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Recurrence of Urothelial Carcinoma in the Penis Treated By Emasculation, A Rare Entity: About A Case

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Abstract **Case Report**

Secondary localization of urothelial carcinoma after cystectomy or total cystoprostatectomy (CPT) on the penis (metastasis) is rare, representing 1 to 8%. Occurring within 18 months of the diagnosis of the primary lesion in about 65% according to the literature. The prognosis is often poor with survival rarely exceeding 20 months. The treatment of cavernous metastases of the penis is multidisciplinary, essentially based on surgery (emasculation or penectomy) and chemotherapy, which provide the best results in terms of overall survival. The authors report a case of metastasis, cavernous location of the penis after cystoprostatectomy in 2016 for high-grade urothelial carcinoma of the bladder, whose anatomy-pathology of the surgical specimen is classified pT3a. The patient underwent emasculation (penectomy plus bilateral total orchiepidymectomy involving the entire scrotum). The authors also discuss the diagnostic, therapeutic and prognostic problems that this secondary location poses.

Keywords: Urothelial carcinoma, Metastases, Corpora Cavernosa, Emasculation.

INTRODUCTION

A rare event in the evolution of bladder carcinoma is that marked by the appearance of cavernous metastasis of the penis. In all cases, the cystectomy specimen shows an extensive or multifocal tumour reaching the bladder neck. Tumour classified as Pt3, pt4 or high grade G3, histo-pronostatic with the presence of vascular emboli. The incidence is 1 to 8% depending on the series.

The first description dates back to 1882 by NEUMANN. Cavernous metastases are often observed within 18 months of the initial tumour in 65% of cases, often in relation to advanced disease. There are multiple routes of dissemination. The prognosis remains poor with survival rarely exceeding 20 months.

We report a case of a 58-year-old patient who underwent Total nephro-ureterectomy (NUT) in 2008 for upper excretory tract tumour, in 2017 CPT for pt1 high grade G3 bladder tumour and on the surgical specimen pt3 with presence of vascular emboli.

OBSERVATION

Mr A. A, 58 years old, followed in our department since 2008 for a tumour of the upper excretory tract having led to a NUT (Pt3), then in 2017 for a recurrence in the bladder the patient has benefited from CPT with extensive Ilio-obturator lymph node dissection with a urinary diversion type ureters to the skin, the pathologist has classified the tumour Pt3a with invasion of the pelvic ureter of 5cm, we noted in the bladder three tumour masses: anterior wall 15mm, upper wall 40 X 30 mm and lateral right of 50 x 27 mm. Another right parietal mass invading the right ureteral meatus and extending to the pelvic urethra on 5 cm high. The operation was simple and the patient was hospitalised for about 15 days. Four years later, the patient presented with an interperineal-scrotal mass.

Regional and locoregional physical examination revealed an arciform endured penis with a feeling of extensive fibrosis and a round, punctiform, shiny wound on the right base of the penis. There is also a large mass in the perineal area at the pelvic urethra.

Lymph node examination revealed no significant abnormalities. Rectal examination was normal.

Biological work-up was requested with correct renal function, haemoglobin level of 14.2 g/dl, platelet level of 199,000/microlitre.

A CT scan was performed on 02/03/2021 finds a tumour process of the perineal and penile urethra invading the corpora cavernosa of 45mm/33mm Fig 1, 2 & 3. A non-progressive adrenal adenoma compared to the 2017 scans. At the thoracic level, two inflammatory, non-specific Barety's lodge nodules.

A biopsy was performed on 07/05/21 in favour of a secondary location of a high-grade urothelial carcinoma of bladder origin (Fig 1).

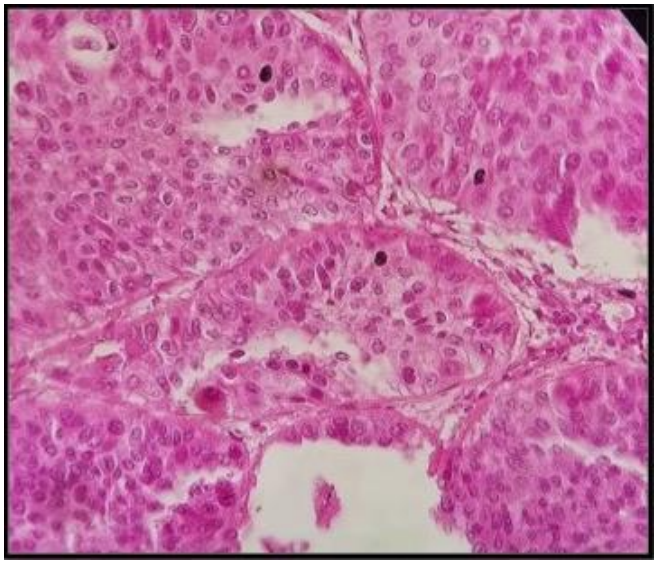

Fig-1: **Microscopic study of the penile lesion showing high grade urothelial carcinoma**

An 18 FDG PET scan was performed on 08/07/2021: in favour of an intense and diffuse hyper metabolism of the penis extended over a height of 115mm, suspected of recurrence.

A focus of hyper metabolism in the hilar and bilateral inters bronchial Barety's lodge of suspected inflammatory origin.

An MRI was performed on 20/07/2021 in favour of a large recurrence of an urothelial tumour involving the urethra along its entire length and invading the left corpora cavernosa with a cystic mass located between the penis and the perineum with nodules of the parietal wall (Fig 2).

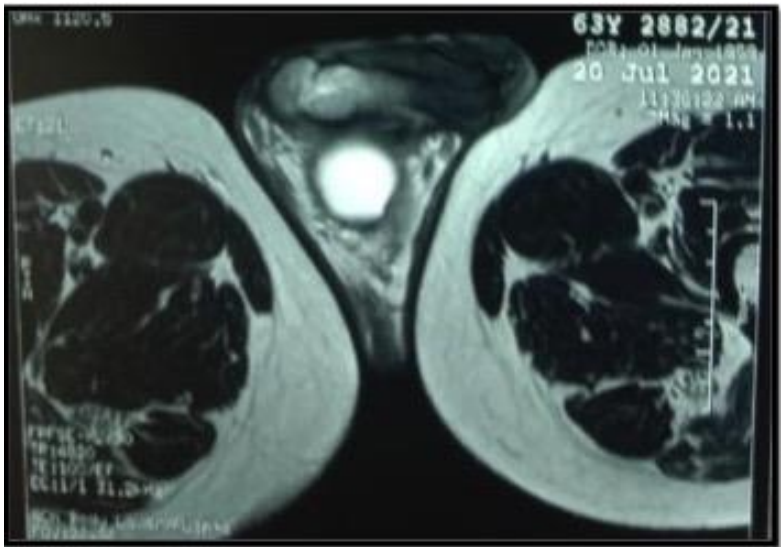

Fig-2: MRI image showing the cavernous lesion and cystic mass located between the penis and the perineum

Following the examinations, and after discussion at the multidisciplinary consultation meetings. The patient was scheduled for emasculation, performed on 29/07/2021(Fig 3).

Fig-3: Image showing the surgical emasculation specimen

The surgical specimen was sent for pathological examination. The surgical specimen was sent for pathological examination finding a high-grade G3 urothelial carcinoma, macroscopically friable, obliterating the urethral lumen and infiltrating the left corpora cavernosa while the right one was spared (Fig 4).

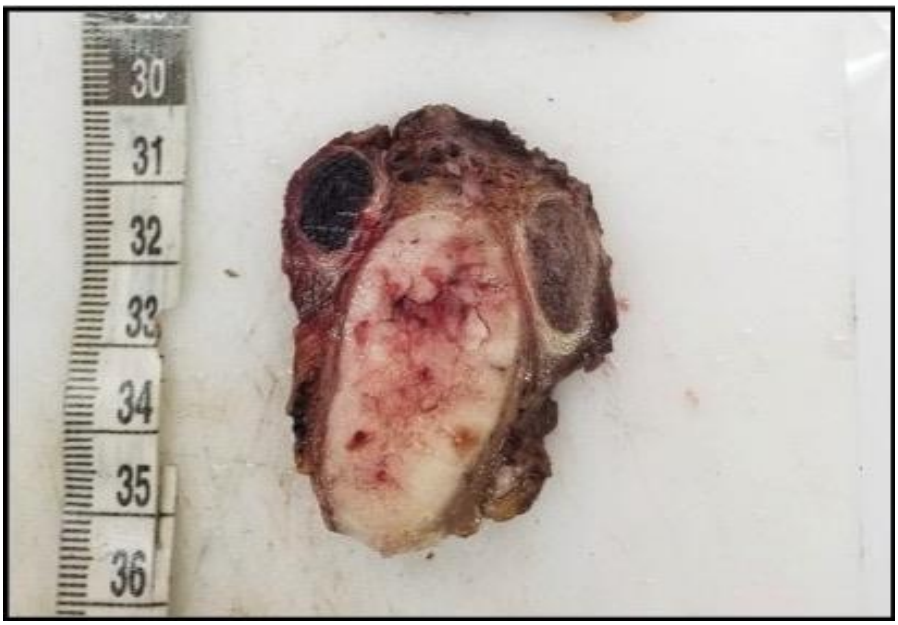

Fig-4: **Image showing the macroscopic appearance of the surgical specimen after fixation**

The patient then underwent several sessions of hyperbaric oxygen therapy to allow better healing (Fig 5).

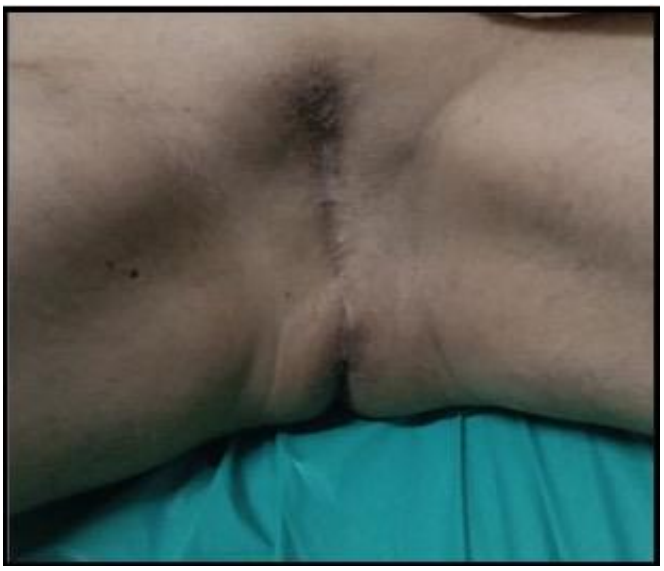

Fig-5: Clinical appearance of the surgical scar after emasculation

DISCUSSION

The first description dates back to 1870 by Hébert of a secondary cavernous location of a rectal carcinoma. Since then, less than 500 cases of penile metastases have been reported in the literature [1]. In the majority of cases, the primary tumour was of urogenital location [2] 75% with bladder cancer in first place 30-35%, followed by prostate cancer 28-30%, kidney cancer 10% and testicular cancer 5%.

Secondary cavernous urothelial carcinomas of the bladder are rare and represent 1 to 8%. They occur more frequently in cavernous bodies than in corpus spungiosum [3] and most often occur within 24 months of the discovery of the primary tumour, more rarely up to 4 years after CPT as in our patient.

The symptomatology in relation to the penile event is dominated by pain, nodular induration of the corpora cavernosa and sometimes a plaque of fibrosis.

Priapism is not unrelated to the penile event and accounts for 38% of cases [2, 3], which can be explained, by local invasion and growth of metastatic cells in the adjacent areas or by neoplastic vascular emboli. Rarely of reflex origin by excitation of the erectile nerve by the tumour process.

Other signs can be observed despite their rarity during the cavernous event, namely acute urine retention [4], haematuria and ulceration of the penis, which is superimposed on our clinical case, and super infections, which can lead to a delay in the diagnosis of the lesions [5].

In a study published by Paquin and Roland, they described the pathways used by the cancer cells to spread to the corpora cavernosa. However, this pathophysiology remains debated. It includes direct extension by contiguity, vascular emboli, and retrograde diffusion by venous or lymphatic route and iatrogenic trauma (RTUV, UIE, RTUP, etc.).

Currently the venous route remains the main route of dissemination of cancer cells and is favoured by the change in blood flow in the dorsal vein of the penis as a result of neoplastic compression and the anastomoses between the bladder, prostate and internal pudendal venous systems.

Arterial dissemination is a possible but very rare route. Instrumental spread by endoscopy is a possible mechanism in bladder or prostate tumours.

The investigations that can be carried out when a cavernous recurrence is discovered are cavernography to confirm the diagnosis and assess the spread of the metastatic disease and for better orientation, but remain invasive: it can be complicated by a haematoma and injection site abscesses. Post-injection fibrosis priapism has been reported.

Ultrasound has its place in penile disorders of inflammatory origin, Lapeyronie's disease and erectile dysfunction, but its use remains limited in cancerous metastases of the penis.

CT has also been used to assess penile metastases; it allows good exploration of the pelvis, but may miss some secondary penile lesions that are limited to the penis, which limits the diagnostic value of CT.

MRI: is a non-invasive examination, more efficient, allows the vision of soft tissues with the possibility of having multiple planes, allows to see directly the penile metastases which present themselves in inhomogeneous forms with hypo signal in T1 and hyper signal in T2.

It then assesses urethral involvement, urethral obstruction and inguinal invasion. MRI is considered the gold standard in the evaluation of metastases. FDG PET is also a diagnostic tool.

The differential diagnosis is with lapeyronie's disease and primary penile cancer (the anamnesis and the shape of the lesion often make the diagnosis evoke) but it is the anatomopathological examinations of the specimen as specified in some studies in the cases of Ucar *et al*., by surgical biopsy that confirm the diagnosis, as was the case with our patient who had 2g of tissue removed.

The treatment of penile metastases is difficult, disappointing, and multidisciplinary and is both etiological and symptomatic.

It also depends on the aetiology of the primary cancer and its sensitivity to different chemotherapies.

Mutthwman *et al*., [6] reported a two-year survival with cisplatin and methotrexate in the treatment of urothelial carcinoma of the bladder.

Radiotherapy is symptomatic and improves comfort and life [6]. Cavernous infiltrations of local anaesthetics are very effective for pain and for 30% of cancer priapism [7].

Excision, which was proposed for cleanliness or in the case of very severe pain, as in our patient's case, is currently, still indicated when the metastasis is single and limited.

Even with surgery and chemotherapy, the prognosis of penile metastases remains poor. The best recurrence-free survival was 2 years with combined surgery and chemotherapy [7].

CONCLUSION

Cavernous metastases of urothelial carcinoma are rare and have a poor prognosis. There may be factors favouring the development of a cavernous metastasis, such as if the patient has had an endourethral instrumental procedure during preoperative bladder resection, if the tumour is locally advanced, multifocal, reaching the bladder neck, with a stage greater than or equal to Pt3, with a histoprognostic grade equal to G3, and with endo-vascular neoplastic emboli (Healthy urethral sections on CPT and healthy prostatic urethra biopsies do not eliminate the risk of penile metastases from penile cancer).

Treatment is multidisciplinary and outcome remains poor with survival rarely exceeding 24 months.

Curative surgery is only appropriate for single and limited penile metastases. The best outcome in terms of overall survival is related to a combination of chemotherapy and surgery.

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