CASE REPORT

Unusual presentation of bladder cancer resurgence and efficacy of radiotherapy

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SUMMARY

A 68-year-old man with a history of bladder cancer presented with perineal pain and penile priapism. The work up showed multiple lesions strictly located in the penis; biopsy confirmed metastases of bladder cancer. Surgery was judged unfeasible and chemotherapy failed to improve symptoms. Radiotherapy was therefore delivered on the whole penis and resulted in a rapid clinical benefit and persistent control of the disease. Penile metastases are very rare and no consensus exists concerning their management; radiotherapy appears as a promising therapeutic option not only to palliate pain but also to control the disease.

BACKGROUND

Urothelial carcinoma represents 90% of bladder cancer and is the fifth most common cancer worldwide. When invading muscle layer, urothelial cancer requires multimodal therapy including chemotherapy and cystectomy; however, despite this aggressive management, more than 50% of patients develop metastases with a median survival not exceeding 13 months and with a 5-year survival of only 15–20%.¹ Metastasis appearing in the penis is very rare and results in uncontrolled symptoms such as pain, priapism, haematuria and urinary retention; as it is often associated with widespread metastatic disease, prognosis is poor and treatment is usually palliative. However, in advanced urothelial cancer with unique penile metastasis, the role of radiotherapy is not clear. We present the case of a patient with urothelial cancer metastasis to the penis; chemotherapy did not improve symptoms, while radiotherapy resulted in a rapid and significant clinical benefit.

CASE PRESENTATION

In January 2013, a 68-year-old man presented with haematuria. His medical history included only hypertension and hypercholesterolaemia. Initial cystoscopy showed a polypoid lesion of the right lateral part of the bladder. Endoscopic biopsies confirmed a high-grade (grade 3) urothelial carcinoma infiltrating the detrusor muscle. Systemic work up showed no distant metastasis. Treatment consisted of neo-adjuvant platinum-based chemotherapy followed by cystectomy. Histological findings confirmed a large high-grade urothelial carcinoma infiltrating the bladder muscle; no metastatic node was found.

In January 2015, the patient was admitted for continuous and uncontrolled perineal pain associated with priapism.

INVESTIGATIONS

Pelvic MRI showed multiple nodules located in the penis, infiltrating the two corpus cavernosum and the corpus spongiosum and reaching the bulbospongiosus muscle (figure 1).

Thoracoabdominal CT did not show any abdominal lymph node nor was any other distant lesion seen.

DIFFERENTIAL DIAGNOSIS

In the context of the patient's recent urothelial cancer, percutaneous biopsies were performed, which confirmed metastases of urothelial carcinoma.

TREATMENT

Surgery was judged unfeasible due to the infiltration of the bulbospongiosus muscle. Chemotherapy with an every-2-weeks regimen including paclitaxel 150 mg/m² and gemcitabine 2000 mg/m² was rapidly started. Despite increasing doses of analgesics such as opioids, corticosteroids and pregabalin and two courses of chemotherapy, no improvement was observed and our patient remained highly symptomatic. During preparation of radiotherapy, a third course of chemotherapy was administered; radiotherapy was then initiated and a dose of 32.5 Gy in 13 fractions was delivered over 3 weeks. Radiotherapy fields were designed to cover the whole penis with sufficient planning target volume margins. The penile pain disappeared 4–5 days after the onset of radiotherapy and the analgesic drugs were decreased progressively. Clinical examination showed a rapid decrease of priapism and penile tenderness. Aside from a mild balanitis (grade 1) induced by radiotherapy, our patient did not present any side effects.

Figure 1 Infiltrating metastatic lesions throughout the penis.

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OUTCOME AND FOLLOW-UP

Further work up after radiotherapy confirmed control of the penile metastases and no other distant lesion. At the time of submission, 6 months after the end of radiotherapy, our patient is still treatment-free with a good quality of life and no radiotherapy-induced side effects.

DISCUSSION

Despite the rich vascularisation of the penis, penile metastasis is a rare entity. Most primary lesions are prostate, bladder, rectal and kidney cancers.^{2–9} Different mechanisms of spread are described, involving the arteriovenous route, the lymphatic system, direct extension and iatrogenic implantation. The majority of penile metastases occur 6–24 months after the detection of the primary tumour. Most patients with penile metastasis present a widespread metastatic disease, poor performance status and a poor prognosis. Around 80% of patients with penile metastasis die within 6 months, irrespective of the primary tumour and treatment.^{5–9} The most frequent symptoms of penile metastasis include pain, penile swelling and priapism due to compromised penile venous drainage. Other symptoms such as urinary retention and haematuria can also occur. In our patient, the metastatic lesions were strictly limited to the penis, which is extremely rare. As penile metastases are described only in case reports, no consensus exists concerning their management. Treatment is usually indicated in a palliated setting and includes mainly chemotherapy, depending on the general health and comorbidities of the patient.^{5–9} Surgery, including partial or total penectomy, is sometimes proposed as a palliative treatment, but is associated with negative psychosocial morbidity.^{10 11} In our case, due to the infiltration of the bulbospongiosus muscle, surgery was judged unfeasible.

Even though the role of radiotherapy remains otherwise unknown in penile metastasis, in our patient, it resulted in rapid control of the symptoms after failure of chemotherapy. The dose our patient received (32.5 Gy) is much higher than that administered in some case reports (20 Gy)¹²; we selected a radiotherapy fractionation scheme that would deliver an efficient dose in a relatively short time and induce efficient control of the disease with low risk of skin toxicity, as radioinduced balanitis can occur even at a palliative dose range. Interestingly, in our patient, radiotherapy resulted not only in rapid pain palliation but also in persistent local control of the disease. Radiotherapy appears thus as a fast, efficient and non-invasive way to improve quality of life of such patients.

Penile metastases are rare but should be investigated in patients with history of genitourinary cancer presenting symptoms such as priapism, penile swelling or penile pain. Little guidance or evidence exists to aid management of metastasis strictly located in the penis. Therefore, radiotherapy could

appear as a promising method in some cases. In our case, radiotherapy resulted in rapid and persistent improvement of symptoms, subjectively and objectively.

Learning points

- ▶ Penile metastasis should be suspected in patients with history of urogenital malignancies presenting with priapism, penile swelling or pain.
- ▶ Penile metastases indicate widespread metastatic disease with a poor prognosis, but in some cases, metastasis is solely to the penis.
- ▶ Radiotherapy should be part of multimodal therapy to ensure a rapid palliation of pain, control of the disease and a quality of life improvement.

Contributors RG, NC and ES were involved in the conceptualisation of the manuscript, collecting patient data, conducting the literature search and drafting the manuscript. MB supervised the radiological collection. All the authors were involved in clinical management of the patient.

Competing interests None declared.

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