CASE REPORT

A rare case of metachronous penile and urethral metastases from a rectal mucinous adenocarcinoma

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SUMMARY

Metastatic lesions in the penis are uncommon in patients with prostate or bladder cancer but penile metastatic lesions from rectal tumours are rare with only 65 cases reported in the literature. We describe the case of a 70-year-old man who developed metastatic lesions within his corpus cavernosum 2 years after being diagnosed and treated for a mucinous adenocarcinoma of the rectum and a year after a wedge resection of an isolated lung metastasis. He proceeded with total penectomy and intraoperatively two skip lesions were also found within the wall of his urethra; histological analysis proved that these were also metastatic lesions. A perineal urethrostomy was formed with the remaining macroscopically healthy urethra. He made a good recovery from his operation and continued his treatment under the oncology team.

BACKGROUND

Penile metastasis from rectal cancer is rare with only 65 cases reported in the literature since its discovery over a century ago. This case report highlights the need to raise awareness of the possibility of penile metastasis in order to allow timely management of the disease. Early treatment with penectomy and chemotherapy may improve patient outcomes and prognosis. This is the first reported case presenting with both penile and urethral metastasis 2 years after treatment for a mucinous adenocarcinoma of the rectum.

CASE PRESENTATION

A 70-year-old man presented with a 6-month history of a lump within the right side of his penile shaft. It became larger over this period and caused significant pain. He did not suffer from any voiding symptoms, urinary tract infections or haematuria. When examined, this lump was hard, painful and was felt beneath the skin involving largely the right corpus cavernosum. It also appeared to be encroaching onto the left side and most of the pendulous part of the penis was most likely involved.

Two years earlier, he was diagnosed with a rectal tumour when he noticed a change in bowel habits. Biopsy of the tumour revealed a poorly differentiated mucinous adenocarcinoma. He underwent neoadjuvant chemoradiotherapy due to the presence of large obturator lymph nodes and mesorectal fat lymphadenopathy in his initial staging CT and MRI.

He proceeded with an anterior resection with loop ileostomy formation and pathological staging

showed vascular invasion in the primary lesion and 6 of 13 lymph nodes contained metastatic disease. After recovery from this procedure, he was treated with three cycles of adjuvant chemotherapy with 5-fluorouracil and oxaliplatin. This was complicated by sensory neuropathy and new-onset diabetes secondary to chemotherapy, resulting in an episode of diabetic ketoacidosis leading to an intensive care unit admission. He also developed a deep vein thrombosis in his right upper limb secondary to his intravascular chemotherapy line and was started on long-term anticoagulant treatment.

After discontinuing his chemotherapy, a restaging CT scan revealed a 1 cm nodule towards the apex of the right lung. A positron emission tomography CT (PET-CT) demonstrated uptake at the site of the pulmonary nodule but no uptake elsewhere, indicating a solitary metastasis. He was therefore started on further chemotherapy, but this was discontinued due to fatigue and weight loss. He underwent a wedge resection of his lung lesion and histological analysis confirmed a metastasis.

INVESTIGATIONS

An urgent MRI of the penis and pelvis revealed an extensive intracorporal tumour affecting both corpora cavernosa from the tips to a distance of 7 cm proximally, essentially involving the pendulous part of the penis (figure 1A). The tumour was mostly present on the right side where there was also expansion of the corpora (figure 1B). A skip lesion was apparent on the MRI approximately 11 cm from the corporal tip in the non-dependent portion of the penis (figure 1C). No other lesions were identified in the urinary tract including the prostate.

A restaging CT scan showed the presence of a new 7 mm lesion in his right lung base but no other intra-abdominal disease.

A tru-cut biopsy was performed for histological analysis of the penile shaft tumour.

DIFFERENTIAL DIAGNOSIS

The biopsy of the penile shaft tumour revealed cores of fibrofatty tissue with extensive infiltration by a mucin-secreting adenocarcinoma. This was found to be related histologically to the colorectal primary tumour.

Further immunohistochemistry was also performed and staining was positive for carcinoembryonic antigen (CEA), cytokeratin 20 and CDX2 adenocarcinoma marker. Cytokeratin 7 staining was negative, proving this was not Paget's-related

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Figure 1 MRI of the penis and pelvis; (A) Intracorporeal tumour involving the pendulous part of the penis. (B) Expansion of the right corpora cavernosa secondary to tumour. (C) Skip lesion in the corpus cavernosum approximately 11 cm from the coronal tip.

adenocarcinoma and confirming the penile lesion as a rectal metastasis.

TREATMENT

All images were reviewed in a supraregional Penile Cancer Network and the penile metastases appeared to be resectable with no obvious extension to the pelvis, prostate or new lymphadenopathy. Following discussion with the patient, a decision was made to proceed with a total penectomy and perineal urethrostomy for both symptomatic control and to improve prognosis. The new solitary 7 mm lung lesion remained under surveillance until the patient recovered from his operation. Preoperative CEA levels were 0.5 µg/L.

Intraoperatively, the urethra was dissected off the penis to allow the formation of a perineal urethrostomy. Two small hard nodules were palpated within the urethral wall and were extraluminal with an intact urethral mucosa. Owing to the proximity of the second nodule to the proximal urethra, this was excised from the urethral wall with an intact mucosa and the defect was closed with sutures longitudinally before mobilising the urethra to the perineum. A total penectomy then took place with removal of the whole of the pendulous part of the penis deep to the pubic bone.

OUTCOME AND FOLLOW-UP

During pathological dissection of the penectomy specimen, multiple ill-defined tan nodules were visible within the corpus cavernosum ranging from 5 to 16 mm in diameter (figure 2A, B). Microscopically, these nodules consisted of deposits of a mucin-producing adenocarcinoma and were located in the stroma and vascular spaces infiltrating the corpus cavernosum and the tunica with no epidermal involvement (figure 3A, B). The skin and deep margin of the specimen did not contain any tumour.

The urethral nodule initially excised from the urethra showed microscopically the same type of tumour, confirming the presence of both corpus cavernosum and corpus

spongiosum metastases from a mucinous adenocarcinoma of the rectum.

The patient recovered from his operation with no complications and well-controlled pain. The findings from his operation were reported to the Colorectal Cancer Network for further follow-up and consideration of chemotherapy.

DISCUSSION

Metastatic involvement of the penis is extremely rare, despite the rich vascularisation between the penis and the neighbouring pelvic organs.¹ The most common sites of primary malignancy are the genitourinary organs, rectum and rectosigmoid areas.¹ Until 2006, 372 cases had been reported in the literature with prostate and bladder cancer being the most common origin of the metastatic lesions at 34% and 30%, respectively.²⁻³ Rectosigmoid metastasis to the penis, however, accounted for only 13% of cases.²⁻³

There have been many proposed mechanisms of how rectal adenocarcinoma metastasises to the penis. However, it is difficult to elucidate the exact mode of spread in individual cases, as penile metastases are usually associated with disseminated disease.²⁻⁴ The most accepted mechanisms of spread are: the *retrograde venous route* due to communications between the dorsal venous system of the penis and the venous plexuses draining the pelvic viscera, the *retrograde lymphatic route*, via *arterial spread*, by *direct extension* and by *implantation* or *secondary to instrumentation*.²⁻⁴

The first case of secondary penile malignancy from an adenocarcinoma of the rectum was reported by Eberth in 1870.⁵ Following a literature search using PubMed database we identified 65 cases in total. A systematic review by Perez *et al*³ identified 40 cases up to 1991 and Chorian *et al*² reported a total of 49 cases in the literature until 2006.³ We found 16 further case reports from 2006 to 2015.¹⁻⁴⁻⁶⁻¹⁹

Penile metastases usually occur as hard lumps within the corpus cavernosum giving the penis a semi-erect appearance and occasionally skin lesions occur.¹ Malignant priapism, penile pain

Figure 2 Macroscopic pathology images of the dissected specimen; (A) Lesion involving approximately the distal 7 cm of the corpus cavernosum (black arrow) and small skip lesions (white arrows). (B) Skip lesion in the corpus cavernosum (white arrow) as identified in figure 1C.

Figure 3 Microscopy with H&E staining; (A) Mucin-producing adenocarcinoma in the corpus cavernosum, ×20 magnification, (B) Mucin-producing adenocarcinoma in the corpus cavernosum, ×100 magnification.

and difficulty in micturition are some of the major presenting symptoms.⁹ Most cases describing metachronous penile metastasis occur approximately 16–26 months after diagnosis and treatment of the colorectal primary.^{1 6–10 12–16 19} One case, however, was reported 4.5 years after treatment and another 26 years later.^{14 17} We identified two cases of synchronous metastasis in the literature which were associated with disseminated disease.^{18 20}

MRI of the penis and pelvis is considered the gold standard for local pelvic and perineal organ staging. It can also detect skip lesions within the corpus cavernosum and neighbouring organ invasion. Repeat imaging with PET-CT allows restaging of the primary cancer and the detection of other metachronous metastases which may guide treatment options following palliative penectomy for symptom control. An open or tru-cut biopsy, however, is needed for accurate histological diagnosis prior to any surgical intervention.

In this case, we also found intraoperatively intraluminal urethral metastases which are even rarer than penile and preputial metastatic lesions. These were not detected on the patient's staging MRI, and as he did not present with any preoperative voiding symptoms or haematuria, a diagnostic cystoscopy was not indicated at the time. There is only one other case report of a 72-year-old man with sigmoid colon adenocarcinoma and synchronous metastatic lesions in his liver, urethra and pubic bone detected with PET-CT.²⁰ This case highlights the diagnostic and prognostic impact of nuclear medicine imaging in oncology, as a solitary metastasis can still be treated radically, as was the case in our patient when he developed a solitary lung metastasis approximately a year earlier.

However, survival generally does not exceed 2 years unless the metastatic disease is limited to the penis.^{9 10} In most cases, the average survival of patients with penile metastases treated with chemoradiotherapy is 8 months due to the subsequent

development of further metastatic lesions in other organs.¹⁴ Since these occurrences are rare, a high index of suspicion is required to detect a penile metastasis when a patient presents with a penile lump. Early diagnosis combined with total penectomy and individualised chemotherapy may improve the quality of life, relieve pain and prolong the life of the patient, but as with any treatment this needs to be tailored to the patient's needs.

Learning points

- ▶ All physicians should have a high index of suspicion for penile metastasis when a patient presents with a new penile lump, malignant priapism or voiding symptoms and has a history of bladder, prostate or bowel carcinoma.
- ▶ CT, MRI and positron emission tomography-CT should ideally include the male genitalia when performing surveillance imaging for any pelvic organ cancer.
- ▶ MRI is the gold standard investigation for any patient presenting with a penile shaft lesion but could miss lesions within the corpus spongiosum.
- ▶ Early diagnosis combined with total penectomy and individualised chemotherapy may improve the quality of life, relieve pain and prolong the life of the patient.

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REFERENCES

- 1 Yildirim M, Coskun A, Pürten M, *et al.* A clinical case of the penile metastasis from the rectal carcinoma. *Radiol Oncol* 2010;44:121–3.
- 2 Cherian J, Rajan S, Thwaini A, *et al.* Secondary penile tumours revisited. *Int Semin Surg Oncol* 2006;3:33.
- 3 Perez LM, Shumway RA, Carson CC III, *et al.* Penile metastasis secondary to supraglottic squamous cell carcinoma: review of the literature. *J Urol* 1992;147:157–60.
- 4 Dorsett F, Hou J, Shapiro O. Metastasis to the penis from rectal adenocarcinoma. *Anticancer Res* 2012;32:1717–19.
- 5 Eberth CJ. Krebismetastasen des corpus cavernosum penis. *Virch Arch* 1870;51:145.
- 6 Brønserud MM, Sørensen FB, Rahr H, *et al.* Penile metastasis from rectum cancer primarily interpreted as Peyronie's disease. *Ugeskr Laeger* 2015;177:120–1.
- 7 Alzayed MF, Artho G, Nahal A, *et al.* Penile metastasis from rectal cancer by PET/CT. *Clin Nucl Med* 2015;40:245–50.
- 8 Luo NQ, Zhang ZH, Ma Y. Penile metastasis from rectal carcinoma: a case report and literature review. *Zhonghua Nan Ke Xue* 2014;20:359–62.
- 9 Persec Z, Persec J, Sovic T, *et al.* Penile metastases of rectal adenocarcinoma. *J Visc Surg* 2014;151:53–5.
- 10 Kimura Y, Shida D, Nasu K, *et al.* Metachronous penile metastasis from rectal cancer after total pelvic exenteration. *World J Gastroenterol* 2012;18:5476–8.
- 11 Yagnik VD. Unusual metastasis from a rectal adenocarcinoma: penile metastasis. *Anz J Surg* 2011;81:844.
- 12 Maestro MA, Martínez-Piñero L, Moreno SS, *et al.* Penile metastasis of rectal carcinoma. Case report and bibliographic review. *Arch Esp Urol* 2011;64:981–4.
- 13 Lee JI, Kang WK, Kim HJ, *et al.* Unusual metastasis from a rectal adenocarcinoma: penile metastasis. *Anz J Surg* 2011;81:102.
- 14 Kúronya Z, Bodrogi I, Lóvey J, *et al.* [Metachronous metastasis from rectal adenocarcinoma to the penis—case report]. *Magy Onkol* 2009;53:263–6.

- 15 Park JC, Lee WH, Kang MK, *et al.* Priapism secondary to penile metastasis of rectal cancer. *World J Gastroenterol* 2009;15:4209–11.
- 16 Murhekar KM, Majhi U, Mahajan V, *et al.* Penile metastasis from rectal carcinoma. *Indian J Cancer* 2007;44:155–6.
- 17 Ketata S, Boulaire JL, Soulimane B, *et al.* Metachronous metastasis to the penis from a rectal adenocarcinoma. *Clin Colorectal Cancer* 2007;6: 657–9.
- 18 Chung TS, Chang HJ, Kim DY, *et al.* Synchronous penile metastasis from a rectal carcinoma. *Int J Colorectal Dis* 2008;23:333–4.
- 19 Appu S, Lawrentschuk N, Russell JM, *et al.* Metachronous metastasis to the penis from carcinoma of the rectum. *Int J Urol* 2006;13:659–61.
- 20 Seo HS, Kim ES, Kim S, *et al.* A Case of Urethral Metastasis from Sigmoid Colon Cancer Diagnostically and Prognostically Indicated by F-18 FDG PET/CT. *Nucl Med Mol Imaging* 2011;45:319–23.

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