A rare hepatic abscess by Streptococcus intermedius complicated with hepatobronchial fistula: a case report

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ABSTRACT

Background: Hepatobronchial fistula is infrequent and secondary to pyogenic abscess caused by *Streptococcus intermedius*. This agent causes infections such as hepatic abscesses, endocarditis, bacteremia, and others. The treatment includes drainage, antibiotic management, and surgical intervention in some cases.

Case Presentation: We describe a case of a 50-year-old male with a pyogenic liver abscess complicated by hepatobronchial fistula, admitted to the Internal Medicine Department with fever, long-lasting chills, and weight loss. The diagnostic approach revealed a pyogenic abscess secondary to *S. intermedius*. After percutaneous drainage, the patient presented purulent expectoration. A cavitogram revealed a hepatobronchial fistula. Treatment with antibiotics and closure of the defect was satisfactory.

Conclusion: This is a case of a hepatic abscess due to *S. intermedius* complicated with hepatobronchial fistula, a rare clinical entity; the opportune hybrid approach (surgical plus interventional and radiological) reduces the risk of a major thoracoabdominal operation, which influenced the good prognosis of our patient.

Keywords: Hepatobronchial fistula, hepatic abscess, Streptococcus intermedius, fever, cavitogram, case report.

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Background

Pyogenic hepatic abscesses are infrequent and rarely communicate with the pleural space. Therefore, only few cases of this entity have been reported in the literature, its frequency is rare at present due to the widespread use of antibiotics [1]. Hepatobronchial fistula can be classified into congenital and acquired. The latter is usually due to hydric cysts and less frequent than pyogenic or amebic hepatic abscesses, hepatic lithiasis, and is rarely a result of hepatic trauma or surgery [2]. Streptococcus intermedius of the Streptococcus anginosus group is the causative agent and is characterized by the formation of focal or multifocal abscesses which disseminate in the systemic circulation [3]. The mainstay of treatment is surgical drainage of abscess as well as correction of the fistula. Over the last decades, some authors have reported on the success of drainage with a guided catheter by computed tomography and ultrasound, followed by long-term intravenous antibiotics [4].

Case Presentation

A 50-year-old male from Mexico City was admitted to our institution with a history of chronic alcoholism and a craniotomy for severe head trauma 14 years prior. He presented with a 2-month evolution of unquantified fever associated with asthenia, chills, and a productive morning cough with whitish sputum; a weight loss of 8 kg (11% body weight loss) was also noted in the 2-month evolution. Faced with febrile syndrome and weight loss, hospital admission was decided; during the first days of continuous hospitalization with fever and chills, the study protocol was started.

Physical examination on admission revealed blood pressure 110/70 mmHg, heart rate 112 beat per minute, temperature 39°C, respiratory frequency 19 minute, head with scarring secondary to craniotomy, skin and mucosae with pallor, chest had good excursion, absent breath sounds over the right subscapular region, as well as dullness on percussion, thus integrating a consolidation syndrome. The abdomen was soft with normal peristalsis present; no pain on palpation; no organomegaly. Rest of the exploration was within normal limits.

Initial laboratory results showed glucose 86.1 mg/dl, creatinine 0.6 mg/dl, total proteins 5.4 gr/dl, albumin 1.9 gr/dl, Alanine aminotransferase (ALT) 48 U/l, alkaline phosphatase 100.7 U/l, Lactic dehydrogenase (LDH) 190 U/l, γ -glutamyl trasferase 91 U/l, total bilirubin 0.78 mg/ dl, sodium 130 mEq/l, and potassium 4.5 mEq/l. Cell blood count showed hemoglobin 8.2 g/dl, Mean corpuscular volume (MCV) 86.8 fl, Mean corpuscular hemoglobin (MCH) 27.3 pg, leukocytes 17,300 with neutrophils 14,992 K/ul, platelets 754,000 K/ul, C-reactive protein 148 mg/l, and sedimentation rate 22 mm/hour.

Chest x-ray revealed an area of consolidation in the basal region of the right chest (Figure 1). Abdominal ultrasound showed an occupying liver lesion in segment VII and



Figure 1. Posteroanterior chest film showing right basal density and an elevation of the right hemidiaphragm.



Figure 2. Hepatic Doppler ultrasound showing the presence of an ill-defined heterogeneous oval lesion of $81 \times 66 \times 64$ mm, avascular, localized in segment VII with signs of extension to the right pleural cavity.

multiple single liver cysts (Figure 2). Triphasic abdominal tomography showed rounded density in hepatic segment VII, $58 \times 7 \times 67$ mm, with a volume of 149.5 ml, compatible with the abscess and with the presence of a fistula to the pleural cavity and right lower lung lobe (Figure 3). In the blood cultures, the growth of *S. intermedius* was obtained on the fifth day, while the sputum cultures for bacteria and mycobacteria were negative.

The patient received treatment with third generation cephalosporin and metronidazole for 6 weeks. In addition, a guided drainage by fluoroscopy with placement of a 12 F catheter was carried out obtaining purulent material, leaving it for drainage (Figure 4). During the procedure the patient presented with a cough with purulent and bloody expectoration. A cavitogram confirmed the presence of a hepatobronchial fistula (Figure 5). Because of the persistence of drainage and evidence of a fistula, surgical management was undertaken through a right posterolateral thoracotomy. At



Figure 3. Triphasic computed tomography showing bilateral pleural effusion with atelectasis and consolidation in the right inferior lobe. Hepatic segment VII showing a round, defined, hypodense collection (27-30 UH) of 58 × 74 × 67 mm, with a volume of 149.5 ml. The contrasted phase showing annular enhancement and communication with the pleural cavity. (A) Late axial. (B) Late coronal. (C) Venous sagittal.



Figure 4. Percutaneous drainage by fluoroscopy.



Figure 5. Cavitography hepatobronchial fistula passing through the right hemidiaphragm.



Figure 6. Ultrasound of hepatic Doppler control showing changes in hepatic echogenicity and absence of lesions.

surgery, there was a diaphragmatic defect which was repaired. The drainage catheter was removed without incidents and with a control ultrasound (Figure 6) with resolution of the lesion. The postoperative period was uneventful and the patient was discharged 4 weeks after hospitalization, completing treatment at home with levofloxacin plus metronidazole oral for 2 weeks.

Discussion

This is the case of a 50-year-old male with the diagnosis of hepatobronchial fistula secondary to a pyogenic hepatic abscess by *S. intermedius*.

Hepatobronchial fistula is an infrequent entity and is even rarer when secondary to a pyogenic abscess caused by *S. intermedius* [1]. This agent belongs to the *S. anginosus* group (also known as *S. milleri* group) a *Streptococcus* viridans subgroup that comprises three different species: *S. anginosus*, *S. intermedius*, and *Streptococcus* con*stellatus* [5]. The members of the *S. anginosus* group are known by their pathogenicity and tendency to form abscesses. The precise responsible virulent factors are not well understood, and it is believed that polysaccharide capsules pyogenic exotoxins and hydrolytic enzymes play an important role [6]. *Streptococcus intermedius* produces a unique form, intermedilysin, which is a cytolytic toxin specific for human cells that seems to be a virulent factor for the liver and other deep-seated abscesses [7].

The members of the *S. anginosus* group are part of the gastrointestinal tract microbiota [8]. They are able to cause a variety of abdominal infections such hepatic abscess, cholangitis, peritonitis, appendicitis, subphrenic abscess, pelvic abscess, abdominal wound infections, and postoperative infections after visceral trauma or elective surgery [8]. Furthermore, they can cause clinical infections in the oral cavity and endodontic, infections of head and neck, central nervous system, chest, as well as endocarditis, bacteremia, and others [8].

Clinical manifestations include fever, which is present in 90% of the cases and abdominal symptoms like pain and tenderness over the right upper quadrant in 50%-75% of the cases. About 50% of the patients with hepatic abscesses show hepatomegaly and jaundice. Other symptoms are nausea, vomiting, anorexia, weight loss, and malaise [9]. Laboratory findings include bilirubin and/or hepatic enzymes' elevation in up to 50% of the cases as well as raised alkaline phosphatase in 67%-90% of the cases [9]. Other alterations include leukocytosis, hypoalbuminemia, and anemia (normocytic and normochromic). These data were present in our patient.

Chest x-rays may show an elevated right hemidiaphragm, a right basal infiltrate, or a right pleural effusion in 25%-35% of the cases [10]. Our patient presented with these images on admission (Figure 1). The presence of a fistula can be demonstrated by the appearance of a "smoke pile" on a plain x-ray. Computed tomography allows the outlining of a hepatic abscess and the assessment of pulmonary involvement, being in some studies more sensitive for hepatic abscesses than echography (95% *vs.* 85%), respectively [10]. Blood cultures are positive in up to 50% of the cases [11]. Several cultures should be obtained, both aerobic and anaerobic, before antibiotic administration. All aspirated material should be Gramstained as well [11].

Zerem and Hadzic [12], in a study of 30 patients, observed that unilocular liver abscesses of less than 5 cm are acceptably treated by percutaneous drainage through catheter placement or needle aspiration. The latter can be repeated in up to 50% of the cases. The catheter should be left in place until drainage is minimal, usually 7 days [12]. On the other hand, abscesses larger than 5 cm are treated by catheter placement or surgery, as was the case in our patient, in whom closure of the diaphragmatic defect was also carried out. Surgery has a higher success rate [12].

In most cases of hepatobronchial fistula, surgical intervention is warranted and is considered a gold standard because of the aggressiveness of the disease, as well as the possibility of a rapid deterioration [13]. The surgical approach through thoracotomy has become the most commonly used. The surgical steps are as follows: adequate subcostal drainage under direct vision, safe closure of the diaphragmatic perforation, decortication of the involved pulmonary lobe, and lobectomy of the devitalized portion due to the fistulous tract [13]. Other procedures may involve decompression and drainage by radiologic endoscopic and interventionists means, which when combined, generally yield a favorable clinical outcome [13].

Liao et al. [14] reported the spontaneous closure of a fistula (up to 60% in posttraumatic ones) after subcutaneous drainage, while others stress that conservative measures have protracted the healing process, requiring a longterm drainage and risk of new sources of infection. Betalactamic antimicrobials are commonly used in infections by S. anginosus, with ceftriaxone being the preferred agent [15]. Further treatment should be governed by the culture results and antimicrobial susceptibility tests. Irrespective of the identified causative agent, treatment with antibiotics should be maintained for 4-6 weeks [16]. Patients who have had a good response to initial drainage should receive 2-4 weeks of parenteral treatment, whereas patients with an incomplete drainage should be treated parenterally for 4-6 weeks [16]. The remaining course can be completed with oral medication according to the culture results and susceptibility with levofloxacin 750 mg per day plus metronidazole [16].

Follow-up x-rays should be carried out in case of persisting clinical symptoms or if the drainage was not satisfactorily carried out [17].

Hepatobronchial fistulas have a mortality risk of up to 10.3% mainly because of surgical complications [17]. The operative mortality rate varies from 5% to 50% in underdeveloped countries to 2%-12% in developed countries and generally depends on underlying comorbidities, extension of the disease, and degree of the inflammatory process [17].

Conclusion

This is a rare case of hepatobronchial fistula secondary to *S. intermedius* infection; although benign in nature, it carries an unacceptable mortality risk of up to 10.3% as mentioned. With the introduction of less invasive techniques, the results have improved significantly. Current data suggest a hybrid approach (surgical plus interventional and radiological) individualized for this rare clinical entity, for each patient according to the etiology and severity of the disease.

List of Abbreviations

fl	femtoliter
gr/dl	grams per deciliter
K/ul	thousands per cubic millimeter
mEq/l	milliequivalent per liter
mg/dl	milligrams per deciliter

mg/l	milligrams per liter
mm/h	millimeters per hour
pg	picogram
S	Streptococcus
U/L	units per liter

Conflict of interest

The authors declare that there is no conflict of interest regarding the publication of this article.

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Consent for publication

Written consent was obtained from the patient.

Ethical approval

Ethical approval is not required at our institution to publish an anonymous case report.

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Summary of the case

1	Patient (gender, age)	Male, 50-year-old
2	Final diagnosis	Hepatic abscess by S. intermedius complicated with hepatobronchial fistula
3	Symptoms	Fever, long-standing bacteremia, and weight loss
4	Medications	Third generation cephalosporin and metronidazole for 6 weeks
5	Clinical procedure	A guided drainage by fluoroscopy with placement of a 12 F catheter was carried out obtaining purulent material. At surgery there was a diaphragmatic defect which was repaired.
6	Specialty	Internal medicine