

A Rare Case of Adenosquamous Carcinoma Variant of Squamous Cell Carcinoma of the Scrotum: A Case Report and Literature Review

Bunchong Seubsang, MD

Division of Urology, Department of Surgery, Warinchamrab General Hospital,
Ubon Ratchathani, Thailand

Abstract

Squamous cell carcinoma (SCC) of the scrotum is rare and adenosquamous carcinoma variant of SCC of the scrotum is very rare. It was the first cancer directly associated with occupational exposure when, in 1775, Percivall Pott described it in chimney sweeps in England. SCC of the scrotum is a tumor that is of interest for clinical and historical reasons. Some previous studies indicate that survival of scrotal cancer depends on tumor histology. Surgery with a negative resection margin offers the best hope of cure as adjunctive therapy has not proved useful. Prognosis correlates with the extent of nodal involvement. We report a case of a 63-year old Thai farmer. He presented with a stage A disease, and underwent right hemiscrotectomy.

Keywords: Scrotum, squamous cell carcinoma of the scrotum, treatment, variant

INTRODUCTION

Squamous cell carcinoma (SCC) of the scrotum is uncommon. Very rare variant of this disease, adenosquamous, differs in histology and prognosis, but the treatment is identical to SCC¹. It was the first cancer to be linked to environmental factors when Percivall Pott discovered its unusually frequent occurrence in chimney sweeps in England. The incidence has been reported to be less than 10 cases per year in the United States⁷. SCC is the most common scrotal malignancy². Survival of scrotal cancer depends on tumor histology³. The established treatment for SCC of the scrotum is wide excision with a 2-3 cm margin. Surrounding subcutaneous tissue should be excised with the primary tumor. Resection of the

scrotal contents is rarely necessary for disease control unless involved with tumor⁷. Here, we present the case of adenosquamous carcinoma variant of SCC of the scrotum in Thai farmers.

CASE PRESENTATION

A 63-year old Thai male farmer by profession was presented with the complaints of a small nodule lesion on anterior aspect of the right scrotum, which slowly enlarged but failed to heal. There was no history of sexually transmitted disease or trauma in the scrotal area.

Physical examination revealed a 5.5 × 1.5 cm irregular ulceroproliferative lesion, no contact

Correspondence address: Bunchong Seubsang, MD, Division of Urology, Department of Surgery, Warinchamrab General Hospital, 46 Moo 6, Warinchamrab, Ubon Ratchathani 34190, Thailand ; Telephone: +66 4526 7259; Fax: +66 4526 7258 ; E-mail : bseubsang5599@gmail.com

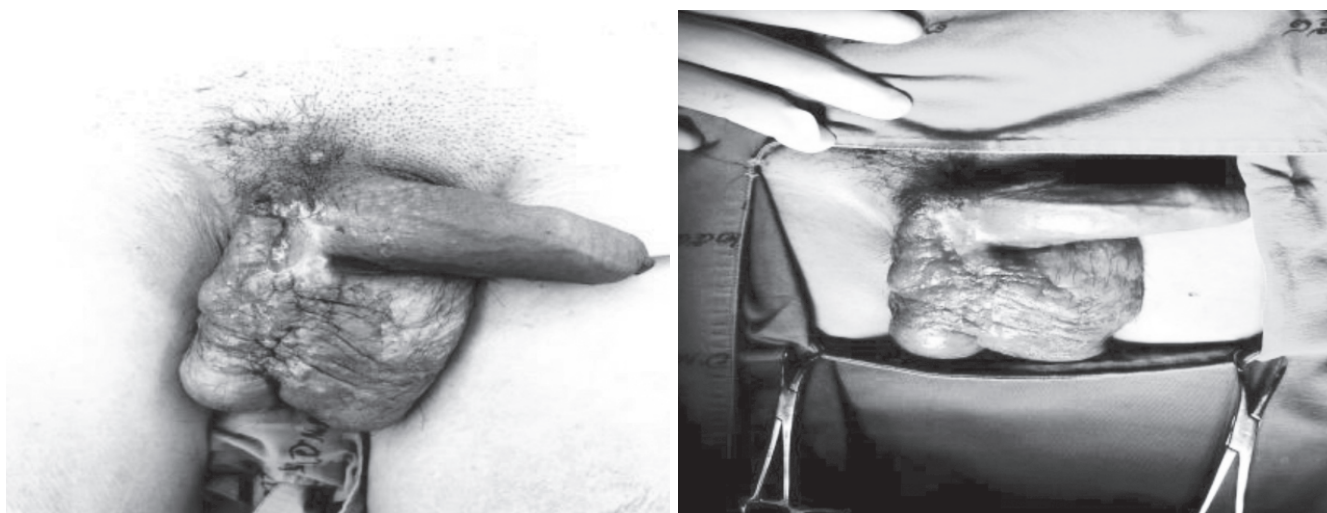


Figure 1 Ulceroproliferative lesion at anterior aspect of the right scrotum



Figure 2 The patient underwent right hemiscrotectomy.



Figure 3 Clinical picture showing well healed right hemiscrotectomy wound

bleeding, no tenderness, hard consistency at anterior aspect of the right scrotum and occupying the right hemiscrotum (Figure 1). There was no associated enlarged inguinal lymph nodes. A wedge biopsy revealed adenosquamous carcinoma.

Ultrasound of whole abdomen showed no evidence of liver metastasis and no enlargement of external iliac chain lymph nodes. Chest X-ray was also normal.

The patient underwent right hemiscrotectomy due to invasion of the tumor to subcutaneous tissue of the right scrotum (Figure 2). The clinical course, post-operative and wound healing, were no complication (Figure 3). The histopathology reported showed adenosquamous carcinoma variant of squamous cell carcinoma of the right scrotum, no involvement of testicular parenchyma and epididymis, free spermatic cord margin, free bilateral margins and free deep, margin (Figure 4).

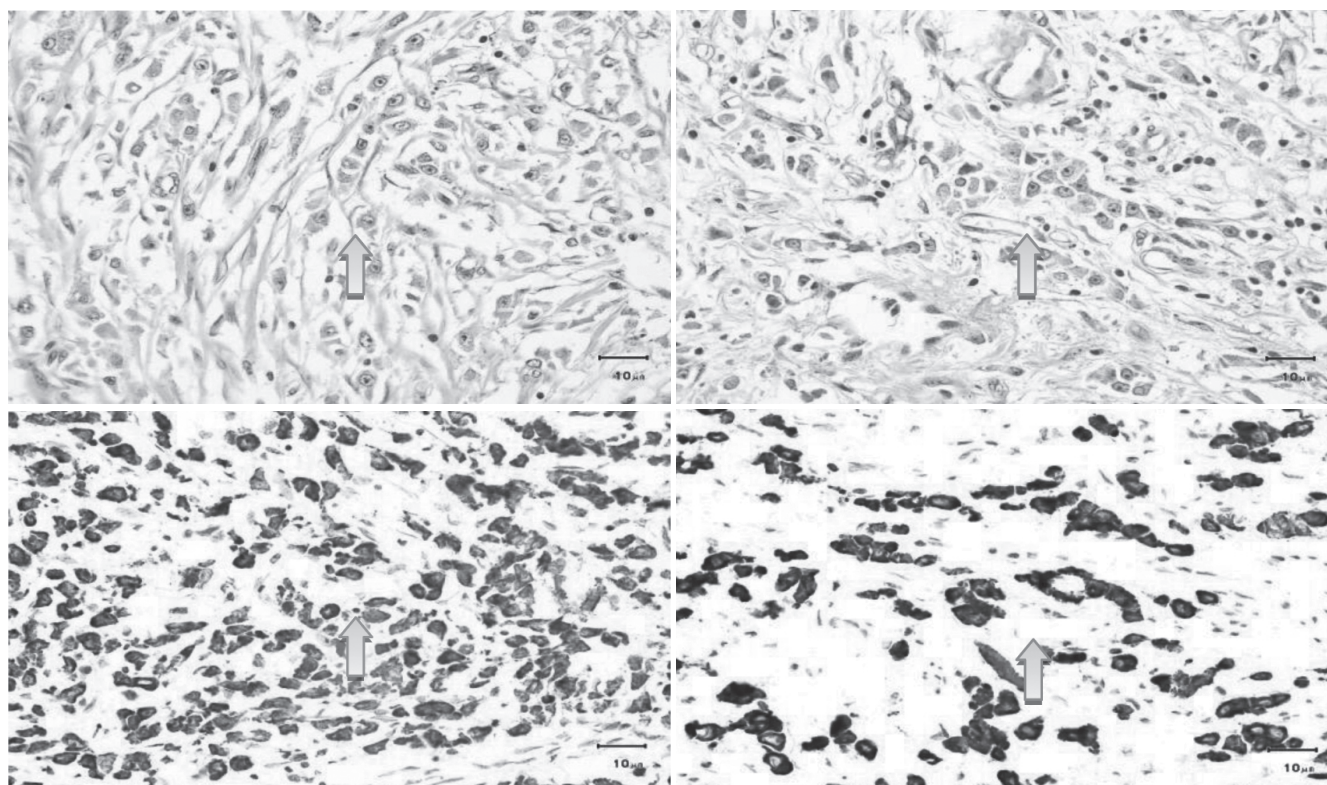


Figure 4 Histological examination of right scrotal mass biopsy showed predominant tumor cells infiltration in cords and severe desmoplasia of the stroma. Some areas of sheets resembling squamous cell carcinoma were noted. Rare well- defined tumor in glands was present. Several lymphatic space invasions were observed. There was skin ulcer with tumor invasion (AE1/AE3: positive).

DISCUSSION

Adenosquamous carcinoma of the scrotum is a rare aggressive variant of SCC of scrotum characterized by the formation of mucin secreting glands. The tumor is composed of invasive tongues and columns of atypical squamous cells, connected with glandular structures with epithelial mucin secretion.

It may be presented as a smooth-surface scrotal nodule or a large, ulcerated, deeply invasive tumor indistinguishable from squamous cell carcinoma or basal cell carcinoma. The clinical course is usually aggressive. The tumors may recur locally or become metastatic. Early detection and superficial location are associated with a better prognosis⁴.

SCC of the scrotum is now very rare. SCC of scrotum was the first cancer linked to occupational exposure. In 1775, Percival Pott described it in chimney sweeps in England. Other occupations that had a preponderance of the disease include people who worked with the distillates of coal and men exposed to mineral oil. Currently, it is rare and most are thought

to be resulted from poor hygiene and chronic irritation⁶.

The overall incidence rate varied around 1.5 per 1,000,000 person per year; most frequently being SCC (27%), basal cell carcinoma (19%) and Bowen's disease (15%). Overall 5-year relative survival was 82%, being 77% and 95% for patients with SCC and basal cell carcinoma, respectively. Overall, 18% of the patients were diagnosed with a second primary tumor².

The mean age of patients is 62 years, ranged from 40 to 73 years. In our case, the age was 63 years. The mean delay in diagnosis for all patients was 22 months, with a range of 2 months to 10 years⁵. In our case, the diagnosis was delayed for 14 months. SCC of the scrotum is frequently presented as a solitary wart or nevus on the scrotum. There seems to be no predilection for any size, if patients with occupational cancer are excluded. Superficial ulceration may follow increase in size or effect of scratching at the lesion. Ulcerated lesions may have a serosanguinous discharge that may become profuse and foul-smell if infected. If an ulcer

is present, it is a malignant squamous ulcer and would have feature expected of such. There may be ipsilateral or bilateral inguinal lymph node enlargement which may be inflammatory or neoplastic or both. In our case the patient was presented with a small nodule on anterior of the right scrotum, which slowly enlarged but failed to heal. There was no associated enlarged inguinal lymph nodes.

SCC of the scrotum tends to remain localized in the scrotal wall, may occasionally involve the adjacent perineal skin but rarely involve the scrotal contents or penis, distant spread is rare⁷. In our case, the lesion occupies the right hemiscrotum, but does not involve testicular parenchyma and epididymis.

The diagnosis is suggested clinically and is confirmed by biopsy of the edge of the scrotal lesion. Refractory scrotal inflammatory lesions should be biopsied to rule out SCC⁷. In our case we confirmed diagnosis by wedge biopsy, and pathological diagnosis was adenosquamous carcinoma variant of SCC.

Scrotal carcinoma staging as proposed by Ray and Whitmor⁶.

Stage A : A1 - Disease localized to the scrotum
: A2 - Locally extensive disease involving adjacent structures (penis, perineum, testis and or cord structures, pubic bone) by continuity but without evidence of metastasis.

Stage B : Regional* metastasis, resectable

Stage C : Regional metastasis, non-resectable

Stage D : Distant metastasis (beyond regional nodes)

*inguinal or ilioinguinal

Our case was in stage A1. Appropriate investigations to exclude any other internal malignancy and distant metastasis include abdominal pelvic ultrasonography and contrast-enhanced computerized tomography of the abdomen and chest. Magnetic resonance imaging (MRI) is also of late being considered a useful tool in the differential of scrotal conditions⁸. In our case, we investigated ultrasonography of whole abdomen, we found no evidence of metastasis.

The established treatment for SCC of the scrotum is wide excision with a 2-3 cm margin. Surrounding subcutaneous tissue should be excised with primary tumor. Resection of the scrotal contents is rarely necessary for disease control unless involved by tumor.

Regarding the treatment of inguinal lymph nodes, the role of regional lymph node dissection remains controversy. Many authors proposed bilateral radical groin dissection to remove micrometastases⁹. In our case we underwent right hemiscrotectomy as the tumor involved skin and soft tissue of right scrotum. We did not perform ilioinguinal dissection because there was no clinically palpable ilioinguinal nodes.

An important prognostic factor is the ability to perform complete excision of the tumor at the time of initial surgery. Prognosis correlates with nodal status. If the inguinal nodes are involved, 5-year survival rate nodes is approximately 25%. There was no survivor if the iliac is effected⁷.

Adjunctive treatment like radiotherapy or chemotherapy or both have not proved useful and only palliation is usually feasible for late stage disease. However, previous studies proposed cisplatin-based chemotherapy combined with bleomycin, methotrexate resulting in stabilization of the disease⁹.

In our case, the diagnosis was established in stage A1. He underwent complete excision and no nodal involvement. His clinical course, post-operative, looked well with favorable prognosis after follow up of three months.

CONCLUSION

We experienced a case of adenosquamous carcinoma variant of SCC of the scrotum, a very rare carcinoma. The diagnosis was confirmed by histology, specific treatment by undergoing right hemiscrotectomy and clinically no metastasis, so our case has a good prognosis. However, patients frequently delay health care consultation; therefore, widespread awareness is essential among the public and medical community of the existence of this disease, because early diagnosis and treatment gives a better prognosis.

REFERENCES

1. Shulstad, Raymond M, Proper, Steven. Squamous cell carcinoma: a review of etiology, pathogenesis, treatment, and variants. J Dermatology Nurses Association 2010;2:12-6.
2. Verhoeven R, Louwman W, Koldewijn E, Demeyere T, Coebergh J. Scrotal cancer: incidence, survival and second primary tumors in the Netherlands since 1989. Br J Cancer 2010;103(9):1462-66.

3. Johnson T, Hsiao W, Delman K, Canter D, Master V. Scrotal cancer survival is influenced by histology: A seer study. World J Urology 2012;31(3):585-90.
4. WHO. WHO classification of tumors: adenosquamous carcinoma skin.
5. Andrews P, Farrow G, Oesterling J. Squamous cell carcinoma of the scrotum: long-term follow up of 14 patients. J Urology 1991;146(5):1299-304.
6. Somnath S, Sanjay D, Suvadip C, Sanjay S. Case report: a rare case of squamous cell carcinoma of scrotum with distant metastasis. Int J Curr Sci Tech 2015;3(7):46-8.
7. Jerome A. A review of the history, epidemiology and treatment of squamous cell carcinoma of the scrotum. Rare Tumor 2009;1(1):47-52.
8. Aparajita M, Pn A Rajdeep S, Sushant V, Vaishali S, Anmol C, Varun J. Case report : squamous cell carcinoma of the scrotum - still an occupational hazard. Indian Association of Occupational Health 2014;18(3):150-52.
9. Rajae N, Yousra A, Hassania A, Fatima Z, Samia A, Nawfel M. Scrotal squamous cell carcinoma: about a rare clinical case and review of literature. Schj Med Case Rep 2015;3 (3): 182-84.

บทคัดย่อ ความชุกของโรคและปัจจัยเสี่ยงของโรคไตเรื้อรังในผู้ป่วยโรคนี้ไต ที่รักษาโดยการผ่าตัดไตข้างใดข้างหนึ่งออกทั้งหมด

บรรจง สืบสังข์, พบ.

แผนกศัลยกรรมระบบปัสสาวะ กลุ่มงานศัลยกรรม โรงพยาบาลวชิรพยาบาล จังหวัดอุบลราชธานี

วัตถุประสงค์: เพื่อศึกษาปัจจัยเสี่ยงที่มีผลต่อการเปลี่ยนแปลงอัตราการกรองของไต (GFR) และความชุกของโรคไตเรื้อรังในการรักษาผู้ป่วยโรคนี้ไต โดยการผ่าตัดไตข้างใดข้างหนึ่งออกทั้งหมด (Nephrectomy)

ผู้ป่วยและวิธีการ: การศึกษาวิจัยครั้งนี้ได้ทำการศึกษาวิจัยแบบย้อนหลัง (Retrospective study) ในผู้ป่วยจำนวน 65 คน ที่ได้รับการวินิจฉัยว่าเป็นโรคนี้ไต และได้รับการรักษาโดยการผ่าตัดไตข้างใดข้างหนึ่งออกทั้งหมดอย่างน้อยเป็นระยะเวลา 1 ปี ก่อนทำการศึกษาวิจัย ได้ทำการศึกษาวิจัยตั้งแต่เดือน มีนาคม 2549 ถึงเดือนกันยายน 2557 การประเมินค่าอัตราการกรองของไตใช้สูตร CKD-EPI (Chronic kidney disease epidemiology collaboration) ทั้งก่อนและหลังการผ่าตัดไตออก ส่วนการวิเคราะห์ทางสถิติเกี่ยวกับปัจจัยเสี่ยงที่มีผลต่ออัตราการกรองของไต ใช้การวิเคราะห์ถดถอยเอกนาม (Univariate regression analysis) และการวิเคราะห์ถดถอยพหุนาม (multivariate regression analysis) ส่วนปัจจัยที่ใช้ในการศึกษาวิจัยครั้งนี้ได้แก่ อายุของผู้ป่วย เพศของผู้ป่วย ดัชนีมวลกาย (BMI) ของผู้ป่วย ค่าอัตราการกรองของไตก่อนการผ่าตัด ระยะเวลาในการติดตามผลการรักษาหลังจากผ่าตัดไตออก และโรคประจำตัวของผู้ป่วย (เบาหวาน, ความดันโลหิตสูง, ภาวะไขมันในกระแสโลหิต)

ผลการศึกษา: พบว่าผู้ป่วยมีความชุกของโรคไตเรื้อรัง 22.64% และพบว่าเพศชายเป็นปัจจัยเสี่ยงต่อการเป็นโรคไตเรื้อรังหลังจากการรักษาโรคนี้ไต โดยการผ่าตัดไตข้างใดข้างหนึ่งออกทั้งหมดอย่างมีนัยสำคัญทางสถิติโดยการวิเคราะห์ทางสถิติ โดยใช้วิธีวิเคราะห์ถดถอยเอกนาม และวิเคราะห์ถดถอยพหุนาม (Odds ratio, 7.619 [1.516 - 38.303], $P = 0.006$; Odds ratio, 7.347 [1.419 - 38.049], $P = 0.017$, ตามลำดับ) การเปลี่ยนแปลงอัตราการกรองของไตไม่มีนัยสำคัญทางสถิติเกี่ยวกับอายุของผู้ป่วย ดัชนีมวลกายของผู้ป่วย ค่าอัตราการกรองของไตก่อนการผ่าตัด ระยะเวลาในการติดตามผลการรักษาหลังจากผ่าตัดไตออก และโรคประจำตัวของผู้ป่วย (เบาหวาน, ความดันโลหิตสูง, ภาวะไขมันในกระแสโลหิต)

สรุปผลการศึกษา: ในการศึกษาวิจัยในครั้งนี้ พบว่าความชุกของโรคไตเรื้อรัง 22.64% และพบว่าเพศชายเป็นปัจจัยเสี่ยงที่มีนัยสำคัญในการเกิดโรคไตเรื้อรังในผู้ป่วยโรคนี้ไต ที่ทำการรักษาโดยการผ่าตัดไตข้างใดข้างหนึ่งออกทั้งหมด