CASE REPORT

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Bleeding hepatico-cutaneous fistula – an unusual complication of the percutaneous liver biopsy

Krvareća hepatično-kutana fistula – retka komplikacija perkutane biopsije jetre

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Abstract

Introduction. An arteriovenous fistula is one of the complications that can occur during percutaneous liver biopsy (PLB). Hepatic arteriovenous fistula with chronic bleeding from the puncture site on the skin is an extremely rare complication following PLB. **Case report.** We present a 35-year-old woman with secondary anemia caused by chronic bleeding at the site of a granuloma caused by a previous liver biopsy performed 7 years ago. The patient was examined and treated for several years due to anemic syndrome. The pathological communication between the right hepatic vein, the anterior sectional branch of the portal vein, and the posterior arterial sectional branch were detected on a computed tomography scan and proven by fistulography. Due to the failed embolization, a laparotomy was performed, where a tumor mass was found in the VI and VII segment of the liver, which communicates with the skin. The tumor mass was removed by atypical resection of the VI and VII liver segments. Due to hemorrhage, reexploration was performed, where the bleeding was found from the surface of the resected liver parenchyma. The patient was released for home treatment two weeks after the last operation. **Conclusion.** Although PLB is a safe procedure, a complication in the form of bleeding sometimes occurs but with spontaneous cessation. In the presented patient, there was a complicated intrahepatic arteriovenous-portal fistula with the formation of communication with the puncture site on the skin. That is the first published case of complications of this type after PLB.

Key words:
biopsy, fine-needle; diagnosis; hemorrhage; liver; surgical procedures, operative; vascular fistula.

Apstrakt

Uvod. Arteriovenska fistula predstavlja jednu od komplikacija koja se može javiti prilikom perkutane biopsije jetre (PBJ). Hepatična arteriovenska fistula sa hroničnim krvarenjem iz mesta uboda na koži predstavlja izuzetno retku komplikaciju PBJ. **Prikaz bolesnika.** Prikazujemo 35-godišnju ženu sa sekundarnom anemijom izazvanom hroničnim krvarenjem na mestu granuloma, izazvanog prethodnom biopsijom jetre rađenoj 7 godina ranije. Bolesnica je nekoliko godina ispitivana i lečena zbog anemijskog sindroma. Patološka komunikacija između desne hepatične vene, prednje sekcijske grane vene porte i zadnje arterijske sekcijske grane je detektovana kompjuterizovanom tomografijom, a patološka vaskularizacija u desnom lobusu jetre je dokazana fistulografijom. Zbog neuspele embolizacije, učinjena je laparotomija i nađena je tumorska masa u VI i VII segmentu jetre, koja je komunicirala sa kožom. Tumorska masa je odstranjena atipičnom resekcijom VI i VII segmenta jetre. Zbog akutne hemoragije, posle dva dana učinjena je reeksploracija, kada je nađeno krvarenje sa površine reseciranog parenhima jetre. Bolesnica je otpuštena na kućno lečenje dve nedelje nakon poslednje operacije. **Zaključak.** Iako je PBJ sigurna procedura, nekada dolazi do komplikacije u vidu krvarenja koje spontano prestaje. Kod prikazane bolesnice je došlo do komplikovane intrahepatične arteriovensko-portne fistule sa formiranjem komunikacije sa mestom uboda na koži. To je prvi opisani slučaj komplikacije te vrste nastale posle PBJ.

Ključne reči:
biopsija tankom iglom; dijagnoza; krvarenje; jetra; hirurgija, operativne procedure; fistula, vaskularna.

Introduction

Liver biopsy is one of the most frequent diagnostic tools not only for liver pathologies but also for other systemic diseases. In particular, percutaneous liver biopsies (PLB) constitute a large portion of all biopsies and are being performed at an increasing rate [1].

The overall incidence of complication after PLB rates from 1.2–6.8%, whereas post-biopsy bleeding represents the most serious complication and occurs in up to 10.9% of cas

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es [2, 3]. Arteriovenous fistula as a complication of PLB has been reported in 5.4%–52% of cases [4, 5]. However, in recent medical literature data, the occurrence of arteriovenous fistulas or arterioportal fistulas following PLB has only been reported in liver transplant recipients [6, 7]. The hepatic arteriovenous fistula, with its communication with skin on the site of skin puncture, has not been reported yet. We present a case with a delayed diagnosis of arteriovenous fistula as a complication of PLB manifested with chronic bleeding from the site of the puncture on the skin.

Case report

A 35-year-old female patient was admitted to our institution with secondary anemia caused by chronic bleeding from a small skin wound in the right upper region of the abdomen. On admission, red blood cells (RBC) was 2.4×10^{12}/L [normal range (nr) $3.8–5.8 \times 10^{9}$/L], hemoglobin of 59 g/L (nr 115–165 g/L), and hematocrit of 37% (nr 37–47%). All other laboratory parameters, including platelets, international normalized ratio (INR), partial thromboplastin time (PTT), liver enzymes, antinuclear antibodies, antineutrophil cytoplasmic antibodies, and parameters of inflammation, were in normal ranges.

It was reported in the patient's medical history that she underwent PLB with a fine needle 7 years ago due to slightly elevated liver enzymes. The histopathological finding suggested mild hepatitis with subsequent exclusions of hepatitis A, B, and C viral infections. After PLB, a granuloma, which occasionally bled, appeared at the puncture site on the skin. Due to the inability to heal, granuloma was treated with local excisions, antibiotics, and local skin transplantations in different healthcare institutions. Following these procedures, rib osteomyelitis has been diagnosed. After the failure of conservative treatment, the patient underwent resections of the right 11th rib and parts of the right rib arch in the region of the 7th and 8th ribs. The skin wound diminished over time but could not heal and close completely. Based on the suggestion of a hematologist and dermatologist, who suspected the existence of an undefined autoimmune disease, the patient received treatment with cyclosporine and pulse doses of methylprednisolone with subsequent corticosteroid therapy. This kind of treatment failed, and the skin wound kept occasionally bleeding for years. Blood transfusions and blood product administration have been indicated periodically to correct the patient's anemic syndrome. The diagnostic procedures indicated by hematologists revealed hypogammaglobulinemia, factor XIII deficiency (46%), and severe $CD4^{+}$ T lymphocyte suppression. Coagulopathy, platelets dysfunctions disorders, and Ehlers-Danlos syndrome have been excluded. Meanwhile, a pathological vascularization in the right liver lobe was observed during one of the computed tomography (CT) scans, and embolization was attempted during selective angiography of the liver. One week before admission to our hospital, the patient experienced increased bleeding from the skin wound.

Upon patient admission to our hospital, blood transfusion, and administration of supportive therapy, a body CT scan revealed pathological communication of the right hepatic vein, portal branch for the anterior liver section, and posterior artery sectoral branch (Figure 1).

Throughout the skin wound, the fistulography was performed and confirmed the pathological vascularization in the right liver lobe. On this radiological examination, a pathological cavity diameter of 40 × 10 mm was shown, which communicated with the arterial and venous system of the right liver lobe (Figure 2).

Due to a previously failed embolization and obvious late complication of PLB, it was concluded that the surgery would represent the safest treatment modality for the patient. During laparotomy, a tumor mass with a diameter of 9 × 6 cm was found in the VI and VII liver segments (Figures 3 and 4). The communication between skin, liver, and tumor mass was evident. The tumor mass was removed with atypical resection of the VI and VII liver segments (Figure 5). Histopathological findings of the specimen showed granulation tissue with chronic inflammation and slight portal and

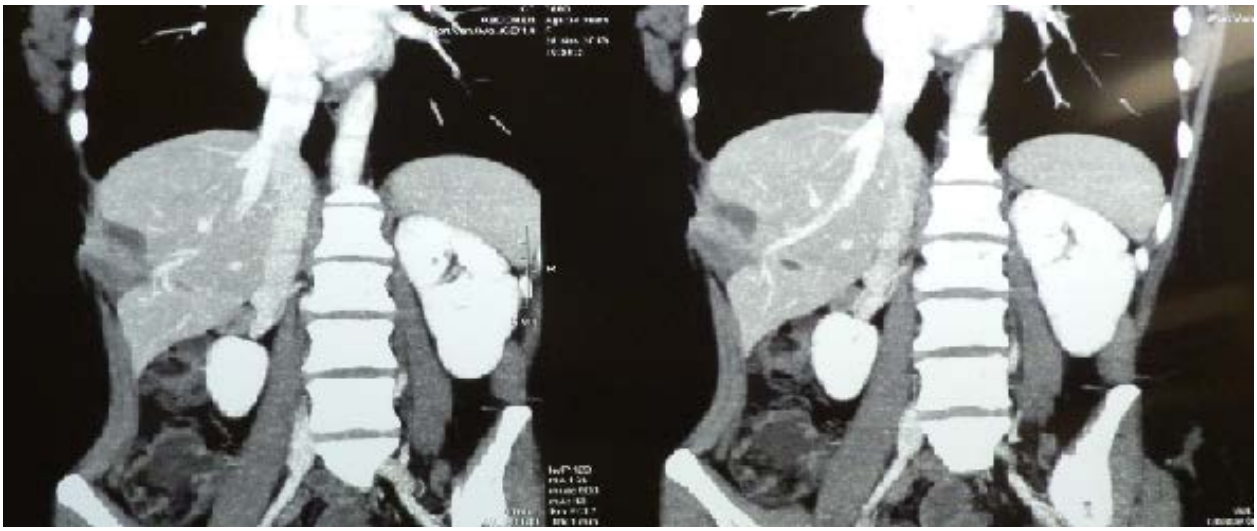

Fig. 1 – Abdominal computed tomography scan with pathological communication between arterial, venous, and portal venous system of the right liver lobe.

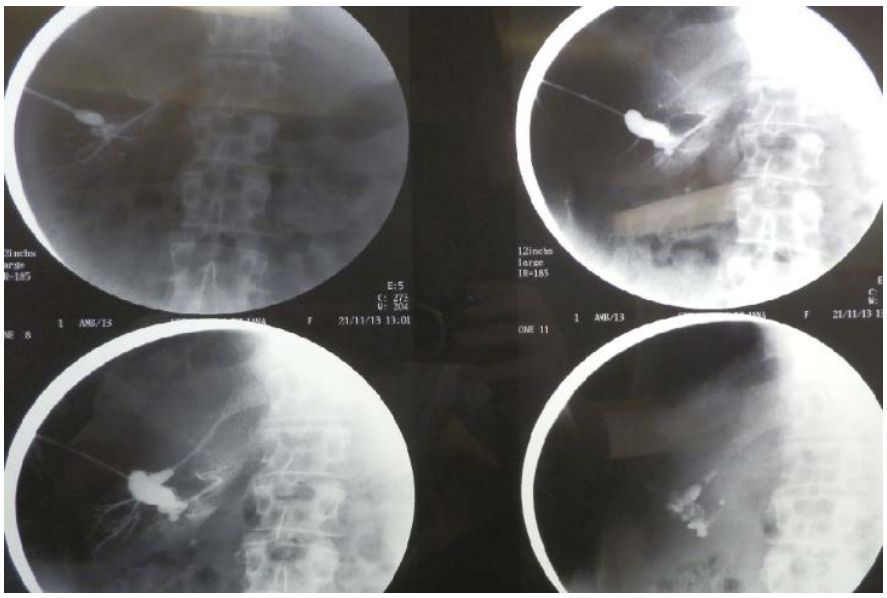

Fig. 2 – The fistulography through the wound of the skin and pathological communication of the arterial and venous system of the right liver lobe.

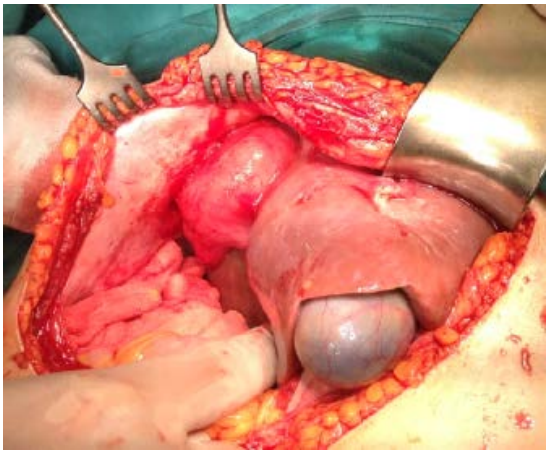

Fig. 3 – Intraoperative finding of tumor formation connected with the right liver lobe.

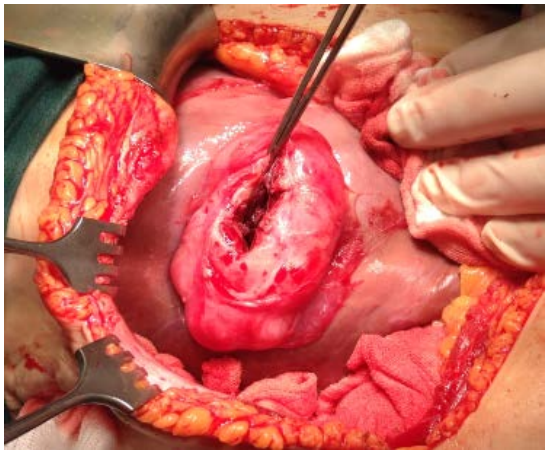

Fig. 4 – Tumor formation with central cavitation and fistula's tract to the liver parenchyma.

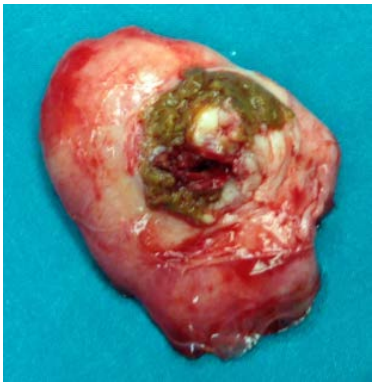

Fig. 5 – Tumor specimen after excision and atypical liver resection.

lobular inflammatory infiltrate. A reexploration was indicated on the second postoperative day due to acute hemorrhage. Bleeding was found at the cut parenchymal surface; afterward, liver segmentectomy was performed. The further postoperative course was uneventful, and the patient was discharged from the hospital two weeks after the first operation. During follow-ups, one and three months after hospital discharge, there were no signs of bleeding, and laboratory findings were normal.

Discussion

Complications after fine needle PLB are not frequent. The most frequent complication following PLB is postprocedural bleeding [2, 3]. Hepatico-cutaneous fistula as a complication of PLB has not been described yet. However, such a complication has been reported after percutaneous radiofrequency ablation of hepatocellular carcinoma [8]. Pathological communication between liver arteries and/or portal system and hepatic veins (arterio-venous fistula) may occur due to the rupture of artery aneurism into the portal system, in primary or secondary liver malignancies, or liver trauma [9]. Iatrogenic arteriovenous liver fistulas have been reported after liver biopsies, percutaneous transhepatic cholangiography, and following transhepatic biliary drainage [9–12]. The first literature data on arteriovenous liver fistula as a complication of PLB was described in 1967 [11]. After that report, several arteriovenous fistulas have been described, suggesting that the incidence of this kind of complication after liver biopsy is much higher than indicated by the scattered case reports [13]. Fortunately, these patients were asymptomatic, and shunts did not persist on the repeated angiography [13]. Other reports suggested that arteriovenous hepatic fistula, including arterioportal fistula, may be quiescent for a considerable time [13, 14]. In addition, a large multicentre study evaluated complications of PLB and did not find any arteriovenous hepatic fistula [12]. However, there are several described cases in the recent literature of intrahepatic arterioportal fistula following PLB [15–17]. Our patient has been asymptomatic for a certain time, with unspecific clinical signs. Perhaps that was the reason for misdiagnosis and inadequate treatment, including the fact that the majority of arteriovenous hepatic fistula could not be seen on single angiography [13, 14]. However, "over-treatment" in the presented patient in this perspective was unnecessary.

In proven hepatic arteriovenous (or arterioportal) fistula, the treatment modality includes embolization and surgical procedure [18, 19]. The specificity of the presented patient was the presence of not only communication between the hepatic artery and portal system but the communication with the right hepatic vein. Moreover, the site of PLB on the skin and skin wound has been transformed in the fistula tract, which communicates with the liver and intrahepatic arteriovenous fistula. Delayed diagnosis in our patient led to the "over-treatment" with multiple surgical procedures and chronic anemia. An accurate diagnosis of hepatico-cutaneous fistula in the presented patient was made with fistulography, one of the simplest diagnostic procedures. Fistulography should not be avoided in favor of imaging procedures such as magnetic resonance imaging and/or CT scan. However, a CT scan was necessary to confirm communication between the branch of the right hepatic artery, right portal, and hepatic vein. Bearing in mind the assumption that fistula embolization would be unsuccessful, our multidisciplinary team decided that surgical treatment was the only safe treatment with the goal of definitive care for our patients. Although parenchymal-sparing liver resections have shown comparable safety and efficacy compared with anatomic resections [20, 21], the patient experienced early postoperative bleeding after atypic liver resection.

Although the PLB is a safe and effective procedure, the rate of bleeding complications ranges between 0% and 25%, with the vast majority of studies reporting rates of bleeding under 2% [3]. If this complication occurs, the bleeding usually stops spontaneously, which was not the case with our patient. Furthermore, our patient had complicated intrahepatic fistula formation along with the communication of the skin site of the puncture. That is the first described case of unusual complication after PLB, and, as such, it is necessary to reconsider all possible consequences of PLB. In this term, with the adequate implementation of diagnostic procedures and timely treatment, it is possible to avoid any unnecessary and inappropriate approaches.

Conclusion

Arteriovenous intrahepatic fistula is a rare complication of PLB and may be complicated by chronic bleeding and a formation of the fistula tract with the site of puncture on the skin. The presented patient is the first one described in the literature with this complication following PLB. Fistulography may be a useful diagnostic tool, and the initial treatment should be embolization of the intrahepatic fistula. In case of failure of embolization, the liver resection should be performed by adapted and individualized liver resection (anatomic or parenchyma sparing) to the site of the intrahepatic fistula.

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