Prenatally diagnosed intra-abdominal testicular teratoma managed with single-stage Fowler-Stephens orchiopexy postnatally: A case report and literature review

HONGGANG FANG[1], GUANGBEI PENG[2], XING LIU[1], SHENGDE WU[1], YI HUA[1], GUANGHUI WEI[1] and DEYING ZHANG[1]

[1]Department of Urology, Children's Hospital of Chongqing Medical University, Clinical Research Center for Child Health and Disorders, Ministry of Education Key Laboratory of Child Development and Disorders, Key Laboratory of Structural Birth Defect and Reconstruction, Chongqing 400014, P.R. China; [2]Department of Urology, Jiangxi Children's Medical Center, Nanchang, Jiangxi 330200, P.R. China

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Abstract. Fetal intra-abdominal testicular tumors diagnosed prenatally are exceedingly rare. However, the increasing utilization of prenatal ultrasound has led to the identification of these uncommon neoplasms. The present study reports the case of a 1-month-old male infant in whom an intra-abdominal mass was first identified on a fetal ultrasound performed at 28 weeks of gestation. Postnatally, the patient underwent complete tumor excision followed by single-stage Fowler-Stephens orchiopexy. The 7 months postoperative follow-up revealed normal findings. To the best of our knowledge, this is the first reported case of prenatally diagnosed intra-abdominal cryptorchidism associated with a mature teratoma, successfully managed with testis-sparing surgery (TSS) using Fowler-Stephens orchiopexy. The present study represents the ninth such case reported worldwide. Although neonatal testicular tumors generally require surgical excision, intraoperative frozen section analysis combined with TSS can effectively prevent unnecessary orchiectomy and enable the potential for bilateral fertility preservation.

Introduction

Primary testicular tumors in children are rare, accounting for ~1% of all pediatric solid tumors, with an annual incidence of 0.5-2 cases per 100,000 boys (1,2). Testicular teratomas in neonates are even more uncommon, with an estimated incidence of 0.015-0.06 cases per million live births (3). A large cohort study has shown that yolk sac tumors are the most common prepubertal testicular neoplasm, comprising ~42% of cases, with >97% classified as malignant. Notably, ~75% of these cases require radical orchiectomy (4). However, current treatment approaches remain limited by the lack of standardized management guidelines and the potential risk of overtreatment in benign cases (5). In the present study, the rare case of a fetal testicular teratoma diagnosed prenatally at 28 weeks of gestation is reported. A comprehensive perioperative assessment, including intraoperative frozen section analysis and histological evaluation of the peritumoral tissue, enabled successful testis-sparing management, thereby avoiding unnecessary orchiectomy and preserving bilateral testicular function and future fertility potential.

Case report

A male infant was delivered vaginally at 39 weeks after a fetal ultrasound performed at 28 weeks revealed a cystic mass superior to the fetal bladder, suspected to be a teratoma. Prenatal ultrasound of the fetus in a 31-year-old primigravida demonstrated a well-defined anechoic area with a thin wall and good sound transmission in the right-superior bladder region, showing a 1.6x1.1 cm irregular hyperechoic focus with punctate echogenic foci and scattered vascular signals on color Doppler flow imaging (CDFI) (Fig. 1A and B); this lesion had not been observed on earlier scans. Postnatal examination revealed an empty right hemiscrotum without palpable abdominal or inguinal masses.

At 1 month of age, ultrasound (April 2025, Children's Hospital of Chongqing Medical University, Chongqing, China) showed a testis-like structure in the right lower abdomen measuring ~2.8x1.8x1.0 cm. Within this structure, an abnormal echogenic lesion ~1.8x1.6x1.0 cm in size was identified, with a heterogeneous echotexture (mixed hyperechoic, isoechoic and small anechoic areas) and detectable blood flow on CDFI, findings consistent with an intra-abdominal undescended right testis containing a teratoma (Fig. 1C and D). Notably, tumor marker analysis showed an elevated serum ferritin level of 473 ng/ml (reference range, 28-365 ng/ml), while α-fetoprotein

Correspondence to: Professor Deying Zhang, Department of Urology, Children's Hospital of Chongqing Medical University, 136 Zhongshan 2nd Road, Yuzhong, Chongqing 400014, P.R. China
E-mail: zdy@hospital.cqmu.edu.cn

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(AFP; 531 ng/ml, reference range 24-4,800 ng/ml), β-human chorionic gonadotropin (<0.3 mIU/ml, reference range <10 mIU/ml), carcinoembryonic antigen (2.63 ng/ml, reference range <5.09 ng/ml), and neuron-specific enolase (35.3 ng/ml; reference range <35.6 ng/ml) were all within normal limits.

Laparoscopy confirmed an intra-abdominal right testis located ~1 cm from the internal inguinal ring. A 2.5x1.5 cm cystic-solid mass was identified in the mid-to-lower portion of the testis, with a thin, intact capsule and soft consistency; a small amount of residual testicular tissue was noted at the upper pole. The testis was delivered through a right lower abdominal incision. After incising the tunica albuginea, the mass was completely excised (Fig. 2A and B). A portion of both peritumoral tissue and the mass was sent for pathological examination. Intraoperative frozen section analysis (5-μm-thick sections, cut at -25˚C, stained with hematoxylin for 1 min and eosin for 15 sec at room temperature, and examined by light microscopy) revealed mature elements from all three germ layers, including abundant glial tissue and cartilage (Fig. 3A). Despite adequate mobilization of the spermatic cord, the testis could only reach the mid-inguinal level. In accordance with the family's request for testicular preservation and supported by a temporary clamping test, which demonstrated well-sustained perfusion without color change or signs of ischemia prior to spermatic vessel ligation, a single-stage Fowler-Stephens orchiopexy was performed (Fig. 2C). Final histopathology confirmed a mature teratoma containing glial and cartilaginous components, without immature elements or tumor infiltration in the adjacent tissue (Fig. 3B and C). At the 7 month follow-up, the infant remained well without complications.

Discussion

Pediatric testicular tumors have two incidence peaks. The first is before age 4, where one-third to half are benign, advocating for testis-sparing surgery. The second is in adolescents (15-18 years), where most germ cell tumors are malignant; the most common type is mixed germ cell tumor, making radical orchiectomy both diagnostic and therapeutic (6). Although teratomas account for ~10% of all pediatric testicular tumors, their occurrence in undescended testes is rare, especially when identified prenatally (7,8). The management of intra-abdominal testicular tumors identified postnatally in neonates generally follows Bolande's principle of the 'malignant transformation grace period', which suggests tumors originating in fetal or early neonatal life are often biologically benign and may undergo spontaneous regression or cytodifferentiation, typically before 6 months of age-reflecting a transient period during which malignant transformation is biologically restrained by the oncorepressive fetal milieu (9,10). For mature teratomas, simple tumor excision during the first month of life (or even antenatally) may be sufficient without the need for radical orchiectomy.

Prenatal diagnosis of intra-abdominal testicular tumors is typically achieved during routine fetal examinations aimed at evaluating testicular position. Under normal embryological development, the testes reach the area near the internal inguinal ring by the fifth month of gestation and begin descending through the inguinal canal by the seventh month. By the eighth month, they typically reach the scrotum. If the penis is clearly visible but the testes are not yet present in the scrotum, cryptorchidism should be suspected (10-12). This supports two potential mechanisms underlying the presence of germ cell tumors in undescended testes during the prepubertal period: i) A prenatal tumor that interferes with normal testicular descent; and ii) inherent cryptorchidism that predisposes the testis to malignant transformation (13-15). Accordingly, some researchers recommend prenatal ultrasonographic evaluation of fetal scrotal testes to improve diagnostic accuracy (11).

When a prenatally detected calcified abdominal mass appears distinct and clearly separated from the kidneys and spine, the differential diagnoses should include neuroblastoma, teratoma, meconium peritonitis, hamartoma and certain infectious masses (11,16,17). The concurrent presence of polyhydramnios, bowel dilatation and ascites is highly suggestive of meconium peritonitis (18). Hamartomas typically present as irregularly shaped masses, with sonographic characteristics that vary depending on the predominant tissue component (such as liver, lung and pancreas); they often present as heterogeneous, hypoechoic or hyperechoic lesions, occasionally accompanied by ascites (19).

Differentiating neuroblastoma from teratoma can be challenging as both may present as solid tumors with or without calcifications (20). Prenatally diagnosed neuroblastoma is the most common malignant solid tumor in the neonatal period; its typical prenatal ultrasonographic appearance is cystic (53%), followed by hyperechoic (31%) and mixed cystic-solid (16%) patterns (21). Up to 93% of fetal neuroblastomas originate in the adrenal gland, displacing the kidney inferiorly and laterally. Associated complications include metastatic lesions, hepatomegaly, fetal ascites and hydrops fetalis (22). Teratomas typically exhibit a thick capsule, well-defined margins and lack local invasion or distant metastasis (23). When diagnosis is uncertain, magnetic resonance imaging can provide additional information. Peripheral fat signals of the abdominal mass show high intensity on both T1- and T2-weighted images, with the mass center appearing hypointense on T1 and hyperintense on T2, indicating cystic and solid components (11). Accurate prenatal diagnosis facilitates timely referral to pediatric surgeons, enabling timely detection and treatment of the tumor to prevent complications such as torsion and malignant transformation (13,24).

Postnatal ultrasonography, particularly high-resolution sonography, offers superior visualization of cryptorchid testes and delineates their anatomical structure; therefore, re-evaluation immediately after birth is recommended. Normal testicular parenchyma exhibits a uniformly homogeneous echotexture, whereas testicular teratomas, similar to teratomas at other sites, appear as complex solid or cystic lesions with well-defined margins (25). Mature teratomas are typically not associated with abnormal elevations in tumor markers; AFP is predominantly secreted by yolk sac tumors and certain embryonal carcinomas, whereas increases in β-human chorionic gonadotropin and lactate dehydrogenase isoform 1 suggest endodermal sinus tumors, yolk sac tumors, immature teratomas, non-seminomatous germ cell tumors and choriocarcinomas (26). Ferritin levels may be physiologically elevated in neonates, making it an unreliable tumor marker (27). Although testicular teratoma may display characteristic

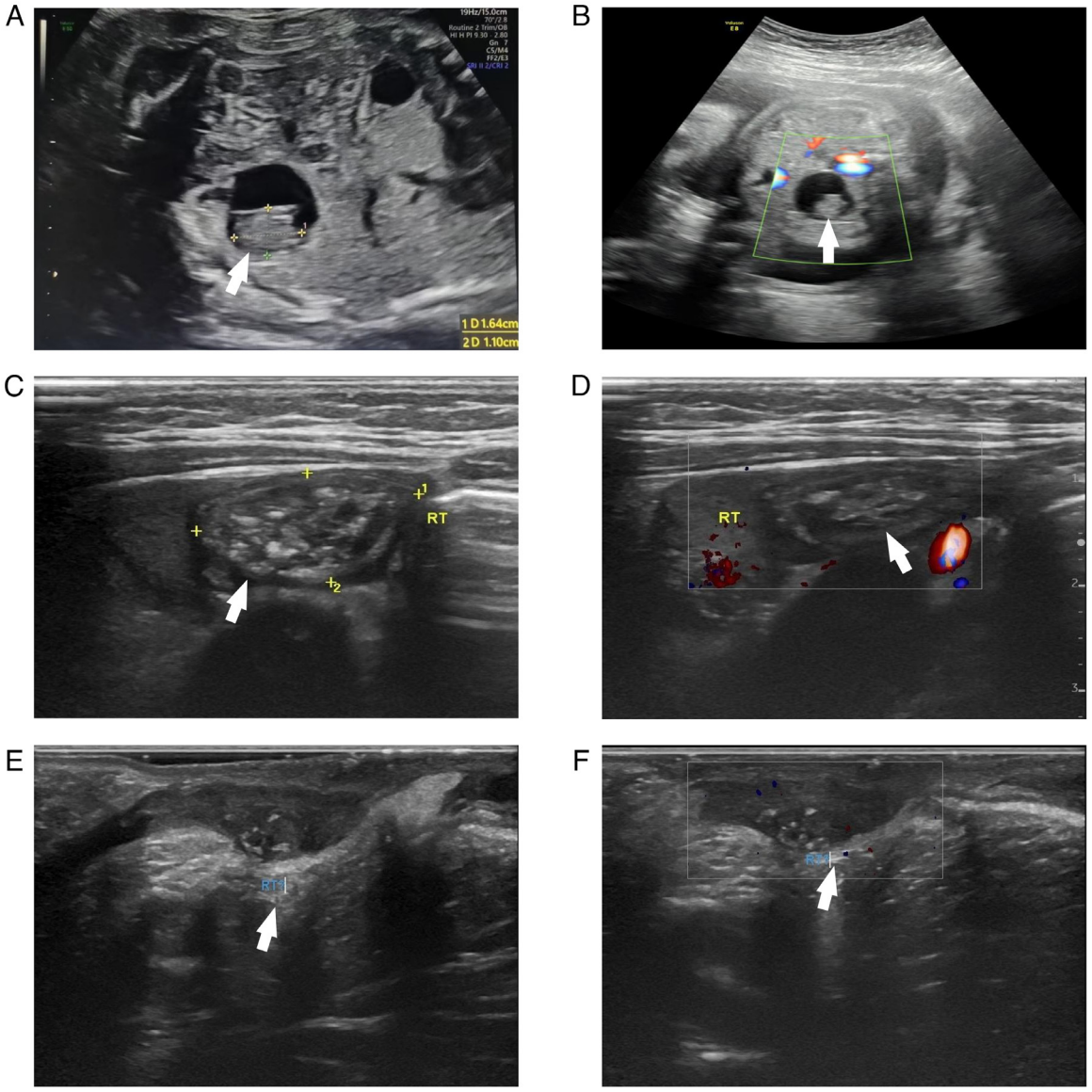

Figure 1. Prenatal, postnatal and postoperative sonographic findings of an intra-abdominal undescended testis with teratoma. Prenatal ultrasound at 28 weeks of gestation. (A) Transabdominal ultrasound showed a heterogeneous, well-circumscribed mass (arrow) in the right lower abdomen. (B) CDFI demonstrates scattered flow signals within the hyperechoic areas. Postnatal ultrasound at 1 month of age. (C) Ultrasound shows the persistence of the complex mass (arrow), consistent with an undescended testis associated with a teratoma. (D) CDFI reveals detectable blood flow signals within the medium-echoic areas. Follow-up ultrasound at 1 month after surgery. (E) Ultrasound shows a small testicular structure located above the scrotum. (F) CDFI shows scattered blood flow signals (arrows).

ultrasonographic features and laboratory tests can imply the histological subtype, intraoperative frozen section examination remains critical for guiding surgical management. A previous study has shown that testis-sparing surgery (TSS) guided by intraoperative frozen section analysis represents a viable alternative to orchiectomy (28).

In previously reported cases of prenatally diagnosed cryptorchidism with teratoma, mass excision was performed without attempting to preserve the testis (Table I), resulting in a partial loss of fertility potential (10,11,13,15,17,23-25). Almekaty *et al* (29) analyzed 42 post-pubertal male patients who had undergone unilateral orchiectomy and found that 45.2% developed postoperative azoospermia; contralateral testicular abnormalities were identified as an independent predictor and the azoospermic group showed significantly lower serum testosterone levels. Although the primary pathology may have contributed to this elevated rate, the detrimental effect of unilateral orchiectomy on fertility is well recognized. Given the excellent prognosis of mature teratomas and the uncertain developmental status of the contralateral testis in neonates, maintaining reproductive and endocrine function while ensuring complete tumor removal should be paramount (30,31). Furthermore, TSS can mitigate negative psychological effects during childhood development, which is essential for long-term mental health and social adaptation (32).

Although TSS aims to preserve endocrine and reproductive function, the capacity of the residual tissue warrants careful consideration. Most studies of radical orchiectomy report postoperative reductions in serum testosterone or increased compensated hypogonadism (33,34). By contrast, reports

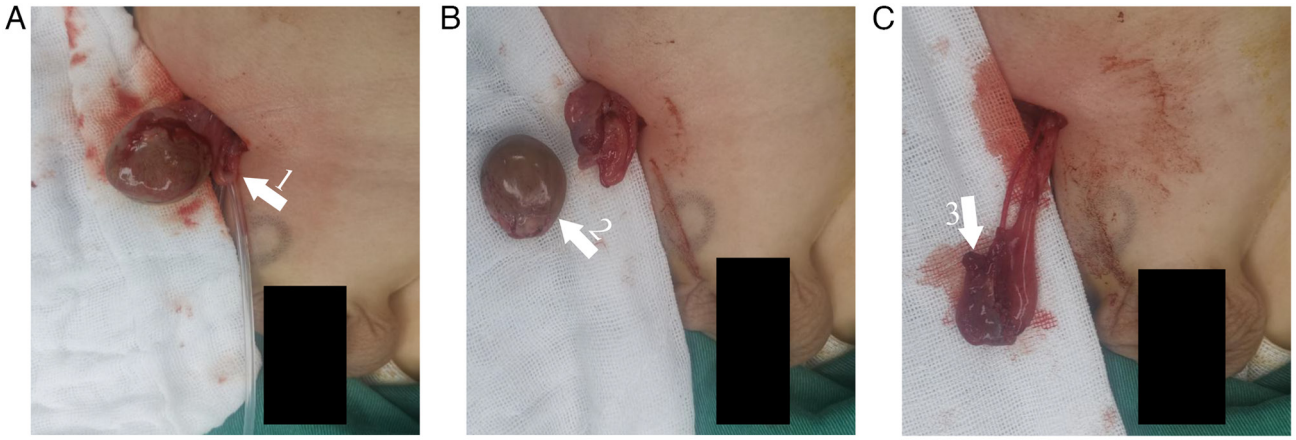

Figure 2. Right tumor excision and right laparoscopic Fowler-Stephens procedure. (A) Intact delivery of the testis and tumor via the testicular gubernaculum (arrow 1). (B) Completely excised tumor specimen (arrow 2). (C) After thorough mobilization, the testis reaches only the mid-inguinal region; spermatic vessels were ligated (arrow 3) before performing Fowler-Stephens orchiopexy.

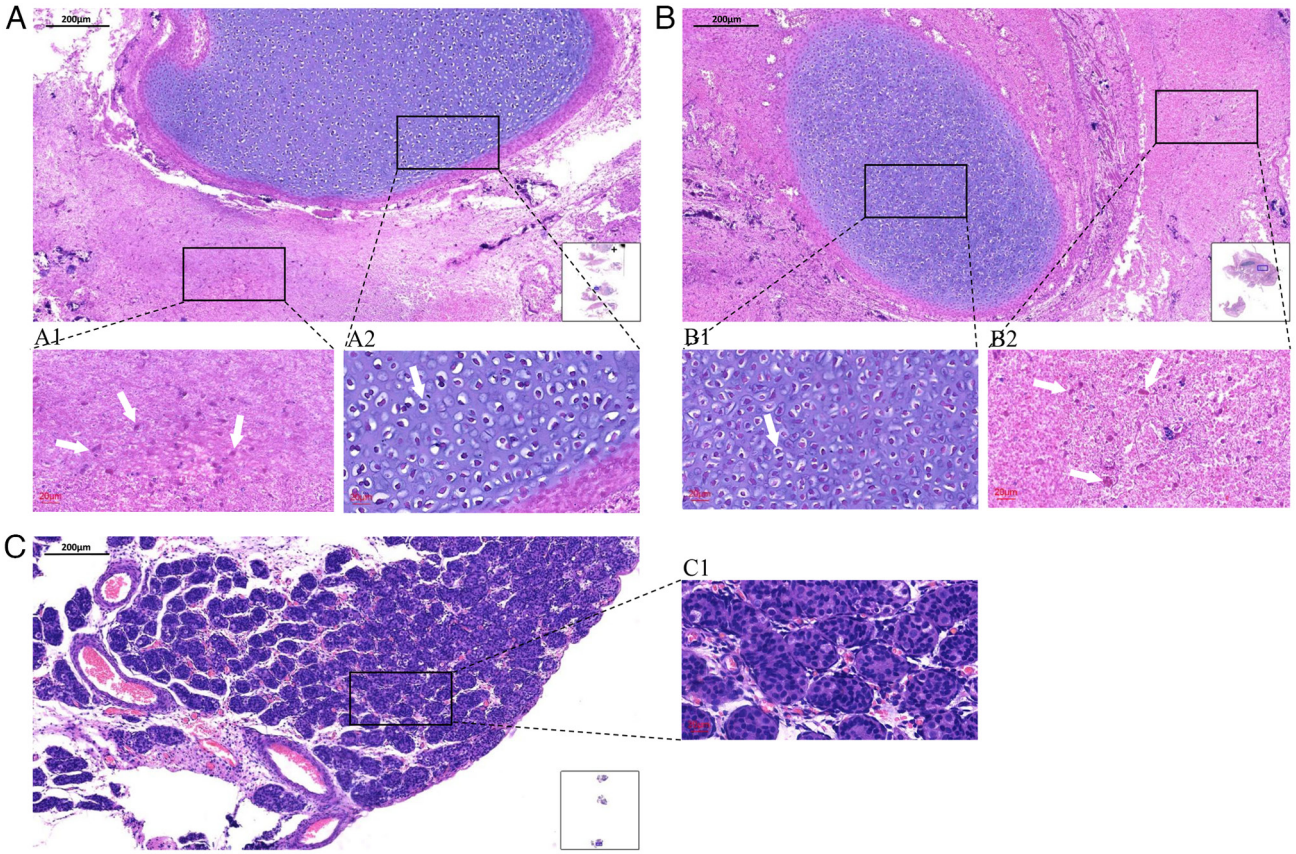

Figure 3. Intraoperative frozen section and postoperative H&E staining of tumor and peritumoral tissue. (A) Intraoperative frozen section showing mature elements from all three germ layers, including (A1) abundant glial tissue and (A2) cartilage cells. (B) H&E staining of tumor tissue demonstrating (B1) abundant cartilage and (B2) glial tissue. (C) H&E staining of peritumoral tissue showing testicular tissue without evident tumor involvement. (C1) magnified view.

show that small Leydig-cell remnants often maintain normal testosterone, so clinically overt hypogonadism after TSS is uncommon and overt testosterone deficiency is rare (35,36). Spermatogenesis is more vulnerable to the adverse effects of the testicular lesion; most patients already have abnormal semen parameters preoperatively. In the largest prospective series of benign lesions, oligo- and asthenozoospermia were common before surgery but did not significantly worsen postoperatively; however, radical orchiectomy was frequently associated with measurable declines in semen quality even without adjuvant therapy (37).

A potential limitation of the present study is the relatively short 7 months follow-up period, which, although adequate to detect early postoperative complications such as testicular atrophy (typically apparent within 3 months) may not completely exclude the possibility of late atrophy

Table I. Cases of prenatal diagnosis of intra-abdominal cryptorchidism associated with teratoma.

First author, year	GA at diagnosis, weeks	Laterality	Prenatal US characteristics at diagnosis	Size of mass at diagnosis, cm	Primary prenatal diagnosis	Mass size at delivery (cm)	Palpable abdominal mass	Surgical procedure and timing	Postoperative follow-up	(Refs.)
Mboyo *et al*, 1997	31	Left	Cystic and solid mass on left side of bladder, empty left scrotum	2.5x2.3	Fetal retroperitoneal teratoma/ cystic neuroblastoma	4.3x5.8	Mass palpable	Excision 13 days postpartum,	Persistent Grade III vesicoureteric reflux at 1 year	(25)
Shih *et al*, 1997	36	Right	Semisolid mass with central echogenic part in front of right fetal kidney and anterior to the bladder, undescended right testis	5x4x3	Fetal retroperitoneal tumor	5x4	Mass palpable	Excision by laparoscopy at 1 month	Normal outcome at 1 year	(10)
Siu *et al*, 2001	30	Right	Cystic intra-abdominal lesion with solid component anterior to the right kidney and superior to the bladder, empty scrotal sacs	2.4	Cystic teratoma in undescended testis	2.5x2.7x1.9	No mass	Excision by laparoscopy at 1 month	Normal postoperative outcome	(24)
Pramanik *et al*, 2011	27	Right	Intra-abdominal mass in right iliac fossa	2.1x1.9	Intra-abdominal mass	Unchanged	Mass palpable	Excision by laparotomy at 5 months	Normal postoperative outcome	(11)
Janda *et al*, 2014	22	Left	Mass adjacent to the bladder, subsequent onset of calcification in mass, absence of vascular flow	1.0x1.2	Intra-abdominal mass	1.8x1.3x1.4	No mass	Excision by laparotomy at 19 days postpartum	Normal postoperative outcome	(13)

Table I. Continued.

First author, year	GA at diagnosis, weeks	Laterality	Prenatal US characteristics at diagnosis	Size of mass at diagnosis, cm	Primary prenatal diagnosis	Mass size at delivery (cm)	Palpable abdominal mass	Surgical procedure and timing	Postoperative follow-up	(Refs.)
Youssef *et al*, 2016	32	Left	Single unilocular anechoic cyst with regular boundary located between the left kidney and urinary bladder in retroperitoneal position, scarcely vascularized small intracystic solid component, empty ipsilateral hemiscrotum	2.0x2.0 x2.2	Cystic teratoma in undescended testis	3	No mass	Excision by laparoscopy 3 days postpartum,	Normal outcome at 1 year	(23)
Arkar *et al*, 2016	36	Left	The lesion, located on the left lateral side of the urinary bladder, was round, cystic with a solid component, and contained a few discrete subcentimeter-sized calcifications	2.0x1.8	Cryptorchid testicular teratoma	2.0x1.9	No mass	Excision by laparotomy 3 days postpartum	Normal postoperative outcome	(17)
Yada *et al*, 2017	33	Right	A mass in the right lower quadrant	3.0x2.0	Intra-abdominal mass	3.0x2.0	Mass palpable	Excision by laparoscopy at 14 days postpartum	Normal outcome at 3 years	(15)

Table I. Continued.

First author, year	GA at diagnosis, weeks	Laterality	Prenatal US characteristics at diagnosis	Size of mass at diagnosis, cm	Primary prenatal diagnosis	Mass size at delivery (cm)	Palpable abdominal mass	Surgical procedure and timing	Postoperative follow-up	(Refs.)
Present report, 2025	28	Right	Single anechoic area with thin wall and good sound transmission located in the upper right bladder, showing irregular hypere-choic areas with small punctate strong echoes within	1.6x1.1	Cryptorchid testicular teratoma	2.8x1.8x1.0	No mass	Excision and primary Fowler-Stephens orchidopexy at 1 month	Normal postoperative outcome	-

GA, gestational age; US, ultrasound.

or tumor recurrence, although these are rare in mature teratomas (38,39). Nevertheless, the mid-term findings provide preliminary evidence supporting the safety and feasibility of this testis-sparing strategy. Long-term monitoring is ongoing and will be reported in future updates.

In the present case, guided by the adequacy of collateral circulation suggested on preoperative ultrasound and confirmed by intraoperative clamping test, a single-stage Fowler-Stephens orchiopexy was performed, although testicular descent was only achievable to the mid-inguinal canal. Single-stage Fowler-Stephens orchiopexy completes repair in a single operation, avoiding a second general anesthetic and repeat-procedure risks, shortening overall treatment time and limiting prolonged intra-abdominal exposure of the testis, however, certain studies report that, compared with two-stage FSO, single-stage procedures generally have lower testicular survival/success rates and higher atrophy rates (40,41). The present case highlights that primary testicular tumors can be detected early, even in the late fetal stage, and that TSS with selective removal of the teratoma is a feasible option in infants. Intraoperative frozen section analysis and histopathological examination of the peritumoral tissue provided critical evidence supporting the safe application of a testis-sparing procedure.

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Availability of data and materials

The data generated in the present study may be requested from the corresponding author.

Authors' contributions

HF, XL, SW, YH and GW analyzed clinical data, performed the literature review, and drafted the manuscript. DZ performed the surgical procedure. GP participated in data analysis. HF and DZ were responsible for revising the manuscript. HF and DZ confirm the authenticity of all the raw data. All authors have read and approved the final manuscript.

Ethics approval and consent to participate

Not applicable.

Patient consent for publication

Written informed consent for the publication of all clinical details and accompanying images was obtained from the patient's legal guardian in accordance with institutional and journal ethical guidelines.

Competing interests

The authors declare that they have no competing interests.

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