

Actinomycosis of the Gallbladder: A Case Report

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Abstract: Actinomycetes are a normal part of the microflora of the cervicofacial region and gastrointestinal tract, but they can cause infections when the normal mucosal barrier is lost due to surgery, trauma or infections. Actinomycosis of the gallbladder is extremely rare. A 70-year-old man with ischaemic heart disease, hypertension, non-insulin dependent diabetes mellitus and hypercholesterolaemia presented with episodes of acute pancreatitis and cholecystitis. CT scan suggested a perforated gallbladder. The liver was normal. At cholecystectomy the gallbladder was contracted with adhesions and a probable cholecystochocho-duodenal fistula. Histology showed an inflamed and fibrotic gallbladder with colonies of Actinomycetes, the so-called 'sulphur granules'. Postoperatively, he has been well.

Keywords: Gallbladder, actinomycosis.

INTRODUCTION

Actinomycosis usually involves the head and neck region, thorax and abdomen. Pelvic actinomycosis may occur in women using intrauterine contraceptive devices. Abdominal actinomycosis is rare, usually occurring in the caecum and appendix. Actinomycosis of the gallbladder is extremely rare; to date only 20 cases involving the gallbladder or bile duct have been reported [1-23]. We report a further case of actinomycosis of the gallbladder and review the literature.

CASE REPORT

A 70-year-old man with one episode of acute pancreatitis in 2002 presented with two episodes of acute cholecystitis in 2004 when a CT scan showed a possible sealed-off perforation of the gallbladder with thickened mucosa and a small stone in the lumen. There was air in the bile duct, but no evidence of subphrenic fluid or pleural effusions. The liver was normal.

He underwent ERCP and sphincterotomy. The common bile duct was found to be dilated, but no stones were seen. Due to medical co-morbidity (ischaemic heart disease, hypertension, non-insulin dependent diabetes mellitus and hypercholesterol aemia) it was decided that the risk for elective resection of the gallbladder was too high at that time. He was treated with intravenous Augmentin and Flagyl for five days.

He remained well for two years, until he again required three admissions for acute abdominal pain during a two-month period. He was diagnosed with acute cholecystitis, but had no evidence of acute pancreatitis on these admissions. CT scan demonstrated a thickened gallbladder containing gallstones, a small out-pouching possibly due to a sealed-off perforation, and gas within the biliary tree.

He underwent laparoscopic cholecystectomy, which was converted to open cholecystectomy due to a very contracted

gallbladder, adhesions and a probable small cholecystochocho-duodenal fistula. He was well postoperatively and follow up in clinic has been uneventful. He did not receive pre- or post-operative antibiotics.

The gallbladder (35 mm in length) was tubular and firm with a spongy cut surface and a small lumen. No stones were submitted with the specimen. Histology showed a fibrotic gallbladder wall with acute and chronic inflammatory cell infiltrates that included abundant foamy macrophages. Colonies of Actinomycetes, the so-called 'sulphur granules', with the characteristic Splendore-Hoeppli phenomenon, were present in the lumen (Fig. 1).

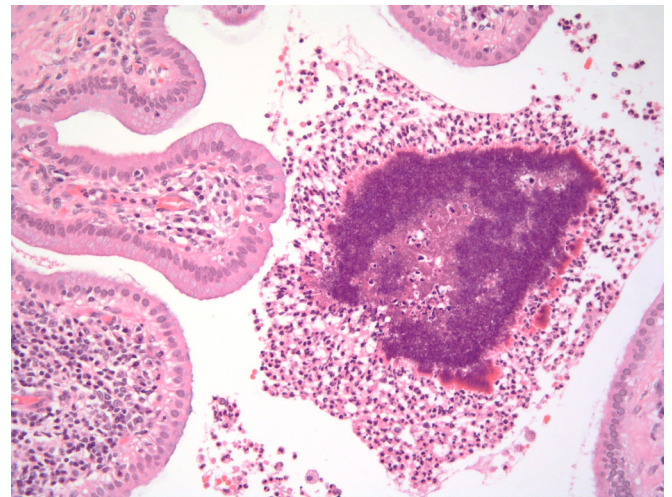


Fig. (1). Colony of Actinomycetes with Splendore-Hoeppli phenomenon surrounded by polymorphs within the lumen of the gallbladder.

DISCUSSION

Actinomycetes are filamentous anaerobic Gram-positive bacteria that are a normal part of the microflora of the cervicofacial region and gastrointestinal tract. They can become pathogenic when the normal mucosal barrier is lost due to surgery, trauma or infections.

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Table 1. Actinomycosis of the Gallbladder and Bile Ducts

No.	Country	Age/Sex	Stones	Associated Diseases	Sites	SG	GS	Culture	Identification
1	UK	47 M	-	Liver tumour	GB*	+	+	?	<i>Actinomyces sp.</i>
2	USA	? F	+	None	GB	+	+	-	<i>Actinomyces sp.</i>
3	Germany	55 F	-	None	GB	+	+	+	<i>A. israelii</i>
4	USA	65 F	+	None	GB	-	+	+	<i>A. naeslundii</i>
5	UK	73 M	-	Amyloidosis	GB*	+	+	-	<i>A. israelii</i>
6	Canada	77 M	+	None	GB	+	+	+	<i>A. israelii</i>
7	USA	86 F	+	Liver abscess	GB*	+	+	-	<i>A. israelii</i>
8	USA	56 M	-	Rheumatoid Arthritis	Duct	-	+	-	<i>A. israelii</i> **
9	Australia	72 F	-	Heart failure, etc	GB	+	+	-	<i>Actinomyces sp.</i>
10	France	72 F	-	None	GB	-	+	+	<i>A. naeslundii</i>
11	France	60 M	+	None	GB	-	+	+	<i>A. naeslundii</i>
12	France	57 F	+	Carcinoma GB, Liver Abscess	GB	-	-	+	<i>A. naeslundii</i>
13	USA	80 F	+	None	Duct	+	+	-	<i>A. naeslundii</i> **
14	India	71 F	+	Glomerulonephritis. Myocard infarct	GB	+	NP	-	<i>Actinomyces sp.</i>
15	Sudan	50 M	+	Diabetes,	GB	+	+	NP	<i>Actinomyces sp.</i>
16	UK ***	70 M	+	Diabetes, Myocardial infarction, etc	GB	+	NP	NP	<i>Actinomyces sp.</i>

GB = Gallbladder, * = liver involved, ** = Confirmed by immunofluorescent stain, NP = not performed, SG = sulphur granules, GS = Gram's stain, *** = present case

To date only 20 cases of actinomycosis of the gallbladder or bile ducts have been reported [1-23]. Table 1 summarises the main clinico-pathological features [1-14]; some cases have not been included for lack of confirmatory evidence [15] or due to insufficient information [16-23].

The diagnosis of actinomycosis is usually based on the demonstration of "sulphur granules" and / or isolation of the organism in cultures. *Actinomyces* species identified are *A. israelii* and *A. naeslundii*. However, more recent methods of identification have identified several new species and resulted in changes in the taxonomy of the genus *Actinomyces* [24, 25]. It is worth noting that some *Actinomyces* species may not form 'sulphur granules' and unless samples are taken for anaerobic culture, the diagnosis may be missed [10].

Infection appears to be due to bacterial reflux from the duodenum into the bile duct. In some cases, the invasion of the gallbladder may have occurred following mucosal damage by chronic cholecystitis. It is believed that lymphatic spread does not occur in actinomycosis due to the large size of the filaments. However, haematogenous spread to the liver is documented. *Actinomyces* do not grow in the presence of bile salts, except for certain strains of *A. naeslundii*. Therefore, the mere presence of sulphur granules in the gallbladder has been considered indicative of invasive disease and not colonization.

Patients may present with fever, malaise and right quadrant pain. The mean age at presentation is 65 years, more commonly in females, and cholelithiasis is present in most cases. Patients are treated with antibiotics. The majority of cases have a favourable outcome, unless associated with underlying malignancy or other multiple medical problems.

Immunosuppression, prolonged antibiotic therapy and corticosteroid therapy are the usual predisposing factors [13].

Actinomycosis must be treated with high doses of antimicrobials. Initial treatment is with high doses of intravenous penicillin for two to six weeks followed by oral therapy with penicillin or amoxicillin for six to twelve months. It is not clear whether cholecystitis can be treated with antibiotics alone. All of the reported cases of gallbladder actinomycosis have been diagnosed after resection. Cytological detection from aspirated bile has been reported [26]. Endoscopic retrograde cholangiopancreatography (ERCP) aspiration of bile for cytology and culture is theoretically sound but needs to be investigated. If gallbladder actinomycosis could be diagnosed by cytology and/or culture of ERCP aspirated bile before surgery, and the patient remained stable, it may be possible to treat with antibiotics alone, as has been reported in other cases of abdominal actinomycosis.

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