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Solid Mesenchymal Hamartoma of the Liver in Adult

This paper presents an unusual solid mesenchymal hamartoma of the liver (MHL) in adult. A well defined solid mass in the left lobe of the liver was found in a 57-year-old female. Preoperative radiologic examinations demonstrated solid mass with multifocal calcifications abutting the gallbladder. By light microscopy, the lesion was composed of dense fibrous stroma with hyalinization, bile ducts and thick-walled vessels without hepatocytes. The solid and hyalinized mesenchymal component would suggest an unusual degenerative change representing a burnt-out MHL.

Key Words: *Mesenchymal hamartoma; Liver; Adult*

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INTRODUCTION

Mesenchymal hamartoma of the liver (MHL) is a rare cystic mass with predilection for infants in their first two years. It seldom occurs beyond the age of five with the average age being 10 to 15 months (1). It is thought to be a developmental anomaly rather than a true neoplasm, and consists of admixture of bile ducts, hepatocytes and mesenchymal tissue (1, 2). MHL rarely occurs in adults, and adult MHL shows series of histological modifications, i.e. progressive loss of hepatocytes, degeneration of bile duct epithelium, and cystic change of the mesenchymal component (3). We report here a unique case of solid MHL in adult showing burnt-out mesenchymal component.

CASE REPORT

A 57-year-old Korean woman visited Korea Cancer Center Hospital because of abdominal discomfort and weight loss of 14 kg during 18 months. Abdominal sonography and contrast-enhanced computed tomography (CT) were performed. The sonogram showed a solid mass containing calcifications, that was located in the medial segment of the left hepatic lobe abutting the gallbladder. On CT scan, the mass showed low attenuation with amorphous calcifications (Fig. 1). Serum alpha-fetoprotein and carcinoembryonic antigen were within normal limits. All viral markers were negative. The patient underwent tumor resection under the impression of gallbladder can-

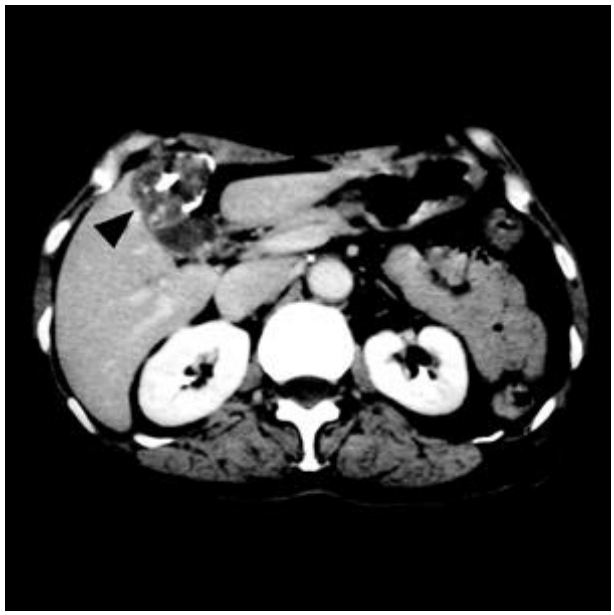

Fig. 1. Contrast-enhanced abdominal CT scan shows a low attenuating mass with central and peripheral calcification (arrow head).

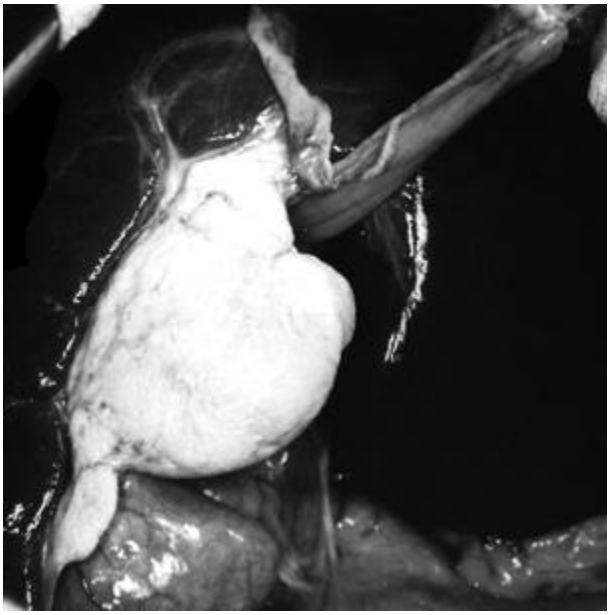

Fig. 2. Solid mass of the liver. A well-demarcated and entirely solid lesion is noted in the inferior edge of the left medial segment.

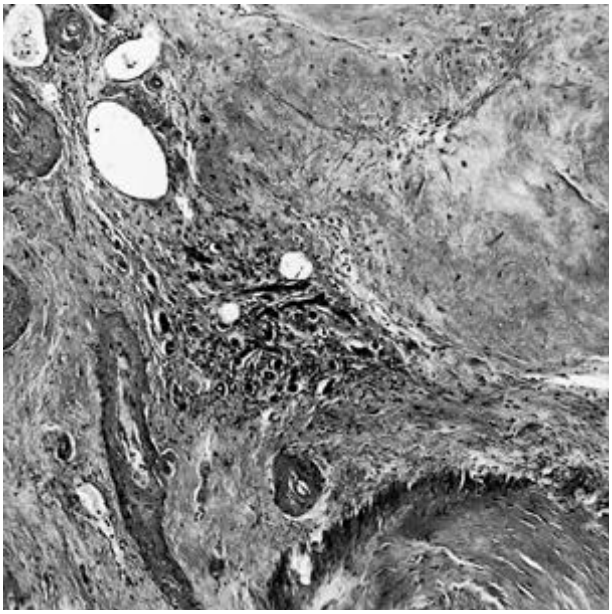

Fig. 3. MHL with solid burnt-out mesenchymal component. Thick-walled vessels and bile ducts are embedded in dense fibrous stroma (H&E, ×40).

cer. In the operative field, the lesion was located at the inferior edge of the medial segment of the liver, which was closely attached to the hepatic bed of the gallbladder. The lesion was solid and well demarcated with no encapsulation (Fig. 2). Many thick-walled anomalous vessels were seen in the lesion. Although much bleeding occurred during the operation, the mass was entirely removed with gallbladder. The patient is alive and well six months after the surgery.

PATHOLOGY

At gross examination, the lesion consisted of a well-circumscribed, non-encapsulated, nodular and solid mass, measuring 6×4×3.5 cm, which was attached to the hepatic bed of the gallbladder. The mass was well delineated from the attached gallbladder. Cut surface of the mass disclosed yellow-gray, firm and vaguely nodular appearance with multifocal calcifications. Light microscopic examination showed an admixture of bile ducts and thick-walled vessels in dense fibrous stroma with hyalinization (Fig. 3). Proliferation of thick-walled vessels showed dystrophic calcification in their wall, focally mimicking a vascular malformation. Typical zonation with a central hypocellular dense collagenous region and surrounding zone of fibrous tissue containing bile ducts was present. A few cystic spaces surrounded by rims of dense collagen had a flattened endothelial lining that stained positively for factor VIII-related antigen with immunoperoxidase method. The epithelial lining of the proliferative bile ducts was low-cuboidal without atypia (Fig. 4). Neither hepatocytes nor hematopoietic elements were identified within the lesion despite of extensive sampling. Hepatocytes could be found only at the hepatic bed portion of the gallbladder where the lesion was attached.

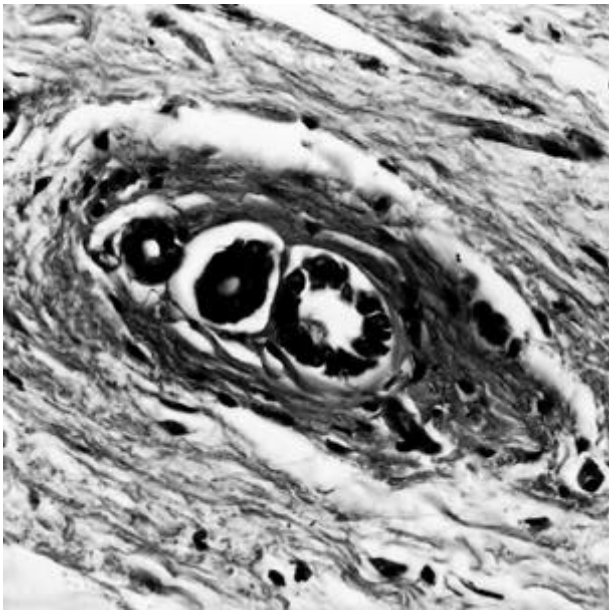

Fig. 4. Small bile ducts are surrounded by rims of dense collagen (H&E, ×200).

DISCUSSION

MHL is a tumor-like lesion, which appears to result from a failure in the normal development of the fetal liver (4). It probably originates from the connective tissue along the portal tracts, and progressive enlargement is the result of cystic degeneration and subsequent fluid accumulation (1). Most of the reported cases described multicystic change of the mesenchymal component with a predilection for infants (1, 2, 4, 5). Only a few cases of MHL in adults have been reported in the literature (3, 6-8). In adult cases of MHL, decrease in non-mesenchymal elements is a major difference in microscopic appearance compared to MHL found in infants (3, 7). Moreover, most of the reported adult cases of MHL were described to have more often cystic change in the mesenchymal area than those of younger patients. Our case shows microscopic appearance that correlates well with previous observations in adult MHL, in that, there is ductal degeneration, dense collagen production, loss of hepatocytes and eccentric thickening of fibrous tissue around vascular spaces (1). However, the peculiar finding of this case is entirely solid and hyalinized mesenchymal component, which is a major difference from other reported cases of MHL in adults (3, 7). In the English literature, only a case of MHL showing macroscopically solid appearance has been reported (8). In that case, the mass consisted of microscopically cystic lymphangiomatous area and fibrous tissue containing bile ducts without mesenchymal hyalinization. The solid burnt-out mesenchymal component seen in the present case is probably indicative of a degenerative process of the lesion. Due to the patients age, history, vague localization with relation to gallbladder, a clinical diagnosis of gallbladder malignancy was considered. However, such a possibility was easily ruled out because there was no mucosal lesion in gallbladder and no atypia in proliferative bile duct epithelia. In our case, the presence of thick-walled vessels warranted differential diagnosis from vascular malformation. However, no arteriolized veins, the presence of bile ducts and collagenous stroma could not explain such a diagnosis.

The correct preoperative diagnosis of MHL is seldom made especially in adults as in our case. Routine laboratory studies and test for liver function are usually normal. Radiological examination will often delineate a mass and may aid in preoperative localization but they seldom lead to a correct diagnosis (4, 9). In our case, the solid nature of the lesion and tight adherence to the hepatic bed of the gallbladder were the obstacles to a correct preoperative diagnosis. In almost all cases, exploratory laparotomy proved to be the definitive diagnostic procedure with subsequent biopsy or resection of the mass. Little is known about the natural history of MHL providing that it be left untreated. With regard to the treatment, complete surgical resection is curative. Although operative mortality may be high due to uncontrollable bleeding, recurrence and malignant transformation have not been reported even when excision is incomplete (1).

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