SCHWANNOMA OF THE GALLBLADDER : REPORT OF A CASE

RYO OHTA1), YUDAI HIRATA1), MASATAKA ONEYAMA1), YASUMASA TAKAHASHI1), YUICHI KAWAHARA1), MASAYA KITAMURA1), MANABU GOTO1), KOJI SEKIKAWA1) and SEIICHI TAKENOSHITA2)

1)Department of Surgery, Institute of Gastroenterology, Kawasaki Saiwai Hospital, Kawasaki, Japan, 2)Second Department of Surgery, Fukushima Medical University School of Medicine, Fukushima, Japan

(Received November 20, 2009, accepted March 1, 2010)

Abstract : We report a schwannoma of the gallbladder in a 58-year-old man who was diagnosed as cholecystolithiasis. He presented with recurrent episodes of abdominal pain in the right upper quadrant. The abdominal computed tomography scan and ultrasonography revealed stones about 15 mm in diameter in the gallbladder. Under the diagnosis of cholecystolithiasis, laparoscopic cholecystectomy was performed. The resected specimen showed chronic cholecystitis and no suspicion of neoplasm. Pathological examination revealed that the tumor consist of spindle cells without atypical appearance at small area of fundus. Immunohistologically, tumor cells were positive for S-100 protein and negative for alpha-SMA and c-kit, the lesion was diagnosed as schwannoma.

Key words : Schwannoma, Gallbladder, Immunohistochemistry

INTRDUCTION

Schwannomas are uncommon neoplasms. They typically arise from neural crest cells which encapsulate the nerve sheath and are sometimes also referred as neurilemmomas. Schwannomas usually occur in the extremities, but can also be found in the trunk, head and neck, retroperitoneum and mediastinum1). It is rarely found in the intraabdomen, although cases involving the small bowel, pancreas, pelvis and rectum have previously been reported2). They are usually asymptomatic and hence discovered incidentally. We report such an extremely rare case of schwannoma of the gallbladder, which was treated by laparoscopic cholecystectomy under the diagnosis of cholecystolithiasis.

太田 竜, 平田雄大, 小根山正貴, 高橋保正, 河原祐一, 北村雅也, 後藤 学, 関川浩司

Corresponding author : Ryo Ohta  E-mail : r-ohta@saiwaihp.org
http://fmu.ac.jp/home/lib/F igaku/  http://www.sasappa.co.jp/online/
A 58-year-old man, presented with recurrent episodes of right hypochondralgia for many years, admitted to our department under the diagnosis of cholecystolithiasis. Physical
examination revealed nothing special except a minimal degree of localized tenderness in the right upper quadrant. He had no history of laparotomy. Blood laboratory data showed normal levels of liver function and slight inflammation. Computed tomography scan showed high-density area and ultrasonography showed high echoic lesions in the gallbladder (Fig. 1 A, B). Because of filled stones in the gallbladder, adequate evaluation in presence of neoplasm was not confirmed.

We made a preoperative diagnosis of cholecystolithiasis and performed laparoscopic cholecystectomy. The gallbladder was surrounded by omentum and appeared to be thickened wall. There was no obvious tumor lesion at the gallbladder (Fig. 2). A specimen showed that the gallbladder had chronic inflammatory change and several combination stones with debris. The fundus of gallbladder revealed marked wall thickness (Fig. 3). Macroscopically, tumor lesion was not detected. Histological examination revealed the well-demarcated tumor 3 mm in diameter mainly composed of spindle-shaped cells without mitotic figures at the fundus of
SCHWANNOMA OF THE GALLBLADDER

There were no atypical cells and no signs of malignancy. Since immunohistochemical study showed that the tumor cells were positive for S-100 protein, and negative for alpha-smooth muscle actin and c-kit (Fig. 4 B, C, D), this tumor was diagnosed as schwannoma of the gallbladder. The postoperative course was uneventful and no additional therapy was performed. Sixteen months after surgery the patient has been doing well without evidence of recurrent disease.

DISCUSSION

Schwannomas are benign neurogenic tumors that arise in the nerve sheaths of the peripheral nerves. Malignant transformation is extremely rare and malignant schwannomas are commonly associated with von Recklinghausen's disease have been reported\(^3\). Histologically benign schwannomas reveal such as absence of mitosis and preservation of spindle shape with large cohesive aggregates of cells distinct from malignant type. They can occur mainly in the head, the neck, and the flexure surfaces of the upper and lower extremities. On other side, the frequency of schwannoma arising in the digestive tract is relatively low and such as biliary tract is extremely rare. To our knowledge, only three similar cases of schwannoma of the gallbladder have been reported\(^4-6\) (Table 1). Clinically, intra-abdominal schwannoma is asymptomatic for a long time, or it is accompanied by nonspecific abdominal pain and discomfort. In the late clinical course, compression of the surrounding organs might be noticed. In the reported cases of schwannoma of the gallbladder, one case revealed obstructive jaundice and the other cases were asymptomatic.

Preoperative diagnosis of intra-abdominal schwannomas appears to be very difficult because these tumors are commonly asymptomatic and are often discovered incidentally. For preoperative diagnosis, computed tomography (CT), ultrasonography, magnetic resonance image (MRI), and angiography are useful for locating the tumor site, but can not be confirmed the definitive diagnosis. CT findings usually show well-defined, round masses with multiple, low-attenuation, and cystic necrotic areas\(^7\). On MRI, schwannomas are present as masses of low signal intensity on T1-weighted images and of high signal intensity on T2-weighted images\(^8\). In our case, the tumor is too small to detect radiological findings and filled stones in the gallbladder prevent adequate evaluation. In the reported cases, small size tumor was also similarly not detected by preoperative examinations\(^3\). The endoscopic findings are almost nonspecific as these tumors appear grossly as submucosal lesions, which are distin-

<table>
<thead>
<tr>
<th>First Author(^3)</th>
<th>Year</th>
<th>Age</th>
<th>Sex</th>
<th>Symptoms</th>
<th>Preoperative diagnosis</th>
<th>Treatment</th>
<th>Location</th>
<th>Size (mm)</th>
<th>S-100</th>
<th>SMA</th>
<th>c-KIT</th>
<th>Antoni type</th>
</tr>
</thead>
<tbody>
<tr>
<td>Yamagiwa(^3)</td>
<td>1991</td>
<td>58</td>
<td>M</td>
<td>Jaundice</td>
<td>Bile duct cancer</td>
<td>Cholecystectomy</td>
<td>Neck</td>
<td>3 × 4</td>
<td>+</td>
<td>NA</td>
<td>NA</td>
<td>A</td>
</tr>
<tr>
<td>Matsuoka(^4)</td>
<td>1996</td>
<td>74</td>
<td>M</td>
<td>Asymptomatic</td>
<td>Adenomyomatosis</td>
<td>Cholecystectomy + hepatectomy</td>
<td>Fundus</td>
<td>10</td>
<td>+</td>
<td>−</td>
<td>NA</td>
<td>A</td>
</tr>
<tr>
<td>Colovic R(^5)</td>
<td>2003</td>
<td>61</td>
<td>F</td>
<td>NA</td>
<td>Tumor of gallbladder</td>
<td>Cholecystectomy</td>
<td>Whole</td>
<td>90 × 65</td>
<td>NA</td>
<td>NA</td>
<td>NA</td>
<td>NA</td>
</tr>
<tr>
<td>Present case</td>
<td>2008</td>
<td>58</td>
<td>M</td>
<td>Asymptomatic</td>
<td>Cholecystolithiasis</td>
<td>Cholecystectomy</td>
<td>Fundus</td>
<td>3</td>
<td>+</td>
<td>−</td>
<td>−</td>
<td>A</td>
</tr>
</tbody>
</table>

NA, no data available; SMA, smooth muscle actin
guishable from other mesenchymal tumor. Though the tumor arising from the alimentary tract can obtain tumor tissue by endoscopic biopsy easily, it is too difficult to obtain biopsy specimen from the gallbladder for the definite diagnosis. Therefore preoperative diagnosis of schwannoma in the gallbladder is limited.

Immunohistological staining is useful for definite diagnosis from stromal and myogenic tumor. Positive staining for S-100 protein and vimentin and negative for alpha-smooth muscle actin, c-KIT and CD34 support the notion that the tumor originated from the schwann cell\(^9\). Schwannomas originating in the digestive tract show distinct histological features that distinguish them from conventional schwannomas, which arise from the central nerves system and soft tissues. Histologically, schwannomas demonstrate a biphasic pattern with areas of Antoni A and B\(^10\). These schwannomas are S-100 protein-positive spindle cell tumors that are composed mainly of Antoni A areas, and generally do not show a nuclear palisading pattern that is usually found in conventional schwannomas\(^11\). In the present case, the tumor showed dominant Antoni A areas and positive staining for S-100 protein, but had nuclear palisading pattern. More cases are needed to determine the pathological correlation between the schwannoma of the gallbladder and other digestive schwannomas.

Schwannomas of the digestive tract have excellent prognosis similar to conventional schwannomas\(^12\). The treatment of choice is complete surgical excision because of diagnostic uncertainty, and the longterm outcome is excellent as these lesions are uniformly benign. However regarding to the malignant potential of schwannomas, it is still controversial fields. At present, due to the uncertainty of preoperative diagnosis, the increasing size of the tumor, and the possibility of malignancy, early surgical excision is considered to be the most acceptable strategy.

REFERENCES

9. Weiss SW, Langloss JM, Enzinger FM. Value of S 100 protein in the diagnosis of soft tissue tumors with particular reference to benign and malignant Schwann cell tumors. Lab Invest,