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Scientific and clinical letters

Leyomioma of the renal capsule: A case report and literature review

Leiomioma de la cápsula renal: presentación de un caso y revisión de la literatura

To the Editor,

Mesothelial tumors are an exceptional finding in the urinary tract. Leiomyoma is the most common of these tumors, and can develop in any structure containing smooth muscle. Radiological studies are unable to unequivocally distinguish these lesions from other malignant renal tumors. Given their exceptional incidence, mesothelial tumors are rarely included in the differential diagnosis of renal masses, despite their capacity to grow to a considerable size. As a result, the diagnosis is usually based on histological evaluation of the surgical piece, as in the case presented in this study.

A 44-year-old woman presented with a history of allergy to penicillin and surgery several years ago in the form of appendectomy and myomectomy. She was referred to our center due to the casual detection of a left renal mass following abdominal ultrasound. The abdomen proved soft and depressible, without signs of peritoneal irritation, and with negative bilateral renal percussion and Guyon test results. The blood and urine tests only revealed microscopic hematuria. Abdominal ultrasound showed a solid mass in the upper pole of the left kidney. Computed axial tomography (CAT) confirmed this finding, revealing a tumor in relation to the upper pole of the left kidney, measuring approximately 32 x 23 mm in size, with discretely diminished contrast uptake, compatible with oncocytoma – though without being able to rule out renal carcinoma. There was no vascular involvement or retroperitoneal or pelvic adenopathies. With a tentative diagnosis of left renal hypernephroma, laparoscopic left renal tumor resection was carried out. The intraoperative of renal bed biopsy proved negative. The postoperative course was uneventful, and the patient was discharged on the second day after the operation. The piece was sent to the Department of Pathology, which after macroscopic (fig. 1) and microscopic evaluation (fig. 2) reported the lesion as corresponding to capsular leiomyoma. The immunohistochemical study of the cells proved positive for actin and desmin, and negative for cytokeratins, S-100 and HMB-45.

After 7 months of follow-up, the patient is asymptomatic and free of disease.

Leiomyomas are included among the benign tumors of connective tissue or mesenchymal origin, and can affect any organ of the genitourinary apparatus containing smooth muscle.

In this context, leiomyomas have been reported in the uterus, kidneys, pelvis, ureter, bladder, urethra, seminal vesicles, prostate gland, epididymis, tunica albuginea, testicles, penis and scrotum[1]. Renal leiomyomas are infrequent tumors, though in necropsy series they have been found to rank second in order of frequency among the renal mesenchymal neoplasms, after intramedullary fibroma[2]. Few cases have been described in the Spanish literature[3-9]. These tumors develop in areas containing smooth muscle, such as the renal capsule (37%), renal pelvis (17%), cortical vessels (10%) and other non-clearly defined areas (37%).

Renal leiomyoma is classified into three types according to the location of the lesion: subcapsular lesions (53%), which are small, multiple and asymptomatic tumors usually constituting casual necropsy findings; capsular lesions, which are generally large, solitary and symptomatic growths (37%); and leiomyoma of the renal pelvis (10%), which is extremely rare – only 7 cases having been reported in the literature to date.

They most often present as small (< 2 cm), cortical, multiple and asymptomatic tumors, and less often as solitary and voluminous growths. Renal leiomyoma is more common in women (66%) and in Caucasians (70%) – presentation being most frequent between the fifth and sixth decades of life. The most common form of manifestation is as a palpable mass (57%) and/or flank pain (53%). The classical triad of renal carcinoma (pain, abdominal mass and hematuria) is only observed in 3.3% of all cases[10]. Leiomyoma has also been associated to tuberous sclerosis, though the renal lesions that most often accompany the latter are angiomyolipomas and cysts. The tumor shows no predilection for one side or

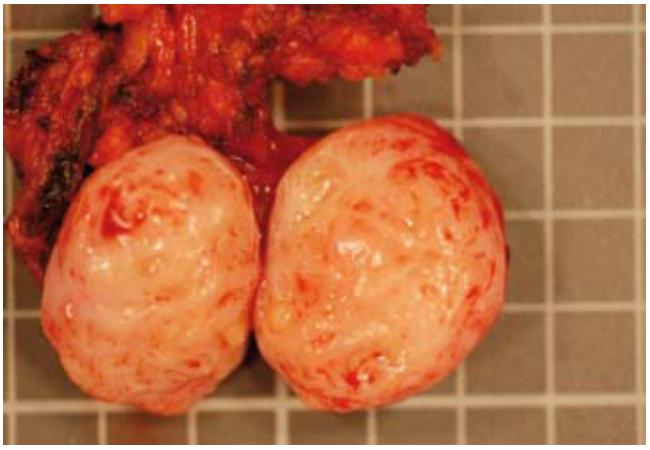

Figure 1 – Radical nephrectomy piece revealing a firm nodule measuring 3.5 x 2.5 x 2 cm in size, and accompanied by 4 cm of adipose tissue. Upon sectioning, a whitish parenchyma of elastic consistency and fascicular appearance is noted.

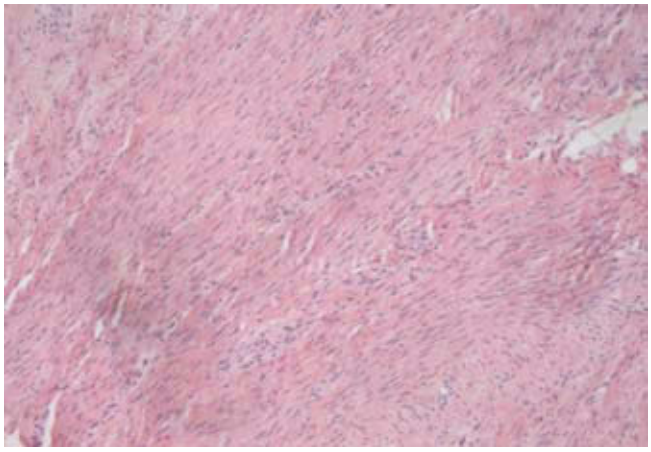

Figura 2 – Leiomyoma is characterized by the presence of fusiform cells distributed in bundles within a lax hyalinized stromal component (fascicular growth). Of note is the absence of mitotic figures and other signs of malignancy.

other, and 74% of all lesions are located in the lower third of the kidney. Leiomyoma is usually of a solid appearance (73%), and occasionally may undergo cystic degeneration (cystic leiomyoma), not always related to sarcomatous degeneration. Bleeding is observed in 17% of all cases, and 20% show irregular calcifications. Small leiomyomas (under 2 cm in size) are often multiple, located at subcapsular level, and generally constitute findings at necropsy. This is the most common presentation, though such lesions only exceptionally constitute clinical findings.

Large, single and symptomatic leiomyomas are extremely rare[8].

The low frequency of these tumors has precluded the description of specific radiological characteristics; this in turn has led to radical surgery in many instances, based on a tentative diagnosis of hypernephroma.

CAT shows certain characteristics of renal leiomyoma, and suspicion of the lesion increases when voluminous renal masses are identified in asymptomatic young women, in the absence of locoregional or distant spreading of the disease. In this context, CAT reveals well circumscribed lesions, predominantly located at subcapsular or peripyelic level, of a solid nature with soft tissue density. However, cystic presentations are sometimes observed, in the absence of contiguous invasion, lymph node involvement of systemic visceral disease. Due to their predominant smooth muscle component, leiomyomas reflect soft tissue density (45 UH) lower than the density of hypernephromas (60-80 UH), as was confirmed in our case. Doppler ultrasound, magnetic resonance angiography and spiral CAT are used to obtain vascular and topographical anatomy information on these tumors. The prognosis of these lesions is excellent, though a cases of metastasis in a tumor presenting the same histological characteristics has been described.

Macroscopically, leiomyoma is a solid tumor of elastic consistency as a result of its muscle and collagen components. The lesion is well delimited, of a white-pearly, white-gray or reddish color upon sectioning, depending on the existing vascularization, and with a fascicular appearance. In this context, the lesion differs from the classical yellow color of renal cell carcinoma or the cinnamon color of oncocytoma. Immunohistologically, leiomyoma proves positive for vimentin, actin, smooth muscle myosin, desmin and basal membrane components – including laminin and type 4 collagen – and negative for low molecular weight cytokeratins (BDK, AE1). Some authors have found HMB-45 expression to determine tumor origin in the renal capsule, versus leiomyomas developing in the renal pelvis. In addition, this marker allows differentiation from leiomyosarcoma, which is negative for HMB-45, i.e., positivity for the latter is indicative of the benign nature of the lesion. HMB-45 positivity fundamentally has been described in tumors of hamartomatous origin, such as angiomyolipoma, rhabdomyoma and lymphangioma.

The microscopic study shows the presence of fusiform cells distributed in bundles within a lax hyalinized stromal component. Of note is the absence of mitotic figures and other signs of malignancy – though some authors have reported sarcomatoid and myxomatous degeneration. Accordingly, the differential diagnosis must be established with leiomyosarcoma, fibroma, angiomyolipoma, congenital mesoblastic nephroma, sarcomatoid renal carcinoma, and in cases of leiomyoma of the renal pelvis, differentiation with respect to schwannoma is required. Epstein-Barr virus is known to be associated to the presence of different tumors – including smooth muscle lesions – in immune depressed individuals. Such tumors have been documented in different organs, including the kidneys. As a result, leiomyoma should be considered when establishing the differential diagnosis of a renal mass in this patient subgroup.

Renal leiomyoma is an infrequent tumor that must be included in the differential diagnosis of renal masses. Strong suspicion is indicated when young women present with voluminous, well circumscribed renal lesions located predominantly at peripheral capsular or peripyelic level,

of a solid nature with soft tissue density, and the absence of contiguous invasion. Since conservative surgery is a feasible alternative to radical nephrectomy in the treatment of patients with putative unilateral renal cell carcinoma measuring under 4 cm in size, a diagnosis of leiomyoma or angiomyolipoma could be established in the piece obtained from such partial surgery (as in our case). Nevertheless, considering the difficulty of clinical suspicion of these tumors, it is not uncommon for the diagnosis to be established from the histological evaluation of a radical nephrectomy piece.

REFERENCES

1. Belis J, Post G, Rochman S. Genitourinary leiomyomas. Urology. 1979;13:424.
2. Xipel JM. The incidence of benign renal nodules (a clinicopathologic study). J Urol. 1971;106:503.
3. Llamazares G, Ibarz L. Leiomioma renal en el adulto. Arch Esp Urol. 1980;4(5):269-72.
4. Cortadellas R, Castellanos RI, Guzmán A. Leiomioma de cápsula renal. Presentación de un caso y revisión de la literatura. Arch Esp Urol. 1992;45:478-80.
5. Montoya MD, García PJ, Gutiérrez JM. El leiomioma renal sintomático. Arch Esp Urol. 1993;46:833-5.
6. Rabade CJ, Fernández JM, Álvarez S. Leiomioma renal. Aportación de un nuevo caso. Actas Urol Esp. 1994;18:816-8.
7. Pereira Arias JG, Ullate Jaime V, Gutiérrez Díez JM, Ateca Díaz-Obregón R, Ramírez Rodríguez MM, Etxezarraga Zuluaga MC, et al. Leiomioma renal voluminoso. Actas Urol Esp. 2001;25(1):81-5.
8. Clemente Ramos LM, Candia Fernández A, Allona Almagro A. Leiomioma renal sintomático: una masa de difícil diagnóstico. Actas Urol Esp. 2003;27(7):546-50.
9. Gómez Pérez L, Budía Alba A, Delgado Oliva FJ, Boronat Tormo F, Pontones Moreno JL, Jiménez Cruz JF. Leiomioma de pelvis renal. Actas Urol Esp. 2006;30 (6):641-3.
10. Steiner M, Quinlan D, Goldman S, Millmond S, Hallowell MJ, Stutzman RE, et al. Leiomyoma of the kidney: presentation of 4 new cases and the role of computerized tomography. J Urol. 1990;143:994-8.

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Late relapse of stage I testicular seminoma after 11 years: successful treatment with chemotherapy alone

Recidiva tardía de seminoma testicular en estadio I a los 11 años: tratamiento satisfactorio con quimioterapia sola

To the Editor,

Approximately 45% of all germ cell tumors are seminomas, and are identified in stage I in most patients.

Radiotherapy – the most common treatment after orchiectomy – affords a relapse-free healing rate of 95% and a global healing rate of 99-100%. Nevertheless, as a result of the systematic use of radiotherapy, many patients receive unnecessary treatment, and concern over the long-term clinical effects of irradiation has led to the application of vigilance protocols in males with stage I disease.

About 15-20% of all patients subjected to vigilance suffer relapse after a median of 15 months, though relapses may occur as long as 10 years after orchiectomy.

Late relapses of germ cell tumors (GCTs) are infrequent. In these cases it has been reported that chemotherapy offers only moderate success, and that surgery may be the preferred treatment option.

The present report describes the case of a male with seminoma relapse 11 years after the initial diagnosis, in which complete remission was achieved with rescue chemotherapy alone.

A 36-year-old male presented in September 1990 with hard swelling of the right testicle during the last month. The lesion was not painful in response to palpation. Serum tumor markers proved negative, and computed tomography (CT) revealed no chest, abdominal or pelvic anomalies.

The patient preferred an intensive vigilance protocol during 5 years after orchiectomy.

Physical examinations were made, together with serum tumor marker tests and chest X-rays every month for one year after surgery, every two months during the second year, every 4 months during the third year, and every 6 months during the fourth year. CT scans were made every three months during the first postoperative year, every 4 months during the