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Retroperitoneal Giant Immature Teratoma Diagnosed at Birth and treated post-neonatally: A Case Report

Toshiko Takezoe[1], Wakako Sumiya[2], Yuichi Mitani[3], Kayoko Ichimura[4], Shoichi Tsuzaka[1], Takashi Tsutsuno[1], Shoko Ogawa[1], Yasuhiro Kondo[1], Kyoichi Deie[1], Hiroshi Kawashima[1]

[1]Department of Pediatric Surgery, Saitama Children's Medical Center, 1-2, Shintoshin, Chuo-ku, Saitama-city, Saitama, 330-8777, Japan

[2]Department of Neonatology, Saitama Children's Medical Center

[3]Department of Hematology and Oncology, Saitama Children's Medical Center

[4]Department of Pathology, Saitama Children's Medical Center

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ABSTRACT

Background: Immature retroperitoneal teratomas (RTs) are rare tumors predominantly diagnosed within the first year of life, with the majority being benign. Complete surgical resection is the primary treatment; however, surgical challenges due to anatomical complexities and perioperative complications often arise, particularly in neonates. This study presents a case of a congenital immature RT identified at birth, where the timing of surgical intervention was critically evaluated through a review of relevant literature.

Case presentation: The patient, a male neonate born at 36 weeks, exhibited abdominal distention and was transferred for further evaluation. Imaging revealed a large retroperitoneal mass, leading to a diagnosis of immature teratoma. An initial biopsy confirmed the absence of malignancy, and surgical resection was delayed to allow for further development of the infant. The tumor, weighing 930 g, was successfully removed while preserving the right kidney, though postoperative urinary peritonitis occurred. The patient recovered and he was discharged with no recurrence after seven months.

Conclusion: This case highlights the importance of carefully timing surgery in neonatal patients with complex conditions to minimize surgical risks.

Keywords: *Immature teratoma, retroperitoneal teratoma, neonatal surgery*

1. INTRODUCTION

Retroperitoneal immature teratomas (RTs) are rare, with more than half identified within the first year of life. [1] Most cases are benign, while malignancies occur in < 15 % of cases. [1–3] Complete surgical removal is curative and remains the primary treatment for immature RTs. [1, 2, 4] However, the surgery remains challenging because of its anatomical features, which are often difficult to manage owing to perioperative complications, particularly in the neonatal period. However, there are no reports that mention the optimal timing for surgical intervention in neonates with congenital immature RT. Therefore, in this study, we report a case of congenital immature RT detected at birth and evaluate the optimal timing for surgical intervention based on a review of relevant literature. This report reviews the clinical presentation, imaging findings, surgical approach, and postoperative outcomes, in addition to a literature review on timing strategies for surgery.

2. CASE REPORT

A male neonate was born at 36 weeks of gestation, weighing 3,692 g. He developed abdominal distention on the day of birth and was transferred to our hospital with suspected fetid peritonitis. Abdominal ultrasonography and computed tomography (CT) revealed a large retroperitoneal mass, consisting primarily of cystic components and a few solid areas (Fig 1). The tumor appeared to completely encompass the right kidney, its vessels, and the right ureter. Alpha-fetoprotein level was high (678,063 ng/mL), and β-human chorionic gonadotrophins were normal (1.25 ng/mL). No other abnormal blood test findings were observed. Based on these findings, an immature teratoma was suspected, and primary resection was considered. However, the localization and size of the tumor posed significant challenges. A mixed gonadal tumor was also considered, for which chemotherapy was expected to reduce the tumor size. Therefore, a tumor biopsy was performed to confirm the diagnosis and reexamine treatment options. Biopsy confirmed an immature teratoma without malignancy, making surgical resection the preferred approach. However, because the child had only mild renal hypertension and maintained stable oral intake, surgery was re-scheduled till after the child had grown. Unfortunately, respiratory disturbances and leg edema caused by tumor growth developed approximately 2 weeks later, prompting the decision to proceed with tumor resection on day 38 of life. During resection, the tumor was found to have originated from the right retroperitoneal space, involving vessels of the right kidney and ureter. Complete tumor resection was performed while preserving the right kidney. Portal vein laceration required vessel repair. The tumor weighed 930 g, with an operative time of 284 min and a blood loss of 890 g during the procedure. Pathological findings confirmed a diagnosis of a grade 3 immature teratoma (Fig 2).

Postoperatively, the patient developed urinary peritonitis, probably owing to an intraoperative ureteral injury, which resolved with conservative treatment. Antihypertensive medications were discontinued 50 days after surgery, and the patient was discharged on day 53. Presently, 7 months post-surgery, no recurrence has occurred, and the child is developing age-appropriately.

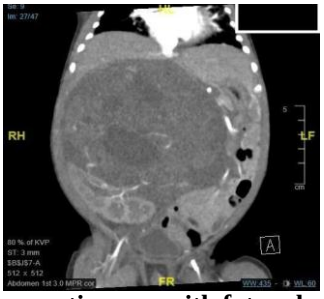

Figure 1 Enhanced CT image showing a large cystic mass with fat and calcification. The right kidney was displaced downward by the tumor, while the right renal artery and ureter penetrated the tumor center.

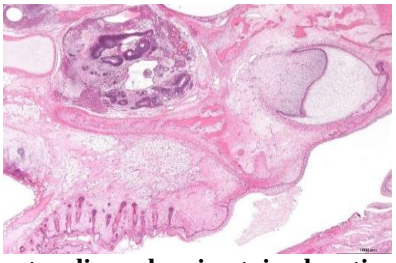

Figure 2 Microscopic findings. Hematoxylin and eosin-stained sections show immature neural, skin, and cartilaginous components characteristic of grade 3 immature teratoma. No malignancy.

3. DISCUSSION

Teratomas are classified as mature or immature based on their histologic composition and degree of maturation or differentiation. [5] RTs constitute 5% of teratomas and primarily occur in infants. [2] Most RTs are mature, with complete surgical resection as the standard treatment. [1] Similarly, surgical excision is an effective treatment for childhood-onset immature teratomas. [5] However, Surgical removal of RTs is typically challenging, with perioperative complication rates ranging from 40% to 100%. [1, 4, 6, 7] T. Yang et al. report that the risk factors for perioperative complications during RT resection encompass the vasculature and distorting organs. [1] In the present case, the tumor was present at birth, but the timing of resection was carefully considered with close follow-up, and surgery was performed on day 38. This approach was considered in anticipation of surgical challenges owing to organ immaturity in the neonatal period. Reports of immature RTs resected in patients < 1 year of age since 2000 were reviewed, examining 25 detailed cases (Table 1). [4, 6, 8–11] The present case (Case 25) was among them. Of the 25 patients, 12 underwent surgery in the neonatal period and 13 in the post-neonatal period. Four postoperative deaths occurred, all in the neonatal period.

Table 1. Clinical characteristics and complications of the 25 patients

♗ ge at case surgery		Sex	Perioperative complications	Late complications	Pathology	Outcome
1[1]	21 days	M	Gastric tear, hypotension	Hiatal hernia recurrence	Immature with YST	Well 2 years
2[1]	1 day	F	Blood loss-2.3L,	N.D. ♗	Immature with YST	Death at day 12
			disseminated intravascular coagulation, sepsis			
3[1]	3 months	F	Common bile duct injury, perioperative sepsis	None	Immature, grade 2	Well 6 months
4[3]	21 days	M	Hypertension	None	Immature	Well 2.5 years
5[8]	14 days	F	Lt nephroureterectomy	None	Immature	Well 7 months
6[9]	21 days	M	Congestive cardiac failure due to VSD	Recurrence	Immature, grade 3	Death after op. for removal of
				(after 5 months of op.)		the recurrent tumor
7[10]	6 months	F	♗ section of the gastric wall resection	None	Immature, grade 2	Well 6 months
8[11]	6 days	F	None	None	Immature, grade 3	Well 1 month
9[11]	1 month	F	Lt renal artery injury	Lt renal dysfunction	Immature, grade 3	Well 9 years
10[11]	2 months	F	None	Small bowel obstruction	Immature, grade 1	Well 8 years
11[11]	5 months	F	None	None	Immature, grade 1	Well 12 years
12[11]	5 months	M	None	None	Immature	Well 3 years
13[11]	6 months	M	None	None	Immature	Well 1 year
14[11]	9 months	M	None	None	Immature, grade 1	Well 3 months
15[4]	0 month	F	Diaphragm injury	None	Immature	Well

16[4]	25 days	M	Hypertension, hypovolemic shock state, brain death	multiple organ failure	Immature, grade 1	Death after 8 months of op.
17[4]	2 months	F	None	None	Immature, grade 1	Well
18[4]	0 month	M	PV injury, amputation of GD	None	Immature, grade 2	Well
19[6]	3 months	M	None	None	Immature, grade 1	Well 18 years
20[6]	0 day	M	Uncontrollable diffuse bleeding leading to death		Immature, grade 2-3	Died a few hours after birth
21[6]	5 months	M	Small caval vein tear	None	Immature, grade 1	Well 12 years
22[6]	1 day	F	None	None	Immature, grade 1	Well 9 years
23[6]	4 months	M	Choledochal tear	None	Immature, grade 1	Well 3.5 years
24[6]	10 days	M	Esophagogastric tear	None	Immature, grade 2	Well 1 year
25	1 month	M	Hypertension,	Right renal atrophy	Immature, grade 3	Well 7 months
			rt ureter injury, PV injury			

YST yolk sac tumor, VSD ventricular septal defect, PV portal vein, GD gastroduodenal artery, N.D. no data available

** 25 cases of retroperitoneal immature teratoma operated on at < 1 year of age, including 24 cases reported in detail since 2000 and this case.*

Intraoperative complications occurred in 10 cases (83%) in the neonatal period, compared to 6 cases (46%) in the post-neonatal period. Long-term complications were similar, with three cases (25%) in the neonatal surgery group and three cases (23%) in the post-neonatal surgery group. Therefore, careful monitoring of clinical manifestations in large tumors, such as in this case, and delaying surgery until organ growth and development before surgery is recommended. Conversely, the necessity of a biopsy remains controversial. In the present case, an open tumor biopsy was performed to evaluate the potential of tumor shrinkage bychemotherapy. Mixed tumors with yolk sac tumors can occur in immature teratomas. Heifetz et al. report that 11 of 28 extragonadal immature teratomas observed at 0–3 years had yolk sac tumors[12]. Neyssa M. M. et al. also report that four of seven patients with RTs under 6 months of age had malignancy complications. Therefore, malignancy complications are often reported in immature teratomas, and the higher the grade of immature teratomas, the higher the complication rate. Complete surgical excision is an effective treatment, even for high-grade immature RTs, and the disease-free rate after resection is high. [13] In cases such as the present one, where an immature teratoma is strongly suspected clinically and radiologically, a biopsy is unnecessary, and careful consideration of the timing for tumor resection surgery is crucial. This study has a limitation. The reviews used in this study were limited to cases with detailed reports and did not include review articles with a larger number of cases. Therefore, further accumulation of case data is required for future analysis.

4. CONCLUSION

Although surgical resection is the standard treatment for large RTs diagnosed at birth, the timing of resection should be carefully considered with close follow-up, anticipating surgical challenges due to the immaturity of neonatal organs.

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