Metastatic Tumor of the Spermatic Cord in Adults: A Case Report and Review

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Metastatic spermatic cord (SC) tumor is extremely rare. Recently, we experienced a case of late-onset metastatic SC tumor from cecal cancer. This case is a 68-year-old man presenting with a painless right SC mass. He had undergone a right hemicolectomy for cecal cancer 6 years ago. Radical orchiectomy and adjuvant chemotherapy with S-1 were performed. No recurrence was found after one year of follow-up. We identified a total of 25 cases, including our case, on a literature search via PubMed from January 2000 to April 2015. The most frequent primary sites of the tumors metastasizing to the SC were the stomach (8 cases, 32%) and the colon (8 cases, 32%), next the liver (2 cases, 8%), and kidney (2 cases, 8%). The majority of the cases underwent radical orchiectomy for the metastatic tumors of the SC. Over half of the cases received adjuvant interventions based on the regimens for the primary tumors. Prognosis in the patients with metastatic tumor of the SC was unfavorable except for late-onset metastasis. In patients with a mass in the SC and a history of neoplasm, especially in gastrointestinal tract, the possibility of metastasis from the primary cancer should be considered.

1. Introduction

Tumors arising from the spermatic cord (SC) are rare and most of these tumors are benign such as lipoma. However, approximately 25% are potentially life-threatening malignant neoplasms [1]. The most common malignant tumors comprise sarcomas such as liposarcoma, leiomyosarcoma, rhabdomyosarcoma, and malignant fibrous histiocytoma and occur as a result of a mutation of a pluripotent mesenchymal cell that transforms into a malignant population clones [2].

On the other hand, metastatic SC tumor is even more unusual [3]. Several investigators have indicated that the most frequent primary tumors metastasizing to the SC and peritesticular tissues have been neoplasms of the stomach and prostate [4]. The timings of the detection of SC metastasis in most previous reports have been synchronous or metachronous, and the majority of cases in the metachronous were found in less than several years after the treatment for primary tumors [4]. Recently, we experienced a patient with cecal cancer recurrence in the SC that occurred on late phase after radical hemicolectomy. Herein, we report this case and a review of the recent literatures.

2. Case Report

A 68-year-old man was referred to the Department of Urology from the Surgery at Higashimatsuyama Municipal Hospital with a right painless inguinal mass in April 2014. He had noticed it 6 months previously and observed an increase of its size. He had undergone right radical hemicolectomy for cecal cancer 6 years ago. Histological examination of the extirpated colon specimen showed moderately differentiated adenocarcinoma (Figure I(a)) with depth of invasion of subserosa, lymphovascular invasion, and metastasis of the paracolic lymph nodes.

On physical examination, an approximately $4 \times 3$ cm palpable relatively fixed painless mass was observed in the right inguinal region. Abdominal computed tomography
Figure 1: Histopathology. (a) Primary cecal cancer reveals moderately differentiated adenocarcinoma. (b) Spermatic cord tumor shows moderately differentiated adenocarcinoma, which is compatible with a metastasis from the cecal cancer. (c) Immunohistochemical staining indicates caudal-type homeobox- (CDX-) 2 positive in the spermatic cord tumor. (d) Immunohistochemical staining shows cytokeratin- (CK-) 20 positive in the spermatic cord tumor.

Figure 2: Abdominal CT. Abdominal CT reveals a 3.7 cm diameter slightly enhanced tumor (arrow) in the right spermatic cord.

(CT) scan showed a heterogeneously slight-enhanced mass with noncapsulated irregular shape, 3.7 cm in diameter, suspicious of extending to the adjunct structures (Figure 2). Serum carcinoembryonic antigen (CEA) value was slightly elevated to 7.0 ng/mL (normal range < 5.0 ng/mL) but the levels of other tumor markers such as carbohydrate antigen 19-9 (CA19-9) and prostate-specific antigen (PSA) were normal. We did not deny that this tumor had been potentially malignant based on the clinical findings.

The patient underwent right radical orchiectomy. During the operation, the tumor was located in the SC and showed relatively invasive growth to the adjunct structures but did not invade the epididymis and testis. The resected specimen involved a 4.5 × 3.5 × 3 cm solid mass, with grayish-white tumor in the cut surface, and was located in the lower part of the SC (Figure 3).

Figure 3: Gross appearance of the resected tumor. Gross examination shows grayish-white mass (arrow) in the cut surface of the resected tumor.
A histological examination of the SC tumor showed moderately differentiated adenocarcinoma (Figure 1(b)). In an immunohistochemical examination, the tumor cells in the SC were stained for caudal-type homeobox- (CDX-) 2 (Figure 1(c)) and cytokeratin- (CK-) 20 (Figure 1(d)). Based on the histopathological and immunohistochemical findings, the SC tumor was compatible with a metastasis from the cecal cancer.

The postoperative course was uneventful. The patient received adjuvant chemotherapy with tegafur/gimeracil/oteracil (S-1) which is a combined oral chemotherapeutic agent and has been doing well without evidence of recurrence for one year following the surgery.

3. Discussion

The SC is an extremely rare site for distant metastasis from a malignant neoplasm. In an autopsy study there were only two metastatic sites (0.01%) of the SC among 13,500 autopsy cases, both of which were from a primary gastric cancer [3]. The most common primary origin of a SC metastasis was the stomach, followed by the prostate, ileum, kidney, and colon as previously reviewed by Algaba et al. [4]. That review is over thirty years old and to our knowledge, there have not been any reviews involving recent cases. We performed a literature search of the case reports on adult metastatic SC tumor via PubMed from January 2000 to April 2015. Table 1 shows identified cases [5–27] including our case since 2000. The mean age of incidence was 61 years (range: 36 to 85 years). The overall incidence was similar on both sites, but with respect to the colon cancers as the primary site the ascending colon and cecal cancers metastasized to the right SC while the descending colon and sigmoid cancers metastasized to the left SC. The most presenting symptoms were a mass in inguinal sites and scrotal swelling, both with and without pain except for one case that was incidentally found in an orchietomy specimen due to the treatment of prostate cancer. The average metastatic tumor size of the SC in the identified 15 cases was 3.6 cm in a diameter (range: 1.6 to 6.5 cm). The most frequent primary origin of the tumor was the stomach (8 cases, 32%) and colon (8 cases, 32%), followed by liver (2 cases, 8%) and kidney (2 cases, 8%), and one case occurring in the small bowel, gastrointestinal tract, pancreas, lung, and prostate each. The average time between diagnosis of primary tumor and the presence of metastasis to the SC in the 15 metachronous cases (60%) was 42 months (range: 2 to 108 months), while seven cases (28%) were synchronously detected and three cases (12%) were found as an occult cancer. The metastatic SC tumors extending to the epididymitis were found in six (32%) of the identified 19 cases, of which two cases (11%) invaded the testis.

The vast majority of cases as well as our case underwent radical orchietomy for the metastatic tumors in the SC, while tumor resection alone with preservation of the testis was found in two cases (8%). Over half of the cases received adjuvant interventions such as chemotherapy, molecular therapy, or hormone therapy based on the regimens for the primary tumors.

The prognosis of a metastatic tumor in the SC has been typically unfavorable as previously reported [4]. In this review, the 2-year survival rate in the postmetastasis to the SC was 36% in a total of the 16 patients identified since 2000, including our case, using the Kaplan-Meier method with a median follow-up duration of 12 months (range: 0.47 to 26 months) (Figure 4). Although there have been a small number of cases and short follow-up duration, the prognosis in patients with a metastatic SC tumor seems to be unfavorable even in the recent cases. However, the patients with late-onset (6 years or more) metastasis to the SC are likely to be a favorable prognosis because four (80%) of the five patients had been alive without recurrence after radical orchietomy with a mean follow-up duration of 16 months.

The mechanisms of metastasis to the SC and paratesticular tissues from primary malignant neoplasms have not been precisely elucidated. However, several possibilities have been proposed. The main routes have been postulated to be vascular and lymphatic routes. Other routes involving retrograde extension through the vessel, either along its lumen or by direct extension via the wall of the vessel, and transperitoneal seeding through the patent tunica vaginalis have been proposed [3, 4]. In our case hematogenous or lymphatic spread may be possible due to the positive lymphovascular invasion as well as the evidence of the paracolic lymph nodes metastasis in the primary neoplasm, and the late recurrence after the treatment of the primary site may be related to the activation of long-lasting tumor dormancy in distant organs including the SC.

In conclusion SC solid masses are usually not considered as SC metastasis from primary neoplasms such as gastrointestinal tract cancers previously treated with a curative intention. We did not initially consider this case to be a metastasis from cecal cancer. However, in patients with solid mass of the SC and a history of neoplasm, especially in the gastrointestinal tract, and even though the primary neoplasm has been treated with a curative intent long time earlier, the solid mass of the SC should be kept in mind in a possibility of metastasis from the primary tumor.
### Table 1: Reports of metastatic tumors of the spermatic cord in adults since 2000.

| Case number | Author (year) | Age | Site | Symptoms | Tumor size (SC in diameter, cm) | Primary site | Duration of detection of SC metastasis from primary site diagnosis | Histopathology (SC tumor) | Involved structure | Treatment (SC tumor) | Prognosis after treatment |
|-------------|---------------|-----|------|----------|--------------------------------|--------------|----------------------------------------------------------------|--------------------------|-------------------|---------------------|--------------------------|
| 1           | Ota et al. [5] (2000) | 51  | Left | Painless scrotal swelling | 2.5 | Stomach | 9 years | Poorly differentiated adenocarcinoma | Epididymis and parietal tunica vaginalis | Radical orchiectomy + chemo (MTX, 5-FU) | Died, 1 year |
| 2           | Polychronidis et al. [6] (2002) | 63  | Left | Painless scrotal swelling | 2 | Colon (sigmoid) | Occult | Mucus-secreting adenocarcinoma | Intact | Radical orchiectomy | ND |
| 3           | Bawa et al. [7] (2003) | 85  | Left | Incidentally found by castration | ND | Prostate | Synchronous | Adenocarcinoma | Vas deferens | Radical orchiectomy + hormone therapy | ND |
| 4           | Salesi et al. [8] (2004) | 62  | Left | Mass in scrotum | ND | Gastrointestinal tract | Occult | Adenocarcinoma | Epididymitis | Orchifunicolectomy + chemo (CDDP, Epirubicin, 5-FU) | Died, 5 months |
| 5           | Bandyopadhyay et al. [9] (2005) | 67  | Right | Mass in groin | ND | Pancreas | Synchronous | Moderately differentiated adenocarcinoma | ND | Radical orchiectomy + distal pancreatectomy | ND |
| 6           | Kaya et al. [10] (2006) | 62  | Left | Painful mass in inguinal site | 4.5 | Lung | Synchronous | Non-small cell adenocarcinoma | Intact | Radical orchiectomy | Died, 2 weeks |
| 7           | Shida et al. [11] (2006) | 75  | Left | Mass in inguinal site | 5 | Colon (ascending) | 2 months | Poorly differentiated adenocarcinoma | Intact | Radical orchiectomy | Died, 6 months |
| 8           | Miyake et al. [12] (2007) | 60  | Right | Mass in inguinal site | 3 | Colon (ascending) | 1 year and 8 months | Moderately differentiated adenocarcinoma | Intact | Radical orchiectomy | ND |
| 9           | Paravastu et al. [13] (2007) | 62  | Left | Painless scrotal swelling | ND | Colon (descending) | Synchronous | Poorly differentiated adenocarcinoma | Intact | Radical orchiectomy + chemo (iri-notecan, fluorouracil, cetuximab) | Alive, 18 months |
| 10          | Galanis et al. [14] (2009) | 80  | Right | Painful mass in inguinal site | ND | Colon (cecum, ascending, sigmoid) | Synchronous | Adenocarcinoma | Intact | Radical orchiectomy | Died, early postoperative period |
| Case number | Author (year) | Age | Site | Symptoms | Tumor size (SC in diameter, cm) | Primary site | Duration of detection of SC metastasis from primary site diagnosis | Histopathology (SC tumor) | Involved structure | Treatment (SC tumor) | Prognosis after treatment |
|-------------|---------------|-----|------|----------|--------------------------------|--------------|---------------------------------------------------------------|--------------------------|-------------------|---------------------|--------------------------|
| 11          | Chang et al. [15] (2009) | 38  | Right| Scrotal enlargement and chronic testicular pain | ND           | Liver                        | 7 months                                               | Cholangiocarcinoma (Klatskin tumor)  | ND                | Biopsy of the spermatic cord tumor careful surveillance | Alive, 5 months          |
| 12          | Correa et al. [16] (2009) | 57  | Left | Mass in inguinal site | 5             | Left kidney                  | Synchronous                                           | Renal cell carcinoma clear cell type | ND                | Radical orchiectomy + radical nephrectomy + Sunitinib | Alive, 1 year           |
| 13          | Schaefer et al. [17] (2010) | 64  | Right | Painful mass in groin and scrotum | 2             | Stomach                      | Synchronous                                           | Signet ring cell carcinoma | Epididymitis and testis | Radical orchiectomy + chemo (paclitaxel, leucovorin, 5-FU: FLF regimen) | Died, 1 year            |
| 14          | Ishibashi et al. [18] (2011) | 71  | Right | Mass in groin | 3.8           | Colon (cecum)                | 1 year                                                 | Well-differentiated adenocarcinoma | Intact            | Radical orchiectomy + chemo (S-1) | Alive, 15 months without recurrence |
| 15          | Chiang et al. [19] (2011) | 57  | Right | Painful hard mass in inguinal site | 2             | Liver                        | 6 years                                               | Hepatocellular carcinoma | Intact            | Radical orchiectomy + adjuvant radiotherapy | Alive, 6 months without recurrence |
| 16          | Mohammadi et al. [20] (2011) | 57  | Left | Painless mass in high scrotal site | 6.5           | Left kidney                  | 3 years                                               | Renal cell carcinoma (clear cell type) | ND                | Tumor resection with preserved testis | Alive, 3 months without recurrence |
| 17          | Al-Ali et al. [21] (2012) | 77  | Left | Inguinal and testicular pain | ND           | Colon (descending)          | 2.5 years                                              | Adenocarcinoma | Epididymitis and capsule of the testis | Radical orchiectomy | ND                          |
| 18          | Lee et al. [22] (2012) | 57  | Left | Mass in inguinal site | 4             | Stomach                      | 3 years                                               | Poorly differentiated adenocarcinoma | Intact            | Radical orchiectomy | ND                          |
| 19          | Watanabe et al. [23] (2013) | 52  | Right | Mass in inguinal and scrotal site | 2             | Stomach                      | 2 years                                               | Poorly differentiated adenocarcinoma | ND                | Radical orchiectomy + chemo (CDDP) | Died, 10 months          |
| Case number | Author (year) | Age | Site | Symptoms | Tumor size (SC) in diameter, cm | Primary site | Duration of detection of SC metastasis from primary site diagnosis | Histopathology (SC tumor) | Involved structure | Treatment (SC tumor) | Prognosis after treatment |
|-------------|---------------|-----|------|----------|-------------------------------|-------------|-------------------------------------------------|--------------------------|------------------|----------------------|------------------------|
| 20          | Valizadeh et al. [24] (2013) | 36  | Right | Painful mass in inguinal site | 1.6 | Small bowel | Occult | Adenocarcinoma | ND | Radical orchiectomy + small bowel tumor resection + chemo (capecitabine plus oxaliplatin regimen) | Alive, 6 months without recurrence |
| 21          | Kanazawa et al. [25] (2013) | 66  | Right | Groin pain | 4.2 | Stomach | 1 year | Moderately differentiated tubular adenocarcinoma | ND | Tumor resection with preserved testis + chemo | ND |
| 22          | Xu and Wang [26] (2013) | 50  | Bil  | Mass in spermatic cords | ND | Stomach | 4 years | Signet ring cell carcinoma | Epididymitis and seminiferous duct | Radical orchiectomy | ND |
| 23          | Kim et al. [27] (2014), Case 1 | 49  | Right | Mass in scrotum with discomfort in spermatic cord | 4 | Stomach | 7 years | Mucinous adenocarcinoma with signet ring cell carcinoma | Epididymitis | Radical orchiectomy + chemo (folinic acid, fluorouracil, oxaliplatin; FOLFOX regimen) | Alive, 26 months without recurrence |
| 24          | Case 2 | 60  | Left | Inguinal pain | 3.5 | Stomach | 6 years | Mucinous moderately differentiated adenocarcinoma | Intact | Radical orchiectomy + adjuvant radiation | Alive, 20 months without recurrence |
| 25          | Present study | 68  | Right | Painless mass in inguinal site | 4.5 | Colon (cecum) | 6 years | Moderately differentiated adenocarcinoma | Intact | Radical orchiectomy + chemo (S-1) | Alive, 12 months without recurrence |

S-1: tegafur/gimeracil/oteracil.
ND: Not documented, SC: Spermatic cord.
Consent
Written informed consent was obtained from the patient for publication of this case report and any accompanying images and extirpated specimens.

Conflict of Interests
The authors declare that there is no conflict of interests as to the publication of this paper.

Authors’ Contribution
Daisaku Hirano contributed to writing the paper including acquisition and analysis of data and surgery. Mizuho Ohkawa contributed to acquisition of data especially regarding prior reported cases. Ryo Hasegawa, Norimichi Okada, and Naoki Ishizuka contributed to surgery. Yoshiaki Kusumi contributed to carrying out the pathologic confirmation. All authors read and approved the final paper.

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