



Immature Gastric Teratoma in a Newborn: A Case Report

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ABSTRACT

A teratoma is the most common germ cell tumor in children; however, a gastric teratoma is very rare. An immature gastric teratoma has malignant potential; therefore, it should be removed surgically and followed up routinely to assess for recurrence by performing imaging studies and estimating serum alpha-fetoprotein (AFP) level. We describe the case of a 2-day-old male neonate with abdominal distension and a palpable mass. He underwent surgical resection of a tumor that was diagnosed as an immature gastric teratoma.

Key Words: Immature teratoma, Stomach neoplasm, Newborn

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INTRODUCTION

Gastric teratoma is a very rare tumor, accounting for <1% of all teratomas diagnosed in children¹⁾. Immature gastric teratomas are even rarer²⁾. Immature gastric teratomas have malignant potential; therefore, it should be removed surgically and followed up routinely. This allows for proper assessment for recurrence by performing imaging studies and estimation of serum alpha-fetoprotein (AFP) level.

We report a 2-day-old male neonate who had no abnormal results on serial antenatal sonography; however, after the birth, he presented with respiratory distress, abdominal distension, and a huge sized intra-abdominal mass diagnosed as immature gastric teratoma, a extremely rare germ cell tumor.

CASE REPORT

A 2-day-old male was transferred to a neonatal intensive care unit at Soonchunhyang University Bucheon Hospital due to tachypnea, cyanosis, and abdominal distension.

The patient was delivered by cesarean section at a gestational age of 41⁺³ weeks due to late term pregnancy and failure to induce labor. His Apgar score was 8 at 1 min and 9 at 5 min. His mother was admitted to another hospital for 1 week because of decreased fetal activity and high serum C-reactive protein (hs-CRP) levels. She had no significant delivery problems

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or past medical history. Serial antenatal fetal ultrasonography was performed, and there were no abnormal findings. At the time of delivery, meconium staining was observed. At 2h postpartum, the neonate's respiration rate increased despite receiving oxygen.

After the transfer to our neonatal intensive care unit on a day after birth, the patient's clinical parameters were as follows: systolic blood pressure (BP), 76 mmHg; diastolic BP, 54 mmHg; mean BP, 68 mmHg; pulse rate, 146 beats/min; and respiratory rate, 56 breaths/min. He exhibited chest wall retractions, desa-

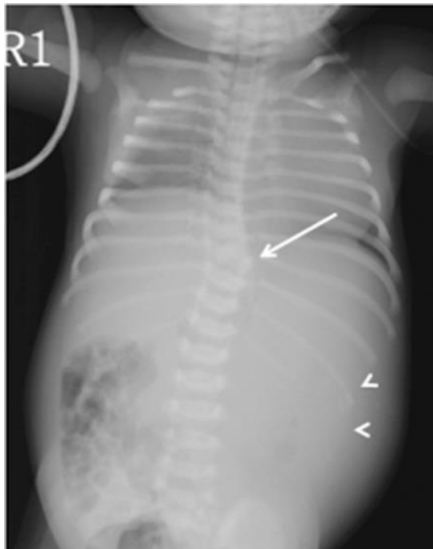


Figure 1. A plain radiograph showing a huge soft tissue mass with internal calcifications (arrowheads) in the left abdomen. Bowel loops are displaced to the right. Hemivertebra of T9 (arrow) is also seen.

uration, and bradycardia due to apnea; a palpable solid mass approximately $10 \times 10 \text{ cm}^2$ in size was found in the left abdomen.

His leukocyte count was 14,800 cells/ μL , hemoglobin was 11.9 g/dL, and hematocrit was 34.9%. His hs-CRP and procalcitonin levels were mildly elevated (0.89 mg/dL and 1.8 ng/mL, respectively); therefore, prophylactic antibiotic treatment (ampicillin+gentamycin) was initiated. A plain radiograph revealed a huge soft tissue mass with internal calcifications (Figure 1).

Abdominal ultrasonography was performed, and a pre-diagnosis of either neuroblastoma (based on the ill-defined left adrenal branch) or teratoma (based on the calcifications) was made (Figure 2). Exploratory laparotomy and excision of the mass were performed on the first day of the patient's transfer. After the incision, surgeons found a massive hematoma in the intra-abdominal cavity. After removing the hematoma, the surgeons found a $10 \times 8 \times 7 \text{ cm}$ -sized, well-defined, movable mass originating from the stomach. The mass seemed to invade the antrum extensively or originate from the antrum. Surgeons performed distal gastrectomy and gastroduodenostomy for radical resection of the mass. There were no anomalies of the other organs (Figure 3). Owing to its gastric origin, neuroblastoma was ruled out. After pathological evaluation, the final diagnosis was immature gastric teratoma, grade 3 (margin free from tumor; immunochemistry: positive glial fibrillary acidic protein, S-100, glypican-3, and AFP tests).

Postoperative laboratory analysis revealed elevated CRP and procalcitonin levels (6.64 mg/dL and 2.3 ng/mL, respectively). Seven days after the operation, histological findings were

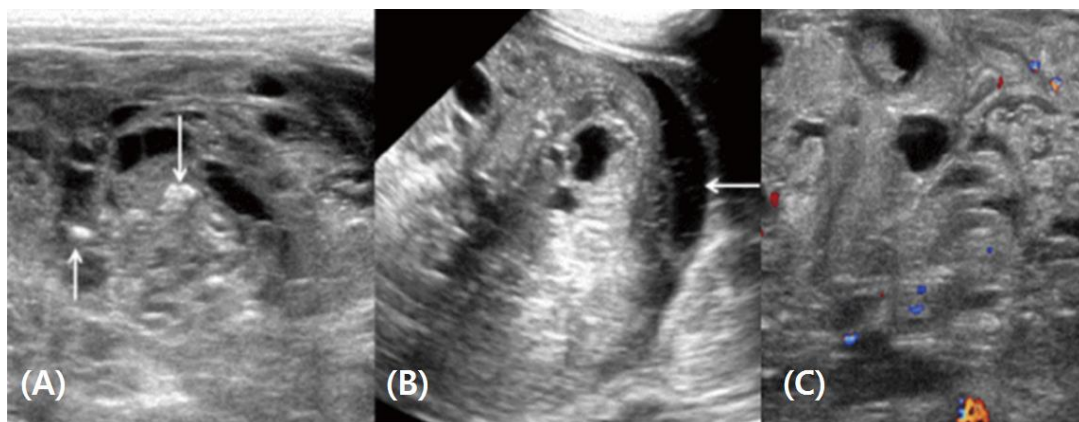


Figure 2. Abdominal ultrasonography: A huge heterogeneous echogenic solid mass extending and crossing the midline in the left abdomen can be seen. (A) The mass exhibits considerable echogenic calcification (arrows), (B) cystic changes (arrow), and (C) increased vascularity.



Figure 3. The well-defined movable mass originated in the distal stomach. The mass was approximately 10×8×7 cm in size. Distal gastrectomy and gastroduodenostomy were performed with radical resection of the mass.

confirmed; his AFP and beta-human chorionic gonadotropin (β -HCG) levels were 17,111 ng/mL and 3.4 mIU/mL, respectively. Five days later, his AFP level was 5,580 ng/mL and β -HCG was 0.6 mIU/mL. Six days later, his AFP level had decreased to 2,078 ng/mL.

After the surgery, the patient had multiple seizure events. The events occurred at 1, 4 and 5 days after operation, and lasted within 5 min. We supposed that he could have hypoxic damage because of meconium aspiration. Therefore, antiepileptic drug treatment (phenobarbital and levetiracetam) was initiated. Brain magnetic resonance imaging (MRI) performed before discharge revealed a possible hypoxic brain injury. An electroencephalogram (EEG) showed a mild occipital multifocal negative sharp wave. Antiepileptic drug administration was continued.

After the surgical site was healed, abdominal magnetic resonance imaging (MRI) was performed. A residual mass-like lesion was observed. Therefore, positron emission tomography-computed tomography (PET-CT) was planned. The patient was discharged, and follow-up evaluations were conducted on an outpatient basis. However, after discharge, his parents wanted to visit another hospital; thus, the follow up study could not be performed.

DISCUSSION

Teratoma is the most common germ cell tumor in children. It commonly arises in the sacrococcygeal (60–65%), gonadal (10–20%), mediastinal (5–10%), and presacral (5%) regions³. Gastric teratomas occur very rarely (<1%)³. Teratomas are more common in boys; the reason for this is unknown⁴.

Most cases are diagnosed based on abdominal distension and the presence of a mass. Occasionally, gastric teratomas are diagnosed owing to respiratory difficulty caused by the mass. Patients present with vomiting, hematemesis, or melena due to mucosal ulceration⁵. In our case, despite the large size of the mass, it was not detected by antenatal fetal sonography, and the patient had respiratory distress with abdominal distension.

Gastric teratomas can arise at any site in the stomach, and most commonly occur in the lesser curvature of the antrum and fundus. Exogastric growth is observed in approximately 60% of cases, and growth into the stomach is observed in 30% of cases. Mixed types are rarely observed⁵. Spontaneous rupture and perforation of a large gastric teratoma have also been reported⁶. Gastric teratomas must be differentiated from neuroblastoma, Wilms' tumor, hepatoblastoma, rhabdomyosarcoma, liposarcoma, and retroperitoneal teratoma⁷. Histologically, the presence of neuroglial tissue can be used to distinguish between mature and immature teratomas. The management of gastric teratoma involves surgical excision of the tumor and the site of attachment to the stomach. Simple excision of gastric teratomas followed by reconstructive gastric surgery is the recommended treatment⁸. This management strategy is usually curative, and partial or total gastrectomy is required only in rare cases and is dependent on the extent of the tumor growth⁹. Studies have shown that complete excision of the mass results in a good prognosis and recurrence-free survival without chemotherapy, or radiotherapy³. Our patient had a large, grade 3 immature gastric teratoma following total excision, and the patient did not undergo chemotherapy or radiotherapy.

It appears that the main risk factor for relapse or regrowth is an incomplete resection of immature teratomas, which may occur in cases of more aggressive or infiltrative tumor growth¹⁰. Further, malignant transformation can occur in the case of immature teratomas, and patients should be closely followed up. Serum AFP is a useful marker of recurrence or the presence of a residual tumor⁶.

In conclusion, gastric teratoma is an extremely rare pediatric tumor and is benign in most cases. It is more likely to occur in males and is diagnosed based on the findings of a palpable abdominal mass. However, it can present with respiratory distress like tachypnea or apnea with mild abdomen distension. Therefore, neonatologist's should examine the baby more carefully after birth, even if the antenatal ultrasonography results were negative. If suspected, an imaging study should be performed. We report this rare case to guide diagnosis of a newborn germ cell tumor and to remind differential diagnosis of abdominal distension in newborns.

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