ORIGINAL ARTICLE

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Gastric teratoma in children

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Abstract Gastric teratoma (GT) comprises less than 1% of all teratomas in children. Though GT in the presence of immature neuroepithelial elements is regarded as malignant, the prognosis is excellent after complete excision of the tumor. Because of its rarity the world literature lacks a large study. Clinical experience with ten cases of GT is presented and discussed. Only one patient was female; the mean age at presentation was 3.2 months. Two cases were immature grade III GT; one of these had infiltrated the left lobe of the liver and the transverse colon while the other had metastasized to the regional lymph nodes and omentum. All the patients underwent complete excision. There were no deaths, and after a mean follow-up period of 4.2 years, all the patients had no recurrence and were healthy. Both the mature and immature types of GT have an excellent prognosis after complete excision of the tumor. Even when the immature type infiltrates surrounding structures, complete excision offers recurrence-free survival without requiring chemotherapy.

Key words Teratoma · Stomach · Child

Introduction

Teratomas are embryonal neoplasms that contain tissue from all three germ layers, namely, ectoderm, endoderm, and mesoderm and are derived from totipotent cells. The majority of the patients present during infancy and early childhood and the tumors may be benign or malignant. The nomenclature is somewhat misleading in that immature teratomas are frequently labeled as malignant, but in childhood they do not behave in a really malignant fashion unless they have foci of malignant germ cells or neural elements, a yolk-sac tumor, neuroblastoma, or medulloepithelioma is present. These elements may be overlooked, as the clusters may be small, may be intermingled with immature tissue, or may not stain for alpha-fetoprotein (AFP). To add to this confusion, structures called fetal or embryonic intestine may occasionally be found in immature teratomas and may be related to elevated AFP levels. In an analysis of 85 benign and malignant teratomas in children, Grosfeld et al. reported that 21% of all teratomas in children were malignant [13].

Gastric teratoma (GT) is extremely rare, comprising less than 1% of all teratomas [4]. We culled the reported cases of GT in the world literature using the MEDLINE database from 1966 onward; a total of 102 cases were found. More than 90% of these were in males. Although the literature is replete with occasional case reports of malignant GT [3, 4, 19], there is only one case report of a GT undergoing malignant transformation [17].

The purpose of this review was to present the authors’ clinical experience during the period 1987–1999 with ten cases, which to our knowledge is the largest series of GTs from a single center. The importance of immature elements and the response to surgical treatment alone even in cases with infiltration of surrounding structures and lymph nodes metastases is discussed.

Materials and methods

Ten patients with GT diagnosed and treated at our institute were all born at full term; nine were males. The mean age at presentation was 3.2 months (range 10–180 days). Eight presented with an abdominal mass and two with abdominal distention and vomiting. Ultrasonography (US) revealed a mixed echogenic mass, and calcifications were noted in four cases. Computed tomography (CT) was done in five cases and revealed a solid and cystic multicirculated tumor of mixed echogenicity and calcifications. Serum AFP was determined by immunoenzymetric assay (AFP Serozyme, Biochem...
Immunosystems) and the results were compared with our laboratory nomogram. On comparison with age-related serum titers, AFP levels were grossly elevated in two cases that turned out to be grade III immature GTs, and the level in one case of a mature GT was just above 2 standard deviations.

Surgical exploration revealed the mass localized to the posterior wall of the stomach in six cases and arising from the posterior wall and extending to the lesser curvature and anterior wall as well in two. All except two tumors were well-encapsulated and without any gross metastases, and complete excision with a rim of normal stomach completed the treatment. The mean follow-up period was 4.2 years. The details of this series are briefly summarized in Table 1. There were no deaths, and histologic examination showed mature teratomas in eight cases and immature grade III teratomas in two, which are discussed here briefly as case reports.

Case 1

A 6-month-old male presented with an abdominal mass noticed by a pediatrician during a routine check-up. Abdominal examination revealed a firm, irregular mass at the left hypochondrium and epigastric region, the dullness of which to percussion was continuous with that of the liver. The liver was palpable up to 3 cm below the costal margin. US showed a large, multiloculated tumor with mixed echogenicity related to the stomach and liver with solid and cystic areas and calcifications. Serum AFP was 100 ng/ml. Laparotomy confirmed a large, multicystic tumor arising from the lesser curvature of the stomach and extending to the anterior wall, infiltrating the left lobe of the liver and transverse colon. Resection of the tumor along with a left hepatic lobectomy and resection and end-to-end anastomosis of the transverse colon was performed. Histologic examination of the tumor revealed a grade III immature teratoma. Serum AFP was absent 12 weeks postoperatively. At 18 months following the surgery, the child was asymptomatic and doing well.

Case 2

A 3-month-old male presented with abdominal distension and poor weight gain. Physical examination revealed a large mass in the umbilical and right lumbar regions. CT showed a large, multiloculated tumor with calcifications displacing the right kidney and bowel loops. Serum AFP was 1750 ng/ml. At laparotomy, a large tumor was arising from the posterior wall of the stomach. The regional lymph nodes were enlarged. Excision of the tumor (Fig. 1) and repair of the stomach was performed. Lymph nodes were also sent for histologic examination. The specimen was reported as a grade III immature GT with metastases to the lymph nodes. An omental biopsy also revealed metastases. Four months after surgical excision, there was no evidence of recurrence of the tumor. Serum AFP was undetectable.

Discussion

Teratomas are commonly observed in the sacrococcygeal region, gonads, mediastinum, and retroperitoneum. The etiology is not well-elicited and may be variable at different sites. Theotipotent cells are found in close proximity to the primitive streak and Hensen’s node in early embryogenesis, and the most widely accepted theory is that these are responsible for teratomas [1]. The site of origin in one of the largest series of benign and malignant teratomas in children was sacrococcygeal in 65%, mediastinal in 10%, gonadal in 11%, and presacral in 4% of cases [13].

GT was first reported by Eusterman and Sentry in 1922 [9]. It constitutes less than 1% of all teratomas in children, and so far 102 cases have been reported in the world literature. GTs are found mostly in male infants [16], and in our series there was only one female. The mean age at presentation in this series was 3.2 months. Presentation is usually as an abdominal mass or distension with or without vomiting. The patient may present with gastrointestinal (GI) bleeding [5, 10, 14], as the intramural component may ulcerate and hemorrhage. Rarely, the child may present with respiratory distress [15].

![Fig. 1 Specimen of gastric teratoma arising from posterior wall of stomach after complete excision](image)

Table 1 Summary of cases comprising this series (Calc: calcification, HPE: histopathological examination, MT: mature teratoma, IMT: immature teratoma, PW: posterior wall of stomach, LC: lesser curvature of stomach, AFP: alpha-fetoprotein, Gr grade, SD standard deviation, Mets: metastases)

<table>
<thead>
<tr>
<th>Patient no.</th>
<th>Age (days)</th>
<th>Cardinal sign</th>
<th>Calc</th>
<th>Site (predominant)</th>
<th>HPE</th>
<th>AFP (ng/ml)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>58</td>
<td>Mass</td>
<td>–</td>
<td>PW</td>
<td>MT</td>
<td>358 (normal)</td>
</tr>
<tr>
<td>2</td>
<td>110</td>
<td>Mass</td>
<td>–</td>
<td>PW</td>
<td>MT</td>
<td>288 (&lt;2SD)</td>
</tr>
<tr>
<td>3</td>
<td>10</td>
<td>Mass</td>
<td>+</td>
<td>LC</td>
<td>MT</td>
<td>2316 (normal)</td>
</tr>
<tr>
<td>4</td>
<td>112</td>
<td>Mass</td>
<td>–</td>
<td>PW</td>
<td>MT</td>
<td>84 (normal)</td>
</tr>
<tr>
<td>5</td>
<td>96</td>
<td>Distension</td>
<td>–</td>
<td>PW</td>
<td>MT</td>
<td>76 (normal)</td>
</tr>
<tr>
<td>6</td>
<td>92</td>
<td>Mass</td>
<td>–</td>
<td>LC</td>
<td>MT</td>
<td>84 (normal)</td>
</tr>
<tr>
<td>7</td>
<td>180</td>
<td>Mass</td>
<td>+</td>
<td>LC, liver, transverse colon</td>
<td>Gr III IMT</td>
<td>100 (&lt;2SD)</td>
</tr>
<tr>
<td>8</td>
<td>94</td>
<td>Mass</td>
<td>+</td>
<td>PW</td>
<td>MT</td>
<td>114 (normal)</td>
</tr>
<tr>
<td>9</td>
<td>90</td>
<td>Distension</td>
<td>+</td>
<td>PW, Enlarged lymph nodes</td>
<td>Gr III IMT, Mets +</td>
<td>1750 (&lt;2SD)</td>
</tr>
<tr>
<td>10</td>
<td>96</td>
<td>Mass</td>
<td>–</td>
<td>PW</td>
<td>MT</td>
<td>122 (normal)</td>
</tr>
</tbody>
</table>