Int J Clin Exp Med 2018;11(10):11317-11319
www.ijcem.com /ISSN:1940-5901/IJCEM0073470

Case Report

Gastrointestinal stromal tumor of the small intestine with penile metastasis: a case report

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Received January 25, 2018; Accepted July 2, 2018; Epub October 15, 2018; Published October 30, 2018

Abstract: Gastrointestinal stromal tumor (GIST) is the most common mesenchymal tumor in the alimentary tract. GISTs most commonly occur in the stomach (60%), jejunum, and/or ileum (30%). To the best of our knowledge, there have been lacking cases reported in the literature about penile metastasis of GIST. Here, we described one case of liver and penile metastases in a patient who had advanced GIST. The patient underwent a partial small intestine resection of the primary tumor with negative margins but no follow-up adjuvant therapies. After 4 years since his enteral tumor resection, multiple liver metastases were found in succession. Then a two-year course of imatinib mesylate (Gleevec) combined with surgical intervention plus percutaneous microwave coagulation therapy (PMCT) was initiated for treatment. He had penile metastasis at 11 years after resection of the primary tumor, and this mass was resected in a short time. This case is worthy to be reported because penile metastasis from GIST is very rare. It might draw more attention to the diagnosis of the genital organ metastases from GISTs in clinical practice despite the shortage of available data so far.

Keywords: Gastrointestinal stromal tumor, metastasis, penis metastasis, case report

Introduction

Gastrointestinal (GI) stromal tumors (GISTs) are considered potentially malignant neoplasms arising from mesenchymal cells and most "clinically relevant" GISTs originate in the stomach (60%), followed by the small intestine (30%), and the colon/rectum (5%) [1]. Liver and peritoneal metastatic lesions are often observed but penile metastasis is rarely found in GIST. Recently there has been a report about penile metastasis of GIST which is most described in literature [2]. However, this is the first report of metastatic penile tumor from GIST (the small intestine in this case) after a long course of treatment for the original disease.

Case report

A 41-year-old male patient came to the clinic on September 7th, 2016, complaining of feeling a penile mass gradually growing for 3 months without obstructive urinary symptoms. A detailed past history was taken at outpatient clinic. He stated he received partial small intestine resection in October 2005 for small intestine GIST but refused any adjuvant therapies or regular follow ups. In 2009, a lesion (2 cm in maximum diameter) which located in the left liver was found as he had pain in the upper quadrant and was treated using imatinib 400 mg/d because the mass biopsy result showed liver metastasis consistent with his original small intestinal stromal tumor and gene sequence analysis showed a KIT exon 11 mutation. However, the liver mass grew up to 6 cm in diameter in 2011, and an unavoidable hepatectomy was done. Unfortunately, multiple liver metastases were confirmed by the upper quadrant MRI arose in the subsequent 12 months, percutaneous microwave coagulation therapy (PMCT) was used afterwards and the patient was followed with imaging studies every six months since that time.

In 2016, presenting with penile tumor, he had physical examination which revealed a 3.5 cm × 2 cm elliptical and solid mass located in the scapus penis. Penile ultrasonography showed a

Figure 1. Penile ultrasonography showed a low echo-level mass, 33 × 13 mm, with a well-defined boundary, regular shape, and irregular inside echo.

low echo-level mass (**Figure 1**). A complete gross excision of the mass was carried out on September 19, 2016. The surgical specimen was a tunicary and firm tumor (**Figure 2**). Histopathological examination showed epithelioid and spindle cells, which was consistent with the primary tumor, less than 5 mitoses at 50 × magnification, and there was partial necrosis (**Figure 3A**). In the immunochemical analysis, CD 117, Dog-1, Nestin, p53, and SMA were positive, whereas CD34, DM, EMA, S-100, and CKpan were negative, and the Ki-67 index was +1% (**Figure 3B**, **3C**). The tumor was completely removed with the total envelope with a negative incisal edge from the cavernosa. After the operation, the patient refused any medical interventions including sunitinib or regorafenib. There was no sign of tumor recurrence in the next 16 months of follow-up until the current time.

Discussion

Most GIST tumors are diagnosed in clinical practice and considered to be potentially malignant tumors, and for those patients who present with metastatic GIST, the median survival is reported to be 19 months [3]. Liver and peritoneal cavity are supposed to be the main metastatic lesions and the lymph nodes metastasis are uncommonly observed [1, 3]. Carlson et al. had reported a case which thought to be a penile metastatic lesion arising from the rectal GIST [2]. Recently, a case of stomach GIST with a solitary simultaneous metastasis in the left axillary lymph node was reported [4]. However, to the best of our knowledge, the case reported here is the first reported case of a GIST with penile metastasis from the small intestine with a long overall survival.

However, the distinct pattern of penile metastasis of gastroenteric tumors is unclear. In another case reported by Zhang et al. it was hypothesized that when tumor cells invaded the liver, they had a higher probability to spread into peripheral blood, and tumor cells could then migrate into the peripheral lymph nodes [5]. Anyway, it is clear that the malignant cells prefer organs which are abundant in blood flow to make the “new home” and the penis is a special organ that has a flexible blood flow rate. It can be assumed that the tumor cells migrate into the corpus cavernosum when an erection occurs whereas they settle and proliferate afterwards.

It is recommended that surgery including macroscopically complete resection can be used for those GISTs that are potentially malignant or malignant tumors confirmed with pathology with the intention of R0 resection. Imatinib is widely used as the first line treatment for metastatic or unresectable KIT-positive GIST like this case, Unfortunately, this patient seemed to be resistant or intolerant to imatinib which may become one of the reasons for him to remain conservative to second-line medical treatment. Recent discoveries have extended our understanding of GIST biology beyond mechanisms of KIT/PDGFRA-inhibitor treatment response and resistance which may explain this situation [6]. For multiple primary or metastatic tumors especially in the liver or lungs, percutaneous microwave coagulation therapy (PMCT) seems to be a feasible option, with less invasiveness and better short-term optimistic results, especially for the patients who are not suitable for open surgery. We used PMCT in this case and

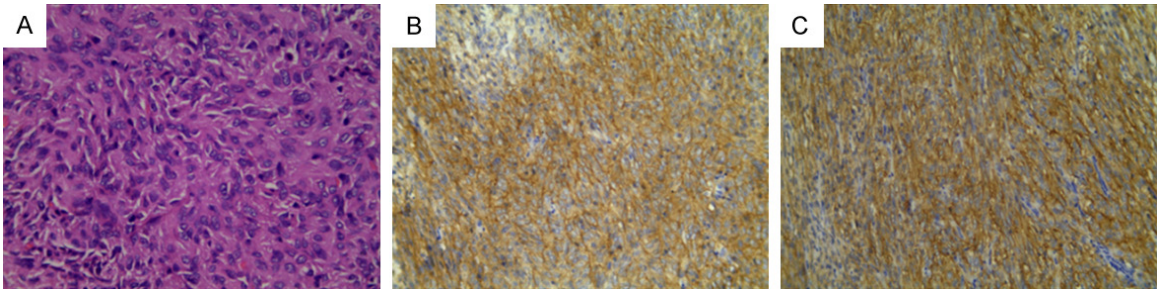

Figure 2. Surgical specimen. A. The tumor was located in the corpus cavernosum of scapus penis. B. The surgical specimen consisted of a 3.5 cm × 1.5 cm large, elliptical, tunicary and firm tumor. C. The cross section of the specimen revealed soft and fresh tissue, and no necrosis on gross examination.

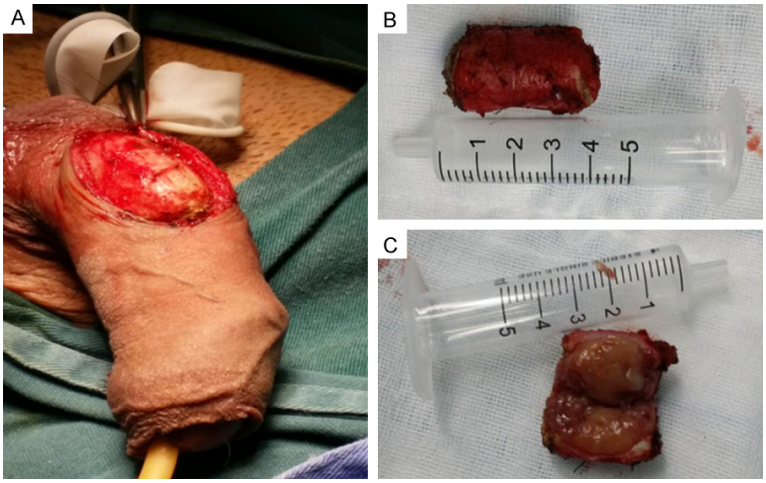

Figure 3. Histopathological examination showed epithelioid and spindle cells. (A, Hematoxylin and eosin staining, original magnification × 400). Immunohistochemical staining showed strong positivity of CD117 (B) and DOG-1 (C) (original magnification × 200).

there was no sign of subsequent liver recurrence.

In conclusion, more attention should be paid to the penile metastases from GISTs in clinical practice, and further studies are necessary to explore the mechanism of metastasis.

Acknowledgements

This work was supported by Zhejiang Provincial Medical and Healthy Science and Technology Projects (Grants No. 2016ZDB013; 2018ZD-047), National Natural Science Foundation of China (Grants No. 81772537; 81374014), Zhejiang Provincial Science and Technology Projects of Traditional Chinese Medicine (Grant No. 2017ZB089), and Zhejiang Provincial Science and Technology Projects (grants no. LGF18H160041, 2017C33212, 2017C33213, and 2015C33264).

Disclosure of conflict of interest

None.

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