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Case Report

A rare case of leiomyosarcoma of infundibulopelvic ligament cause renal colic: a case report

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Abstract: Primary leiomyosarcoma is an extremely rare malignant tumor in infundibulopelvic ligament. A 49-year-old female patient presented with left renal colic for 10 days. Ultrasonography and computerized tomography (CT) imaging demonstrated soft tissue density mass lesions in the lower ureter. The mass was completely excised and retroperitoneal lymphadenectomy was done with the open approach and was verified as a primary leiomyosarcoma by post-surgical histopathological examination. The patient was alive without recurrence, lymphadenopathy and other complications above 18 months. We report the first case of primary leiomyosarcoma in infundibulopelvic ligament. Moreover, early diagnosis and surgical operation are essential.

Keywords: Leiomyosarcoma, infundibulopelvic ligament, immunohistochemical, renal colic

Introduction

Leiomyosarcomas are tumors of the smooth muscle cells that may originate in any location, but most often arise in the uterus, gastrointestinal tract and soft tissue [1]. Primary infundibulopelvic ligament tumor is extremely rare, and there were no report about infundibulopelvic ligament leiomyosarcoma in English literature. Here, a case of primary leiomyosarcoma of the infundibulopelvic ligament was reported in a 49-year-old woman.

Case report

A 49-year-old female patient presented with complaints of left renal colic for 10 days in September 2015. There was no history suggestive of any other system involvement. Physical examination significantly related to percussion pain of left renal region. Urine and blood routine examination showed that there were no abnormalities. Ultrasonography and computerized tomography (CT) imaging demonstrated a density of soft tissue (2.7×2.4 cm) in the lower margin of the fourth lumbar vertebra (L4) in lower ureter, left upper ureter and renal pelvis expansion combined with hydronephrosis, and left retroperitoneal lymphadenopathy (**Figure 1**). Ureteroscopy revealed that the ureteral mucosal of lower ureter was smoothly, the ureter was compressed by the mass outside the ureter and the lower ureter was become stricture.

After discussing the illness with the patient and his family members, they required for traditional open surgical treatment. Then we underwent open approach for the mass. During the operation, after opening the retroperitoneum, carefully separate the tumor, and the pedicle of the tumor was derived from the infundibulopelvic ligament, the ureter was seen that passed under the tumor. The mass was separated without difficulty from the surrounding organs, which was complete excised, together with the proximal infundibulopelvic ligament and adipose tissue. And retroperitoneal lymphadenectomy was done.

Macroscopically, the size of the solid mass was 3.5×3.0×2.5 cm (**Figure 2A**). Which on histopathology showed a high grade fascicular spindle cell sarcoma (**Figure 2B**, **2C**), with a mitotic rate of 18 per 10 high power fields (18/10 HPF). Immunohistochemistry was strongly positive for vimentin, caldesom and smooth muscle actin (SMA) (**Figure 2D-F**), and it was focally

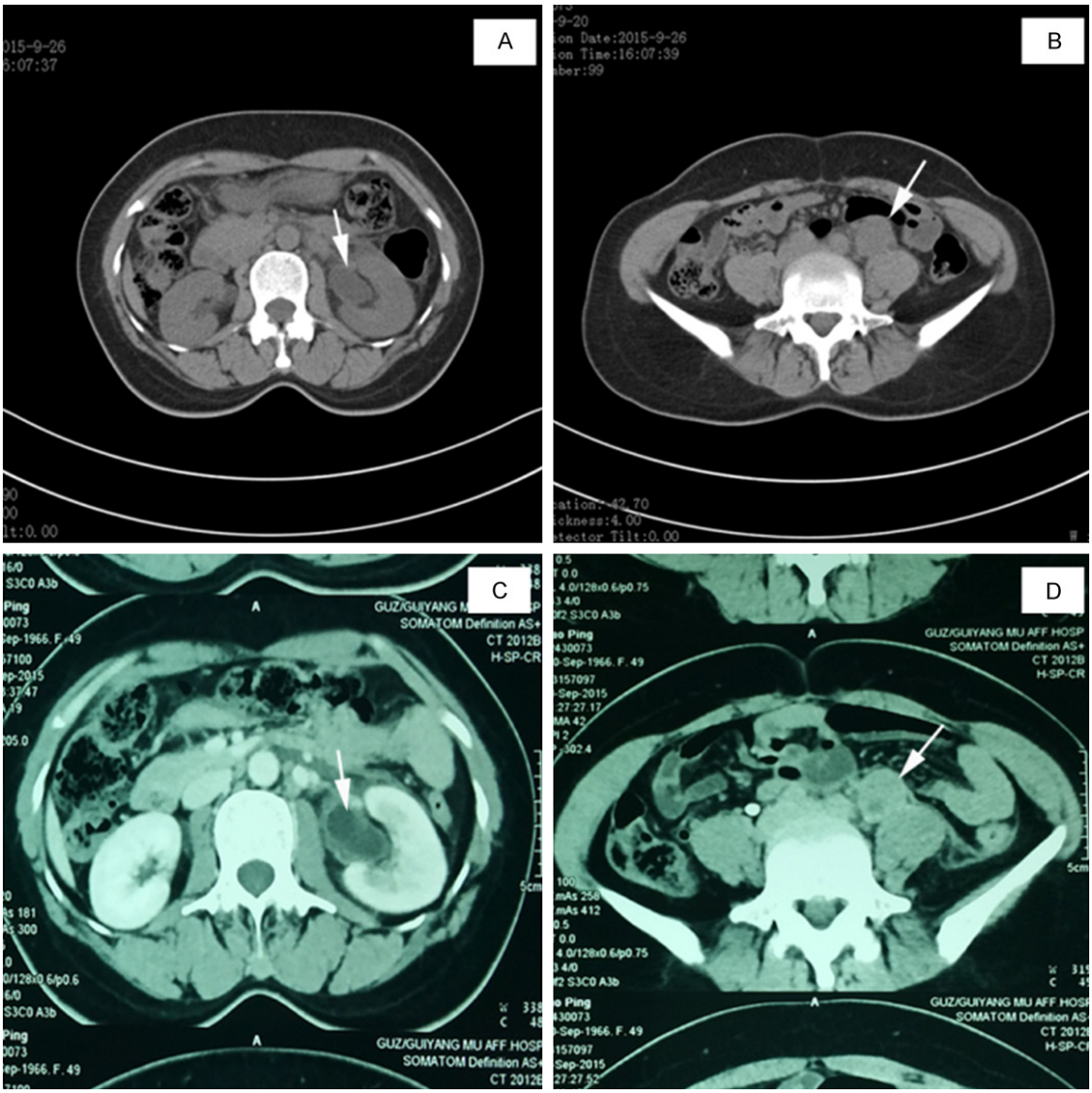

Figure 1. Computerized tomography (CT) imaging demonstrated a density of soft tissue (2.7×2.4 cm) in the lower margin of the fourth lumbar vertebra (L4) in lower ureter, left upper ureter and renal pelvis expansion combined with hydronephrosis, and with left retroperitoneal lymphadenopathy. A, B. Computed tomography scan. C, D. Enhanced Computed tomography scan.

positive for EMA (**Figure 2G**). The tumor cells were negative for CD34, CK, ALK and S100. The patient was discharged, with an advice of having regular follow up, and patient was disease free at 18 months of follow-up. The renal colic and hydronephrosis were disappearance, and no retroperitoneal lymphadenopathy was detected.

Discussion

Leiomyosarcoma is a mesenchymal tumor that has been observed to originate from the smooth muscle cells. Leiomyosarcomas occur more frequently as compared with liposarcomas and rhabdomyosarcoma [2]. On macroscopical examination, these tumors are usually rubbery in consistency, well circumscribed, with a grey, yellow or white appearance [3]. This tumor section was in a grey appearance. Microscopically, spindle shaped smooth muscle bundles arranged into interlacing fascicles are seen [4, 5]. Mitosis may be very high and foci of necrosis may be seen. We recorded a high mitotic rate of 18 per 10 high power fields,

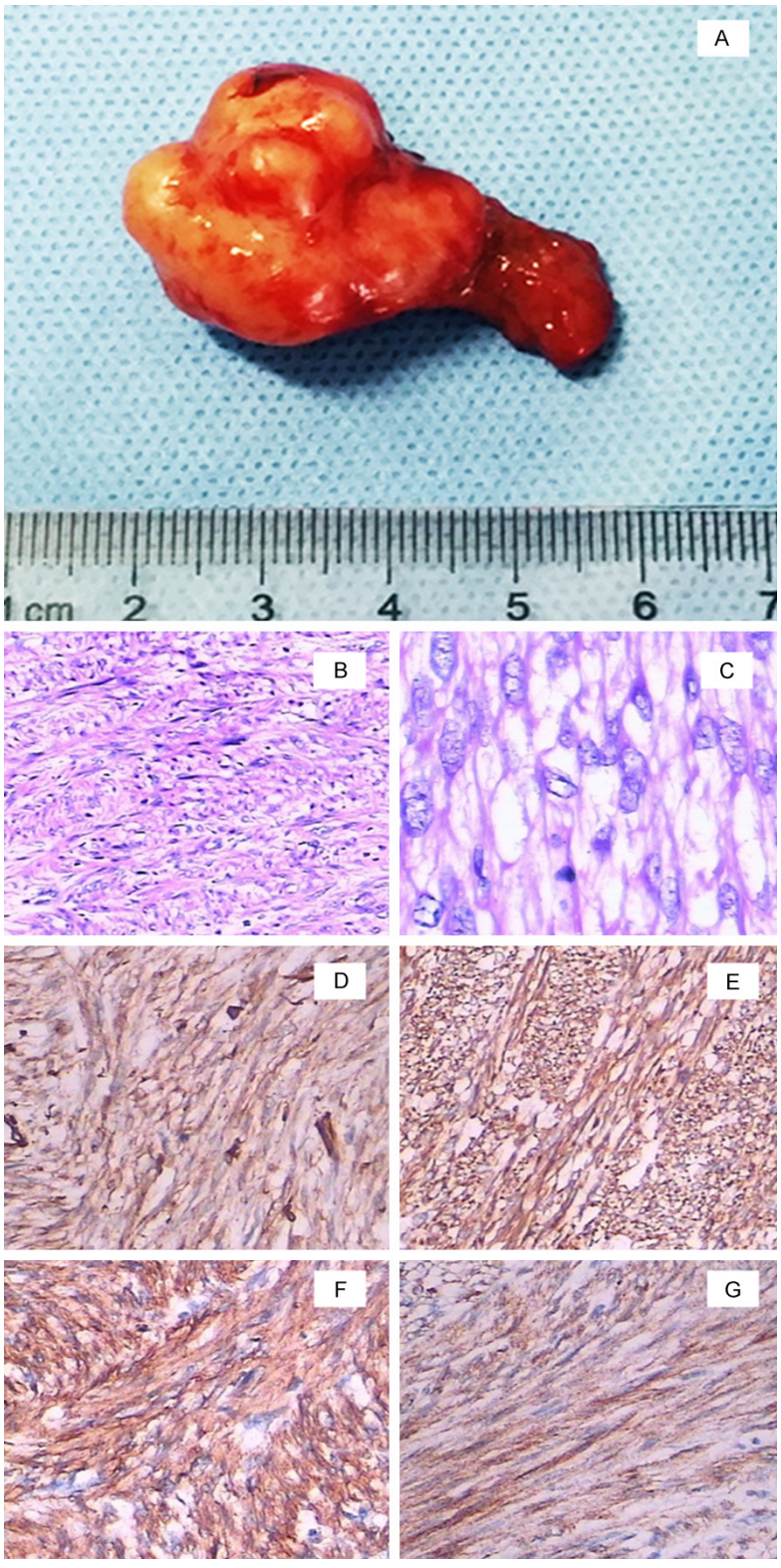

Figure 2. A. The tumor following open approach; B, C. High grade spindle cell tumor arranged in interlacing fascicles (×40, ×400). D. Spindle cells showing diffuse strong positivity for vimentin (×200). E. Caldesom positivity (×200). F. Smooth muscle actin (SMA) (×200). G. Focally positive for EMA (×200).

with many atypical mitoses. Immunohistochemistry is very essential for arriving at a definitive diagnosis, the tumor cells in our case were positive for vimentin, caldesom and smooth muscle actin (SMA), focally positive for EMA, and negative for CD34, CK, ALK and S100. These were in concordance with the findings as previous studies [1, 5, 6].

Broad ligament is divided into two layers, the upper edge (2/3) of which is free and packaged the fallopian tube (umbrella part without peritoneal covered), while the external 1/3 part of which named infundibulopelvic ligament or suspensory ligament covering ovary, ovarian artery and vein among it [7, 8]. Primary infundibulopelvic ligament tumor is extremely rare, except a case about hemangioma of the infundibulopelvic ligament in Chinese. And this 36 years old woman was found a 6.0×5.0 cm hemangioma in the right infundibulopelvic ligament according postoperative pathology [9], and there were no other reports. In this case, we found primary leiomyosarcoma of infundibulopelvic ligament, result in the left upper ureter and renal pelvis expansion combined with hydronephrosis, and cause the patient with left renal colic.

Treatment of this rare malignancy is predominantly surgical, either wide excision or amputation; however, the approach should be individualized. Complete resection is associated with better survival [10, 11]. Lymph node

disease is very rare, but if it is present, it indicates a high rate of distant spread, particularly in the lungs, liver and brain [12]. In this case, the mass was completely excised, together with the proximal infundibulopelvic ligament and adipose tissue, and retroperitoneal lymphadenectomy was done. Pre- or post- operative radiation has not proved its value in reducing loco-regional recurrences or in increasing survival rates. Radiotherapy has been used for palliation, with chemotherapy being reserved for cases of disseminated disease. And no adult urological sarcoma with disseminated disease was fully responsive to use of several chemotherapy regimens [13]. There was no radiotherapy or chemotherapy for this patient.

Conclusion

We are presenting this case of infundibulopelvic ligament high grade leiomyosarcoma as it is a common soft tissue tumor which occurs in an uncommon place, and an immunohistochemical work up is very essential for rendering this rare diagnosis. Complete surgical resection with adjuvant radio- and chemotherapy in selected patients may improve the prognosis of patients. This case reminds us to add leiomyosarcoma of infundibulopelvic ligament to the list of the causes of renal colic.

Disclosure of conflict of interest

None.

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