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A case of traumatic neuroma of the gallbladder in the absence of previous surgery and cholelithiasis.

Junji Matsuoka* Noriaki Tanaka† Kazushi Kojima‡

Kenichi Takai** Kazuo Hamaya†† Akira Gochi‡‡

Yasuaki Kamikawa§ Kunzo Orita¶

*Okayama Central Hospital,

†Okayama University,

‡Okayama Univeristy,

**Okayama University,

††Okayama Saiseikai General Hospital,

‡‡Okayama University,

§Okayama University,

¶Okayama University,

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Junji Matsuoka, Noriaki Tanaka, Kazushi Kojima, Kenichi Takai, Kazuo Hamaya, Akira Gochi, Yasuaki Kamikawa, and Kunzo Orita

Abstract

We experienced a patient with traumatic neuroma of the gallbladder with no history of gallbladder surgery or cholelithiasis. A 74-year-old man was referred to our department after a gallbladder tumor was incidentally discovered during a preoperative screening examination for prostate hypertrophy. Ultrasonography, MRI, CT and endoscopic retrograde cholangiography revealed a protuberant lesion of the gallbladder. Laparoscopic cholecystectomy was attempted but adhesion between the liver and duodenum forced us to convert to open laparotomy. Cholecystectomy and adjacent liver tissue resection was performed. Diagnosis was made by frozen histology during operation. It revealed no malignancy. Postoperative pathological examination revealed traumatic neuroma associated with inflammation. To our knowledge, this is the first reported case of gallbladder neuroma without a history of gallstones or surgery in the English and Japanese literature since 1980. This traumatic neuroma should be considered in a differential diagnosis in treating gallbladder neoplasm, even in the absence of an operative history or cholelithiasis.

KEYWORDS: traumatic neuroma, gallbladder

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Brief Note

A Case of Traumatic Neuroma of the Gallbladder in the Absence of Previous Surgery and Cholelithiasis

Junji MATSUOKA*, Noriaki TANAKA[a], Kazushi KOJIMA, Kenichi TAKAI, Kazuo HAMAYA[b], Akira GOCHI[a], Yasuaki KAMIKAWA[a] and Kunzo ORITA[a]

Okayama Central Hospital, [a]First Department of Surgery, Okayama University Medical School and [b]Okayama Saiseikai General Hospital Okayama 700, Japan

We experienced a patient with traumatic neuroma of the gallbladder with no history of gallbladder surgery or cholelithiasis. A 74-year-old man was referred to our department after a gallbladder tumor was incidentally discovered during a preoperative screening examination for prostate hypertrophy. Ultrasonography, MRI, CT and endoscopic retrograde cholangiography revealed a protuberant lesion of the gallbladder. Laparoscopic cholecystectomy was attempted but adhesion between the liver and duodenum forced us to convert to open laparotomy. Cholecystectomy and adjacent liver tissue resection was performed. Diagnosis was made by frozen histology during operation. It revealed no malignancy. Postoperative pathological examination revealed traumatic neuroma associated with inflammation. To our knowledge, this is the first reported case of gallbladder neuroma without a history of gallstones or surgery in the English and Japanese literature since 1980. This traumatic neuroma should be considered in a differential diagnosis in treating gallbladder neoplasm, even in the absence of an operative history or cholelithiasis.

Key words: traumatic neuroma, gallbladder

A traumatic neuroma arising from the biliary tract is rare and most patients have a history of surgery and the tumor arises in the stump of the cystic duct or in the choledochus where operations were performed. Traumatic neuroma is postulated to be one of the causes of postcholecystectomy syndrome with such clinical manifestations as jaundice, fever and abdominal pain. Differential diagnosis from bile tract malignancies is also needed to obviate radical surgery, but it is often difficult to diagnose preoperatively especially if there is no evidence of previous surgery. This paper describes a patient with gallbladder neuroma who was treated surgically under suspicion of gallbladder carcinoma because of the local findings and the lack of previous surgery or bile duct disorders. Frozen histological examination during operation revealed no malignant findings and cholecystectomy with adjacent liver dissection was performed. The cause of neuroma of the gallbladder has been speculated so far to be caused by chronic inflammation together with cholelithiasis. However, no gallstones were found in this case. Traumatic neuroma should be considered in a differential diagnosis even in patients without previous surgery or cholelithiasis.

A 74-year-old man was admitted to the department of urology, Okayama Central Hospital on November 15, 1995, hoping for treatment on his prostatic hypertrophy. He had never undergone surgery and never suffered from abdominal pain nor liver disorder. He underwent transurethral resection (TUR) of the prostate on November 22, 1995. Pathological examination revealed nodular hyperplasia of the prostate without evidence of malignancy. Preoperative systemic screening before TUR was performed and a neoplastic lesion of the gallbladder was revealed. Ultrasonography showed an atrophic gallbladder and a hyperechoic lesion in the middle portion of the gallbladder. MRI showed a protuberant well-margined mass in the base of the gallbladder (Fig. 1a). CT examina-

* To whom correspondence should be addressed.

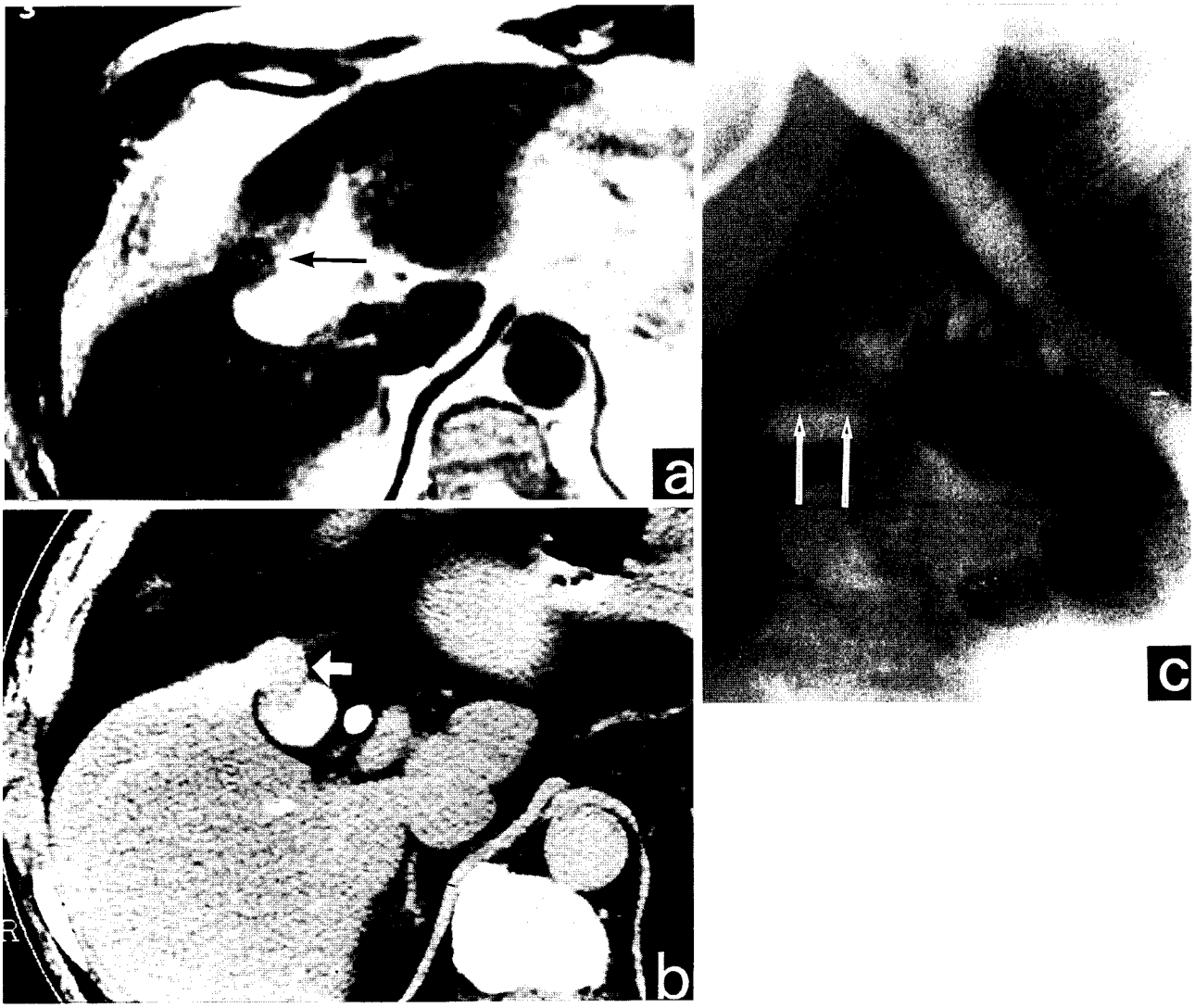

Fig. 1 Preoperative digonostic imagings.
a: T2-weighted MRI examination revealed a mass lesion (arrow) which is not enhanced by gadolinium chloride injection; **b**: CT examination showed a well-marked tumorous lesion in the fundus of the gallbladder (arrow). The border of the tumor was clear but the tumor seemed to invade into the liver; **c**: Endoscopic retrograde cholangiography revealed a mass inside the gallbladder (arrows) without cholelithiasis. The margin and the surface of the tumor was clear and smooth.

tion also revealed the existence of the mass (Fig. 1b) which is not enhanced by the contrast medium. The border of the tumor margin was clear but invasion of the tumor to the liver was suspected. The colon and the duodenum were suspected to adhere the gallbladder. Endoscopic retrograde cholangiography (ERC) showed a mass inside the gallbladder (Fig. 1c). The cystic duct was patent and the passage of the contrast medium into the gallbladder was smooth. The margin of the mass inside the gallbladder was well marked and no invasive tendency was suggested. The intrahepatic and common bile ducts were clear. Laboratory data on admission showed no remarkable change including tumor markers: Ca 19-9 was 36 u/ml (normal value, less than 37 u/ml) and CEA was 0.8 ng/ml (normal value, less than 2.5 ng/ml). On December 12, 1995, the patient underwent cholecystectomy under suspicion of gallbladder adenomyomatosis.

Initially, laparoscopic cholecystectomy was attempted

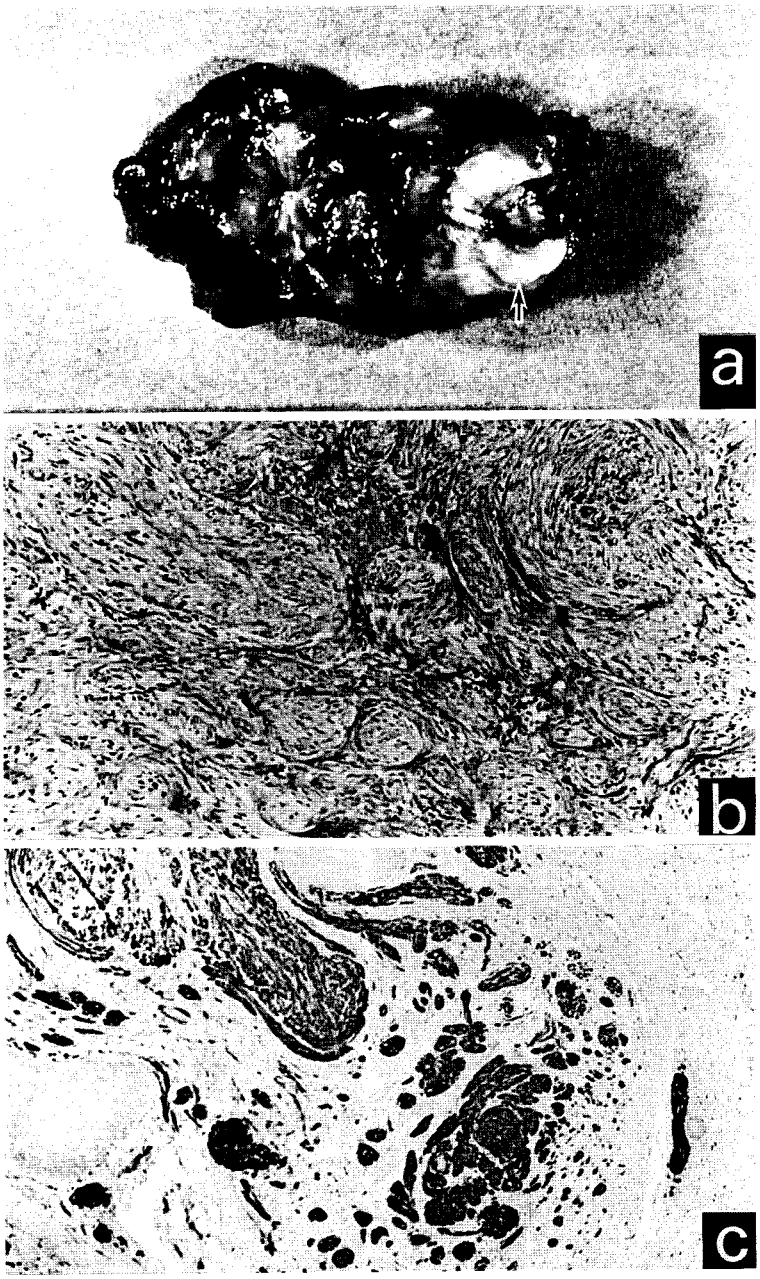

Fig. 2 Postoperative pathological findings.
a: Resected cholecyst showed a gray-white tumor (arrow) in the fundus without cholelithiasis; **b**: Pathologic examination revealed proliferated fascicles (HE staining, × 10); **c**: Positive immunohistochemical staining for S-100 proved the tumor to be of neural origin (× 10).

in this patient. However, duodenal adhesion to the cholecyst and the liver was too severe to complete the procedure, and cholecystectomy by converted open laparotomy was performed. The gallbladder was surrounded by the duodenum and the adhesion of the duodenum to the liver was severe. The fundus of the gallbladder was markedly fibrotic and a hard tumor was palpated. The margin was unclear and the growth of the tumor seemed

to be invasive. Gallbladder carcinoma was highly suspected. Cholecystectomy and resection of the liver around the fundus of the gallbladder was performed. Resection of the adjacent liver tissue was performed in order to have a clear margin of the hard mass. There were no stones in the gallbladder. The area of the Hartman's pouch showed no evidence of inflammation and the ligation and cut of the cystic duct was easy. During the operation, histological examination using frozen specimens was performed and no evidence of malignancy was revealed. Therefore, no further operation was performed.

The resected cholecyst was atrophic in appearence and a gray-white elevated lesion in the fundus was seen (Fig. 2a). Postoperative pathological examination revealed poorly circumscribed proliferation of a tangled small fascicles in a dense fibrous matrix and peri-fascicular perineum developed well in the submucosa to the submucosal area (Fig. 2b). They tended to proliferate around dilated Aschoff-Rokitansky sinus. The covering mucosa was extensively eroded. Immunopathological study revealed that these fascicles were strongly positive for S100 protein (Fig. 2c) and vimentin (not shown), but not for alpha-smooth muscle antigen and desmin (not shown).

The postoperative course was not eventful and he was discharged on December 28, 1995. He has been doing well since the discharge.

The development of amputation neuroma after biliary tract surgery is well recognized, and these tumors have been suggested to be the cause of the symptom that occasionally follow cholecystectomy. Such an association, however, is debatable (1, 2). Injury to nerves commonly results in the development of neuromas. Traumatic neuroma is caused by the overgrowth of the proliferation of axis cylinders, nerve sheaths, and fibroblasts (2). The region of the cystic duct has a rich supply of nerves from both sympathetic and parasympathetic trunks, which ramify around it as the choledochal plexus. The common lesion of the traumatic neuroma is therefore located in the cystic duct and choledochus (1, 2). In our case, the neuroma was found in the fundus of the gallbladder. Traumatic neuroma of the gallbladder is very rare. Three patients with traumatic neuroma of the gallbladder have been reported so far (4–6): They had previous operative history on cholecyst (4), selective vagotomy (5) and no history of operation but choledocholithiasis (6). Traumatic neuroma is not necessarily caused by trauma or surgery; instead, it may be caused by chronic inflammation as reported by Daneshvar (7). Histology revealed the existence of the chronic persistent inflammation in all three cases. In this case, histological examination also revealed the existence of chronic inflammation and eroded mucosa. Since there was extensive and deep proliferation of Aschoff-Rokitansky sinus in the wall of the gallbladder and the nerve fascicles tended to conglomerate around the sinus, it is possible that the peripheral nerves in these area are damaged by the preexistent destructive inflammation involving the sinus. Superficial mucosal erosion can not damage the nerve fiber to produce amputated neuroma, because the nerves near the mucosal surface are too thin to produce massive abnormal regeneration of the nerve. In this case, although the patient himself did not notice, he seemed to have experienced severe inflammation of the gallbladder. Proliferation of the neural tissue was observed around the Ashoff Rokitansky sinus, suggesting that inflammation was present at the deep layer of the gallbladder wall where thick nerve was present. It was clearly indicated that not only operative trauma but also inflammation cause so-called traumatic neuroma.

The clinical manifestation of the traumatic neuroma is symptom associated with obstruction by tumor. In case of the traumatic neuroma of the gallbladder, the symptom is often caused by the existing gallstones. In this case, no symptoms were clear because there were no stones in the gallbladder. However, the erosion of the mucosa suggests the previous existence of inflammation in the gallbladder. The adhesion of the duodenum to the gallbladder may contribute to the inflammation but it is not clear.

A preoperative diagnosis was not made in any of the three reported cases. In our case, adenomyomatosis was initially suspected. Unnecessary surgery should be avoided in benign disease such as traumatic neuroma or adenomyomatosis. In our case, operative local findings were similar to those associated with gallbladder cancer. Tumor was very hard, adhesion was severe, the tumor seemed to be invasive and dissection from the liver bed was very hard. Intraoperative histological examination seemed to be the only way to avoid oversurgery.

Amputation neuroma of the gallbladder, although rare, should be considered in a differential diagnosis in patients who are suspected of having gallbladder cancer. The recognition of this entity may obviate unnecessary radical surgery. Histological examination during surgery seems to be the best way to diagnose this clinical entity.

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