

Wilms' Tumor Metastatic to Bilateral Testes at Presentation: Case and Review of the Literature

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A 7-year-old male with bilateral palpable testicular masses was found to have metastatic stage IV Wilms' tumors associated with both left renal and lung lesions. The patient was treated successfully with testicular-sparing bilateral partial orchiectomies, radical nephrectomy, chemotherapy, and radiation, with 8 years free of recurrence. This is the only reported case of bilateral testicular tumors from metastatic Wilms' tumor that were present at the initial presentation. A review of the literature of all previous reported cases of metachronous or related testicular metastatic Wilms' tumors is reported. *UROLOGY* 79: 899–901, 2012. © 2012 Elsevier Inc.

Wilms' tumor is the most common primary malignant renal tumor of childhood. It is estimated that 12% of Wilms' tumor patients will have evidence of metastases at initial presentation and 80% of these will be to the lung.¹ Specifically, testicular or paratesticular metastasis of Wilms' tumor is extremely rare, with only 6 reported cases in the literature, to our knowledge. This is the first report of bilateral testicular metastasis of Wilms' tumor at initial presentation.

CASE REPORT

A 7-year-old male was referred for bilateral testicular enlargement, a 25-cm left renal mass, and multiple pulmonary metastases concerning for Wilms' tumor (Fig. 1A,B). Bilateral partial orchiectomy specimens stained positive for WT1 protein. After pathologic Wilms' tumor diagnosis, neoadjuvant cyclophosphamide, vincristine, and doxorubicin were given. The patient concomitantly received radiotherapy to the entire lung, abdomen, and bilateral testes with a resultant 63% reduction in size of the left renal mass over 2 months. Subsequent abdominal exploration with negative contralateral renal biopsy, left radical nephrectomy, and retroperitoneal lymph node sampling was consistent with stage IV Wilms' tumor. Seven rounds of adjuvant chemotherapy were administered over 7 months.

Follow-up over 8 years has included serial imaging and management of bilateral hydroceles at which time suspicious lesions of tunica albuginea were excised and showed no malignancy. He has since exhibited sexual develop-

ment to Tanner stage 5 without further findings of residual or *de novo* testicular masses. However, he exhibits bilateral testicular atrophy with borderline primary hypogonadism, which is currently managed medically.

COMMENT

Review of the literature finds 6 reports of testicular metastases that were not present at the time the primary Wilms' tumor was diagnosed (Table 1).²⁻⁷

The rare spread of Wilms' tumor to the testicles has been theorized to be hematogenous, retrograde by direct extension, and/or transcoelomic if a patent processus vaginalis is present.⁶ There is likely not a single mechanism that accounts for the metastases seen in all of these cases, although the management has been approached in a similar fashion. We would postulate that our case was caused by hematogenous metastatic spread given the concomitant pulmonary disease. Partial orchiectomies were performed because both testicles were involved and there was normal testicular tissue that was uninvolved with the masses. Partial orchiectomy has shown to be of benefit to the patient when compared with radical surgery by reducing the need for androgen replacement, lessening psychological stress, and preserving fertility, with a durable cure rate in germ cell tumor patients.⁸ However, hypogonadism is a long-term sequela after irradiation, the severity of which is dose-dependent,⁹ for most adult survivors of childhood cancers.¹⁰

CONCLUSIONS

This is the first report of metastatic Wilms' tumor to bilateral testis at initial presentation. A testicular-sparing approach with bilateral partial orchiectomies, radical nephrectomy, chemotherapy, and irradiation resulted in

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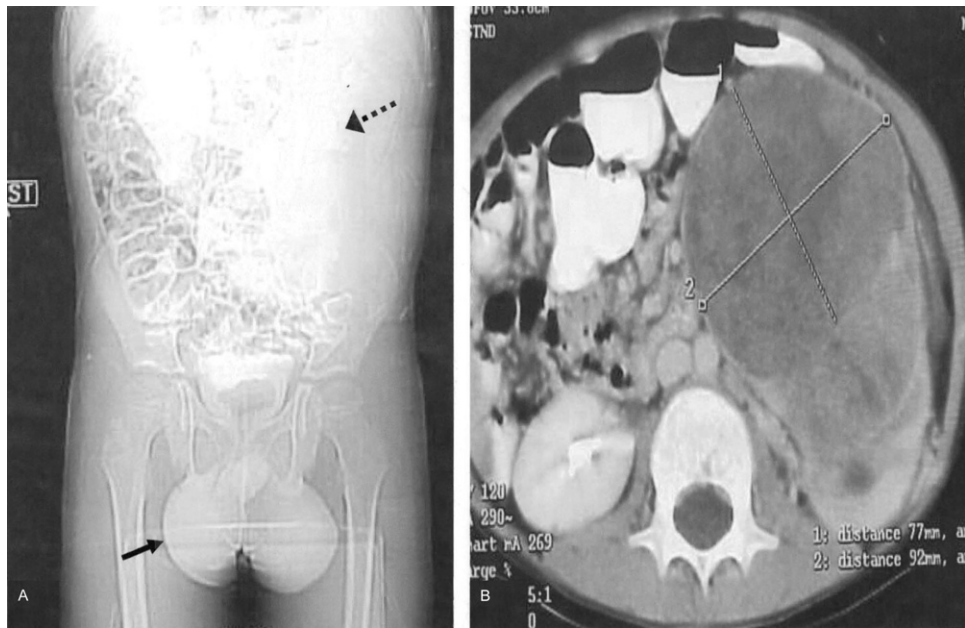


Figure 1. (A) Kidney-ureter-bladder x-ray at presentation showing testicular enlargement (solid arrow) and left-sided abdominal mass (dotted arrow). (B) Computed tomography image showing abdominal mass arising from the left kidney.

Table 1. Literature review of previous asynchronous cases of Wilms' tumor testicular metastases

Citation	Location			Timing	Testicular Metastasis Treatment			Follow-up
	Primary Wilms' tumor	Testis			Orchiectomy	Chemo	XRT	
Dew (1928) ²	Left	Left		6 mo after nephrectomy	(+)	(-)	(-)	Died of metastatic disease in 4 mo
De Camargo et al (1988) ³	Right	Left (epididymis)		7 mo after nephrectomy	(+)	(+)	(+)	4 mo
Sauter et al (1990) ⁴	Left	Left		3 mo after nephrectomy	(+)	(+)	(+)	NA
Tröbs et al (2002) ⁵	Right	Right		Hydrocele at presentation, but orchiectomy 11 mo after nephrectomy	(+)	(+)	(-)	8 y
Aydin et al (2006) ⁶	Left	Right (paratesticular)		Hydrocele at presentation, but orchiectomy >6 mo after nephrectomy	(+)	(+)	(+)	22 mo
Daher et al (2010) ⁷	Right	Bilateral		Left testicular mass, right hydrocele >3 mo after nephrectomy; biopsy revealed bilateral Wilms' tumor	(-) Undecided at time of report	(+)	(+)	NA

XRT = external beam radiation therapy.

successful Wilms' tumor treatment and 8 years free from recurrence, with hypogonadism as a late sequela, however. For future cases, we would urge the diligent and

aggressive approach to clinically apparent hydroceles involved with abdominal malignancies. Furthermore, the risk of hypogonadism should be considered and discussed

with the patient as a likely consequence of chemotherapy and/or testicular irradiation.

References

1. Davidoff AM. Wilms' tumor. *Curr Opin Pediatr.* 2009;21:357-364.
2. Dew H. Sarcomatous tumors of the testicle. *Surg Gynecol Obstet.* 1928;46:447-458.
3. de Camargo B, Pinus J, Lederman H, et al. Intrascrotal metastasis from Wilms' tumor. *Med Pediatr Oncol.* 1988;16:381-383.
4. Sauter ER, Schorin MA, Farr GH Jr., et al. Wilms' tumor with metastasis to the left testis. *Am Surg.* 1990;56:260-262.
5. Tröbs RB, Friedrich T, Lotz I, et al. Wilms' tumour metastasis to the testis: long-term survival. *Pediatr Surg Int.* 2002;18:541-542.
6. Aydin GB, Ciftçi AO, Yalçın B, et al. Paratesticular metastasis from Wilms tumor associated with a hydrocele. *Pediatr Blood Cancer.* 2006;47:97-99.
7. Daher P, Raffoul L, Riachy E, et al. Bilateral testicular metastasis from Wilms' tumor. *Eur J Pediatr Surg.* 2010;20:415-416.
8. Zuniga A, Lawrentschuk N, Jewett MA. Organ-sparing approaches for testicular masses. *Nat Rev Urol.* 2010;7:454-464.
9. Kinsella TJ, Trivette G, Rowland J, et al. Long-term follow-up of testicular function following radiation therapy for early-stage Hodgkin's disease. *J Clin Oncol.* 1989;7:718-724.
10. Romerius P, Ståhl O, Moëll C, et al. Hypogonadism risk in men treated for childhood cancer. *J Clin Endocrinol Metab.* 2009;94:4180-4186.