

Case Report

Squamous cell carcinoma arising from suprapubic cystostomy: report of two cases and a narrative review of literature

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ABSTRACT

Squamous cell carcinoma arising from Marjolin's ulcer of the suprapubic cystostomy tract is a rare entity that usually occurs in patients with a history of spinal cord injuries and a chronic indwelling catheter. We present 2 cases of this rare entity with the clinical exams needed for diagnosis and treatment according to individual characteristics of each case. Marjolin's ulcer is a cutaneous malignancy that arises from injured skin. The most frequent type of malignancy identified on histopathologic examination is squamous cell carcinoma (80-90%). It can develop from long-standing scars, chronic wounds, pressure ulcers, osteomyelitis, and burns. Only cases have been reported about Marjolin's ulcer with squamous cell carcinoma at the cystostomy cite. Treatment regimens have not been standardized; cases have received individualized treatments, usually with excision, radiation, or a combination of both. There is insufficient evidence to aid in the understanding of the etiology. In addition, there is no consensus on its optimal treatment and follow-up schemes.

Keywords: Squamous cell carcinoma, Cystostomy, Urinary bladder neoplasms, Spinal cord injuries

INTRODUCTION

Squamous cell carcinoma (SCC) of the bladder is rare in the general population, accounting for less than 5% of all bladder tumors. Nevertheless, it is more frequent in the context of chronic inflammation and infection, as is the case of patients with chronic indwelling urinary catheters, bladder stones, diverticula or schistosomiasis, especially in endemic regions.¹ The first case of SCC of the suprapubic cystostomy tract (SCT) was published in 1993 by Stroumbakis et al.² The patient was an eighty-year-old male who required suprapubic cystostomy due to urethral stricture, and 5 years later he presented with SCC at the cystostomy site with involvement of the entire abdominal wall, but without bladder involvement. Subsequently, and to date, only 14 cases of this condition have been reported around the world.

CASE REPORT

A 71-year-old male with a history of spinal cord injury (SCI) due to a traffic accident in 1969, living with a suprapubic catheter since 1970 (50 years). In 2012, he presented with an exophytic mass at the cystostomy site. A biopsy reported a keratinizing, well-differentiated, infiltrating SCC, without vascular or perineural invasion. One of the ink-marked section edges was in contact with the carcinoma. Bladder involvement was ruled out by cystoscopy and cross-sectional imaging. Plastic surgery was performed for wide tumor resection and reconstruction of the abdominal wall, leaving 1 cm margins. The final pathology report was negative for malignancy. Eight years later, he presented with a new 4x3 cm erythematous, exophytic mass at the same location. A new punch biopsy revealed a keratinizing,

moderately differentiated SCC. Cross-sectional images and endoscopic evaluation of the lower urinary tract ruled out bladder involvement. Computed tomography (CT) of the abdomen and chest found nonspecific bilateral inguinal nodes and a mediastinal lymphadenopathy.

Wide local resection and partial cystectomy were performed in addition to radioisotope-guided sentinel lymph node (SLN) biopsy of the inguinal nodes. It was necessary to perform an en-bloc resection of the cystostomy with adjacent tissue including skin, subcutaneous tissue, fascia, and a partial cystectomy. He required reconstruction of the abdominal wall with primary closure of the defect and placement of prosthetic mesh to reduce the risk of ventral hernia after the procedure. The sentinel tracer (99m-Technetium-nanocolloid, radioactivity 30 MBq) was preoperatively injected peritumorally on the day of surgery 4 hours before the operation. Lymph nodes identified as SLNs by the gamma probe were dissected and sent for pathological analysis. Histopathological macroscopic findings included an exophytic lobulated mass with a central depression, of approximately 4x3.5 cm. Specimen obtained from partial resection of the bladder wall was adhered to the depth of the surgical piece. A fistulous tract was found that crosses the subcutaneous tissue, with a tumor that extends along the fistulous tract to a depth of 4 cm (Figure 1).

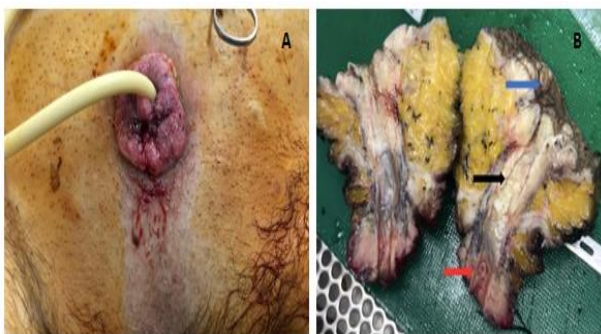


Figure 1: Macroscopic aspect of the tumor. (A) Abdominal mass (60 mm diameter) surrounding a suprapubic catheter. (B) Macroscopic findings included a fistulous tract that crosses the subcutaneous tissue (black arrow) and communicates the tumor on the skin surface (blue arrow) with the bladder (red arrow).

Microscopic evaluation (10X and 100X) showed a well-differentiated keratinizing SCC originating in the cystostomy tract. The invasion extended to the space of Retzius, with an invasion depth of 4 cm, but without bladder involvement. Lymphovascular invasion was found (Figure 2). Regarding SLNs, the right inguinal node was negative for malignancy, while the left inguinal node showed parenchymal metastases of SCC. Subsequently, given that left SLNs were positive, an ilioinguinal lymphadenectomy was performed. The histopathology report was negative for malignancy, so

adjuvant management with radiotherapy was not considered necessary.

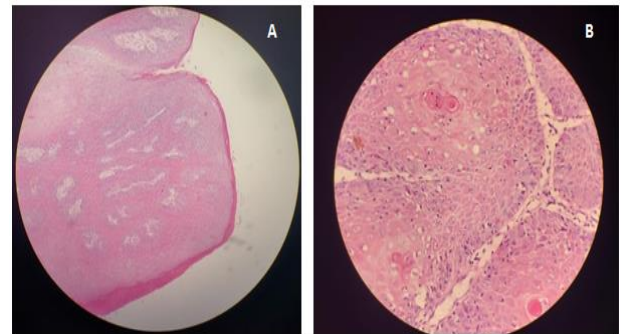


Figure 2: Microscopic findings (hematoxylin and eosin stain). (A) Well-differentiated SCC (B) A high-power microscopic view shows rare nuclear pleomorphism and evident keratinization.

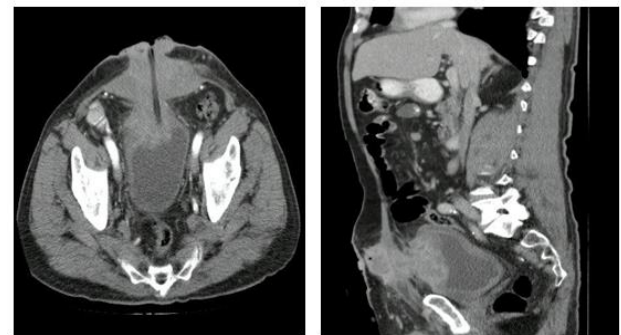


Figure 3: Computed tomography of the abdomen and pelvis, Bladder mass with extension to the soft tissues of the abdominal wall and skin in the cystostomy tract.

An 81-year-old male with a history of benign prostatic obstruction and chronic bladder outlet obstruction with urinary retention was user of a cystostomy tube for over 20 years due to multiple comorbidities that made him a poor surgical candidate. The patient presented with an exophytic mass at the cystostomy site with inflammatory changes characterized by purulent discharge and erythema. A CT of the abdomen and pelvis reported an irregular, multilobulated mass that extended from the anterior wall of the bladder to the soft tissues of the abdominal wall along the cystostomy tract. This mass measured approximately 92x119 mm. The cystostomy tube was surrounded circumferentially by the aforementioned mass. Additionally, suspicious bilateral inguinal nodes were found (Figure 3). A punch biopsy was performed. Microscopic evaluation (10X and 100X) showed an ulcerated, moderately-differentiated, keratinizing SCC, extending to the deep reticular dermis (at least 6 mm), with margins compromised by tumor (Figure 4). Interventional radiology performed biopsy of the left inguinal node, which was positive for malignancy.

Table 1: Published cases of SCC of the suprapubic cystostomy tract.

Author	Published year	Age (years)	Duration suprapubic cystostomy (years)	Bladder involvement	T stage	Treatment	Survival
Stroumbakis et al	1993	80	5	(-)	T4	Radiation and excision	Not described
Stokes et al	1995	50	25	(+)	T4	Excision	Dead at 8 months after surgery
Schaafsma et al	1999	80	5	(-)	T4	Wide excision of mass and partial cystectomy	Dead at 5 months after surgery
Gupta et al	2000	40	20	(+)	T4	Radical cystoprostatectomy, en bloc pubectomy and excision mass with ileal conduit	Survival at a follow-up of 3 months
Ito et al	2011	58	35	(-)	T4	Palliative external radiation therapy	Survival at a follow-up of 6 months
Manzo-Perez BO, et al	2012	34	3 (months)	(+)	T4	Bilateral percutaneous nephrostomy	Dead at 30 days after diagnosis
Chung et al	2013	56	9	(+)	T4	Radiation	Dead at 6 months after radiation
Massaro et al	2014	55	38	(-)	T4	Excision and resection of the tumour of the extra vesicle bladder wall	Not described
Massaro et al	2014	85	9 (months)	(+)	T4	Wide excision of mass and partial cystectomy	Not described
Ranjan et al	2015	68	20	(+)	T4	Radiotherapy	Deat at 4 months after radiation
Zhang et al	2015	61	29	(-)	T4	Radiotherapy	Survival at a follow-up of 3 years
Boaz et al	2015	65	3 (months)	(-)	T3	Penile amputation, wide local excision of scrotum, radical urethrocystoprostatectomy and pelvic lymphanectomy	Survival at a follow-up of 6 months
Subramaniam et al	2017	88	25	(-)	T4	Wide local excision en bloc	Survival at a follow-up of 6 months
Khadhoury et al	2018	53	20	(+)	T4	Excision en bloc with partial cystectomy and colostomy	Survival at a follow-up of 8 months

A chest CT scan ruled out distant metastasis. After a multidisciplinary clinical meeting and considering the patient's functional status and multiple comorbidities, it

was decided to offer the patient palliative external radiation therapy and supportive care.

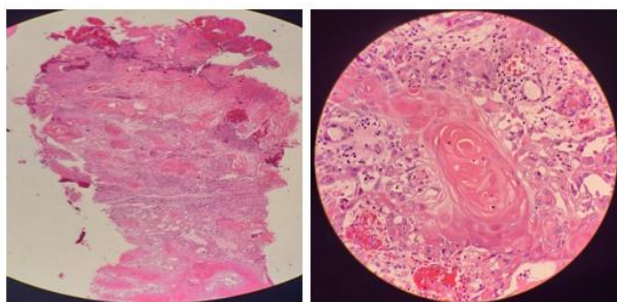


Figure 4: Microscopic findings (hematoxylin and eosin stain), histopathology slide of the biopsy specimen, showing the ulcerated, moderately-differentiated SCC with abundant keratin pearls.

DISCUSSION

Marjolin's ulcer is a cutaneous malignancy that arises from injured skin. The most frequent type of malignancy is squamous cell carcinoma (80-90%); however, other types like basal cell carcinoma (9.6%) and melanoma (2.4%) have also been reported. Its physiopathology is not well defined; nevertheless, it has been proposed that irritation, repetitive re-epithelization, local damage to the skin's immune mechanisms, and genetic predisposition may be involved.³ The most frequent age of presentation is between 30 to 76 years with a mean of 51 years. Most often it originates from burn scars (65%), followed by venous ulcers and trauma scars. The most frequent location is in the lower limbs (62%), followed by the head, face, upper extremities, and torso. Usually, these neoplastic lesions develop in scar tissue between 11 and 41 years (average of 28 years).⁴ There is up to 36% of cases with depth extension with involvement of adjacent tissue. Clinical lymphatic involvement is present in up to 30%, and 7% of the cases are pathologically confirmed. Also, 8% of the patients present with distant metastases.⁴ The incidence of Marjolin's ulcer is approximately 1% to 2% in burn scars. Regarding the incidence of squamous cancer at the cystostomy site, only cases have been reported in this regard and this is why these cases are so relevant. It is usually diagnosed in elderly patients, and it implies a complete en-bloc resection of the tissue surrounding it. It is important to consider primary closure and placement of a prosthetic mesh to repair the abdominal wall defect after the en-bloc resection to avoid future hernias. Complete lymph node excision is an important part of treatment. SLN resection has a high sensitivity to evaluate lymph node metastasis. When complete resection cannot be performed or there are poor prognostic factors, chemotherapy and radiotherapy are an option; however, this option is performed in selected patients due to the risk of radionecrosis.³ In literature on Marjolin's ulcers, 71% of patients are treated with complete tissue resection, amputation in 24%, radiotherapy in 2%, and chemotherapy in less than 1%. Treatment with radiotherapy and chemotherapy does not show a clear benefit and there is still no clear indication.

Similarly, there is no clear benefit with immunomodulatory therapy with cemiplimab.⁴

Kaufman et al. reported that squamous metaplasia is more likely to occur in patients with indwelling catheters for more than 10 years than in those with less than 10 years of use (80% vs. 42%).⁵ The clinical importance of keratinization of squamous metaplasia in the context of permanent bladder catheters remains unclear, but some studies have associated this entity with the development of invasive SCC.⁶ The most common bladder cancer in patients with SCI is SCC (33-46.9%), followed by urothelial carcinoma (31.3-55%) and adenocarcinoma (9.4-10%).⁷ The incidence of SCC of the bladder in patients with permanent catheters for more than 10 years is 10%.² In a study of 48 patients, the average time between SCI and the first diagnosis of bladder cancer was 22.6 years.⁶ The risk of SCC does not decrease with intermittent catheterization since cases have been reported in these patients.^{6,8} The average time from cystostomy catheter insertion and SCC presentation is 31 years.¹ Cancer manifestations appear after a variable latent period of 5 to 25 years, the latency being longer (20-25 years) in young age group and shorter in old age (5-10 years), which supports a strict surveillance protocol in elderly people with chronic catheterization.^{9,10} In SCC of the suprapubic cystostomy tract, the bladder may or may not be compromised by cancerous cells, which will determine the appearance of symptoms (hematuria or positive urine cytology). It is not clear how bladder involvement affects prognosis.⁹ If there is no bladder involvement, the clinical presentation is an irregular skin ulcer or a spontaneous opening of a previously closed fistula.⁹ Of all the reported cases, six have shown bladder involvement.^{9,11-15} Nine of the patients underwent a wide excision of the surrounding skin and the suprapubic tract; of these, six had partial cystectomy, while three underwent radical cystectomy.^{1,2,10,12-14,16,17} Only one patient received preoperative radiation therapy.² Four cases were treated with palliative radiotherapy; one of these cases because the patient had comorbidities that contraindicated management with chemotherapy, and the others rejected other types of treatment.^{6,9,11,18} Two of these cases died at 4 and 6 months, while the other two cases were asymptomatic and without evidence of metastasis at follow-up at 6 months and at 3 years.^{6,9,11,18} One patient with metastasis to bone and right lung base did not accept management with radiotherapy or chemotherapy, therefore, the patient was treated with bilateral nephrostomy and died 30 days after diagnosis (Table 1).¹⁵

Treatment regimens have not been standardized; cases have received individualized treatments, usually with excision, radiation, or both. Regarding other therapies described as neoadjuvant radiotherapy, there is no sufficient evidence to support their routine use. In the case of chemotherapy, an adequate tumor response has not been observed in order for it to be recommended.¹ Management with chemotherapy is considered in patients

with lymph node metastases. In cases where it is contraindicated, external radiation therapy should be considered as a palliative treatment, with which partial remission of the disease and good palliation of symptoms have been described.⁶ To date, no long-term survival benefits have been reported as a result of radiotherapy or chemotherapy.¹¹ There is no consensus on the optimal follow-up strategy. Some authors have recommended annual cystoscopy with biopsy of suspicious intravesical lesions.¹ However, a normal cystoscopy does not exclude bladder involvement.¹³ The prognosis is poor, probably because most patients present with advanced disease at the time of diagnosis.¹¹

CONCLUSION

SCC originating from a Marjolin's ulcer of the SCT is rare. As in the primary SCC of the bladder, it probably arises from chronic inflammation of the tract and surrounding skin. Due to the extreme rarity of this cancer, there is insufficient evidence to aid in the understanding of the etiology, nor there is consensus on its optimal treatment and follow-up schemes. Paying attention to any suspicious sign, including changes in the skin around the cystostomy site is crucial, especially in the presence of an indwelling suprapubic catheter. Given the rarity of these cases and the lack of experience regarding treatment, we recommend these cases to be referred to specialized multidisciplinary oncological clinics for definitive treatment.

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