Rare Case of Immature Gastric Teratoma

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Introduction: Teratomas are rare and complex tumors with components from more than one of the germ cell layers. Teratomas range from benign, well-differentiated (mature) cystic lesions to those that are solid and malignant (immature). The incidence of all teratomas is estimated at 1:10,000–1:20,000 newborns. Gastric teratomas represent only 1-2% of all teratomas.

Case presentation: We report a case of gastric teratoma of a 2 month-old boy who presented with abdominal distension. Diagnosis was established by physical examination, ultrasonography and computed tomography. The tumor measured 13/10/5.5 cm and weighed 390 grams and was surgically excised. Histological examination revealed an immature gastric teratoma. We also reviewed existing clinical and genetic data on gastric teratomas. Association of gastric teratomas with other congenital anomalies or tumors is very rare. Reported cases include: Beckwith-Wiedemann syndrome (involved:11p15, IGF2), Hodgkin disease (developed 3 yrs. post-resection) and focal neuroblastoma. Recent theories include extraembryonic cells; also have been hypothesized to originate from pluripotent cells present in the gastric wall.

Conclusion: Gastric teratomas are extremely rare tumors. Complete resection induce a good outcome.

Keywords: teratomas, genes, embryonic tumors, gastric tumors

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circumferential gastrectomy. A 3.5 cm gastric incision was subsequently closed by transverse gastrotomy.

The macroscopic pathologic findings were: multinodular solid and cystic areas; non-continuous tumor capsule (Figure 6). Tumor sections revealed many serous and serosanguineous cysts; the solid areas had a white-yellow color, containing regions of calcification and bone formation and many small cysts with transparent walls; separately a nodular elastic process was identified, 2.1/1.5/0.9 cm in dimension, with cysts within solid area.

Microscopic pathologic findings were:
– Solid and cystic areas of proliferative tissues derived from all three germ layers were identified: squamous, intestinal and respiratory epithelium, glandular structures, pilous follicles, muscle, cartilage, bony and adipose tissue, dental and nervous tissue, choroid plexus, mesenchymal tissue and ganglionar nervous cells.
– Neuroectodermal areas were localized as rosettes, neural tubes or glial areas (immature nervous tissue fields/slide).
– Some cysts had papillary projections covered by clear cells or cylindric epithelium. There were small regions containing clear cytoplasm cells forming pseudoglands or trabeculae.
– The specimen contained gastric wall with submucosal hemorrhagic foci covering diverse tissues proliferation (epithelium, cartilage, bone, nervous tissue) similar with the tissue found within the tumor.
- The histopathological specimen was consistent with a 3rd grade, immature gastric teratoma (Figure 7).

**Evolution.** The patient presented a good post-operative course with normal intestinal transit time and the surgical wound cured per-primam. Clinical follow-up at 1 month, 6 months and 1 year revealed good progress with normal somatic development and normal intestinal transit time.

**Discussion**

Teratomas are rare embryonic tumors with components from all three germinal layers. (1). The first case was reported by Eustermann and Sentry in 1922. Teratomas range from benign, well-differentiated (mature) cystic lesions to those that are solid and malignant (immature). Immature teratomas have varying degrees of immature fetal tissues.
The malignant type contains at least one of the malignant germ cell elements. Immature teratomas are graded from 1 to 3 by the amount of immature tissue contents (neural elements) and by the degree of mitotic activity. The incidence of all teratomas is estimated at 1:10,000–1:20,000 newborns.

Gastric teratoma is an extremely rare location: only 1-2% of all teratomas, i.e. less than 1:1,000,000 newborns. Little more than 100 cases are reported in the literature, out of which only about 10% are of immature type. More than 90% of all reported cases are males. Clinical presentation generally occurs within the first 3 months of life but there are few reports of delayed diagnosis in older children and in adolescents [2]. We only found six published cases of gastric teratoma in adults. The most common manifestation of gastric teratomas is abdominal distension, vomiting and an abdominal mass palpated on exam; other described associated signs: gastrointestinal bleeding (hematemesis or melena), constipation, dyspnea, anemia.

Diagnosis of gastric teratoma is made by US and/or CT. Teratomas have a characteristic heterogeneous appearance, including cystic areas with solid components and calcifications. Prenatal diagnosis can be made by fetal US and maternal serum AFP.

Differential diagnosis include: neuroblastoma, Wilms tumor, hepatoblastoma, liposarcoma; lipoblastoma, rhabdomyosarcoma, retroperitoneal teratoma.

Treatment usually consists in: surgical resection of the entire tumor, or partial gastrectomy may be necessary, as described in our case. Recurrence of tumor is not described after complete resection, and adjuvant therapy (e.g., chemotherapy or radiotherapy) is not needed.

Possible complications and prognosis: Gastric teratomas are usually benign tumors and malignancy is extremely rare (only three cases reported) [1,3,4]. Elevated serum AFP levels may be the only alerting sign of the presence of a malignant teratoma. Correct interpretation of serum AFP levels in infants is difficult because of wide physiologic variation. Acute severe or prolonged bleeding is uncommon but a serious complication. However, in our case, post-operative prognosis was excellent.

Genetical aspects: Association of gastric teratomas with other congenital anomalies or tumors is very rare. Reported single cases include: Beckwith-Wiedemann syndrome [5], Hodgkin disease (developed 3 years post-resection) [6] and focal neuroblastoma [7]. No familial cases are reported. Recent theories of teratoma origin include extraembryonic cells other than embryonic stem cells, conjoined and maldeveloped twins, and undetermined cell types. Gastric teratomas also have been hypothesized to originate from pluripotent cells in the gastric wall.

Conclusions
Gastric teratoma is an extremely rare tumor (only 100 reported cases) benign tumor of childhood and the immature type is more rare (not more that 10 cases). This is the first case reported in Romania. The most common physical sign of gastric teratoma is abdominal distension and palpable abdominal mass and abdominal CT is the elective imaging modality of diagnosis. The mature or immature type is defined by histological exam. After complete resection, a good evolution is expected in all cases.

References