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Case Study of the Month

Urethra Reconstruction Following Resection of Penile Leiomyosarcoma

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Abstract

Leiomyosarcoma is a rare tumor, particularly in genital area. We present a case of penile leiomyosarcoma. Surgical treatment involved subcutaneous penectomy, preserving a sensate skin envelope, bilateral groin dissection and perineal urethrostomy. Reconstruction of the urethra and soft tissue was carried out using a free radial forearm flap. We believe that subcutaneous penectomy should be considered as a treatment option in selected cases of penile tumor as this facilitates urethral reconstruction.

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1. Case report

A 56-yr-old man with leiomyosarcoma of the penis underwent subcutaneous penectomy and radical inguinal lymphadenectomy bilaterally (Catalona). Subcutaneous penectomy included resection of the corpus spongiosum, corpora cavernosa, urethra, glans, and tunica albuginea. The phallic preservation technique left a sensate penile skin tube. Histology showed a 5-cm tumor of one corpus cavernosum infiltrating the tunica albuginea and the second corpus cavernosum in a mass of spindle cell, malignant, mesenchymal tumor (Fig. 1). Immunohistochemical expression of vimentin by spindle tumor cells was shown (Fig. 2). The tumor was negative for CD31 and factor 8. This was graded as a mitotic active leiomyosarcoma G2 with clear resec-

tion margins. The inguinal lymph glands were free of tumor.

The patient was referred 13 mo after tumor resection for definitive reconstruction. On examination he had a preserved skin tube with sensate penile skin but no erectile soft tissue or urethral component and no glans (Fig. 3). The urethral remnant formed a perineal urethrostomy, with the patient retaining control over the internal urethral sphincter.

The patient's reconstructive requirements included reconstruction of the penile urethra, reconstruction of the glans, and the provision of sufficient soft tissue volume and turgidity to allow intercourse. A single-stage penile and urethral reconstruction using a partly de-epithelialized forearm flap within the preserved sensate penile skin tube was planned to best meet these requirements.

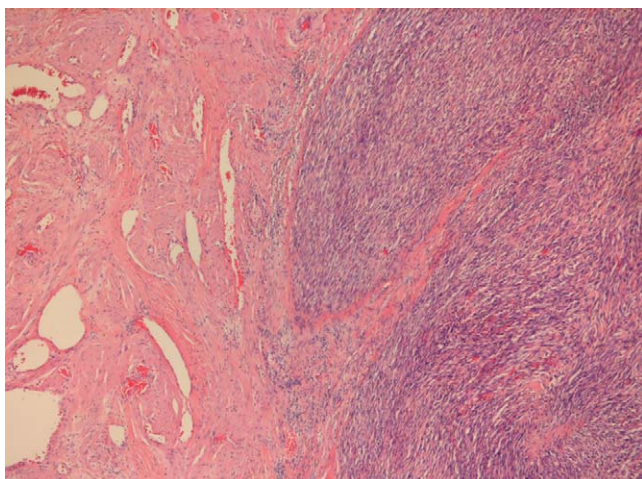


Fig. 1 – Tumor of corpus cavernosum infiltrating the tunica albuginea and the second corpus cavernosum in a mass of spindle cell, malignant, mesenchymal tumor.

Surgery was performed under general anesthesia with two operative teams. One team recreated the defect to be reconstructed by resecting the severe scar formation inside the penile tube (Fig. 4) and prepared the proximal urethra in the perineal region for urethral anastomosis. The femoral vessels were prepared as recipient vessels.

Because no skin was required for external reconstruction, the design of the radial forearm flap was limited to a longitudinal skin paddle to allow tubularization for urethral reconstruction, in addition to a distal transverse paddle for glans reconstruction. The remaining flap skin was de-epithelialized. This preserved the maximum volume of tissue and ensured pliability of the flap during tubularization.

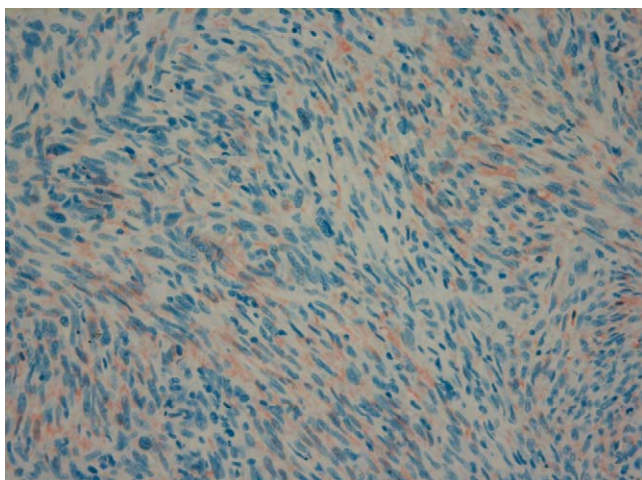


Fig. 2 – Immunohistochemistry. Cytoplasmic expression of smooth muscle actin (SMA) by spindle tumor cells is shown.



Fig. 3 – Patient with total loss of the penis except the sensate penis skin tube.

After the radial forearm flap was raised, the longitudinal skin paddle was tubularized around a catheter (Fig. 5). An additional layer was sutured over the neourethra to maximize waterproofing of the suture line. The de-epithelialized skin and fascia were then wrapped around the neourethra to provide sufficient bulk to reconstruct the penile shaft. The distal skin paddle was shaped to recreate the glans.

Two sensory nerves were included with the forearm flap to provide innervation of the neopenis glans. The flap was raised in a standard fashion and inserted through the penile skin tube. Care was taken to avoid kinking and avulsion of the pedicle. The arterial anastomosis was performed end-to-side and the venous anastomosis in an end-to-end manner. The nerve coaptation was performed to branches of the external pudendal nerves.

The urethral anastomosis was performed to the proximal urethral remnant and the radial forearm fascia was wrapped around the anastomosis to provide waterproofing of the repair. Following inset, the flap perfused well, and a satisfactory aesthetic result was achieved (Fig. 6). The donor site was closed with a full-thickness skin graft from the left lateral groin.

The flap transfer was successful without postoperative complications. After the catheter was removed, the patient was able to void urine on postoperative day 9; he was discharged on postoperative day 10. The patient retained total control over the internal urethral sphincter and was able to void successfully from the tip of the reconstructed penis with no evidence of fistula or stricture.

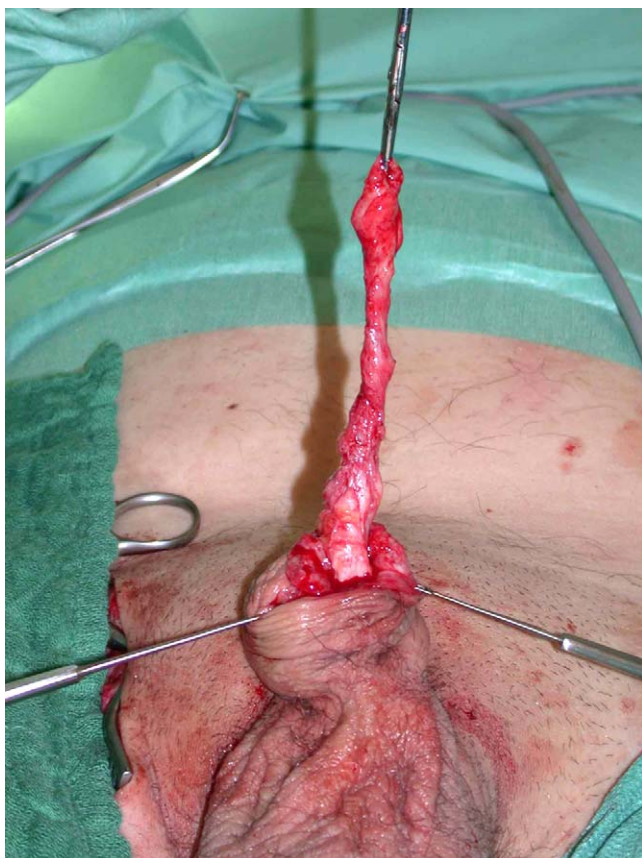


Fig. 4 – Scar excision intraoperatively.

The aesthetic appearance and position of the reconstructed penis was very natural because of the preserved penis skin, which retained sensitivity. Because the native penile skin tube was preserved, the future insertion of a permanent erectile prosthesis remained possible. This was declined by the



Fig. 5 – Urethra construction with creating a tube over a silver catheter.



Fig. 6 – Flap brought in the preserved penis skin, with good distal perfusion.

patient, who had sufficient turgidity to perform intercourse. The patient remains free of any local recurrence or metastasis 41 mo following tumor resection (28 mo after penile reconstruction).

EU-ACME Question

Please visit www.eu-acme.org to answer the below EU-ACME question on-line (the EU-ACME credits will be attributed automatically). The answer will be given in *Case Study of the Month: Part 2*, which will be published in next month's issue of *European Urology*.

Question:

At immunohistochemistry, leiomyosarcoma usually does NOT stain positively for which of the following markers:

- A. Smooth muscle actin
- B. S 100
- C. HMB 45
- D. Vimentin



Previous Month's Discussion and Answer

Urethra Reconstruction Following Resection of Penile Leiomyosarcoma: Part 2

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1. Discussion

Reviewing the literature there is a general agreement about certain goals in penile reconstruction [1–5]. Ideally the neophallus should be natural in appearance and it should possess erogenous and tactile sensation. The neourethra should extend to the distal tip and permit voiding from a standing position. The reconstructed penis should be large enough to accommodate an erectile prosthesis for intercourse if required.

The first published penis reconstruction was a local flap procedure by Bogoras in 1936 [1]. Improved urinary and sexual function has been accomplished with recent microsurgical techniques [6]. Microsurgical reconstruction offers the possibility of penile and urethral reconstruction using cutaneous, fasciocutaneous, musculocutaneous, or osseocutaneous free flaps [6–8]. Definitive reconstruction can be accomplished as either a single-stage or multiple-stage procedure depending on requirements for urethral prelamination or penile prosthesis.

With the described procedure we satisfactorily accomplished these requirements as a one-step delayed reconstruction without the need for additional procedures [1,2,7]. In this case the radial forearm free flap has the following advantages [3,4]: it is easily accessible and has constant and secure blood supply, the skin is thin and pliable and it is capable of accommodating a permanent erectile prosthesis for intercourse. The main disadvantage is an unsightly scar at the skin grafted donor site in a region that is frequently exposed.

Subcutaneous penectomy has previously been described in the treatment of penile urethral carcinoma [9,10]. We believe this is the first occasion that subcutaneous penectomy has been described in the treatment of penile sarcoma, and the first time subcutaneous penectomy has been reconstructed in this manner. At follow up, 41 months postoperatively, the patient has no sign of local recurrence or distant metastasis.

When managing soft tissue tumor of the penis, certain information is required in addition to patient age and general health, including specific information about the tumor site, grade, histopathologic features, and antigenic profile as evidenced by immunohistochemistry. Leiomyosarcoma usually stains positive for smooth muscle markers such

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as desmin, vimentin, and smooth muscle actin (SMA), in addition poorly to occasionally staining positive for cytokeratin markers such as S 100. But leiomyosarcoma usually does not stain positively for HMB 45, which is more commonly a marker for melanoma [11].

Leiomyosarcoma of the penis is a rare tumor [12,13], which may be either superficial or deep. The literature shows that patients with deep tumors tend to develop metastatic disease, whereas superficial tumors have a better prognosis [6,8] with a low rate of local recurrence and metastasis. Long-term survival is reported. Compartmental resection using subcutaneous penectomy is a possible means of tumor treatment in penile leiomyosarcoma. This allows the maximal amount of native skin to be preserved. Oncologic principles of sarcoma surgery dictate that the tumor should be resected including one intact anatomic plane [13]. Because the penis represents a coalescence of multiple anatomic planes from the trunk, we believe that subcutaneous penectomy meets these requirements by providing adequate local control [10] and still leaving an intact and viable penile skin envelope.

If radical oncologic surgery is performed to the penis, it is worth considering subcutaneous penectomy [9,10] because this potentially offers a more acceptable cosmetic, psychological, and reconstructive outcome during cancer removal [10]. This facilitates the reconstructive process, because the main focus of reconstruction is aimed at the urethra. Only sufficient skin for tubularization of the urethra is required and fascia included in the flap is merely used to waterproof the repair and provide bulk inside the preserved penile skin tube. Reconstruction of the glans may be accomplished at the same time.

When using a de-epithelialized radial forearm flap in total penis reconstruction, the competing needs of skin for urethral reconstruction and external skin reconstruction are avoided. In this case of reconstruction following previous subcutaneous penectomy, urethral tubularization was easily obtained. The patient was satisfied with the appearance following reconstruction and was able to void while standing, without leak. He also had preserved erogenous sensation and was able to perform intercourse.

Although delayed reconstruction was performed, immediate reconstruction should also be possible and has the potential advantages of reducing scarring within the penile skin tube and minimizing the negative effect on body image.

EU-ACME question

At immunohistochemistry, leiomyosarcoma usually does NOT stain positively for which of the following markers:

- A. Smooth muscle actin
- B. S 100
- C. HMB 45
- D. Vimentin

Correct answer: C: Leiomyosarcoma usually does not stain positively for HMB 45.

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