Penile Cancer Incidentally Detected During Circumcision: A Case Report
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Citation

Abstract
This report presented the case of an 83-year-old male with penile cancer detected accidentally during circumcision for the purpose of facilitating the insertion of resecting scope for bladder carcinoma. This penile cancer was too close to the collum glandis to be removed with a sufficient surgical safety margin. After due consideration, we performed the circumcision only 2 mm away from the tumor. The pathological examination of the bladder tumor revealed urothelial carcinoma, G2, pTa, and that of the penile tumor revealed SCC, pTaN0M0. We performed a punch out biopsy at six points around the surgical margin at one week postoperatively. However, we did not detect any malignant tissue in the biopsy specimen. This patient did not want additional postoperative treatment, and was followed-up this patient without treatment. This patient is still alive with no recurrence of any tumor at 1 year after the operation.

INTRODUCTION
Cancers of the penis are uncommon tumors that are often devastating for the patient and frequently diagnostically and therapeutically challenging for the urologist. Although rare in North America and Europe, penile malignant neoplasms constitute a substantial health concern in many African, South American, and Asian countries. Penile cancer accounts for 10% to 20% of all male malignant neoplasms among uncircumcised tribes of Africa and in uncircumcised Asian cultures1,2. However, recent reports suggest the incidence of penile cancer to be decreasing in many countries, and the reasons are unclear, but they may be related in part to increased attention to personal hygiene. However, urologists often accidentally detect complete phimosis in elderly males during examinations for other urological diseases in countries that do not practice neonatal circumcision. Urologists perform circumcision routinely in the hope of curing complete phimosis. Circumcision is a common and easy procedure for urologists, and therefore unexpected events are uncommon.

However, on occasions when urologists incidentally detect penile tumors during circumcision, it may be that they do not remember how long they maintained the surgical margin around the tumor. It is not rare that penile tumors are detected incidentally during circumcision of elderly males; however, few case reports regarding such cases exist in the English literature. The reason for this might be the prevalence of neonatal circumcision. For urologists, the therapeutic algorithm is difficult to remember because the incidence of penile cancer is rarer than that of other urological cancers. In cases of pre-scheduled operations for penile cancer, urologists have adequate time to recollect the therapeutic algorithm. However, in cases of incidental circumcision during other operations, for example, transurethral surgery, urologists tend to not anticipate the probability of penile tumor, and it is expected that urologists are puzzled by its sudden appearance.

This paper reports a case of penile tumor that was incidentally diagnosed during the circumcision of an elderly male’s true phimosis.

CASE REPORT
An 83-year-old male visited our urological department with micturition pain. Abdominal ultrasound did not reveal any residual urine, bladder stones or a bladder tumor. Urine cytology was class II. Urinary analysis revealed pyuria, and he was diagnosed with urinary infection and prescribed antibiotic drugs. His pyuria did not improve, and cystoscopy was performed a few days later. Preparation for urethral anesthesia revealed that his preputial hole was too narrow to insert a flexible cystoscope (Figure 1). We were not able to turn over his preputium. His penis showed complete phimosis. His preputial hole was dilated using a metal...
bougie and he then underwent flexible cystoscopy. Cystoscopy revealed a tiny bladder tumor (Figure 2). A physical examination revealed no abnormal findings other than a slightly enlarged prostate. Magnetic resonance imaging of the pelvis did not reveal any lymph node swelling or invasion of bladder tumors into the smooth muscle layer of the bladder. Therefore, he underwent transurethral resection of the bladder tumor (TUR-Bt) and circumcision at the same time, under lumbar anesthesia. Circumcision was performed first. Incising of the prepuce and drawing the drapes revealed a 8 mm verrucous tumor on the excess preputium (Figure 3). However, this tumor was located within only 3 mm from the annular groove. Ocular inspection revealed conclusive penile cancer, but the preoperative explanation did not include the possibility of detecting penile cancer. Therefore, he did not undergo partial amputation and the excess preputium was resected only 2 mm away from tumor. He then underwent TUR-Bt. Pathological investigation of the bladder tumor revealed urothelial carcinoma, G2, pTa, and that of the penile tumor revealed squamous cell carcinoma (SCC) and pTa (Figure 4). The surgical margin was negative. We therefore diagnosed the patient with penile carcinoma, SCC, pTaN0M0 and urothelial carcinoma, G2, pTaN0M0. The penile wound did not recover immediately and became ulcerous. A remnant tumor on the ulcerous site was suspected and punch-out biopsies were obtained from six sites at the center of the ulcerous site. However, no remnant tumor was detected in any of the biopsy specimens. SCC antigen in a blood sample before circumcision was 0.8 ng/ml (< 1.5 ng/ml). Although adjuvant therapy was recommended, the patient did not want to undergo this procedure. The patient is currently being treated as an outpatient at 1 year after presentation.
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Figure 3
Figure 3. Intraoperative finding and excised specimen revealed a verrucous tumor on the excess preputium.

Figure 4
Figure 4. Histological findings demonstrated squamous cell carcinoma. Magnification ×100, (b) Magnification ×400

DISCUSSION
The incidence of penile cancer varies according to circumcision practices, hygienic standards, phimosis, the number of sexual partners, human papillomavirus infection, exposure to tobacco products, and other factors.3-6

Neonatal circumcision is well established as a prophylactic measure that virtually eliminates the occurrence of penile carcinoma because it eliminates the closed preputial environment where penile carcinoma develops. Complete phimosis in Japanese children is conservatively treated without repeated balanoposthitis and ballooning since true phimosis is a non-lethal disease, and complete phimosis in adults are treated only if they opt for circumcision.

However, complete phimosis has a definite disadvantage to pseudo phimosis for early diagnosis of penile cancer. Older patients might be embarrassed to seek appropriate treatment, and therefore circumcision for complete phimosis might be necessary in Japan too. Kelly et al reported adult male surgical circumcision reduces HIV infection in men and is recommend by the WHO for incision in comprehensive health care.7 Complete phimosis in adult males should no longer be allowed to be left untreated.

Penile tumor was detected during circumcision in the current case. The patient was circumcised only 2 mm away from tumor. The question arose in this case as to whether or not a partial penectomy was indicated for a radical cure. In the EAU penile cancer guidelines of 2009, Giorgio et al. stated that negative surgical margins are imperative for penile-conserving treatments, the pathologic assessment of surgical margins is recommended and, in general, a margin of 3 mm is considered safe in cases of Tis, Ta and T1. In cases of T2, the authors stated that partial amputation with a tumor-free margin is considered the standard treatment and a surgical margin of 5-10 mm is considered safe. Moreover, in cases of T3 and T4, total penectomy with perineal urethrostomy is the standard surgical treatment. Parkin et al. reported that patients with T3 and T4 are rare (i.e., 5% in Europe, 13% in Brazil).8 In cases of penile cancer detected incidentally during circumcision, tumors found earlier would be expected to be either benign or in the early stages of penile cancer. Therefore, urologists should perform physical examinations carefully. Moreover, performing partial penectomy without a pathological diagnosis is dangerous because of the possibility that the tumors are benign, such as condylomas.

A search of PubMed revealed no reported cases of penile cancer detected during circumcision for complete phimosis in elderly males. This is because neonatal circumcision has been prevalent in English-speaking countries, and complete phimosis cases in elderly males tend to be very rare.

In the current case, we should have struggled to obtain an additional surgical margin of 1 mm because penile cancer localized within the preputium has little hope of being more than a T1 stage. If this tumor had not localized within the preputium, we could have performed only a tumor biopsy and TUR-Bt.

According to the EAU penile cancer guidelines of 2009, local recurrence during the first two years following treatment with penile preserving surgery has been reported in up to 30% of patients. Follow-up visits are advised every three months in the first two years following penile-preserving treatment. After that, follow-up visits are advised every six months, provided that the patient and his partner have been thoroughly instructed to examine the penis regularly and to return if any abnormality is observed. The patient must continue to perform regular self-examinations even after five years of follow-up visits. Following amputation, a less frequent time interval of follow-up visits every six months during the first two years and every one year during the subsequent three years is advised. The risk of local recurrence is not more than 5%.
Regarding lymph node metastases, regional recurrence occurs more frequently within two years after inguinal lymphadenectomy and sentinel node biopsy. A stringent follow-up with an ultrasound investigation of the groin every three months for two years and every six months during the subsequent three years is advised. Patients managed with a wait-and-see policy have a higher risk of recurrence (9%) than patients staged surgically for negative nodes (2.3%), whether the surgery is performed by traditional lymphadenectomy or dynamic sentinel node biopsy.

Patients treated for lymph node metastases have an increased risk of recurrence (19%). Therefore, if urologists have difficulty remembering the EAU penile cancer guidelines because the frequency of penile cancer is very low, we should recommend that patients receive follow-up visits every three months during the first two years and every six months during the subsequent three years.

Generally, urologists do not tell patients about the risk of penile cancer before circumcision. Moreover, the possibility is extremely low, and the patients might not provide informed consent for partial penectomy. The preoperative explanation for performing circumcision for the purpose of facilitating insertion of the resecting scope was simpler than usual. However, urologists must remember that penile tumors might be detected incidentally during circumcision in situations where the preputium is covered completely. Moreover, urologists must retain knowledge regarding penile cancer in preparation for the accidental detection of penile tumors.

In conclusion, complete phimosis of elderly males is not rare in countries that do not practice circumcision. As patients grow older, the potential for transurethral surgery increases and, in those cases, circumcision might be performed at the same time. Therefore, we must acquire knowledge regarding penile cancer on a daily basis.

References
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