International Journal of Nature and Life Sciences

IJNLS

https://dergipark.org.tr/tr/pub/ijnls

e-ISSN: 2602-2397 https://doi.org/10.47947/ijnls.1487910

Case Report

Rare Cause of Pelvic Mass: Hepatocellular Adenoma

Mücahit Ünal [1*], Şener Balas [2]

[1] Sakarya Yenikent State Hospital, General Surgery, Sakarya, Türkiye; https://orcid.org/0000-0001-9941-5166

[2] Dr. Abdurrahman Yurtaslan Oncology Research and Training Hospital, General Surgery, Ankara, Türkiye; https://orcid.org/0000-0002-9154-3179

*** Corresponding author:** mucahitunal78@gmail.com

Received: May 22, 2024

Accepted: May 28, 2024

Online Published: June 1, 2024

Citation:

Ünal, M., Balas, Ş. (2024). Rare cause of pelvic mass: hepatocellular adenoma. International Journal of Nature and Life Sciences, 8 (1), 61-66.

Abstract: Introduction: Hepatocellular adenoma (HA) is a rare solitary tumor originating from liver cells. There have been no reported cases with pelvic localization in the literature. In this article, we present a case of HA presenting with a pelvic mass.

Case: A 24-year-old male patient was found to have an intraabdominal mass on ultrasound during routine health screening. MRI (Magnetic Resonance Imaging) showed a solid mass lesion with pelvic localization, extending towards the upper abdomen, measuring approximately 16.5x8 cm, with sharp margins and well-defined borders, primarily suggestive of sarcomatous pathologies (rhabdomyosarcoma? / embryonal sarcoma?). Exploration revealed a mass resembling liver tissue attached to the left lobe of the liver via the falciform ligament, which was excised along with the ligament. The pathology of the specimen was consistent with adenoma. The patient was discharged on the third day postoperatively. Follow-up MRI at six months postoperatively did not reveal any issues.

Discussion and Conclusion: In patients with no significant etiological factors such as oral contraceptive use or anabolic androgen use, genetic studies and identification of possible environmental etiological factors from appropriate case series will contribute significantly to the literature. Our case demonstrated that even in male patients, HA should be considered in the differential diagnosis of intraabdominal masses.

Keywords: Hepatocellular adenoma, Pelvic mass, Sarcoma.

1. Introduction

Hepatocellular adenoma (HA hepatic adenoma, liver cell adenoma, etc.) is a rare benign tumor originating from hepatocytes in the liver. It predominantly affects young women who use oral contraceptives during their reproductive years. HA is commonly found in the right lobe of the liver and is typically solitary in 70-80% of cases. The size of these tumors can range from 1 cm to 30 cm. They are rarely found outside the liver (Grazioli et al. 2001).

The incidence of hepatocellular adenoma has shown a significant increase since the widespread use of oral contraceptives in the 80s and 90s (Sherlock,1975; Klatskin, 1977; Baum, et al., 1973). Another contributing factor to this rise in incidence is the increased utilization of imaging techniques in patients presenting with nonspecific abdominal pain (Grazioli et al., 2001).

HA is prominently associated with the use of oral contraceptives, anabolic androgen use, and glycogen storage diseases. Pregnancy and diabetes mellitus are also linked to HA as other etiological factors (Sherlock,1975; Klatskin, 1977; Baum, et al., 1973; Bioulac-Sage et al., 2017). HA can be seen in both type 1 and type 3 glycogen storage diseases, with reported rates of 51% and 25%, respectively. However, the exact mechanism of pathophysiology remains unclear (Fujiyama et al., 1990; Labrune et al., 1997; Talante et al., 1994). Aplastic anemia, Fanconi syndrome treatment, and the use of anabolic androgens by transgender adults or bodybuilders for muscle mass gain have been associated with the development of HA (Nakao et al., 2000; Touraine et al., 1993; Coombes et al., 1978).

HA can be clinically asymptomatic or present with episodic pain in the epigastrium or right hypochondrium. Sudden-onset severe abdominal pain accompanied by hypotension suggests intra-abdominal bleeding due to HA rupture, which can be fatal if left undiagnosed and untreated (Rubin and Mitchell, 1996; Bieze et al., 2014).

Liver function tests are often normal in laboratory evaluations. Alpha-fetoprotein (AFP) levels are typically within normal range except in cases of malignant transformation. While determining the exact risk of malignant transformation is challenging, it is reported to range between 8% and 13% in the literature. In cases of larger tumor size and elevated AFP levels, malignancy should be considered (Foster and Berman, 1994).

The diagnosis of hepatocellular adenoma (HA) is established through clinical evaluation, imaging techniques, and/or surgical resection. Due to the risk of bleeding, thick needle tissue biopsies or fine needle aspiration biopsies are not recommended.

2. Case

A 24-year-old caucasian male patient presented to an external healthcare facility for a routine check-up without any active complaints. An abdominal ultrasound revealed an intra-abdominal mass, prompting a referral to our hospital's general surgery clinic. The patient denied any history of medication use, and there was no history of smoking or alcohol consumption.

Upon physical examination, the patient appeared well, with no abnormalities noted during head and neck examination or cardiovascular and respiratory system assessment. Neurological examination revealed the patient to be conscious, alert, and cooperative, with a Glasgow Coma Scale score of 15. Cranial nerves were intact, and deep tendon reflexes were bilaterally equal and normal, with no sensory deficits detected. Examination of the urogenital system revealed bilaterally normal testes, with no penile lesions or inguinal lymphadenopathy noted. No inguinal hernias were detected. Gastrointestinal examination revealed a non-tender abdomen with no signs of defense or hepatosplenomegaly. However, both superficial and deep palpation revealed a mobile mass filling the pelvis.

Laboratory investigations revealed normal results for the following parameters: Complete blood count: Within normal limits. Erythrocyte sedimentation rate: 4 mm/hour (0-20 mm/hour). International Normalized Ratio (INR): 1.04 (0.8-1.22). Activated Partial Thromboplastin Time (APTT): 29.4 seconds (25.1-36.5 seconds).Hepatitis serology: Negative. Aspartate Aminotransferase (AST): 31 U/L (3-50 U/L). Alanine Aminotransferase (ALT): 34 U/L (3-50 U/L). Gamma-Glutamyl Transferase (GGT): 21 U/L (0-55 U/L). Total bilirubin: 0.64 mgdL-1 (0.3-1.2 mgdL-1). Direct bilirubin: 0.17 mgdL-1 (0-0.2 mgdL-1). Electrolytes: Within normal reference ranges.

An abdominal CT scan performed at an external center and rereported at our hospital's radiology clinic revealed a solid mass lesion located in the right lower quadrant, extending from the paraumbilical region to the midline of the abdomen, in proximity to the anterior abdominal wall. The lesion is approximately 14x8x12 cm and demonstrates heterogeneous contrast enhancement with scattered cystic areas. The mass displaced the right ureter, and caused partial obstruction. Additionally, there is mild compression on the inferior vena cava, affecting the superior mesenteric artery (SMA) and superior mesenteric vein (SMV) with mild displacement of adjacent bowel loops. The lesion exhibits well-defined margins without evidence of significant invasion into surrounding tissues.

Following the CT findings, the patient underwent abdominal MRI, which revealed a solid mass lesion located in the pelvic region with extension towards the upper abdomen. The lesion is approximately 16.5 x 8 cm and demonstrates sharp margins. It appears hypointense on T1-weighted images and intermediate to high signal intensity on T2-weighted images, with overall homogeneous signal intensity and scattered areas of heterogeneity. diffusion-weighted imaging shows restricted diffusion within the lesion. dynamic post-contrast series demonstrate diffuse homogeneous enhancement, primarily suggestive of sarcomatous pathology (Figure 1).

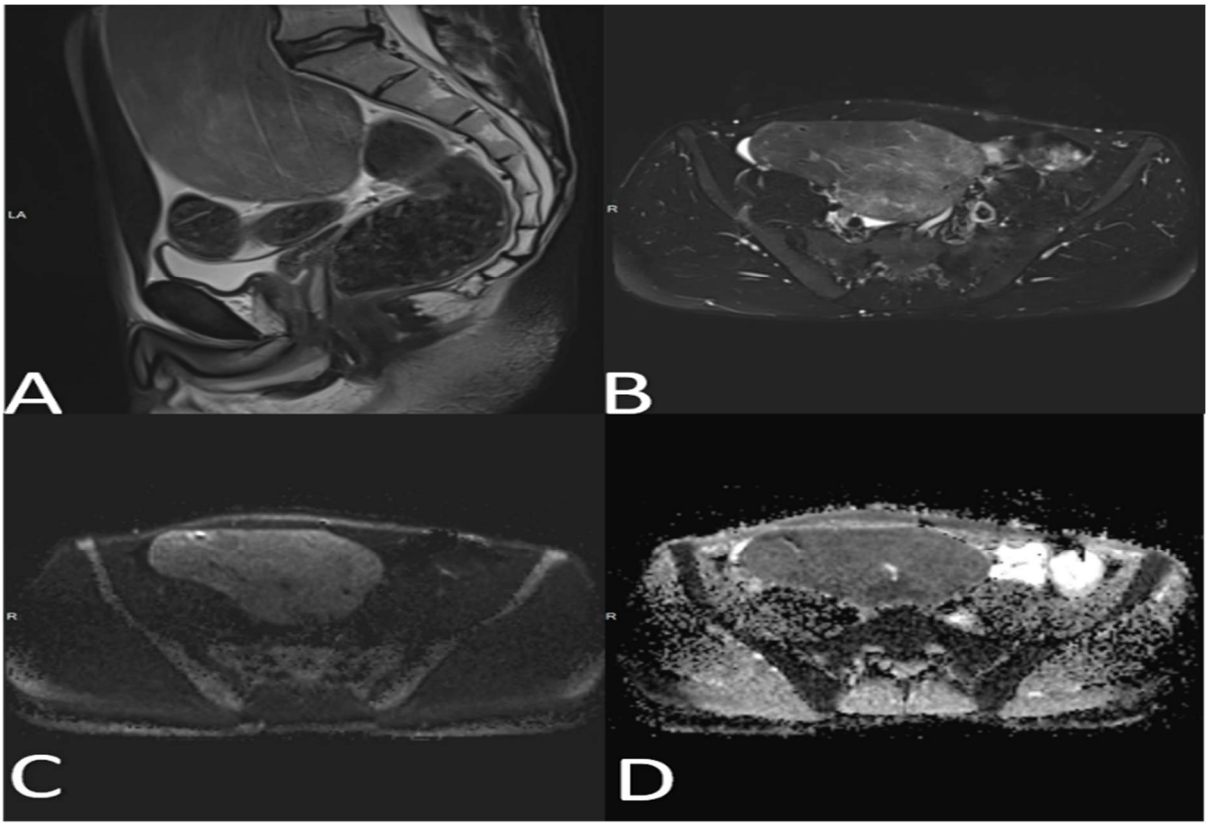

Figure 1. MRI Findings:A: Sagittal plane T2 weighted series B: T2 transverse plane C,D: Diffusion MRI images.

Surgical intervention was decided. Intraoperatively, it was observed that the mass originated from the inferomedial aspect of the left lobe of the liver, with vascular involvement noted. However, the mass was found to have no significant attachment to surrounding tissues apart from the hepatic pedicle (Figure 2). Complete excision of the mass and falciform ligament was achieved during the surgical procedure. The patient was discharged on the 4th postoperative day, following an uneventful recovery period.

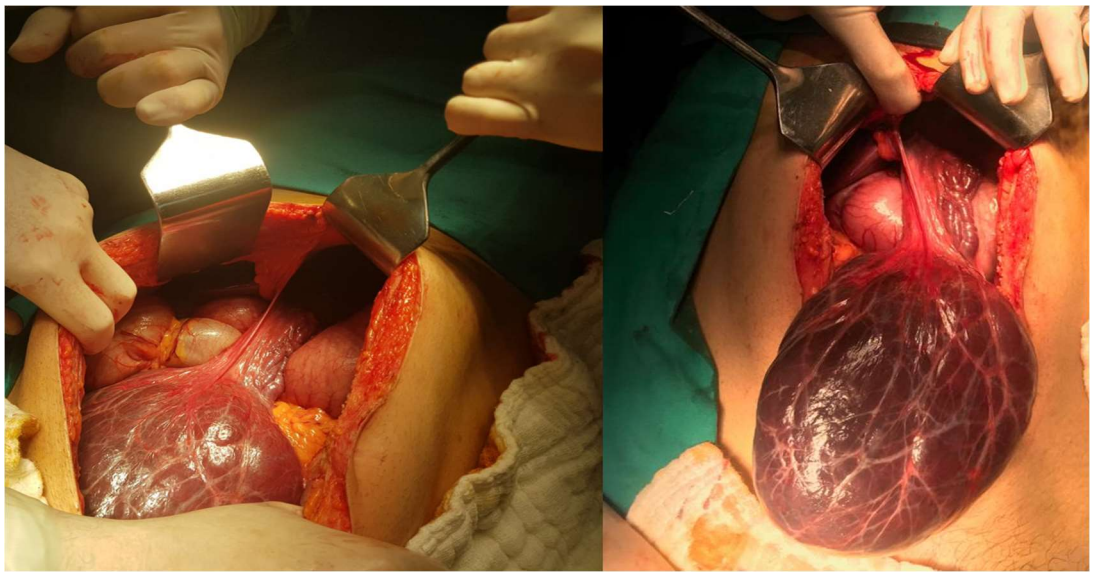

Figure 2. Intra operative images: hepatic adenoma attached to liver with falciform ligament.

Post-operative Pathology: Upon immunohistochemical staining, Beta-catenin and glypican were found to be negative. CD34 staining revealed vascularization particularly around vascular structures in the parenchyma. CK7 staining showed absence of bile ducts in most areas, with some areas containing well-developed small bile ducts and isolated hepatocytes showing focal bile duct metaplasia. No siderosis was observed upon iron staining, and no specific features were noted on PAS and dePAS staining. Copper staining showed no accumulation. Retikulin staining showed a generally preserved reticulin pattern. Ki67 proliferative index was determined to be 1% (Figure 3). Following the postoperative pathology evaluation, the pelvic mass was reported as hepatocellular adenoma.

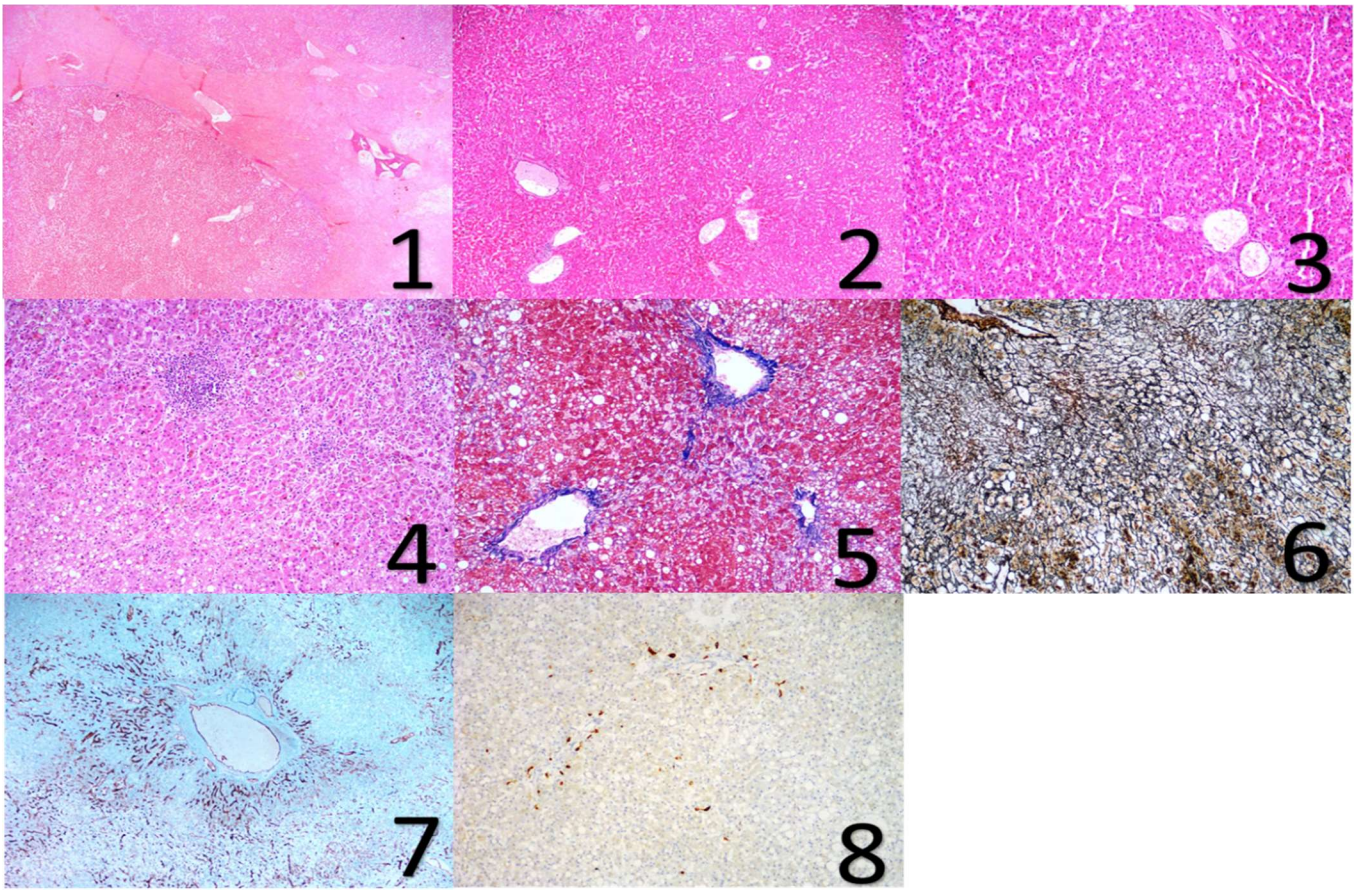

Figure 3. Pathology. 1,2,3,4: In H&E stained sections of mass lesion, in liver parenchyma resembling nearly normal architecture with hyalinized septa descending from the capsule, congested ectatic veins without bile ducts in portal areas distant from the capsule, and chronic inflammation were observed. (H&E X20, H&E X400, H&E X100, H&E X100) 5,6: These areas showing mild fibrosis and preserved focal collapse with a reticulin pattern. (Masson's trichrome X100, Reticulin X100) 7,8: Vascularization in vessels and surrounding tissue was observed in CD34 staining, and absence of bile ducts with staining in isolated hepatocytes was observed in CK7 staining. (CD34 X40; CK7 X100).

3. Discussion

In the approach to intra-abdominal masses with pelvic extension, hepatocellular adenoma should be considered, especially in male patients who typically lack gynecological etiologies. Additionally, in differential diagnosis, preoperative thick and thin needle biopsies should be carefully planned for suspected sarcomatous lesions, taking into account the patient's history, clinical laboratory findings, and imaging modalities.

Our case has no known etiological factors of hepatocellular adenoma. After discharge, the patient and his first-degree relatives were scheduled for colonoscopy, considering germline mutations in the APC and P53 genes cause to familial adenomatous polyposis coli. Referral for genetic testing was also made (Bala et al., 2004). In patients without clear etiological factors such as oral contraceptive use and anabolic androgen

use, identifying possible environmental etiological factors through genetic research and sufficient case series will contribute significantly to the literature.

Heterozygous mutation in the HNF-1a, gene resulting in MODY Type 3 (Maturity-Onset Diabetes of the Young), a form of adult-onset diabetes seen in young individuals, was found related to HA (Reznik et al., 2004; Bacq et al., 2003). Our patient was referred to the endocrinology clinic. Following investigations, no dysregulation of blood sugar was found. The patient, whose mother is diabetic, was planned for follow-up and monitoring. The relationship between hepatocellular adenoma and MODY Type 3 should be considered both in patients with hepatocellular adenoma and those with MODY Type 3.

4. Conclusion

In the differential diagnosis of pelvic masses, especially when sarcomatous lesions are considered, the importance of clean surgical margins in resection becomes evident once again. Our case highlights the necessity of considering hepatocellular adenoma even in male patients with such lesions. The surgical procedure performed not only provided a diagnosis but also constituted the most crucial stage of treatment for the patient.

Conflicts of Interests
Authors declare that there is no conflict of interests

Financial Disclosure
Authors declare no financial support.

Statement contribution of the authors
This study's experimentation, analysis and writing, etc. all steps were made by the authors.

References

1. Bacq, Y., Jacquemin, E., Balabaud, C., Jeannot, E., Scotto, B., Branchereau, S., Laurent, C., Bourlier, P., Pariente, D., de Muret, A., Fabre, M., Bioulac-Sage, P., & Zucman-Rossi, J. (2003). Familial liver adenomatosis associated with hepatocyte nuclear factor 1alpha inactivation. Gastroenterology, 125 (5), 1470-1475. https://doi.org/10.1016/j.gastro.2003.07.012
2. Bala, S., Wünsch, P. H., & Ballhausen, W. G. (1997). Childhood hepatocellular adenoma in familial adenomatous polyposis: mutations in adenomatous polyposis coli gene and p53. Gastroenterology, 112 (3), 919-922. https://doi.org/10.1053/gast.1997.v112.pm9041254
3. Baum, J. K., Bookstein, J. J., Holtz, F., & Klein, E. W. (1973). Possible association between benign hepatomas and oral contraceptives. Lancet (London, England), 2 (7835), 926-929. https://doi.org/10.1016/s0140-6736(73)92594-4
4. Bieze, M., Phoa, S. S., Verheij, J., van Lienden, K. P., & van Gulik, T. M. (2014). Risk factors for bleeding in hepatocellular adenoma. The British Journal of Surgery, 10 1(7), 847-855. https://doi.org/10.1002/bjs.9493
5. Bioulac-Sage, P., Sempoux, C., & Balabaud, C. (2017). Hepatocellular adenoma: Classification, variants and clinical relevance. Seminars in Diagnostic Pathology, 34 (2), 112-125. https://doi.org/10.1053/j.semdp.2016.12.007
6. Coombes, G. B., Reiser, J., Paradinas, F. J., & Burn, I. (1978). An androgen-associated hepatic adenoma in a trans-sexual. The British Journal of Surgery, 65 (12), 869-870. https://doi.org/10.1002/bjs.1800651212
7. Foster, J. H., & Berman, M. M. (1994). The malignant transformation of liver cell adenomas. Archives of Surgery (Chicago, Ill. : 1960), 129 (7), 712–717. https://doi.org/10.1001/archsurg.1994.01420310044007
8. Fujiyama, S., Sato, K., Sakai, M., Sato, T., Tashiro, S., & Arakawa, M. (1990). A case of type Ia glycogen storage disease complicated by hepatic adenoma. Hepato-Gastroenterology, 37 (4), 432-435.
9. Grazioli, L., Federle, M. P., Brancatelli, G., Ichikawa, T., Olivetti, L., & Blachar, A. (2001). Hepatic adenomas: imaging and pathologic findings. Radiographics: a review publication of the Radiological Society of North America, Inc, 21 (4), 877-894. https://doi.org/10.1148/radiographics.21.4.g01jl04877

10. Klatskin G. (1977). Hepatic tumors: possible relationship to use of oral contraceptives. Gastroenterology, 73 (2), 386-394.
11. Labrune, P., Trioche, P., Duvaltier, I., Chevalier, P., & Odièvre, M. (1997). Hepatocellular adenomas in glycogen storage disease type I and III: a series of 43 patients and review of the literature. Journal of Pediatric Gastroenterology and Nutrition, 24 (3), 276-279. https://doi.org/10.1097/00005176-199703000-00008
12. Nakao, A., Sakagami, K., Nakata, Y., Komazawa, K., Amimoto, T., Nakashima, K., Isozaki, H., Takakura, N., & Tanaka, N. (2000). Multiple hepatic adenomas caused by long-term administration of androgenic steroids for aplastic anemia in association with familial adenomatous polyposis. Journal of Gastroenterology, 35 (7), 557-562. https://doi.org/10.1007/s005350070081
13. Reznik, Y., Dao, T., Coutant, R., Chiche, L., Jeannot, E., Clauin, S., Rousselot, P., Fabre, M., Oberti, F., Fatome, A., Zucman-Rossi, J., & Bellanne-Chantelot, C. (2004). Hepatocyte nuclear factor-1 alpha gene inactivation: cosegregation between liver adenomatosis and diabetes phenotypes in two maturity-onset diabetes of the young (MODY)3 families. The Journal of Clinical Endocrinology and Metabolism, 89 (3), 1476-1480. https://doi.org/10.1210/jc.2003-031552
14. Rubin, R. A., & Mitchell, D. G. (1996). Evaluation of the solid hepatic mass. The Medical clinics of North America, 80 (5), 907-928. https://doi.org/10.1016/s0025-7125(05)70473-9
15. Sherlock S. (1975). Hepatic adenomas and oral contraceptives. Gut, 16 (9), 753-756. https://doi.org/10.1136/gut.16.9.753
16. Talente, G. M., Coleman, R. A., Alter, C., Baker, L., Brown, B. I., Cannon, R. A., Chen, Y. T., Crigler, J. F., Jr, Ferreira, P., Haworth, J. C., Herman, G. E., Issenman, R. M., Keating, J. P., Linde, R., Roe, T. F., Senior, B., & Wolfsdorf, J. I. (1994). Glycogen storage disease in adults. Annals of Internal Medicine, 120 (3), 218-226. https://doi.org/10.7326/0003-4819-120-3-199402010-00008
17. Touraine, R. L., Bertrand, Y., Foray, P., Gilly, J., & Philippe, N. (1993). Hepatic tumours during androgen therapy in Fanconi anaemia. European Journal of Pediatrics, 152 (8), 691-693. https://doi.org/10.1007/BF01955250