



Metastasis to The Tunica Vaginalis as an Initial Manifestation of Occult Hepatoid Type Gastric Cancer: A Case Report

Tunika Vajinalis Metastazı ile Prezante Olan Okkult Hepatoid Tip Mide Kanseri: Olgu Sunumu

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ABSTRACT

Paratesticular metastases are extremely rare. Metastases in this area usually appear in the advanced stages of a known malignancy, but are sometimes the first manifestation, mimicking a primary testicular tumor. In this article, we present a patient with metastasis to the right tunica vaginalis testis who had scrotal swelling and pain as the first manifestation of an alpha-fetoprotein (AFP)-producing hepatoid type gastric cancer.

Key words: alpha-fetoprotein; hepatoid type gastric adenocarcinoma; tunica vaginalis

ÖZET

Paratestiküler metastazlar oldukça nadirdir. Bu alana olan metastazlar çoğunlukla bilinen kanserin ileri evreleri şeklinde izlenmekle birlikte nadiren de olsa primer testiküler tümörü taklit edebilir. Bu makalede, alfa fetoprotein üreten hepatoid tip mide kanserinin, skrotal ağrı ve şişliğin eşlik ettiği tunika vaginalis metastazı şeklinde prezante olması sunulmaktadır.

Anahtar kelimeler: alfa fetoprotein; hepatoid tip mide kanseri; tunika vaginalis

Introduction

Both primary and metastatic, benign or malignant tumours of the tunica vaginalis testis are extremely rare^{1,2}. Only a few cases of metastasis to the tunica vaginalis testis have been reported. The prostate, stomach, rectum, cecum, and colon are the most common primary sites for tunica vaginalis testis metastasis²⁻⁷.

We report an extremely rare case of alpha-fetoprotein (AFP)-producing hepatoid type gastric cancer, presenting with scrotal swelling as the first manifestation due to metastasis to the tunica vaginalis testis. Computed tomography (CT) and magnetic resonance imaging (MRI) findings are reviewed.

Case Report

A 51-year-old man who had been suffering from scrotal pain and swelling for one month was admitted to our urology department. His laboratory tests revealed high serum AFP levels (49.11 ng/ml, range 0–8.04 ng/ml) and he was investigated for a likely malignant testicular tumour. Ultrasonography showed no testicular mass, and the only finding was diffuse thickening of tunica vaginalis testis and spermatic cord with hydrocele in the right side. MRI showed diffuse thickening and irregularity of the right spermatic cord and tunica vaginalis testis, and hydrocele. However, the appearance of the testis and epididymis was normal (Fig. 1).

Therefore, the patient was investigated for a potential primary tumour which might have metastasized to the tunica vaginalis testis. An abdominal CT scan showed peritoneal carcinomatosis with some peritoneal fluid. In addition to these findings, mild gastric wall thickening at antrum was revealed (Fig. 2). Subsequently, a gastroscopy was performed, mucosal oedema especially at the antrum and the body of stomach was observed, and serial biopsies were obtained. A concurrent biopsy from the tunica vaginalis testis was performed for definitive diagnosis.

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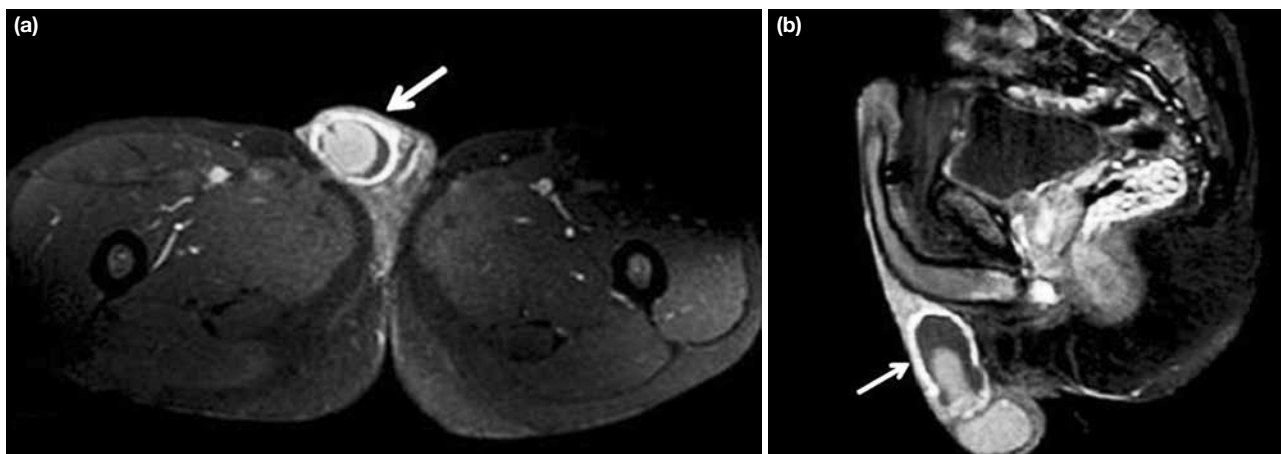


Figure 1. a, b. Contrast-enhanced axial T1-weighted MR image with fat-suppression showed intensive enhancement and irregular thickening of the right tunica vaginalis (a, arrow). Sagittal plane of contrast-enhanced T1-weighted image with fat-suppression showed heterogeneous enhancement of the tunica vaginalis testis (b, arrow) and accompanying hydrocele.

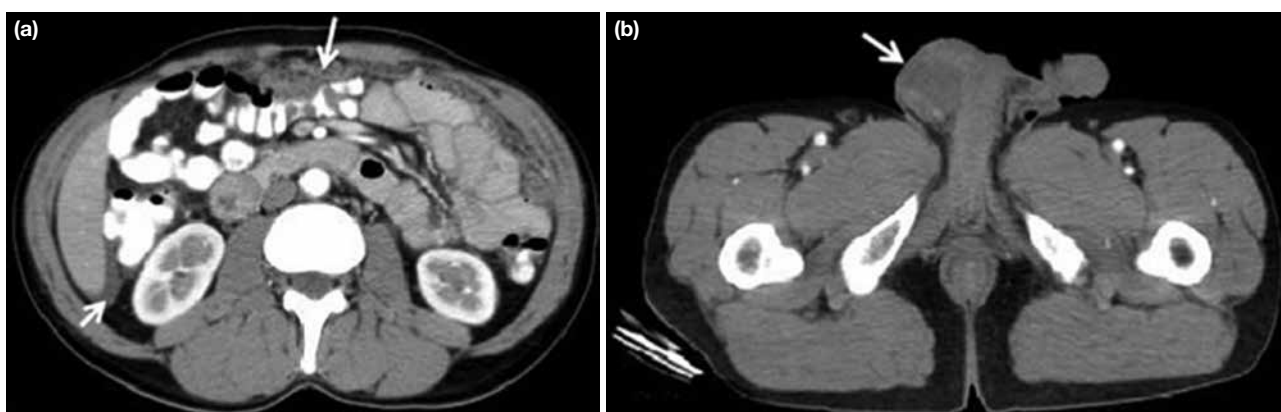


Figure 2. a, b. 51-year-old male with right tunica vaginalis metastasis from gastric cancer. Axial contrast-enhanced CT scan showed peritoneal thickening, omental cake appearance and subhepatic fluid (a, arrow). Heterogeneous enhancement and irregular thickening of the right tunica vaginalis is seen on the lower sections (b, arrow).

Histopathologic examination revealed neoplastic cell nests and abortive glands within the tunica vaginalis (Fig. 3), while immunohistochemical study showed that the tumour cells were positive for pan-cytokeratin (pan-CK), AFP and negative for PLAP, hCG, CD117, Melan A, and S100 (Fig. 4). Specimens of gastric mucosa had neoplastic cells with eosinophilic cytoplasm and round nuclei, occasionally exhibited obvious nucleoli and had positive staining for AFP. The findings were compatible with hepatoid type gastric cancer (Fig. 5). In the light of biopsy results, the patient was diagnosed with a metastatic hepatoid type gastric cancer to the tunica vaginalis testis.

Discussion

The tunica vaginalis is a mesothelium-lined sac that results from closure of the superior portion of the

processus vaginalis of the peritoneum. It has a visceral layer and an outer parietal layer that line the internal spermatic fascia of the scrotal wall, with a potential cavity between the two layers⁸. Primary tumours of the tunica vaginalis testis, whether benign or malignant, are extremely rare. Adenomatoid tumour, mesothelioma or benign intraepithelial cyst may originate from mesothelial cells of the tunica vaginalis. Benign or malignant mesenchymal tumours, lymphomas or serous borderline tumours are the other rare primary tumours of the tunica vaginalis⁹.

Malignant mesothelioma is the most common primary tumour of the paratesticular region with an epithelial growth pattern¹⁰. Tunica vaginalis is the most common site of origin, but the tumour may also arise from the spermatic cord and the epididymis^{11,12}.

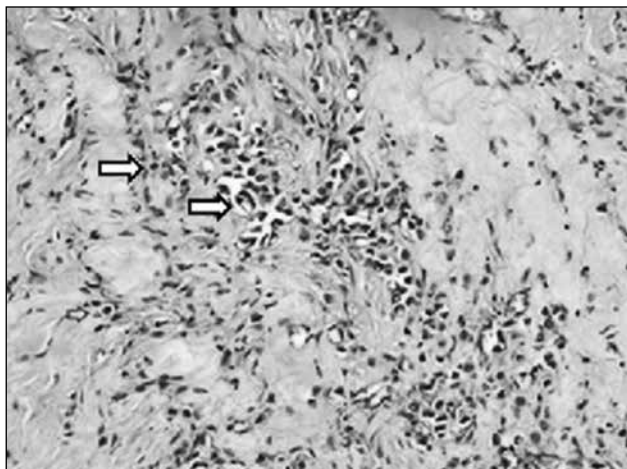


Figure 3. Neoplastic cells (arrows) showed cell groups and abortive glandular within tunica vaginalis (H&Ex200).

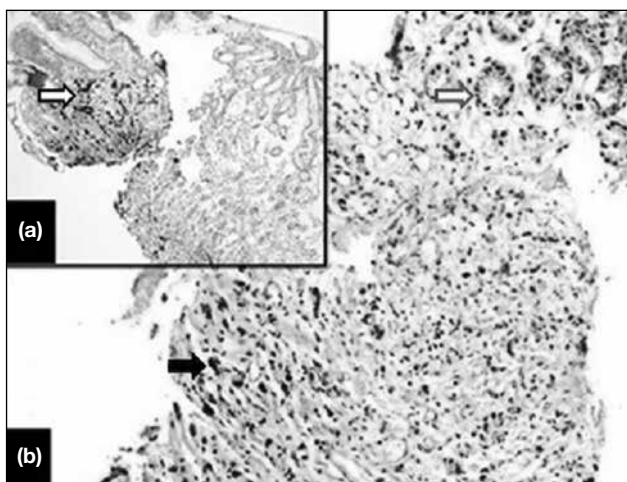


Figure 5. a, b. Tumour cells within gastric mucosa are positive with α -feto protein (a, arrow) (AFPx100). Neoplastic cells (b, black arrow) showed cell groups and abortive glandular within gastric mucosa (b, white arrow) (H&Ex200).

Metastases to the tunica vaginalis are also rare and generally encountered as an incidental autopsy finding¹³. Secondary paratesticular tumours are usually diagnosed at advanced stages of the primary malignancy. However, they may be presented as an initial manifestation of an occult primary neoplasm, as shown in our case. Similarly, a few cases have been reported in the literature^{3,14–16}.

The most common primary sites of metastasis to the paratesticular tissue include the prostate gland, kidney, stomach, colon, ileum, pancreas, lung, and breast¹⁷. The colon is the most common primary site in the gastrointestinal tract¹³. Hematogeneous or lymphatic spread

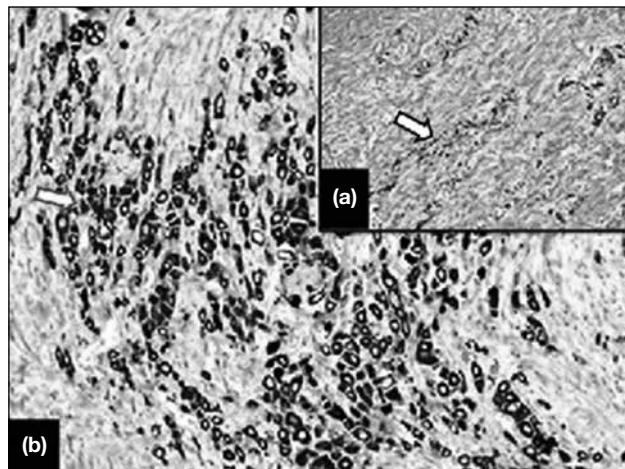


Figure 4. a, b. Tumour cells within tunica vaginalis are focally positive with AFP (a, arrow) (AFPx200). Tumour cells within tunica vaginalis are positive with pan-cytokeratin (b, arrow) (pan-CKx200).

are the main routes of metastasis to the paratesticular region; the other ways of spread include direct extension from adjacent organs (particularly prostatic carcinoma), retrograde extension through vas deferens, either along its lumen or as a direct extension, retrograde venous or lymphatic extension, arterial embolism, and trans-peritoneal seeding through a patent tunica vaginalis¹⁸. In our case, the route of transperitoneal seeding was suspected, particularly in view of the peritoneal carcinomatosis.

It is well known that serum AFP, β -hCG, and LDH levels are elevated in primary testicular neoplasms. However, it is extremely rare for the metastases to the testis and paratesticular region. As reported in our case, hepatoid type of gastric adenocarcinoma is one of the few exceptions that could be associated with increased plasma AFP levels¹⁹.

A number of reports concerning AFP-producing hepatoid type of gastric cancer associated with multiple liver metastases have been published. However, to our knowledge, this is the first description of a hepatoid type of gastric cancer metastasis to the tunica vaginalis testis. In differential diagnosis, AFP-producing germ cell tumours of testis are needed to be distinguished from metastatic cancers of this region²⁰. Imaging features are non-specific, and attention must be paid because similar findings may be seen both in primary tunica vaginalis tumours and in other metastatic tumours of this region. Nevertheless, multiplanar imaging capability of MRI allows precise

demonstration and localization of a mass, including its anatomic relationship to the surrounding structures. Gadolinium-based contrast material may increase the conspicuity of a tumour by showing differential enhancement.

Metastatic tumours to this region is poor, because they are usually diagnosed in the setting of disseminated disease²¹. In our case the patient died within eight months after the diagnosis.

In conclusion; the possibility of a hepatoid type gastric adenocarcinoma metastasis may be considered in the differential diagnosis list when a patient is admitted with acute scrotal swelling and high level of serum AFP. As shown in our case; the triad of elevated levels of AFP, testis swelling without testicular tumour should alert the physician about the risk of the rare hepatoid gastric adenocarcinoma metastasis.

References

1. Plas E, Ried CR, Pflüger H. Malignant mesothelioma of the tunica vaginalis testis: review of the literature and assessment of prognostic parameters. *Cancer* 1998;83:2437–46.
2. Lee J, Kang SC, Ban JH, Shin DS, Yeo JK, Yoon DH, et al. Metastatic Tumour of Tunica Vaginalis Testis with Hydrocele in a Patient with Gastric Cancer. *Korean J Urol* 2007;48:667–9.
3. Yeo JK. Scrotal Hydrocele as the First Clinical Manifestation of Occult Gastric Cancer. *Korean. J Urol* 2009;50:1151–3.
4. Kageyama Y, Kawakami S, Li G Kihara K, Oshima H, Teramoto K. Metastatic tumour of spermatic cord and tunica vaginalis testis from gastric cancer: a case report. *Hinyokika Kyo* 1997;43:429–431.
5. Yasuhide T, Heizo Y, Katsusuke N. Metastatic Tumour of the Tunica Vaginalis Testis Arising from Rectal Cancer: A Case Report. *Nishinohon J Urol* 1999;61:245–7.
6. Ruiz JM, Sanchis CM, López PC, Navarro HP, Moreno MJ, Sánchez AS, et al. Metastasis to the tunica vaginalis testis from a primary mucinous tumour of the cecum. *Arch Esp Urol* 2010;63:235–8.
7. Zheng J, Lu G, Xu C, Song B, Zhou Z. Metastatic Colon Adenocarcinoma to the Tunica Vaginalis Testis Presenting As Hydrocele. *BJUI* 2012.
8. Garriga V, Serrano A, Marin A, Medrano S, Roson N, Pruna X. US of the tunica vaginalis testis: anatomic relationships and pathologic conditions. *Radiograph* 2009;29:2017–32.
9. Woodward PJ, Schwab CM, Sesterhenn IA. Extratesticular scrotal masses: radiologic-pathologic correlation. *Radiograph* 2003;23:215–40.
10. Srigley JR, Hartwick RW. Tumours and cysts of the paratesticular region. *Pathol Ann* 1990;25:51–108.
11. Carp NZ, Petersen RO, Kusiak JF, Greenberg ML. Malignant mesothelioma of the tunica vaginalis testis. *J Urol* 1990;144:1475–8.
12. Umekawa T, Kurita T. Treatment of mesothelioma of the tunica vaginalis testis. *Urol Int* 1995;55:215–7.
13. Meacham RB, Mata JA, Espada R, Wheeler TM, Schum CW, Scardino PT. Testicular metastasis as the first manifestation of colon carcinoma. *J Urol* 1988;116:621–2.
14. Irisawa C, Yamaguchi O, Shiraiwa Y, Kikuchi Y, Irisawa S, Irisawa C. A case of metastatic tumour of the spermatic cord from gastric carcinoma. *Hinyokika Kyo* 1989;35:1807–9.
15. Polychronidis A, Tsolos C, Sivridis E, Botaitis S, Simopoulos C. Spermatic cord metastasis as an initial manifestation of sigmoid colon carcinoma: report of a case. *Surg Today* 2002;32(4):376–7.
16. Lioe TF, Biggart JD. Tumours of the spermatic cord and paratesticular tissue. A clinicopathological study. *Br J Urol* 1993;71:600–6.
17. Algaba F, Santaularia JM, Villavicencio H. Metastatic tumour of the epididymis and spermatic cord. *Eur Urol* 1983;9:56–9.
18. Lee KY, Park SJ, Moon SK, Kim HC. Unilateral spermatic cord metastasis from gastric cancer: a case report. *Iran J Radiol* 2012;9(4):217–9.
19. Ota T, Shinohara M, Tanaka M, Itakura H, Munakata A, Kinoshita K, et al. Spermatic Cord Metastases from Gastric Cancer with Elevation of Serum hCG-β: A Case Report. *Japanese J Clin Oncol* 2000;30(5):239–40.
20. Vlachostergios PJ, Voutsadakis IA, Barbanis S, Karasavvidou F, Papandreou CN. AFP-producing hepatoid adenocarcinoma of the stomach: a casereport. *Cases J* 2009;2:9296.
21. Algaba F, Santaularia JM, Villavicencio H. Metastatic tumour of the epididymis and spermatic cord. *Eur Urol* 1983;9(1):56–9.