

# RECURRENT CHOLANGIOCARCINOMA PRESENTING AS OVARIAN KRUKENBERG TUMOR

Chuan-Chi Shih<sup>1</sup>, Yuh-Cheng Yang<sup>1,2,3,4</sup>, Jerry Cheng-Yen Lai<sup>4</sup>,  
Yu-Hui Huang<sup>1</sup>, Tao-Yeuan Wang<sup>5</sup>, Kung-Liahng Wang<sup>1,3,6\*</sup>

<sup>1</sup>Department of Obstetrics and Gynecology, Mackay Memorial Hospital, <sup>2</sup>Taipei Medical University,

<sup>3</sup>Mackay Medicine, Nursing and Management College, Departments of <sup>4</sup>Medical Research and

<sup>5</sup>Pathology, Mackay Memorial Hospital, and <sup>6</sup>National Taipei College of Nursing, Taipei, Taiwan.

## SUMMARY

The ovary is a common metastatic site for many neoplastic transformations of both gynecologic and non-gynecologic origins. The morphologic and clinical similarities between metastatic and primary tumors of the ovary often render confusion for clinicians at diagnosis. The bile duct is an extremely rare source of metastases (Krukenberg tumor). We present here a rare case of recurrent cholangiocarcinoma with metastatic adenocarcinoma of the ovary. [International Journal of Gerontology 2008; 2(2): 76–78]

**Key Words:** bile duct cancer, cholangiocarcinoma, Krukenberg tumor, neoplasm metastasis, ovarian neoplasms

## Introduction

Ovarian metastases can account for as many as 30–40% of all ovarian malignancies<sup>1–3</sup>. The potential for metastatic tumors to mimic the clinical and morphologic appearance of primary ovarian tumors is well known to surgical pathologists<sup>2</sup>. Correct diagnosis and successful management of patients with these diseases often rely on the degree of distinction between primary and metastatic tumors. Most of the recent literature emphasizes various origins for the tumors metastasizing to malignant ovarian tumors. Ovarian metastatic tumors originating from primary tumors in the gastrointestinal tract are collectively known as the Krukenberg tumors. The most common non-gynecologic primary tumors are frequently observed in the large intestine<sup>1,4</sup> or breast<sup>5,6</sup>. Cholangiocarcinoma or bile duct cancer metastasizing to the ovary is extremely rare. It is readily apparent that its retroperitoneal location and low

histologic specificity in the ovary often leads to a misdiagnosis. Limited clinical cases of this disease type make any further attempt in the accurate diagnosis based on clinical, radiologic and morphologic symptoms even more difficult. We present here a rare case of a woman with recurrent cholangiocarcinoma, presenting as an ovarian Krukenberg tumor. It is hoped that this case report can contribute to the understanding of this unusual malignancy.

## Case Report

A 61-year-old postmenopausal woman came to the outpatient department of gastroenterology because of abdominal distension and malaise for 1 month. Abdominal examination revealed distension and a palpable adnexal mass. Her past medical history indicated the diagnosis of cholangiocarcinoma with mesocolic and mesenteric metastases 2 years previously. She had received surgery and 15 courses of 5-fluorouracil and leucovorin as postoperative adjuvant chemotherapy until 10 months previously, followed by oral uracil-tegafur until presentation. Initial laboratory studies did not reveal abnormal findings, except elevation of tumor markers (CA-125 assay was 546.4 U/mL). She was referred



\*Correspondence to: Dr Kung-Liahng Wang, Department of Obstetrics and Gynecology, Mackay Memorial Hospital, 92, Section 2, Chung-Shan North Road, Taipei, Taiwan.  
E-mail: kl421229@ms6.hinet.net  
Accepted: February 10, 2008

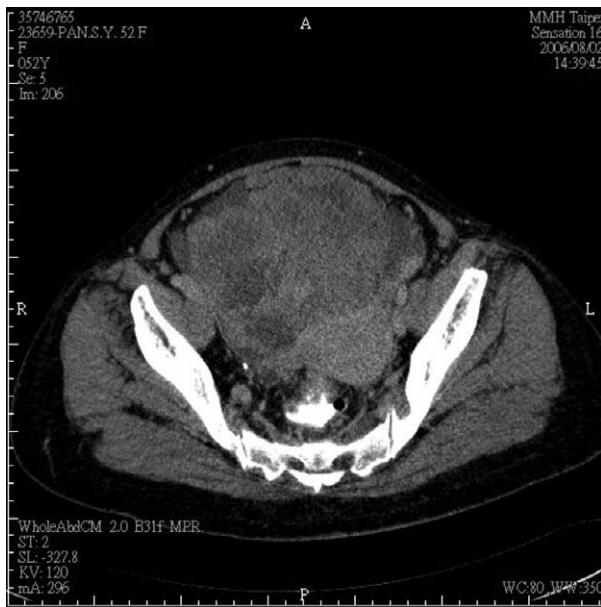


Figure 1. A heterogeneously enhancing mass in the lower pelvic region.

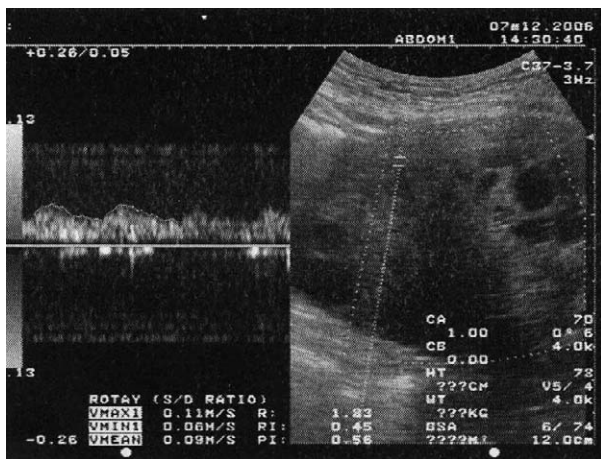


Figure 2. Complex cystic mass of 9×10 cm in size with septum and power Doppler blood flow (resistance index, 0.4).

to the outpatient department of gynecology for further evaluation after computed tomography of the abdomen showed a pelvic mass and massive ascites (Figure 1). Ultrasound examination revealed a complex cystic mass, measuring about 9×10 cm in size with septum, and power Doppler blood flow (resistance index, 0.4; Figure 2). Exploratory laparotomy revealed a bilateral ovarian tumor measuring 10×9 cm and 6×4 cm for the right and left ovarian tumors, respectively, and intra-peritoneal tumor implants. Bloody ascites of 2,000 mL in the abdominal cavity were also noted and sampled for cytologic examination. A surgical specimen of the ovaries indicated metastatic adenocarcinoma upon frozen

section analysis. Bilateral salpingo-oophorectomy and partial omentectomy were performed, and the associated pathology report was consistent with a diagnosis of metastatic adenocarcinoma compatible with cholangiocarcinoma. No further adjuvant treatment was carried out postoperatively as a result of rapid progression of carcinomatosis and intestinal obstruction. She died 5 months after operation.

## Discussion

We present here a rare case of a woman with cholangiocarcinoma, manifested as a pelvic mass as the result of ovarian metastasis. The exceptional rarity of ovarian metastatic tumors of bile duct origin in the literature initially led us to regard this case as a primary ovarian tumor. Being an unlikely and unfamiliar ovarian metastasis, the possibility of cholangiocarcinoma was precluded from our initial clinical diagnosis. The ovarian gross appearance was similar to those of primary ovarian tumors. However, in conjunction with the patient's past medical history of cholangiocarcinoma, the presentation of bilateral ovarian tumors and the frozen section analysis of the resected specimen allowed us to conclude that this was metastatic adenocarcinoma of the ovary. Metastatic tumors of the ovary often render clinical confusion in diagnosis of primary ovarian tumors<sup>5,7</sup>. An important clue that differentiates primary tumors from metastatic tumors is bilaterality. Fewer than 10% of primary ovarian neoplasms are bilateral<sup>7</sup>, and unilateral involvement of the ovarian metastases can be a good indication of primary ovarian cancer. A further classification has successfully subcategorized unilateral involvement with tumor sizes of less than 10 cm as metastatic, and those of more than 10 cm as primary ovarian cancers<sup>8</sup>. Other predominant features of metastatic tumors often involve the presence of signet ring cells, surface involvement by tumor cells, and extensive extraovarian tumor, whereas features of primary ovarian neoplasms include smooth external surfaces as well as a complex papillary pattern<sup>7</sup>. The management of patients with Krukenberg tumors after gastric cancer by metastasectomy has been shown to reveal significant improvement in overall and progression-free survival<sup>9-12</sup>. However, performing metastasectomy did not seem to improve the management of the patient with recurrent cholangiocarcinoma presenting as an ovarian Krukenberg tumor in this clinical case

report. We believe that a combination of cautious clinical evaluation and pathologic examination that allows us to better differentiate metastatic adenocarcinoma from primary ovarian carcinoma at diagnosis is extremely important. Further studies are required to characterize and improve the outcome of this rare disease.

## References

1. Webb MJ, Decker DG, Mussey E. Cancer metastatic to the ovary: factors influencing survival. *Obstet Gynecol* 1975; 45: 391–6.
2. Young RH, Scully RE. Metastatic tumors of the ovary. In: Kurman RJ, ed. *Blaustein's Pathology of the Female Genital Tract*, 5<sup>th</sup> edition. New York: Springer, 2002; 1063–101.
3. Yada-Hashimoto N, Yamamoto T, Kamiura S, Seino H, Ohira H, Sawai K, et al. Metastatic ovarian tumors: a review of 64 cases. *Gynecol Oncol* 2003; 89: 314–7.
4. Moore RG, Chung M, Granai CO, Gajewski W, Steinhoff MM. Incidence of metastasis to the ovaries from nongenital tract primary tumors. *Gynecol Oncol* 2004; 93: 87–91.
5. Mazur MT, Hsueh S, Gersell DJ. Metastases to the female genital tract: analysis of 325 cases. *Cancer* 1984; 53: 1978–84.
6. Petru E, Pickel H, Heydarfadai M, Lahousen M, Haas J, Schaidt H, et al. Nongenital cancers metastatic to the ovary. *Gynecol Oncol* 1992; 44: 83–6.
7. Lee KR, Young RH. The distinction between primary and metastatic mucinous carcinomas of the ovary: gross and histologic findings in 50 cases. *Am J Surg Pathol* 2003; 27: 281–92.
8. Seidman JD, Kurman RJ, Ronnett BM. Primary and metastatic mucinous adenocarcinomas in the ovaries: incidence in routine practice with a new approach to improve intraoperative diagnosis. *Am J Surg Pathol* 2003; 27: 985–93.
9. Kim HK, Heo DS, Bang YJ, Kim NK. Prognostic factors of Krukenberg's tumor. *Gynecol Oncol* 2001; 82: 105–9.
10. Cheong JH, Hyung WJ, Chen J, Kim J, Choi SH, Noh SH. Surgical management and outcome of metachronous Krukenberg tumors from gastric cancer. *J Surg Oncol* 2004; 87: 39–45.
11. Cheong JH, Hyung WJ, Chen J, Kim J, Choi SH, Noh SH. Survival benefit of metastasectomy for Krukenberg tumors from gastric cancer. *Gynecol Oncol* 2004; 94: 477–82.
12. Ayhan A, Guvenal T, Salman MC, Ozyuncu O, Sakinci M, Basaran M. The role of cytoreductive surgery in nongenital cancers metastatic to the ovaries. *Gynecol Oncol* 2005; 98: 235–41.