a large mass in the right upper and lower quadrants and left inferior quadrant with subtle calcifications (arrows) and bowel loops displacement. (Bottom right: magnification)
Figure 2: 6-month old male with gastric teratoma. Oblique projection of the stomach demonstrates a rounded filling defect, with irregular contours at the antrum. (Upper GI series, oblique view, single contrast, barium sulphate 60% weight/volume, 85 ml).

Figure 3: 6-month old male with gastric teratoma. Abdominal plain film showing the stomach filled with air and abnormally displaced, there is also abnormal displacement of small bowel loops filled with barium. (Upper GI series, antero-posterior recumbent projection, single contrast, barium sulphate 60% weight/volume, 85 ml).

Figure 4: 6-month old male with gastric teratoma. Axial CT with intravenous contrast of the abdomen demonstrated a large mass with soft tissue, fat, fluid and calcium densities arising from the stomach with a large exogastric component and displacing liver and small bowel aside. (16-MDCT Siemens Sensation scanner, protocol: 80 kVp, 50 mAs, slice thickness 3 mm, pitch 1.25; iopromide 300 mg of iodine /ml, 2 ml/kg).

Figure 5: 6-month old male with gastric teratoma. Coronal reformation of contrast enhanced CT showing the large exogastric component of a stomach depending mass with mixed density. (16-MDCT Siemens Sensation scanner. Protocol: 80 kVp, 50 mAs, slice thickness 3 mm, pitch 1.25; iopromide 300 mg of iodine/ml, 2 ml/kg).
Figure 6: 6-month old male with gastric teratoma. Perioperative photography demonstrating a large exogastric mass (M= Mass) depending from the lesser curvature (S=stomach).

Figure 7: 6-month old male with gastric teratoma. Histological section of the tumor demonstrates intestinal mucosa (arrow), epidermis (large arrowhead), keratin (asterisk) and brain fibrillar stroma (small arrowhead), findings consistent with components originated from the three germ layers. (stain: hematoxylin/eosin with 10X magnification)

Figure 8: 6-month old male with gastric teratoma. Histological section of the tumor demonstrates hyaline cartilage (asterisk) and bone (arrowhead), elements derived from mesoderm. (stain: hematoxylin/eosin with 20X magnification)
<table>
<thead>
<tr>
<th>GASTRIC TERATOMA</th>
<th>ABDOMINAL X-RAY</th>
<th>US</th>
<th>CT</th>
<th>MRI</th>
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<tbody>
<tr>
<td>Epi-meso gastric mass with calcific densities inside.</td>
<td>Cystic or solid mass with sharply marginated and smooth borders, and augmented in intensity through transmission. Cystic areas with inhomogeneous and moderate-intensity echoes separated by thick septations. Focal high-intensity echoes with acoustic shadowing, characteristic of calcification.</td>
<td>Well defined, mixed cystic and solid mass with calcifications and fat density.</td>
<td>The sebaceous component of has very high signal intensity on T1-weighted images, similar to retroperitoneal fat. The signal intensity of the sebaceous component on T2-weighted images is variable, usually approximating that of fat.</td>
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| WILMS TUMOR | Abdominal mass displacing bowel loops medially. Effacement and/or displacement of kidney shadow. | Large, lobulated, somewhat circumscribed mass, with heterogeneous echotexture but overall slightly hyperechoic. | Heterogeneous but predominantly solid mass, containing regions of low attenuation suggesting hemorrhage or necrosis. | Low signal intensity on T1-weighted images and high signal intensity on T2-weighted images. MR imaging also permits assessment of caval patency and multifocal disease. |

| ANGIOMYOLIPOMA | Heterogeneous mass with effacement of the psoas shadow. Effacement and/or displacement of kidney shadow. | Heterogeneous mass with highly echogenic nonshadowing foci, which correlate with the presence of fatty elements. | Renal mass with intramural variable amounts of fat present. Spontaneous hemorrhage presenting as irregular high density images in non-contrast scans is a common complication. | T1- weighted images: high signal intensity mass, hypointense in fat suppression sequences. T1- weighted images + Gd: Variable enhancement, depending on vascularity. |

Table 1: Differential diagnosis table for cystic abdominal masses [9, 11, 15-21]
### Table 1 (continued): Differential diagnosis table for cystic abdominal masses [9, 11, 15-21]

<table>
<thead>
<tr>
<th>MESENTERIC LYMPHANGIOMA</th>
<th>US</th>
<th>CT</th>
<th>MRI</th>
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<td>Usually bilateral elevation of diaphragm. Peripheral displacement of bowel loops.</td>
<td>Cystic, multiseptated masses with lobules that may be anechoic or contain internal echoes or sediment, with fluid-fluid levels caused by debris.</td>
<td>Cystic masses, with attenuation patterns ranging from fluid (if the contents are serous) to that of fat (if the contents are chylous). Uncommonly, cyst contents may be hemorrhagic and show high attenuation on non-enhanced CT scans. Proximal bowel dilatation, due to partial obstruction, may be present.</td>
<td>Serous contents appear hypointense on T1-weighted images and hyperintense on T2-weighted images. Cysts with hemorrhagic or fatty contents appear hyperintense on T1- and T2- weighted images.</td>
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<thead>
<tr>
<th>NEUROBLASTOMA</th>
<th>US</th>
<th>CT</th>
<th>MRI</th>
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<tr>
<td>Flank mass with stippled calcifications are present on up to 30%.</td>
<td>Inhomogeneously echogenic mass. In masses with fine calcifications, images show diffuse, increased echogenicity. Hemorrhagic or necrotic areas in the tumor appear as hypoechoic or anechoic areas.</td>
<td>The tumors often appear lobulated and typically have a heterogeneous appearance on contrast-enhanced CT. About 80-90% show stippled calcifications. Rarely, invasion into the lumen of blood vessels.</td>
<td>Typically hypointense on T1-weighted images and hyperintense on T2-weighted images. When contrast material is administered, the tumor exhibits inhomogeneous enhancement.</td>
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<tr>
<th>STROMAL TUMORS (GIST)</th>
<th>US</th>
<th>CT</th>
<th>MRI</th>
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<td>Elevation of the left hemidiaphragm and a soft-tissue mass indenting or displacing the gastric air shadow. Rarely, calcification may occur and be visible on abdominal radiographs.</td>
<td>Heterogeneous cystic or solid mass with sharply marginated borders.</td>
<td>Cavitary mass with extension into the gastrohepatic or gastrosplenic ligaments. Peripheral enhancement pattern. Lesions with extensive hemorrhage or necrosis. Calcification is an unusual feature of GISTs.</td>
<td>The degree of necrosis and hemorrhage greatly affects the signal-intensity pattern. The solid portions of tumor are typically low signal intensity on T1-weighted images, are high signal intensity on T2-weighted images, and enhance after administration of gadolinium. Areas of hemorrhage within the tumor will vary, depending on the age of the Hemorrhage.</td>
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</table>
ETIOLOGY | Teratomas are germ cell neoplasms which belong to a heterogeneous group of diverse histology.
---|---
INCIDENCE | 2.4 per million in children under 15 years of age, and they account for approximately 1% of childhood malignancies.
GENDER RATIO | Male predominance.
AGE PREDILECTION | Most cases occur in children before one year of age.
RISK FACTORS | Unknown
TREATMENT | In most of reported cases, performing a partial gastrectomy seems to be enough treatment. In few patients total gastric resections were performed with good results.
PROGNOSIS | Good prognosis, gastric teratomas are benign in nature.
FINDINGS ON IMAGING | Mixed cystic and solid mass with calcification inside and fatty component.

**Table 2:** Summary table for gastric teratoma

### ABBREVIATIONS

- **US:** Ultrasound
- **CT:** Computed tomography
- **MRI:** Magnetic Resonance Imaging
- **MDCT:** Multidetector Computed Tomography
- **Gd:** Gadolinium
- **GIST:** Gastrointestinal Stromal Tumor

### KEYWORDS

Teratoma, neoplasm, stomach, germ cell tumor