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

Yavuz Metin1, Nurgül Orhan Metin1, Oğuzhan Özdemir1, Hakki Uzun2, Recep Bedir3

1Recep Tayyip Erdoğan University Faculty of Medicine, Training and Research Hospital, Department of Radiology; 2Department of Urology; 3Department of Pathology, Rize, Turkey

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 izleneceği birlikte nadiren de olsa primer testiküler tümörü taklit edebilir. Bu makalede, alfa feto-protein üreten hepatoid tip mide kanserinin, skrotal ağrı ve şişliğin eşlik ettiği tunika vaginalis metastazı şeklinde prezante olması sunulmaktadır.

Anahtar kelimeler: alfa feto-protein; hepatoid tip mide kanseri; tunika vaginalis

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 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.
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 layers\(^8\). 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 vaginalis\(^9\).

Malignant mesothelioma is the most common primary tumour of the paratesticular region with an epithelial growth pattern\(^10\). Tunica vaginalis is the most common site of origin, but the tumour may also arise from the spermatic cord and the epididymis\(^11,12\).
Metastases to the tunica vaginalis are also rare and generally encountered as an incidental autopsy finding. 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. The most common primary sites of metastasis to the paratesticular tissue include the prostate gland, kidney, stomach, colon, ileum, pancreas, lung, and breast. The colon is the most common primary site in the gastrointestinal tract. Hematogeneous or lymphatic spread 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 vaginalis. 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 levels. 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 region. 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

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

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).

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).
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 disease. 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