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CASE REPORT

Case Report

Soft Tissue Metastasis from Immature Teratoma of the Testis

Second Case Report and Review of the Literature

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Abstract

Background Testicular cancer, like other histopathologic types, commonly metastasizes to the lungs, liver, and brain. Spread to soft tissue, however, is rare with only four cases with seminoma reported. However, one case with metastasis of testicular immature teratoma to soft tissue was documented previously.

Case Description We report the case of a 38-year-old man with recurrent immature teratoma of the testis who presented with a painless soft tissue mass in the left thigh previously treated with standard chemotherapy. After removal of the soft tissue mass, his serum alpha-fetoprotein level had returned to the normal range.

Literature Review To our knowledge, this is the second case of immature teratoma of the testis metastasized to soft tissue.

Purposes and Clinical Relevance We suggest that for a man with testicular cancer who has a soft tissue mass, metastasis of soft tissue from testicular cancer and other solid malignancies should be considered in the differential diagnosis of a soft tissue mass together with primary soft tissue sarcoma.

Introduction

Metastases to soft tissue are uncommon and easily can be confused with primary soft tissue sarcoma clinically and histopathologically [4, 14]. The differential diagnosis between a metastatic neoplasm and a primary soft tissue sarcoma is critical, because their treatment is markedly different. Although soft tissue metastasis can represent the initial manifestation of a primary malignancy, it usually is seen as late evidence of recurrence in advanced cancer [4, 13, 17]. Its incidence is rare, as low as 0.16% and 0.8% [6, 7].

Most primary testicular tumors originate from germ cells. Germ cell tumors are classified for clinical purposes into two major groups: seminomas or nonseminomas. Pure seminoma is the most common testicular tumor and generally has a more favorable prognosis than nonseminomas. Nonseminomas include embryonal carcinoma, teratoma, choriocarcinoma, and yolk sac tumor. Mature teratomas contain well-differentiated tissue and may include ectodermal, endodermal, or mesodermal elements. Immature teratomas are characterized by primitive, poorly differentiated tissue [1]. The most common sites of metastasis from testicular cancer are the lungs, liver, and brain [1, 5]. Choroidal [15], eye [11], gingiva, skin [2], and kidney also

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have been reported as uncommon sites for metastasis. Although metastasis of soft tissue from the various cancer types, including the lung, breast, kidney, and colon, have been documented comprehensively [4, 6, 7, 13, 17], soft tissue metastasis of testicular cancer rarely has been reported because there are only four patients with seminoma and one with immature teratoma [8, 10, 14].

We describe the second case of an immature teratoma of the testis that metastasized to soft tissue as the manifestation of recurrence. We also present a review of the literature.

Case Report

A 38-year-old man presented with two months history of painless enlargement of the left testis. He had noted gradual enlargement of his testicular mass during a period of several months. Physical examination was normal except for a 7 × 5-cm nontender mass in the left testis. Serum alpha-fetoprotein (AFP) was 42 ng/mL (normal, 0–7 ng/mL), beta-human chorionic gonadotropin (ß-HCG) was normal, and serum lactate dehydrogenase (LDH) also was in the normal range. Ultrasonography of the scrotum revealed a nonhomogeneous 6.8 × 5.2-cm mass in the left testis with varying echogenicity. We subsequently performed a left radical orchiectomy. Histopathologic examination of the testicular mass showed a pure immature teratoma measuring 9 × 7 × 5 cm and no invasion of adjacent structures. Two weeks after surgery, the serum AFP level returned to the normal range postoperatively. Subsequent CT scans of the thorax and abdominopelvis were normal. Because of these findings, the diagnosis of Stage IA immature teratoma of the testis was made. He had no subsequent chemotherapy or radiation therapy.

The patient's cancer remained in remission for 3 years. He then was referred to our clinic with a 1-month history of headache, cough, and chest pain. Physical examination revealed normal system findings except for mildly decreased breathing sounds in the right lower lung site. Initial laboratory results were as follows: AFP 834.9 ng/mL, ß-HCG 0.436 IU/mL (normal, < 7 IU/mL), LDH 521 U/L (normal, 240–480 U/L), erythrocyte sedimentation rate 119 mm/hour, C-reactive protein 15 mg/L, leukocytes 10,500/mm^3, platelets 480,000/mm^3, hematocrit 32.2%, and MCV 91.4 fL. Other laboratory values, including blood urea nitrogen, creatinine, sodium, potassium, calcium, ALT, AST, alkaline phosphatase, glucose, total protein, albumin, total bilirubin, and bilirubin, were within normal limits. A thorax CT revealed a 6 × 9-cm mass in the lower lobe of the right lung, multiple millimeter-sized metastatic nodules in the anterior and posterior segments of the upper lobe of the right lung, and three 1.5-cm mediastinal lymph nodes. Abdomen and pelvic CT revealed multiple metastatic lesions that measured 2.5 cm in maximal diameter in the liver. In addition, MRI of the brain showed a 6 × 4 × 4-cm metastatic mass in the left occipital lobe with surrounding edema. Because of cranial metastasis, the patient first was treated with whole-brain radiation therapy. Thereafter, chemotherapy with bleomycin, etoposide, and cisplatin was started. After completion of two cycles of chemotherapy, his symptoms improved and the serum AFP level returned to the normal range. Therefore, chemotherapy was continued with the same protocol. There was no evidence of clinical relapse after completion of four courses of chemotherapy.

Four years after the initial diagnosis, he presented with a 2-month history of a painless soft tissue mass in the left thigh 8 months after presenting with metastatic disease. He stated he otherwise felt well. On physical examination, a 4 × 4-cm diameter soft tissue mass in the left thigh was detected. The serum AFP level was 20.8 ng/mL. Other laboratory values were within normal ranges. MRI of the thigh revealed a 4 × 3 × 2.5-cm diameter soft tissue mass that was hyperintense on T2-weighted and hypointense on T1-weighted images in the medial side of the left thigh (Fig. 1). PET/CT scans showed a soft tissue mass in the left thigh with increased FDG activity in this site for malignancy. No increased FDG activity was detected in the other sites.

Fine needle aspiration cytology of the left thigh mass was inconclusive. Therefore, a Tru-cut biopsy was performed. Histopathologic examination of the biopsy specimen revealed the component of immature cartilage in fibrous stroma and cytokeratin positivity with a glandular

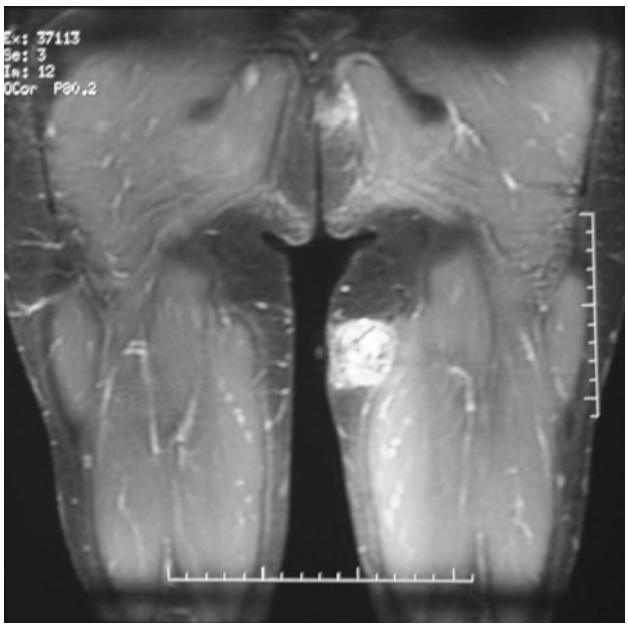

Fig. 1 MRI of the thigh shows a 4 × 3 × 2.5-cm diameter soft tissue mass, which was hyperintense on T2-weighted and hypointense on T1-weighted images in the medial side of the left thigh.

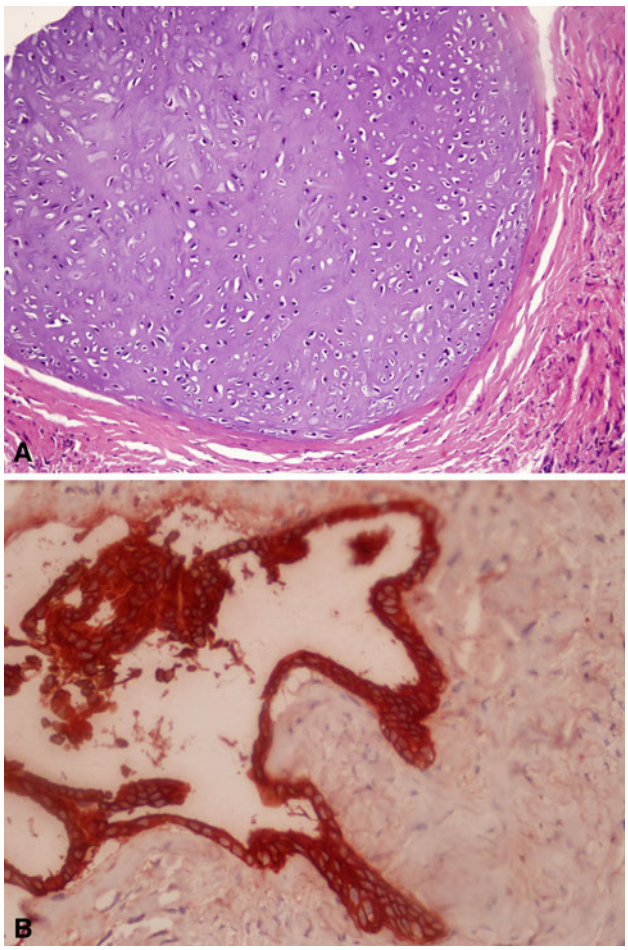

Fig. 2A–B (**A**) A Tru-cut biopsy of the soft tissue lesion shows the component of immature cartilage in fibrous stroma (Stain, hematoxylin and eosin; original magnification, ×20). (**B**) There is high nuclear cytokeratin positivity with a glandular component in the tumor cells (Stain, immunohistochemistry cytokeratin; original magnification, ×40).

component (Fig. 2). The diagnosis of soft tissue metastasis of an immature teratoma of the testis was made. Therefore, a wide excision of the metastatic thigh lesion was performed. We observed no gross residual tumor and the margins were negative for tumor after excision. The serum AFP level declined postoperatively. Surveillance was initiated with history and physical examination, serum tumor markers (AFP and ß-HCG), and CT of the abdomen, pelvis, and thorax every 3 months for the first 3 years. At last followup at 10 months after excision of the soft tissue mass, he had no specific symptoms and his disease has remained in remission; the patient's serum AFP level was within normal limits 10 months postoperatively.

Discussion

Compared with other organs of the body, including the lungs, liver, and brain, soft tissue is a rare site for metastasis in testicular cancer [5, 8, 14, 15]. For all solid tumors, incidence of soft tissue metastasis is low [6, 7]. Metastases to soft tissue usually are detected in patients with advanced disease; they rarely occur in a primary unknown tumor. Furthermore, it can be the source of diagnostic confusion with a soft tissue sarcoma [7]. In the majority of patients, soft tissue metastasis may be the first sign of the disease; however, it usually is seen as a manifestation of relapse in advanced cancer. Multiple case reports and small series of patients with associated soft tissue metastases have been documented [4, 6, 16, 17].

The most commonly reported primary malignancies to result in soft tissue metastasis are lung, kidney, and colon carcinoma [3, 12, 17], but only five cases of soft tissue metastases from testicular cancer have been reported. Four of the patients had seminoma histology and only one patient had immature teratoma [8, 10, 14]. In the first case and in our case, the thigh was the site of soft tissue metastasis from the seminoma. Plaza et al. reported the abdominal wall, back, thigh, chest, and shoulder were commonly affected sites in the majority of patients with soft tissue metastasis in solid malignancies [14] (Table 1).

It is unknown whether metastasis was present on initial presentation in Patients 1 and 5. However, in the other three patients with seminoma and our patient, soft tissue metastasis was a feature of recurrence of disease. Previous reports suggest soft tissue metastasis is relatively common after relapse [4, 6, 17], as was the case with our patient. In contrast, soft tissue metastasis as the initial manifestation of the disease has been detected in as much as 21.1% of cases other solid tumors [14]. Soft tissue metastases frequently present with a painful mass, which is distinct from primary sarcomas in which the mass often is painless [4, 9]. However, our patient had a painless mass, whereas Patient 2 presented with vague discomfort attributable to a lump in the right upper arm.

In soft tissue metastasis, treatment options include observation, radiotherapy, chemotherapy, and excision. Excision may be indicated for selected patients with isolated soft tissue metastases, especially after a long disease-free interval [4]. Our patient was treated with wide excision because of isolated soft tissue metastasis. Chemotherapy or radiotherapy was not instituted because he did not have visceral metastasis and residual tumor. Patient 2 (Table 1) also was treated with subtotal excision and radiotherapy. The treatment for Patient 1 was not described. The other patients (Patients 3-5) were treated with chemotherapy.

This is the second reported case of a testicular immature teratoma metastasizing to soft tissue. In patients with testicular immature teratoma who presented with a painful or painless soft tissue mass, soft tissue metastasis of testicular cancer should be considered in the differential diagnosis of a soft tissue mass as a primary soft tissue sarcoma. Our

Table 1. Features of patients with soft tissue metastasis of testicular cancer

Patient number	Study	Age of patients (years)	Gender	Histology	Soft tissue site of metastasis	Tumor size	Metastasis as the initial presentation	Time between primary tumor diagnosis and metastasis	Treatment
1	Plaza et al. [14]	54	Male	Seminoma	Thigh	NA	NA	NA	NA
2	Hans et al. [8]	43	Male	Seminoma	Right upper arm	7 × 7 cm	Yes	6 years	Surgery, RT
3	Husband et al. [10]	NA	NA	Seminoma	Left psoas muscle	NA	Yes	NA	CT
4	Husband et al. [10]	NA	NA	Seminoma	Left psoas muscle	NA	No	NA	CT
5	Husband et al. [10]	NA	NA	Immature teratoma	Left iliac and middle gluteal muscles	NA	Yes	NA	CT
6	Current study	38	Male	Immature teratoma	Thigh	4 × 3 × 2.5 cm	Yes	4 years	Surgery

CT = chemotherapy; RT = radiotherapy; NA = not available.

patient highlights the importance of distinguishing soft tissue metastasis from primary soft tissue sarcoma. The treatment strategy in metastasis of testicular cancers is markedly different because testicular tumors are more chemosensitive compared with primary soft tissue sarcomas.

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