Science Letters ISSN 2345-5463 – An International Triannually Journal

Case Report
August 2017 | Volume 5 | Issue 2 | Pages 163-166

ARTICLE INFO

Received
March 03, 2017
Accepted
May 06, 2017
Published
August 15, 2017

***Corresponding Author**
Zijun Liu
E-mail
liuzijundoctor@sina.com
Phone
+86-18951670241

Keywords
Extrapancreatic
Intrapancreatic
Intraoperative ultrasonography
Manual palpation
Glucose monitoring

How to Cite
Manandhar S, Ma Y, Yang SY, Xi PC, Liu ZJ. Pelvic retroperitoneum insulinoma-a rare clinical entity: case report. Sci Lett 2017; 5(2):163-166

Open Access

Pelvic Retroperitoneum Insulinoma – A Rare Clinical Entity: Case Report

Suhreet Manandhar, Yong Ma, Shiyong Yang, Pengcheng Xi, Zijun Liu*

Department of General Surgery, Nanjing First Hospital Affiliated to Nanjing Medical University, 68-Chang Le Road, Jiangsu, Nanjing, China

Abstract

This present report describes a rare case of extrapancreatic insulinoma in a 53-year-old female who was admitted to the hospital and was managed surgically. The patient had typical clinical features of insulinoma for two years, which was only relieved by oral intake of glucose. Prior to her admission to the hospital, she had visited other hospitals where a diagnosis of insulinoma was made on the basis of her clinical features for which she underwent blind resection of the tail of the pancreas. Postoperatively, her symptoms exaggerated and hence, she was brought to our hospital where we were able to precisely locate and completely excise the tumor from the pelvic retroperitoneum with the help of preoperative and intraoperative localization techniques. With this report, we would like to stress on the need of precise localization of insulinoma to avoid blind resections and to ensure complete removal of the tumor to prevent recurrences.

Scan QR code to see this publication on your mobile device.

Introduction

Insulinomas are rare neuroendocrine tumors of the pancreas [1]. Most insulinomas are benign, solitary, and intrapancreatic in origin and <2 cm in diameter (90% of cases) [2]. These tumors generally occur sporadically but can also be associated with multiple endocrine neoplasia type 1 (MEN 1) [3]. Patients often present with symptoms of hypoglycemia as shown in Table 1. Whipple's triad, [hypoglycemia (plasma glucose <50 mg/dL), neuroglycopenic symptoms and prompt relief of symptoms following the administration of glucose] is the hallmark of diagnosis [3]. Surgical excision of the tumor is the treatment of choice for the definitive cure of the disease [4]. Here, we report an extremely rare case of extrapancreatic insulinoma, which was precisely localized and excised completely.

Table 1 Symptoms of hypoglycemia.

Neuroglycopenic symptoms	Adrenergic symptoms
Confusion	Diaphoresis
Blurred vision	Palpitations
Seizures	Tremors
Altered behavior	Weight Gain
Stupor or Coma	Hyperphagia

Case presentation

This case is about a 53-year-old female patient, married, had a history of episodes of dizziness, palpitation, diaphoresis, tremors of hands, confusion and on and off the onset for last two years, which was relieved by the oral intake of glucose. Upon a visit to the local hospital, she was presented with a typical Whipple's triad. Her plasma glucose level was less than 2.8 mmol/L (normal range: 3.90-6.44 mmol/L) and had symptoms of hypoglycemia. The symptoms were relieved by the intravenous glucose administration. Although computed tomography (CT) of upper abdomen did not reveal any obvious mass in the upper abdomen, a diagnosis of insulinoma was made on the basis of her clinical features. She underwent abdominal exploration and blind resection of the tail of the pancreas was performed. Histopathological examination of the specimen showed the mass of 0.2 cm diameter, suggesting a neuroendocrine tumor. However, postoperatively, her symptoms did not resolve and aggravated. She also had one episode of syncopal attack. Hence, she visited our hospital where a contrast-enhanced computed tomography (CECT) of abdomen and pelvis revealed four masses in right side of the pelvic cavity as shown in Fig. 1. She denied the history of seizures, galactorrhea and diabetes but she noticed weight gain of 7.5 kg compared to last year. She had no family history of diabetes, thyroid and pituitary diseases. However, she had a history of excision of rectal carcinoid done six years back. She denied the history of alcohol consumption and cigarette smoking.

The laboratory investigations showed plasma glucose level of 2.44 mmol/L with an insulin level of 51.33 uU/ml (normal Insulin range: 2.6-24.9 uU/ml). She underwent surgical exploration again in our hospital. No obvious masses were seen in the abdominal and pelvic cavity. However, manual palpation and with the use of intraoperative ultrasonography (IOUS), the tumor was localized in pelvic retroperitoneum. The tumor was hence carefully excised with intraoperative frozen section suggestive of insulinoma. Hence, we continued to excise remaining masses similarly with a total of five masses excised from the pelvic retroperitoneum with the largest about 3cm × 3cm and the smallest about 0.8cm × 0.8cm in size as shown in Fig. 2. Intraoperatively, plasma glucose level was closely monitored and plasma glucose and insulin levels measured at end of surgery were within the normal range. Postoperatively, her plasma glucose level was persistently high. Hence, regular insulin was started with continuous plasma glucose monitoring. Her plasma glucose level gradually reduced and insulin was eventually stopped once her blood glucose level was within the normal range. Histopathological examination of the specimen showed typical features of insulinoma as shown in Fig. 2. She was discharged after 14 days of hospital stay with a normal range of plasma glucose level. Her plasma glucose level was also within normal range after six months of follow-up period.

Discussion

Insulinomas are rare neuroendocrine tumors with the incidence of 1-4 people per million in general population [1]. Almost all insulinomas are intrapancreatic with extremely low incidence of extrapancreatic insulinoma (<2%) [2]. Extrapancreatic insulinoma cases reported have mainly been in duodenum, ileum, splenic hilum, gastrocolic ligament, lung and cervix [2]. Our case of pelvic retroperitoneum insulinoma was atypical and a rare presentation. Once suspected, localization of tumor is the key to successful management of the patient. Preoperative localization of tumor is not only helpful in the

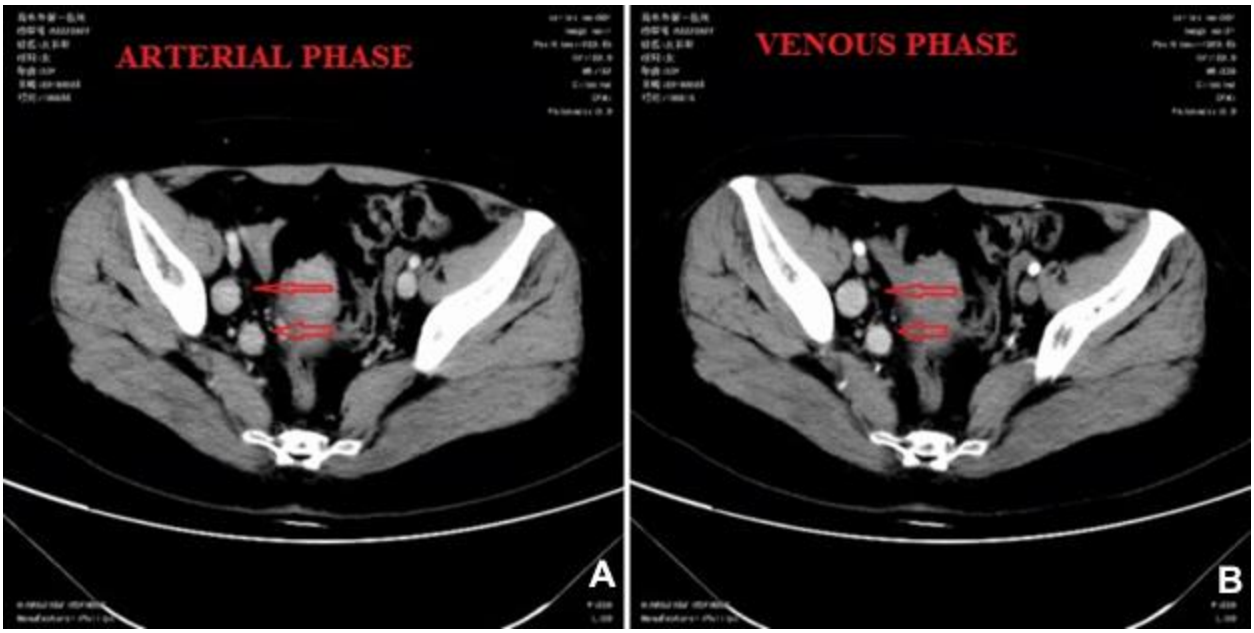

Fig. 1 Contrast enhanced CT scan of abdomen and pelvis. (**A**) Arterial phase showing two masses in the right side of the pelvic cavity (red arrow) and (B) venous phase showing two masses in the right side of pelvic cavity (red arrow).

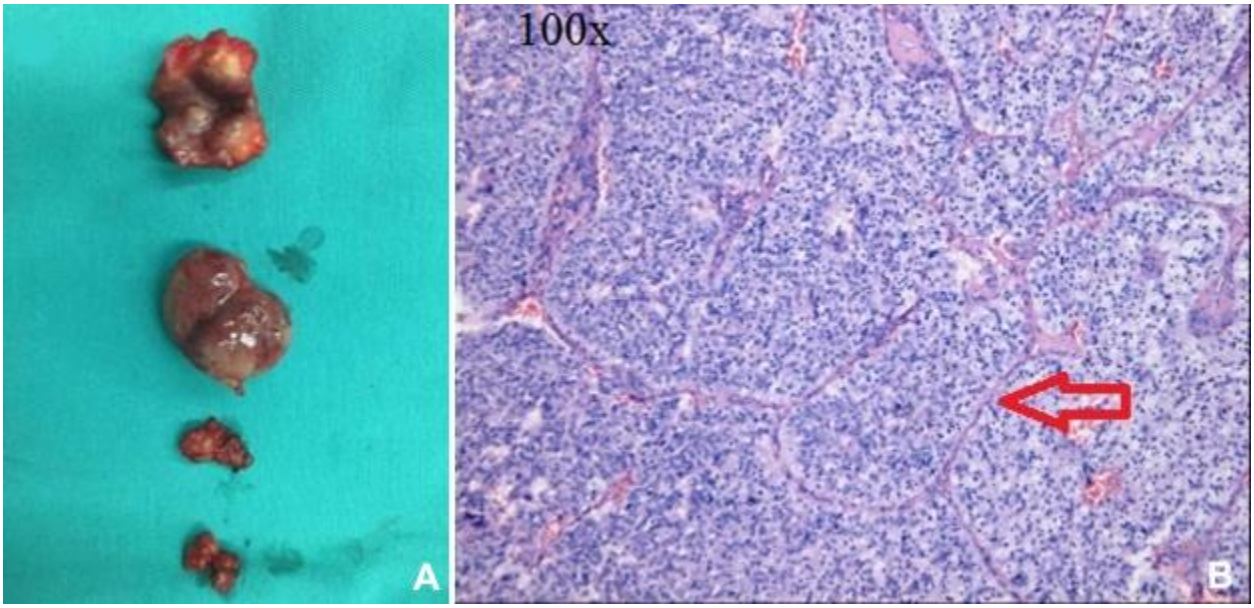

Fig. 2 Gross pathology (**A**) and histopathological examination (**B**) showing tumor cells arranged in nests and separated by thin fiber blood vessels (red arrow).

planning of surgery but also for the evaluation of metastatic diseases [3, 4]. Most commonly used methods of preoperative localization are computed tomography (CT), magnetic resonance imaging (MRI) and somatostatin receptor (SSTR) scintigraphy [5, 6]. Another important localization method is manual palpation and use of intraoperative ultrasonography (IOUS), which many surgeons consider the best method of localization, helps not only in localizing the tumor and surgical excision but also in understanding its relationship with surrounding structures [7]. Our patient had undergone blind resection of the tail of pancreas in other hospitals prior to admission in our hospital as surgeons from other hospital were unable to localize the tumor which did not relieve her symptoms.

Hence, precise localization of tumor should be done to prevent unnecessary blind resection of the pancreas. Surgery is golden standard of care for most insulinomas with cure rates of 77-100% and prognosis dependent on stage and features of the tumor [7, 8]. As recurrences are high, it is essential to ensure complete removal of the tumor [7, 8]. Measurement of plasma glucose and insulin level during and after surgery helps in ensuring complete excision of the tumor. Intraoperative plasma glucose monitoring, although infrequently used, has a sensitivity of 87% (based on the increase in plasma glucose level of 30 mg/dL（1.66mmol/L in sample taken before and after resection) [9]. Amikura et al. [9] in a study concluded that the combined monitoring of peripheral blood glucose and peripheral and portal immunoreactive insulin (IRI) are helpful in ensuring the complete removal of the insulinoma. In our patient, we noticed an increase in plasma glucose level following resection of tumor; however, her plasma glucose and insulin level were within the normal range at the end of the surgery. Hence, intraoperative plasma glucose monitoring and insulin level monitoring plays a key role in ensuring the complete excision of the tumor.

Conclusions

Insulinoma is rarely met in clinical practices. Once suspected, localization of the tumor is the cornerstone for the successful management of the patients. Although the incidence of extrapancreatic insulinoma is low, it should not always be ruled out in patients to avoid blind resection of the pancreas.

Conflict of Interest

All authors declare that they have no conflict of interest.

References

[1] Shin JJ, Gorden P, Libutti SK. Insulinoma: pathophysiology, localization and management. Fut Oncol 2010; 6(2):229-237.

[2] Okabayashi T, Shima Y, Sumiyoshi T, Kozuki A, Ito S, Ogawa Y, et al. Diagnosis and management of insulinoma. World J Gastroenterol 2013; 19(6):829-837.

[3] Tucker ON, Crotty PL, Conlon KC. The management of insulinoma. Br J Surg 2006; 93(3):264–275.

[4] Taye A, Libutti SK. Diagnosis and management of insulinoma: current best practice and ongoing developments. Dove Press 2015; (5): 125—133.

[5] Poornima V, Mahale A, Kumar A, Subas Chandra S, Paudel K. Insulinoma - A case report and review of diagnostic modalities. J, Indian Acad Clin Med 2008; 9(1): 53-6

[6] S Patel S, Narwari M, Parekh D, Shah V. Insulinoma: case report and review of diagnostic and treatment modalities. J Assoc Phys India 2013; 61(6):423–6.

[7] Tarchouli M, Ali AA , Ratbi MB , Belhamidi Ms , Essarghini M , Aboulfeth EM, et al. Long-standing insulinoma: two case reports and review of the literature. BMC Res Notes 2015; (8):444

[8] Vasikasin V, Watthanatham J, Napatharatip P, Termmathurapoj S. Giant insulinoma in a 15-year-old man: a case report. Int J Surg Case Rep 2016; 24:135-138.

[9] Amikura K, Nakamura R, Arai K, Kobari M, Matsuno S. Role of intraoperative insulin monitoring in surgical management of insulinoma. J Laparoendoc Adv Surg Tech 2004; 11(4):193-199.