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LETTER TO THE EDITOR

Male Health

Sequential testis sparing surgery of simultaneous bilateral testicular tumors with different cell types in a Chinese infant: an uncommon presentation

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Dear Editor,

Patients of bilateral pediatric testicular tumors and bilateral testicular teratoma or yolk sac tumor (YST) were rarely reported, and there was still no standard therapy. However, synchronous bilateral testicular tumors with different histology were a quite rare situation. We reported the first case of successful radical orchiectomy in the left scrotum and testicular sparing surgery in the right testis sequentially. No atrophy or residual tumor in the right testis and no recurrence in the left scrotum were found after 7 years of follow-up. Ethical approval was obtained from the Institutional Review Board of Children's Hospital of Shanghai, which was affiliated to the Shanghai Jiao Tong University, School of Medicine, Shanghai, China. Informed consent was obtained from his parents.

A 19-month-old infant presented to Children's Hospital of Shanghai outpatient clinic with the chief complaint of a painless mass in the left scrotum in 2010. The bilateral testicular tumors were visualized by ultrasound and magnetic resonance imaging (Figure 1a). Serum alpha-fetoprotein (AFP) was 2122.3 ng l⁻¹ (reference range: 0–5.3 ng l⁻¹). The parents of the infant refused bilateral surgery simultaneously and the patient underwent left radical orchiectomy in the first stage. The mass on the left side were completely enucleated and final pathology confirmed that the specimen was diagnosed as yolk sac tumor (YST) (Figure 1d and 1e). The patient was considered the clinical Stage I YST and no further treatment was given. Follow-up ultrasonography at 1 month postoperatively revealed that no evidence of disease was observed in the left scrotum and testicular mass in the right scrotum did not show significant changes. The AFP levels were within normal range, meanwhile. The patient presented again 7 months later after the first surgery with the lump in the right testis. Ultrasound and magnetic resonance imaging showed the well-delineated, anechoic cystic mass located in the lower part of the right testis, the dimension of the right cystic mass being 0.6 cm × 0.5 cm × 0.6 cm in size, and the absence of left testis (Figure 1b and 1c). There were no enlarged lymph nodes in the retroperitoneum and inguinal region was normally defined. Serum AFP was 1.4 ng l⁻¹ and CEA was 2.74 ng l⁻¹ (reference range: 0–5.00 ng l⁻¹). Serum neuron-specific enolase (NSE) was 31.79 ng l⁻¹

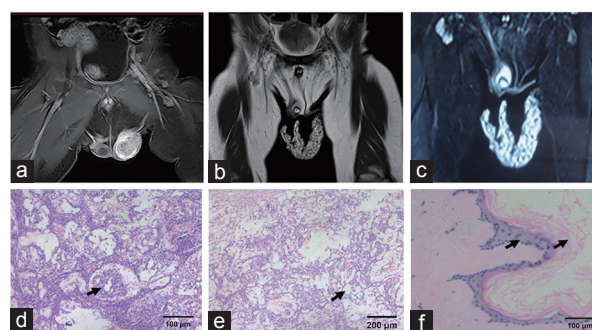


Figure 1: (a) Bilateral pediatric testicular tumors before surgery. (b) Seven months after left radical orchiectomy on T1WI. (c) T₂-trim MRI sequences visualize the right testicular tumor image after the first surgery. (d) Endodermal sinus-like in the left testicular mass (Schiller-Duval body). (e) Microcystic pattern in the left testicular mass. (f) Corneous flakes and squamous epithelial cells in the right testicular mass. T1WI:T1 weighted image; MRI: magnetic resonance imaging.

(reference range: 0–16.3 ng l⁻¹). The testis was explored by right inguinal incision with control of the spermatic cord, and an incision was made in the tunica albuginea; the mass was enucleated finely. The mass showed well-delineated, white, and round-like cystic finding intraoperatively. The frozen section analysis of biopsies was declared mature teratoma. The incision of tunica albuginea was examined for bleeding, closed with interrupted 6-0 polyglactin sutures, and placed back into the scrotum. Pathology report revealed cystic mature teratoma of the testis (Figure 1f). During the 7 years of follow-up, normal contour and no atrophy or residual tumor of the testis were observed after the second stage (ultrasound revealed the testis measuring 1.4 cm × 1.0 cm × 0.9 cm at 4 years and 1.6 cm × 1.1 cm × 1.0 cm at 7 years postoperatively).

In a previous study, 68% of the patients had benign tumor. Before puberty, the proportion of benign tumors was 77%. After the age of 13 years, the proportion of benign tumors decreased to 38%.¹ Previous study confirmed the relative frequencies of the histologic types of testicular tumor in boys.² YST was the most common type and followed by teratoma. Choriocarcinomas and seminomas were exceedingly uncommon. During boyhood, YST was most common in infancy and in child under 2 years old especially. YST often characterized by hypervascular solid mass in the ultrasonography and the elevation of serum AFP level.³ It was usually palpated and presented as an

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Table 1: Published studies that describe bilateral testicular yolk sac tumor in children

Study number	Reference	Patient age (month)	Pathologic diagnosis	Clinical stage	Treatment
1	Royal <i>et al.</i> , 1994 ⁷	24	Right: mixed germ cell tumor (95% YST) Left: teratoma	Not available	Bilateral orchiectomy
2	Yokomizo <i>et al.</i> , 2001 ⁸	19	Right: YST Left: Y	Stage I	Bilateral orchiectomy
3	Luo <i>et al.</i> , 1998 ⁹	7	Right: teratoma Left: YST	Stage I	Bilateral orchiectomy

YST: yolk sac tumor

asymptomatic testicular enlargement. Teratoma was the most popular benign tumor in prepubertal children.²

The treatment of testicular tumors in childhood had evolved during the last several decades. Until 1980, the popular opinion was that almost all tumors were malignant, and the orchiectomy should be proposed.¹ Bilateral orchiectomy had several negative implications on endocrine system in addition to infertility. With the progressing of ultrasound, the sensitivity in ruling out the mass was near 96.6% and could indicate the exact tumor position in the testis.⁴ Intraoperatively, frozen sections were highly accurate for differentiating malignant from benign tumor. Several studies reported that there was no discordance between frozen section and definitive histology.⁵ Consequently, testis sparing surgery (TSS) was proposed among pediatric surgeons. Shukla *et al.*⁶ reported their experiences of TSS for teratoma. There was no recurrence reported after TSSs, and patient's outcomes of long-term follow-up were without atrophy or orchialgia.

Cases of bilateral pediatric testicular tumors and bilateral testicular teratoma or YST were rarely reported. We learned from literature that there were limited reports of bilateral pediatric testicular yolk sac tumors (Table 1). Royal *et al.*⁷ reported a 2-year-old male child with synchronous bilateral testicular tumors received bilateral radical inguinal orchiectomy. The final pathology was mixed with germ cell tumor (95% YST) of the right and mature teratoma of the left. Yokomizo *et al.*⁸ reported a 19-month-old infant of bilateral YSTs accepting sequentially radical orchiectomy during 14 months. Luo *et al.*⁹ reported a 7-month-old case of synchronous YST in the left testis and mature teratoma in the right treated with bilateral orchiectomy in 1998. Koski *et al.*¹⁰ reported a patient treated using bilateral testicular sparing surgery. The examinations postoperatively showed good residual parenchyma bilaterally, symmetrical testicles with minimal postoperative changes, and good blood flow. Changes in the maternal hypothalamic-pituitary-adrenal (HPA) axis may lead to long-term adverse health outcomes for the offspring, and maternal endocrine dysregulation can impact child neuroendocrine-immune (NEI) function by altering the sensitivity of inflammatory immune processes to the inhibitory effects of cortisol.^{11,12}

To our knowledge, this was the first case of bilateral testicular tumors with different cell types managed with radical orchiectomy and successful testis sparing surgery sequentially. Examination after long-term follow-up postoperatively showed good result. For this patient, one-stage surgery with orchiectomy and TSS may be an alternative choice.

AUTHOR CONTRIBUTIONS

XXL carried out the studies, participated in the design of the study, performed the surgery, and participated in the draft of the manuscript. FC, SLL, YCH, YQL, YC, and HZS participated in study design and coordination and helped to draft the manuscript. HX conceived of the study and provided the academic guidance. All authors read and approved the final manuscript.

COMPETING INTERESTS

All authors declare no competing interests.

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