Isolated inguinal lymph node metastasis in early stage testicular seminoma: a case report and review of the literature

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Summary. Inguinal lymph node metastasis in patients with testicular seminoma is exceptionally infrequent. We here present a case of early stage (stage 1A) testicular seminoma developing inguinal lymph node metastasis without involvement of the epididymis or tunica vaginalis according to a biopsy specimen. A 31 year-old male patient presented with a left testicular mass. Post-orchiectomy biopsy materials revealed a classical type of seminoma with clear surgical margins. The primary tumor was limited to the testicle without involvement of the epididymis or tunica vaginalis (pT1). Following surgery, clinical staging was compatible with stage 1A seminoma and close surveillance without any further treatment was planned by way of management. However, the patient came back with inguinal lymph node metastasis 5 months after surgery. We report this case as an important and conflicting clinical issue that requires to be elucidated since lymph node metastasis in an early stage of testicular seminoma is unexpected.

Key words: early stage seminoma, lymph node metastasis, testicular cancer

Introduction

Testicular cancer is a rare malignancy accounting for 1% of all cancer cases in men (1). Seminoma is the most common germ cell tumor of the testis comprising 40-45 % of all germ cell tumors (2). Stage 1 seminoma is the most frequently reported seminomatous testicular cancer constituting 80 % of all seminomas and 40 % of all testicular cancers (3). Most patients with stage 1 seminoma are treated by orchiectomy with curative intent. Current treatment methods following surgery for early-stage seminoma include surveillance, radiotherapy or carboplatin-based chemotherapy depending on the risk status of the patient (4). One of the most important advantages of surveillance in these patients who are not

expected to relapse is that it avoids unnecessary treatments such as chemotherapy or radiotherapy which may also increase the risk of developing a second cancer and other complications. As recurrences in patients with early stage seminoma mostly occur in retroperitoneal lymph nodes, inguinal lymph node involvement as the initial recurrence site is extremely unusual without a history of inguinal surgical intervention. Clinical management of these patients is unclear (1).

Case Report

A 31 year-old male patient underwent partial orchiectomy after presenting with the complaint of a

painless mass in the left testis. Post operative pathological examination of the biopsy materials revealed a classical type of seminoma with clear surgical margins without lymphovascular invasion. The primary tumor was 2 cm diameter and limited to the testicle without involvement of the epididymis or tunica vaginalis (pT1) (Figure 1). Tumor markers and lactate dehydrogenase were within the normal range and imaging workup including abdominal computed tomography showed no distant metastasis. The patient was diagnosed as stage 1A seminoma according to AJCC -2010 guidelines (American Joint Committee on Cancer) and a close follow-up without any treatment was planned according to the National Comprehensive Cancer Network guideline version 1.2015. However, the patient re-presented with new swelling and pain in the left inguinal area. Superficial ultrasound showed two hypoechoic-heterogeneous, lobulated contoured, solid masses sized 26x20 mm and 23x17 mm in the superior and inferior part of the left inguinal canal. Positron emission tomography identified increased pathological FDG uptake (SUV Max: 12.5 and 9.8) in the 2 masses defined in ultrasonography, indicating a possible recurrence of seminoma. Pathological findings from the excisional biopsy performed from the inguinal masses were consistent with metastasis of the seminoma (Figure 2). The patient was discussed at a multidisciplinary team meeting including a urologist, radiation oncologist, pathologist, radiologist and medical oncologist, and systemic chemotherapy treatment was planned since the inguinal area was accepted as the site of systemic involvement rather than a local spread. However, the patient died of neutropenic fever following the first cycle treatment of Bleomycin, Etoposide and Cisplatin (BEP) chemotherapy.

Discussion

Testicular lymphatics usually drain into the retroperitoneal lymph nodes through the gonadal veins between the thoracic and lumbar vertebrae. Iliac and inguinal lymph node involvement in testicular cancer is usually developed by retrograde lymphatic spread as a result of bulky retroperitoneal masses. In general, lymphatics of the hips and lower abdomen skin including the scrotum drain into the superficial inguinal

lymph nodes, while the legs and deep penile structures drain into the deep inguinal lymph nodes. However, in cases of testicular lymphatic damage or spermatic cord injury following surgery or scrotal incision, lymphatics are capable of developing new collaterals, leading to direct inguinal invasion (5). Hence, primary inguinal and iliac lymphatic involvement is infrequent and generally associated with some predisposing factors such as epididymal tumoral invasion, tunica vaginalis or vas deferens involvement (6). Although the incidence of inguinal lymph node metastasis in patients who have a history of surgery for testicular tumor or orchidopexy is uncertain, it has been reported to be 2% to 10% (7, 8) in various series. When considering the frequency according to the histological subtype, inguinal metastasis was found to be significantly higher in non-seminomatous testicular cancer than in seminoma (4.9% vs 0.5%, respectively) (7). Metastasis to the inguinal lymph nodes in testicular cancer may occur due to the following mechanisms: scrotum or tunica vaginalis involvement of the primary tumor, prior inguinal or scrotal surgery, retrograde lymphatic spread due to massive lymph nodes (1). Besides, spermatic cord (funiculus spermaticus) involvement due to tumoral infiltration is a common cause in patients who have not undergone any previous operation. Thus, in order to reduce the risk of recurrence, the pathologist should also perform a further examination of the spermatic cord. By contrast, the fact that our case was pure stage 1A seminoma and did not carry any risk factors including lymphovascular invasion, tunica vaginalis and spermatic cord invasion that might increase the risk of recurrence or metastasis, made this a highly unique phenomenon in our patient. One another reason for developing a risk of recurrence in seminoma may be associated with spillage of tumor cells during surgery for testicular cancer. An inguinal approach rather than scrotal incision should be performed to prevent tumoral contamination of the scrotal skin and also to avoid exposure of tumor cells to the inguinal lymph nodes. Inguinal incision is therefore performed to reduce the risk of tumor spillage and recurrence (7). On the other hand, there are limited data regarding the beneficial effect of inguinal lymphadenectomy whether the inguinal lymphatics are palpable or not in testicular tumors (8). Although prophylactic lymphadenectomy is rarely

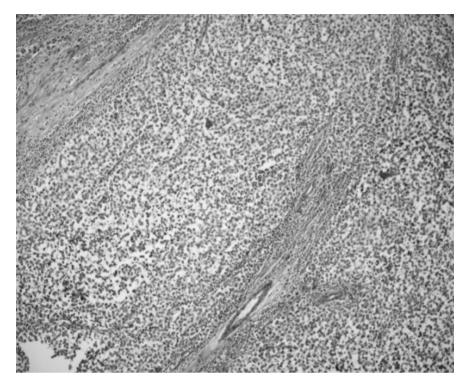


Figure 1. Image of seminoma in testis; Neoplastic cells showing nucleoli specificity with an eosinophilic cytoplasm divided by thin fibrous septas and infiltrated by lymphocytes. 40x10 magnification in hematoxylin eosin stained sections.

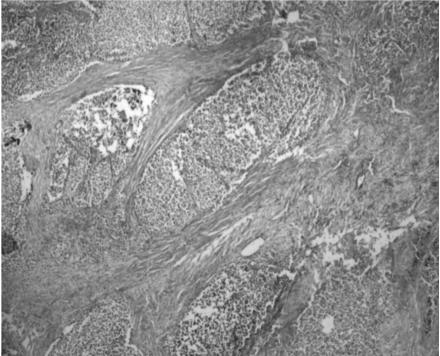


Figure 2. Image of tumor metastasis in inguinal lymph node; Neoplastic cells showing nucleoli specificity with an eosinophilic cytoplasm infiltrating the lymph node with a diffuse pattern. 10x10 magnification in hematoxylin eosin stained sections. The structure of the lymph node can not easily be selected due to excessive tumoral infiltration.

mentioned in the literature, this approach has been reported to have no significant effect on recurrence (6).

All in all, radical inguinal lymphadenectomy is not recommended in testicular cancer because of the rarity of inguinal lymph node metastasis. An initial workup of excisional or fine needle biopsy should be carried out in the presence of inguinal lymph node metastasis.

The optimal treatment for testicular cancer presenting with inguinal lymph node metastasis has remained a therapeutic challenge. Stein et al. recommend the approach of local excision with or without chemotherapy in this group of patients (9). van Ahlen et al. suggest adjuvant chemotherapy treatment following local tumor excision (10). However, inguinal nodal involvement in our case was considered as a distant metastasis site at a multidisciplinary team meeting, and hence the indication was for systemic chemotherapy treatment. On the other hand, radiotherapy may be considered as an appropriate option in such cases, though there is not enough information in the literature regarding which management is most appropriate for patients with inguinal nodal metastasis.

In conclusion, the issue of inguinal nodal metastases on the basis of no risk of recurrence is uncertain and remains to be elucidated in testicular tumors, particularly seminoma. In such cases, in order to reduce the potential risk of contamination and to minimize the possibility of inguinal metastases, patients should be considered for new surgical techniques and also given close surveillance following surgery.

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