Dear Editor,

We present here a case report of “Krukenberg tumor” in male. In 1896, Friedrich Krukenberg (1871–1946), a German gynecologist and pathologist, described what he presumed as a new type of primary ovarian neoplasm. The true metastatic nature of this lesion was established 6 years later and termed as “Krukenberg tumor”. Now, the term “Krukenberg tumor” has been used either as a broad definition to indicate all metastatic tumors to the ovaries or to describe just metastatic tumors from the gastrointestinal tract containing the typical signet ring cell with intracellular mucin. Our case, as an analog in male, has criteria to be called “Krukenberg tumor” in male.

A 73-year-old male patient, who had a history of right inguinal testicle, was admitted to our department for right inguinal and left abdominal pain. Physical examination revealed a firm right testis that was located in the superficial inguinal pouch. On the other side, the left testis was normal. The physical examination also revealed that there was a palpable mass in the left lower abdomen. The whole abdominal computed tomography indicated a cryptorchid testis in the right groin (Figure 1) and a huge tumor at the descending colon (Figure 2). Colonoscopy was then performed showing a mass at the descending colon with intestinal stenosis. The biopsy of the colon mass indicated moderately differentiated adenocarcinoma. The laboratory tests of tumor markers such as carcinoembryonic antigen, CA 19-9, and alpha-fetoprotein were all in the standard range. Left hemicolectomy and right-sided orchiectomy were performed. During the operation, insufficient obliteration of the peritoneal gap in the abdominal inguinal ring with right cryptorchid testis was identified. There were no ascites, hepatic metastasis, or abdominal cavity metastasis, while the right cryptorchid testis was normal. The peritoneal cavity was washed with normal saline, and during the operation, retrograde spermiduct extension was suspected. Histopathological examination confirmed moderately differentiated adenocarcinoma of the colon mass (Figure 3) and right cryptorchid testicular metastatic adenocarcinoma (Figure 4) which was histologically similar to the colonic origin. The colon tumor had extended through the peritoneum. The surface of the right cryptorchid testis was involved (Figure 4), while the internal organization was not involved (Figure 5). Metastasis to three lymph nodes was also found in 14 examined mesenteric lymph nodes. The patient received postoperative chemotherapy that consisted of 12 cycles of mFOLFOX6. He was doing well during follow-ups and remained no recurrence at 20 months after surgery.

Reports on metastatic cancer of the testis, as analog Krukenberg tumor in male, are rare. Cryptorchid testicular metastasis, as a special type of testicular metastasis, scarcely has been reported. We searched the medical literature using the MEDLINE/PubMed database from 1950 to 2013. Our search yielded two cases of cryptorchid testicular metastasis, both from colon cancer. Rampa et al. reported a case of metastatic carcinoma from the sigmoid colon to the left cryptorchid testis. In another Spanish paper, Alvarez Pérez et al. reported a case of metastasis of carcinoma from the colon to cryptorchid testis. We described here for the first time, a metastasis of descending colon adenocarcinoma to the right cryptorchid testis.

Various pathways have been supposed as routes of cancer spread leading to the development of Krukenberg tumor. These include lymphatic spread, hematogenous spread, direct invasion, peritoneal seeding, etc. Al-Agha and Nicolas considered that retrograde lymphatic spread is the most likely route of metastasis. But Jun and Park reported the incidence of hematogenous recurrence was the highest. Other less common pathways of metastasis to the ovary occur via the peritoneum and vasculature. In male, the route for the spread of metastasis to the testes might be similar. Some possible pathways have been reported, including a retrograde lymphatic extension, direct invasion or extension from the original lesion, arterial and venous embolization from the tumor, and retrograde spermiduct extension. Our case showed that the testicular maldescending led to an insufficient obliteration of the peritoneal gap in the abdominal inguinal ring and generated a patent processus vaginalis. This might provide a potential route for gastrointestinal tumors spread through the patent processus vaginalis to testis. Therefore, our case provided a new pathway for the development of “Krukenberg tumor” in male.

The incidence of “Krukenberg tumor” in male is lower than that in female. Yada-Hashimoto et al. found that metastatic tumors accounted for 21.1% (64/304) of malignant ovarian tumors. While metastatic carcinoma to the testes is extremely rare, the incidence rates vary from 0.02% to 2.5%. Such a big difference might be explained in two reasons at least. One reason is that the sufficient obliteration in the abdominal inguinal ring prevents tumor transperitoneal seeding on testis in most
males. The other reason is that the lower temperature of the scrotum may be an unacceptable environment for growth of metastatic tumor cells, which was also supported by the study of Smallman and Odedra. In the present case, the male who had cryptorchid or maldescended testis, lost these advantages, therefore, having the same chance to develop “Krukenberg tumor” as female. Further studies are required to characterize this rare disease.

AUTHOR CONTRIBUTIONS
QX cared for the patients, collected the clinical information and drafted the manuscript. YWY helped to collect the pathological data. XLL, LRD, JGS, and LBW revised the manuscript and participated in critical discussion. All authors read and approved the final manuscript.

COMPETING INTERESTS
The authors declare no competing interests.

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