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Case of testicular nonseminomatous germ cell tumor cancer soft tissue seeding after inguinal orchiectomy

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ABSTRACT

A 29-year-old patient presented to his primary care provider complaining of a painful right inguinal swelling. He was referred for inguinal hernia repair, but during surgery, an enlarged necrotic-appearing testicle was observed and removed. Pathology demonstrated a mixed non-seminomatous germ cell tumor (NSGCT) with evidence of tumor violation. After receiving BEPx3 for elevated post-operative AFP his tumor markers normalized. On surveillance, he was found to have several palpable masses around his inguinal incision. On soft tissue excision he was found to have residual teratoma within his soft tissues. We review the literature on germ cell tumor seeding and atypical recurrences.

1. Introduction

Testicular cancer is the most common cancer among males 15–40 years old. The pattern of spread of testicular cancer is predictable with retroperitoneal lymph node involvement being one of the primary landing sites. Inguinal orchiectomy is an essential component of testicular cancer diagnosis and management. Recent reports of out-of-field atypical recurrences after minimally invasive retroperitoneal surgery for primary and post-chemotherapy non-nonseminomatous germ cell tumor (NSGCT) have raised concerns for possible seeding of tumor due to pneumoperitoneum. In this report we review a case of testicular cancer seeding of subcutaneous soft tissues due to likely tumor violation during an open inguinal hernia repair. This and other reports suggest that GCTs have the ability to seed if oncologic principles are violated, regardless of surgical approach.

2. Case presentation

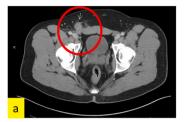
A 29-year-old male with a history of an undescended right testis s/p orchidopexy at age 2 presented with sharp pain and swelling in his right testicle. He was diagnosed with a right inguinal hernia and descended testis bilaterally on physical exam after evaluation with a general

surgeon and was scheduled for an open right inguinal hernia repair. No pre-operative imaging was obtained. Per report, intra-operatively there was no evidence of an inguinal hernia. A possible testicle was noted in the inguinal canal which appeared necrotic from presumed torsion and the decision was made to perform an orchiectomy. Pathology demonstrated a 5 cm mixed germ cell tumor (65% embryonal carcinoma, 15% yolk sac tumor, 15% teratoma, and 5% choriocarcinoma) with lymphovascular and tunica vaginalis invasion. However, pathology report demonstrated unclear surgical margins and no epididymis or spermatic cord noted in the specimen, raising concern for potential tumor violation during the procedure.

Pre-operative serum tumor markers (STMs) were not obtained. 3 week post-operative serum tumor markers were significant for Beta-HCG: 2 IU/L, AFP: 42 ng/mL (H), and LDH 165 U/L. Computed tomography (CT) of the chest, abdomen, and pelvis 3 weeks after surgery was negative for retroperitoneal lymphadenopathy, but notable for soft tissue nodularity in the right inguinal region concerning for residual malignancy (Fig. 1a). The patient was subsequently started on bleomycin, etoposide, and cisplatin (BEP). His AFP increased to 191 after his first cycle of BEP, and he subsequently went on to complete 3 total cycles of BEP. After his third cycle of BEP, his serum tumor markers were within normal limits and a CT abdomen/pelvis demonstrated a 1.2×1.4

Abbreviations: NSGCT, Non-seminomatous germ cell tumor; BEP, Bleomycin, etoposide, cisplatin; STM, Serum tumor marker.

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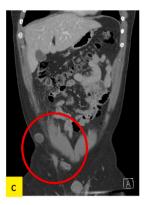


Fig. 1. (a) CT abdomen/pelvis 4 months after initial orchiectomy and pre-chemotherapy; post BEPx3 CT abdomen pelvis axial (b) and sagittal (c) images. Growing subcutaneous nodules are highlighted in red. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

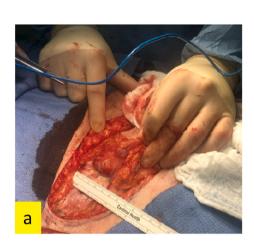
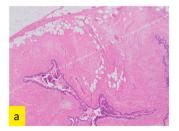


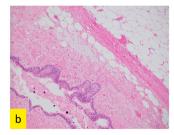


Fig. 2. Intra-operative photos demonstrating (a) visible teratoma within the specimen and (b) final internal ring, external oblique fascia and soft tissue resection defect.

cm soft tissue density in the right inguinal canal. This mass was resected by his surgeon under ultrasound-needle guidance and pathology demonstrated residual epididymis and vas deferens with no residual malignancy. He started surveillance 5 months after initial surgery with serial imaging and serum tumor markers. About 14 months after his original orchiectomy, he noted a palpable soft tissue mass in the right groin which was associated with pain. CT demonstrated several nodules within the soft tissues of the groin and peripherally enhancing fluid density with the subcutaneous fat of the right lower pelvis near the inguinal canal (Fig. 1b and c). Serum tumor markers remained within normal limits.

At this point, he was referred to our institution for further workup and care for presumed local recurrence of his tumor. His exam was notable for palpable, mobile nodules in the right inguinal region close to his prior surgical scar. Ultimately, the decision was made to proceed with wide local excision of his right inguinal scar and soft tissue masses. A 20×8 cm skin excision down to the fascia and internal inguinal ring was performed. The residual proximal cord, prior surgical scars and small portion of abnormal appearing fascia were resected with the soft tissue specimen (Fig. 2a and b). Due to the size of the defect, the deep epigastric vessels were resected with the specimen and, therefore, plastic surgery performed a right pedicled anterolateral thigh flap. The





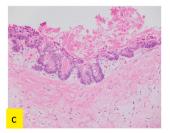


Fig. 3. (a) Teratoma involving fibroadipose tissue. H&E 40x. (b) Teratoma involving soft tissue with glandular and focal cartilaginous differentiation H&E 100x. (c) Teratoma with glandular differentiation, recapitulating gastrointestinal-type glands with goblet cells. H&E 200x.

patient tolerated the procedure well and was discharged home on postoperative day 3. Pathology demonstrated multiple soft tissue nodules containing teratoma (up to 3.1 cm) with some teratoma within the soft tissue (no lymphatic tissue identified) involving the excised fascia around the internal inguinal ring (Fig. 3a–c). All resection margins were negative. At 15 months after surgery, imaging and serum tumor markers have remained normal.

3. Discussion

Testicular cancer has a predictable pattern of lymphatic spread. Adherence to guideline-directed multidisciplinary care in testicular cancer has led to significant improvements in survival. Recent studies evaluating the role of minimally invasive (robotic/laparoscopic) approaches for the management of retroperitoneal lymph node metastases has raised concerns for the development of atypical recurrences. In a recent report, Calaway, et al. described 4 cases of atypical out-of-field recurrences (e.g., perinephric, pericolic) within the abdomen after robotic retroperitoneal lymph node dissection. Notably, the median time to recurrence was 9 months. Furthermore, recent published randomized trials in the treatment of localized cervical cancer demonstrated lower rates of disease-free survival and overall survival in minimally invasive surgery vs open abdominal radical hystorectomy. 3,4

Our case report and others⁵ demonstrates the ability of testicular GCT to seed soft tissues when the tumor is violated. This case and other recently reported cases demonstrate two points to highlight, 1) GCT can seed tissues and 2) violation of oncologic principles is likely more important than surgical approach (open vs minimally invasive). The appropriate surgical approach should be chosen based on experience and every attempt made to avoid tumor violation. Guiding principles should include proper handling of lymphatic tissues, avoid division of lymph nodes, and placement of tumors/lymph nodes within extraction

bags rather than removing through extraction ports.²

4. Conclusion

Tumor violation during orchiectomy could lead to soft tissue seeding of NSGCT. This case report and small published case series suggest atypical recurrences of GCTs can occur when an open or minimally invasive technique is used and oncologic principles are violated.

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Author contributions

JV wrote the case report and reviewed literature; JM provided histology images and interpretations; JA, CD, and AS edited case report and provided guidance.

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