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Renal inflammatory myofibroblastic tumour presenting with a large retroperitoneal abscess

Ginimal Bandaralage Kavinda Deshapriya Bandara, Umesh Jayarajah and Vitharanage Srimantha Dewsiri Rodrigo

Abstract

Inflammatory myofibroblastic tumour arising from the kidney is a rare occurrence. In this case report, we present a rare case of inflammatory myofibroblastic tumour arising from the kidney diagnosed after the presentation with a large retroperitoneal abscess for the first time in literature. A 55-year-old woman with diabetes mellitus presented to us with painful lump in the left lumbar region of the back for 1-week duration. On examination, there was a firm, diffuse lump in the left lumbar region of the back. Her inflammatory markers were high, but the serum creatinine and blood urea were within the normal range. Abdominal ultrasonography showed a distorted left kidney with a heterogeneous mass consisting cystic and solid components measuring approximately $7 \times 9 \times 8$ cm in size. A contrast-enhanced computed tomography scan showed an $11 \times 9 \times 9$ cm-sized low-density mass posterior to the left kidney with multiple contrast-enhancing septations which appeared posterior to but separate from the left kidney within the left renal fascia. There was breeching of the Gerota's fascia with extension posteriorly up to subcutaneous tissue. Since the radiological opinion was in favour of a complex perinephric abscess, open drainage of the abscess was performed after failed attempts of ultrasound-guided drainage. The biopsy of the abscess wall was suggestive of a renal cell carcinoma and radical nephrectomy was planned. Due to tumour invasion, the radical nephrectomy was combined with a splenectomy and the specimen sent for histology. It showed an inflammatory myofibroblastic tumour or pseudotumour with the periphery showing ulceration and abscess formation. The patient had an uneventful recovery following surgery. Thus, we report the first case of renal inflammatory myofibroblastic tumour presenting with a large retroperitoneal abscess extending to the subcutaneous tissue plane. Final diagnosis was made only after radical surgery which was curative.

Keywords

Inflammatory myofibroblastic tumour, pseudotumour, kidney, renal neoplasm, abscess, case report

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Introduction

Inflammatory myofibroblastic tumours (IMTs) are a benign group of tumours that can arise in almost any part of the body. These tumours can present as a single mass or multiple masses with polymorphous inflammatory cell infiltrate and variable amounts of fibrosis, necrosis, granulomatous reaction and myofibroblastic spindle cells.[1]

In relation to the urinary system, the commonest site is the urinary bladder.[2] IMTs arising from the kidney are rare. Between 1972 and 2019, only 49 cases of IMTs of the kidney have been reported.[2]

These cases have typically presented with abdominal pain, lower back pain, constitutional symptoms and gross haematuria. Of the above 49 cases, 13 have been diagnosed incidentally when imaging studies were performed for unrelated indications.[2] An IMT of the kidney presenting with a large retroperitoneal abscess giving rise to a subcutaneous lump in the lumbar region is an extremely rare phenomenon. Such occurrence has not been reported in the literature. Therefore, we aimed to describe this rare atypical presentation of renal IMT.

Department of Surgery, District General Hospital, Chilaw, Sri Lanka

Corresponding Author:
Umesh Jayarajah, Department of Surgery, District General Hospital, Putlam District, Chilaw 61000, Sri Lanka.
Email: umeshe.jaya@gmail.com

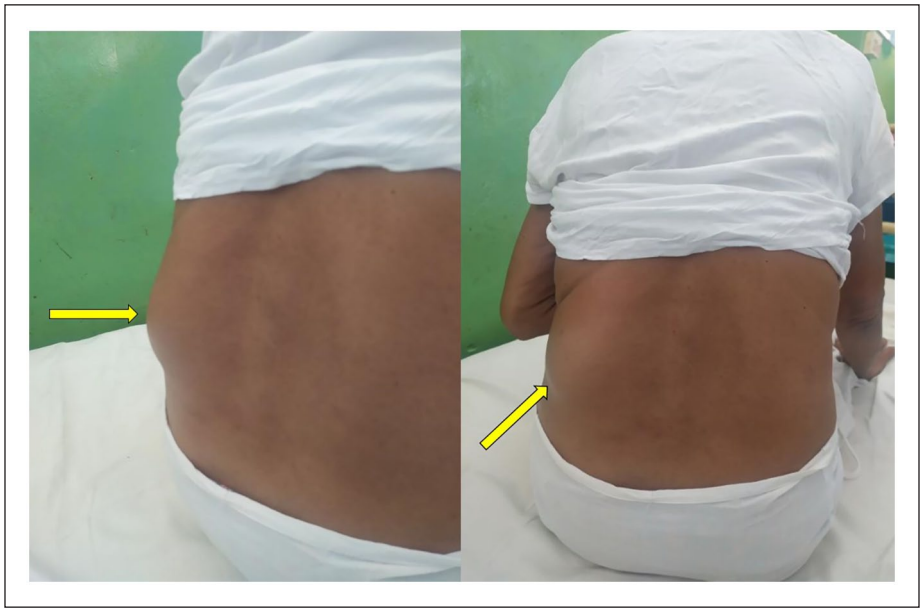

Figure 1. Image shows the subcutaneous lump (arrow) in the left loin region.

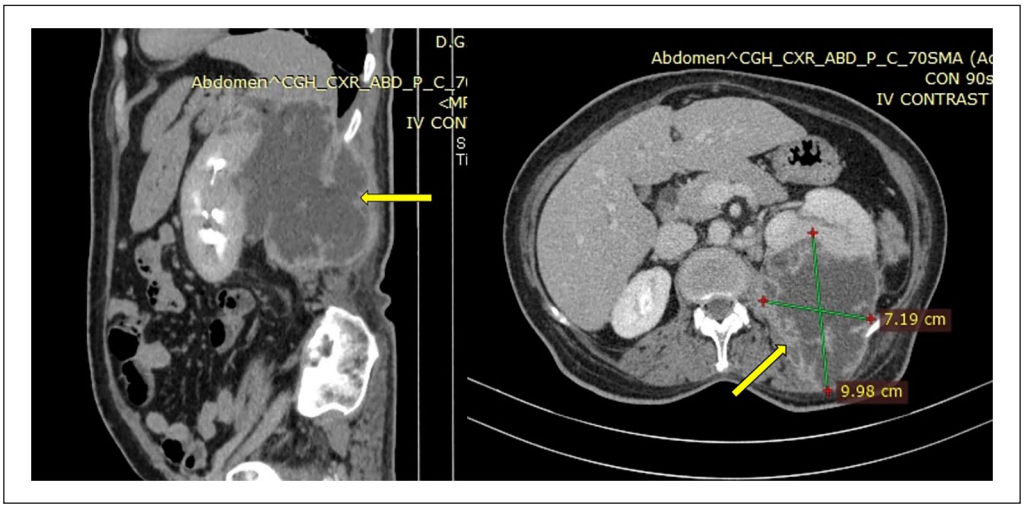

Figure 2. CECT abdomen, sagittal and axial cuts showing a large retroperitoneal abscess (arrow) posterior to the left kidney, extending to the subcutaneous tissue plane.

Case presentation

A 55-year-old woman with diabetes mellitus on insulin presented with a painful lump in the left lumbar region for 1-week duration. She had no history of fever but had loss of appetite. Urine output was normal. On examination, she had a diffuse lump in the left lumbar region (Figure 1). It was approximately 8 cm in diameter with a smooth surface and firm consistency. The abdomen was soft and no masses were palpable.

Initial investigations revealed a white cell count of 16.40×10^9/L, C-reactive protein level of 90.6 mg/L and erythrocyte sedimentation rate of 145 mm for the first hour. Serum creatinine and blood urea levels were within normal range. Abdominal ultrasonography showed a distorted left kidney with a heterogeneous mass consisting cystic and solid components measuring approximately $7 \times 9 \times 8$ cm in size. There was no hydronephrosis, and the psoas muscles appeared normal. The right kidney was normal.

A contrast-enhanced computed tomography (CECT) scan of the chest and abdomen showed an $11 \times 9 \times 9$ cm-sized low-density (0–35 Hounsfield units) mass arising from the posterior aspect of the left kidney with multiple contrast-enhancing septations (Figure 2). There was breeching of the Gerota's fascia with extension posteriorly up to subcutaneous tissue. The mass was extending medially to origin of the

psoas and erector spinae muscles and laterally to abdominal wall muscles. There were no associated fat densities, foci of air, contrast-enhancing soft tissue lesions or calcifications. Left renal vessels were normal and there was no regional lymphadenopathy. CECT chest was normal, showing no signs of a systemic or metastatic disease. Radiological opinion was that it was a complex perinephric abscess.

Since liquefaction was noted at the posterior subcutaneous part of the mass during a follow-up ultrasound study, an ultrasound-guided aspiration of the contents was arranged. Ultrasound-guided aspiration yielded 20 mL of pus and a 7-French gauge catheter was inserted into the abscess cavity with a collection bag for free drainage. The samples were sent for pyogenic culture, tuberculosis (TB) culture and polymerase chain reaction test (TB-PCR). The drain was left in situ for around 5 days to allow free drainage of pus. However, the drainage was minimal due to the thick consistency of the pus and the size of the lump remained the same.

Pyogenic culture was positive for coliforms with sensitivity to first-line antibiotics, and the TB-PCR and culture were negative. The patient was already on culture-sensitive intravenous antibiotics (intravenous co-amoxiclav). Since there was no satisfactory resolution of the abscess despite this management, an open drainage of abscess was performed under general anaesthesia.

Abscess was drained through a transverse incision in the lumbar region. It was a multiloculated abscess and 450 mL of pus was drained during the procedure. Few samples of the abscess wall were sent for histology. The histology of the abscess wall showed fragments of tissue containing diffuse sheets of clear cells with slightly irregular nuclei, sinusoid-like vessels and infiltration of fat and fibro-collagenous tissue by sheets of clear cells. The features were suggestive of a renal cell carcinoma.

A multi-disciplinary meeting was conducted with the input of the surgeon, oncologist, pathologist and radiologist with regard to further management. A decision was made to perform a left-sided radical nephrectomy. A midline laparotomy was performed. The tumour and the abscess tissue were found to be infiltrating the capsule of the spleen, posterior abdominal wall and the diaphragm. She underwent a radical nephrectomy and a splenectomy. The part of the tumour adhered to the diaphragm was excised and the defect was approximated. A left intercostal tube was inserted.

Macroscopic assessment of the specimen showed a tumour measuring 80 × 50 × 46 mm in the superior pole and extending posteriorly. The tumour was seen to be protruding posteriorly through the capsule of the kidney into perinephric fat breaching Gerota's fascia. The renal sinus and the pelvicalyceal system were not involved. Tumour was situated 7 mm away from the hilum and the pelvicalyceal system. The remaining kidney was normal and the cut surface of the adrenal gland was yellowish and homogeneous. The cut surface of the tumour was cystic, yellowish and fibrous.

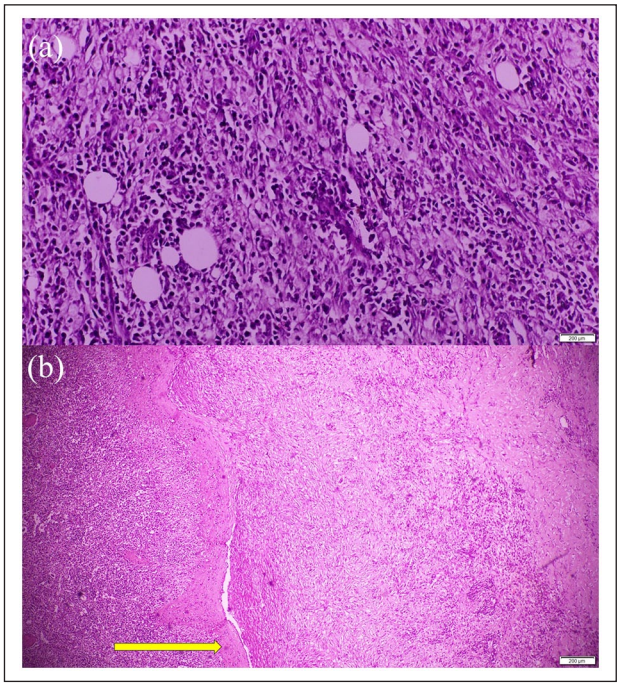

Figure 3. H and E stain with (a) ×40 and (b) ×10 magnifications showing loosely arranged spindle-shaped cells. The arrow in (b) shows the splenic capsule with no evidence of tumour invasion.

The tumour was composed of loosely arranged spindle-shaped cells resembling fibroblasts, histiocytes, plasma cells, lymphocytes and abundant thin-walled vessels (Figure 3). The periphery of the tumour showed ulceration and abscess formation. It was seen to be pushing the normal kidney tissue and the splenic capsule, but there was an invasive growth pattern in some of the areas with extension to skeletal muscle. There was no nuclear atypia, tumour necrosis or mitoses. The adrenal gland, hilar vessels, ureter and the pelvicalyceal system were normal. Splenic tissue showed disruption of the capsule with haemorrhage and reactive lymph nodes.

On immunostaining, the spindle cells were positive for smooth muscle actin (SMA) focally and CD68 (diffusely). The tumour cells were negative for vimentin and desmin and the Ki-67 proliferation index was <5%. The histological findings were compatible with an IMT or a pseudotumour with the periphery showing ulceration and abscess formation.

Post-operatively, the patient was managed in the intensive care unit and she had an uneventful recovery. Vaccinations were given for the post-splenectomy prophylaxis of infections after 2 weeks. The patient was followed up for 18 months and an ultrasound scan did not reveal a recurrence. The patient was well and asymptomatic.

Discussion

IMTs, which are also commonly referred to as pseudotumours, have earned its name due to its ability to mimic a

malignancy both clinically and radiologically.[3] They are a rare group of tumours originally described in the lung but later identified in multiple extrapulmonary anatomical sites.[4] In the urogenital system, the commonest site is the bladder, but isolated cases of IMT of kidney, renal pelvis and ureters have been reported.[5] The pathogenesis of IMTs is still obscure, but inflammatory reaction secondary to surgery, trauma, infection and even malignancy can be the cause.[6] As evident from the intra-operative and histopathological findings, surface ulceration of the tumour and necrosis may have led to the large retroperitoneal abscess formation.

There is an isolated case report of a perirenal abscess presenting with posterior abdominal wall gangrene with blackish discolouration of the skin of the left side posterior abdominal wall.[7] However, the abscess was not associated with a malignancy and the patient had a complete recovery after the drainage of the abscess.[7] Our case remains the first of its kind for a renal IMT with a large retroperitoneal abscess appearing in the lumbar region as a subcutaneous lump. Therefore, the awareness of these findings would educate the practicing clinicians, radiologists and pathologists about this rare manifestation of renal IMT. There have been isolated case reports of renal carcinomas presenting as an abscess. These included upper tract urothelial cancers or clear cell renal cell carcinomas.[8,9] These reports highlight that the presence of pus should not be attributed to an infective pathology and a malignancy should always be suspected.[8,9] Furthermore, due to the rising incidence of renal malignancies and genitourinary TB in the Sri Lankan context, it is essential that histological assessment and mycobacterial cultures are obtained.[10-12]

The diagnosis of an IMT pre-operatively is challenging because they lack specific clinical and radiological features.[2] Even pre-operative biopsies have failed to provide the correct diagnosis.[13] In most instances and in our case, the final diagnosis was usually made after nephrectomy.[14] In the management, surgical resection of the lesion is recommended as the first line of treatment for IMTs except in the cases of single kidney, bilateral masses or kidney insufficiency. Targeted therapy, chemotherapy, corticosteroids and non-steroidal anti-inflammatory drugs are other modalities of treatment.[2] The long-term prognosis of these tumours remains favourable with no evidence of recurrence or metastatic disease.[5]

Conclusion

We report the first case of an unusual presentation of a renal IMT, a large retroperitoneal abscess extending to the subcutaneous tissue plane. In our case, pre-operative diagnosis was challenging and the final diagnosis was reached after surgical resection. Radical excision of the tumour with drainage of the abscess was curative.

Author contributions

G.B.K.D.B. collected relevant information and formulated the case report with guidance from U.J. and V.S.D.R. U.J. and V.S.D.R. finalized the case report. All authors read and approved the final manuscript.

Availability of data and materials

All data used to support the findings of this study are included within the article.

Consent for publication

Written informed consent was obtained from the patient for anonymized information to be published in this article.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship and/or publication of this article.

Ethical approval

Our institution does not require ethical approval for reporting individual cases or case series.

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Informed consent

Written informed consent was obtained from the patient for anonymized information to be published in this article.

ORCID iDs

Ginimal Bandaralage Kavinda Deshapriya Bandara https://orcid.org/0000-0002-6869-8017
Umesh Jayarajah https://orcid.org/0000-0002-0398-5197

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