Intrapancreatic abscess due to arteriovenous malformation involving the entire pancreas: A case report and review of the literature

Ahmad Almalki
General Surgery, Aseer Central Hospital, Abha, Saudi Arabia

ABSTRACT

INTRODUCTION: The described case is a patient with pancreatic arteriovenous malformation involving the entire pancreas and complicated with intrapancreatic abscess.

PRESENTATION OF CASE: A 47 years-old was suffering from episodes of abdominal pain and vomiting with multiple hospital visits without reaching a diagnosis for four months. Contrast-enhanced computed tomography scan (CECT) done which shows a 1.6 x 1.4 cm fluid collection was seen in unicentric process of the pancreas. Magnetic resonance imaging (MRI) demonstrates abnormal vasculature involving the pancreas. Therefore patient diagnosed as a case of pancreatic arteriovenous malformation (P-AVM), which confirmed by Selective Computed Tomography Angiogram (CT Angiogram).

DISCUSSION: Normal investigations in the first attempts can lead to miss pancreatic arteriovenous malformation (P-AVM), and can lead to serious and fatal.

CONCLUSION: Early diagnosis and treatment of (P-AVM) is very important even in asymptomatic patients. Conservative treatment in complicated and big P-AVM appears ineffective as surgical resection.

© 2018 The Author. 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

Pancreatic arteriovenous malformation (P-AVM) is an extremely rare entity, even more rare to involve whole parts of the pancreas [1]. Although the diagnosis of P-AVM is easy after imaging modalities development, it can be missed in asymptomatic patient and lead to fatal complications [2]. There are several articles report a successful treatment with transcatheter arterial embolization (TAE), and radiotherapy, but surgical resection appears to be the only curative treatment [3]. The reported case is a patient complains abdominal pain and vomiting, successfully diagnosed and treated for complicated pancreatic arteriovenous malformation (P-AVM).

The work has been reported in line with the SCARE criteria [4].

2. Case report

A 47 years-old male presented to emergency department complains intermittent, progressive epigastric abdominal pain associated with vomiting for the last four months. He has a history of multiple hospital visits which endevory time with normal laboratory and radiological work up. In our hospital, his abdomen was soft and lax, there was moderate epigastric tenderness. Laboratory work including pancreatic enzymes was unremarkable. The patient admitted under general surgery as a case of abdominal pain for investigation. Contrast-enhanced computed tomography scan (CECT) done which shows a 1.6 x 1.4 cm fluid collection was seen in unicentric process of the pancreas (Fig. 1A) which was not found in a previous CT done by other hospital three weeks before. Also, there was hypervascularity around tail of the pancreas with a draining veins noticed to joint portal vein confluence (Fig. 1B). A course of Tienam 1 g and metronidazole 500 mg every 8 h was established.

After three days, magnetic resonance imaging (MRI) done which demonstrates the previously mentioned collection as lobulated mass measuring 1.8 x 1.6 cm with air pocket at the center (Fig. 2A). There were abnormal and extensive arterial and venous vasculature within and surrounding the pancreas were seen with several draining veins to the portal venous system (Fig. 2B). The vasculature cannot be explained as simple collaterals, therefore patient diagnosed as a case of pancreatic arteriovenous malformation (P-AVM). Selective Computed Tomography Angiogram (CT Angiogram) done to confirm the diagnosis which demonstrates enhancement of portal venous system during arterial face, and bazar vasculature as P-AVM involving the entire pancreas (Fig. 3). After ten days, a follow up MRI done to follow the mass. The study shows interval increase in size measuring 2.4 x 2.3 cm with multiple gas pockets in the mass center (Fig. 4). Decision was made to take the patient for surgical resection. Intraoperative the pancreas was hard, there were dilated and tortuous abnormal vessels within and around the pancreas which extending to splenic hilum. The mass was palpable at unci- nate process of the pancreas and apart from surrounding structures. Patient undergone to total pancreatectomy with splenectomy.

E-mail address: almalki@mil.gov.sa

https://doi.org/10.1016/j.ijscr.2018.03.026
2210-2612 © 2018 The Author. 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/).
Fig. 1. A (left): Contrast-enhanced computed tomography scan (CECT) shows a 1.6 × 1.4 cm fluid collection located at the uncinate process of the pancreas (red arrow). B (right): Contrast-enhanced computed tomography scan (CECT) hypervascularity around tail of the pancreas (red arrow) and a draining vein joint portal vein confluence (green arrow).

Fig. 2. A (left): Magnetic resonance imaging (MRI) demonstrates a lobulated mass measuring 1.8 × 1.6 cm with gas pocket at the center (red arrow). B (right): Magnetic resonance imaging (MRI) demonstrates an abnormal vasculature involving entire pancreas, as well as the nidus of AVM can be seen (red arrow).

Fig. 3. Computed Tomography Angiogram (CT Angiogram) demonstrates enhancement of portal venous system during arterial face (green arrow), and bazar vasculature as P-AVM involving the entire pancreas; head, body, and tail (red arrows).
Sectioning of the head of the pancreas revealed a solid yellowish soft to firm inflammatory mass measuring $5.5 \times 4 \times 4$ cm, with a central cystic necrotic area about 1 cm in diameter. Microscopically; there is focal vascular proliferation, pancreatic ducts proliferation, parenchymal nicrosis, fibrosis, with chronic inflammatory cell infiltrate (Fig. 5). Postoperative day fourteen, patient improved and discharged to be followed in out-patient clinic. The patient was registered in diabetic center to be evaluated regularly for diabetes and pancreatic enzymes supplements. Follow up CT shows no post-operative complications.

3. Discussion

Pancreatic arteriovenous malformation (P-AVM) first reported in 1986 by Halpern et al., and defined as a vascular anomaly in which blood flows from the arterial system directly into the portal venous system without passing through the capillaries in the pancreas [2]. It is a very rare anomaly with less than 1% incidence of all gastrointestinal vascular anomalies. Frequently involve the head of the pancreas (up to 60%), then body and tail, very rare to involve the entire pancreas [1].

By reviewing the literature in the English language, no more than 130 reported case of P-AVM including cases described secondary in articles with a different main title. There are two articles reporting four cases of P-AVM which involving the entire pancreas; First three cases reported by Ki Byung Song et al., in a study of sixty nine patients from 1999 to 2010 [2]. The fourth case reported by Shimizu K et al., in 2013 [5].

Pancreatic arteriovenous malformation is asymptomatic in general (>95%) and diagnosed accidentally. Most common symptoms reported in symptomatic patients are gastrointestinal bleeding, followed by epigastric pain which radiates to the back in some cases [1]. These symptoms are not specific and most of the time P-AVM diagnosed after performing radiological studies. Patient in this case came with a long history of abdominal pain and vomiting episodes and many hospital visits without reaching a diagnosis. Normal laboratory investigation can lead to missing the diagnosis in first presentations. However, we couldn’t ignore his history and we admit him for more workup. Otherwise, only a few cases of P-AVM were reported with pancreatitis and elevated pancreatic enzymes.

The gold standard imaging to diagnose P-AVM is angiography. Anyway, other imaging modalities include computed tomography (CT) scans, doppler ultrasonography, and magnetic resonance imaging (MRI) are very useful for diagnosis [6]. In this case, Contrast-enhanced computed tomography scan (CECT) demonstrates draining veins into the portal venous system, then it was very clear to make the diagnosis of P-AVM after performing magnetic resonance imaging (MRI).

Diagnosis of hereditary hemorrhagic telangiectasia (HHT) in a patient with chronic gastrointestinal bleeding or visceral vascular malformations should be ruled out. Hereditary hemorrhagic
telangiectasia (HHT) or Osler-Rendu-Weber disease is an autosomal dominant hereditary disease associated with multi-organs vascular abnormalities. According to Curaçao criteria, HHT considered "probable" when two of the following criteria are present and "definite" when three or four criteria are present: epistaxis, telangiectasia, visceral vascular malformations, and a first degree relative with HHT [7]. This patient was negative for Curaçao criteria except for his P-AVM, therefore, HHT was ruled out.

Complications such as gastrointestinal bleeding and rupture of the esophageal varices were reported to be fatal in 30–50% of P-AVM cases. Therefore, treatment of an asymptomatic P-AVM patient is recommended as early prevention of portal hypertension. Portal hypertension once developed cannot be treated even with surgical resection [2].

Another complication of P-AVM is pseudocyst formation. Shu Cheng Chou et al. performed a study on eighty nine P-AVM case, he reported 3.4% of the cases were complicated by pseudocyst [8]. Actually, in our literature all reported pancreatic collections in patients with P-AVM were mentioned as pseudocysts, while Revised Atlanta Classification for pancreatic fluid collections divides them according to the duration of collection forming, type of pancreatitis and presence of infection to several types: acute peripancreatic fluid collection in simple pancreatitis with duration less than four weeks, acute necrotic collection in necrotic pancreatitis with duration less than four weeks, pseudocyst in simple pancreatitis with duration more than four weeks, walled-off pancreatic necrosis in necrotic pancreatitis with duration more than four weeks, and pancreatic abscess in infected collection [9]. In this patient pancreatic enzymes were not elevated and there is no sign for acute pancreatitis. After performing the first CT scan, there was a fluid collection developed within three weeks, therefore the course of antibiotics was established empirically. After performing MRI which shows gas pockets as golden sign for intrapancreatic abscess, then in the follow up MRI which shows interval increasing in mass size (2.4 × 2.3) and gas pockets number within a very short period (10 days), we believed it is persistent intrapancreatic abscess forming inflammatory mass as a complication of P-AVM.

Non-surgical treatment modalities of P-AVM such as transcatheter arterial embolization (TAE), and external radiation can be useful in small non complicated P-AVM, or patients with high surgical risk. Otherwise, surgical resection appears to be the only effective treatment in complicated, and multiple malformations with less recurrence rate [3].

4. Conclusion

Although most of pancreatic arteriovenous malformation (P-AVM) are asymptomatic, misdiagnosis can lead to serious and fatal complications such as gastrointestinal bleeding and pancreatitis. Therefore, early detection and treatment are important.

Conflict of interests

No conflicts of interest.

Funding

No funding resource.

Ethical approval

For case report our Institute exempted to take ethical approval.

Consent

Informed consent for the publication of this work has been taken by the patient.

Author contribution

Single author.

Registration of Research Studies

Research registration box does not apply.

Guarantor

Dr. Ahmad M. Almalki.

References

[1] Jernej Vidmar, Mirko Omeje, Rok Dežman, Peter Popovic, Thrombosis of pancreatic arteriovenous malformation induced by diagnostic angiography: case report, BMC Gastroenterol. 16 (2016) 68.
[2] S. Rajesh, Amar Mukund, Vikram Bhatta, Ankur Arora, Transcatheter embolization of pancreatic arteriovenous malformation associated with recurrent acute pancreatitis, Indian J. Radiol. Imaging 26 (2016) 95–98.
[3] Ki Byung Song, Song Cheol Kim, Jae Bemm Park, Young Hoon Kim, Young Soo Jung, Myung Hwan Kim, et al., Surgical outcomes of pancreatic arteriovenous malformations in a single center and review of literature, Panreas 41 (2012) 388–396.
[4] R.A. Agha, A.J. Fowler, A. Saetta, I. Barai, S. Rajmohan, D.P. Orgill, for the SCARE Group, The SCARE statement: consensus-based surgical case report guidelines, Int. J. Surg. 34 (2016) 180–186.
[5] Kanichiro Shimizu, Yoshimitsu Sunagawa, Kotaro Ouchi, Takuji Mogami, Junta Harada, Kumihiro Fukud, External beam radiotherapy for angiographically diagnosed arteriovenous malformation involving the entire pancreas, Jpn. J. Radiol. 31 (2013) 760–765.
[6] Asma Alnajjar, Ahmed Abu-Zaid, Dina A. Al-onem, Daniah S. Aloufi, Ayman Azzam, Tarek Amin, Concurrent pancreatic head and tail arteriovenous malformations in a 40-year-old gentleman: the first published report, J. Pancreas 15 (2014) 269–273.
[7] Yuijiro Nishioka, Nobuhisa Akamatsu, Yasuhiko Sugawara, Junichi Kaneko, Junichi Arita, Yoshihiro Sakamoto, et al., Hereditary hemorrhagic telangiectasia with hepatic vascular malformations, Case Rep. Med. 2015 (2015) 517818.
[8] Shu-Cheng Chou, Yi-Ming Shyr, Shin-E Wang, Pancreatic arteriovenous malformation, J. Gastrointest. Surg. 17 (2013) 1240–1246.
[9] Ken Zhao, Sharon Z. Adam, Rajesh N. Keswani, Jeanne M. Horowitz, H. Frank, Acute pancreatitis: revised Atlanta classification and the role of cross-sectional imaging, AJR Am. J. Roentgenol. 205 (2015) 32–41.

Open Access

This article is published Open Access at sciencedirect.com. It is distributed under the JJSCR Supplemental terms and conditions, which permits unrestricted non commercial use, distribution, and reproduction in any medium, provided the original authors and source are credited.