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Case Report

Management of retroperitoneal teratoma in infants younger than one-year-old

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Abstract: The study was conducted to evaluate the clinical results of surgery treatment for retroperitoneal teratoma in infancy. The study group consisted of consecutive patients (The patients who are diagnosed consecutively in our hospital and we got the complete case information). Less than 1-year-old (median age, 4.2 months) who underwent resection of retroperitoneal teratoma with vascular involvement. The procedures were performed between 2002 and 2011. Vessels involved by teratoma were artery and vein. Patient data were prospectively gathered in a database and retrospectively analyzed. 42 patients with retroperitoneal teratoma were involved, all of them underwent tumor resection. 39 patients were mature teratoma, 2 patients were grade I immature teratoma and grade II immature teratoma was in one patient. The most common vascular involvement pattern was vein only at 64.3%. Arterial and vein and arterial only involvements were observed in 28.6% and 7.1% of the cases, respectively. Vascular repair was done in 22 patients in the resection. All the tumors were not malignant, and nobody recurred. There were no deaths, and no child has hypertension or thrombosis post operation. The most common complication is hemorrhage. The median follow-up is 3.5 years (interquartile range, 3 to 4.375 years). The retroperitoneal teratoma could surround major vessels, making resection difficult. An acceptable surgical risk underlines the value of en bloc resection of the lesions. The oncologic outcome of retroperitoneal teratoma is positive especially with early diagnosis and treatment.

Keywords: Retroperitoneal teratoma, tumor resection, infant

Introduction

Retroperitoneal teratoma is an uncommon entity in infancy [1]. Due to the retroperitoneal location, these tumors can grow to be quite large before signs or symptoms are detected. Because of the location and massive size, major vessel anatomy may be distorted. Vascular injuries are well-recognized surgical complications with urgent repair. But retroperitoneal teratoma would difficult to recurrent, and they were shown to be caused by local recurrence. Local control is therefore important in the treatment of infant patients. Surgical resection is the mainstay treatment to retroperitoneal teratoma.

In a subset of patients, retroperitoneal teratoma involves major venous or arterial blood vessels. In order to reduce recurrence, patients with locally resectable disease should undergo aggressive operation. In this clinical setting, teratoma surgery represents a challenge in terms of treatment concept and technique. Furthermore, detailed analysis has shown that only few clinical data are available to answer the question of how blood vessel involvement influencing resectability, surgical approach, and the further course in infant. Due to the restricted number of clinical series, the clinical result of blood vessel involvement by retroperitoneal teratoma is not understood very well.

The aim of this study was to assess vascular involvement in retroperitoneal teratoma of patients under one-year-old, to suggest a treatment algorithm. Immediate-term outcomes were analyzed for retroperitoneal teratoma surgery performed during a 10-year period.

Case presentation

Inclusion criteria

A retrospective review of consecutive patients operated at the Department of Oncological Sur-

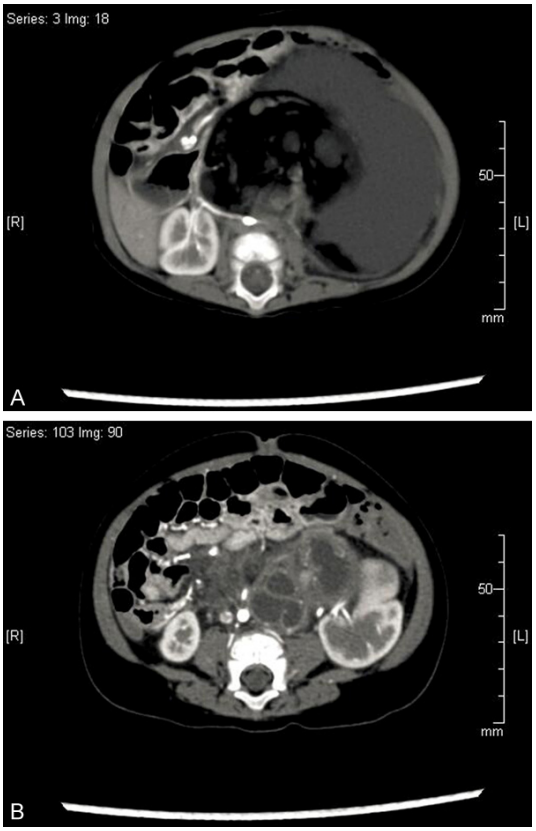

Figure 1. A, B. CT images show a 3-month girl with retroperitoneal teratoma. The tumor includes left renal arteries and reaches the level of the superior mesenteric artery.

gery, Children's Hospital of Chongqing Medical University from January 2002 to December 2011 with the diagnosis of retroperitoneal teratoma younger than 1-year-old were included in the analysis. Tumors were studied preoperatively by ultrasonography and computed tomography (CT). The pathologists assessed the resected specimens histopathologically. The present study was approved by the Ethics Committee of Chongqing Medical University.

A total of 42 patients younger than 1-year-old with retroperitoneal teratoma were included. The median age was 4.2 months (1 month to 11.5 months). 32 of the 42 children were girls, and 27 were younger than 6 months and 13 were younger than 3 months at the time of operation. Patients took the examination of the chest, abdominal and pelvic computed tomography (CT) scan and abdominal ultrasound (**Figures 1**, **2**). CT scan revealed the cystic and/or with solid mass or calcification in the retroperitoneum. Types of vascular involvement

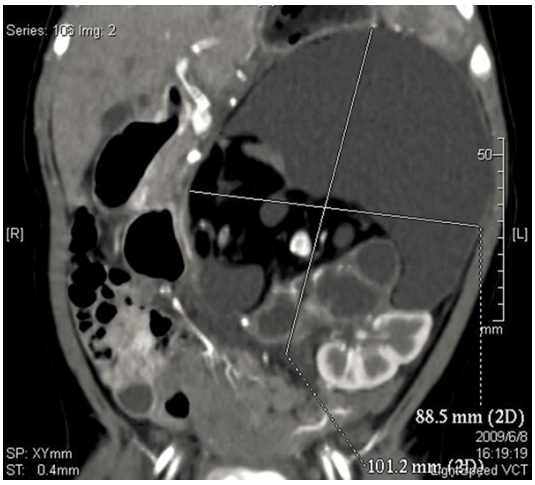

Figure 2. Coronal reconstruction of the preoperative contrast enhanced CT scan shows the tumor and gross displacements of left kidney.

were assessed preoperatively by CT scans. It was classified as follows: tumor involving major arteries and veins; tumor affecting only arterial blood vessels; tumor involving the veins without altering an artery. The most common vascular involvement pattern was vein only at 64.3% (27 cases). Arterial and vein and arterial only involvements were observed in 12 cases (28.6%) and 3 cases (7.1%) of the cases, respectively. The vessels include inferior vena cava, renal vessels, superior mesenteric vessels and iliac vessels. The tumors in 36 cases (85.7%) were >8 cm in diameter. The largest lesion was with a tumor diameter of about 14 cm. The serum level of tumor marker alpha-fetoprotein (AFP) was normal in most of the patients. β-Human Chorionic Gonadotropin (hCG) and lactate dehydrogenase (LDH) were normal in all of the patients (**Table 1**).

Patients have taken exploratory laparotomy which was undertaken under full intubated general anaesthesia with arterial and venous pressure monitoring. A urinary catheter and gastric tube are also inserted. The abdomen is opened through a transverse supra-umbilical incision. On whichever side the tumor arises, the colon is reflected medially to display the tumor. If most of the tumor is on the left side, the spleen and pancreas are also mobilized and all the viscera are placed sideward. Display all the major vessels partly which traverses the tumor, then separate the tumor and the tunica adventitia of major blood vessels. The viscera have to be carefully mobilized off the surface of the lesion.

Table 1. Tumor marker of patients with retroperitoneal teratoma

	Age	Mean ± SD	Cases	Female	Male
AFP ng/ml	1 month	6122±2918.7	3	3	0
	<3 months	161.7±86.6	10	7	3
	<6 months	23±15.6	14	11	3
	<12 months	11.8±25.1	15	11	4
HCG ng/ml		0.69±0.51	42	32	10
LDH IU/L		131.3±51	42	32	10

Once the vessels and viscera are free, the tumor is completely removed. The vessels and viscera are subsequently cleared circumferentially, after which the tumor may be removed. No arterial was reconstructed. Venous reconstructions were usually performed by the appropriate method, such as primary anastomosis, or a venous patch. Vascular repair was done in 22 patients (52.4%) in the resection. Patch was performed in 21 patients (50%). Vascular reconstructed in 1 of the patients (2.4%) with right iliac vein involvement. All tumors are located in the retroperitoneum, with preponderance on the left side (29 cases, 69%). 83.3% (35 cases) of the tumors contained calcification. Each patient has received copious volumes of intravenous fluids intraoperatively. The operating time ranged from 3 to 5.5 hours (median, 4.25 hours). Hemorrhage was the most common complication, the intraoperative bleeding was 54.40±30.02 ml. There was no mortality during operation.

Pathologic analysis of the resected specimens revealed the exclusive presence of teratoma in all cases. All patients were diagnosed with the first, and histopathologic examination of the specimens showed that mature teratoma without a malignant germ-cell tumor component in 39 patients, grade I immature teratoma in two, and grade II immature teratoma in one. No one was grade III immature and malignant teratoma. One patient with immature teratoma (grade II) had slightly increased AFP levels before tumor resection, but two weeks after operation, AFP value had dropped to normal. The patient with grade II immature teratoma did not have postoperative chemotherapy. None of the patients in this study had local recurrence during the fellow up period. No wound infection postoperation. Prolonged bowel occurred in 4.7% of the cases. Various unspecific symptoms such as abdominal distension, irritability, noisy crying were recorded.

Discussion

Retroperitoneal teratoma is one of the most common primary retroperitoneal tumors in the pediatric population. The treatment of retroperitoneal teratoma is complete surgical excision, because teratoma may result in the degeneration. Malignant teratoma may cause a rise of tumor marker in serum such as AFP, and the prognosis of some patients with teratoma malignant transformation was poor [2]. Also the bulky teratoma was associated with increased difficulty in dissecting the tumor mass from major vascular structures, the involvement of major vascular structures is no longer considered a contraindication for the resection of retroperitoneal teratoma. En bloc excision is clearly an option when vascular encasement is present. In our study, the incidence of retroperitoneal teratoma in females is triple that in males. It is different from the reported retroperitoneal teratoma had a 2:1 female to male incidence, and most are benign, with a malignancy rate of 7% [3]. Maybe the small sample size yielded the different results. Patient in our research is under 1-year-old, and we did not find malignant tumor in these cases. The differential diagnosis of retroperitoneal teratoma in infant includes nephroblastoma, neuroblastoma, ovarian tumors and lymphangioma. Valeria Solari et al reported a 4 months female child with this tumor who underwent a transverse laparotomy. And they reported the focus were “early assessment of the typically anterior and unique venous anatomy, resection of occluded veins where necessary and centripetal tracing of vessels from peripherally displaced organ” [4]. Preoperative scans provided some useful information. In our study ultrasound scanning confirmed the cystic or solid mass and renal blood flow, but did not reveal the distorted vascular anatomy. CT but not magnetic resonance imaging was used in our cases because of rapid access and short scanning times to assessment of the tumor and the arterial and venous anatomy. Niall M et al reported 6 children with retroperitoneal teratoma that involved surrounding structures. And major retroperitoneal vessels were intimately involved with the tumor. They found bleeding was unavoidable [5]. Some surgeon used polytetrafluoroethylene (PTFE) tube in an aortic-aortic connection, or in an aortic-iliac connection in adult patients with

growing teratoma syndrome underwent vascular surgery with standard retroperitoneal lymph node dissection [6]. Although major vessels were sacrificed in 22 of our 42 cases, we did not have to revert to replacement techniques to preserve supply and drainage of blood to the major organs. No malignant component was found in our study, there was less tissue adhesion between tumor and peripheral organs. Most of the vessel damage was slight, just one patient's right iliac vein was needed reconstructed. And considering growth of the child, we did not use prosthetic vascular replacement in selected case. With these tumors, most of the vessel and organs are surrounded, making resection difficult and increasing operative risk. It was a significant challenge to display of the major vessels and excise the retroperitoneal teratoma. Other research reported the experience with retroperitoneal teratoma in 10 patients over a 5-year period. They reported retroperitoneal teratoma had no apparent connection to the retroperitoneal organs and were easy to be excised. And histologic evidence of grade III immature teratoma or malignancy demanded aggressive postoperative chemotherapy to prevent local recurrence [7]. In our institution, the treatment of extragonadal mature and immature germ cell tumor after radical excision is observation. No chemotherapy was used in our research.

Retroperitoneal teratoma is rare and difficult to early diagnose in infancy because of nonspecific signs and symptoms. On time antenatal examination and child health care is necessary in order to get early diagnosis and treatment. The bulletin of a statistic of health and family planning programme of China in 2010 and 2012 showed the rate of maternal antenatal examination and child health care under 3-year were 92.2%, 94.1%, 93.7% and 77.2%, 81.5%, 84.6% respectively from 2009 to 2011 in China [9]. It was means that almost a quarter of children under 3 years old did not have regular child care and physical examination. In our study only 5 (11.9%) of them were detected an abdominal mass when they took child care. And other patients did not have regular physical examination when they were born. Two of the patients were detected when prenatal examination, they took surgery one month after birth. Other patients come to hospital because of abdominal distention, pneumonia, feeding difficulty, emesis or diarrhea. Recommendation on examination in the nearby Maternal and Child Health centers for pregnant woman and young child is necessary. Early detection and diagnosis of teratoma is the most efficient way to improve the prognosis and reduce medical expenses. Once the diagnosis is made, surgical management is the therapy of choice.

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Disclosure of conflict of interest

None.

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