Adenosquamous Carcinoma of the Liver: A Case Report

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A case of primary adenosquamous carcinoma of the liver is reported. A large cystic mass with irregular enhancing wall was seen in the left lobe of the liver; multiple hepatic cysts and metastatic nodules were scattered in both lobes. Sonoguided needle biopsy was performed and on histopathology, adenosquamous carcinoma was diagnosed. The radiologic and pathologic relationships of this unusual neoplasm are discussed.

Index Words: Liver neoplasms, CT
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Primary adenosquamous carcinoma (ASC) of the liver is a very rare; on histology, a tumor is seen to contain both malignant squamous and glandular components. In the literature in English, only 23 cases of this neoplasm, including our case, have been reported since the first report in 1975 (3-4). No case has been reported in Korea. It is suggested that ASCs may arise from malignant transformation of squamous metaplasia in preexisting cholangiocarcinoma (1-3), from metaplastic squamous bile duct epithelium, or from congenital or non-parasitic hepatic cyst (4).

Case Report

A 72-year-old woman was admitted to our hospital suffering from epigastric pain, a sensation of chilling, and generalized weakness. She had a history of hepatitis C. Her vital signs were unremarkable except for fever. On physical examination, a fist sized, slightly tender mass was palpated in the epigastrium. Abnormal laboratory data included leukocytosis (15,700/dL), and decreased hemoglobin level (8.1 g/dL). Other laboratory studies including a Papanicolaou smear, were negative.

A helical computed tomographic (CT) scan of the liver performed at another hospital five months previously demonstrated multiple, variable-sized cystic lesions in both hepatic lobes, the walls of which were thin, even in the largest one (on the left), which had a diameter of 7 cm. (Fig. 1). A helical CT scan in our hospital revealed that the wall of the larger cystic lesion in the left hepatic lobe was thickened and enhanced (Fig. 2A), while other cystic lesions had not changed. In addition, multiple low attenuating lesions in the liver were noted; these indicated intrahepatic metastases as well as multiple lymphadenopathy in the porta hepatis, and in the peripancreatic, aortocaval, and paraaortic areas. Ultrasonography (US) showed that the cystic lesion

Fig. 1. Initial spiral liver CT reveals multiple hepatic cysts in both lobes with especially large one in left lobe (arrowheads).
had a similar thickened wall containing echogenic debris (Fig. 2B). No abnormalities were seen on chest radiograph, barium enema, and pelvic US. Automated gun biopsy was performed under US guidance aiming at the thickened wall of the largest cystic lesion. On histopathologic examination, tumor cells forming a tubular structure suggesting adenocarcinoma and well differentiated squamous cells with keratin pearl formation were found in a section of biopsy specimen, adenosquamous carcinoma was confirmed (Fig. 3).

**Discussion**

Primary ASC is in the gallbladder, extrahepatic bile ducts, and pancreas is unusual; in the liver it is particularly rare. In the literature in English, only 23 cases of this neoplasm, including our case, have been reported since the first report in 1975 (3-4).

As described above, ASCs consist of both adenocarcinoma and squamous cell carcinoma (SCC), ASC of the liver is thus considered to be a special type of cholangiocarcinoma. The pathogenesis of the squamous component in ASC is unclear; however, although various theories and experiments have been described (1-4). According to the literature, squamous metaplasia may arise from benign bile duct epithelium, from epithelium of a congenital or non-parasitic hepatic cyst, or from cholangiocarcinoma. Moore et al (5) reported a case of adenosquamous carcinoma of the liver, which was confirmed by histopathologic examination. However, the precise pathogenesis of squamous metaplasia in ASC remains unclear.

**Fig. 2.** Follow up spiral liver CT (A) reveals the wall of largest cystic lesion in left hepatic lobe thickened and enhanced (arrows). Intrahepatic and lymph node metastases were developed (arrowheads). US (B) reveals cystic lesion had thickened wall containing fine echogenic debris.

**Fig. 3.** The microscopic finding exhibits sheets of tumor cells occasionally forming tubular structure (arrow) and well differentiated squamous cell carcinoma frequent keratin pearl formation (arrowhead). (HE staining X100)
orted a case of ASC arising in a biliary cystadenocarcinoma, and also suggested that biliary hamartoma, which is believed to be the precursor of congenital hepatic cyst and biliary cystadenoma and cystadenocarcinoma, could be associated with ASC. Iemura et al (1) experimentally proved the metaplastic transformation in the nude mouse of adenocarcinoma to adenosquamous and squamous carcinoma. In addition, continued irritation by chronic infection, stones, and/or Clonorchis sinensis (2, 4) may be an etiologic factor, leading to metaplastic change in the biliary epithelium. The rarity of ASC of the liver may be simply explained by the fact that these irritative factors are less frequent in intrahepatic bile ducts than in the gallbladder.

It is well known that due to earlier metastasis, the prognosis of cholangiocarcinoma is worse than that of hepatocellular carcinoma. Furthermore, ASC generally has a worse prognosis than pure adenocarcinoma, because the existence of a squamous component may indicate an accelerated growth rate (2, 4). According to the literature, most of ASCs in the liver showed rapid growth, extensive necrosis, and early lymphatic and hematogenous metastases, resulting in a hopeless survival rate (2–4).

According to analyses in the literature of radiologic and macroscopic appearances, ASC might be either cystic or solid, but solid tumors were highly necrotic (2–5). A case of ASC diagnosed as liver abscess and treated by drainage and antibiotics was reported (5). In our case, the wall of the largest cystic lesion became thicker during five-months period, suggesting that the wall of a hepatic cyst was replaced by malignant cells, rather than that the tumor was largely necrotic. The limitation of our case is that we were unable to obtain on exact histopathologic diagnosis of this cystic lesion before squamous metaplasia. It is possible that the underlying histopathology of this cystic lesion was that of either a hepatic cyst or a unilocular cystadenoma or cystadenocarcinoma. In out case, multiple variable sized cystic lesions were at first scattered throughout the liver. It thus seems reasonable that the lesions should all be considered as hepatic cysts rather than as single unilocular biliary cystadenoma or cystadenocarcinoma among hepatic cysts, or as multiple biliary cystadenoma or cystadenocarcinoma.

In conclusion, it is believed that ASC in our case arose from a hepatic cyst, and that wall thickening and enhancement in such cysts can be a sign of malignant change.

References

1. Iemura A, Yano H, Mizoguchi A, Kojiro M. A cholangiocellular carcinoma nude mouse train showing histologic alteration from adenocarcinoma to squamous cell carcinoma. Cancer 1992; 70: 415-422
### 방사선 해부학(신경계 및 근골격계)

<table>
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<td>휴식</td>
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