

World Journal of *Clinical Cases*

World J Clin Cases 2021 January 16; 9(2): 291-520



OPINION REVIEW

- 291 Continuity of cancer care in the era of COVID-19 pandemic: Role of social media in low- and middle-income countries
Yadav SK, Yadav N

REVIEW

- 296 Effect of a fever in viral infections – the ‘Goldilocks’ phenomenon?
Belon L, Skidmore P, Mehra R, Walter E
- 308 Overview of bile acid signaling in the cardiovascular system
Zhang R, Ma WQ, Fu MJ, Li J, Hu CH, Chen Y, Zhou MM, Gao ZJ, He YL

MINIREVIEWS

- 321 Gut microbiota and inflammatory bowel disease: The current status and perspectives
Zheng L, Wen XL

ORIGINAL ARTICLE**Retrospective Cohort Study**

- 334 Effective immune-inflammation index for ulcerative colitis and activity assessments
Zhang MH, Wang H, Wang HG, Wen X, Yang XZ

Retrospective Study

- 344 Risk factors associated with acute respiratory distress syndrome in COVID-19 patients outside Wuhan: A double-center retrospective cohort study of 197 cases in Hunan, China
Hu XS, Hu CH, Zhong P, Wen YJ, Chen XY

META-ANALYSIS

- 357 Limb length discrepancy after total knee arthroplasty: A systematic review and meta-analysis
Tripathy SK, Pradhan SS, Varghese P, Purudappa PP, Velagada S, Goyal T, Panda BB, Vanyambadi J

CASE REPORT

- 372 Lateral position intubation followed by endoscopic ultrasound-guided angiotherapy in acute esophageal variceal rupture: A case report
Wen TT, Liu ZL, Zeng M, Zhang Y, Cheng BL, Fang XM
- 379 Perioperative mortality of metastatic spinal disease with unknown primary: A case report and review of literature
Li XM, Jin LB

- 389** Massive gastric bleeding - perforation of pancreatic pseudocyst into the stomach: A case report and review of literature
Jin Z, Xiang YW, Liao QS, Yang XX, Wu HC, Tuo BG, Xie R
- 396** Natural history of inferior mesenteric arteriovenous malformation that led to ischemic colitis: A case report
Kimura Y, Hara T, Nagao R, Nakanishi T, Kawaguchi J, Tagami A, Ikeda T, Araki H, Tsurumi H
- 403** Coil embolization of arterioportal fistula complicated by gastrointestinal bleeding after Caesarian section: A case report
Stepanyan SA, Poghosyan T, Manukyan K, Hakobyan G, Hovhannisyanyan H, Safaryan H, Baghdasaryan E, Gemilyan M
- 410** Cholecystoduodenal fistula presenting with upper gastrointestinal bleeding: A case report
Park JM, Kang CD, Kim JH, Lee SH, Nam SJ, Park SC, Lee SJ, Lee S
- 416** Rare case of fecal impaction caused by a fecalith originating in a large colonic diverticulum: A case report
Tanabe H, Tanaka K, Goto M, Sato T, Sato K, Fujiya M, Okumura T
- 422** Intravitreal dexamethasone implant – a new treatment for idiopathic posterior scleritis: A case report
Zhao YJ, Zou YL, Lu Y, Tu MJ, You ZP
- 429** Inflammatory myofibroblastic tumor successfully treated with metformin: A case report and review of literature
Liang Y, Gao HX, Tian RC, Wang J, Shan YH, Zhang L, Xie CJ, Li JJ, Xu M, Gu S
- 436** Neonatal isovaleric acidemia in China: A case report and review of literature
Wu F, Fan SJ, Zhou XH
- 445** Malignant solitary fibrous tumor of the greater omentum: A case report and review of literature
Guo YC, Yao LY, Tian ZS, Shi B, Liu Y, Wang YY
- 457** Paratesticular liposarcoma: Two case reports
Zheng QG, Sun ZH, Chen JJ, Li JC, Huang XJ
- 463** Sinistral portal hypertension associated with pancreatic pseudocysts - ultrasonography findings: A case report
Chen BB, Mu PY, Lu JT, Wang G, Zhang R, Huang DD, Shen DH, Jiang TT
- 469** Epstein-Barr virus-associated monomorphic post-transplant lymphoproliferative disorder after pediatric kidney transplantation: A case report
Wang Z, Xu Y, Zhao J, Fu YX
- 476** Postoperative complications of concomitant fat embolism syndrome, pulmonary embolism and tympanic membrane perforation after tibiofibular fracture: A case report
Shao J, Kong DC, Zheng XH, Chen TN, Yang TY
- 482** Double-hit lymphoma (rearrangements of MYC, BCL-2) during pregnancy: A case report
Xie F, Zhang LH, Yue YQ, Gu LL, Wu F

- 489** Is sinusoidal obstructive syndrome a recurrent disease after liver transplantation? A case report
Liu Y, Sun LY, Zhu ZJ, Wei L, Qu W, Zeng ZG
- 496** Portal hypertension exacerbates intrahepatic portosystemic venous shunt and further induces refractory hepatic encephalopathy: A case report
Chang YH, Zhou XL, Jing D, Ni Z, Tang SH
- 502** Repair of a severe palm injury with anterolateral thigh and ilioinguinal flaps: A case report
Gong HY, Sun XG, Lu LJ, Liu PC, Yu X
- 509** Indirect inguinal hernia containing portosystemic shunt vessel: A case report
Yura M, Yo K, Hara A, Hayashi K, Tajima Y, Kaneko Y, Fujisaki H, Hirata A, Takano K, Hongo K, Yoneyama K, Nakagawa M
- 516** Recurrent inverted papilloma coexisted with skull base lymphoma: A case report
Hsu HJ, Huang CC, Chuang MT, Tien CH, Lee JS, Lee PH

ABOUT COVER

Editorial Board Member of *World Journal of Clinical Cases*, Dr. Mukul Vij is Senior Consultant Pathologist and Lab Director at Dr Rela Institute and Medical Center in Chennai, India (since 2018). Having received his MBBS degree from King George Medical College in 2004, Dr. Vij undertook postgraduate training at Sanjay Gandhi Postgraduate Institute of Medical Sciences, receiving his Master's degree in Pathology in 2008 and his PDCC certificate in Renal Pathology in 2009. After 2 years as senior resident, he became Assistant Professor in the Department of Pathology at Christian Medical College, Vellore (2011), moving on to Global Health City as Consultant Pathologist and then Head of the Pathology Department (2013). (L-Editor: Filipodia)

AIMS AND SCOPE

The primary aim of *World Journal of Clinical Cases* (*WJCC*, *World J Clin Cases*) is to provide scholars and readers from various fields of clinical medicine with a platform to publish high-quality clinical research articles and communicate their research findings online.

WJCC mainly publishes articles reporting research results and findings obtained in the field of clinical medicine and covering a wide range of topics, including case control studies, retrospective cohort studies, retrospective studies, clinical trials studies, observational studies, prospective studies, randomized controlled trials, randomized clinical trials, systematic reviews, meta-analysis, and case reports.

INDEXING/ABSTRACTING

The *WJCC* is now indexed in Science Citation Index Expanded (also known as SciSearch®), Journal Citation Reports/Science Edition, PubMed, and PubMed Central. The 2020 Edition of Journal Citation Reports® cites the 2019 impact factor (IF) for *WJCC* as 1.013; IF without journal self cites: 0.991; Ranking: 120 among 165 journals in medicine, general and internal; and Quartile category: Q3.

RESPONSIBLE EDITORS FOR THIS ISSUE

Production Editor: Jia-Hui Li; Production Department Director: Yu-Jie Ma; Editorial Office Director: Jin-Lai Wang.

NAME OF JOURNAL

World Journal of Clinical Cases

ISSN

ISSN 2307-8960 (online)

LAUNCH DATE

April 16, 2013

FREQUENCY

Thrice Monthly

EDITORS-IN-CHIEF

Dennis A Bloomfield, Sandro Vento, Bao-gan Peng

EDITORIAL BOARD MEMBERS

<https://www.wjnet.com/2307-8960/editorialboard.htm>

PUBLICATION DATE

January 16, 2021

COPYRIGHT

© 2021 Baishideng Publishing Group Inc

INSTRUCTIONS TO AUTHORS

<https://www.wjnet.com/bpg/gerinfo/204>

GUIDELINES FOR ETHICS DOCUMENTS

<https://www.wjnet.com/bpg/GerInfo/287>

GUIDELINES FOR NON-NATIVE SPEAKERS OF ENGLISH

<https://www.wjnet.com/bpg/gerinfo/240>

PUBLICATION ETHICS

<https://www.wjnet.com/bpg/GerInfo/288>

PUBLICATION MISCONDUCT

<https://www.wjnet.com/bpg/gerinfo/208>

ARTICLE PROCESSING CHARGE

<https://www.wjnet.com/bpg/gerinfo/242>

STEPS FOR SUBMITTING MANUSCRIPTS

<https://www.wjnet.com/bpg/GerInfo/239>

ONLINE SUBMISSION

<https://www.f6publishing.com>

Coil embolization of arterioportal fistula complicated by gastrointestinal bleeding after Caesarian section: A case report

Suren Agho Stepanyan, Tigran Poghosyan, Karen Manukyan, Gagik Hakobyan, Hayk Hovhannisyan, Hayk Safaryan, Elena Baghdasaryan, Manik Gemilyan

ORCID number: Suren Agho Stepanyan 0000-0003-1772-8738; Tigran Poghosyan 0000-0002-5932-1736; Karen Manukyan 0000-0002-2247-0404; Gagik Hakobyan 0000-0001-7876-9096; Hayk Hovhannisyan 0000-0002-0469-1692; Hayk Safaryan 0000-0002-9309-4709; Elena Baghdasaryan 0000-0002-2793-233X; Manik Gemilyan 0000-0003-0939-4889.

Author contributions: Stepanyan SA supervised the surgical care of patient, designed and drafted manuscript, edited text and figures, and organized manuscript language proofreading; Poghosyan T performed coil embolization and wrote manuscript text; Manukyan K performed endoscopic variceal band ligation and wrote manuscript text; Hakobyan G performed Gastroenterology consultation and wrote manuscript text; Hovhannisyan H performed abdominal drainage and wrote manuscript text; Safaryan H was attending surgeon of the patient and wrote manuscript text; Baghdasaryan E performed and interpreted CT imaging and wrote manuscript text; Gemilyan M performed Gastroenterology consultation and follow-up, wrote and edited manuscript text, organized references, formatted manuscript according to journal

Suren Agho Stepanyan, Hayk Safaryan, Department of Surgery No. 1, Yerevan State Medical University, Yerevan 0025, Armenia

Tigran Poghosyan, Department of Vascular Surgery, National Center of Oncology, Yerevan 0052, Armenia

Karen Manukyan, Department of Endoscopy, Mikaelyan University Hospital, Yerevan 0052, Armenia

Gagik Hakobyan, Manik Gemilyan, Department of Gastroenterology and Hepatology, Yerevan State Medical University, Yerevan 0025, Armenia

Hayk Hovhannisyan, Department of Surgery, Masis Medical Center, Masis 0801, Armenia

Hayk Safaryan, Department of Surgery, Mikaelyan Institute of Surgery, Yerevan 0052, Armenia

Elena Baghdasaryan, Department of Radiology, Armenia Medical Center, Yerevan 0078, Armenia

Corresponding author: Manik Gemilyan, MD, Assistant Professor, Department of Gastroenterology and Hepatology, Yerevan State Medical University, 2 Koryun st, Yerevan 0025, Armenia. mgemilyan@yahoo.co.uk

Abstract

BACKGROUND

Most intrahepatic arterioportal fistulae (IAPF) are acquired. The few cases of congenital fistulae are diagnosed in infants and children.

CASE SUMMARY

We report a 31-year-old female patient presenting with haematemesis and melena three weeks after delivering her second child. The patient had a 20-year history of abdominal distention and nausea. IAPF, along with splenomegaly and ascites, was found by Doppler sonography and confirmed by computed tomography angiography. The patient was treated with endovascular coil embolization, resulting in occlusion of the fistula.

CONCLUSION

This was an unusual case of possible congenital IAPF that manifested during a second pregnancy and was complicated by portal hypertension.

standards, and submitted manuscript; all authors have reviewed and agreed on the final version of manuscript.

Informed consent statement:

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Conflict-of-interest statement: The authors have no conflict of interest to disclose.

CARE Checklist (2016) statement:

The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

Open-Access: This article is an open-access article that was selected by an in-house editor and fully peer-reviewed by external reviewers. It is distributed in accordance with the Creative Commons Attribution NonCommercial (CC BY-NC 4.0) license, which permits others to distribute, remix, adapt, build upon this work non-commercially, and license their derivative works on different terms, provided the original work is properly cited and the use is non-commercial. See: <http://creativecommons.org/licenses/by-nc/4.0/>

Manuscript source: Unsolicited manuscript

Specialty type: Medicine, research and experimental

Country/Territory of origin:

Armenia

Peer-review report's scientific quality classification

Grade A (Excellent): A
Grade B (Very good): B
Grade C (Good): C
Grade D (Fair): 0
Grade E (Poor): 0

Received: July 9, 2020

Peer-review started: July 9, 2020

First decision: October 27, 2020

Revised: November 26, 2020

Accepted: December 10, 2020

Article in press: December 10, 2020

Key Words: Intrahepatic fistula; Coil embolization; Portal hypertension; Case report; Occlusion; Arterioportal fistula

©The Author(s) 2021. Published by Baishideng Publishing Group Inc. All rights reserved.

Core Tip: Our study describes an unusual case of possible congenital intrahepatic arterioportal fistula that manifested during a second pregnancy and was complicated by portal hypertension. Coil embolization of the fistula provides a less invasive and effective therapeutic option in this case. Only 35 cases of congenital intrahepatic arterioportal fistula were reported until 2015, and no case in the peripartum setting was described.

Citation: Stepanyan SA, Poghosyan T, Manukyan K, Hakobyan G, Hovhannisyanyan H, Safaryan H, Baghdasaryan E, Gemilyan M. Coil embolization of arterioportal fistula complicated by gastrointestinal bleeding after Caesarian section: A case report. *World J Clin Cases* 2021; 9(2): 403-409

URL: <https://www.wjgnet.com/2307-8960/full/v9/i2/403.htm>

DOI: <https://dx.doi.org/10.12998/wjcc.v9.i2.403>

INTRODUCTION

Intrahepatic fistulae are categorized as arteriovenous, arterioportal and portosystemic. These three types of fistulae represent anomalous communications between the hepatic artery, portal vein and hepatic vein. Arterioportal fistulae can be intrahepatic (IAPF) and extrahepatic (EAPF). Optimal management of these fistulae has not been established due to the rarity of the anomalies^[1-4].

IAPF was first reported by Gryboski *et al*^[5] in 1967, but its aetiology remains unclear. Most IAPFs are acquired; fewer than 10% of cases are congenital, and they are diagnosed mostly in infants and children^[6].

These fistulae have the potential to damage the portal vein and bowel and should be treated immediately after confirmation of the diagnosis. Congenital IAPF is typically caused by vascular malformation. Acquired IAPF and EAPF are more common and seen in hepatocellular carcinoma, ruptured hepatic artery aneurysm, post-liver biopsy, iatrogenic injury, blunt or penetrating trauma, interventional hepatic procedure, hepatectomy, Kasai portoenterostomy, segmental liver transplantation, cirrhosis, biliary atresia, and hereditary haemorrhagic telangiectasia^[7-11].

Interventional radiology has a key role in the treatment of congenital arterioportal fistulae (APF)^[12]. Congenital and acquired APFs require coil embolization or surgical intervention that involves hepatic artery ligation^[13,14]. The treatment of APF has shifted from surgical ligation or hepatic resection to an endovascular-first approach^[15].

Currently, the treatment of choice for most arterioportal fistulae is trans-arterial coil embolization. Symptomatic IAPFs and EAPFs can be occluded using steel coils, microcoils, polyvinyl alcohol and/or microspheres, and detachable balloons^[13,14]. Embolization of APF is technically feasible and effective and can be considered the first-choice therapy in selected patients^[16]. The curative embolization of congenital IAPF is usually achieved in unilobar lesions^[13,17,18]. For management of bilobar or complex lesions, a combination of embolization and surgery or a primary surgical approach is preferred^[19-21].

CASE PRESENTATION

Chief complaints

A 31-year-old woman was referred to our clinic from a district hospital after two episodes of haematemesis and melena requiring transfusions.

History of present illness

There was no history of trauma, iatrogenic causes or tumour. A month prior, the

Published online: January 16, 2021**P-Reviewer:** Coco D, Rong GH**S-Editor:** Gao CC**L-Editor:** A**P-Editor:** Wang LL

patient had experienced the first episode of gastrointestinal bleeding during the 38th wk of her second pregnancy. After 7 d of conservative therapy (i/v fluids and observation), a caesarean section was performed without any complications.

History of past illness

Of interest, the patient reported a history of periodical abdominal distention since she was 10 years old and nausea since she was 20 years old. An abdominal ultrasound two years prior had revealed hepatomegaly and splenomegaly.

Personal and family history

Personal and family history was unremarkable.

Physical examination

The physical examination on admission revealed abdominal distension. The patient's pulse rate was 110 beats/min, and the blood pressure was 90/60 mmHg.

Laboratory examinations

Laboratory results are shown in [Table 1](#). There was no encephalopathy detected. Viral and autoimmune hepatitis markers were negative.

Imaging examinations

As seen on Doppler ultrasound, arterial inflow to the fistula occurred *via* a hypertrophied right hepatic artery draining into a dilated venous varix of the right branch of the portal vein. The portal vein showed arterialization with reversal of flow. There was associated splenomegaly detected. Approximately 4000 mL of free fluid was detected in the abdomen.

Computed tomography (CT) angiography confirmed the ultrasound findings, showing a round irregular vascular malformation with a size of 28.0 mm × 27.1 mm × 22.4 mm in segment 5 of the liver above the gallbladder with a shunt between the right hepatic artery and right portal vein ([Figure 1A](#)). The right hepatic artery near the vascular malformation was 11.8 mm (normal 3-5 mm), the right portal vein was 23.6 mm (normal 4-6 mm) and the fistula was 12 mm in diameter. Upper gastrointestinal endoscopy showed multiple large oesophageal varices as the cause of bleeding.

FINAL DIAGNOSIS

Intrahepatic arterioportal fistula complicated by portal hypertension and acute upper gastrointestinal bleeding from esophageal varices.

TREATMENT

Endoscopic band ligation of oesophageal varices was performed. There was no bleeding after the procedure.

On the 11th day of hospitalization, the ascites became severe, and the abdominal cavity was drained with a 10-Fr pigtail catheter. On the 12th day of hospitalization, endovascular coil embolization of the arterioportal fistula was performed under local anaesthesia and mild intravenous sedation through the right femoral artery route using a 5F Cobra catheter (Terumo Corporation, Tokyo, Japan). Images were obtained of the celiac arteries, and further images were obtained of the right hepatic artery. The right hepatic artery was dilated, with the widest dilation of 12.2 mm. Continuation showed aneurysmal dilatation. The fistulous connection was 12 mm in diameter ([Figure 2A](#)). The decision was made to close the distal segments of the right hepatic artery by embolization with metallic coils (MRAY Cook medical, Bloomington, IN, United States), two coils of size 12-3 and six coils of size 8-5 ([Figure 2B](#)).

Following placement of coils, the flow gradually decreased. With stable vitals and an alert state of consciousness, the patient was transferred to the intensive care unit (ICU). The embolization procedure was completed without complications.

Table 1 Laboratory markers on admission and after treatment

Laboratory marker	On admission	Day 17 after coil embolization	Normal range	Unit
RBC	1.99	2.6	3.5-5.6	$\times 10^{12}/L$
Haemoglobin	53	92	112-152	g/L
WBC	10.3	7.5	4.0-9.0	$\times 10^9/L$
Platelets	113	214	180-400	$\times 10^9/L$
INR	2.12	1.9	0.9-1.5	-
Albumin	28.9	41	35-52	g/L
Bilirubin-total	12.7	1.17	0.1-1.2	mg/dL
AST	85	15	0.1-50.0	IU/L
ALT	37	17	0.1-41.0	IU/L
Creatinine	143	90	44-97	$\mu\text{m}/L$
Glucose	7.9	5.6	4.1-6.1	mm/L

RBC: Red blood cell; WBC: White blood cell; INR: International normalized ratio; AST: Aspartate aminotransferase; ALT: Alanine aminotransferase.

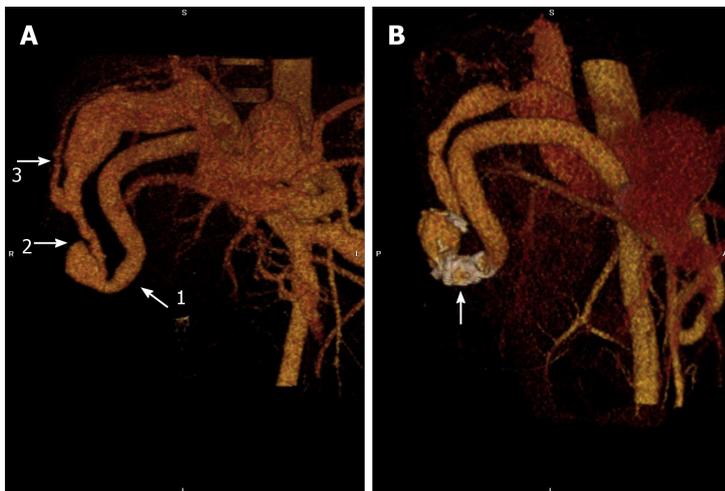


Figure 1 Contrast-enhanced computed tomography scan of the upper abdomen before and after coil embolization. A: Three-dimensional computed tomography reconstruction demonstrated the hypertrophied right hepatic artery (11.8 mm, white arrow 1), the large arterioportal fistula (12 mm in diameter, white arrow 2), and the right portal vein (23.6 mm, white arrow 3). There is a round irregular vascular malformation (28 mm \times 27.1 mm \times 22.4 mm) around the fistula; B: There was a reduction in the right hepatic artery (9.5 mm), arterioportal fistula (5.8 mm) and right portal vein (11.1 mm) after embolization. The site of coil embolization in the distal part of the right hepatic artery is visible (white arrow).

OUTCOME AND FOLLOW-UP

The patient's symptoms and laboratory markers improved after the procedure (Table 1). On day 20 after the procedure, a second upper gastrointestinal endoscopy was performed, revealing scars after ligation of variceal veins in the middle and lower portions of the oesophagus. There were varices (0.7 cm in diameter) near the Z-line, and a second ligation procedure was performed.

The postoperative outcome of the patient was excellent. At 1 mo of follow-up, the patient had no signs of portal hypertension or malabsorption. Compared to before the procedure, follow-up CT angiography 2 years later revealed significantly less blood flow from the hepatic artery towards the portal system and a reduction in the size of the liver and spleen. Along with a reduction in the size of the right hepatic artery (9.5 mm), fistula (5.8 mm) and right portal vein (11.1 mm), no migration of the coil was noted (Figure 1B).

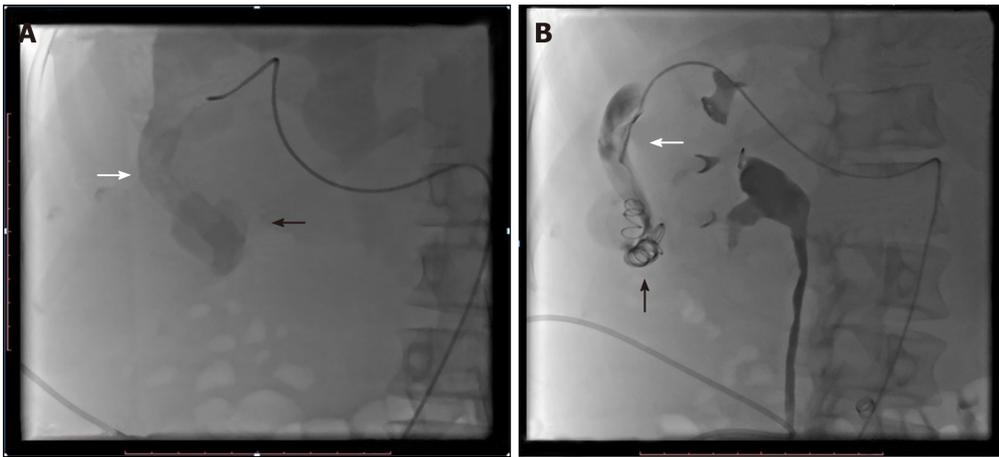


Figure 2 Selective right hepatic angiogram before and after coil embolization. A: Angiography demonstrated the hypertrophied right hepatic artery (white arrow) and vascular malformation (black arrow) around the arterioportal fistula. B: Angiography showed the catheter in the enlarged right hepatic artery (white arrow). The distal segments of the right hepatic artery were occluded using two 12-3 and six 8-5 coils (black arrow).

DISCUSSION

Acquired IAPF is more common than congenital IAPF and is seen in cases of disease and trauma of the liver and its vessels^[22]. Congenital arterioportal fistulae in adults are very rare. If hepatic arterioportal fistula is revealed in adulthood, it is difficult to establish the cause as congenital^[23].

In 2015, only 35 cases of congenital IAPF were reported, and no case in the peripartum setting was described Zhang *et al*^[11] (2015) presented 4 cases (65-year-old female, 33-year-old female, 74-year-old male, and 43-year-old male patients) of congenital IAPF. There was no history of trauma or liver disease. The clinical manifestations were haemorrhage from oesophageal varices in two cases, ascites in one case and encephalopathy in one case. In all cases, endovascular embolization was performed^[11].

Patients with IAPF may be asymptomatic or present with haemobilia, portal hypertension, or mesenteric steal syndrome^[15]. Our patient did not have a history of abdominal trauma or any previous procedures related to the abdominal cavity. The present case was considered to be congenital because of its symptomatic presentation in childhood (periodical abdominal distention and nausea) and lack of any secondary factors. While the patient had preserved synthetic function, the worsening of her condition was related to portal hypertension and bleeding from oesophageal varices during the 38th week of her second pregnancy.

Ward *et al*^[15] (2015) described a case of extrahepatic arterioportal fistula in a 40-year-old woman with a history of multiple pregnancies and no previous trauma. The fistula was formed between the left hepatic artery off the left gastric artery and the left portal vein. Endovascular embolization was performed with Amplatzer vascular plugs I. APF formation after visceral artery aneurysm rupture, often in the setting of pregnancy, is an alternative precipitating event and possibly the aetiology in this patient.

The present case is rare because clinical manifestations of portal hypertension started at the end of the second pregnancy. We could not identify cases in the literature describing the manifestation of IAPF during pregnancy.

Non-invasive examinations such as ultrasonography (US), colour Doppler US, CT or magnetic resonance imaging (MRI) are helpful in these cases. Catheter angiography is the confirmative diagnostic method for arterioportal fistulae^[7]. In our case, Doppler US, CT angiography and catheter angiography were performed before the procedure, and the type, site and size of the arterioportal fistula were detected. As bleeding from oesophageal varices is life-threatening, endoscopic ligation and haemostasis play an important role in the treatment.

The treatment of IAPF includes percutaneous trans-arterial embolization, implicated-hepatic artery surgical ligation, partial hepatectomy and liver transplantation. Interventional radiological treatment is considered the preferred procedure^[11]. Overall, slightly more than half of the cases of IAPFs are unilobar lesions. In this setting, curative embolization of intrahepatic arterioportal fistula is usually performed^[12,13,24].

There is a wide range of embolic agents and devices available, including ethanol, Gelfoam, steel coils, detachable balloons, a combination of micro-coils and N-butyl 2-cyanoacrylate (NBCA), Amplatzer vascular plugs I, and a combination of Guglielmi detachable coils with NBCA injection^[11,15].

In the present case, a single fistula between the right hepatic artery and the right portal vein was discovered. This finding allowed for successful treatment with coil embolization. Post-embolization angiogram showed occlusion of the arteriovenous fistula. In this report, we emphasize fistulae management and endovascular treatment. Embolization provides a less invasive and effective therapeutic option in this case.

CONCLUSION

IAPF is a rare but treatable cause of portal hypertension, and it may cause gastrointestinal bleeding and ascites. Embolization of IAPF is technically feasible and effective and can be considered the first-choice treatment in selected patients. This case highlights an unusual cause of IAPF that manifested during a second pregnancy and was complicated by portal hypertension.

ACKNOWLEDGEMENTS

The authors wish to extend their acknowledgements to Dr. Poghosyan A who performed the ultrasound and Doppler assessment of the patient, and to Dr. Shekherdimian S, MD (UCLA Mattel Children's Hospital, Los Angeles, United States) for reviewing this paper and giving valuable input.

REFERENCES

- 1 **Wu L**, Zhao L, Lu Y, He L, Hu X. Interventional embolization of congenital intrahepatic shunts in children. *Pediatr Radiol* 2016; **46**: 541-547 [PMID: 26637318 DOI: 10.1007/s00247-015-3497-3]
- 2 **Paley MR**, Farrant P, Kane P, Heaton ND, Howard ER, Karani JB. Developmental intrahepatic shunts of childhood: radiological features and management. *Eur Radiol* 1997; **7**: 1377-1382 [PMID: 9369502 DOI: 10.1007/s003300050304]
- 3 **Gallego C**, Miralles M, Marín C, Muyor P, González G, García-Hidalgo E. Congenital hepatic shunts. *Radiographics* 2004; **24**: 755-772 [PMID: 15143226 DOI: 10.1148/rg.243035046]
- 4 **Chandrasekharan R**, Kp S, Moorthy S, Kulkarni C. Traumatic hepatic arteriohepatic venous fistula managed with selective coil embolization: a case report. *BJR Case Rep* 2017; **3**: 20150512 [PMID: 30363278 DOI: 10.1259/bjrcr.20150512]
- 5 **Gryboski JD**, Clemett A. Congenital hepatic artery aneurysm with superior mesenteric artery insufficiency: a steal syndrome. *Pediatrics* 1967; **39**: 344-347 [PMID: 6018965]
- 6 **Vauthey JN**, Tomczak RJ, Helmberger T, Gertsch P, Forsmark C, Caridi J, Reed A, Langham MR Jr, Lauwers GY, Goffette P, Lerut J. The arterioportal fistula syndrome: clinicopathologic features, diagnosis, and therapy. *Gastroenterology* 1997; **113**: 1390-1401 [PMID: 9322535 DOI: 10.1053/gast.1997.v113.pm9322535]
- 7 **Eastridge BJ**, Minei JP. Intrahepatic arterioportal fistula after hepatic gunshot wound: a case report and review of the literature. *J Trauma* 1997; **43**: 523-526 [PMID: 9314320 DOI: 10.1097/00005373-199709000-00024]
- 8 **Gabriel S**, Maroney TP, Ringe BH. Hepatic artery-portal vein fistula formation after percutaneous liver biopsy in a living liver donor. *Transplant Proc* 2007; **39**: 1707-1709 [PMID: 17580227 DOI: 10.1016/j.transproceed.2007.03.099]
- 9 **Sachdeva R**, Yapor M, Schwersenz A, Mitty H, Norton K, Rosh J, Borcich A, Benkov K, LeLeiko NS. Massive variceal bleeding caused by a hepatic artery-portal vein fistula: a manifestation of hepatocellular carcinoma in a 12-year-old. *J Pediatr Gastroenterol Nutr* 1993; **16**: 468-471 [PMID: 8391075 DOI: 10.1097/00005176-199305000-00022]
- 10 **Tanaka H**, Iwai A, Sugimoto H, Yoshioka T, Sugimoto T. Intrahepatic arterioportal fistula after blunt hepatic trauma: case reports. *J Trauma* 1991; **31**: 143-146 [PMID: 1986122 DOI: 10.1097/00005373-199101000-00029]
- 11 **Zhang DY**, Weng SQ, Dong L, Shen XZ, Qu XD. Portal hypertension induced by congenital hepatic arterioportal fistula: report of four clinical cases and review of the literature. *World J Gastroenterol* 2015; **21**: 2229-2235 [PMID: 25717263 DOI: 10.3748/wjg.v21.i7.2229]
- 12 **Teplisky D**, Tincani EU, Lipsich J, Sierre S. Congenital arterioportal fistulas: radiological treatment and color Doppler US follow-up. *Pediatr Radiol* 2012; **42**: 1326-1332 [PMID: 22699373 DOI: 10.1007/s00247-012-2443-x]
- 13 **Raghuram L**, Korah IP, Jaya V, Athyal RP, Thomas A, Thomas G. Coil embolization of a solitary

- congenital intrahepatic hepatoportal fistula. *Abdom Imaging* 2001; **26**: 194-196 [PMID: 11178699 DOI: 10.1007/s002610000116]
- 14 **Zhang WG**, Li Z, Ding PX, Ren JZ, Ma J, Zhou PL, Wang ZG, Han XW. Endovascular treatment of an unusual primary arterioportal fistula complicated by cavernous transformation of the portal vein caused by portal thrombosis. *Ann Vasc Surg* 2014; **28**: 491.e5-491. e8 [PMID: 24368181 DOI: 10.1016/j.avsg.2012.10.032]
 - 15 **Ward TJ**, Marin ML, Lookstein RA. Embolization of a giant arterioportal fistula requiring multiple Amplatzer vascular plugs. *J Vasc Surg* 2015; **62**: 1636-1639 [PMID: 24840744 DOI: 10.1016/j.jvs.2014.04.031]
 - 16 **Nie L**, Luo XF, Li X. Gastrointestinal bleeding caused by extrahepatic arterioportal fistula associated with portal vein thrombosis. *World J Gastroenterol* 2012; **18**: 6501-6503 [PMID: 23197898 DOI: 10.3748/wjg.v18.i44.6501]
 - 17 **Kumar N**, de Goyet Jde V, Sharif K, McKiernan P, John P. Congenital, solitary, large, intrahepatic arterioportal fistula in a child: management and review of the literature. *Pediatr Radiol* 2003; **33**: 20-23 [PMID: 12497231 DOI: 10.1007/s00247-002-0764-x]
 - 18 **Ozyer U**, Kirbas I, Aytekin C, Hasdogan B. Coil embolization of a congenital intrahepatic arterioportal fistula: increasing experience in management. *Pediatr Radiol* 2008; **38**: 1253-1256 [PMID: 18690425 DOI: 10.1007/s00247-008-0957-z]
 - 19 **Guzman EA**, McCahill LE, Rogers FB. Arterioportal fistulas: introduction of a novel classification with therapeutic implications. *J Gastrointest Surg* 2006; **10**: 543-550 [PMID: 16627220 DOI: 10.1016/j.gassur.2005.06.022]
 - 20 **Sutcliffe R**, Mieli-Vergani G, Dhawan A, Corbally M, Karani J, Heaton N. A novel treatment of congenital hepatoportal arteriovenous fistula. *J Pediatr Surg* 2008; **43**: 571-573 [PMID: 18358306 DOI: 10.1016/j.jpedsurg.2005.07.005]
 - 21 **Tannuri AC**, Tannuri U, Lima FR, Ricardi LR, Leal AJ, da Silva MM. Congenital intrahepatic arterioportal fistula presenting as severe undernutrition and chronic watery diarrhea in a 2-year-old girl. *J Pediatr Surg* 2009; **44**: e19-e22 [PMID: 19853734 DOI: 10.1016/j.jpedsurg.2009.07.027]
 - 22 **Hirakawa M**, Nishie A, Asayama Y, Ishigami K, Ushijima Y, Fujita N, Honda H. Clinical outcomes of symptomatic arterioportal fistulas after transcatheter arterial embolization. *World J Radiol* 2013; **5**: 33-40 [PMID: 23494252 DOI: 10.4329/wjr.v5.i2.33]
 - 23 **Van Way CW 3rd**, Crane JM, Riddell DH, Foster JH. Arteriovenous fistula in the portal circulation. *Surgery* 1971; **70**: 876-890 [PMID: 5124667]
 - 24 **Norton SP**, Jacobson K, Moroz SP, Culham G, Ng V, Turner J, John P. The congenital intrahepatic arterioportal fistula syndrome: elucidation and proposed classification. *J Pediatr Gastroenterol Nutr* 2006; **43**: 248-255 [PMID: 16877994 DOI: 10.1097/01.mpg.0000221890.13630.ad]



Published by **Baishideng Publishing Group Inc**
7041 Koll Center Parkway, Suite 160, Pleasanton, CA 94566, USA
Telephone: +1-925-3991568
E-mail: bpgoffice@wjgnet.com
Help Desk: <https://www.f6publishing.com/helpdesk>
<https://www.wjgnet.com>

