Case Report

Immature gastric teratoma in an infant: A case report

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ABSTRACT

Immature gastric teratoma is an extremely rare embryonal neoplasm. Although teratomas have been identified in most body regions, the sacrococcygeal is the most commonly involved site, followed by cervicofacial, mediastinal, gonadal, and retroperitoneal regions. The stomach is rarely involved, accounting for <1% of teratomas. Most of the cases occur in infants. The rarity of this disease in unusual sites makes the diagnosis difficult. We report an unusual case of an immature gastric teratoma in an infant who clinically presented with upper respiratory tract infection. Contrast-enhanced computed tomography abdominal was performed which revealed a large exogastric mass. Histopathological examination confirmed it to be an immature gastric teratoma. This has an excellent prognosis if treated promptly. Complete surgical excision is usually curative. Hence, awareness of this entity in unusual sites like the stomach is required to make the right diagnosis. Until date, only 35 cases of immature gastric teratomas have been reported in the published literature. We, therefore, report one such case in the stomach of a neonate.

Key words: Gastric teratoma, Immature, Infant

Teratoma is defined as a germ cell tumor composed of tissues derived from the ectoderm, endoderm, and mesoderm in various locations, including the gonads, intracranium, anterior mediastinum, retroperitoneum, and sacrococcygeal. Stomach is the rarest site of extragonadal teratoma and comprises <1% of all teratomas diagnosed worldwide [1,2]. Gastric teratoma was first reported by Eusterman and Sentry in 1922 [3]. Gastric teratomas are benign in nature. In infants, immature teratomas have a good prognosis. The literature review revealed only around 100 cases of gastric teratoma [1]. Immature gastric teratoma is even rarer and only 35 cases have been reported in the literature. All the cases reported are single case reports [1,2,4-6].

Herein, we report the case of a 27-day-old male baby who presented with a mass in the left hypochondrium, which was diagnosed as an immature gastric teratoma on the basis of histological features.

CASE REPORT

A 27-day-old boy presented to the outpatient department with a complaint of upper respiratory tract infection. On examination, an incidental mass was detected in the left hypochondrium. Contrast-enhanced abdominal computed tomography revealed a large exogastric, solid, and cystic mass arising from the greater curvature of the stomach (Fig. 1.1). The tumor was completely excised.

Operative findings showed that the tumor (Fig. 1.2) was arising from the posterior wall of the stomach with around 20% intragastric component and the rest in the lesser sac displacing the pancreas, spleen, and extending up to the diaphragm with no obvious infiltration. The tumor was carefully dissected, the posterior wall of the stomach was resected with a clear possible margin of more than a centimeter from the visible tumor and the stomach was reconstructed.

Grossly, the tumor measured 7.6 cm × 6.2 cm × 3.7 cm. Cut surface of the tumor was solid and cystic with cysts ranging in diameter from 0.1 cm × 0.1 cm to 2.1 cm × 1 cm, filled with gelatinous material. Cut surface of the solid area was gray-white, glistening with focal gritty areas.

Histopathological examination showed a tumor composed of elements from all three germ layers. The cysts were seen lined by respiratory (Fig. 1.3) and intestinal epithelium (Fig. 2.2), both being of endodermal origin. Focally, squamous lining with dermal adnexa was seen, being of ectodermal origin. Foci of mature bone (Fig. 1.4) and cartilage (Fig. 1.5) were seen along with adipose tissue (Fig. 1.4) and smooth muscle (Fig. 2.1) of mesodermal origin. Mature glial tissue (Fig. 2.3) was seen in a few areas being composed of neutrophil along with immature neural elements forming rosettes (Fig. 2.4) and lining tubules at places. The immature cells were round to ovoid with scanty cytoplasm and

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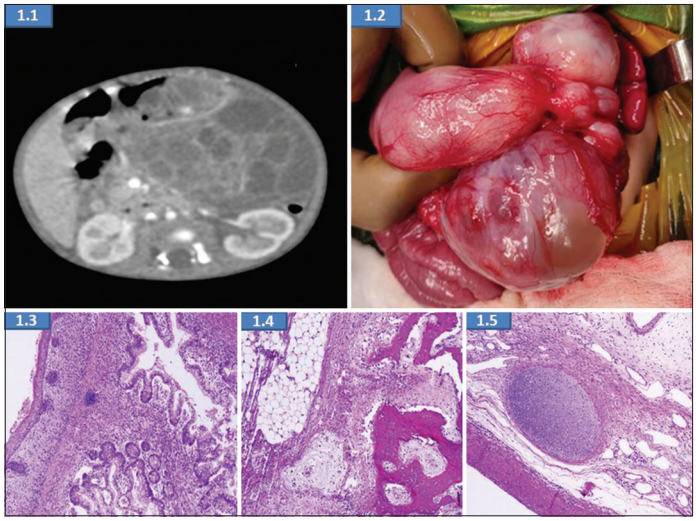

Figure 1: (1.1) Contrast-enhanced computed tomography shows solid and cystic exogastric mass. (1.2) Intraoperatively, an exogastric solid and cystic mass arising from the greater curvature of the stomach. Histopathological examination showing (1.3, H and E, 10× low power) endodermal component: Respiratory epithelium, (1.4, H and E, 10× low power) mesodermal component: Adipose tissue, bony trabeculae, and (1.5, H and E, 10× low power) cartilage

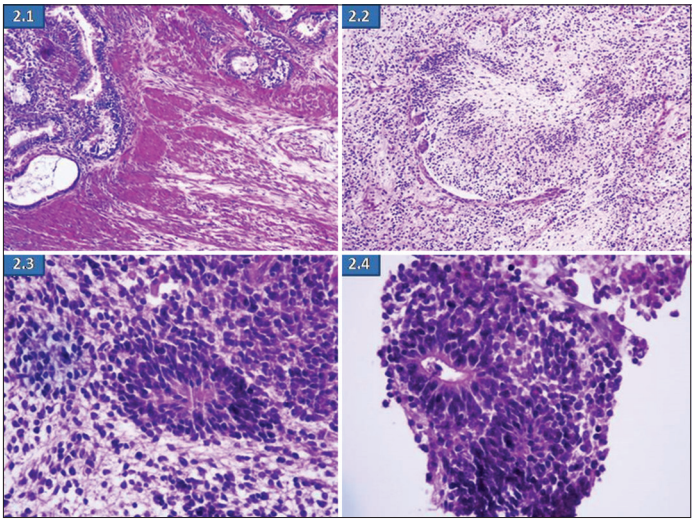

Figure 2: Histopathological examination showing (2.1, H and E, 10× low power) mesodermal component: Muscle spindles, (2.2, H and E, 10× low power) endodermal component: Colonic mucosal lining, (2.3, H and E, 20× high power) ectodermal component: Mature neural element, and (2.4, H and E, 20× high power) immature neural element: Rosette

had hyperchromatic nuclei with occasional mitoses. With these features, a diagnosis of an immature gastric teratoma was made.

The infant made an uneventful recovery but presented 5 months later with a complaint of post-operative adhesive intestinal obstruction which was managed by laparotomy and adhesiolysis. He is on regular follow-up with frequent ultrasounds and serum alpha-fetoprotein level monitoring. The patient has normal growth and development.

DISCUSSION

Teratoma is the most frequent tumor among germ cell neoplasms in children. In infancy and childhood, the most common site of teratomas are the sacrococcygeal region (60–65%), gonadal (10–20%), mediastinal (5–10%), presacral (5%), and rarely intracranial, retroperitoneal, and cervical [7]. Although gastric teratomas can occur at any age, approximately 94% of reported cases have involved infants or neonates [8,9]. There is a striking male predominance of gastric teratoma with only 6.7% of cases occurring in females [10,11].

In most of the cases, the chief complaints are abdominal distension and a palpable lump, but sometimes respiratory difficulty can be caused by upward displacement of the diaphragm by the tumor especially when large [12], as in this case.

Gastric teratomas are usually located on the posterior wall or greater curvature of the stomach [1]. The majority of gastric teratomas are exogastric masses, representing approximately 60% of the cases, while endogastric growths are present in about 30% of the cases [13]. Mixed exogastric and endogastric growths are rare. In our case, around 20% of the tumor was endogastric.

Differential diagnosis of these abdominal lesions in the pediatric age group includes neuroblastoma, Wilms tumor, hepatoblastoma, rhabdomyosarcoma, liposarcoma, and retroperitoneal teratoma [13]. While most of these are round cell tumors, the presence of a combination of ectodermal, endodermal, and mesodermal components, both mature and immature help, concludes a diagnosis of teratoma. Wherever required, specific immunohistochemical stains could be used to confirm the diagnosis in correlation with the clinical findings [14].

Histopathologically, gastric teratomas can be mature or immature. The diagnosis of mature versus immature teratoma is based on the presence of immature tissue, especially immature neural tissue as in other sites. Mature gastric teratomas can contain mature glial tissue along with other derivatives of all three germinal layers. Immature teratoma is diagnosed if immature tissue, especially immature neuroectodermal tissue, is found forming tubules and rosettes as seen in this case [15]. Unlike ovarian teratomas which have an established grading system based on the extent of immature neuroectodermal tissue (Norris grading system), a similar grading system has not yet been described in the gastric location. The presence of immature elements, however, does not have a bearing on the prognosis. Rarely, these tumors may be associated with elevated serum alpha-fetoprotein levels indicative of an associated yolk sac component [16]. However, this may not be demonstrable even after extensive sampling of the tumor.

Surgical excision is curative in gastric teratomas. Follow-up in these cases consists of regular observation and serum alpha-fetoprotein measurement to monitor for recurrence or malignant transformation. In the unusual case of rising alpha-fetoprotein level after surgical resection, chemotherapy is recommended [1]. Prognosis is excellent with complete excision and primary closure of the gastric wall defect. Recurrence of the tumor is rarely seen after complete resection. Thus, adjuvant chemotherapy or radiotherapy is not routinely recommended [14].

CONCLUSION

Immature gastric teratoma is an extremely rare tumor of childhood, and almost all cases are benign. Pre-operative diagnosis may

be challenging, because of its rare location. The presence of immature neuroectodermal elements on histopathology, confers the diagnosis of immature teratoma, however, does not impact the prognosis if excised completely. With very few cases reported previously worldwide, we report a case of immature gastric teratoma in a 27-day-old neonate.

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