



Contents lists available at ScienceDirect

## International Journal of Surgery Case Reports

journal homepage: [www.casereports.com](http://www.casereports.com)

## Therapeutic strategies for refractory variceal bleeding due to percutaneous liver biopsy: A case report

Chikwendu Ede<sup>a,\*</sup>, John Cantrell<sup>b</sup>, Jose Ramos<sup>b</sup><sup>a</sup> Department of Surgery, University of the Witwatersrand, 7 York Road, Parktown, Johannesburg, 2193, South Africa<sup>b</sup> Donald Gordon Medical Centre, University of the Witwatersrand, South Africa

## ARTICLE INFO

## Article history:

Received 27 February 2021

Received in revised form 27 March 2021

Accepted 28 March 2021

Available online 7 April 2021

## Keywords:

Arterioportal fistula

Liver biopsy

Varices

Bleeding

Case report

## ABSTRACT

**INTRODUCTION AND IMPORTANCE:** Variceal bleeding due to intrahepatic arterioportal fistula is an unusual complication of percutaneous liver biopsy. As majority of variceal bleeding are cirrhotic in origin, the rare occurrence of an acquired intrahepatic arterioportal fistula presents a therapeutic dilemma.

**CASE PRESENTATION:** We report the case of a 57-year-old female with refractory variceal bleeding that occurred six years after a percutaneous liver biopsy. As part of the workup for placement of Transjugular Intrahepatic Portosystemic Shunt, a computed tomography hepatic arteriography was performed. This revealed a large arterioportal fistula in left lobe of liver. Variceal bleeding was controlled following successful embolisation of the arterioportal fistula.

**CLINICAL DISCUSSION:** Persistent intrahepatic arterioportal fistula can result in portal hypertension and variceal bleeding. This is a rare complication of percutaneous liver biopsy that warrants consideration as an aetiology of portal hypertension with variceal bleeding. The therapeutic strategy for refractory bleeding due to intrahepatic arterioportal fistula is different from cirrhotic portal hypertension and requires trans-arterial embolisation of the fistula.

**CONCLUSION:** This case highlights the need to consider arterioportal fistula as an aetiology of portal hypertension as therapeutic strategy in refractory variceal bleeding is different from cirrhotic portal hypertension.

© 2021 The Author(s). Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

## 1. Introduction

An intrahepatic arterioportal fistula (IAPF) is an abnormal communication between a branch of hepatic artery and a portal venous radicle. An increasing role for interventional radiology has given rise to reports of acquired IAPF [1]. Most of these IAPFs are not symptomatic and will close spontaneously [2]. However, persistent IAPF can cause portal hypertension resulting in variceal bleeding. In a patient that has undergone a percutaneous liver biopsy and presents with variceal bleeding IAPF should be considered as an aetiology. Herein, we report the case of a patient with refractory variceal bleeding due to IAPF successfully managed by angiographic embolisation. This work has been reported in line with the SCARE criteria [3].

**Abbreviations:** IAPF, intrahepatic arterioportal fistula; CTHA, computed tomography hepatic arteriography; TIPS, Trans-jugular Intrahepatic Portosystemic Shunt.

\* Corresponding author.

**E-mail addresses:** [chikwendu.ede1@wits.ac.za](mailto:chikwendu.ede1@wits.ac.za) (C. Ede), [cantrell.j@hotmail.com](mailto:cantrell.j@hotmail.com) (J. Cantrell), [jramos@icon.co.za](mailto:jramos@icon.co.za) (J. Ramos).

<https://doi.org/10.1016/j.ijscr.2021.105852>

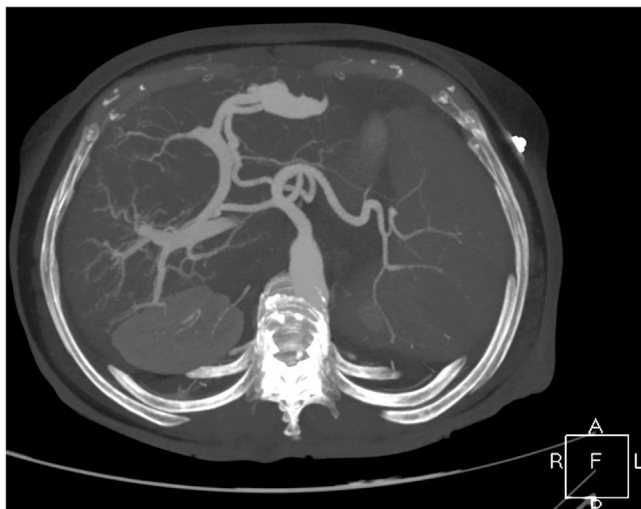
2210-2612/© 2021 The Author(s). Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

## 2. Case presentation

A 57-year-old female was transferred to the hepatobiliary unit at Donald Gordon Medical Centre, Johannesburg, South Africa on 4th December 2020. The patient was referred for Trans-jugular Intrahepatic Portosystemic Shunt (TIPS) having failed endoscopic management of bleeding fundal varices. Her history was that of percutaneous core needle liver biopsy performed six years previously, to investigate for chronic cholestasis. The tissue yield was determined to be adequate, and the histology revealed diffuse oedema of portal tracts with chronic inflammatory infiltrate rich in plasma cells. There was loss of bile ducts in a significant number of portal tracts. In the larger portal tracts where the original bile ducts were present, there was mild periductal fibrosis. In the presence of elevated anti-mitochondrial antibody, the morphologic features were considered to be in favour of primary biliary cholangitis. She had not required endoscopic retrograde cholangiopancreatography or percutaneous biliary intervention. On detailed questioning she admitted to moderate alcohol consumption for many years and had abstained completely in the last 3 months. She reported an episode of haematemesis and melaena 3 months previously but did not go to hospital due to Covid-19-related local lockdown restrictions. Haematemesis recurred 3 weeks prior to admission and she



**Fig. 1.** Arterial phase of dynamic computed tomography showing large arterioportal fistula in left lobe of the liver.

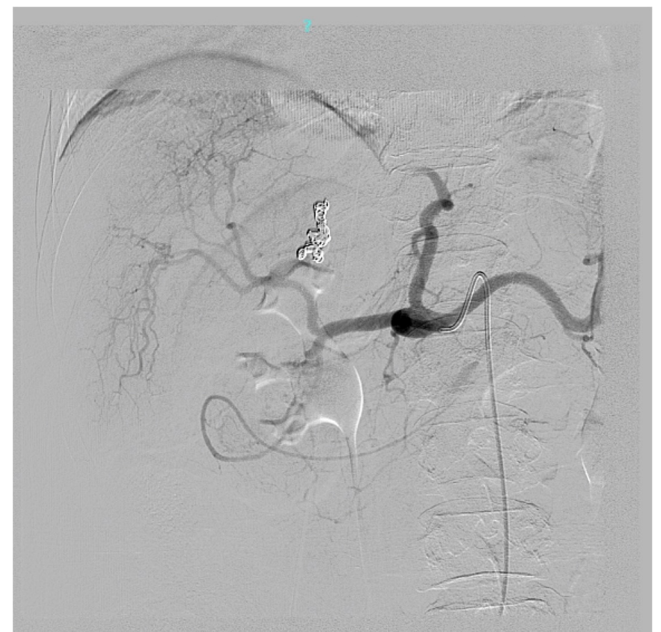


**Fig. 2.** Maximum intensity projection in arterial phase showing early enhancement of the portal vein.

underwent gastroscopy at another institution. Oesophageal varices were noted and about 5 bands applied (Six-shooter Multi-bands, Cook Medical, Europe). She did not require any blood transfusion. Upper gastrointestinal bleeding recurred 2 days prior to her admission to our institution and repeat gastroscopy by the referring gastroenterologist reported bleeding large fundal varices. No endoscopic intervention was performed. Two units of packed red cells were transfused, and octreotide infusion therapy commenced. She was then transferred for further management and possible TIPS. On arrival at our institution her haemodynamic status was noted to be normal. She was not jaundiced. Her abdomen was soft, non-tender with a palpable splenomegaly. Pertinent laboratory parameters were haemoglobin 10 g/dl; platelet 114 000 g/l; alkaline phosphatase 184 iu/l; gamma glutamyl transferase 58 iu/l; alanine aminotransferase 48 iu/l; aspartate aminotransferase 50 iu/l; international normalized ratio 1.1; and serum albumin 40 mg/dl. A computed tomography hepatic arteriography (CTHA) (CT 128; Koninklijke Philips N.V), that was performed as part of the TIPS workup, showed a large IAPF in the left lobe of the liver. It was supplied by a branch of the left hepatic artery and a branch of the hypertrophied left gastric artery, and it drained into the left portal vein (Figs. 1–3). There were no obvious features of hepatic cirrhosis on CTHA. She subsequently underwent catheter-directed angiogra-

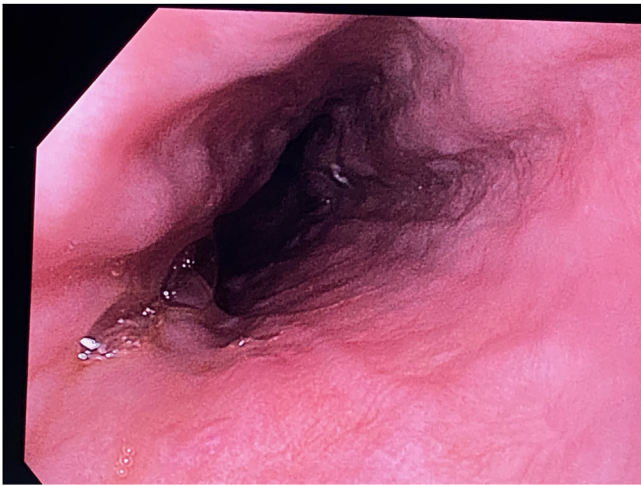


**Fig. 3.** Volume rendering showing an intrahepatic arterioportal fistula supplied by branches from left gastric artery and a branch of left hepatic artery.



**Fig. 4.** Completion angiogram showing microcoils successfully deployed and disappearance of the arterioportal fistula.

phy and embolisation in the interventional radiology department. This embolization was performed by an interventional radiologist. Firstly, cannulation of right femoral artery was performed under local anaesthesia and conscious sedation. The hepatic branches of the left gastric artery were localized and embolised, then proceeded to embolise distal left hepatic artery. All embolisation was performed through a microcatheter (Cantata; Cook Medical, Europe) with interlock detachable coils, 4 mm–6 mm in diameter, 8 mm–20 mm in length (Boston Scientific, Natick, MA, USA). Completion angiogram showed a good result with occlusion of the fistula and no opacification of the portal vein (Fig. 4). Post-intervention overnight observation in the intensive care unit was uneventful. A planned gastroscopy (EVIS videogastroscope; Olympus, Tokyo, Japan), was performed 72 h after embolisation by a hepatobiliary surgeon and showed residual grade II oesophageal varices extending for about



**Fig. 5.** Endoscopy following angiographic embolisation showing grade II oesophageal varices with no stigmata of recent bleed.

10 cm at distal oesophagus. There were no red wale signs and no cherry-red spots (Fig. 5). The stomach showed severe portal hypertensive gastropathy in proximal half, but no obvious fundal varices were noted. There were no other signs to suggest recent bleeding. Five bands (SmartBand™, Intelligent Endoscopy, NC, USA) were placed in the residual oesophageal varices. She was discharged the next day and has been scheduled for a repeat gastroscopy in six months. The patient has been followed up by telephone on two occasions since discharge and she reported no new event of upper gastrointestinal bleeding.

In this particular patient, it is felt that hepatic cirrhosis is unlikely, and that the portal hypertension was most likely due to the IAPF which led to variceal bleeding.

### 3. Discussion

Current practice of percutaneous liver biopsy requires image-guidance to achieve precision and reduce complications. Over 90% of complications following percutaneous liver biopsy occur within 24 h of the procedure, but development of IAPF is uncommon [4,5]. A review by Iwaki et al. in 2012 showed that only 30 cases of symptomatic IAPF have been reported [6]. Most IAPFs are small, asymptomatic, and appear to close spontaneously within a month [2,4]. Long-term events like portal hypertension and variceal bleeding are rare. Variceal bleeding is a sequela of portal hypertension that is said to occur by two mechanisms: either an increase in intra-hepatic vascular resistance or increase in portal blood flow [7]. In this index case, a high pressure hepatic arterial blood is directed into the portal circulation bypassing the hepatic sinusoids. This creates an increase in hepato-venous pressure gradients that results in development of varices, and subsequent bleeding.

Variceal bleeding is the most dreaded complication of portal hypertension with six-week mortality that approaches 25% in the setting of liver cirrhosis [8]. Most cases of variceal bleeding are attributable to cirrhotic portal hypertension, and IAPF is not usually considered an aetiology. Hence a high index of suspicion is required. A history of previous hepatic intervention or abdominal trauma is suggestive. The findings of thrill or bruit in the epigastric region on physical examination are helpful. Management guidelines for variceal bleeding due to cirrhotic portal hypertension are applied across board irrespective of the aetiology of variceal bleeding. It entails initial resuscitation, pharmacologic therapy combined with endoscopic intervention in the acute setting [7]. This strategy will provide effective control of variceal

bleeding in 80% of cases irrespective of aetiology [6]. However, in individuals with IAPF, endoscopic therapy alone is not sufficient, angiographic embolisation is recommended as definitive therapy to control variceal bleeding [1,4,6,9]. Endovascular treatment by way of catheter-directed trans-arterial embolisation is an attractive option as it is minimally invasive. The goal of treatment is to occlude fistula-feeding vessel, and thereby reduce the high portal blood flow. Embolisation may be achieved with coils as in the index case, Gelfoam sponge, detachable balloon, or liquid embolic agents. Procedure-related complications include non-target embolisation, hepatic infarction, and liver abscess. In situations where endovascular treatment fails to control variceal bleeding, alternative interventions have been recommended. Surgical ligation of a second or third order branch of hepatic artery is an option that is achieved through division of liver parenchyma to reach the area of the fistula. This procedure is technically challenging and not usually practiced. Hence in terms of operative management, resection of the part of the liver that contains the fistula may be a better option [5,10]. Liver transplantation has been performed as salvage therapy in refractory cases [2,9]. However, the availability of donor organs for liver transplantation is a huge limitation.

### 4. Conclusion

Although percutaneous liver biopsy is relatively safe, its attendant complications though rare may be life threatening. Given that angiography is not performed routinely after a percutaneous liver biopsy, this case highlights the need to consider IAPF, an aetiology of portal hypertension as therapeutic strategy in refractory variceal bleeding is unique. Future studies should evaluate risk factors for development of IAPF in patients that are scheduled for hepatic interventional procedures.

### Declaration of Competing Interest

None to declare.

### Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

### Ethical approval

Ethics approval obtained from Human Research Ethics Committee, University of the Witwatersrand, South Africa. Ethics number: M2011162.

### Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

### Author contribution

CJ conceptualized the idea and wrote the manuscript. JA and JC made changes to the manuscript. All authors read and approved the final version of the manuscript.

### Registration of research studies

Not Applicable.



**Guarantor**

Chikwendu Ede; FCS (SA), MMed.

**Provenance and peer review**

Not commissioned, externally peer reviewed.

**Acknowledgements**

None.

**References**

- [1] A. Kumar, C.K. Ahuja, S. Vyas, N. Kalra, N. Khandelwal, Y. Chawla, et al., Hepatic arteriovenous fistulae: role of interventional radiology, *Dig. Dis. Sci.* 57 (October (10)) (2012) 2703–2712.
- [2] J.N. Vauthey, R.J. Tomczak, T. Helmberger, P. Gertsch, C. Forsmark, J. Caridi, et al., The arterioportal fistula syndrome: clinicopathologic features, diagnosis, and therapy, *Gastroenterology* 113 (October (4)) (1997) 1390–1401.
- [3] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, A. Kerwan, A. Thoma, et al., The SCARE 2020 guideline: updating consensus Surgical CAse REport (SCARE) guidelines, *Int. J. Surg.* 84 (December) (2020) 226–230.
- [4] E.A. Guzman, L.E. McCahill, F.B. Rogers, Arterioportal fistulas: introduction of a novel classification with therapeutic implications, *J. Gastrointest. Surg.* 10 (April (4)) (2006) 543–550.
- [5] F. Piccinino, E. Sagnelli, G. Pasquale, G. Giusti, Complications following percutaneous liver biopsy. A multicentre retrospective study on 68,276 biopsies, *J. Hepatol.* 2 (2) (1986) 165–173.
- [6] T. Iwaki, H. Miyatani, Y. Yoshida, K. Matsuura, Y. Suminaga, Gastric variceal bleeding caused by an intrahepatic arterioportal fistula that formed after liver biopsy: a case report and review of the literature, *Clin. J. Gastroenterol.* 5 (April (2)) (2012) 101–107.
- [7] G. Garcia-Tsao, J.G. Abraldes, A. Berzigotti, J. Bosch, Portal hypertensive bleeding in cirrhosis: risk stratification, diagnosis, and management: 2016 practice guidance by the American Association for the study of liver diseases, *Hepatology* 65 (1) (2017) 310–335.
- [8] B.E. Fortune, G. Garcia-Tsao, M. Ciarleglio, Y. Deng, M.B. Fallon, S. Sigal, et al., Child-Turcotte-Pugh class is best at stratifying risk in variceal hemorrhage: analysis of a US multicenter prospective study, *J. Clin. Gastroenterol.* 51 (May/June (5)) (2017) 446–453.
- [9] X. Huang, W. Zhang, S. Chen, C. Liu, R. Sheng, F. Li, et al., Therapeutic evaluation of arterio-portal fistula-related gastroesophageal variceal bleeding, *Oncotarget* 8 (September (39)) (2017) 66584–66592.
- [10] H. Takata, H. Makino, T. Yokoyama, H. Maruyama, A. Hirakata, J. Ueda, et al., Successful surgical treatment for intrahepatic arterioportal fistula with severe portal hypertension: a case report, *Surg. Case Rep.* 5 (April (1)) (2019) 67.

**Open Access**

This article is published Open Access at [sciencedirect.com](https://www.sciencedirect.com). It is distributed under the [IJSCR Supplemental terms and conditions](#), which permits unrestricted non commercial use, distribution, and reproduction in any medium, provided the original authors and source are credited.