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Case Report

Successful transcatheter arterial antimicrobial and steroid therapy for refractory liver abscess in chronic granulomatous disease: A case report and review of literature☆

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ABSTRACT

Hepatic abscess in chronic granulomatous disease (CGD) is very refractory and frequently requires multiple surgeries with frequent morbidities. Although surgical interventions are often required, patients are often not able to have surgery for various reasons. We present the case of a 21-year-old man with recurrent hepatic abscess in CGD. We could not provide surgical interventions due to the lack of a fluid cavity and the patient's refusal, and therefore we administered transcatheter arterial antimicrobial and steroid therapy. He did not have any exacerbation for more than 18 months after the final transcatheter treatment. This is the first reported case of successful transcatheter arterial antimicrobial and steroid therapy for refractory hepatic abscess in CGD. Although the patient's burden and medical cost were not inconsequential, this case shows that the transcatheter arterial antimicrobial and steroid therapy may be a treatment option for patients who are not candidates for surgical interventions.

1. Introduction

Chronic Granulomatous Disease (CGD) is a primary immunodeficiency disorder in which phagocytes are not able to destroy catalase-positive microorganisms, such as *S. aureus*, *E. coli*, *Klebsiella*, *Serratia*, and *Candida*, due to failure of the phagocytic cells' respiratory burst as a result of reduced levels of nicotinamide dinucleotide phosphate (NADPH) oxidase complex [1]. CGD is caused by missense, nonsense, frameshift, splice, or deletion mutations in the genes for p22phox, p40phox, p47phox, p67phox (autosomal CGD), or gp91phox (X-linked CGD) [2,3]. Patients frequently experience recurrent infections involving the lymph nodes, lungs, soft tissue, bones, and liver [4–7].

Hepatic abscess in patients with CGD is usually refractory. Treatment options include surgery, percutaneous drainage, and systemic steroid administration [7–9]. In some case reports, interferon gamma, granulocyte colony-stimulating factor, and percutaneous transhepatic alcoholization have been shown to be effective [10–13]. Although surgical interventions are chosen in many cases, some patients must choose non-surgical options since surgery is invasive with severe complications, and drainage is difficult without a fluid lesion. Despite recent advances in the treatment of CGD, it is usually very difficult to treat hepatic abscess in CGD patients [14]. We report here the first successful case of transcatheter arterial antimicrobial therapy for refractory liver abscess in CGD, and propose that it may be a promising treatment option.

2. Case report

A 21-year-old Japanese man who had been diagnosed with CGD presented with fever and right upper quadrant pain over the previous 4 days. He had been suspected of CGD because he had had a

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history of multiple admissions due to pneumonia, lymphadenitis, and skin infections since he was an infant, and because his senior brother had also been diagnosed with CGD. The genetic analysis revealed that he had a gp91phox deficiency. He has had prophylactic trimethoprim/sulfamethoxazole and itraconazole since then. The liver ultrasound at the time of the presentation showed a hypoechoic lesion with an irregular echo level inside. Computed tomography (CT) revealed a 9-cm liver mass with peripheral ring enhancement and a central hypodensity lesion without any liquid cavity in S4/8 (Fig. 1). Laboratory tests showed aspartate aminotransferase 21 U/L, alanine aminotransferase 19 U/L, alkaline phosphatase 359 U/L, total bilirubin 0.39 mg/dL, albumin 4.0 g/dL, erythrocyte sedimentation rate 71 mm/h and C-reactive protein 6.73 mg/dL. Aspiration was not performed, and no culture was taken due to the lack of a fluid lesion. He was admitted and treated with oral trimethoprim/sulfamethoxazole, intravenous ceftazidime, and fluconazole. After 7 weeks of treatment with intravenous antibiotic, he was discharged with improved clinical symptoms and inflammatory markers with some ultrasonographic improvements. After discharge, we followed him by ultrasonography with oral trimethoprim/sulfamethoxazole and itraconazole prophylaxis. However, symptoms of liver abscess recurred 8 months later. Magnetic Resonance Imaging (MRI) revealed a 5 cm mass with an irregular border in S4/8 (Fig. 2). He was again treated with intravenous ceftazidime for 6 weeks and oral minocycline for 3 weeks in addition to oral trimethoprim/sulfamethoxazole and itraconazole. Because the treatment regimen was not effective, it was switched to intravenous piperacillin/tazobactam (PIPC/TAZ) and fluconazole, but his radiographical findings and inflammatory markers did not resolve. We thoroughly discussed treatment options (surgical excision, drainage, and systemic steroids), which the patient rejected. Eventually, the patient and his family agreed to transcatheter arterial antimicrobial therapy as described below.

We moved a catheter through the hepatic artery, and confirmed a distended A4 branch (Fig. 3). Next, we administered antimicrobials into the right hepatic artery and A4 branch. After 5 series of transcatheter arterial PIPC/TAZ 4.5 g and 100 mg fluconazole, and 4 series of PIPC/TAZ and 125 mg (2 mg/kg) methylprednisolone (mPSL) therapy once a week to cover *S. aureus*, gram-negative bacilli, and Candida (the antimicrobial regimen is discussed later), the mass lesion subsided substantially. We added 3 additional series of PIPC/TAZ and mPSL every 2–4 weeks afterwards.

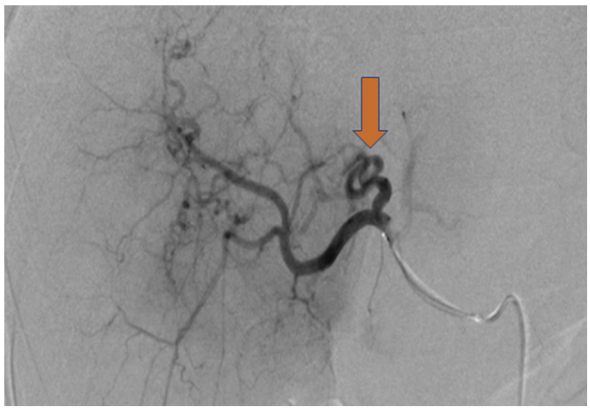

Fig. 3. Angiography showed a developed A4 branch of the hepatic artery (arrow).

Two months after we stopped transcatheter arterial antimicrobial therapy, mild inflammatory findings were seen on MRI without any clinical symptoms. We performed an additional 4 series of transcatheter arterial PIPC/TAZ and mPSL treatment once a week and then 2 series every other week. The radiological findings improved again (Fig. 4), and he did not show any exacerbation clinically or radiologically for more than 16 months after the final transcatheter therapy. The treatment schedule in the case is shown in Fig. 5.

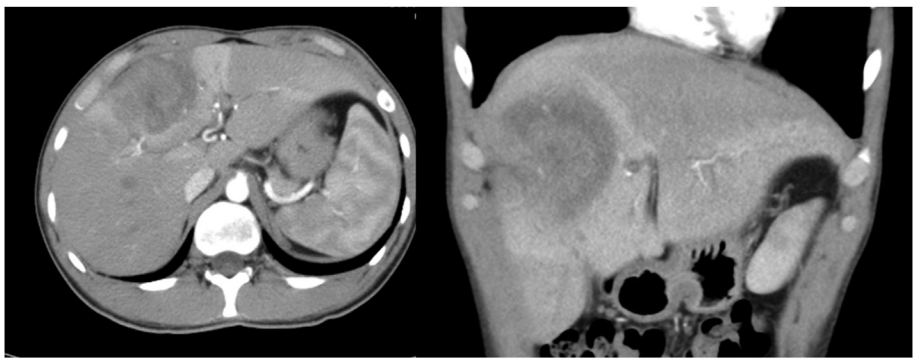

Fig. 1. Enhanced computed tomography (CT) showed abscess mainly in S4 and S8 with peripheral ring enhancement and central hypodensity measuring 9 cm in the craniocaudal dimension.

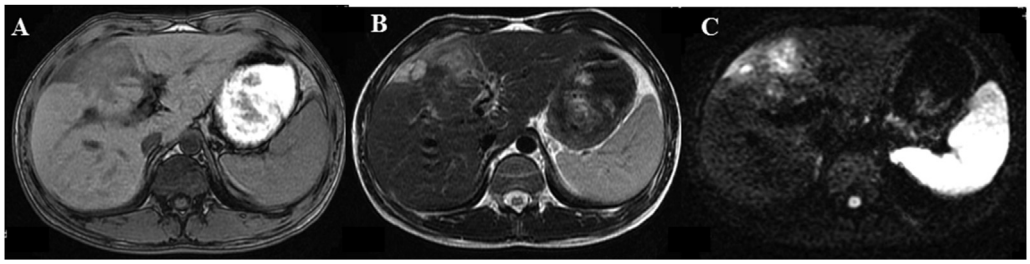

Fig. 2. Magnetic resonance imaging (MRI) revealed an irregular mass measuring 9 cm that showed hypointensity in T1(A), and hyperdensity in T2(B)/DWI(C).

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