Ciliated Foregut Cyst of the Gallbladder: Report of a Case

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Introduction

Gallbladder cysts are rare, and most are considered to be acquired in genesis, arising in association with a Rokitansky-Aschoff sinus (RAS). Only a few cases of congenital gallbladder cysts have been described. We present an extremely rare case of a gallbladder cyst that originated from the foregut remnants in an asymptomatic 37-year-old woman. To our knowledge, only three similar cases have been reported: one in Japan1 and the other two in Western countries.2,3 We review the clinico-pathological aspects of this unusual entity and discuss the usefulness of radiological imaging by computed tomographic arteriography (CT-A).

Case Report

A 37-year-old woman was referred to our hospital after screening ultrasonography (US) revealed a tumor in the gallbladder. She had not experienced any abdominal symptoms and laboratory data were normal. Drip infusion cholangiography revealed a filling defect with a smooth margin, fixed to the right wall of the well-opacified gallbladder. An endoscopic retrograde cholangiography was unsuccessful. Abdominal US showed a well-demarcated round cystic mass, 1.3 × 1.3 cm in size, in the body of the gallbladder, which contained high-echoic foci. The cyst wall was thin and smooth, and protruded into the lumen of the gallbladder. There were no moving strong echoes or acoustic shadows, indicating stones or calcifications, or comet-like echoes suggestive of RAS (Fig. 1). Simple abdominal CT showed a mass with slightly enhanced density, about 1.2 cm in diameter, in the body of the gallbladder. Dynamic CT with a bolus injection of contrast medium showed that the lesion was slightly enhanced, with a CT value ranging from 49 to 58 Hounsfield unit (HU) (Fig. 2). Selective celiac angiography failed to demonstrate

Abstract

We report the rare case of a gallbladder cyst arising from the foregut remnants. A 36-year-old woman was referred to our hospital after screening ultrasonography (US) detected a tumor in the gallbladder. On admission, she was well and her blood analyses were all normal. US showed a cystic mass with internal high-echoic lesions, and computed tomography (CT) demonstrated a protruding tumor with slight enhancement in the gallbladder. Angiography provided no additional information; however, sequential CT-arteriography (CTA) clearly demonstrated that this tumor was a cystic lesion. Surgical exploration was performed, first because of the difficulty in establishing a definite diagnosis, and also because the patient wanted the tumor removed. The resected specimen contained a unilocular cystic tumor that looked like a submucosal tumor. Histologically, the wall of the cyst was lined by ciliated stratified columnar epithelium with interspersed goblet cells and underlying smooth muscle fibers. The mass was finally diagnosed as a congenital ciliated foregut cyst of the gallbladder. Cysts of the gallbladder are uncommon and the majority are acquired. To our knowledge, this represents only the fourth report of a ciliated foregut cyst of the gallbladder in the literature. Although rare, an awareness of this entity could allow a preoperative diagnosis to be made, whereby surgical exploration may be avoided. CT-A is a very useful diagnostic tool, especially when the nature of the tumor presents a difficult differential diagnosis.

Key words Ciliated foregut cyst · Gallbladder cyst

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pathological findings in any phase. Sequential CT-A was performed via a catheter in the common hepatic artery, which clearly demonstrated that the wall of the mass was enhanced similarly to the gallbladder wall, but no enhancement was seen in the internal lesions of the mass (Fig. 3). These results indicated that the tumor was cystic rather than solid, based on which we suspected a cystic lesion, such as duplication of the gallbladder. Because it was so difficult to establish a definite diagnosis, and since the patient wanted the tumor removed, we decided to perform an exploratory laparotomy with cholecystectomy.

At laparotomy, a firm mass was felt in the gallbladder wall, which looked otherwise normal. No other lesions were found in the abdominal cavity, and a simple cholecystectomy was carried out.

Macroscopically, a unilocular cyst, $2.4 \times 1.6 \times 0.8$ cm in size, was found in the body of the gallbladder. This mass did not appear to communicate with the lumen of the gallbladder and looked like a submucosal tumor (Fig. 4). No stones were found in the gallbladder, the surface of which was smooth and even. The cut surface of the tumor revealed a cystic mass containing mucin with no tumorous solid lesion. Regrettably, a sample of mucin was not taken for analysis.

Microscopically, the solitary cyst was separated from the muscular layer and no communication with the gallbladder lumen was found. The cyst was lined by ciliated pseudostratified columnar epithelium with interspersed mucin-producing columnar epithelium, and the outside was circumscribed by a leiomyomuscular layer (Fig. 5). No Rokitanski-Ashoff sinus (RAS) or adenomyomatosis was detected in the gallbladder. Immunohistochemical