

Wilm's tumor during pregnancy: report of laparoscopic removal and review of literature

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REHMAN J, CHUGHTAI B, GURU K, KHAN SA, ADLER HL, MILLER F. Wilm's tumor during pregnancy: report of laparoscopic removal and review of literature. *The Canadian Journal of Urology*. 2008;15(4):4180-4183.

tumor among children. Few cases of Wilm's tumor have been reported in women during pregnancy. The authors present a rare case of a pregnant female, who underwent laparoscopic excision of a large Wilm's tumor. The authors have also provided a review of the current literature.

Wilm's tumor, or nephroblastoma, is a common renal

Key Words: Wilm's tumor, pregnancy, review

Introduction

Although a common renal tumor in children, Wilm's tumor has been reported in less than 200 cases worldwide.¹⁻¹⁰ Children tend to present with a palpable abdominal mass, while flank pain and/or gross hematuria tend to be the presenting complaints in adults. Approximately 20% of cases are metastatic at the time of diagnosis.¹¹ Pregnancies complicated by Wilm's tumors are extremely rare. In fact, less than

seven such cases have been reported¹⁻⁷ within the last 20 years. Furthermore, the majority of these cases were stage III or higher. The incidence in pregnant and non-pregnant groups is similar, despite the relative immunosuppression associated with pregnancy. Pain is the second most common presenting symptom in pregnant patients (50%), as compared with 41% of the non-pregnant population and is almost universally associated with a palpable mass. Walker et al suggested the pain reported by patients may be due to the mild hydronephrosis that accompanies pregnancy.¹² Flank pain during pregnancy is more commonly attributed¹³ to urinary calculus, pyelonephritis, or uterine irritability. Although rare, hematuria in pregnancy may be the first indication of Wilm's tumor, especially when presented with a palpable mass.

Accepted for publication March 2008

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Case report

A 36-year-old Hispanic female, with a history of ectopic pregnancies and bilateral fallopian tubal surgical repairs, experienced an episode of gross hematuria during the third trimester, following an in vitro fertilization (IVF) pregnancy. An ultrasound of the kidneys revealed a large heterogeneous left renal mass, measuring approximately 18 cm x 9 cm, which was confirmed by MRI, Figure 1. The patient was scheduled for an elective nephrectomy 1 week following delivery. However, 5 days after a normal spontaneous vaginal delivery of a healthy female infant, the patient presented complaining of a 2-day history of left flank pain. Physical examination revealed left CVA tenderness and a palpable left subcostal mass. An abdominal CT scan confirmed a large heterogeneously enhanced mass in the left kidney, Figure 2. The patient underwent a laparoscopic left radical nephrectomy with no complications. Histopathological examination revealed Wilms' tumor, with a very dominant tubulopapillary pattern, although other components were present, Figure 3. The immunocytochemistry staining excluded primitive neuroectodermal tumor (PNET) and/or other entities confused with adult Wilms' tumors, Figure 4. Surgical margins were negative. The National Wilms' Tumor Study Group concurred, and graded the tumor as stage II because of necrosis and vascular invasion. The patient underwent 18 weeks of chemotherapy, consisting of vincristine and dactinomycin.



Figure 1. MRI scan showing the huge enhancing renal mass.

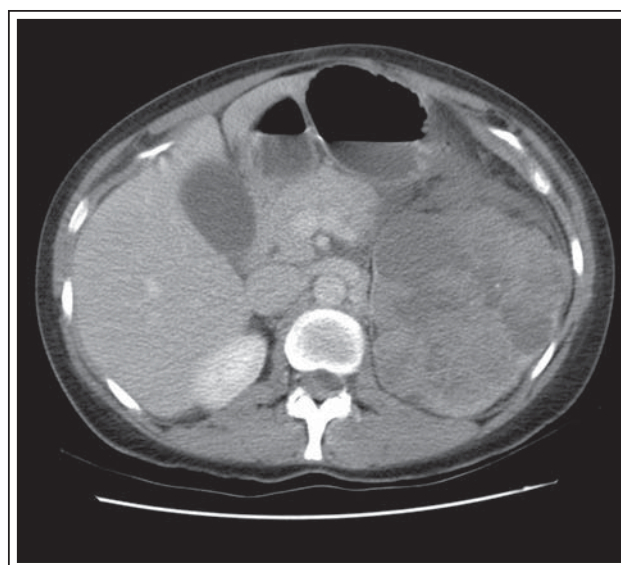


Figure 2. CT scan of kidney showing the huge enhancing renal mass.

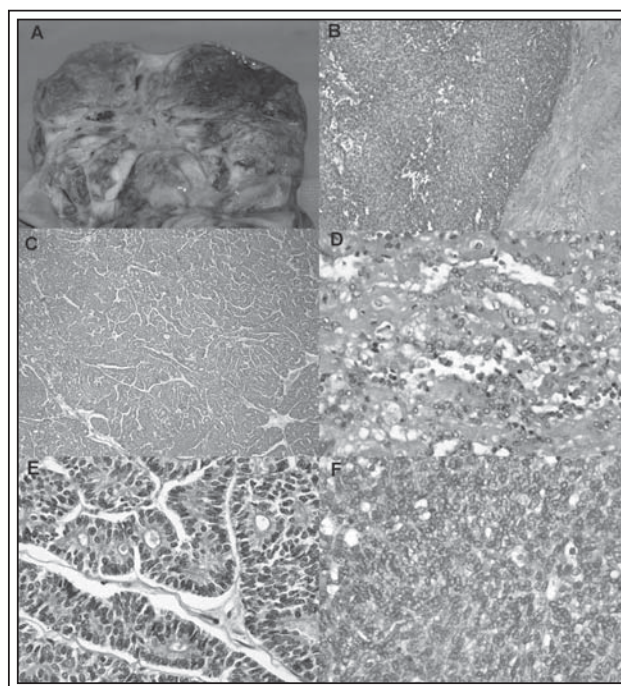


Figure 3. Pathology. A) Gross photograph of bivalved tumor involving the entire upper portion of the photograph. B) Blastema portion of tumor, low power, showing demarcation from capsule (H&E X4). C) Tubulopapillary component, which made up over 75% of tumor (H&E X6). D) Area of necrosis with poorly differentiated tumor element (H&E X200). E) Epithelial component with well-differentiated tubules (H&E X400). F) Blastema (H&E X200).

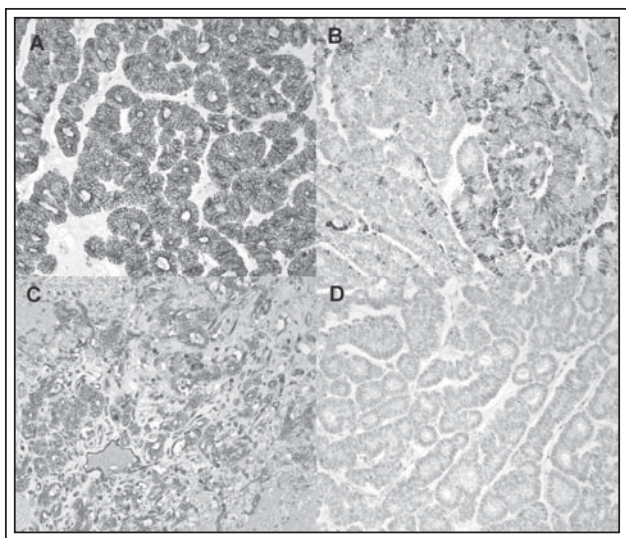


Figure 4. Immunostaining (immunoperoxidase detection system). A) Pancytokeratin was strongly positive. B) Vimentin was focally positive. C) CD99 (Ewing's sarcoma marker) was positive in a few very small areas. D) Neuron specific enolase, leukocyte common antigen (CD45), S-100, synaptophysin, and muscle actin were not detected.

Discussion

The diagnosis of an adult Wilm's tumor, based on the findings of currently available diagnostic methods, is considered only presumptive until the surgical specimen can be studied. The application of therapeutic protocols for pediatric, as well as adult patients, has not obtained the same percentage of cure or prognosis. A large multi-center study is warranted to confirm the foregoing. Misdiagnosis, PNET, synovial sarcoma, and other malignancies misclassified as Wilms' tumors contribute to worse prognoses in adult patients. In this case, other possibilities were excluded through differential diagnosis.

The non-invasive nature of ultrasound, absent radiation, renders it an ideal initial imaging for screening the maternal urinary system. Ultrasound has 85% sensitivity in detecting renal masses greater than 3 cm, which is equal to that of IVP with tomography. Ultrasound is more sensitive than IVP (82% versus 52%) for detecting lesions between 2 cm and 3 cm. Because of the aforementioned advantages, Smith et al¹⁴ suggest, the use of ultrasound to screen the urinary system of pregnant women with hematuria, recurrent or refractory urinary tract symptoms, flank pain, or a palpable flank mass. Ultrasound also differentiates between solid or cystic masses.

There is no proven safe threshold dose of radiation exposure to the fetus. At an average dose of 10 mGy, similar to the exposure of a limited intravenous pyelogram, statistical analysis revealed¹⁵ the following relative risk values: 1.6 for leukemia, 3.2 for childhood solid cancers, and overall risk of 2.4 for all childhood malignancies. Although IVP and abdominal CT are the standard diagnostic modalities used in evaluating a renal mass in a non-pregnant patient, alternative methods are appropriate during pregnancy. The MRI urogram has replaced intravenous urograms and nuclear scans, as risks and effects of intravenous contrast agents and radiopharmaceuticals crossing the placenta are unknown. In cases of hemorrhage or tumor infarction of large or small tumors, when nephrectomy is declined by the patient, angiography may be used therapeutically for embolization.¹⁶ It may also be used to better define vascular anatomy, and is dependent on the tumor's extensiveness and the surgeon's experience. It is not routinely recommended.

Radical nephrectomy is the standard surgical approach for treating Wilm's tumor during pregnancy. There have been no reported pregnancy losses due to radical nephrectomy. For a mass diagnosed in the second trimester, it is advisable to continue the pregnancy until 28 weeks, test for fetal lung maturity and then perform radical nephrectomy. At 28 weeks gestation, if the fetus were to be delivered, neonatal survival rates of over 90% can be expected. Delivery of the infant would only be necessary if labor ensues. If the diagnosis is made near term, surgery can be safely postponed until after delivery.¹⁷

A simultaneous cesarean section is not routinely recommended during radical nephrectomy, nor is labor automatically induced, since the kidney may be delivered through a flank incision. In cases complicated by severe hypertension, spontaneous tumor rupture, heavy bleeding, or a necessary transabdominal approach complicated by uterine size, a cesarean section should be performed initially.¹⁸ Cesarean section or induction of labor may also be appropriate if the fetus has reached age of viability.

The advent of laparoscopy as a treatment modality for renal masses during pregnancy is an interesting option. Preliminary results of laparoscopy during pregnancy are encouraging,¹⁹ but multi-institutional prospective studies are needed to determine its efficacy and safety. □

References

1. Ney C, Posner AC, Ehrlich JC. Tubular adenoma of the kidney during pregnancy. Report of a patient and angiographic studies. *Obstet Gynecol* 1971;37:267.

2. Borisova-Khromenko VM, Maletin AG, Sukovatitsin AM. Wilms' tumor in pregnant women. *Akush Ginekol (Mosk)* 1981;55.
3. Davis LW. Wilms' tumor complicating pregnancy: report of a case. *J Am Osteopath Assoc* 1987;87:306.
4. Bozeman G, Bissada NK, Abboud MR et al. Adult Wilms' tumor: prognostic and management considerations. *Urology* 1995;45:1055.
5. Swierz J, Stawarz B. Wilms' tumor in a 22-year old woman during pregnancy. *Pol Tyg Lek* 1994;49:198.
6. Singh NP, Anuradha S, Choudhry D et al. An unusual tumour in a post-partum woman. *Postgrad Med J* 1999;75:61.
7. Wynn T, Ruymann FB, King DR et al. Second pregnancy-associated Wilms tumor 16 years after the first one. *Med Pediatr Oncol* 2003;40:120.
8. Eserksy G. Wilms' tumor in the adult: Review of literature and report of three additional cases. *J Urol* 1947;58:397.
9. Livermore G. Wilm's tumor in adult; report of ten cases. *J Urol* 1953;70:141.
10. Feller N, Kenda M. Adult Wilm's tumor of the kidney. *Dapim Refuim* 1967;26:129.
11. D'Angio GJ, Evans A, Breslow N et al. The treatment of Wilms' tumor: results of the Second National Wilms' Tumor Study. *Cancer* 1981;47:2302.
12. Walker JL, Knight EL. Renal cell carcinoma in pregnancy. *Cancer* 1986;58:2343.
13. Loughlin KR, Ker LA. The current management of urolithiasis during pregnancy. *Urol Clin North Am* 2002;29:701.
14. Smith DP, Goldman SM, Beggs DS et al. Renal cell carcinoma in pregnancy: report of three cases and review of the literature. *Obstet Gynecol* 1994;83:818.
15. Harvey EB, Boice JD Jr, Honeyman M et al. Prenatal x-ray exposure and childhood cancer in twins. *N Engl J Med* 1985;312:541.
16. Shah J, Jones J, Miller MA. et al. Selective embolization of bleeding renal angiomyolipoma in pregnancy. *J R Soc Med* 1999;92:414.
17. Loughlin KR. The management of urological malignancies during pregnancy. *Br J Urol* 1995;76:639.
18. Kobayashi T, Fukuzawa S, Miura K et al. A case of renal cell carcinoma during pregnancy: simultaneous cesarean section and radical nephrectomy. *J Urol* 2000;163:1515.
19. Kim WW, Chon JY, Chun SW et al. Laparoscopic procedures during the third trimester of pregnancy. *Surg Endosc* 2000;14:501.