CASE REPORT

Primary apocrine adenocarcinoma of the scrotum with distant metastasis: a case report

Altug Tuncel, MD,[1] Tezcan Sezgin, MD,[1] Nuket Uzum, MD,[2] Omur Ataoglu, MD,[3] Ali Atan, MD[1]

[1]Ministry of Health, Ankara Numune Research and Training Hospital, Third Department of Urology, Ankara, Turkey

[2]Mikro-Pat Pathology Laboratory, Ankara, Turkey

[3]Gazi University School of Medicine, Department of Pathology, Ankara, Turkey

TUNCEL A, SEZGIN T, UZUM N, ATAOGLU O, ATAN A. Primary apocrine adenocarcinoma of the scrotum with distant metastasis: a case report. The Canadian Journal of Urology. 2009;16(5):4860-4862.

A 53-year-old man presented with weakness, loss of weight, pain in upper and lower extremities, and back pain. He had an intermittent abscess like discharge from a left hemi scrotal lesion. Thoraco abdomino pelvic computerized tomography revealed diffuse, multiple and hypodense lesions in the liver parenchyma. Bone scan showed multi metastatic disease of the bone. Complete resection of the scrotal lesion was performed. In histopathological examination, apocrine adenocarcinoma was diagnosed.

Key Words: apocrine adenocarcinoma, scrotum, metastasis

Introduction

Apocrine adenocarcinoma is an extremely rare primary cutaneous malignant neoplasm. It is established as apocrine derivates and it can be differentiated from benign apocrine adenomas, and sweat gland tumors of eccrine origin.[1] The disease most commonly occurs in the axilla, which contains high number of apocrine gland. But it can also arise from apocrine glands in vulva, external auditory meatus, eyelid, scalp, wrist, anterior chest, and nipple.[2-5]

In the English published literature, scrotal localization of the disease has not been encountered yet. We report the first case of advanced stage primary apocrine adenocarcinoma to occur on the scrotum.

Case report

A 53-year-old man presented with weakness, loss of weight, pain in upper and lower extremities, and back pain over a 6 month period. In his medical history, the patient had intermittent abscess like discharge

Accepted for publication March 2009

Acknowledgment
The authors would like to thank Yasemin Eskirecepoglu for her excellent technical assistance during immunohistochemical study.

Address correspondence to Dr. Altug Tuncel, Birlik mahallesi, 74. sokak, 20/2, 06610, Cankaya, Ankara, Turkey

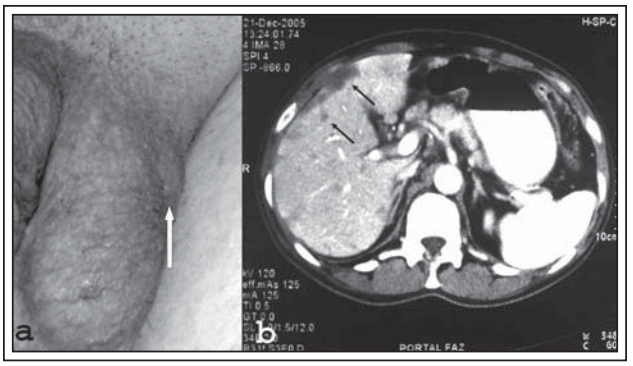

Figure 1. a) Scrotal lesion. b) Liver metastasis in the computerized tomography.

from a left hemi scrotal lesion. The lesion had been available for approximately 5 years and it showed a slow progressive growth without pain.

In physical examination, there was a firm, non tender hyperemic lesion (3 cm x 2.5 cm x 2 cm) protruding from the left hemi scrotum, Figure 1a. The right hemi scrotum was normal. On the left inguinal region, maximum 2 cm x 2 cm in diameter, multiple lymph nodes were palpated. No abnormal findings were found in the penile and testes examination. In the biochemical analysis, aspartate aminotransferase was 69 u/l (normal range, 0 to 50 u/l); alanin aminotransferase was 80 u/l (normal range, 0 to 40 u/l); gamma glutamiltransferase was 352 u/l (normal range, 5 to 64 u/l); alkaline phosphatase was 2429 u/l (normal range, 40 to 150 u/l); total protein was 58 g/l (normal range, 64 to 83 g/l). Although serum prostate antigen, alpha fetoprotein, beta human chorionic gonadotropin, carcinoembryogenic antigen (CEA), and CA 15-3 levels were in normal ranges, CA 19-9 was 462.9 u/ml (reference range, < 39 u/ml).

Cranio thoraco abdomino pelvic computerized tomography revealed diffuse, multiple and hypodense lesions in the liver parenchyma, Figure 1b. There were no abnormal findings in the brain and thorax. Also, whole body bone scan showed increased activity accumulations on the vertebraes, ribs, pelvic region, upper and lower extremities, Figure 2a. These findings consisted with metastatic disease of the bone.

Complementary examinations were performed to exclude another metastasis or malignancy: 1) in the esophago gastro duodenoscopic examination, esophagitis, erosive bulbitis and duodenitis were detected; 2) colonoscopy was normal except for internal and external hemorrhoids.

An incisional biopsy of the left inguinal lymph nodes and complete resection of the scrotal lesion with 2 cm peripheral normal skin were performed.

Histopathological examination

The tumor was located in papillary and reticular dermis that extended to the subcutaneous tissue with infiltrating margins. There was no infiltration in the epidermis. The tumor was arranged in solid sheets and glandular pattern. The cells were round to oval vesicular, hyperchromatic and pleomorphic nuclei with abundant eosinophilic to homogeneous granular cytoplasm. Some cells had nucleoli. The glands demonstrated cribriform features and focal comedo type necrosis, Figure 2b. Relatively few mitotic figures were seen. The stroma was infiltrated with mononuclear, mainly with lymphocytic inflammatory cells. These findings consisted with apocrine adenocarcinoma. The immunohistochemical study revealed a positive staining with pankeratin, CEA and no staining with S-100. Gross cystic disease fluid protein-15 (GCDFP-15) revealed positivity in tumor cells. The biopsy of left inguinal lymph node showed tumor metastasis. The morphology of infiltrating cells was similar in that of the scrotal tumor, Figure 2c.

He did not want to pursue further therapy. He died 9 months after the diagnosis.

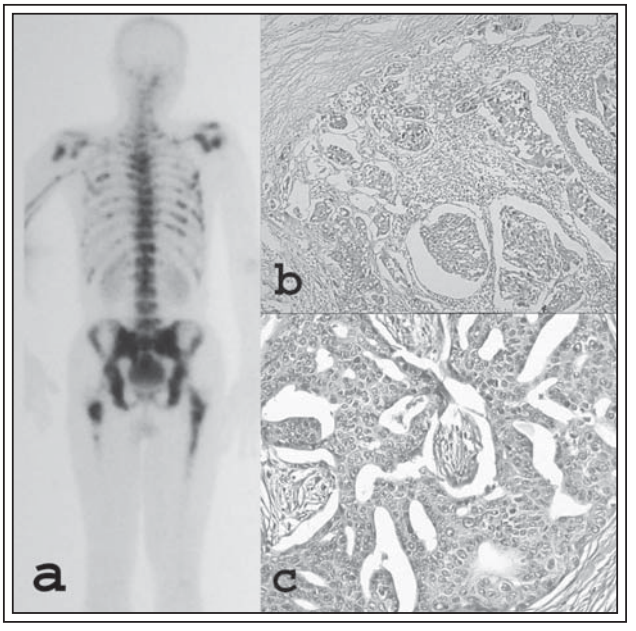

Figure 2. a) Bone metastasis in the whole body bone scan. b) The tumor cells have eosinophilic cytoplasm and vesicular, pleomorphic nuclei forming cribriform features and focal comedo-type necrosis (H&Ex200). c) Small tumor cell groups in metastatic lymph node (H&Ex100).

Discussion

Apocrine adenocarcinoma is an uncommon skin neoplasia with few cases being described in the literature. Because of their rarity, we do not have sufficient knowledge about their clinical occurrence and treatment modalities.

This type of tumor presents as a single or multinodular solid or cystic masses ranging from 1.5 cm to 8.0 cm in diameter.[2,3,6] In history, the lesion generally available for less than a year and the age of the patients usually greater than 50 years.[2,4,7]

Apocrine adenocarcinomas are not encapsulated tumors. They may show different morphology including papillary, anastomosing tubular, solid, and cord like patterns. The nuclei are usually vesicular. Decapitation secretion is found on the apical portion of the cytoplasm of some apocrine secretory cells.[5] In our case, the pattern was mainly glandular and partially solid. Moreover, the cells have abundant eosinophilic, homogeneous to finely granular cytoplasm.

To diagnose apocrine adenocarcinoma, one must exclude metastatic origin. A transitional zone between adenocarcinoma and hyperplastic or adenomatous lesion demonstrating the precursors are helpful in the diagnosis. This finding is also important in differentiating apocrine adenocarcinomas from metastatic lesions.[5,7] We did not observe such a transition in our case.

Immunohistochemistry may be helpful in excluding metastatic cancer. Apocrine adenocarcinoma is immunoreactive for CEA, cytokeratins, GCDFP-15, and negative for S-100.[4,8] This staining pattern was also observed in our case.

Extramammary Paget's disease is another important issue in the differential diagnosis. Extramammary Paget's disease affects less commonly the male genital or perianal area. Histologically, Paget cells are found with in the epidermis. In some instances, the Paget cells are found to be limited to the epidermis.[9] In our case, the Paget cells were not observed in the epidermis.

Wide surgical excision of the lesion is recommended in treatment of apocrine adenocarcinoma.[2-7] In the literature, some cases were treated with adjuvant radiation therapy, but there are not enough reported cases to draw conclusions about the real impact of this combined treatment on patients' survival.[5] There is no report in the international literature on chemotherapy for apocrine adenocarcinoma.

The prognosis of apocrine adenocarcinoma depends on the degree of tumor differentiation.[1] Walker *et* al reported follow up in 10 cases and tumor growth was rapid in patients with unfavorable histopathological results and local lymph node positivity.[2] In our case, advanced stage primary apocrine adenocarcinoma of the scrotum was diagnosed and he did not accept further treatment.

Conclusions

To our knowledge, this case was the first report of an apocrine adenocarcinoma of the scrotum. We believe that this tumor should be taken into account in differential diagnosis in patients with scrotal lesion. □

References

1. Warkel RL, Helwig EB. Apocrine gland adenoma and adenocarcinoma of the axilla. *Arch Dermatol* 1978;114(2):198-203.
2. Warkel RL. Selected apocrine neoplasms. *J Cutan Pathol* 1984;11(5):437-449.
3. Cruz DJ. Sweat gland carcinomas:a comprehensive review. *Semin Diagn Pathol* 1987;4(1):38-74.
4. Zehr KJ, Rubin M, Ratner L. Apocrine adenocarcinoma presenting as a large ulcerated axillary mass. *Dermatol Surg* 1997;23(7):585-587.
5. Vandeweyer E, Hubert A, Renard N. Apocrine adenocarcinoma of the nipple:a case report. *Acta Chir Belg* 2004;104(4):476-478.
6. Paties C, Taccagni GL, Papotti M, Valente G, Zangrandi A, Aloi F. Apocrine carcinoma of the skin. A clinicopathologic, immunocytochemical, and ultrastructural study. *Cancer* 1993;71(2):375-381.
7. Nishikawa Y, Tokusashi Y, Saito Y, Ogawa K, Miyokawa N, Katagiri M. A case of apocrine adenocarcinoma associated with hamartomatous apocrine gland hyperplasia of both axillae. *Am J Surg Pathol* 1994;18(8):832-836.
8. Mazoujian G, Margolis R. Immunohistochemistry of gross cystic disease fluid protein (GCDFP-15) in 65 benign sweat gland tumors of the skin. *Am J Dermatopathol* 1988;10(1):28-35.
9. Kirkham N. Tumors and cysts of the epidermis. Lever's histopathology of the skin. In: Elder DE, Elenitsas R, Johnson BL, Murphy GH (eds). Philadelphia: Lippincott Williams & Wilkins; 2005:805-858.