

Exogastric Mature Teratoma in an Infant: A Case Report¹

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Gastric teratomas are extremely rare neoplasms that have clinical characteristics such as a male predominance and a benign nature. We experienced a case of mature gastric teratoma occurring in a 12-month-old boy, who presented with an exogastric cystic mass, and this was confirmed by complete excision with subsequent pathologic examination. We report here on the radiologic and pathologic findings of an exogastric mature teratoma in an infant.

Index words : Stomach, neoplasms
Teratoma
Infants, gastrointestinal tract

Gastric teratoma is an extremely rare neoplasm. This tumor has clinical characteristics such as a male predominance and a benign nature. These characteristics of gastric teratoma are not shared with the teratomas found at other sites (1). To our knowledge, 102 cases of gastric teratomas have been reported worldwide (2, 3). In the most current literature, 2 cases occurring in a pediatric patient with an immature gastric teratoma have been reported (1, 4, 5). We report here on a case of exogastric mature teratoma that occurred in a 12-month-old boy.

Case Report

A 12-month-old boy was admitted because of a palpable mass and abdominal distension that he had experi-

enced for the previous 2 months. He did not have evidence of hematemesis, melena and fever. The abdominal radiograph showed a large soft tissue mass with irregular central calcifications; the mass was in the left upper quadrant of the abdomen, which displaced the stomach and bowel gas to the right side (Fig. 1A). US revealed a large, mixed-echoic, multi-septated cystic mass with central echogenic solid areas that showed acoustic shadowing (Fig. 1B). Contrast enhanced abdominal CT at the level of the stomach showed about a 16 × 12 × 10 cm heterogenous exogastric, multi-septated cystic mass with irregular shaped calcifications and fat components, and the mass arose from the anterior wall of the body portion of the stomach (Fig. 1C). The mass was in contact with abdominal wall anteriorly and it displaced the stomach to the posterior and right lateral side. The characteristic finding of the tethering point on the anterior gastric wall suggested the stalk of the mass. Our first impression was germ cell tumor such as gastric teratoma, and the differential diagnosis with a low probability was neuroblastoma or stromal tumor of the stomach with necrosis.

A pre-operative diagnosis of gastric teratoma was based on the presence of intratumoral calcification, fat

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and the mixed cystic and solid components of the mass.

Complete excision of the mass with partial wedge resection of the anterior wall of the stomach was done. On gross examination, the round exogastric mass, which had originated from the anterior wall of the gastric body, was $16 \times 12 \times 7$ cm in size. The tethering point of the stomach that was shown on the abdominal CT scan corresponded with the stalk of the mass. The cut section

of the tumor showed multi-septated cystic areas that were filled with serous and mucous material, keratin, hair, bone, brain and fat tissue (Fig. 1D). Histologically, the tumor contained chondroid tissue, squamous epithelium, neural tissue and keratin without any immature component. The pathologic diagnosis was mature teratoma of the stomach.

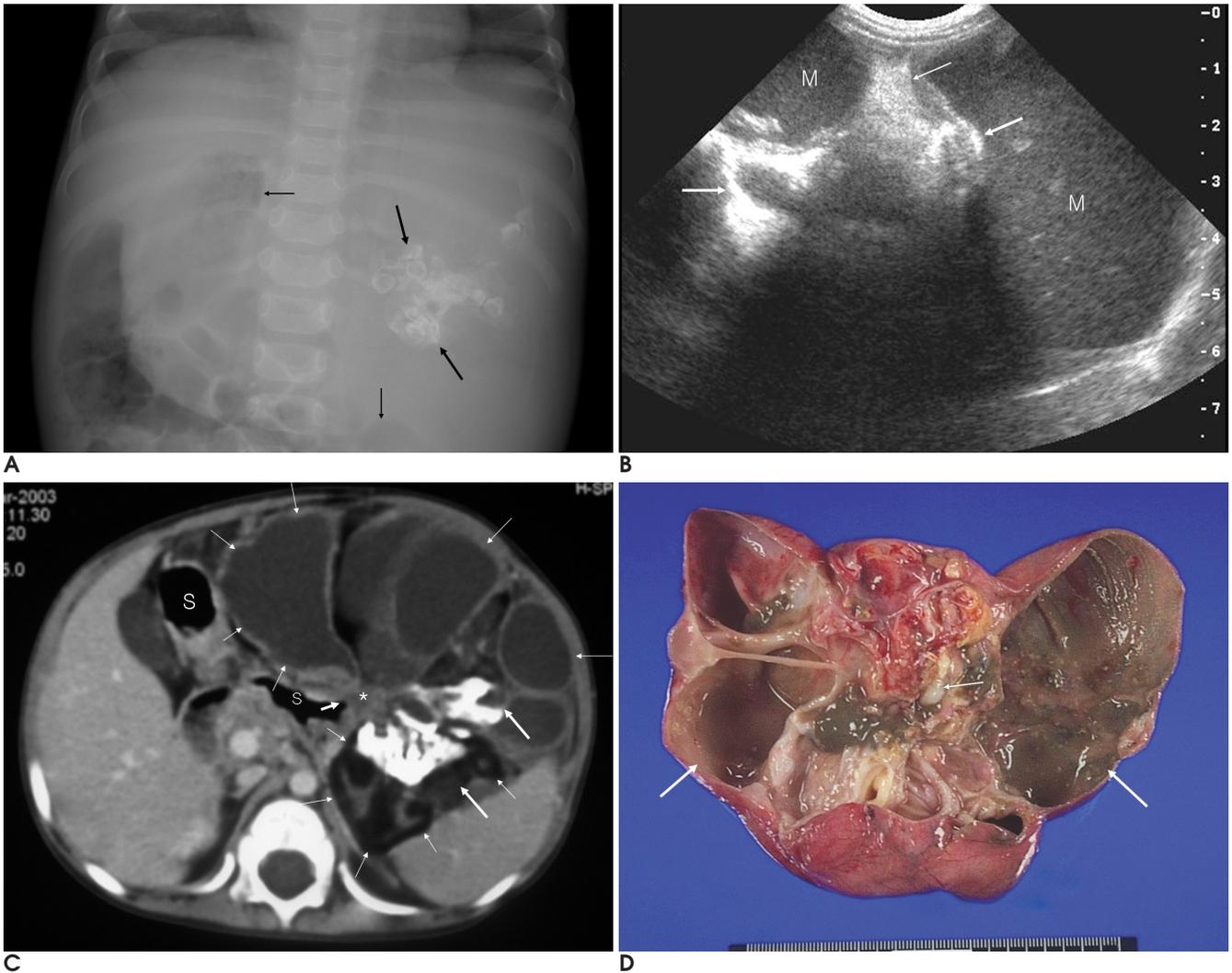


Fig. 1. Mature gastric teratoma in a 12-month-old boy.

A. Radiograph of the abdomen shows a large mass with ovoid and irregular shaped calcifications (large arrows) in the left upper quadrant, and it caused inferior displacement of the bowel loops and right displacement of the stomach (small arrows).

B. Longitudinal US of the abdomen shows a large, multi-septated cystic mass (M) in the left upper abdomen with echogenic solid (fat) portions (large arrow) and strong echogenic (calcifications) foci with acoustic shadows (small arrows).

C. Contrast enhanced abdominal CT at the level of the stomach shows about a $16 \times 12 \times 10$ cm heterogenous exogastric, multi-septated cystic mass (small thin arrows) arising from the anterior wall of the body portion of the stomach, along with irregular shaped calcifications and fat components (large arrows). The tethering point on the anterior gastric wall (small thick arrow) indicates the stalk of the mass (asterisk). The stomach (S) is displaced to the right side.

D. Cut section of the tumor shows multi-septated cystic areas (large arrows) filled with serous and mucous material, keratin, hair, bone (small arrow), brain and fat tissue.

Discussion

Teratomas may arise from the gonads or from extragonadal sites (2). There are various sites of origin for teratoma of infancy and childhood. The more common sites of origin are the sacrococcygeal area (60 - 65%) and the gonads (10 - 20%) and the relative uncommon sites are the mediastinum (5 - 10%), the presacral area (5%), the head, neck and the retroperitoneum (<5%) (3). The incidence of gastric teratoma in infancy and childhood is approximately 1% of all pediatric teratomas. Since the first case of gastric teratoma was reported in 1922, only 102 such cases have been reported in the literature (1 - 3). In the most current literature, 2 cases occurring in pediatric patients (a 3-month-old boy and a neonate male) with immature gastric teratomas have been reported (1, 4, 5), but ours is a case of exogastric mature teratoma that developed in an infant.

Although gastric teratomas can occur at any age, most cases were reported in infants or neonates (approximately 94%) (2, 6). The most common gender for all gastric teratomas is male (approximately 90%) (7). Almost all gastric teratomas have been benign, except for one case involving malignant transformation that was reported by Matsukama et al. (6). Where histological examination indicates that a tumor is immature, malignancy may or may not ensue, although neither metastasis nor recurrence has been reported.

Teratoma is different from dermoids in that teratoma shows derivatives of all three germ layers, and dermoids are neoplasms containing derivatives of only two germ layers (7). The etiology of gastric teratoma is not yet known. The accepted histogenesis of gastric teratoma involves germ cell theory: extragonadal teratomas originate from migrated totipotential germ cells (6).

The clinical symptoms of gastric teratoma are palpable mass (75% of reported cases) and/or abdominal distension (56%). It causes respiratory distress in infants, whereas it causes epigastric discomfort, hematemesis and/or melena in adults (2, 3). These symptoms were related to the large polypoid, exo/endogastric growth pattern and surface ulceration. The tumor can be classified by the growth pattern. The most common growth form is exogastric (65%), followed by exo/endogastric (26%) and endogastric (9%) (1). In our case, only an exogastric mass with symptoms of palpable mass and abdominal distension was disclosed.

Although the plain radiographic findings such as a soft

tissue mass with calcification are nonspecific, it can suggest the possibility of teratoma. US and CT are more specific tools that show not only a heterogeneous mass containing varying amounts of cystic and solid components, but they can also show fat and calcification, suggesting a diagnosis of gastric teratoma. In our case there was definite evidence of characteristic findings such as fat and calcification, and also the mixed cystic and solid components of the mass suggested teratoma. Tethering to the anterior wall of the stomach on CT was a helpful finding to evaluate the origin site of teratoma.

When imaging studies show a solid and cystic mass with calcifications in the left upper quadrant in an infant, neuroblastoma should be included in the differential diagnosis. Neuroblastoma is a malignant tumor of primitive neural crest cells that occurs more often in children under 2 years of age. The median age at diagnosis is 22 months, and more than 95% of cases are diagnosed by 10 years of age (8). This tumor shows as a heterogeneously echogenic mass in the suprarenal area with anechoic areas on US; a large heterogeneous solid mass with calcifications (about 80 - 90%) (9) and low attenuation areas of necrosis or hemorrhage are seen on CT (10). It frequently shows uncommon cystic change in neonates, but it tends to be solid even in the older child (11).

This gastric teratoma gave us several diagnostic clues that were different from neuroblastoma. It was an intraperitoneal mass that displaced the bowel loops and stomach posteriorly without vascular encasement, and it was a multi-septated cystic mass with irregular large calcifications and fat components. Based on the imaging findings, the possibility of other tumors such as Wilms' tumor, hepatoblastoma, rhabdomyosarcoma and liposarcoma was low (12).

In conclusion, gastric teratoma is an extremely rare, benign tumor in childhood. It most commonly occurs in male infants as a palpable abdominal mass. Radiologic evaluation can demonstrate the gastric origin of the mass and the characteristic findings such as fat, calcification and the mixed cystic and solid component of the mass, which are all helpful to exclude other masses in an infant from the differential diagnosis.

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