First report of clear cell renal carcinoma metastasizing to the ischiorectal fossa 17 years after radical nephrectomy: an additional reason for lifelong follow-up

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ABSTRACT

Renal cell carcinoma has a high propensity for metastatic spread. There are several case reports of metastatic renal cell carcinomas associated with rare metastatic sites, in many cases more than ten years after the initial diagnosis. We present a 60-year-old man with perianal pain and a mass in the ischiorectal space, revealed by computed tomography. The patient had a history of clear cell renal carcinoma operated on 17 years ago. A wire localization surgical excision of the ischiorectal fossa mass was performed. The pathological report revealed a metastatic clear cell renal carcinoma. To our knowledge, this is the first case of a clear cell renal carcinoma metastasizing to the ischiorectal fossa reported in the literature. We therefore recommend that any newly discovered mass in any site of a patient with a history of renal cell carcinoma should be carefully explored and biopsied.

KEYWORDS

Renal cancer – Metastasis – Ischiorectal – Clear cell renal carcinoma

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Introduction

Renal cell carcinoma accounts for approximately 3% of all adult malignancies. It has a high tendency to metastasize, with unpredictable patterns and sites of spread. Late metastases more than ten years after excision of the primary tumour have been reported.¹ We report a case of renal cell carcinoma metastasizing to the ischiorectal fossa; to our knowledge, this is the first case reported in the literature.

Case history

A 60-year-old Caucasian male presented at our department of general surgery with a recent onset of mild perianal pain. He had a past medical history of a clear cell renal carcinoma of stage III (T3aN1M0), for which he underwent left radical nephrectomy 17 years ago. Five years ago he was diagnosed with a renal cell carcinoma metastasis in the pancreas, for which he underwent distal pancreatectomy. He then followed a best supportive care protocol. At a recent regular laboratory imaging and clinical follow-up, no disease was revealed. Clinical examination of the

perianal area and digital rectal examination were without palpable findings, except for a mild increase in perianal pain, mainly on the left perianal area.

Because of the patient's perianal symptoms and medical history, abdominal computed tomography (CT) was performed, which revealed a solid mass, 1.5cm in diameter, in the left ischiorectal fossa (Fig 1). Surgical excision of the mass with wire localization guidance was decided.

Preoperative laboratory tests were normal. Wire localization of the mass was performed using a Kopans 20G-15cm lesion localization needle (Cook Medical, IN, USA), with the patient in the prone position under local anaesthesia and CT guidance (Fig 1). In the operating room, the patient was placed in the high lithotomy position and the excision was performed under general anaesthesia. A perianal incision from 1 to 5 o'clock was made. A yellow-brownish, poorly defined mass measuring 3 × 3 × 2cm was excised from the left ischiorectal space under wire localization guidance (Fig 2). The surgical incision was sutured. The postoperative course was uneventful, and the patient returned home the next day.

Pathology examination of the mass revealed metastasis from clear cell renal carcinoma, Fuhrman grade 2 with

free surgical margins (Fig 3). The oncologists proposed a treatment cycle of sunitinib 50mg orally once a day for four weeks, followed by two weeks drug-free. Treatment of nine 6-week cycles was completed successfully, without complications. The patient is now on a follow-up protocol including clinical, laboratory and imaging studies every three months. He is still alive without disease progression after 38 months.

Discussion

To our knowledge, this is the first report to describe a renal cell carcinoma metastasis in the ischiorectal space. Neoplasms within the ischiorectal fossa originate from direct extension of primary tumours of the pelvis. Metastatic tumours of distal organs via haematogenous or lymphatic spread have been reported.^{2,5} Primary tumours within the ischiorectal space have also been described. The most common types are aggressive angiomyxomas, liposarcomas, lipomas and plexiform neurofibromas.²

Surgical resection for stage I–III renal cell carcinoma represents the standard of care, but 40–50% of patients develop metastatic disease. The time of renal cell carcinoma recurrence after nephrectomy is an important factor for a comprehensive follow-up plan. Most recurrences of renal cell carcinoma occur within the first five years, but recurrences ten years or more after initial nephrectomy have been reported.¹

Our patient developed a pancreatic renal cell carcinoma metastasis 12 years after initial nephrectomy and a solitary nodule metastasis in the ischiorectal fossa 17 years after initial nephrectomy. These data may suggest that follow-up for life is rational. The surveillance plan should be

individualized, however; the National Comprehensive Cancer Network suggests surveillance imaging beyond five years as clinically indicated.⁴ Our patient followed an almost annual imaging surveillance with CT scan after the first five years from initial nephrectomy until he was diagnosed with pancreatic renal cell carcinoma metastasis.

The most common sites of renal cell carcinoma metastases are lung (45%), bone (30%), lymph node (22%), liver (20%), adrenal gland (9%) and brain (8%). Sountoulides and colleagues, in their review study, described some rare metastatic sites of renal cell cancer.⁵ Head and neck region metastases may be linked to renal cell carcinoma in 15% of cases, with the nose and paranasal sinuses being the most commonly affected. Other extremely rare head and neck sites include the orbit, parotid gland, tongue, tonsils and thyroid gland. The heart is another rare site of renal cell carcinoma metastasis. These metastases have good prognosis if surgically excised. Skin metastasis mimic common dermatological problems and thus are not easily identified. Renal cell cancer skin metastases occur mainly in the head, neck and trunk, usually at a late stage of disease. Metastases to muscles are a rare incident. Only three cases of renal cell carcinoma metastases to joints have been reported, all presented as acute monoarthritis. Metastasis to the ovaries, uterus, prostate and testis are rare. A probable mechanism of tumour spread to these

Figure 1 Wire localized mass in the ischiorectal space, revealed by computed tomography

Figure 2 Intraoperative findings

Figure 3 Histopathological image. In the inset, the characteristic histological features of the Fuhrman nuclear grade 2 clear cell renal carcinoma are evident (arrows) (original magnification 40×).

organs is via retrograde venous embolization of tumour cells through the ovarian vessels that end in the caval vein or the left renal vein.⁵ In our case, a similar mechanism may explain the metastasis in the ischiorectal space via the internal pudendal veins, which end in the internal iliac veins and then to the inferior caval vein.

The targeted therapy for the management of metastatic renal cell carcinoma with agents such as sunitinib, bevacizumab and sorafenib has been used in clinical

settings with encouraging results. Nevertheless, these drugs induce disease stabilization with eventual disease progression. Our patient developed a new renal cell carcinoma metastasis in the fifth year of best supportive care treatment. Considering the recent ischiorectal space metastasis, he was put on sunitinib with follow-up every three months.

Conclusions

Metastases of renal cell carcinoma can be observed in various organs and areas of the body, even decades after initial diagnosis of renal cell carcinoma. Therefore, any newly discovered mass in any site of a patient with a history of renal cell carcinoma should be carefully explored and biopsied. The ischiorectal space is a potential site for renal cell carcinoma metastasis.

References

1. Abara E, Chivulescu I, Clerk N *et al*. Recurrent renal cell cancer: 10 years or more after nephrectomy. *Can Urol Assoc J* 2010; **4**: E45–E49.
2. Tappouni RF, Sarwani NI, Tice JG *et al*. Imaging of unusual perineal masses. *Am J Roentgenol* 2011; **196**: W412–W420.
3. Llauger J, Palmer J, Pérez C *et al*. The normal and pathologic ischiorectal fossa at CT and MR imaging. *Radiogr Rev Publ Radiol Soc N Am* 1998; **18**: 61–82, 146.
4. NCCN Clinical Practice Guidelines in Oncology: Kidney Cancer Version 2. National Comprehensive Cancer Network; 2020. https://www.nccn.org/professionals/physician_gls/pdf/kidney.pdf
5. Sountoulides P, Metaxa L, Cindolo L. Atypical presentations and rare metastatic sites of renal cell carcinoma: a review of case reports. *J Med Case Rep* 2011; **5**: 429.