

## Intrahepatic biliary cystadenoma: Is there really an almost exclusively female predominance?

Ho Hyun Kim, Young Hoe Hur, Yang Seok Koh, Chol Kyoon Cho, Jin Woong Kim

Ho Hyun Kim, Young Hoe Hur, Yang Seok Koh, Chol Kyoon Cho, Department of Surgery, Chonnam National University Medical School, 519-809 Gwangju, South Korea

Jin Woong Kim, Department of Radiology, Chonnam National University Medical School, 519-809 Gwangju, South Korea

**Author contributions:** Kim HH wrote the paper; Koh YS and Cho CK contributed equally to this work; Hur YH designed the research and supervised the paper; Kim JW performed a radiologic evaluation.

**Correspondence to:** Young Hoe Hur, MD, Department of Surgery, Hepatobiliary and Pancreatic Tumor Clinic, Chonnam National University Hwasun Hospital, 160, Ilsim-ri, Hwasun-eup, Hwasun-gun, Jeollanam-do, 519-809, South Korea. [surgihur@naver.com](mailto:surgihur@naver.com)

Telephone: +82-61-3797646 Fax: +82-61-3797661

Received: March 8, 2011 Revised: April 8, 2011

Accepted: April 15, 2011

Published online: July 7, 2011

### Abstract

Biliary cystic tumors, such as cystadenomas and cystadenocarcinomas, are rare cystic tumors of the liver, accounting for less than 5% of all intrahepatic cysts of biliary origin. Biliary cystadenomas have been known to occur predominantly in women (> 85%), and 38%-44% of biliary cystadenocarcinomas have occurred in males. We wrote this letter to comment on a brief article (*World J Gastroenterol* 2011 January 21; 17(3): 361-365) regarding a case of intrahepatic biliary cystic neoplasm treated with surgery. The adenoma-carcinoma sequence is the possible mechanism of carcinogenesis. If the carcinogenesis of biliary cystadenocarcinoma occurs in the adenoma-carcinoma sequence, we believe that the male-to-female ratio of cystadenoma should be higher than the incidence rate that has been reported to date.

© 2011 Baishideng. All rights reserved.

**Key words:** Biliary cystadenoma; Cystadenocarcinoma; Carcinogenesis; Incidence

**Peer reviewers:** Giammarco Fava, MD, Department of Gastroenterology, Università Politecnica delle Marche, Ancona, via Gervasoni 12, 60129 Ancona, Italy; Selin Kapan, Associate Professor of General Surgery, Sadi Konuk Training and Research Hospital, Department of General Surgery, Kucukcekmece, Istanbul 34150, Turkey

Kim HH, Hur YH, Koh YS, Cho CK, Kim JW. Intrahepatic biliary cystadenoma: Is there really an almost exclusively female predominance? *World J Gastroenterol* 2011; 17(25): 3073-3074 Available from: URL: <http://www.wjgnet.com/1007-9327/full/v17/i25/3073.htm> DOI: <http://dx.doi.org/10.3748/wjg.v17.i25.3073>

### TO THE EDITOR

Biliary cystic tumors, such as cystadenomas and cystadenocarcinomas, are rare cystic tumors of the liver, accounting for less than 5% of all intrahepatic cysts of biliary origin. Biliary cystadenomas have been known to occur predominantly in women (> 85%), and this almost exclusively female predominance suggests a strong hormonal influence. Emre *et al*<sup>[1]</sup> reported nine patients with intrahepatic biliary cystic liver neoplasms, all of them were female. And 38%-44% of biliary cystadenocarcinomas occur in males with a higher mean age compared with cystadenomas<sup>[2]</sup>.

In spite of the improvement in imaging techniques, the differential diagnosis of simple hepatic cysts and intrahepatic biliary cystadenoma is still problematic.<sup>[1]</sup> If the malignancy is suspected, surgery is recommended; if benign disease is suspected, many clinicians might misdiagnose cystadenomas as simple cysts and recommend observation and/or follow-up examinations rather than surgical treatment. Cases of biliary cystic neoplasm reported in the literature are diagnosed mostly based on pathologic findings after operation. Thus, we examined whether there may be a bias toward the gender-related incidence of biliary cystadenoma.

Between May 2004 and December 2009, 10 patients underwent surgery for intrahepatic biliary cystic neoplasm at Chonnam National University Hospital, Gwangju, Korea. Eight patients had biliary cystadenomas, and two had cystadenocarcinomas. The patients with cystadenomas consisted of five females (62.5%) and three males (37.5%). Both patients with cystadenocarcinomas were males. In our report, the female predilection of biliary cystadenoma is much weaker than in other reports<sup>[1,2]</sup>.

The role of biliary cystadenoma in the pathogenesis of biliary cystadenocarcinoma is controversial. Transformation into a cystadenocarcinoma has been reported, although it is rare<sup>[3]</sup>. Thus, the adenoma-carcinoma sequence is a possible mechanism of carcinogenesis. Biliary cystadenomas have been known to occur predominantly in women. If the carcinogenesis of biliary cystadenocarcinoma occurs in the adenoma-carcinoma sequence, we believe that the male-to-female ratio of cystadenoma should be higher than the incidence rate that has been reported until now. The current knowledge and predictions

about intrahepatic biliary cystic neoplasms are based on a limited number of case reports. The precise mechanisms of carcinogenesis remain unknown. Thus, accumulation of larger groups of patients and further examinations will be necessary to analyze the pathogenesis and incidence of intrahepatic biliary cystic neoplasms (cystadenomas and cystadenocarcinomas).

## REFERENCES

- 1 **Emre A**, Serin KR, Ozden I, Tekant Y, Bilge O, Alper A, Güllüoğlu M, Güven K. Intrahepatic biliary cystic neoplasms: Surgical results of 9 patients and literature review. *World J Gastroenterol* 2011; **17**: 361-365
- 2 **Vogt DP**, Henderson JM, Chmielewski E. Cystadenoma and cystadenocarcinoma of the liver: a single center experience. *J Am Coll Surg* 2005; **200**: 727-733
- 3 **Kubota E**, Katsumi K, Iida M, Kishimoto A, Ban Y, Nakata K, Takahashi N, Kobayashi K, Andoh K, Takamatsu S, Joh T. Biliary cystadenocarcinoma followed up as benign cystadenoma for 10 years. *J Gastroenterol* 2003; **38**: 278-282

S- Editor Tian L L- Editor Ma JY E- Editor Ma WH