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Rectal gastrointestinal stromal tumor with metastasis to the penis: Case report and review of literature

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ABSTRACT

We report the case of a 51-year-old gentleman with previously diagnosed gastrointestinal stromal tumor (GIST) of the rectum with metastasis to the penis. The patient underwent abdominoperineal resection of the primary tumor with negative margins and completed a three-year course of imatinib mesylate (Gleevec). Forty months after resection of his rectal tumor, the patient presented to his urologist with worsening testicular pain, mild lower urinary tract obstructive symptoms, and nocturia. A pelvic MRI revealed the presence of an ill-defined mass in the right perineum extending from the base of the penis to the penoscrotal junction. Biopsy of this mass was consistent with metastatic GIST. To our knowledge, this is the first report of metastatic GIST to the penis.

1. Introduction

Gastrointestinal stromal tumors (GIST) are mesenchymal neoplasms that account for approximately 1–3% of all malignant gastrointestinal tumors [1]. Clinical characteristics of the tumor are dependent on the anatomic location, size, and aggressiveness of the tumor. Primary GISTs originate most commonly in the stomach [2], but primary GISTs arising from the pleura, retroperitoneum, omentum, and mesentery have also been reported [3]. GISTs typically metastasize to intra-abdominal locations, with common metastatic sites being the liver, omentum, and the peritoneal cavity [4]. Metastasis of GISTs to lymph nodes and extra-abdominal sites via lymphatics is rare [5].

Imatinb mesylate (Gleevec) has proven to be a safe and efficacious adjunct in the treatment of KIT (CD117)-positive GISTs and has been used for the last 15 years for the treatment of metastatic GISTs [6,7]. A literature review of the metastatic potential of GISTs identified one case of a "lump at the base of the penis" thought to be a metastatic lesion arising from a primary rectal tumor [8]. Although there have been no reported cases of metastatic GIST to the corpora of the penis, cases of GIST metastases to the prostate from primary rectal tumors are documented [9–11]. In this report, we describe the first documented case of metastatic GIST to the penis.

2. Presentation of case

A 51-year-old Caucasian male was found to have a 6.5 × 6 cm rectal mass just proximal to the sphincter complex on screening colonoscopy. Biopsy of the mass demonstrated GIST with mitotic figures. The patient was started on neoadjuvant imatinib therapy to downstage the tumor prior to surgical resection. Four months after initiating treatment, an MRI of the pelvis revealed excellent response to imatinib, as the rectal mass had reduced in size to 3.8 × 3.1 × 2.2 cm. After a total of eight months of neoadjuvant imatinib therapy, the patient underwent an abdominoperineal resection (APR). The patient did well peri-operatively. Pathology of the surgical specimen revealed a gastrointestinal stromal tumor 2.8 cm in greatest extent, 80–90% hyalinization with scarce cellularity (Figs. 1 and 2). The lesion involved the mucosa, submucosa, muscularis propria, and perirectal soft tissue. Mitotic rate of <1/50hpf was visualized. Perineural invasion was identified, but lymphovascular invasion was not. Negative margins were attained, and eleven of eleven lymph nodes collected were negative for malignancy. Imatinib therapy was continued postoperatively.

One year after APR, a surveillance colonoscopy was performed, and no recurrent or metachronous lesions were found. The patient was followed with imaging studies every six months. CT scans of the abdomen and pelvis obtained over following three years revealed no evidence of intra-abdominal or pelvis abnormalities or evidence of recurrent or metastatic disease. Imatinib was discontinued after a total of three years of treatment.

Three and a half years after APR, the patient presented with complaints of obstructive urinary symptoms, erectile dysfunction, and progressively worsening testicular pain radiating towards his rectum. A physical examination identified no gross lesions of the

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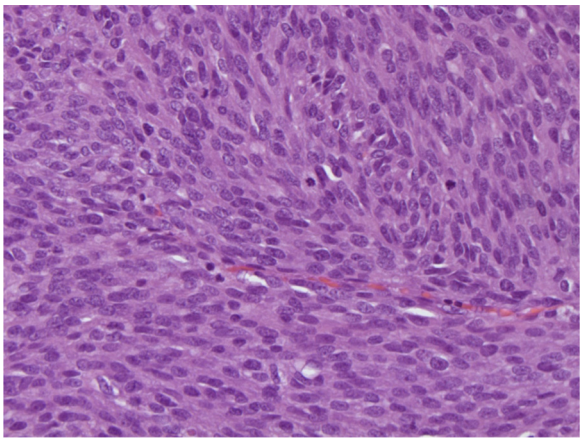

Fig. 1. Microscopic analysis of the rectal GIST. Tumor cells are spindled, forming fascicles with increased mitotic rate.

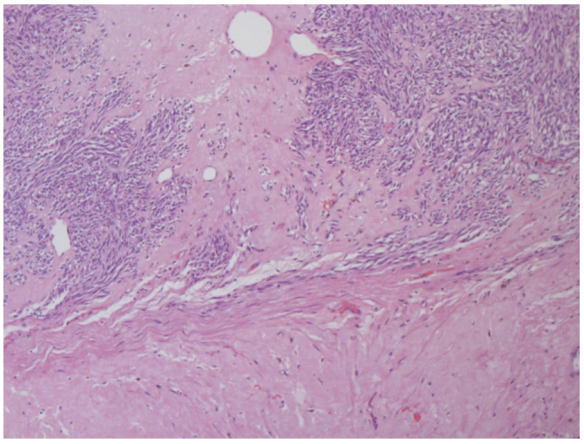

Fig. 2. Microscopic analysis of the rectal GIST. The tumor demonstrates large areas of hyalinization secondary to therapy with imatinib.

penis. A CT scan of the abdomen and pelvis was unremarkable, and cystoscopy showed no evidence of obstruction or abnormality of the urethra or bladder. The patient was treated for suspected epididymitis vs. prostatitis.

The patient continued to experience worsening right testicular, right hip, and perineal pain, which was moderately relieved by application of a heating pad and sitz baths but not by narcotic pain medication. On examination, the proximal penile shaft was tender to palpation, and an ill-defined mass was noted in the right perineum extending from the base of the penis to the penoscrotal junction. A pelvic MRI revealed an $8.4 \times 4.3 \times 3.6$ cm enhancing mass in the right corpus cavernosum (Figs. 3 and 4). The patient was referred to Urologic Oncology. Needle biopsies of the penile lesion were obtained, and histologic evaluation of these specimens revealed a cellular spindle cell tumor with mitoses, positive CD117 immunostain, and negative for desmin, findings which are consistent with metastatic GIST.

Medical and surgical options were discussed with the patient, who opted to continue imatinib therapy, as opposed to undergoing surgery. Three months after resuming imatinib, the patient reported decreased perineal pain and ability to obtain an erection with the use of a vacuum device. Four months after resuming imatinib, an MRI of the pelvis demonstrated that the metastatic penile lesion had decreased in size (Fig. 5). The patient was then receptive to undergo surgical resection of the right corpus cavernosum. Pathological examination of the excised tumor revealed metastatic

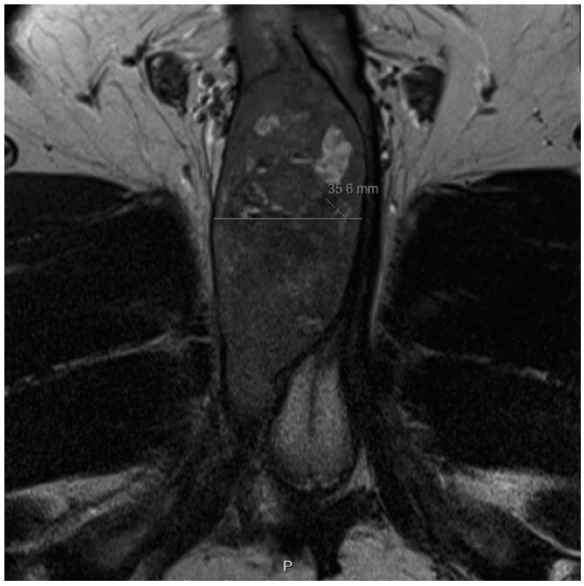

Fig. 3. Axial view of pelvis MRI. Axial view of the pelvis on MRI revealed a right corpus cavernosum mass.

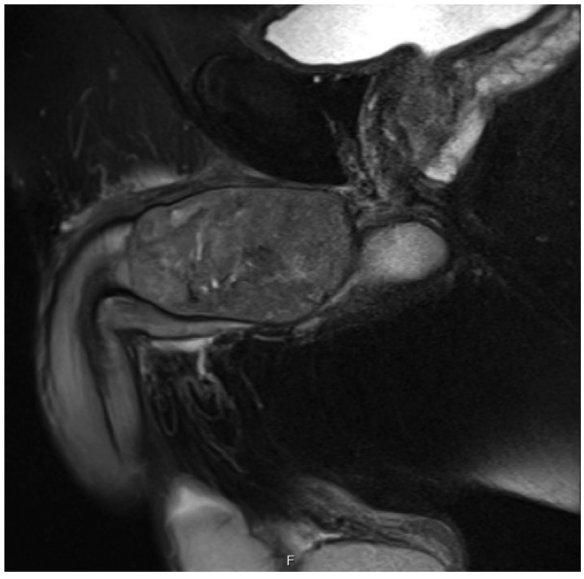

Fig. 4. Sagittal view of pelvis MRI. MRI of the pelvis demonstrated the right corpus cavernosum mass causing external compression of the urethra, leading to the patient's lower urinary tract obstructive symptoms.

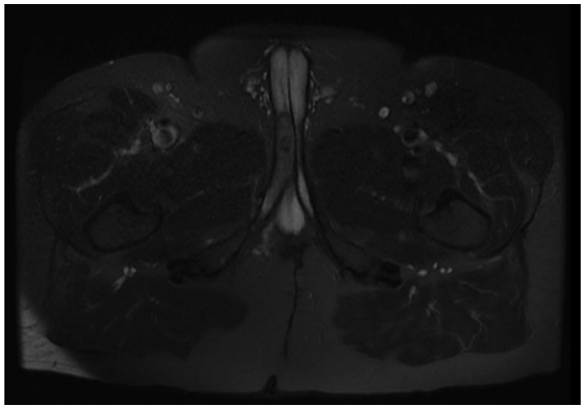

Fig. 5. Axial view of follow-up MRI. MRI of the pelvis obtained four months after resuming imatinib demonstrated decrease in the size of the right corpus cavernosum mass from $8.4 \times 4.3 \times 3.6$ cm to $4.9 \times 1.8 \times 1.5$ cm.