Klatskin Tumor with Spermatic Cord Metastasis: A Case Report

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Malignant spermatic cord tumor is quite uncommon. Metastatic spermatic cord tumor is even rarer. Here we report a case of metastatic spermatic cord tumor from a common hepatic duct tumor (Klatskin tumor). A 38-year-old man presented with right scrotal enlargement and chronic testicular pain. He had a Klatskin tumor (cholangiocarcinoma) stage IIIa, and underwent an extended right hepatectomy with resection of the extrahepatic bile duct, and portal vein and reconstruction by Roux-en-y hepatico-jejunostomy one year before this presentation. Scrotal ultrasound revealed a moderate hydrocele with clean content. Scrotal exploration showed turbid fluid and a fibrotic hyperemic spermatic cord. Excision biopsy of the spermatic cord was done, and the pathology revealed adenocarcinoma. The primary cholangiocarcinoma tumor and the spermatic cord tumor showed identical histologic patterns. Klatskin tumor with spermatic cord metastasis was diagnosed. To our knowledge, this case represents the first such report in the literature. *(Chang Gung Med J 2009;32:104-7)*

Key words: spermatic cord tumor, Klatskin tumor, adenocarcinoma, hepatic duct tumor

Malignant spermatic cord tumor is quite rare. The majority of tumors found in this area are of mesoblastic origin.(1) Metastatic spermatic cord tumor is even rarer. Here, we report a case of a spermatic cord metastasis from a common hepatic duct tumor (Klatskin tumor). To our knowledge, this case represents the first such report in the literature.

CASE REPORT

A 38-year-old man came to Chang Gung Memorial Hospital due to right scrotal enlargement and chronic testicular pain for several weeks. He had a Klatskin tumor (cholangiocarcinoma) and had undergone an extended right hepatectomy, resection of the extrahepatic bile duct, resection of the portal vein and reconstruction with Roux-en-y hepatico-jejunostomy seven months prior to this admission. Pathology revealed stage IIIa cholangiocarcinoma. Physical examination results showed a normal testis and epididymis but a fibrotic spermatic cord. A scrotal ultrasound revealed a moderate hydrocele with clean content (Fig. 1) Under the impression of tumor metastasis with reactive hydrocele, scrotum exploration was performed. It showed turbid fluid and a fibrotic hyperemic spermatic cord (Fig. 2). Fluid cytology and excision biopsy of spermatic cord were done. The fluid cytology was negative for malignant cells. Pathology of the spermatic cord biopsy revealed large and small neoplastic glands in a sclerotic stroma. The primary liver tumor and this spermatic cord tumor shared identical neoplastic glands with intra-cytoplasmic mucin and moderate nuclear pleomorphism (Fig. 3). Therefore, a diagnosis of

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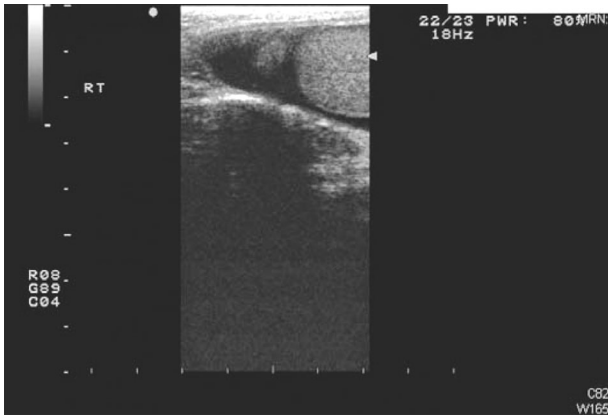

Fig. 1 Scrotal ultrasound reveals a moderate hydrocele with clean content.

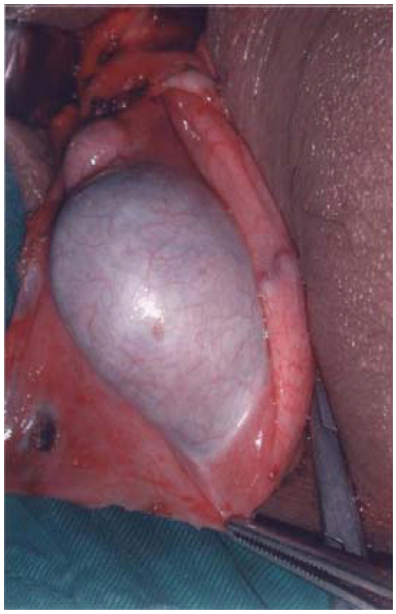

Fig. 2 Scrotum exploration shows turbid fluid and a fibrotic hyperemic spermatic cord.

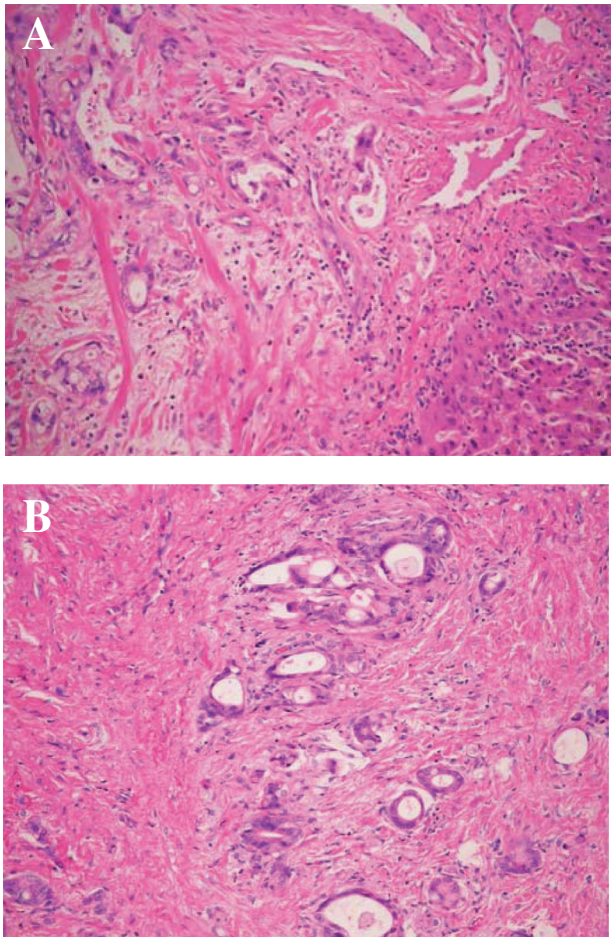

Fig. 3 (A) The primary liver tumor shows invasive large and small neoplastic glands with cytoplasmic mucin in a sclerotic stroma. (H&E stain, original magnification, x 200). (B) The metastatic tumor in the spermatic cord tumor shows neoplastic glands identical to those in the hepatic tumor with a sclerotic stroma. (H&E stain, original magnification, x 200)

Klatskin tumor with spermatic cord metastasis was made. The patient is alive and has been under careful surveillance for about 5 months up to the time of this report.

DISCUSSION

Klatskin tumor is an adenocarcinoma of the hepatic duct confluence. It was first described by Klatskin in 1965.[2] Cholangiocarcinoma is a relatively rare tumor, accounting for 2% of all diagnosed cancers.[3] Tumors involving the hepatic hilus are a challenge for surgeons due to a poor prognosis in most cases. Gerhards et al. showed a 5-year survival rate of less than 20%.[4] Patients with Bismuth type I & type II seem to have more favorable survival then those with type III.[4]

The spermatic cord is an extremely rare site for distant metastasis from a malignancy. Komeda et al. revealed that the most common metastasis to the spermatic cord is from the stomach (70.4%).[5] Other primary sources, such as the colon (28.9%) pancreas

(15.8%), bowel (13.2%) rectum (7.9%) bladder, lung and brain, have also been reported.[6-9] Most gastric primary sources were reported in Japan,[5] which may be due to the high incidence of gastric cancer in that country. Clinically, most patients with metastatic spermatic cord tumors suffer from a painless scrotal mass, although a lower inguinal mass and enlargement of the testis can occur. Hydrocele, hernia and testis tumor are the most common misdiagnoses of the tumor.[1] Typically, spermatic cord tumor presents as firm, non-tender, and not transluminate, and can be separated from the testis. The mechanism of Klatskin tumor metastasis to the spermatic cord is via retrograde lymphatic spread, retrograde venous spread or direct invasion. Retrograde lymphatic spread may be the most likely route. The local invasion of a Klatskin tumor induces lymphatic drainage obstruction. It may lead to dilation of the lymphatic vessels, and then the lymphatic valve is insufficient to maintain unidirectional flow of the lymph. The backflow may allow tumor retrograde metastasis to the spermatic cord. Clinically, our patient demonstrated an enlargement of the scrotum and swelling, and a firm spermatic cord was noted. Tumors with spermatic cord metastasis are in an advanced stage and grade; the prognosis is poor. Algaba et al. reported that the average survival from the diagnosis of metastasis was 9.1 months.[10] Chemotherapy or radiotherapy is controversial for the tumor with mestastasis. Mansfield et al. showed no benefit from palliative therapy.[3] Kuvshinoff et al. reported that in patients with Klatskin tumor who received radiotherapy, the mean survival time was only 14.5 months.[11] Three cases of bile duct tumor metastasis to the male genital organ have been reported, with two cases of metastasis to the testis[12,13] and one to the epididymus.[14] In conclusion, when a patient with a primary intra-abominal tumor presents with chronic testicular pain or a hydrocele, metastasis should be considered and further studies should be done.

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Klatskin 腫瘤合併精索轉移：病例報告

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惡性精索腫瘤相當少見，轉移性精索腫瘤更是少見。這裡我們報告一總膽管惡性腫瘤轉移至精索的案例。一位 38 歲病患因右側陰囊腫大併右睪丸疼痛至本院求診。病患曾在一年前因 klatskin 腫瘤接受切除手術。陰囊超音波顯示陰囊積水。病患接受陰囊探查手術且對精索加以切片。病理報告發現與先前總膽管惡性腫瘤有相同組織型態。Klatskin 腫瘤合併精索轉移因此確立。就我們所知，此爲第一例相關報告。(長庚醫誌 2009;32:104-7)

關鍵詞：精索惡性腫瘤，Klatskin 腫瘤，腺瘤，膽管惡性腫瘤

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