Penile Metastasis from Esophageal Squamous Carcinoma after Curative Resection

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The penis is an uncommon site of metastasis; with only about 300 cases reported in literature. The majority (75%) of primary tumors are located in the pelvis, and they arise from the genitourinary tract and rectum. We report on a patient with esophageal squamous carcinoma who underwent a curative resection and later developed metastatic nodules over the penis and perineum. We believe this is the first instance of this unusual presentation. He was offered palliation with weekly taxanes and had symptomatic relief with this regimen. (Ann Thorac Cardiovasc Surg 2008; 14: 238–241)

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Introduction

Esophageal carcinoma is an aggressive disease with a 5-year survival of 20%–42%1,2 in most series reporting the results of curative resection for locally advanced disease. As the extent of surgical resection increases, with supraradical procedures such as three field lymphadenectomy, the patterns of recurrence are expected to change. Patients who undergo a pathologically curative resection as evinced by negative margins are very likely to fail outside the thorax at distant sites, predominantly affected by hematogenous dissemination.2 The penis is one such site of metastases, as demonstrated by our case. Secondary tumors of the penis are an uncommon occurrence, despite its prolific blood supply, and only about 300 cases have been reported so far.3 A thorough search of published English literature, failed to reveal any other case of esophageal squamous carcinoma, metastatic to this site; we believe ours is the first reported case with this unusual presentation.

Case Report

A 51-year-old male smoker presented to us with a 2-month history of progressive dysphagia. Physical examination revealed a well built man with no palpable cervical adenopathy and excellent effort tolerance. A polypoid tumor extending from 28 to 35 cm was visualized on upper gastrointestinal endoscopy, and a biopsy confirmed it to be a squamous carcinoma. Computed tomography (CT) revealed a localized circumferential thickening of a 4 cm segment of the infracarinal esophagus and paraesophageal adenopathy. Endoscopic ultrasound staged it as uT3N1, the tumor breaching the muscularis with periesophageal nodes. The esophageal mass appeared resectable with intact fat planes all around. He underwent a total esophagectomy with a total mediastinal nodal dissection and abdominal (D2) nodal dissection. The alimentary canal was reconstituted with a gastric tube routed along the posterior mediastinum. The procedure was uneventful except for inadvertent splenic injury necessitating splenectomy. He made an uneventful recovery and was discharged on the 13th postoperative day.

Histopathological examination of the resected specimen showed moderately differentiated squamous carci-
nomma of the middle third of the esophagus breaching the muscularis propria and extending into the adventitia. The circumferential resection margin was focally positive, and there were lymphovascular emboli. Four of 29 lymph nodes dissected showed metastases. The disease was staged as pT3pN1. He received adjuvant external beam radiotherapy (50 gray, 25 fractions) which was well tolerated. A CT scan of the thorax and abdomen performed 2 months after radiotherapy was completed (5 months after surgery) showed a normal postoperative status.

Four months later he presented to us with painful nodules over the penis and perineum. He was in good general condition, and his physical examination revealed hard nodules over the glans penis and the shaft, extending onto the perineum. A fine needle aspiration from one of the nodules confirmed them as metastases from the primary in the esophagus (Fig. 1). Positron emission tomography (PET) scan with CT fusion demonstrated intense tracer uptake (SUV 10.2) involving the perineum in the midline, the root, shaft, and glans penis (Figs. 2 and 3). There were also areas of moderately increased uptake in the left hilar, subcarinal, and left supraclavicular regions. The rest of the whole body survey was unremarkable; notably, the lungs and liver were free of disease.

We offered him palliative chemotherapy (weekly Paclitaxel, 80 mg/m²), because of the pain associated with the penile metastases. At last follow-up, 6 months after initial metastatic presentation (15 months from surgery), he had good symptomatic relief.

Comment

Penile metastases are an uncommon occurrence with slightly more than 300 cases reported. Pelvic malignancies, especially those originating from the genitourinary tract and the rectosigmoid, account for 3/4ths of cases, the rest accounted for by extrapelvic primaries. In a comprehensive review of the 300 reported cases, the most common malignancies metastasizing to the penis in order of frequency were the bladder, prostate, rectum, and rectosigmoid areas, and kidney in 32%, 30%, 13%, and 8% of the cases, respectively. The same review noted that only 16 metastases originated in primary sites above the diaphragm. The extrapelvic organs where the primary originated include the larynx, kidney, pancreas, and lung, and no case of esophageal primary metastasizing to the penis has been reported.

The common modes of presentation are priapism, penile induration, or nodules over the penis. Given the rich vascularity of the penis and the end arterial nature of its supply, it is surprising that metastases do not occur with greater frequency at this site. Penile involvement is usually a marker of widespread hematogenous dissemination with a median survival of less than 6 months after diagnosis. At autopsy, widespread dissemination is noted, and the incidence of isolated penile deposits is sufficiently rare as to excite clinical attention. One-third of all penile metastases are synchronous, whereas the remaining two-thirds are detected a mean of 18 months after discovery of the primary tumor.

This being the first reported case with this unusual presentation, we could find no relevant literature to support the plausible route(s) of metastasis from the esophagus to the penis. Retrograde spread through the pelvic veins has been put forth to explain metastases from pelvic primaries, such as bladder and rectal carcinoma to the penis. Retrograde lymphatic dissemination has also been described, again for pelvic primaries. In sites distant to the penis, such as in this case, arterial dissemination seems to be the likely mode of dissemination; and this has been described by Paquin and Roland.

Our patient had a dominant recurrence in the penis and perineum, associated with failure regionally in the sites not addressed at primary surgery, including the cervical nodes and the opposite mediastinal nodes.

Esophageal cancer remains a lethal disease despite advances in surgical extent and technique with a medi-
Fig. 2. Coronal positron emission tomography-computed tomography (PET-CT) fusion images showing uptake along the penis, indicating recurrent disease.

Fig. 3. Sagittal positron emission tomography-computed tomography (PET-CT) fusion images showing a plaque of disease along the penile shaft and perineum.
an 5-year survival of only 10%, with 20%–40% 5-year survival in localized cases, where curative resection is achieved. Patterns of failure after surgery include local (anastamotic), regional (involving the lymphatic basins), and distant metastases because of blood-borne spread. The lung is the most common organ involved in distant failure, followed by the skeletal system and liver.8) Esophageal cancer is known to metastasize to exotic sites, though these are mostly anecdotal in nature; including the orbit, maxilla, temporal bone, skeletal muscle, pineal gland, skin, and spleen.

The advent of powerful imaging modalities such as the PET scan improves assessment of distant failures and perhaps enables the detection of clinically silent sites of recurrence.

The rarity of penile metastases and the varied origin preclude any firm treatment algorithms. The treatment is essentially palliative, and the combination of radiotherapy with chemotherapy produces only occasional responses.9) Aggressive surgical excision, which may amount to a penectomy, can palliate symptoms, achieve local control, and offer some semblance of cure in patients with disease localized to the penis. The prognosis in most cases, however, is dismal with 80% of patients dying within 6 months because of disseminated disease.

References