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Case Report

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Bilateral Wilms' Tumor Metastasis to Right Spermatic Cord

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ABSTRACT

Wilms' tumor (WT) is the most common primary renal tumor in children. Common sites of metastases are lungs, liver and regional lymph nodes. Testicular and paratesticular metastasis due to WT have been reported but it is extremely rare. We report a 33-month -old male with bilateral WT and metastasis to right spermatic cord.

KEY WORDS: Bilateral Wilms' tumor, Metastasis, Spermatic cord, Paratesticular

INTRODUCTION

Wilms' tumor (Nephroblastoma) is the second common abdominal malignancy and most common malignant renal tumor in children. Often it presents with asymptomatic mass and is usually discovered by parents incidentally while bathing or dressing. Involvement of both kidneys either at presentation or subsequently were seen in 5% to 7% of cases, but the majority of bilateral WT present with synchronous disease at diagnosis.[1] Testicular and paratesticular metastasis due to WT have been reported but it is extremely rare.[2, 3] Just 8 case reports were found in our literature review. Here we report a 33-month -old male with bilateral WT and metastasis to right spermatic cord.

CASE REPORT

The patient is a 33-month-old male whom his mother had discovered right flank mass at the time of dressing one week before admission. Abdominal examination revealed right and left flank mass and mild distention. His physical examination was otherwise unremarkable except blood pressure 130/90 mmHg. The patient was one of two identical twins. He had not congenital anomalies and the other twin was normal. Two months earlier, he had undergone herniorrhaphy for right inguinal hernia. Ultrasound of the abdomen revealed a 73×79 mm and 75×78mm heterogeneous mass without calcification in the right and left kidney, respectively and lymph node in Para-aortic area with 25mm in diameter. Contrast-enhanced computed tomography (CT) scan of the abdomen findings were similar to that of ultrasound without evidence for inferior vena cava and renal vein involvement. Chest CT scan did not show evidence of metastases. Then open biopsies from both kidneys were performed. The histopathology showed infiltration of both kidneys with blastemal cell predominant component, vascular invasion in the tumoral tissue and favorable histology without anaplasia (Figure. 1A).

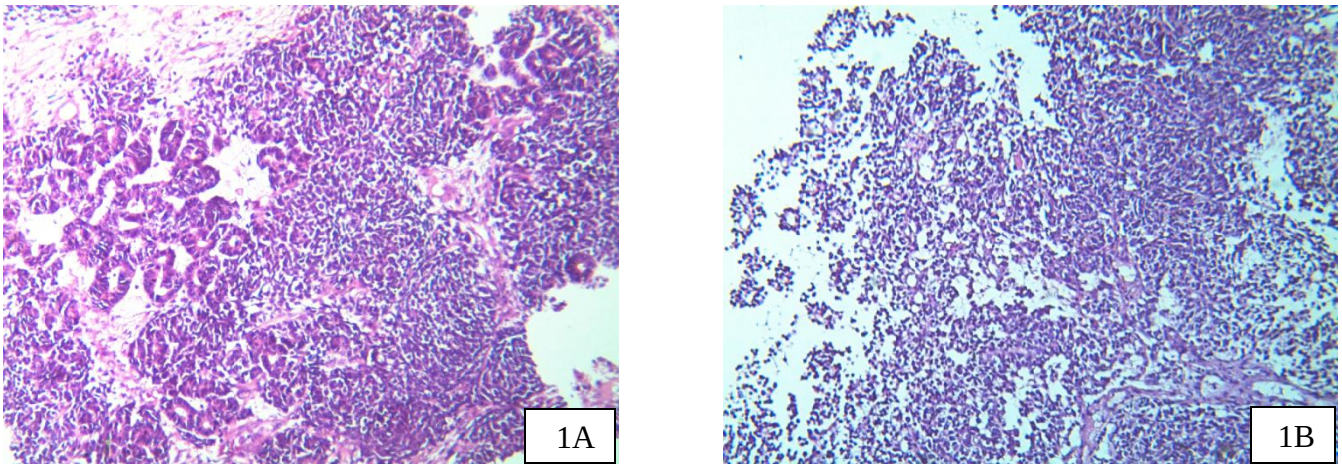

Figure 1: 1A) Histopathological examination showing the neoplastic cells (blastemal component is predominant) from right kidney. **1B)** Spermatic cord biopsy illustrating infiltration by neoplastic cells of Wilms tumor

Para-aortic lymph node biopsy was positive for tumoral involvement. Complete blood count with differential, liver and renal function tests were all within normal range. Erythrocyte sedimentation rate was 89mm/hr. Diagnosis was consistent with stage V favorable-histology Wilms' tumor and treatment was initiated with chemotherapeutic agents including vincristine, actinomycin D, and doxorubicin. Eight weeks following treatment the tumor size of both sides decreased more than 50% and ultrasound demonstrated a 28×40mm and 34×36mm tumor mass in the right and left kidney, respectively. Blood pressure returned to normal range. He underwent right radical nephrectomy and immediately received abdominal irradiation (1080 cGy in 10 fraction dose). Chemotherapy continued with previous drugs. Two months later, ultrasonography revealed a mass measuring 69×69mm in right renal fossa, but tumor size in the left side was without any significant change. Subsequently, the chemotherapy regimen changed to ifosfamide, crboplatin and etoposide (ICE). Following the fourth cycle of ICE, at routine physical examination a right scorotal mass on spermatic cord was noted. Ultrasound revealed a spermatic cord mass measuring 1×1.5×2cm without testicular involvement. The patient then underwent exicisional biopsy and histologic examination suggested WT metastases to spermatic cord (Figure. 1B). Two weeks later, lungs CT scan showed multiple nodules and on the ultrasound examination, liver metastases and widely disseminated tumor spread in abdomen were identified. Chemotherapy with ICE regimen continued but unfortunately, he died 2 months later owing to progressive disease and distant metastases.

DISCUSSION

The most frequent sites of metastases due to WT are lungs, liver and regional lymph nodes. Rarely metastases are seen in other sites. WT metastases to testicular or partesticular is extremely rare and only eight cases have been reported in the literature so far (Table 1).

Leukemia and lymphoma are the most common malignancy that metastases to the testis.[2-4] In rare instances, WT may arise from extra renal regions including scrotom.[5] Different mechanisms have been proposed for testicular and paratesticular metastases due to WT such as direct tumor cell spread from abdomen along the patent processus vaginalis by gravity, spermatic vein involvement by tumor cells, lymphatic or hematogeneous.[2, 6]

The first case of testicular metastasis due to WT was reported in 1928 by Dew H in a 27-month-old boy. Metastasis to left testis in this case occurred 6 months after nephrectomy with direct spread of the tumor cells through left spermatic vein.[7] The last case had reported in 2011 by Kajbafzadeh et al., in a 5-year-old boy. In this

patient, metastases to the right testis occurred 8 months after ipsilateral nephrectomy and the mechanism of this was right spermatic cord involvement along processsus vaginalis.[2] A review of previous reports demonstrated that in four patients of them left testis involvement existed, Whereas, only in 3 cases the involved kidney were on the left side[7-10] and four patients had right testis involvement while only in 3 cases kidney involvement were in the same side.[2-4, 11] In all of these case reports, unilateral WT was present and our case is the first report of bilateral WT with paratesticular metastasis. In the present case, 8 months after diagnosis metastasis to the spermatic cord was occurred. Possible mechanism of this is patent processus vaginalis. However, hematogeneous route can be considered. Although, testicular and paratesticular tumor are rare in children, any solid scrotal mass should be considered malignant until proved otherwise especially when malignant neoplasm exists outside the testis. Therefore, we recommended routine testes examination in patients with WT during treatment and follow-up period.

Table1. Reported Patients with Testicular and/or Paratesticular Metastasis due to WT

Author/ Year of report	Age of patient	Side of Wilms tumor	Scrotal metastases	Metastasis to	Outcome
Kajbafzadeh AM et al., 2011	5- year-old	Right	Right	Spermatic cord	Lost F/U
Aydin GB et al., 2006	3- year-old	Left	Right	Tunica vaginalis and albuginea	NED
Trobs RB et al., 2002	3 –year-old	Right	Right	Testis	NED
Ouattara K et al., 1992	3-year-old	Right	Right	Testis, paratesticular tissues	NA
Sauter ER et al., 1990	13-month-old	Left	Left	Spermatic cord, veins, testis	NA
De Camargo B et al., 1988	5-year-old	Right	Left	Epididymis	Lost F/U
Yadav K et al., 1977	9-month-old	Left	Left	Spermatic cord	NED
Dew H, 1928	27-month-old	Left	Left	Spermatic cord, epididymis, testis	died

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