Ovarian-type Serous Papillary Tumor of the Paratestis Presenting with Calcified Lymph Node Metastases: A Rare Entity

Hilal Sahin, Recep Savas[1], Deniz Nart[2], Burak Turna[3]

Department of Radiology, Tepecik Training and Research Hospital, Departments of [1]Radiology, [2]Pathology and [3]Urology, Faculty of Medicine, Ege University, Izmir, Turkey

Abstract

A 65-year-old male patient was referred to our hospital due to calcified lymphadenopathies in the neck. Positron emission tomography/computed tomography demonstrated a cystic mass in the right scrotum with slightly increased fluorodeoxyglucose uptake in its wall. In addition, calcified hypermetabolic lymph nodes were found in the neck and abdomen. After the operation, the patient was diagnosed as metastatic ovarian-type serous papillary tumor of the paratestis and he was treated with chemotherapy. This case demonstrates that when calcified lymph nodes are present in a male, he should be examined carefully for the primary malignancy which can be determined in an unusual location such as the scrotum.

Keywords: *Calcification, fluorodeoxyglucose positron emission tomography/computed tomography, lymph node metastasis, paratestis, serous neoplasm, tunica vaginalis*

Introduction

Malignant lesions arising from paratesticular region are rare and diagnostically challenging. Ovarian-type serous tumors are the most common subtype, with <50 cases reported in the literature so far.[1] Most of those paratesticular serous neoplasms are borderline serous tumors which behave in a benign fashion.[2] On the other hand, serous carcinomas of the paratesticular region are extremely rare, and they also have metastatic potentials as their ovarian counterparts. However, metastasis is reported as a late event for those tumors in the literature.[1,3] In this case report, we present a male patient with ovarian-type serous papillary carcinoma of the tunica vaginalis with calcified abdominal and cervical lymph node metastases at initial presentation.

Case Report

A 65-year-old male patient presented with swelling in the left supraclavicular region. Ultrasonography (USG) examination showed enlarged lymph nodes, with central amorphous calcification in the left posterior cervical region. The thyroid gland was normal sonographically. The biopsy of lymph nodes revealed high suspicion for malignancy; however, the primary site of malignancy was not determined by fine needle aspiration biopsy.

An ^{18}F-fluorodeoxyglucose (FDG) positron emission tomography/computed tomography (PET/CT) scan was performed to search the primary malignancy. Increased FDG uptake was seen in the left posterior cervical and supraclavicular calcified lymph nodes (maximum standard uptake value [SUV_{max} = 4.5]), the right posterior cervical noncalcified lymph node (SUV_{max} = 2.6), and paraaortic–paracaval calcified lymph nodes (SUV_{max} = 2.8) [Figure 1]. In addition, a huge cystic mass with peripherally increased FDG uptake (SUV_{max} = 2.6) was demonstrated in the right scrotum.

Genitourinary examination revealed a firm 10 cm right scrotal mass. Routine complete blood count, blood chemistry, beta-human chorionic gonadotropin and alpha-fetoprotein levels, and urine analysis were unremarkable. Scrotal USG demonstrated an 11-cm paratesticular cystic mass with hyperechoic papillary projections protruding into the cyst lumen [Figure 2]. There was slightly mobile echogenic material in the cyst, causing a semisolid appearance without showing any vascularity in Doppler sonographic imaging. The right testis was seen as a compressed distinct

How to cite this article: Sahin H, Savas R, Nart D, Turna B. Ovarian-type serous papillary tumor of the paratestis presenting with calcified lymph node metastases: A rare entity. Clin Cancer Investig J 2018;7:234-7.

Address for correspondence:
Dr. Hilal Sahin, Department of Radiology, Tepecik Training and Research Hospital, Yenişehir, Izmir, Turkey.
E-mail: hilalcimen@gmail.com

Access this article online

Website: www.ccij-online.org

DOI: 10.4103/ccij.ccij_97_18

Quick Response Code:

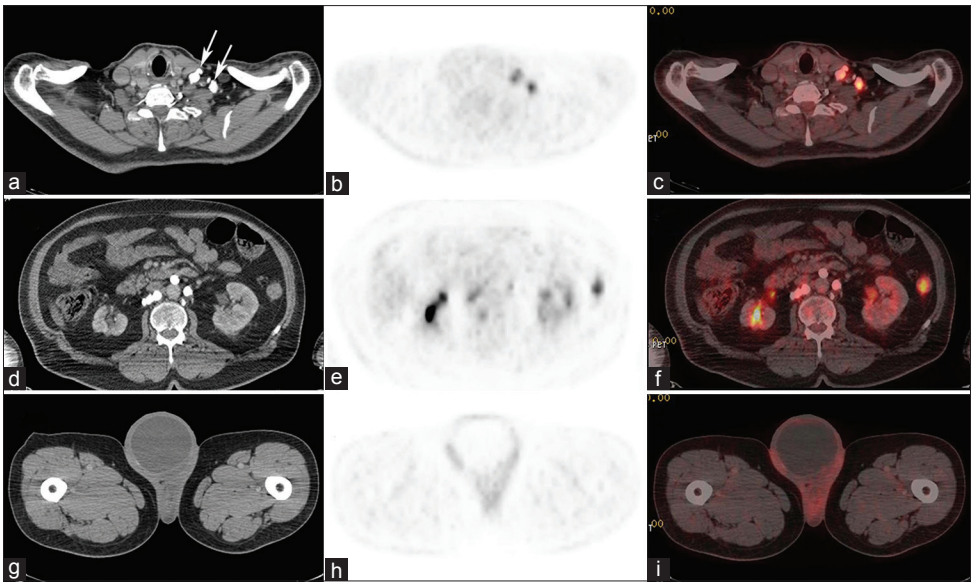

Figure 1: 18F-fluorodeoxyglucose positron emission tomography/computed tomography images showing left supraclavicular calcified lymph nodes with avid fluorodeoxyglucose uptake (arrows) (a-c) and paraaortic calcified lymph nodes with slight fluorodeoxyglucose uptake (d-f). A cystic mass is seen in the right scrotum with slightly increased fluorodeoxyglucose uptake (g-i)

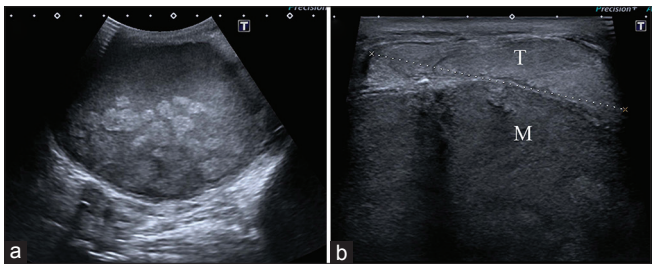

Figure 2: Ultrasonography of the right scrotal cystic mass shows hyperechoic papillary structures with echogenic debris (a). The right testis (T) is seen as a compressed structure with homogeneous echogenicity adjacent to the scrotal mass (M) (b)

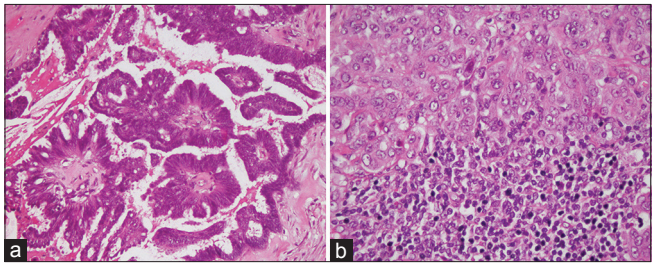

Figure 3: Photomicrograph showing a multinodular tumor with well-defined papillary glandular architecture lined by cells with irregular nuclei and prominent nucleoli (H and E, ×40) (a). A cluster of tumor cells and psammoma bodies are seen in the metastatic lymph node (H and E, ×100) (b)

structure adjacent to the cystic mass. The left testis and the scrotum were sonographically normal.

The patient underwent right radical orchiectomy with paratesticular mass excision and left cervical lymph node excision in the same session. Pathological examination revealed malignant epithelial neoplasm both in the paratesticular mass and cervical lymph nodes [Figure 3]. Immunohistochemical profile was positive for estrogen receptor, cytokeratin 7 (CK7), Wilms' tumor 1, Ber-EP4, cancer antigen 125 (CA-125), and PAX8 and negative for calretinin, CK5/6, CK20, thyroglobulin, thyroid transcription factor-1, and carcinoembryonic antigen [Figure 4]. The final diagnosis was ovarian-type serous papillary carcinoma originating from tunica vaginalis, with lymph node metastases. The patient was treated with three cycles of a chemotherapeutic regimen, including paclitaxel and carboplatin. Although CA-125 level was slightly high after the operation (42 U/ml), following chemotherapy treatment, it became normal. The patient was alive without recurrence in 12-month follow-up.

Discussion

Serous papillary carcinomas of the paratesticular region are rare malignant tumors which are seen in a wide age group, ranging from 6 to 87 years old.[4,5] Most patients present with hemiscrotal swelling or a testicular mass associated with hydrocele. These tumors share similar morphologic, immunohistochemical, and ultrastructural features with serous tumors arising from the female genital tract.[6] The histogenesis of those tumors is still a matter of debate. Favorite theories suggest the origin of those tumors either from Mullerian rests in paratesticular soft tissue or appendix testis, or from tunica vaginalis which shows Mullerian metaplasia.[2,3]

Pathological and immunohistochemical features of ovarian-type serous papillary carcinomas of the paratesticular region are well defined in the pathology literature; however, radiological features of those tumors are less emphasized. Ultrasonographic features may range from a hydrocele to multiloculated cystic heterogeneous masses. A solid mass in a hydrocele can also be seen.[7]

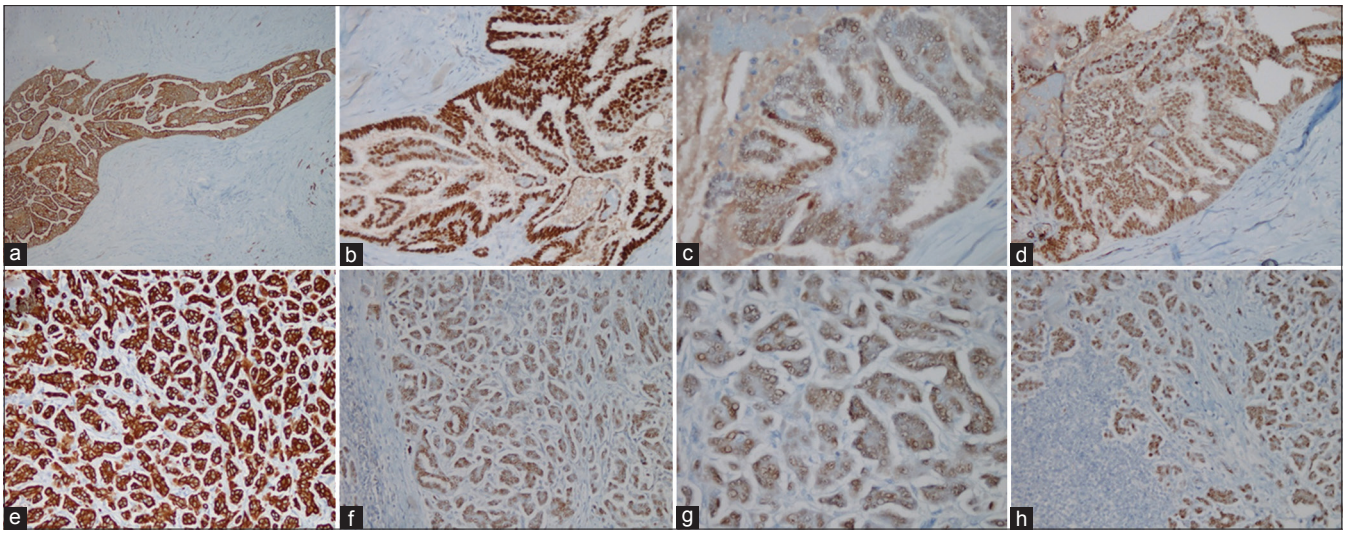

Figure 4: Immunohistochemistry showing the tumor in the upper line and the lymph node in the lower line. Both of them were positive with cytokeratin 7 (a and e), estrogen receptor (b and f), PAX-8 (c and g), and Wilms' tumor 1 stain (d and h) regarding the epithelial Mullerian origin

In addition, foci of calcification may occasionally be seen sonographically.[1,8] In our case, hyperechoic papillary nodular projections were demonstrable in the scrotal mass; however, there was not any visible calcification.

The majority of the reported serous paratesticular tumors are borderline serous tumors which do not tend to recur or metastasize. Conversely, serous carcinomas have the potential to metastasize, but this is reported as a late event in the literature with at least 4-year interval between the initial presentation and development of clinically apparent metastatic disease.[1,3] Of the four reported metastatic cases, two had cervical lymph node metastases and two had diffuse abdominal metastases additionally with leptomeningeal metastasis in one, in the follow-up period.[1,3,9] The case presented herein is unique in that, to our knowledge, it is the first case of ovarian-type serous papillary carcinoma of the paratestis with metastases at the initial presentation.

None of the paratesticular serous adenocarcinoma cases in the literature, up to now, have relevant PET/CT data. In our case, both the primary tumor and metastases had slightly increased SUV_{max} values, however it was less than their ovarian counterparts. The median SUV_{max} for serous ovarian cancer was found as 6.92 in the study of Tanizaki *et al*.[10] However, SUV_{max} value was 2.6 in the periphery of the primary tumor in our case. This finding is of great interest to us since it may be related to different tumor properties of paratesticular serous adenocarcinomas apart from their ovarian counterparts. Unfortunately, PET/CT imaging features of those groups of tumors in males are not defined in the literature according to our knowledge. Still, PET/CT has an important value in those tumors in investigating metastases.

Calcification in metastatic lymph nodes is another interesting point in our case. When calcified cervical lymphadenopathy is determined in a patient, papillary thyroid cancer should be ruled out at first.[11] Then, other primary tumors with calcification potential should be searched such as mucinous colorectal cancers. In the reported case of serous papillary adenocarcinoma of the tunica vaginalis by Blumberg *et al.*, and in our case, calcification was present in metastatic lymph nodes.[3] Ovarian serous cystadenocarcinoma has a high incidence of calcified metastases which are predominantly psammomatous.[12] Paratesticular counterparts may also behave in a similar way, as in our case, regarding serous papillary nature.

Conclusion

Paratesticular serous papillary carcinoma of the tunica vaginalis is an extremely rare tumor with possible metastatic potential. Radiologists and clinicians should keep the possibility of paratesticular serous papillary carcinoma in mind when calcified lymph node metastases are found in the body. FDG PET/CT imaging is helpful in those cases in the assessment of distant metastasis.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Ma YT, Chaudhri S, Cullen MH. Metastatic serous carcinoma of the testis: A case report and review of the literature. Case Rep Oncol 2011;4:246-9.
2. Amin MB. Selected other problematic testicular and paratesticular lesions: Rete testis neoplasms and pseudotumors, mesothelial lesions and secondary tumors. Mod Pathol 2005;18 Suppl 2:S131-45.
3. Blumberg HM, Hendrix LE. Serous papillary adenocarcinoma of the tunica vaginalis of the testis with metastasis. Cancer 1991;67:1450-3.
4. Kurian RR, Prema NS, Belthazar A. Paratesticular papillary serous cystadenocarcinoma – A case report. Indian J Pathol Microbiol 2006;49:36-7.
5. Anchala PR, Dhir R, Parwani AV, Zynger DL. Immunohistochemical profile of paratesticular serous papillary adenocarcinoma and tunica vaginalis facilitates distinction from malignant mesothelioma. Int J Surg Pathol 2011;19:692-8.
6. Narang V, Sood N, Garg B, Aulakh BS. Paratesticular papillary serous cystadenocarcinoma: A rare entity in Indian population. Indian J Pathol Microbiol 2014;57:614-6.
7. Gerscovich EO, Naderi S, Gandour-Edwards RF. Serous papillary carcinoma of the tunica vaginalis testis. J Ultrasound Med 2011;30:418-20.
8. Pease GF, Chou PM. Serous carcinoma of the testicle and the paratesticular tissue: Case report and literature review. Urology 2018;111:183-5.
9. Jones MA, Young RH, Srigley JR, Scully RE. Paratesticular serous papillary carcinoma. A report of six cases. Am J Surg Pathol 1995;19:1359-65.
10. Tanizaki Y, Kobayashi A, Shiro M, Ota N, Takano R, Mabuchi Y, *et al.* Diagnostic value of preoperative SUVmax on FDG-PET/CT for the detection of ovarian cancer. Int J Gynecol Cancer 2014;24:454-60.
11. Ahuja AT, Ying M, Ho SY, Antonio G, Lee YP, King AD, *et al.* Ultrasound of malignant cervical lymph nodes. Cancer Imaging 2008;8:48-56.
12. Wen J, Miao Y, Wang S, Tong R, Zhao Z, Wu J, *et al.* Calcification: A Disregarded or ignored issue in the gynecologic tumor microenvironments. Int J Gynecol Cancer 2018;28:486-92.