Isolated Hepatic Actinomycosis Mimicking Cholangiocarcinoma: a case report and literature review

FU-CHIEH HSU¹  Tzu-Cheng SU²  ALBERT D. YANG¹  CHUNG-YING LIAO¹  KWO-WHEI LEE¹

Department of Radiology¹, Department of Pathology², Changhua Christian Hospital, Changhua, Taiwan

ABSTRACT

Isolated hepatic actinomycosis (IHA) is a rare infectious disease, which presents as a space-occupying lesion. We present a case of pathologically proven liver actinomycosis. Ultrasonography and computed tomography scan revealed a large solitary liver mass. The mild heterogeneously enhancing mass appeared as a hypointense area on a T1-weighted image and as a hyperintense area on a T2-weighted image, with a peripherally enhancing rim, and mimicked the appearance of a peripheral cholangiocarcinoma (PCC) on magnetic resonance imaging (MRI). However, the mean apparent diffusion coefficient (ADC) value ($0.78 \times 10^{-3} \text{ s/mm}^2$) of the mass obtained from diffusion-weighted MRI (DW-MRI) was significantly lower than that of PCCs. IHA is characterized by extension of the mass to the abdominal wall and the presence of numerous honeycomb-like septa within the mass. IHA generally responds well to antibiotic therapy but is easily misdiagnosed. Knowledge of the radiological features of IHA is essential for avoiding delayed treatment and unnecessary surgery.

CASE REPORT

A 55-year-old woman was referred to our department from a local clinic, where an initial diagnosis of a liver tumor had been suggested on the basis of ultrasonography (US) findings. She had a low-grade fever (body temperature, 37.8°C) at the time of presentation. Her clinical history was unremarkable, except for a total abdominal hysterec-

Actinomycosis is a chronic suppurative infection, characterized by abscess formation, draining sinus tracts, granulated tissues, and fibrosis [1]. Liver involvement is rare and has been reported in only 3% of all actinomycosis cases [2]. The clinical and radiological features of actinomycosis usually overlap with those of inflammatory pseudotumors (IPTs) and neoplasms [3, 4]. Diffusion-weighted magnetic resonance imaging (DW-MRI) is an imaging technique that provides tissue contrast by measuring the diffusion properties of water molecules within the tissue. The results of recent studies have suggested that the ADC value is useful in characterizing focal liver lesions [5, 6, 7]. To date, no studies have reported the use of DW-MRI for diagnosing actinomycosis. We found that diffusion-weighted imaging (DWI), when used with ADC values, may be a useful technique for distinguishing hepatic actinomycosis from malignant tumors.

Correspondence Author to: Kwo-Whei Lee
Department of Radiology, Changhua Christian Hospital, Changhua, Taiwan
No. 135, Nan-Xiao Street, Changhua 500, Taiwan
mass in the right hepatic lobe (S5/8), without significant blood flow. Imaging with a multidetector computed tomography (CT) scanner (LightSpeed 16; GE Healthcare) showed a mild heterogeneously enhancing mass in the arterial phase, which was surrounded by a low-density peripheral rim and contained with laminated septa extending to the right anterior abdominal wall (Fig. 1a). In the delayed venous phase, progressive and persistent enhancements were observed in the peripheral rim and in the numerous fine septa of the mass (Fig. 1b).

MRI (1.5-T MR system; Magnetom Verio, Siemens) showed a large liver mass that had extended to the anterior abdominal wall. The mass appeared as a heterogeneous hypointense area on a T1-weighted image and as a slightly hyperintense area on a T2-weighted image (Fig. 2). With 3 different b values set at 100, 500, and 1000 s/mm², the results from axial DW MRI and the ADC map showed water restriction of the mass (Fig. 3). The mean ADC value (0.78 × 10⁻³ mm²/s²) was measured as follows: (1) The axial image with the largest diameter was selected. (2) A circular region-of-interest (ROI) was made by placing a maximal circle entirely inside the mass, but not exceeding the mass margin so as to avoid inclusion of the peripheral wall of the mass. Dynamic contrast-enhanced fat-saturated T1-weighted MRI showed a mild heterogeneously enhancing mass in the arterial phase and numerous fine septa within the mass.

**Figure 1.** a. Arterial phase CT scan shows a mild heterogeneously enhancing mass surrounded by a thick, low-density peripheral rim (arrows), with laminated septa (star) extending to the anterior abdominal wall. b. Delayed phase CT scan shows progressive enhancement of fine septa (stars) within the mass and a thick peripheral rim (arrows).

**Figure 2.** a. The mass is seen with low signal intensity on a T1-weighted MR image and high signal intensity on a T2-weighted MR image b.
and a thick peripheral rim in the delayed phase, indicating progressive enhancement (Fig. 4).

US-guided needle aspiration biopsy was performed twice, and on both occasions, xanthogranulomatous inflammation was observed. Under the radiological impression of a possible peripheral cholangiocarcinoma (PCC), the patient underwent an extended right lobectomy. An ill-defined whitish tumor was found (Fig. 5a). Microscopically, actinomycetes were identified in the fibrotic and xanthogranulomatous inflammation background and were accompanied with dense infiltration of inflammatory cells and foamy histocytes (Fig. 5b). The patient was discharged in stable conditions.

**DISCUSSION**

Actinomycosis was first described in 1877, as filaments radiating from a central mass. It is a chronic suppurative and granulomatous disease caused by Actinomyces israelii [1]. The cause of abdominal actinomycosis is thought to be linked to a source of previous intestinal infection, trauma, abdominal surgery, or long-standing use of an intrauterine device [8]. When the source of the infection is not found, the condition is considered primary or isolated hepatic actinomycosis (IHA) [8]. In the case of our patient, no underlying infection source was identified except for a total abdominal hysterectomy performed 2 decades earlier.

**Figure 3.** High signal intensity is seen on the diffusion-weighted MR image **a**, and low signal intensity is seen on the ADC image **b**.

**Figure 4.** Dynamic fat-saturated MR images show numerous fine intramass septa (stars) and a thick peripheral rim (arrows), indicating progressive enhancement.
IHA can appear as multiple liver abscesses, disseminated microabscesses, or a mass-like lesion [8, 9]. On reviewing the published literature, they were often misdiagnosed as cholangiocarcinomas or metastatic tumors [3, 4, 8-10]. In most cases, resemblance to actinomycosis in other parts of the body, extension to the adjacent organs was a common finding. This finding may be associated with the specific proteolytic enzymes produced by actinomycetes [11]. Thus, early treatment of IHA is important to avoid infiltration to adjacent organs [12, 13]. In many reported IHA cases, invasion to the subcapsular region of the liver was seen, which caused pleural effusion or even pericardial effusion [8]. However, in the case under study, no pleural effusion or pericardial effusion was detected.

IHA commonly shows numerous fine enhancing septa after intravenous contrast administration [4, 10, 12]. This was observed in our patient, and such findings can be attributed to the development of multiple loculated and heavy inflammatory collections that are separated by fine fibrous septal tissues and granulation, thereby causing progressive enhancement and resulting in a honeycomb-like appearance within the mass [4, 12].

A key consideration in the differential diagnosis of IHA is periphery cholangiocarcinoma (PCC), which involves peripheral enhancement in the mass margin [4, 7]. PCC also involves a fibrous stroma admixed with coagulative necrosis, cell debris, and mucin, leading to prolonged septa-like enhancement [4]. These radiological findings overlap with those for hepatic actinomycosis and cholangiocarcinoma. Chan et al. stated that DWI is a useful noninvasive imaging technique for differentiating hepatic abscesses from cystic and necrotic tumors [5]. In an evaluation of 134 patients with 50 healths, 25 cholangiocarcinoma, and 59 focal hepatic lesions showed that the average ADC value of cholangiocarcinoma was $1.56 \pm 0.23 \text{ (s/mm}^2\text{)}$ [7]. The lower mean ADC value in our case ($0.78 \times 10^{-3} \text{ s/mm}^2$) could be because of the contents of the actinomycotic mass, which consisted of inflammatory cells, bacteria, necrotic tissue, and proteinaceous exuded plasma, which has a very high viscosity [5].

Ring enhancement of the liver is also observed in hepatic metastases [4, 14]. The patient’s history of primary malignant tumors and the absence of linear fibrotic septa within the mass were characteristic of liver metastasis, thus helping us differentiate it from IHA [15].

A definite diagnosis of IHA should be made if sulfur granules or recovery of Actinomyces colonies are observed on microscopic examination [10]. Needle aspiration or even core biopsy may, as in our case, only show whether inflammatory or fibrous cells are present, without identification of the infectious organism [4]. The response to antibiotic therapy, usually penicillin alone or in combination with clindamycin or ciprofloxacin, was generally excellent [8]. Surgical resection is considered an option for ruling out malignancy or in the case of antimicrobial treatment failure [13].

In conclusion, IHA is a rare granulomatous disease, which is usually misdiagnosed as liver cancer. Extension to

**Figure 5**

5a. A whitish lobulated mass seen in the resected right hepatic lobe. 5b. Actinomycotic colonies in the matrix of sulfur granules (black arrows) and club-shaped filaments (white arrows) that form a radiating rosette pattern are identified on microscopic examination.
adjacent organs and the presence of numerous honeycomb-like septa within the mass are characteristics of IHA. Recently, more articles have discussed the role of DWI and the ADC value in differentiating liver lesions. The low mean ADC value found for our patient was significantly different from that of PCC, thereby helping us make a correct diagnosis.

REFERENCE