JKAU: Med. Sci., **Vol. 20** No. 3, pp: 91-100 (2013 A.D. / 1434 A.H.)
DOI: 10.4197/Med. 20-3.8

Immature Gastroesophageal Teratoma: Second Case Report and Literature Review

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Abstract. Gastric and gastroesophageal teratoma are rare tumors that involve the stomach and gastroesophageal zone, both are of the same pathological nature. In reviewing literature, nearly 110 cases were reported, but the teratoma that involved the gastroesophageal area is extremely rare as only one case of mature gastroesophageal teratoma has been reported previously. Presented is the second case of gastroesophageal teratoma; a case of 18-days-old male patient with large immature gastroesophageal teratoma which was recognized as abdominal mass on antenatal ultrasound, but patient presented five days after noticing the abdominal mass by his mother. Discussion with the relevant technical considerations in the management of the teratoma originating from or involving the gastro-esophageal junction was considered.

Keywords: Teratoma, Gastric, Gastroesophageal, Immature antenatal, Neonatal.

Introduction

Teratoma in general is divided to gonadal and extra gonadal. Sacrococcygeal teratoma is the most common type of extra gonadal teratoma. Gastric teratoma (GT) and gastroesophageal teratoma (GET) are rare[1-3], and represent the same pathological lesion. However, the

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Accepted for publication: 26 February 2013. Received: 11 December 2012.

teratoma of gastroesophageal zone is most rare as only one case of a male patient with mature gastroesophageal teratoma was previously reported[4].

The present report is a case study of an 18-days-old male patient with large immature GET with relevant technical considerations in the management of the teratoma originating from or involving the gastro-esophageal junction; reviewing the literature on GT and GET as being the same pathological entity.

Case Report

The patient was an 18-days-old Saudi male, a product of full term pregnancy and delivered by cesarean section, was re-admitted to our hospital due to abdominal distention. Abdominal mass was recognized ultrasonically during antenatal period, but was noticed by the mother 5 days prior to presenting to the outpatient clinic. The mass was obvious in left side of the abdomen, with no feeding intolerance or fever, his weight on admission was 3.5 kg.

On examination, the patient was vitally stable, not in distress, and not jaundiced or pale. Abdominal examination revealed generalized abdominal distention and visible swelling at upper abdomen (Fig. 1). Palpation confirms a huge firm mass involved in the upper part of the abdomen, more to the left side; otherwise the rest of the abdomen is soft and lax. Systemic review and examinations were unremarkable.

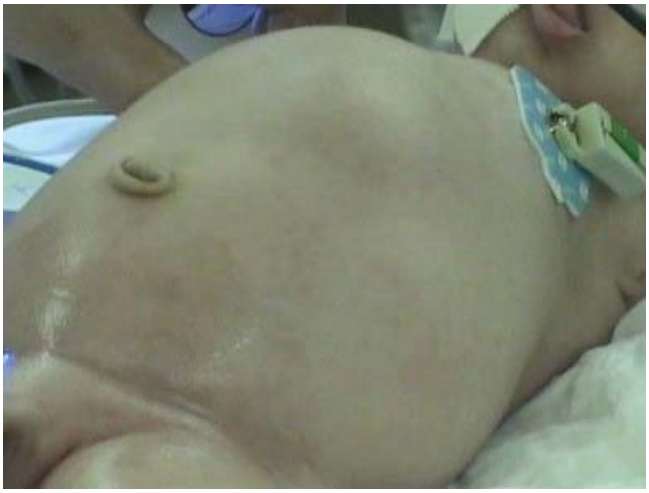

Fig. 1. Patient with abdominal distention and swelling at upper abdomen.

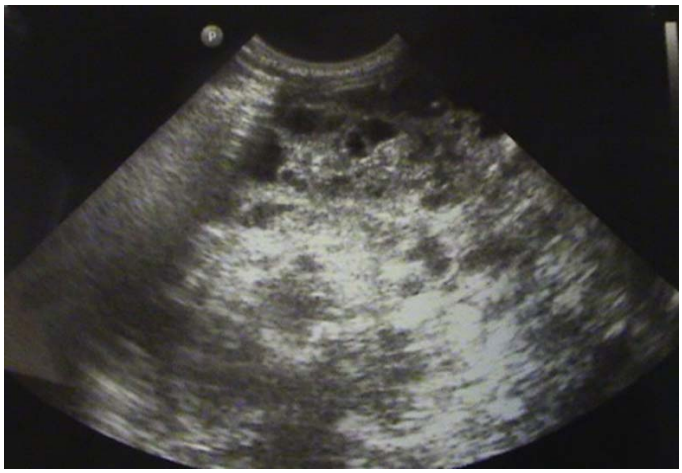

Fig. 2. Abdominal ultrasound showed a large abdominal multicystic mass occupying the whole abdomen.

Investigations were done including complete blood count (CBC), urea and electrolytes (U&E), coagulation profiles, all within normal limit apart of α-fetoprotein, which was very high (2249 IU/mL dropped post operatively in one month, 104 IU/mL and at 6 months to 0.7 IU/mL at 5 years remain 0.5 IU/mL). Radiological study was performed, *i.e*., plain abdominal X-ray which showed a mass with gross calcification occupying the upper and central abdomen. Abdominal ultrasound showed a large central abdominal multicystic mass occupying the whole abdomen displacing the liver laterally and obscuring the midline structure (Fig. 2). Computerized axial tomography (CT) scan abdomen and chest with oral and intravenous contrast revealed a huge well circumscribed cystic lesion (7.8 x 8.2 x 11 cm) in the anteriorly left side and central part of the abdomen with foci of calcification reaching to the diaphragmatic level, pushing the stomach anteriorly and the transverse colon, and the Lt kidney downwards (Fig. 3).

The patient underwent exploratory laparotomy through left upper transverse incision, which revealed multilocular mass around 10 X 10 X 4.5 cm, with cystic and solid components. Hence, arising from the posterior gastric and lower esophageal wall, and pushing the stomach anteriorly (Fig. 4), and weighing 550 grams.

Operative procedure: Excision of the mass was done with part of the upper posterior gastric and lower esophageal wall which has been sent to

frozen section. Histopathology assessment revealed free margin and no malignancy cells were detected. Then, repair of the stomach and esophagus were done over 14F nasogastric tube (NGT) with 4/o vicryl. On fifth day postoperatively, patient started oral feeding.

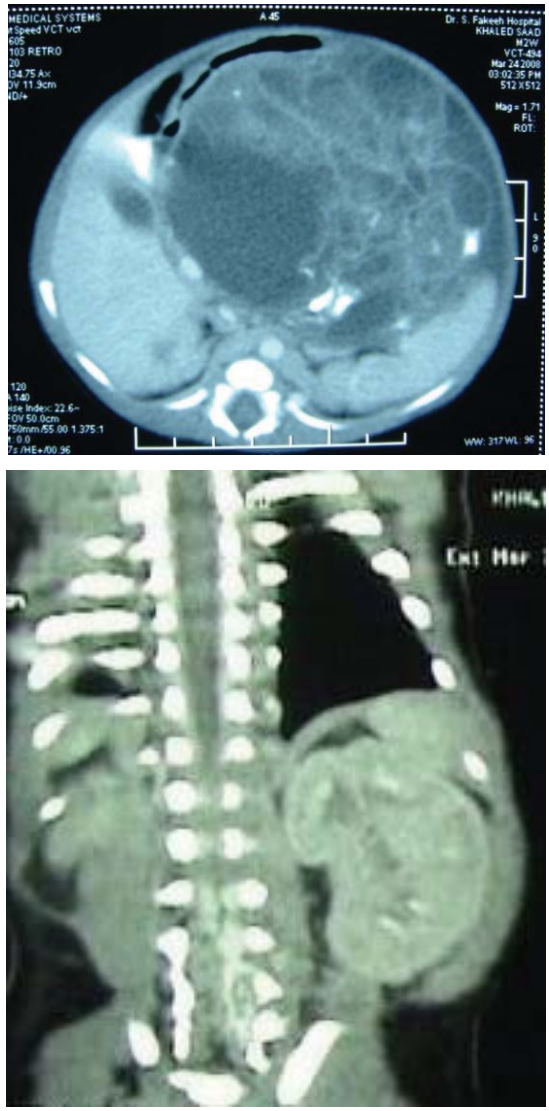

Fig. 3. **CT scan abdomen and chest with oral and intravenous contrast, which revealed huge mass involving the posterior aspect of the stomach and abdominal part of the esophagus, pushing the stomach anteriorly, it contained cystic and solid components with multiple foci of calcification.**

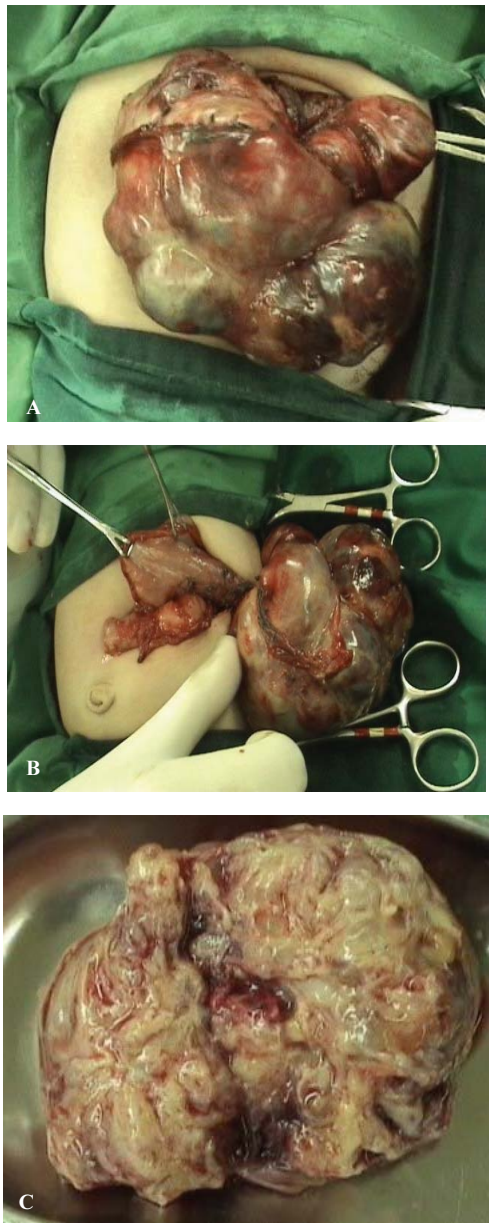

Fig. 4. **Exploratory laparotomy done with the finding of: A) Large lobulated tumor mass10 X 10 X 4.5 cm with cystic and solid component attached to posterior wall of the stomach and the intra-abdominal esophagus; B) The mass separated easily from the posterior wall of the stomach with adherence at the gastro-esophageal area indicating that the mass originating from the gastroesophageal area; C) The excised mass bisected showing the cystic and solid component.**

Histopathology study revealed the picture of immature teratoma, grade II with the immature elements representing 10% of the sampled surface (neuroepithelial tissues) (Fig. 5).

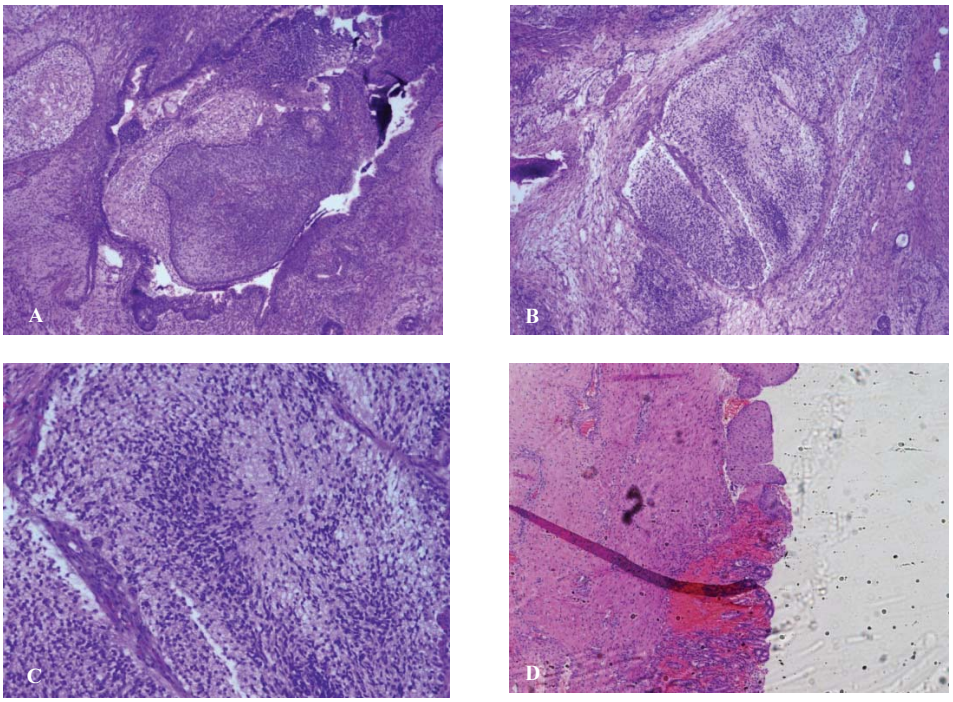

Fig. 5. Histopathological study of the tumor mass was done with the finding of: A) The slid showing tissues of ectodermal, mesodermal and endodermal origin LPF; B) The slid showing immature elements LPF (neuroepithelial tissues); C) The slid showing immature elements HPF (neuroepithelial tissues); D) The slid showing the gastroesophageal junction which was excised with the GET originating from that area.

Discussion

Teratoma is defined as a tumor having the three embryonic layers (endoderm, mesoderm, and ectoderm) that arise from totipotent primordial germ cell. Gastric and gastroesophageal teratoma (GT-GET) most commonly affecting male infant [1-3]. The first gastric teratoma in a female infant was reported in 1979[5], since then, few female cases reported thereafter[6,7].

Gastric and gastroesophageal teratoma (GT-GET) are rare tumors of the same pathological nature, however, the involvement of the gastro-

esophageal junction is very rare. The first case of gastric teratoma was reported in 1922 by Eusterman and Sentry[8], while the first case of gastroesophageal teratoma was reported in 1968 by Giacomoni and Zaffaroni which was mature teratoma[4]. The present case is the second GET, but with immature neuroepithelial tissues.

Two cases were previously reported in Saudi Arabia of such pathology but both where limited to stomach; one was mature while the other was immature teratoma[9,10].

Abdominal mass and abdominal distention is the most common presentation in GT-GET, but can present with hematemesis due to erosion of gastric mucosa and vomiting due to gastric outlet obstruction. Furthermore, a large tumor can cause premature labor and dystocia[3,10-13]. Spontaneous rupture of large teratoma of stomach was also reported[14,15]. Most of them arising from the posterior wall of the stomach at greater curvature; however, our teratoma involved the posterior wall of stomach and lower esophagus. Mostly, separable from the stomach, except at the gastroesophageal zone, which confirms that was originated from that zone. It is also common to originate from greater curvature; three cases were reported arising from lesser curvature[16,17]. Antenatal ultrasound can detect the fetal abdominal masses as seen in our patient; however, the diagnosis was not made except when the neonate presented with obvious abdominal mass with high level of the tumor marker (α-fetoprotein) and the radiological finding of plain abdominal X-ray. The more detailed information seen on abdominal ultrasound, CT scan abdomen and chest (cystic and solid component, gross calcification the location and dimensions), the diagnosis was further confirmed at exploratory laparotomy and finally histopathologically .

Gastric and gastroesophageal teratoma (GT-GET) are almost always of a benign nature[3]. Although, malignancy has been reported as teratoma with evidence malignancy or malignant transformation in immature teratoma[7,18,19]. The typical teratoma predominantly exogastric, which was found in our case, was having both solid and cystic areas. Total excision and primary closure of the gastric or gastroesophageal wall is the treatment of choice, and usually curative with excellent prognosis[3,7,11,13]. Recurrence and malignancy are rare, even with local infiltration or nodal metastasis[3]. Partial, subtotal or total

gastrectomy has been reported depending on the location and to the extent of the teratoma[16]. Adjuvant chemotherapy and radiotherapy are not required[15].

The involvement of the gastro-esophageal junction dictates specific considerations during the excision of the mass in relation to involvement of the vagal nerves. Hence, might necessitate gastric drainage procedure if the vagal nerves were divided, if the esophageal wall involved in the resection, then it is necessary to preclude the esophageal narrowing and the gastro-esophageal reflux, which can be evaluated by utilizing the contrast swallow and meal when patient develops any obstructive or gastroesophageal reflux symptoms. However, in the present case, we precluded involvement of the vagal nerves intra-operatively, and no symptom of stenosis or reflux was occurred.

In conclusion, presented is the second case of gastroesophageal teratoma with immature neuroepithelial tissues grade II with 5 years follow up with no recurrence. However, since the immature GT-GET is potentially a malignant tumor, regular follow-up of the patient with tumor markers (α-fetoprotein) is recommended, to preclude residual teratomata tissue, recurrence or malignant transformation, and then, utilize further imaging studies accordingly. Adjuvant chemotherapy and radiotherapy is not required.

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ورم عجائبي معدي مريئي مكتمل البضج، تقرير حالة ومراجعة للأدبيات الطبية

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المستخلص. الورم العجائبي المعدي والمعدي المريئي من الأورام النادرة وهي ذات طبيعة واحدة من الناحية النسيجية المرضية إلا أن الورم المعدي المريئي شديد الندرة حيث وجدنا في الأدبيات الطبية حالة واحدة من الورم المعدي المريئيي مقابل (١٠٠) حالة ورم معدي فقط، ونقدم هنا الحالة الثانية من الورم المعدي المريئي في مولود عمره (١٨) يوماً وقد سبق رصد هذا الورم أثناء الحمل بالأشعة الصوتية ثم تمت المتابعة والتشخيص بعد الولادة بفحص الموجات فوق الصوتية والأشعة السينية والمقطعية ومؤشر الورم (البروتين الجنيني أ). ونناقش هذه الحالة من الورم العجائبي المعدي المريئي مع مراجعة الأدبيات الطبية لهذا النوع من الأورام مع الأخذ في الاعتبار لبعض النقاط التقنية الجراحية التي يجب مراعاتها عند استئصال الورم الناشيء أو الممتد إلى المنطقة المعدية المريئية.