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SCIENTIFIC LETTER

Ectopic intrapelvic liver as an exceptional cause of female infertility: A case report[☆]



Hígado ectópico intrapélvico como causa excepcional de infertilidad femenina: Reporte de un caso

Liver position abnormalities or the presence of ectopic liver tissue are considered rare entities that are asymptomatic, incidental findings. Incidence is from 0.24 to 0.56%, according to case reports in laparoscopic surgery and autopsy results.¹

Reported sizes vary from millimeters to centimeters. Colan et al. classified them into four types: type 1, an ectopic liver not connected to the mother liver but to the bladder or abdominal ligaments; type 2, microscopic ectopic liver frequently found on the bladder wall; type 3, large accessory hepatic lobe joined to the mother liver by a pedicle; and type 4, small hepatic lobe joined to the mother liver.²

An ectopic liver implies exposure to factors that predispose to carcinogenesis. An incidence of hepatocellular carcinoma is reported in 30% of cases and is commonly related to vascular and biliary tree abnormalities.^{3,4}

There are only two case reports in the literature on an intrahepatic gallbladder in a pelvic liver. The two patients were diagnosed incidentally and had a history of omphalocele repair in infancy. In those cases, the right and left hepatic ducts joined to form the common hepatic duct that took a cranial direction to enter into the duodenal ampulla in the habitual location.^{5,6} We present herein the case of an ectopic liver with an intrahepatic biliary tree located in the pelvis that was found incidentally during the approach in managing female infertility.

Clinical case

A 23-year-old female patient under study for female infertility was referred from the gynecology department for a gastroenterology evaluation to rule out liver disease due to abnormal structures incidentally found in an abdominal ultrasound carried out as part of the female infertility protocol. The pertinent personal pathologic history included

abdominal wall repair due to gastroschisis in infancy and a spontaneous abortion at six weeks of gestation, two years prior. The rest of the past medical history was unremarkable. Physical examination revealed the presence of abdominal scarring along the mid-line, peristalsis, a slightly mobile, well-delineated, palpable mass in the hypogastrium, with smooth, regular edges, that was nonpainful upon palpation, with dull sounds upon percussion. Laboratory tests included liver function tests that reported total bilirubin 0.8 mg/dl, alkaline phosphatase 46 mg/dl, gamma-glutamyl transpeptidase (GGT) 75 mg/dl, aspartate aminotransferase (AST) 27 mg/dl, alanine aminotransferase (ALT) 36 mg/dl, and albumin 4.1 mg/dl.

The abdominal ultrasound report stated: “hepatic gland with notable increase in its dimensions, showing homogeneous echogenicity and echotexture and no intrahepatic or extrahepatic biliary tree dilation. The choledochus measures 2.5 mm and the portal vein has an interior diameter of 5 mm. The gallbladder has a thin wall, and its interior is anechoic; the pancreas cannot be evaluated due to overlapping of the bowel segments. The right kidney cannot be evaluated at this time, and only the right renal fossa was located. Diagnostic impression: grade III hepatomegaly, probable agenesis vs. right renal ectopia.”

Due to the data found in the initial imaging study, abdominal-pelvic angiotomography was ordered. In the simple phase, it showed absence of the liver in the right hypochondrium and location of the liver in the hypogastrium along the pelvis, with a maximum diameter of 187 mm and the presence of a Riedel lobe as an anatomic variant (Fig. 1). Both kidneys were displaced at the level of the diaphragm and the pancreas was rounded. In the arterial phase with maximum intensity projection (MIP) and volume rendering technique (VRT) reconstructions (Fig. 2), absence of the celiac trunk is shown, finding the solitary proximal emergence of the splenic artery and the distal emergence of the hepatic artery, which was subdivided into right and left arteries. The superior mesenteric artery had no alterations in its course or diameter. Magnetic cholangioresonance showed intrahepatic biliary integrity.

Conclusion

Hepatic tissue can migrate to different sites during embryogenesis. The errant liver is described as asymptomatic but has also been described as a cause of intra-abdominal bleeding and malignancy.¹ It is a rare condition resulting after the repair of congenital defects of the abdominal wall.

During the gynecologic approach, an ovarian hormone profile was carried out and was within normal parameters.

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Figure 1 Coronal view of the abdominal-pelvic tomography scan in the contrast-enhanced arterial phase showing the pelvic location of the liver, with the presence of a Riedel lobe.

The imaging studies revealed integrity of the ovaries and uterus. Given that no lesions were identified and there were no liver function test alterations, a liver biopsy was not performed, and we concluded that metabolic liver function was normal. The case has been followed for one year and the patient has not achieved pregnancy, nor has she presented with complications related to liver function.

After a review of the literature, we found case reports on patients diagnosed with gastroschisis and ectopic liver. However, we found no direct association of infertility with an errant liver, given that there are no case reports of said relation in the literature, even though infertility evaluation led to the discovery of that variant in our patient.

Many considerations need to be taken into account in relation to the present case. We are facing a little-known area with respect to the course a pregnancy, if viable, could take under these circumstances, as well as the effect the product could have on the liver. Thus, greater evidence on the topic is needed, as well as on the association between the two entities.

Ethical considerations

No experiments were conducted on animals or humans in relation to this research. The patient gave her informed consent to publish her clinical data. The present study was authorized by the ethics department of the *Hospital General De Occidente* for the review of the data and images obtained from the case report, following the current regulations on

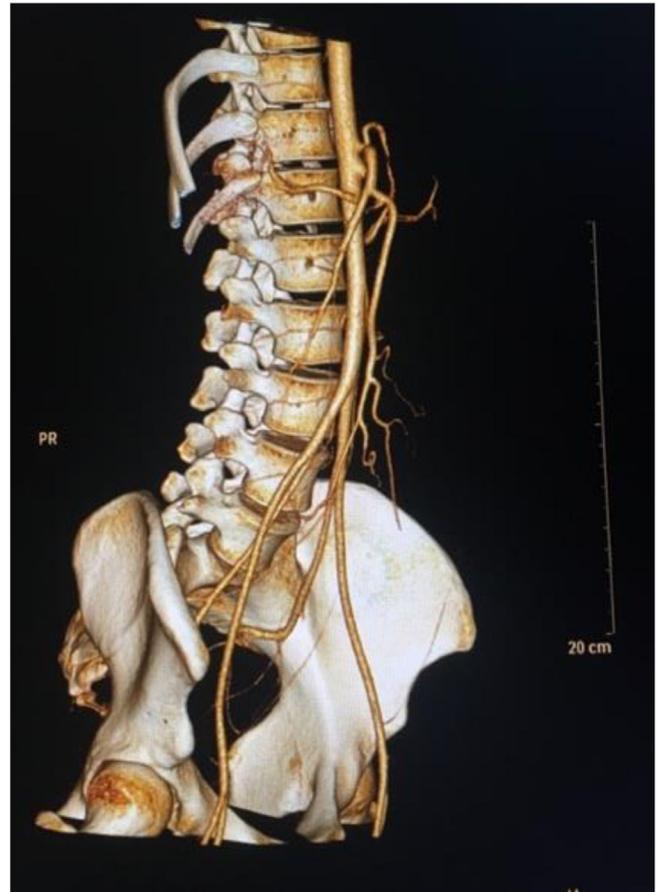


Figure 2 Computerized axial angiography image with maximum intensity projection (MIP) and 3D volume rendering technique (VRT) reconstructions in the arterial phase showing the characteristics of the celiac trunk and hepatic arteries passing into the pelvic region.

data protection and research on humans contemplated in the World Medical Association Declaration of Helsinki. No identifying data, images, or personal data of the patient were used. The patient authorized the presentation of this case report through informed consent.

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Conflict of interest

The authors declare that there is no conflict of interest.

References

1. Zonca P, Martinek L, Ihnat P, et al. Ectopic liver: different manifestations, one solution. *World J Gastroenterol.* 2013;19:6485–9, <http://dx.doi.org/10.3748/wjg.v19.i38.6485>.
2. Mathis R, Stodghill J, Shaver T, et al. Cholecystectomy of an intrahepatic gallbladder in an ectopic pelvic liver: a case report and review of the literature. *Case Rep Surg.* 2017;2017:3568768, <http://dx.doi.org/10.1155/2017/3568768>.

3. Puthenpurayil K, Blachar A, Ferris JV. Pelvic ectopia of the liver in an adult associated with omphalocele repair as a neonate. *Am J Roentgenol.* 2001;177:1113–5, <http://dx.doi.org/10.2214/ajr.177.5.1771113>.
4. Iber T, Rintala R. Intrapulmonary ectopic liver. *J Pediatr Surg.* 1999;34:1425–6, [http://dx.doi.org/10.1016/s0022-3468\(99\)90028-3](http://dx.doi.org/10.1016/s0022-3468(99)90028-3).
5. Sakarya A, Erhan Y, Aydede H, et al. Ectopic liver (choristoma) associated with the gallbladder encountered during laparoscopic cholecystectomy: a case report. *Surg Endosc.* 2002;16:1106, <http://dx.doi.org/10.1007/s00464-001-4251-5>.
6. Caygill CPJ, Gatenby PAC. Ectopic liver and hepatocarcinogenesis. *Eur J Gastroenterol Hepatol.* 2004;16:727–9, <http://dx.doi.org/10.1097/01.meg.0000131037.92864.df>.

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Ulcerative proctitis associated with lymphogranuloma venereum[☆]



Proctitis ulcerada asociada a linfogranuloma venéreo

The differential diagnosis of proctitis in men who have sex with men (MSM) tends to be difficult, given that it includes numerous infectious, inflammatory, and even traumatic causes. Lymphogranuloma venereum (LGV) is a sexually transmitted disease caused by *Chlamydia trachomatis* (*C. trachomatis*). It usually manifests first as an ulcerated, painless papule in the genitals, then as inguinal lymphadenopathy, and finally as distal proctitis.¹ In relation to late diagnosis, disease progression can result in severe complications, such as rectal stricture, obstruction, and perforation.^{1,2}

We present herein a 35-year-old patient, with a history of HIV diagnosed in 2012 in relation to Epstein Barr-associated meningitis, currently treated with highly effective antiretroviral therapy with raltegravir 400 mg and tenofovir/emtricitabine 300/200 mg. He had a CD4+ lymphocyte count of 248 cells and an undetectable viral load, and in addition, was identified as an asymptomatic carrier of hepatitis B infection.

He was admitted to the hospital due to clinical symptoms of intense pain in the rectoanal region of 3-month progression, painful defecation, straining, and tenesmus, associated with frequent episodes of rectal bleeding. In the systems review, the patient stated having occasional fever peaks, asthenia, adynamia, hyporexia, myalgias, and arthralgias.

Upon physical examination, the presence of pain in the hypogastrium, with no peritoneal irritation, stood out. The perianal evaluation revealed a deep posterior anal fissure, with marked edema of the anal canal. No adenopathies were palpated in the inguinal region, nor were there lesions on

the skin. Due to the patient's medical history, coinfection with other sexually transmitted diseases or opportunistic infections was ruled out. A VDRL test and IgM for Epstein-Barr virus were ordered, along with rectosigmoidoscopy, to evaluate the mucosa and anal canal and take biopsies.

The rectosigmoidoscopy revealed severe inflammatory changes and deep inflammatory ulcers with irregular edges that compromised the middle and distal rectum, with anal canal involvement (Fig. 1A-C). Biopsies were taken to identify the causal agent. Included in the pathology study was abundant lymphoplasmacytic infiltrate of the mucosa, with no viral cytopathic changes, with atrophy, and no dysplasia. Direct testing with techniques for mycobacteria, cytomegalovirus, and fungi was negative, as were the Thayer-Martin agar for *Neisseria gonorrhoeae* infection and the PCR for fungi and mycobacteria, and so PCR in *C. trachomatis* tissue was ordered. The VDRL serologic test for syphilis was reactive at 16 dilutions. Thus, in addition to treatment with 100 mg, every 12 h, of oral doxycycline, 2.4 million units of benzathine penicillin was administered weekly for 3 weeks.

Two weeks later, the patient was readmitted to the emergency service for abdominal pain, with scant rectal bleeding. A computed axial tomography scan and rectosigmoidoscopy were ordered. The first image ruled out perforation and associated collections. The rectosigmoidoscopy revealed significant improvement of the inflammatory changes, as well as ulcers in the process of healing (Fig. 2A-C). After symptom control, the patient was released and completed the treatment with doxycycline in 21 days.

Infectious proctitis in MSM, especially those with a history of HIV, is varied. The most frequent pathogens are *Neisseria gonorrhoeae*, *C. trachomatis*, the herpes simplex virus, and *Treponema pallidum*.^{2,3} In an Australian study, differences in the prevalence of the causal agents of infectious proctitis in MSM were found, according to their immune status.³ The most frequent causal agent was the herpes simplex virus in men that had a history of HIV infection, whereas LGV was the most frequent in men that were HIV-negative. No statistically significant differences related to HIV status regarding symptoms were found in that study. LGV procti-

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