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CASE REPORT

Urethral Carcinoma Metastatic to Bone: A Case Report and Literature Review

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Distant urethral carcinoma metastasis is a very rare event. The following discussion presents a unique case of urethral carcinoma (adenocarcinoma) metastatic to bone and reviews the literature regarding this condition. © 1996 Academic Press, Inc.

INTRODUCTION

Urethral carcinoma is an uncommon malignancy with fewer than 1800 cases reported in the literature [1-3]. While many authors [4-10] state that their patients died of "wide metastasis," they fail to document the sites of metastases. Thus, an accurate survey of the types of metastasis cannot be obtained. Clearly, however, bony metastasis is a rare entity; the earliest documented case was reported by Riches and Cullen [11] in 1951. We present what we believe to be the first documented case of a distant bony metastasis in a patient with urethral adenocarcinoma and review the literature regarding this uncommon condition.

CASE REPORT

A 59-year-old African–American female presented in early 1993 with a 5-month history of urinary frequency, urgency, and occasional urge incontinence and subsequently developed urinary retention and proximal muscle weakness. She was evaluated by a neurosurgeon who noted herniated discs on MRI at C4–C5, C7–T1, and T3–T9 with some cord effacement at T5–T6. However, this was not felt to be the etiology of her diffuse proximal muscle weakness. She was diagnosed with polymyositis via muscle biopsy, at which point she was started on prednisone (80 mg every day weaned down to 15 mg every day).

Three months later, she developed hematuria and underwent cystoscopy, bladder biopsies, and anterior vaginal wall biopsy which revealed a well-differentiated adenocarcinoma. On physical examination she had an indurated mass extending from the base of the bladder wall to the distal one-third of the urethra and to the lateral vaginal wall on the right side and midline on the left. A subsequent CT scan revealed an anterior vaginal wall mass extending from the mid urethra to the base of the bladder, pelvic side walls, with some streaking in the perirectal area. There was no adenopathy, and a chest X ray did not reveal lesions suspicious for metastasis.

On 3/1/93, she underwent an anterior exenteration, bilateral pelvic lymph node dissection, left oophorectomy, and creation of continent cutaneous urinary diversion. Pathological report of the specimens noted moderately differentiated adenocarcinoma of the urethra involving the full thickness of the urethral wall and extending into the bladder trigone (Fig. 1). The margins were clear anteriorly and laterally, and there was no evidence of malignancy in the pelvic lymph nodes.

One year later, the patient reported bilateral groin pain, worse on the right, extending to the anterior thigh, leg, and dorsum of her foot, and in 8/94 she also complained

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FIG. 1. Micrograph of biopsy specimen at initial surgical resection revealing moderately differentiated adenocarcinoma ($125\times$, hematoxylin and eosin stain).

FIG. 4. Microscopic evaluation of a sternal biopsy specimen revealing metastatic adenocarcinoma (125×, hematoxylin and eosin stain).

| Urethral Carcinoma Metastatic to Bone | | | | | | |
|---------------------------------------|------|-----------------|--------|-------------------------------------|--------------------|-------------------------|
| Author | Year | No. of patients | Sex | No. of metastases to bone (rate) | Types of carcinoma | Location of metastases |
| Riches [11] | 1951 | 15 19 | M F | 2 (13%) 1 (5%) | 1 SC, 1 TC TC | 1 ribs, 1NS 1 NS |
| Grabstald et al. [16] | 1966 | 79 | F | 3 (3.8%) | 2 AC, 1 SC | 1 pubis, 2 NS |
| Kaplan <i>et al.</i> [21] | 1967 | 11 | М | 1 (9.1%) | UN | NŜ |
| MacKenzie and Whitmore [8] | 1968 | 6 | М | 3 (50%) | 2 EPI | 1 pubis |
| Pointon and Poole-Wilson [13] | 1968 | 10 | М | 1 (10%) | MEL | C2 vertebra |
| Antoniades [24] | 1969 | 41 | F | 1 (2.4%) | NS | NS |
| Waller and Robertson [27] | 1969 | 1 | М | 1 (100%) | SC | Lateral femoral condyle |
| Peterson et al. [12] | 1973 | 49 | F | 2 (4%) | NS | Vertebrae |
| Ray et al. [25] | 1977 | 23 | М | 1 (4%) | EPI | NS |
| Tesluk [28] | 1981 | 3 | F | 1 (33%) | AC | Pelvis |
| Klein et al. [29] | 1983 | 12 | М | 1 (8%) | AC | Pubic rami |
| Hahn et al. [17] | 1991 | 14 | F | 1 (7%) | SC | NS |

 TABLE 1

 Urethral Carcinoma Metastatic to Bone

Note. NS, not specified; AC, adenocarcinoma; EPI, epidermoid; MEL, melanoma; SC, squamous cell; TC, transitional cell; UN, undifferentiated.







FIG. 2. Computerized tomogram of the pelvis demonstrating a right-sided ossified mass.



FIG. 3. Technetium-99 bone scan revealing increased uptake in the right pelvis and lower sternum.

of sternal pain and a palpable lump in the lower sternum. Radiologic work-up including CT scan revealed an ossified mass at the right obturator foramen with increased uptake on bone scan most indicative of myositis ossificans, but a neoplasm could not be ruled out (Fig. 2). A bone scan also revealed an area of increased uptake at the lower sternum and xyphoid process (Fig. 3). A CTguided biopsy of both sites was performed in 10/94 which revealed recurrent urethral adenocarcinoma in the groin and metastatic adenocarcinoma to the sternum (Fig. 4). In 11/94, she underwent 15 cycles of radiation therapy (3500 rads) to both the right obturator fossa and sternum.

The patient continues to have right groin and sternal

pain managed by oral narcotics and remains independent at home with her husband.

DISCUSSION

Urethral carcinomas are uncommon [12-14]. Less than 1800 cases have been reported in the literature [2, 3]. Male urethral carcinoma was first described by Thiaudierre in 1834 [9] and female urethral carcinoma by Boiven and Deuges in 1833 [2]. Female urethral carcinoma accounts for 1% of all female genital tract tumors and 0.02% of all malignancies occurring in females [14-19]. It is the only urological cancer more common in females than in males with a 4:1 to 10:1 ratio [2, 13, 20]. Age of incidence peaks in the fifth and sixth decades with a range of 4-91 years old [1, 3, 6, 13, 16, 18, 20-22].

The male urethra is composed of three parts, prostatic, penile, and bulbocavernous with transitional-cell carcinoma, squamous cell carcinoma, and adenocarcinomas arising more frequently in each, respectively [20, 21, 23]. Distal/anterior tumors have a better prognosis [7, 12, 18–20] and are treated with partial urethrectomy [7]. The female urethra is divided into distal one-third and proximal two-thirds, with squamous cell carcinoma most commonly arising from the distal one-third and adenocarcinomas and transitional cell carcinoma arising from the proximal two-thirds [13, 19, 20, 22]. In both sexes, squamous cell carcinoma, which in turn is more frequently encountered than adenocarcinoma [3, 12, 13, 20, 21, 24].

The etiology of urethral carcinoma is unknown; however, chronic irritation [1, 21], stricture formation [4, 16, 20], and gonorrheal infection [5, 21, 25] have all been implicated.

Metastases can occur through the lymphatics 14 to 30% of the time [5, 26] or hematogenously [1]; however, metastasis to bone does not correlate positively with lymph node metastasis [2, 16, 20]. Few cases of urethral carcinoma metastasis to bone have been documented in the literature (Table 1). We were able to find a total of 10 males and 9 females with average ages of 54 and 58, respectively. The most common type was squamous cell carcinoma which is also the most common urethral carcinoma. There were 10 nonspecified bony metastases, 5 distant bony metastases, and 4 local bony metastases (pelvis), 8 of which were biopsyproven either at time of surgery or at postmortem. Four of the 19 cases of bony metastasis were adenocarcinoma, all of which had either local or nonspecified metastases. Therefore, the present case is the first to document biopsy-proven distant bony metastasis of urethral adenocarcinoma.

Urethral carcinoma metastasis to bone should be suspected in patients with a known primary who present with localized pain and palpable masses if the bone is superficial. The patients may or may not have palpable lymph nodes on examination. A bone scan and CT scan may be helpful in identifying the metastatic lesion. REFERENCES

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