A case of recurrent acute cholecystitis caused by *Actinomyces odontolyticus*, rare actinomycosis

Kento Furuya1*, Kenta Ito1, Kyohei Sugiyama2, Akitsugu Fujita3, Hideyuki Kanemoto3 and Toshio Shimada1

**Abstract**

**Backgrounds:** *Actinomyces* species are gram-positive, obligate anaerobic rods and are rare causes of cholecystitis. Because *Actinomyces* species are anaerobic bacteria, it is difficult for *Actinomyces* to survive in bile apart from *A. naeslundii*. We experienced a case of recurrent acute cholecystitis caused by *A. odontolyticus*.

**Case presentation:** A patient had been diagnosed with acute cholecystitis and treated one month before and after that, admitted to our hospital because of recurrent cholecystitis. Gram stain of the bile revealed gram-positive rods and gram-positive cocci. We found *A. odontolyticus* and MRSA in bile culture and MRSA in blood culture. We administered piperacillin-tazobactam and then changed it to ampicillin-sulbactam and vancomycin. The patient underwent laparoscopic cholecystectomy and was discharged safely.

**Conclusions:** To our knowledge, this is the first case of cholecystitis caused by *A. odontolyticus*. Cholecystitis caused by *Actinomyces* species is rare. In addition, we may overlook it with the low positivity of bile cultures of *Actinomyces*. Whenever the cholecystitis recurs without any obstruction of the biliary tract, we should search for the gram-positive rods hidden in the bile, such as *A. odontolyticus*, as the causative organism, even if the bile culture is negative.

**Keywords:** *Actinomyces odontolyticus*, Cholecystitis, Gram stain, Ampicillin/sulbactam

**Backgrounds**

*Actinomyces* species are gram-positive, obligate anaerobic rods that colonize human’s upper respiratory tract, gastrointestinal tract, and female reproductive organs [1]. More than 30 species of *Actinomyces* have been identified to date and *A. israelii* is the most common pathogen [1]. In actinomycosis, intra-abdominal infections account for about 20%, and the most common site of infection is the ileum. There are few cases of cholecystitis due to *Actinomyces* species reported. To the best of our knowledge, our case must be the first case of cholecystitis caused by *A. odontolyticus*.

**Case presentation**

A 75-year-old Japanese man came to our hospital with fever, chills, and right hypochondrial pain. One and three years ago, this patient was already diagnosed twice with acute cholangitis and cholecystitis and then underwent endoscopic retrograde biliary drainage thirty-nine days before admission to our hospital. We demonstrated gram-positive rods, and gram-negative rods with Gram stain; namely *A. hydrophilia* and *E. faecalis* grew in the culture with no gram-positive rods. When the patient was transferred to our hospital, his body temperature was 39.9 °C, and he had tenderness in the right costal region. Blood tests showed white blood cell counts 23,700 /μL, C-reactive protein...
23.2 mg/dL, aspartate aminotransferase 63 U/L, alanine aminotransferase 51 U/L, alkaline phosphatase 233 U/L, and γ-glutamyl transpeptidase 82 U/L. Contrast-enhanced CT of the abdomen showed an enlarged and multifocal gallbladder, intrahepatic perforation and abscesses around the gallbladder, some perforating into the right lobe of the liver and forming liver abscesses. But there was no obstruction of the biliary tract (Fig. 1). We diagnosed him with acute cholecystitis and liver abscess and then administered piperacillin/tazobactam. On the second day of admission, we decided to put cholecystectomy on hold to prioritize treatment of the liver abscess, and we implemented percutaneous transhepatic gallbladder drainage. Gram stain of the bile revealed gram-positive rods and gram-positive cocci (Fig. 2), which we identified as *A. odontolyticus* and MRSA. We also detected MRSA in the blood culture collected on the first day of admission. Judging from the culture results, we changed piperacillin/tazobactam to ampicillin/sulbactam and vancomycin. Blood cultures were negative on the fourth day of hospitalization. We performed laparoscopic cholecystectomy on the 14th day of hospitalization. The pathology of the gallbladder showed no evidence of malignancy. The patient got well. On the 20th day of hospitalization, we changed antibiotics to amoxicillin/clavulanate and linezolid, and after that, the patient was discharged safely. We administered antibiotics for a total of 5 weeks. After antibiotics were completed, he had no acute cholecystitis and liver abscess recurrence.

**Discussion and conclusion**

*Actinomyces* species are gram-positive, obligate anaerobic bacteria that infect various parts of the human organs [2]. The most common site of *Actinomyces* infections is a cervicofacial area, and other locations include the central nervous system, chest, and pelvis. Hepatobiliary infection is rare and accounts for 5% of all Actinomyces infections [2].

Only 22 cases of cholecystitis caused by *Actinomyces* species have been reported in English literature (Table 1). As far as we know, this is the first case in which *A. odontolyticus* was the causative organism [3–15]. *A. naeslundii* was the most common causative

**Table 1** Characteristics of the cholecystitis caused by *Actinomyces* species [3–15]

| Characteristic                        | Value |
|---------------------------------------|-------|
| Number of cases—no.                   | 22    |
| Male:female                           | 1:1   |
| Average of age—year [range]           | 63.1 [41–86] |
| Gall stone—no. (%)                    | 14 (63.6) |
| Liver abscess—no. (%)                 | 2 (9.1) |
| Species—no.                           |       |
| *A. naeslundii*                       | 7     |
| *A. israelii*                         | 3     |
| Not identified                        | 12    |
| Gram stain positive—no.               | 12    |
| Bile culture positive—no.             | 6     |
| Past history of cholecystitis—no. (%) | 6 (27.3) |
| Average of treatment duration—days [range] | 109 [5–270] |
organism seen in 7 cases [3–5], and A. israelii in 3 cases [6, 7]. The other 12 cases were pathologically diagnosed with Actinomycosis. These results were consistent with the fact that it was difficult for Actinomyces other than A. naeslundii to survive in an environment of bile salts [8]. In this case, we detected A. odontolyticus in bile cultures. We assume that cointfected MRSA consumed oxygen and created an anaerobic environment where A. odontolyticus could grow. When A. naeslundii was the causative organism, it was the single pathogen in 6 of 7 cases (85.7%) [3–5]. On the other hand, in the cases of A. odontolyticus, 2 of 3 cases (66.7%) were infected with another bacteria; Pseudomonas aeruginosa was detected in one case [6], and Pseudomonas aeruginosa and Haemophilus aphrophilus were seen in another case [7].

Gram stain found gram-positive rods in 12 cases [8–10]. Of the 12 cases, six (50%) were positive for Actinomyces in culture [3, 10]. The mean age of the 22 patients was 63.1 years old, 14 cases (63.6%) had gallstones [3–6, 8, 10–14], and only 2 cases (9.1%) had a liver abscess [6, 15]. Of the 22 patients, six had a history of cholecystitis. Furthermore, one recurrent cholecystitis a few weeks after ending treatment was later proven to be caused by Actinomyces, as in our case [11]. Once the source of infection is controlled, the recommended treatment duration for acute cholecystitis is generally up to 7 days, even in severe cases [15]. However, in the case of cholecystitis caused by Actinomyces, the mean duration was 109 days in 22 patients because the appropriate duration of treatment for Actinomyces cholecystitis has not been established. Many cases underwent the treatment for long periods, like other actinomycosis such as lung infections.

Gram stain for Actinomyces visualizes and characterizes the presence of gram-positive rods with an elongated radial pattern [1]. However, as in another case of liver abscess caused by A. odontolyticus reported earlier [16], in this case, A. odontolyticus did not reveal a Gram stain with a typical elongated radial pattern. Thus, it may be difficult to distinguish A. odontolyticus from other gram-positive rods by Gram stain of bile. Furthermore, considering the low positive rate of bile culture in cholecystitis caused by Actinomyces, we may miss cholecystitis caused by Actinomyces. Even if the culture were negative, Actinomyces species should be considered the causative microorganism.

In conclusion, this is the first report of acute cholecystitis caused by A. odontolyticus. As cholecystitis related to A. odontolyticus is a rare condition, we may have been overlooked it because of the low positivity of bile culture and the absence of the typical elongated radial pattern on Gram stain. Furthermore, missing Actinomyces may be related to recurrent cholecystitis that may recall us Actinomyces infection.

Abbreviations
MRSA: Methicillin-resistant Staphylococcus aureus; CT: Computed tomography.

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Author contributions
All authors meet the ICMJE authorship criteria. KF was involved in literature review, planning the study and writing of the manuscript. KL, KS, AF, HK and TS were involved in the patient's care. All the authors interpreted the data, drafted and critically revised the manuscript. All authors read and approved the final manuscript.

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Competing interests
The authors declare that they have no competing interests.

Author details
1Department of Clinical Laboratory Medicine, Shizuoka General Hospital, Kitaandou 4-27-1, Aoi-ku, Shizuoka, Japan. 2Department of Pharmacy, Shizuoka General Hospital, Kitaandou 4-27-1, Aoi-ku, Shizuoka, Japan. 3Department of Surgery, Shizuoka General Hospital, Kitaandou 4-27-1, Aoi-ku, Shizuoka, Japan.

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