

A rare intra-abdominal mass in a newborn: gastric teratoma

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ABSTRACT

Gastric teratomas are rare intra-abdominal tumors. They are frequently seen in the neonatal period. A palpable mass is the most common clinical sign. Although radiologic imaging plays a role in diagnosis, in most cases the final diagnosis is made by histopathologic examination. In this report, we present a newborn male patient diagnosed with immature gastric teratoma. A male infant was referred to the clinic due to bloating and a palpable abdominal mass. Ultrasonography revealed a complex solid cystic mass in the abdominal and pelvic region. Magnetic resonance imaging was recommended. MRI revealed a multilocular, multiseptated, complex lesion. Given the radiologic findings, a gastric teratoma was suspected. The patient underwent surgery. On histopathologic examination, an immature grade 3 teratoma was diagnosed. Gastric teratomas are extremely rare tumors and must be considered in the differential diagnosis of abdominal masses during the neonatal and infant period.

Keywords: Teratoma, stomach, magnetic resonance imaging, neoplasm, pathology

INTRODUCTION

Gastric teratoma is an extremely rare tumor. The first case of gastric teratoma was reported by Eusterman and Sentry in 1922. About 100 cases have been described in the literature.¹ Most cases have been detected in the neonatal period. The majority of lesions are exogastric in location. Gastric teratomas are often of the mature type. It can sometimes be difficult to make a preoperative diagnosis.^{1,2}

This case report discusses the radiologic and pathologic findings of a patient diagnosed with a gastric teratoma due to an intra-abdominal complex cystic lesion in the neonatal period.

CASE

A male infant born with a birth weight of 2289 grams and a height of 47 cm to a 32-year-old mother at 34 weeks' gestation and 2 days' gestation had a postnatal APGAR score of 4-6. The patient was taken to the neonatal intensive care unit where cardiopulmonary resuscitation was performed as the peak heart rate was below 60/minute. Physical examination revealed a distended abdomen and a palpable abdominal mass, so an abdominal radiograph and ultrasonography (US) were performed. The abdominal radiograph showed soft tissue radiolucency in the left abdomen and displacement of

the bowel structures to the right (**Figure 1a**). The ultrasound examination revealed a multilocular, multiseptated, complex cystic solid mass that almost completely filled the abdominal and pelvic region. Abdominal magnetic resonance imaging (MRI) was performed to characterize the lesion. MRI showed a multilocular, multiseptate, expandable, complex mass that was hypointense on T1-weighted (T1W) images and hyperintense on T2-weighted (T2W) images. The mass filled the entire abdominal cavity and compressed the liver, spleen and intestinal structures. The stomach could not be distinguished. Diffusion of its solid components was limited on diffusion-weighted images. The septal structures showed contrast uptake in the T1W post-contrast series (**Figure 1b, 1c, 1d**). Given the imaging findings, a cystic teratoma was primarily considered as a differential diagnosis. Preoperative examination of the patient revealed thrombocytopenia, decreased urine output and an elevated D-dimer level. His alpha-fetoprotein level was within normal age limits. After preoperative treatment, the patient was taken for surgery. At laparotomy, the cystic component of the mass was initially noted. It was found that the integrity of the lesion was disturbed and bleeding occurred on the medial side of the cystic component. The spleen was connected to the upper left part of the lesion. It was observed that the mass was associated with the smaller curvature of the stomach.

On examination of the gastric fundus, a bony structure was palpated. The mass was explored and then a gastric repair and total splenectomy were performed.

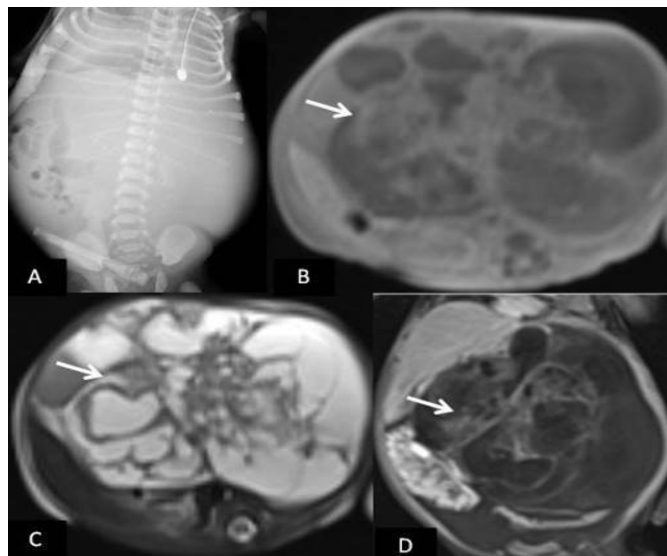


Figure 1. (A) A plain abdominal radiograph shows soft tissue radiolucency on the left side of the abdomen and a shift of the bowel structures to the right. Axial (B) T1W and (C) T2W MR images show a complex mass with a cystic solid structure located predominantly on the left side of the abdominal cavity. (D) A coronal post-contrast T1W MR image shows marked contrast enhancement of the solid components of the lesion

In the pathology report, the macroscopic examination of the material from the partial gastric resection showed a tumor measuring 10x9x6 cm and weighing 200 g, which was adjacent to the stomach wall and included the spleen on one side. The sections of the tumor consisted of patchy friable and hemorrhagic solid areas and cystic structures. Microscopic examination revealed mature elements of all three germ layers (fibrous tissue, cartilage, bone, squamous epithelium, lining epithelium of the airway and gastrointestinal tract, neuroglial tissue, choroid plexus) as well as a high proportion of immature neuroectodermal components and rosette formations (Figure 2, 3). The histopathology of the lesion was classified as an immature grade 3 teratoma. The patient died on the fifth day of his life due to respiratory distress. The patient's parents had given their written consent for this study.

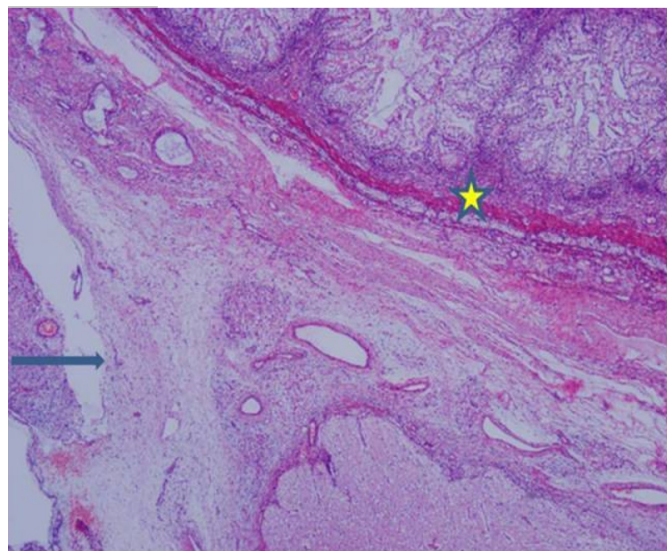


Figure 2. Microscopic view of the tumor in the submucosa of the stomach (H&E; X40) (Asterisk; gastric mucosa and arrow; gastric teratoma)

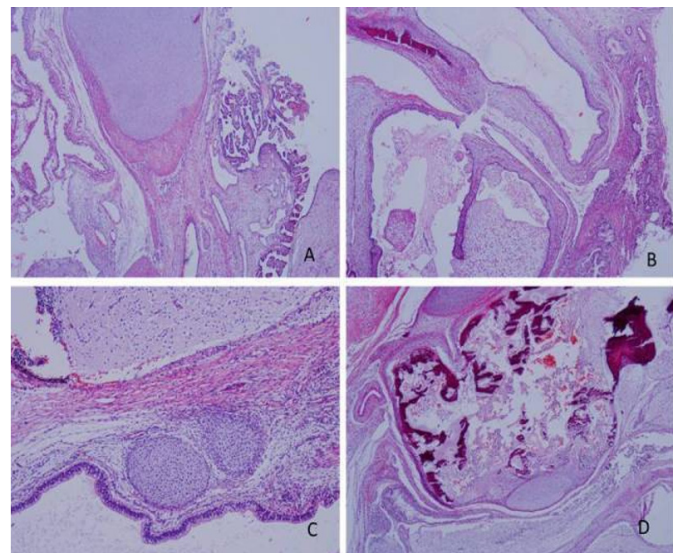


Figure 3. Microscopic images of mature elements from different areas: A: cartilage, epithelial elements and choroid plexus. B: Bone, cystic formations lined by squamous epithelium, the intestinal epithelium on the right side. C: Mature neuroglial tissue, cartilage, columnar ciliated epithelium underneath. D: Bone and bone marrow elements, cartilage and epithelial elements (A-D: H&E; X40).

DISCUSSION

Teratomas are tumors that arise from the three germ layers. They usually have a gonadal or extragonadal localization. Extragenital teratomas occur more frequently in newborns, infants and young children. Gastric teratomas are the most common gastrointestinal teratomas, although they account for only 1% of all body teratomas. Clinically, most patients complain of a palpable mass in the epigastrium and on the left side of the abdomen (75%), abdominal distension and vomiting. In some cases, however, other symptoms such as gastrointestinal bleeding or stomach perforation have also been reported.¹ Other rare findings include peritonitis due to gastric perforation and tumor rupture.² Gastric teratomas frequently contain cystic and solid components. The tumors are usually located exogastrically. The most common site is the posterior wall of the stomach, followed by the lesser curvature of the stomach and the anterior wall of the stomach.^{3,4}

Radiologic imaging studies often show a complex mass composed of solid and cystic components. Although half of all gastric teratomas show calcification on radiography, this is a nonspecific sign. US shows a heterogeneous mass with mixed echogenicity. Contrast-enhanced CT can be used to show the location, relationship to neighboring organs and vascular structures, and metastatic status of a mass lesion. While MRI shows a heterogeneous mass with cystic and solid components, it cannot adequately determine calcification. MRI can clearly determine the size of a tumor mass and differentiate its solid and cystic components.⁵⁻⁷

The most important indicator for the prognosis of teratomas is the histopathological grade of the tumor. Mature teratomas are considered benign and are classified as grade 0. Immature teratomas are classified according to the Norris classification system from grade 1 to grade 3, taking into account the content of immature tissue (mostly neural elements) and the degree of mitotic activity. Pure grade 1 and 2 teratomas have the potential to become malignant, while pure grade 3 teratomas have the potential to metastasize.^{8,9}

The differential diagnosis of gastric teratoma includes gastric stromal tumors, neuroblastoma, ganglioneuroblastoma, ganglioneuroma, mesoblastic nephroma, Wilms tumor, rhabdomyosarcoma, pancreatic cyst, hepatoblastoma, liposarcoma, meconium pseudocyst, and retroperitoneal teratoma. When solid, cystic and calcified intra-abdominal lesions are detected in the left upper quadrant of the abdomen in the pediatric age group, congenital neuroblastoma and gastric teratoma should be considered first in the differential diagnosis. Gastrointestinal stromal tumors are the most common stomach tumors in childhood. However, they are rare in newborns. They are usually characterized by cystic necrotic areas and rare calcifications and present as focal polypoid, primarily exogastric masses with intragastric extension but without intralesional fat.⁴⁻⁹ The association of gastric teratomas with Beckwith-Wiedemann syndrome and peritoneal gliomatosis has also been described in the literature.¹⁰

Complete surgical resection is sufficient for the treatment of mature teratomas and grade 1 immature teratomas. Complete resection followed by close surveillance and follow-up is recommended for grade 2 and 3 immature teratomas, while chemotherapy is indicated for recurrence.^{7,8,11} When a mature teratoma contains intestinal tissue or an immature teratoma is present, the serum alpha-fetoprotein level is abnormally elevated in an age-dependent manner. Alpha-fetoprotein and beta-chorionic gonadotropin levels aid in the detection of a tumor or its recurrence and malignant transformation during follow-up.^{8,9}

CONCLUSION

Gastric teratoma is a very rare tumor that occurs mainly in the neonatal and infant period. In our case, a large, complex cystic intra-abdominal mass was discovered in the neonatal period and diagnosed by advanced imaging techniques. A gastric teratoma should always be considered in the differential diagnosis when a large intra-abdominal mass is discovered in the neonatal period. Early diagnosis and surgical removal are the mainstays of treatment, although immature teratomas may require more aggressive follow-up and additional oncologic treatments.

ETHICAL DECLARATIONS

Informed Consent

The patient's parents signed a free and informed consent form.

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

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Author Contributions

All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

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