Actinomycosis of the Gallbladder Mimicking Carcinoma: a Case Report with US and CT Findings

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We describe a case of actinomycosis of the gallbladder mimicking carcinoma. Sonography showed a hypoechoic mass replacing gallbladder lumen and engulfing a stone; contrast-enhanced computed tomography showed a heterogeneously enhanced thickened gallbladder wall with subtle, disrupted luminal surface enhancement, which formed a mass. As a result of the clinical and radiologic presentation, our impression was of gallbladder carcinoma. Actinomycosis should be included in the differential diagnosis when sonography and computed tomography findings show a mass engulfing the stone in the gallbladder and extensive pericholecystic infiltration with extension to neighboring abdominal wall muscle.

Index terms:
Gallbladder, actinomycosis
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Actinomycosis is a chronic suppurative and granulomatous disease that is characterized by the formation of multiple abscesses, draining sinuses, abundant granulation and dense fibrous tissue. The disease is most frequently caused by Actinomyces israelii. These organisms are gram-positive anaerobic bacteria, and are considered opportunistic pathogens associated with infection, trauma or surgery. These events allow them to cross mucosal barriers as these organisms are normally present in healthy individuals, especially in the oral cavity, gastrointestinal tract, and female genital tract (1). In abdominopelvic actinomycosis, aggressive perilesional infiltration with a tendency to cross fascial planes or boundaries and extend to the abdominal wall has been well described as an important radiologic finding (1). Actinomycosis of the gallbladder is an extremely rare disease; only 21 cases have been reported in the English literature (2–7). Moreover, a diagnosis of actinomycosis of the gallbladder is difficult because this condition can be confused with carcinoma (2, 8). We report here on a rare case of actinomycosis of the gallbladder that presented as a mass by sonography and computed tomography (CT).

CASE REPORT

A 65-year-old man presented with an intermittent right upper quadrant abdominal pain of 15 days duration. He was afebrile and had no history of abdominal surgery or trauma. On physical examination, the patient showed mild tenderness of the right upper abdominal quadrant. Leukocytosis was absent. Liver function tests showed transaminase and total bilirubin levels at normal levels, but an elevated γ-glutamyl transferase level (101 U/L). Levels of serum alpha-fetoprotein, carcinoembryogenic antigen, cancer antigen 19–9, and cancer antigen 72–4 were all within their normal ranges.

Abdominal sonography showed a hypoechoic mass replacing gallbladder lumen and engulfing a gall stone with an indistinct margin between the mass and adjacent liver

(Fig. 1A).

Unenhanced and contrast-enhanced arterial- and portal-phase multidetector-row helical CT of the gallbladder was performed under the impression of gallbladder carcinoma. Contrast-enhanced CT scans obtained during arterial- and portal-phases showed a heterogeneously enhanced and markedly thickened walls of the gallbladder body and fundus with an impacted gallstone and subtle, disrupted luminal surface enhancement, which formed a mass (Figs. 1B−D). A second gallstone was also observed in the gallbladder, and intramural calcification was noted (Fig. 1B). The mass of the gallbladder showed direct hepatic involvement and a perihepatic extension beyond the gallbladder (Figs. 1C, D). The mass had infiltrated to the neighboring transversus abdominis muscle and gastric antrum with fatty infiltration around the gallbladder and in

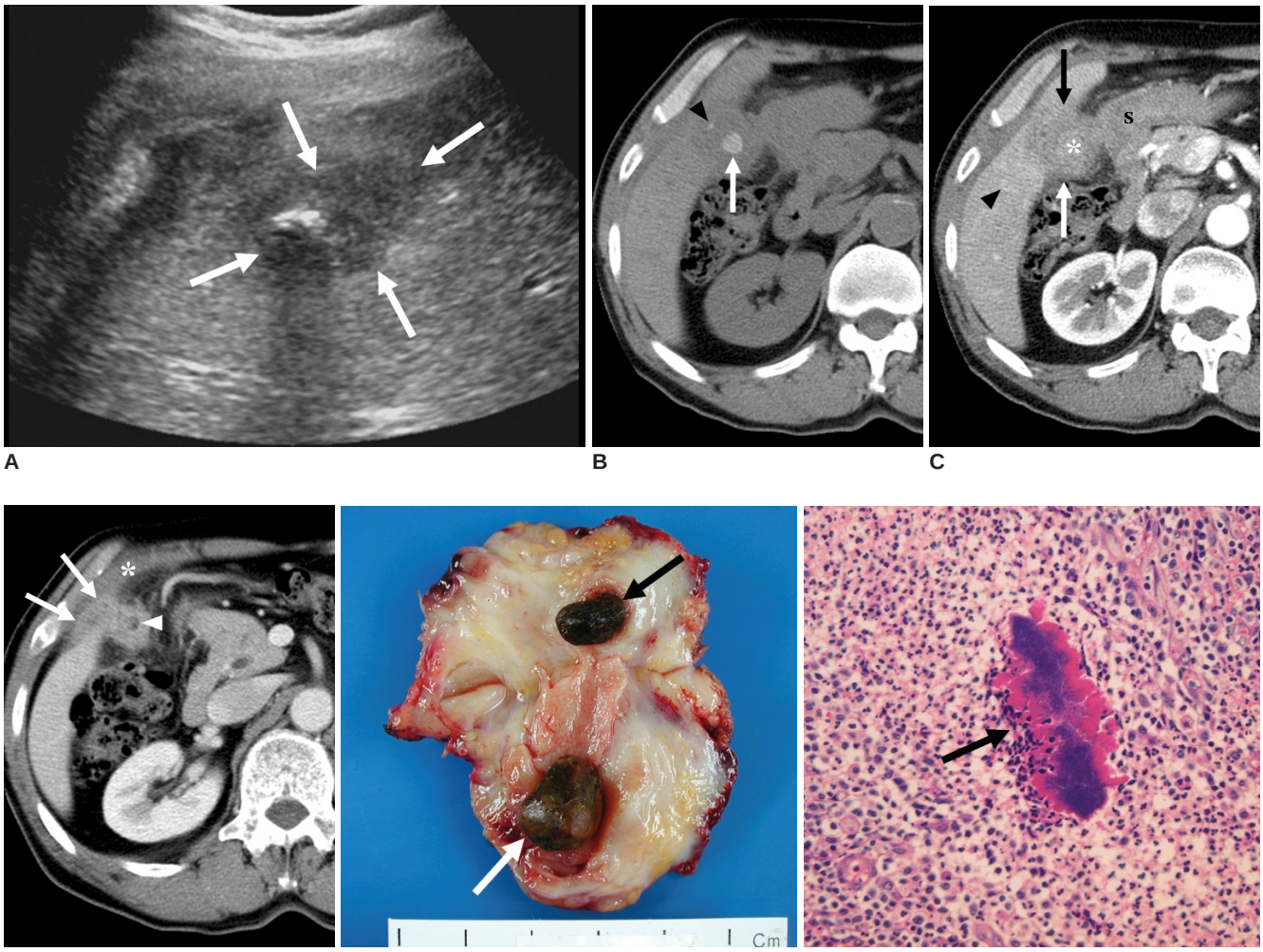

Fig. 1. A 65-year-old man with actinomycosis of the gallbladder which presented as a mass.
A. Sonograph shows a hypoechoic mass (arrows) replacing gallbladder lumen and engulfing a gallstone with infiltration of surrounding liver.
B. Unenhanced CT scan shows the impacted gallstone (arrow) and calcification (arrowhead) in the thickened gallbladder wall. The other gallstone is in the neck of the gallbladder (not shown).
C. Contrast-enhanced CT scan obtained during the arterial phase at the same level as **B**, shows a heterogeneously enhanced and markedly thickened gallbladder wall (arrows) with infiltration of surrounding liver, stomach, and pericholecystic fatty tissue. Faint, disrupted luminal surface enhancement around the stone (asterisk) and hepatic parenchymal enhancement (arrowhead) adjacent to the thickened gallbladder wall are evident. S = stomach.
D. Contrast-enhanced CT scan obtained during the portal phase at a level 1 cm below **C** shows disrupted luminal surface enhancement of the thickened gallbladder wall (arrowhead) with pericholecystic fatty infiltration, perihepatic extension of the soft tissue mass (arrows), and mild thickening of the transversus abdominis muscle (asterisk).
E. Photograph of the cut surface of the resected gallbladder shows markedly thickening of gallbladder body and fundus walls and a fibrotic appearance. Two brown stones (arrows) are indicated.
F. Photomicrograph of the thickened gallbladder wall shows inflammatory cell infiltration with fibrosis, and a sulfur granule (arrow) containing tangled filamentous bacilli, which is compatible with Actinomyces. (H & E stain, ×400).

the greater omentum (Figs. 1C, D). Pericholecystic lymph node enlargement was noted. In addition, hepatic parenchymal enhancement was observed adjacent to the gallbladder in arterial phase CT (Fig. 1C), and this was followed by isoattenuation, relative to the normal liver, during the portal phase. As a result of the clinical and radiologic presentation, our impression was of gallbladder carcinoma with direct involvement of adjacent structures with omental seeding. We did not suspect actinomycosis of the gallbladder. Diagnostic laparotomy was then performed. Operatively, the diseased gallbladder was found to be perforated with severe adhesion to liver and greater omentum. Surgical findings initially appeared to be consistent with advanced carcinoma of the gallbladder. However, the intraoperative biopsy of a frozen section revealed inflammation with no evidence of carcinoma. Cholecystectomy was next performed.

The cut section of the resected gallbladder revealed that the gallbladder body wall and fundus were diffusely thickened with two gallstones, and had a fibrotic appearance (Fig. 1E). Microscopic examination of the thickened wall showed inflammatory cell infiltration with fibrosis and intramural sulfur granules that contained tangled filamentous bacilli compatible with Actinomyces (Fig. 1F). There was no evidence of gallbladder carcinoma.

After surgery, the patient was treated with penicillin intravenously for 10 days, followed by oral therapy with amoxicillin for six months with no specific symptoms or abnormal laboratory findings.

DISCUSSION

Actinomycosis is most commonly caused by the gram-positive anaerobic bacterium Actinomyces israelii, other species include A. viscosus, A. odontolyticus, A. naeslundii, A. meyeri, and A. gerencseriae, which rarely produce disease.

In clinical practice, abdominopelvic actinomycosis is rarely suspected, though it is one of the most frequent mimics of carcinoma. Although 20% of human actinomycosis occur in an abdominopelvic region; most commonly in the ileocecal region, including the appendix and less commonly in another organs including ovaries, liver, gallbladder, and pancreas; actinomycosis of the gallbladder is an extremely rare finding (2). This condition may present as cholecystitis, biliary colic, pancreatitis, and a neoplastic mass with abdominal pain, fever, weight loss, a palpable mass, and laboratory abnormalities, such as, leukocytosis with an elevated erythrocyte sedimentation rate, and elevated levels of serum alkaline phosphatase, bilirubin and/or amylase (2, 5−7).

Although the mode of spread of abdominopelvic actinomycosis is not fully understood, direct spreading into adjacent tissue appears to be most common after the organism has penetrated through a breached mucosal barrier. Hematogenous spread may also occur, but lymphatic spread is said not to occur because of the large size of the organism. It has been demonstrated that actinomycosis is unable to grow in the presence of bile salts (3, 5, 7). Thus, it is possible that the organism spread to the gallbladder in our case via the hematogenous route. However, we consider that gallstone induced mucosal injury and retrograde spread from the duodenum explain the most commonly accepted pathophysiology.

Mass formation and an aggressive infiltrative nature with a tendency to cross fascial planes or boundaries, involve multiple compartments, and extend to the abdominal wall are considered important radiologic features of actinomycosis (1). However, a preoperative diagnosis of abdominal actinomycosis is difficult; less than 10% of cases are detected preoperatively (7). Moreover, no reported case of actinomycosis of the gallbladder has been diagnosed prior to laparotomy or histology (2−7). As shown by the present case, actinomycosis of the gallbladder presenting as a mass with extensive infiltration into surrounding structures by sonography and CT is very difficult to differentiate from gallbladder carcinoma and chronic inflammation, especially from xanthogranulomatous cholecystitis, as radiologic features overlap considerably (9). In addition, actinomycosis of the gallbladder can co-occur with gallbladder carcinoma (2).

Owing to clinical and radiologic similarities of gallbladder carcinoma and benign gallbladder disease, especially complicated acute or chronic cholecystitis and xanthogranulomatous cholecystitis, in cases where a correct diagnosis is impossible, either close clinical and radiological follow-up or imaging-guided aspiration or biopsy may be useful to establish the suitability of nonoperative treatment (10, 11). However, on the other hand, surgical exploration may be necessary to eliminate the risks of false-positive or false-negative percutaneous aspiration or biopsy results or to alleviate fear of malignancy.

In our case, subtle and disrupted luminal surface enhancement of the thickened gallbladder wall and hepatic parenchymal enhancement adjacent to the diseased gallbladder were observed by contrast-enhanced CT. However, gallbladder carcinoma may accompany these findings (9), which thus, may not be helpful for the differentiation of gallbladder carcinoma and actinomycosis of the gallbladder. Intramural calcification was also noted in our case, but this finding too may be seen in gallbladder carcinoma (12).

Actinomycosis of the gallbladder may be diagnosed by aspiration or percutaneous needle biopsy, but Actinomyces cultures can yield a false negative result in up to 76% of abdominal actinomycosis cases. Moreover, the sulfur granules typically seen in actinomycosis are present in only 50% of cases, and can be formed by other microorganisms, including Staphylococcus, Streptomyces, Aspergillus, and Nocardia species (7). Therefore, surgical exploration may be required to differentiate actinomycosis from gallbladder carcinoma, and for the debridement of necrotic tissue.

Reports issued from the 1980s increasingly support that medical therapy alone, without surgical exploration, is usually sufficient for cure, irrespective of extensive actinomycosis (13, 14). Currently, the cure rate of actinomycosis is high, and deformity and death are infrequent, because antibiotic based therapy has greatly enhanced prognosis for all forms of actinomycosis. Thus, although a diagnosis of actinomycosis, especially when it mimics a malignancy as in our case, is rarely considered, CT findings, such as, extensive inflammatory infiltration, a tendency to cross fascial planes or boundaries, the involvement of multiple compartments, and extension to the abdominal wall are helpful for differentiating actinomycosis of the gallbladder and malignancy. Subsequently, an adequate preoperative diagnosis based on aspiration or biopsy may allow the adoption of a chemotherapeutic approach rather than surgical exploration.

In conclusion, actinomycosis of the gallbladder is extremely rare and its preoperative diagnosis a challenge because it sometimes mimics gallbladder carcinoma. Therefore, although non-specific, actinomycosis should be included in the differential diagnosis when sonographic and CT findings show a mass engulfing a stone in the gallbladder with extensive pericholecystic infiltration and extension to neighboring abdominal wall muscle.

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