Annals of Clinical Case Reports

Case Report
Published: 21 Aug, 2017

Actinomycosis of Gallbladder: A Diagnostic Dilemma

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Abstract

We report a rare case of gallbladder actinomycosis in an 88 years old man. The patient presented with a clinical feature of an acute cholecystitis, and a radiological finding mimicking a gallbladder carcinoma. The diagnosis of actinomycosis was made by histopathological examination of the surgical specimen, which is often the case in such infection. The surgery was followed by a long-term antibiotic treatment to prevent recurrence.

Keywords: Actinomycosis; gallbladder; cholecystectomy; penicillin

Introduction

Actinomycosis is a chronic suppurative and granulomatous disease characterized by its capacity to invade surrounding tissues, development of multiple abscesses, granulation and dense fibrous tissue [1]. The infection is frequently caused by *Actinomyces israelii* [2]. Actinomycosis of the gallbladder is an extremely rare disease; less than 50 cases have been reported in the literature [3]. Here we report a new case of actinomycosis of the gallbladder mimicking a gallbladder carcinoma.

Case Presentation

An 88-years-old North African-man presented with fever and nausea and a history of three months of intermittent right upper quadrant pain which became severe 2 days before his hospital admission. Past medical history showed hypertension and diabetes. Clinical examination revealed moderate dehydration, pyrexia (39°C) and tachycardia (118 beats/min), conjunctive jaundice and a 10 cm tender mass in the right upper abdominal quadrant.

Blood tests showed high levels of white cell count (16 000/mm^3) and C-reactive protein (56 mg/dL). Hepatic blood test showed an elevated alkaline phosphatase (480 IU/L) and gamma glutamyl transpeptidase (570 IU/L). Total and conjugated bilirubin levels were slightly high (2.1 mg/dL and 1.1 mg/dL respectively) and serum albumin was low (17 g/L). For tumor markers, carcinoembryonic antigen and carbohydrate antigen 19-9 were normal (6.8 ng/mL and 1 U/mL respectively).

Ultrasonography reviewed an acute calculous cholecystitis and a lesion in the gallbladder, which was isoechoic to surrounding parenchyma. A Computed Tomography (CT scan) of the abdomen was performed and showed a mass mimicking a gallbladder carcinoma (Figure 1). The wall of the gallbladder was thickened and stones were found in the main bile duct. Intraoperative findings suggested gallbladder cancer (Figure 2). The patient underwent a cholecystectomy with choledochotomy and placement of a Kehr drain after extraction of bile duct gallstones. Because of the emergent context, a cholecystectomy was carried out awaiting the histological results and the complementary imaging before deciding the necessity of further resections. Opening of the surgical specimen found a parietal thickening with the presence of multiple small abscesses (Figure 3). Histologic examination was consistent with peri-gallbladder necrosis containing numerous bacterial colonies of *Actinomyces* species (Figure 4). A diagnosis of gallbladder actinomycosis was made. The patient made an uneventful recovery and was started on a 6-months cure of penicillin. Ona year after surgery, no recurrence has been observed.

Discussion

Actinomycosis is an infection caused by a gram-positive anaerobic bacterium: *Actimyces* species [1,3]. There are no less than 30 species of *actinomyces*, the most common is *Actimyces Israelii*. Other species can produce infection in humans such *A. Viscosus A. naeslundii, A. odontolyticus, gerencseriae* and *A. meyeri* [1]. The bacterium is common commensal in the mucous membrane

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Received Date: *03 Jul 2017*
Accepted Date: *18 Aug 2017*
Published Date: *21 Aug 2017*

Citation:
Elmekkaoui A, Bennani A, Soufi M, Bouziane M, Ismaili Z. Actinomycosis of Gallbladder: A Diagnostic Dilemma. Ann Clin Case Rep. 2017; 2: 1422.

ISSN: 2474-1655

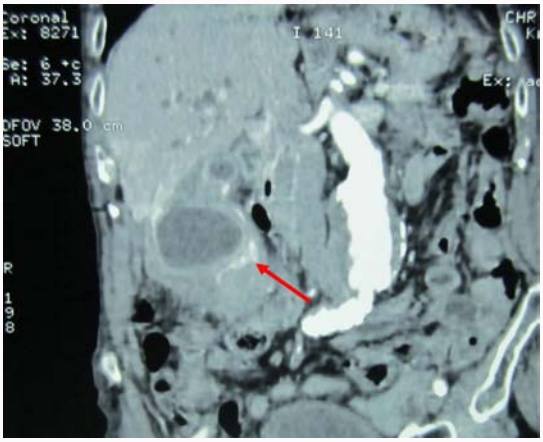

Figure 1: Gallbladder mass mimicking a carcinoma.

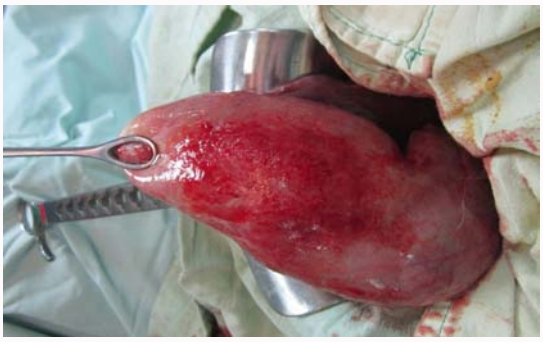

Figure 2: Per-operative gallbladder aspect.

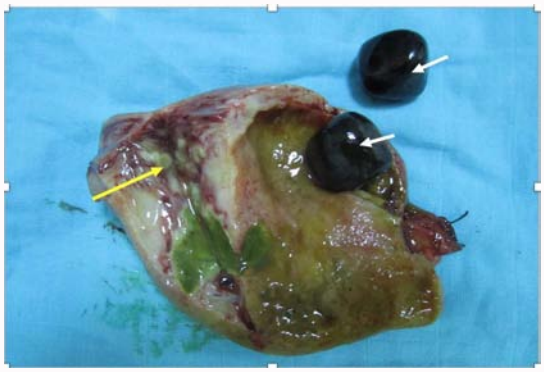

Figure 3: Parietal thickening with presence of multiple small abscesses (long yellow arrow) and stones (white arrows).

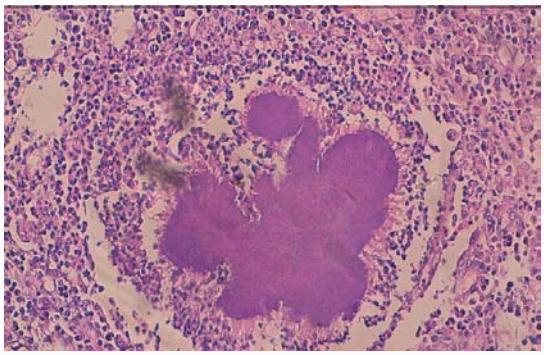

Figure 4: Chronic inflamed granulation tissue with many characteristic 'sulphur granules' of Actinomycosis, composed of basophilic radiating filaments (hematoxilin eosin safran X10).

of oral cavity, intestinal tract and female genitalia. *Actinomyces* were once considered fungi because of their branching filaments [4]. Latter, taxonomic studies confirmed that *Actinomyces* are bacteria rather than fungi by virtue of their lack of nuclear membrane or cell wall chitin, reproduction by fission, insensitivity to amphotericin B and sensitivity to penicillin [5,6]. But the question is still debated; recently Rothschild and al. evaluated *actinomyces* spp and, surprisingly, found that it showed great similarities to fungal infections, bringing into question its current classification as a bacterium [3,7].

Actinomyces are microorganisms of low pathogenicity and require mucosal barrier disruption caused by trauma, surgery (e.g. perforated acute diverticulitis or appendicitis), foreign body ingestion (e.g. fish or chicken bone), irradiation, or local inflammatory to produce disease [2]. Other risk factors are described, like immunosuppression condition, neoplastic disease, diabetes and intrauterine contraceptive devices (IUDs) [2]. Furthermore, actinomycosis is generally a polymicrobial infection, associated with other bacteria in 65% of cases [8]. Copathogenes may assist in the spread of infection by inhibiting host defenses and reducing local oxygen tension. Once infection is established a subacute-to-chronic disease is produced and spread to surrounding tissues without regard for anatomic barriers. The end result is a chronic, indurated, suppurative infection or a granulomatous inflammation and frequent development of multiple abscesses; especially in abdominal site. The fibrotic walls of the mass before suppuration may be confused with a neoplasm [2].

The most common localization of this infection is the cervicofacial. Ten to 20% occur in abdomino-pelvic region, which is the most indolent and non-specific presentation [3]. The infection involves usually the ileocecal region mimicking Crohn disease or tuberculosis. Actinomycosis of the gallbladder is a very rare condition; about 50 cases are reported in the medical literature [3,8]. In the gallbladder, appendicitis is considered the primary source of actinomycosis, but infection may follow a perforated intestinal ulcer or even a blunt trauma. Retrograde spread of actinomycetes from the duodenum through the common bile duct or hematogenous spread have been considered as a possible route of infection [4].

There are no specific clinical or radiological signs of the infection. A preoperative diagnosis is difficult; less than 10 % of cases are detected preoperatively [1]. In the case of gallbladder, no reported case of actinomycosis of the gallbladder has been diagnosed prior to laparotomy [1]. Moreover, gallbladder actinomycosis can be presented as a chronic cholecystitis but more frequently as an acute disease [3]. Ultrasonography and CT scan usually reveal a tumour-like, infiltrative mass that enhances with contrast, mimicking a gallbladder adenocarcinoma [8]. There is no specific, valid serological test for diagnosis. A direct cyto-bacteriological examination of the bile collected intra-operatively, carried out by an informed biologist, or at best an anaerobic culture could show the organism in 50% of cases; but in the latter case it can take several weeks which is not very useful for such cases [9]. The presence, in microbiological study, of a non–acid-fast, a gram-positive organism with filamentous branching is suggestive of the diagnosis [2]. It is very common for the diagnosis to be made only after the pathological examination of the surgical specimen.

After a cholecystectomy, prolonged antimicrobial therapy (6 to 12 months) is recommended for patients with all clinical forms of

actinomycosis to prevent disease recrudescence. However, adequate drainage is indicated if abscesses are present [2]. The treatment of choice is high dose intravenous penicillin followed by oral penicillin alone (i.e., amoxicillin, piperacillin) or combined with β-lactamase inhibitor (i.e., clavulanate, tazobactam) to cover others aerobic and anaerobic co-pathogens [2,9].

Conclusion

Gallbladder actinomycosis is a very rare condition that mimics a wide variety of intra-abdominal complains such as malignant disease. The diagnosis is difficult and usually post-operative, made by the histological examination of the cholecystectomy specimen. The treatment needs long term antibiotics to avoid recurrence.

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