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DOES RETROPERITONEAL LYMPHADENECTOMY FOR TESTICULAR GERM CELL TUMOR REQUIRE A DIFFERENT APPROACH IN THE PRESENCE OF HORSESHOE KIDNEY?

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ABSTRACT

Purpose: We report our management of stage II testicular nonseminomatous germ cell tumor in 2 patients with horseshoe kidney and discuss the technical challenges posed by this renal fusion anomaly. The embryology and clinical anatomy of horseshoe kidney are discussed with particular reference to the anomalous vascular pattern and routes of testicular lymphatic drainage in this setting. Modifications and innovations of the standard technique of retroperitoneal lymphadenectomy in the presence of horseshoe kidney are discussed in light of our experience with these patients at 2 major tertiary care cancer centers. The significance of contemporary advanced noninvasive radiological techniques, such as helical computerized tomographic angiography with digital 3-dimensional reconstruction and magnetic resonance angiography, in the surgical planning and safe performance of surgery is emphasized.

Materials and Methods: Two young male patients treated at 2 major American teaching hospitals who had coexistent stage II testicular nonseminomatous germ cell tumor and horseshoe kidney underwent salvage retroperitoneal lymph node dissection.

Results: There was no evidence of recurrence in these 2 patients 12 and 15 months after surgery, respectively.

Conclusions: Horseshoe kidney poses special technical problems during retroperitoneal lymphadenectomy for testicular tumors due to anomalous renal and intra-abdominal vascular patterns. Helical computerized tomography angiography is useful for meticulous surgical planning and the safe performance of surgery in this setting.

KEY WORDS: testis, kidney, lymph node excision, abnormalities, germinoma

Fusion anomalies of the kidneys are of 2 types, namely horseshoe kidney and its variants, and crossed fused ectopia. In 1522 da Carpe first documented horseshoe kidney[1] but the first detailed description of the anomaly was provided in 1665 by Botallus.[2] Horseshoe kidney is probably the most common fusion anomaly. In more than 90% of cases fusion occurs along the lower pole. It is of 2 types, that is symmetrical (midline fusion) and asymmetrical (L-shaped) horseshoe kidney with the isthmus lying slightly lateral to the midline (lateral fusion) in the latter condition.

The incidence of horseshoe kidney is 1/400 to 1/600 to 1,800 individuals.[3] The male-to-female ratio is approximately 2:1. Germ cell tumors of the testis occur at a yearly frequency of 2.2/100,000 males in the United States. The association of germ cell tumor of the testis with horseshoe kidney has been reported in 1.3% of patients who undergo retroperitoneal lymphadenectomy for testicular tumors, although this rate reflects a tertiary center referral population.[4] This association is significant, in that retroperitoneal lymphadenectomy poses special problems due to intra-abdominal anatomical variations, anomalous vasculature, renal anomalies and occasionally calices traversing the isthmus. These factors demand innovative technical modifications.

Since the last report of this topic by Sogani and Whitmore more than 20 years ago,[4] advances in imaging technology have made it possible to achieve preoperative vascular imaging and surgical planning by helical computerized tomography (CT) angiography, while invasive techniques, such as aortography and inferior venacavography, are seldom indicated. Valuable multiphasic high resolution vascular images can now be obtained by subsecond, multidetector helical CT data acquisition and 3-dimensional (D) reconstructed angiography.[5] Simultaneous acquisition of multiple slices results in decreased scan time with decreased collimation. The latter results in increased z axis resolution, providing optimized image quality and the best 3-D vascular resolution. The vessels are visualized free of artifacts, resulting in a dramatic improvement and acceptance of CT angiography by the surgeon. Rapid 3-D post-processing of helical CT angiography provides an improved depiction of abnormal vascular pattern, which is of paramount importance while operating on patients with renal fusion anomalies. Therefore, the vascular diagnostic protocol in the presence of horseshoe kidneys needs reassessment in the face of contemporary advanced imaging techniques. We present our management of testicular nonseminomatous germ cell tumors in 2 patients with concomitant horseshoe kidney. The technical challenges posed by this rare association, the impact of 3-D CT angiography on surgical planning and modifications to achieve safe and effective retroperitoneal lymphadenectomy in these patients are discussed.

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CASE HISTORIES

Case 1. A 22-year-old male presented with an asymptomatic left testicular mass. General physical examination was normal except for mild gynecomastia. Physical examination of the scrotum revealed a nontender, mobile and firm left testicular mass. The left spermatic cord, contralateral testis and spermatic cord were normal. There was no significant lymphadenopathy. Abdominal examination, review of systems and digital rectal examination were normal. Serum α-fetoprotein (AFP) and β-human chorionic gonadotropin (β-HCG) were elevated at 123 ng./ml. and 652 mIU/ml. (normal 0 to 5 and 0 to 1, respectively).

Left inguinal orchiectomy revealed a mixed nonseminomatous germ cell tumor of 85% embryonal carcinoma, 5% yolk sac tumor and 10% mature/immature teratoma with scattered syncytiotrophoblastic cells. Post-orchiectomy tumor markers at 5 weeks remained high at AFP 161 ng./ml. and β-HCG 112 mIU/ml. Liver function tests and serum creatinine were normal. CT of the abdomen revealed small aortocaval and left periaortic lymphadenopathy, and an incidental horseshoe kidney. Clinical stage was T1N1MX.

The patient received 3 courses of bleomycin, etoposide and cisplatin chemotherapy. After chemotherapy abdominal CT revealed enlarging (3.4 cm.) aortocaval and left periaortic lymph node masses (stage T1N2M0). Helical CT angiography with 3-D reconstruction showed 5 arteries feeding the horseshoe kidney with 2 each arising close to each another from the abdominal aorta and supplying the renal parenchyma on either side, and a central artery arising from the abdominal aorta below the root of the inferior mesenteric artery with individual branches to each half of the isthmus (fig. 1). Six weeks later AFP was 8.3 ng./ml. and β-HCG was undetectable in serum.

Salvage retroperitoneal lymphadenectomy was performed via a midline transperitoneal incision. Complete bilateral retroperitoneal lymph node dissection including paracaval, interaortocaval and periaortic lymphadenectomy was performed by the split and roll technique (fig. 2). The isthmus required retraction upward and anterior to facilitate en bloc resection of the lymph node packet. All 5 arteries supplying the horseshoe were identified and preserved. A large lymph node mass identified in the suprahilar region abutting the left adrenal gland was resected and submitted separately for histopathological examination.

Histopathological evaluation revealed metastatic cystic mature teratoma in the interaortocaval, left para-aortic and left suprarenal lymph nodes. The lymph node mass was 3.5 cm. in largest diameter and involved adjacent soft tissue. No tumor was identified in the pericaval lymph nodes. The patient is currently on regular followup with tumor markers and abdominal CT. There was no evidence of recurrence at the time of this report.

Case 2. A 24-year-old male presented with an asymptomatic left testicular nodule. General physical examination was normal. Physical examination of the scrotum revealed a nontender, mobile and firm left testicular mass. The left spermatic cord, contralateral testis and spermatic cord were normal. There was no significant lymphadenopathy. Abdominal examination, review of systems and digital rectal examination were normal. Serum AFP and β-HCG were elevated at

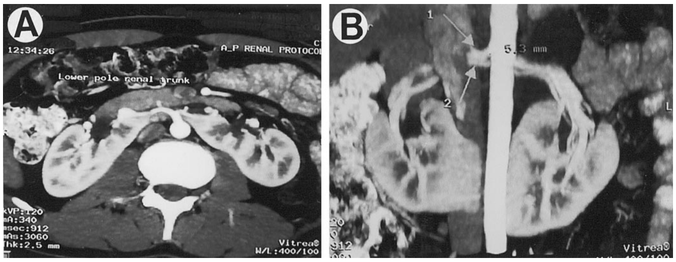

FIG. 1. Case 1. Helical CT with vascular 3-D reconstruction demonstrates anomalous vascular pattern. *A*, accessory central artery arises from abdominal aorta below inferior mesenteric artery root with individual branches to each half of isthmus. *B*, 2 renal arteries (*1* and *2*, arrows) arising from abdominal aorta supply upper portion of horseshoe kidney on each side.

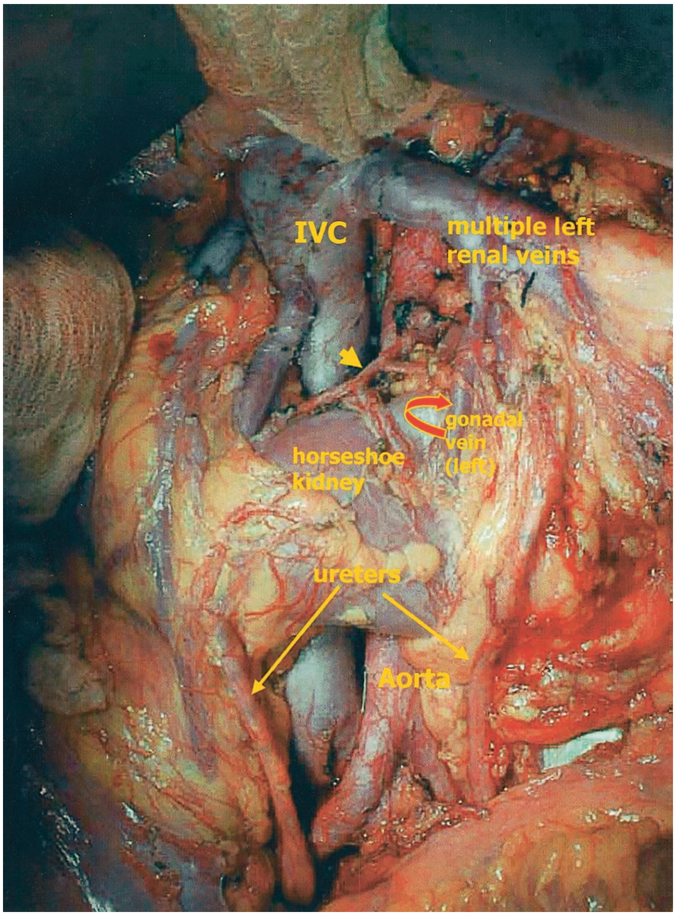

FIG. 2. Case 1. Operative findings of aberrant central artery (short arrow) from aorta with individual branches to each half of isthmus and left gonadal vein draining into left renal vein. *IVC*, inferior vena cava.

1,102 ng./ml. and 383 mIU/ml. (normal 0 to 8 and 0 to 5, respectively). Scrotal ultrasound demonstrated a 6 cm. heterogeneous intratesticular mass in the left testicle.

Left radical orchiectomy revealed a mixed germ cell tumor consisting of immature teratoma, embryonal carcinoma and yolk sac tumor. Chest x-ray was normal. Abdominal CT showed bulky retroperitoneal lymphadenopathy (stage T1N2M0) and an incidental horseshoe kidney.

The patient was initially treated with 3 cycles of cisplatin, etoposide and bleomycin chemotherapy. At followup evaluation tumor markers had returned to normal levels. However, followup CT demonstrated a 3.1 cm. residual left periaortic mass (stage T1N1M0) (fig. 3). CT angiography with 3-D digital reconstruction revealed 4 renal arteries feeding the horseshoe kidney with 1 artery to each moiety and 2 supplying the isthmus (fig. 4). No venous anomaly was evident in this patient.

Exploration was performed via a midline transmesenteric approach. The retroperitoneum was opened below the ligament of Treitz without extensive mobilization of the mesentery root. Retraction of the mesentery provided access for left template retroperitoneal lymphadenectomy. The left periaortic lymph node mass in the horseshoe kidney hilum was resected with care taken not to injure the aberrant arteries supplying the isthmus, anterior displaced ureters and isthmus (fig. 5, *A*). Resection of the periaortic and interaortocaval lymph nodal packet was satisfactorily achieved through the transmesenteric window (fig. 5, *B*).

Postoperatively the patient did well. Histopathological examination of the resected lymph node mass revealed mature teratoma. At the time of this report followup tumor markers

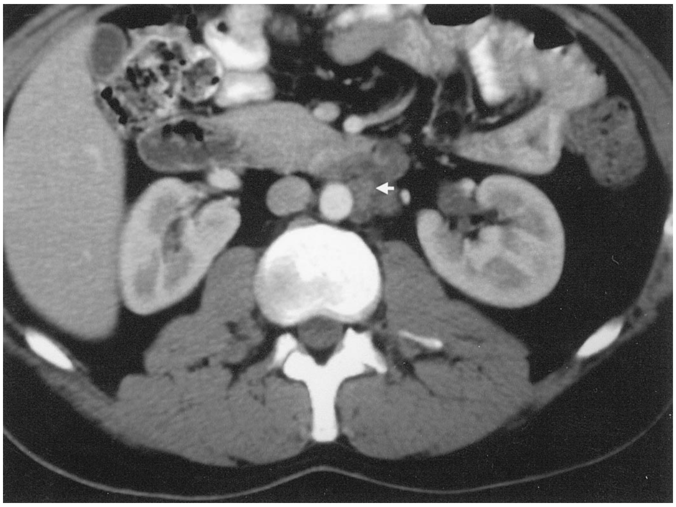

FIG. 3. Case 2. CT shows residual retroperitoneal lymphadenopathy (arrow).

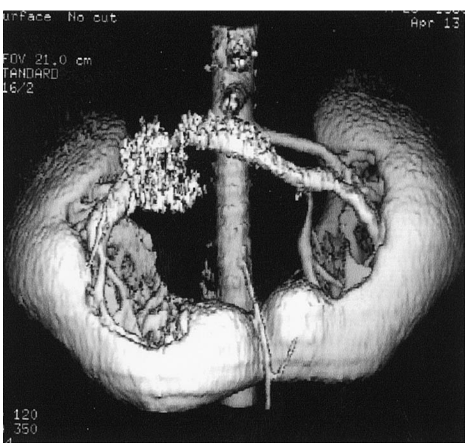

FIG. 4. Case 2. Digitally enhanced CT angiogram reveals anomalous vascular pattern with location of 4 arteries supplying horseshoe kidney, including 1 each to main renal parenchyma on either side and 2 supplying isthmus.

were normal at AFP 4.1 ng./ml. and β-HCG less than 2 mIU/ml. There was no evidence of recurrence.

DISCUSSION

Abnormal fusion of the renal units in a horseshoe kidney is believed to occur at the 5 to 12 mm. embryo stage, when the kidneys are in the true pelvis.[6] The horseshoe kidney lies at a position (L3 to L5) lower than normal since the ascent of its isthmus is prevented by the inferior mesenteric artery root. Medial rotation of the renal pelvis is also prevented and the ureters enter the pelves higher up than in normal kidneys, arising from the inner border of the upper part of each kidney and passing over the anterior surface of the isthmus. The ureter may be ectopic or retrocaval. The majority of horseshoe kidneys have a thick parenchymatous isthmus usually located anterior to the aorta and inferior vena cava, and rarely behind or between these vessels.

Horseshoe kidneys show great variation in the origin, number and size of renal arteries and veins. Only a third have a single renal artery per side.[6–8] Accessory vessels usually feed the lower pole of the renal units and they may arise from the aortic bifurcation and/or inferior mesenteric, common iliac and external/internal iliac arteries. The inferior mesenteric artery may arise from the aorta above the isthmus and cross anterior to the isthmus.

Compared with aberrant/accessory renal arteries, venous anomalies are uncommon. There may be a double inferior vena cava at the lumbar level or in extreme dysembryogenesis the inferior vena cava may be completely absent.[9] A persistent left sacrocardinal vein results in the formation of a double inferior vena cava at the lumbar level. The renal veins may drain into the posterior segment of the duplicated inferior vena cava.[4] The right ureter may travel between the 2 limbs of the inferior vena cava. When the inferior vena cava is absent, the right subcardinal vein fails to connect with the liver and shunts its blood directly into the right supracardinal vein. Therefore, the blood from the caudal part of the body reaches the heart by way of the azygos vein and superior vena cava.[9] The gonadal vein may drain into the azygos vein and superior vena cava in such circumstances. In low-lying asymmetrical horseshoe kidneys the confluent renal vein may drain into the left common iliac vein.[6,10]

Accurate visualization of the abnormal renal vasculature is extremely important for meticulous preoperative planning and it serves as a roadmap for the safe performance of surgery. It can be achieved by helical CT with 3-D digital reconstruction, magnetic resonance angiography or 3-D power Doppler ultrasound.[11]

The testicular lymphatics comprise 2 sets, namely the superficial and deep sets. The superficial lymphatics commence on the surface of the tunica vaginalis and the deep lymphatics commence in the epididymis and testicular body. They form 4 to 8 collecting trunks that ascend with the testicular vessels in the spermatic cord anterior to the psoas major muscle.[12] At the level where the spermatic vessels cross the ureter the lymphatics deviate from the spermatic vessels medial and terminate in the lateral aortic and preaortic groups of the para-aortic (lumbar) lymph nodes.

Evolution of the retroperitoneal lymphadenectomy template from suprahilar bilateral through infrahilar bilateral to modified unilateral templates has resulted in improved postoperative morbidity, while maintaining the efficiency and efficacy of full bilateral dissection. Post-chemotherapy residual masses may be resected with modified template dissection without leaving tumor residue.[13,14] As in our case 1, bilateral retroperitoneal lymphadenectomy has been recommended for post-chemotherapy bulky retroperitoneal disease because metastatic deposits may then be widespread.[13,15] Suprahilar dissection is performed only when palpable residual tumor extends above the renal vessels.[13]

Since the testicular lymphatics follow the testicular vessels, it is reasonable to believe that testicular lymphatic drainage in patients with horseshoe kidney does not differ from that in normal individuals except in rare instances of an absent or duplicated inferior vena cava. In patients with an absent inferior vena cava testicular lymphatic drainage may be unpredictable and follow the anomalous azygos venous system with possible drainage into the periaortic lymph nodes above the level of the normal origin of the renal arteries. However, to our knowledge no embryological, anatomical or clinical evidence is available to support this hypothesis. The testicular lymphatics may drain into abnormally located lymph nodes between the 2 limbs of the bifurcated inferior vena cava.[4] Sogani and Whitmore advocated bilateral retroperitoneal lymphadenectomy with extension of the upper level of dissection to the level of the pancreas in the presence of anomalous venous drainage due to the unpredictability of lymphatic drainage.[4]

The horseshoe kidney may be mobilized superior and medial by incising the peritoneum along the lateral kidney border.[4] Complete retroperitoneal lymphadenectomy can usually be achieved without dividing the isthmus, which needs careful mobilization and retraction anterosuperior or inferior depending on its arterial supply. Contrary to past belief[10,16] division of the isthmus is not necessary and it is contraindicated in the presence of functioning isthmic paren-

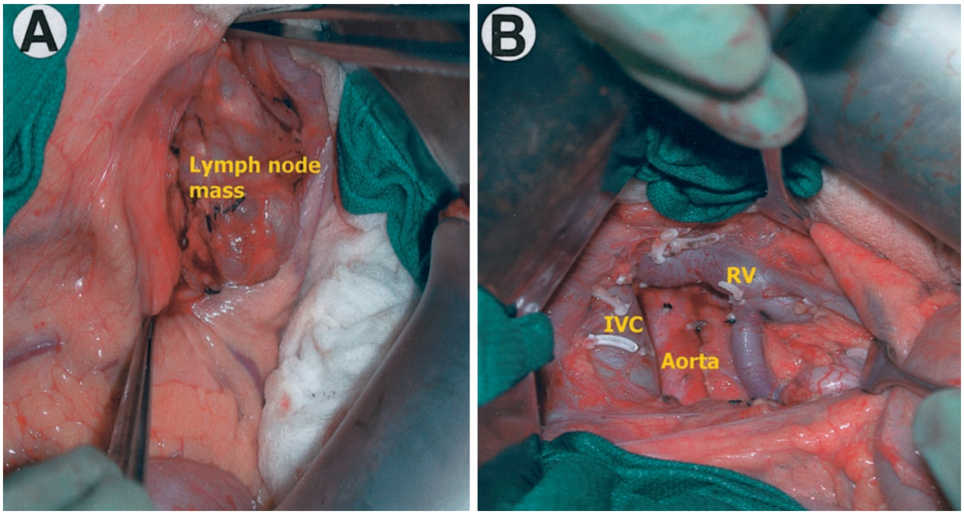

FIG. 5. Case 2. *A*, retroperitoneal lymph node mass. *B*, mass was completely resected off of great vessels. *IVC*, inferior vena cava. *RV*, left renal vein.

chyma to avoid hemorrhage and urinary fistula. To achieve lymphadenectomy one can usually reach behind the isthmus, around and between the aorta and inferior vena cava (paracaval, interaortocaval and periaortic lymph node dissection). One should be wary of the anterior crossing inferior mesenteric artery during mobilization of the isthmus. Resection of the lymph node packet around the common iliac vessels may be facilitated by mobilizing the kidney and isthmus in the superomedial direction.

In our case 1 histopathological evaluation revealed that the left suprarenal lymph node mass was metastatic cancer. However, it was difficult to establish whether the suprarenal lymph nodes were involved by proximal cancer spread from the lower lumbar lymph nodes or independently by a separate set of testicular lymphatics deviating from the others and draining into the suprarenal lymph nodes. Due to this unpredictable nature of lymphatic drainage in patients with anomalous venous drainage it may be prudent to consider extension of the template to the suprarenal level. However, confirming embryological and correlative clinicopathological evidence is lacking. In patients without major vascular (particularly venous) anomalies one may follow the standard template for lymph node dissection without compromising cancer control, as in our patient 2. In those with retroperitoneal lymphadenopathy due to seminoma it may be safer to perform retroperitoneal lymphadenectomy.[17]

CONCLUSIONS

The rare coexistence of retroperitoneal lymph node disease due to testicular tumors and horseshoe kidney poses a technical challenge to the surgeon and mandates individualized innovative modifications to achieve safe and effective lymph node dissection. Meticulous preoperative vascular evaluation and surgical planning are now facilitated by helical CT with 3-D digital reformatting or magnetic resonance angiography. Complete retroperitoneal lymphadenectomy could be achieved with careful mobilization of the isthmus and kidney with preservation of the vascular supply but without dividing the isthmus. Further anatomical and clinicopathological studies are warranted to determine whether the template for lymphadenectomy must be extended above the level of the renal arteries in patients with extreme intra-abdominal venous anomalies, such as an absent inferior vena cava.

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