A hitherto undescribed benign mesenchymal polyp of the gallbladder: edematous angiomyolipoma-like polyp

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Abstract

We report a case of two peculiar gallbladder polyps in a sixty-four year old male who presented with symptomatic cholelithiasis. Cholecystectomy was performed, which revealed two polyps measuring 0.6 cm and 1.9 cm, located in the body of the gallbladder. Microscopic examination of the polyps showed composite mesenchymal lesions with vascular proliferation of small-to-medium sized arterioles, myoid stroma, and lipomatous periphery. The myoid component was characterized by wisps of bland smooth muscle fibers loosely separated by proteinaceous and focally myxoid matrix. The surface of the polyps was lined by a single layer of bland epithelial cells. The unique histomorphologic features differentiate the lesions from other known mesenchymal polyps of the gallbladder. We propose the name "edematous angiomyolipoma-like polyp" for these rare lesions given their histomorphologic similarity to angiomyolipoma. (Acta gastroenterol. belg., **2016**, 79, **371-374**).

Key words : gallbladder, angiomyolipoma, polyp

Introduction

The increased use of abdominal ultrasound has led to a concomitant rise in the detection of polypoid lesions of the gallbladder. The prevalence of gallbladder polyps identified by ultrasound in Western countries ranges from 1.4-6.7% (1,2,3). Although the majority of the polypoid lesions are benign, imaging studies alone cannot reliably predict the presence of malignancy or premalignant lesions. Currently size is the best predictor of malignancy; thus, cholecystectomy is recommended for gallbladder polyps greater than 1 cm in size (4). Pathologists must not only be familiar with the common benign and malignant gallbladder polypoid lesions, but should also be cognizant of rare entities to minimize diagnostic errors. Here we report a case of two mesenchymal polyps of the gallbladder that has not been described in the literature to the best of our knowledge.

Clinical history

The patient was a sixty-four year old male who initially presented to his primary care physician with intermittent, non-radiating right upper quadrant abdominal pain without nausea or vomiting. Abdominal ultrasound demonstrated two polyps in the gallbladder, the largest of which measured 1.5 x 1.2 cm with internal vascularity, and the smaller polyp measuring 0.7 x 0.6 cm (Figures 1A and 1B). Multiple benign-appearing simple cysts were noted throughout the liver, the largest measuring 1.6 cm in diameter. No intra- or extrahepatic biliary dilation was seen. No mass or cystic lesions were documented in the kidneys and pancreas. Upon review, these lesions were first incidentally seen on CT scan in 2012, measuring roughly 1 cm. His past medical history was significant for non-invasive high grade papillary urothelial carcinoma status post transurethral resection 14 years ago, tubular adenoma of the descending colon resected in 2008, cerebral palsy, hepatitis A, left breast gynecomastia, osteoporosis, arthritis, and gastroesophageal reflux disease. The patient did not have a history of tuberous sclerosis. The patient had no known drug allergies. Family history and social history were non-contributory. His medications included baclofen, clonazepam, ibandronate, pantoprozole, tamsulosin, vitamin D, and vitamin E.

The patient was referred to general surgery clinic, where he was recommended to undergo elective laparoscopic cholecystectomy due to the size of the polyp. Lipid panel and preoperative liver function tests were all within normal limits. The cholecystectomy was performed without any complications and the recovery course was unremarkable. The patient had been doing well for more than one year after the surgery.

Pathologic Findings

The resected gallbladder measured 10.0 x 4.5 x 2.5 cm. Opening showed two yellow gelatinous polyps in the body measuring 0.6 x 0.4 x 0.2 cm and 1.9 x 1.5 x 0.5 cm. The gallbladder wall measured from 0.1 to 0.2 cm in thickness. No gallstones or other lesions were identified.

Histologically, both polyps of the gallbladder displayed similar histologic features. They were mucosa-based and lined by a single layer of bland epithelial cells. The periphery of the polyps appeared lipomatous. There were frequent small-to-medium sized arterioles scattered in an edematous and hypocellular matrix (Figures 2A and 2B).

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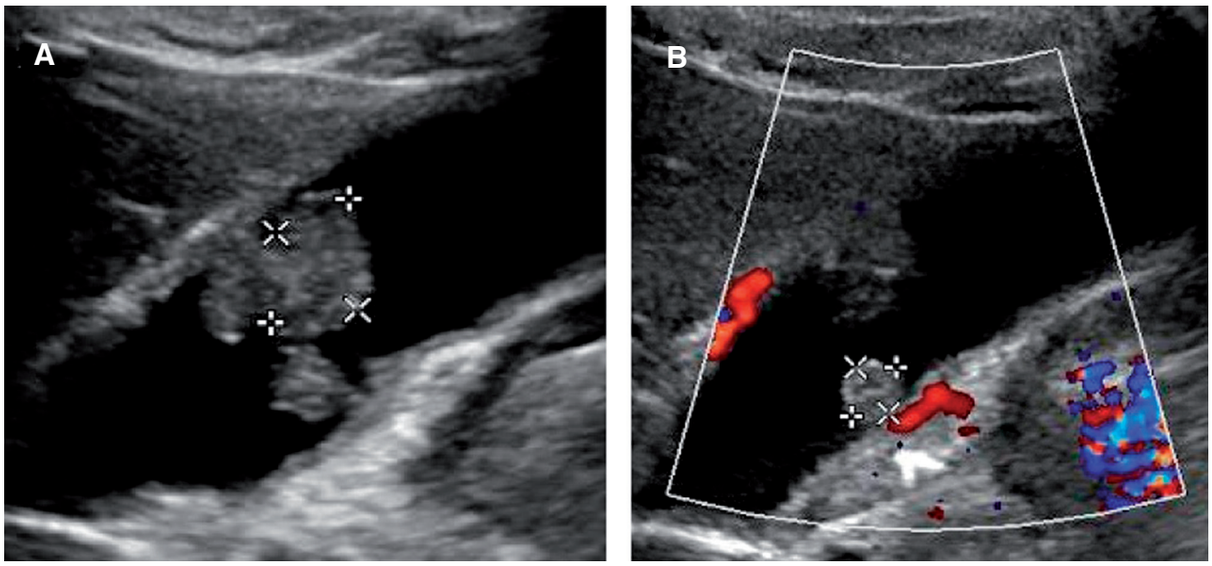

Fig. 1. — Abdominal right upper quadrant ultrasound showing
(A) greyscale image of a hyperechoic lesion without acoustic shadowing within gallbladder lumen, suggestive of gallbladder polyp (inside crosshairs) measuring 1.5 x 1.2 cm, and (B) color Doppler image of a separate gallbladder polyp, measuring 0.7 x 0.6 cm. Images obtained with curved transducer.

Wisps and strands of bland smooth muscle fibers were present, which appeared to spin off the arterioles and radiate towards the periphery (Figure 2C). The muscle fibers were loosely separated by proteinaceous and focally myxoid fluid. Mild bland spindle cell proliferation and rare inflammatory cells were also noted (Figure 2D). No significant nuclear atypia or mitosis was appreciated in spindle cells. No necrosis was seen. No epithelioid cells were identified. Immunohistochemical stain for S100 demonstrated scattered positive cells at the periphery of the polyps, which appeared to represent adipocytes (data not shown). No HMB-45 or MART-1 positive cells were demonstrated by immunohistochemistry (data not shown).

Discussion

Mesenchymal tumors constitute approximately 1% of all biliary tract neoplasms, which include a variety of benign and malignant neoplasms (5). The most common non-neoplastic polypoid lesions of the gallbladder are cholesterol polyps, followed by adenomyomas and inflammatory polyps (6,7), which may mimic cancer clinically and/or radiographically (5). One study found no association of the gallbladder polyps with patient's age, sex, body weight, number of pregnancies, or use of exogenous female hormones (1). Cholesterol polyp (cholesterolosis) has a prevalence of 9-26% (8), which has no sex predilection and is not associated with any risk factors for gallstone formation (1). It seldom reaches a size of greater than 1 cm, and is characterized histologically by abundant lipid-laden macrophages within the lamina propria with a polypoid configuration (5). Adenomyoma (adenomyomatous hyperplasia or adenomyomatosis) is a form of diverticular disease, which is more common in women. It has three morphological types: localized (fundal), segmental, and diffuse, with the localized type being the most common. The association of segmental type with malignancy has been controversial (9, 10). Histologically, adenomyoma is characterized by smooth muscle proliferation associated with invaginated glandular elements (11,12). Fibroinflammatory polyp is typically composed of granulation tissue, edematous stroma, and admixed inflammatory cells in variable amounts (3). However, the stroma can be variable in appearance, ranging from a striking fibrotic appearance (thus termed "fibrous polyp") to markedly edematous (5). Inflammatory polyp is almost always associated with cholecystitis (7).

The gallbladder polyps in the current case were composed of vascular, myoid and lipomatous components in an edematous stroma. The possibility of angiomyolipoma was thus considered. However, immunostains for HMB-45 and MART-1 were both negative, providing no support for this diagnosis. Angiomyolipoma belongs to the perivascular epithelioid cell tumor (PEComa) family, which includes a group of related mesenchymal neoplasms composed of perivascular cells characteristically showing smooth muscle and melanocytic differentiation (13). Only a couple of gallbladder PEComas have been reported, with both cases showing nests of uniform epithelioid cells with round to ovoid nuclei, small nucleoli, and clear to granular lightly eosinophilic cytoplasm; histomorphologic features that are lacking in our case (13,14). Some tumors in the PEComa family, such as

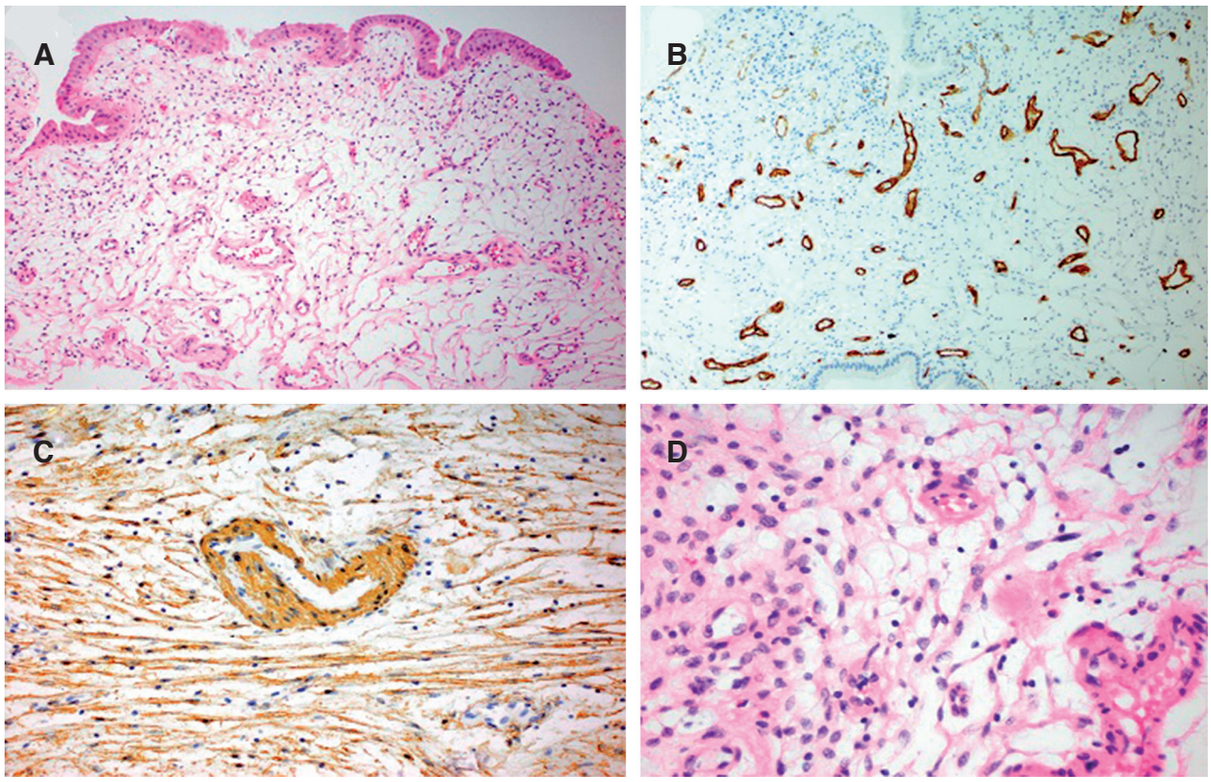

Fig. 2. — The mucosa-based polyp demonstrates edematous stroma with scattered small and medium sized arterioles and peripherally based lipomatous features. The overlying epithelium consists of a single layer of bland epithelial cells (A; hemotoxylin-eosin stain, original magnification 100x). Immunohistochemical staining for CD34 highlights the arterioles (B; original magnification 100x). Immunohistochemical staining for smooth muscle actin reveals wisps of muscle fibers radiating from the arterioles to the periphery (C; original magnification 200x). Mild bland spindle cell proliferation and rare inflammatory cells were noted (D; hemotoxylin-eosin stain; original magnification 400x).

renal angiomyolipoma, are known to be associated with tuberous sclerosis complex, which is an autosomal dominant disorder caused by mutations in the *TSC1* or *TSC2* genes (14). Our patient does not have a history of tuberous sclerosis.

To our knowledge, there has been a single case report in Japanese literature describing a benign fibroepithelial polyp of the gallbladder (15). Although this reported polyp has some similar histologic features to our case, such as edematous stroma, our case lacks a prominent glandular component, but contains prominent vascular and myoid components instead.

In summary, we report a peculiar case of two gallbladder polyps characterized by a composition of mesenchymal elements which include a peripherally oriented lipomatous region and central proliferation of small-to-medium sized arterioles with myoid fibers in an edematous matrix. Although they appear to consist of the similar components as an angiomyolipoma histologically, negative HMB-45 and MART-1 immunoreactivity does not support the diagnosis. We feel that the name "edematous angiomyolipoma-like polyp" best describes this rare but particular entity. The histologic features of the lesion appear benign, which is supported by a benign clinical course during postoperative follow up for >12 months.

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