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Insulinoma: A Rare Neuroendocrine Tumour - Case Report

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Abstract

Introduction: *Insulinoma is one of the uncommon neuroendocrine tumours. Most of these are benign although less than 10% of them may be malignant. Resulting hyperinsulinaemia initiates frequent attacks of hypoglycaemia which affect patient's quality of life and it may be life threatening. Surgery is the mainstay of treatment for this uncommon tumour.*

Case Report: *We reported about a lady with episodic and repetitive symptoms of hypoglycaemia for 2 years, who was diagnosed as a case of pancreatic insulinoma. The localization of tumour was done by endoscopic ultrasound scan preoperatively and bimanual examination during operation. The enucleation of tumour via the open surgical exploration was successfully done.*

Discussion: *Although the working diagnosis of insulinoma can be obtained by thorough history taking, definite diagnosis may be a great challenge for clinician. The gold standard biochemical tests for diagnosis of insulinoma are measurement of plasma glucose, insulin, C-peptide, and proinsulin during a 72-hour fasting. To date, the only chance of cure for insulinoma is surgery and medical treatments are only reserved for the patients who are unfit for surgical operation. Current clinical practice for managing neuroendocrine tumors often depends on the surgeon's discretion due to the lack of clear guidelines.*

Conclusion: *The meticulous surgical technique with tailored surgical decision making and multimodal perioperative care is crucial for the successful outcome in the management of patient with pancreatic insulinoma.*

Keywords: *insulinoma, hypoglycaemia, enucleation, pancreas.*

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INTRODUCTION

Insulinoma is one of the uncommon neuroendocrine tumours detected in pancreas that affects 1–2 per million patients worldwide. The peak incidence occurs in the fifth decade of life. Most of these are benign although less than 10% of them may be malignant. Often, this tumour may be associated with multiple endocrine neoplasia (MEN) syndrome. Resulting hyperinsulinaemia initiates frequent attacks of hypoglycaemia which affect patient's quality of life and it may be life threatening (Hoskovec, D., et.al 2023). Surgery is the mainstay of treatment for this uncommon tumour, which demands precise localization, effective surgical access, meticulous techniques and proper perioperative care (Maggio, I., et.al 2020; Haisley, K. R., et.al 2019). Here, we report a case of insulinoma which was successfully treated by surgery.

CASE REPORT

A lady in her 40s was referred to our surgical clinic with the history of episodic and repetitive symptoms including diaphoresis, tremors, palpitations, and occasional loss of consciousness for 2 years. She also noticed a weight gain and a chronic weakness. Her symptoms were relieved with eating sweets or taking intravenous glucose injection by a general practitioner around her residence. She denied family history of endocrine disease and malignancy. She was noticed to have neuroendocrine tumour in the pancreas which was confirmed by endoscopic ultrasound (EUS) and fine needle aspiration (FNA) biopsy in another hospital. The CT scan examination done before EUS revealed unremarkable. Her Body Mass Index (BMI) was 34.9 kg/m2. Her abdominal examination was unremarkable. Her biochemistry tests demonstrated a low initial glucose level at 46 mg/dl (70–11 mg/dl), a high plasma insulin level at 70.4 µ IU/ml (2.6–24.9 µ IU/ml), and a high C-peptide level at 6.76 ng/ ml (0.8–4.2 ng/ml). Prolonged supervised fasting test was applied and produced symptomatic hypoglycaemia with hyperinsulinemia. Screening of urine for sulfonylurea was negative. Immunohistochemistry test for tumour cells showed a positive staining for synaptophysin. Preoperative localization of tumour with ultrasonography showed a 2 cm well-defined lesion in uncinate process of pancreas measuring with no intra-abdominal lymph nodes. The hormonal studies of serum parathormone, adrenocorticotropic hormone (ACTH), cortisol, thyroid hormone and serum calcium were unremarkable. Because of the resource limitation, we could not perform the MEN gene assay for this patient. According to her functional status and biological behavior, she was diagnosed as a case of pancreatic insulinoma. The enucleation of tumour via the open surgical exploration was performed with upper midline incision. The lesser sac was opened and Kocherization of the duodenum was done. Complete mobilization and bimanual palpation of the pancreas allowed to detect a firm and well-circumscribed nodule at the uncinate process. Enucleation was performed by gentle blunt dissection using sponge on holder and bipolar diathermy after putting stay sutures (Figure-1).

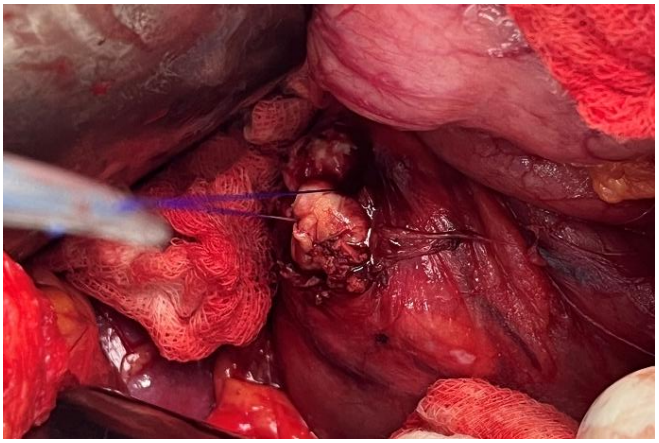

Figure (1) Enucleation of the tumor from uncinate process of pancreas

The meticulous haemostasis was performed with bipolar diathermy. Drainage tube was inserted.The gross examination of the specimen revealed an encapsulated pancreatic mass measuring 18 into 18 mm with homogenous surface with small cystic area in cut section (Figure 2 & 3).

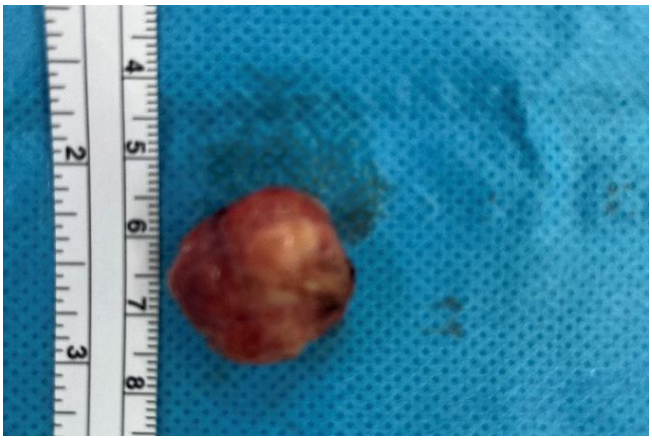

Figure (2) Gross appearance of enucleated insulinoma

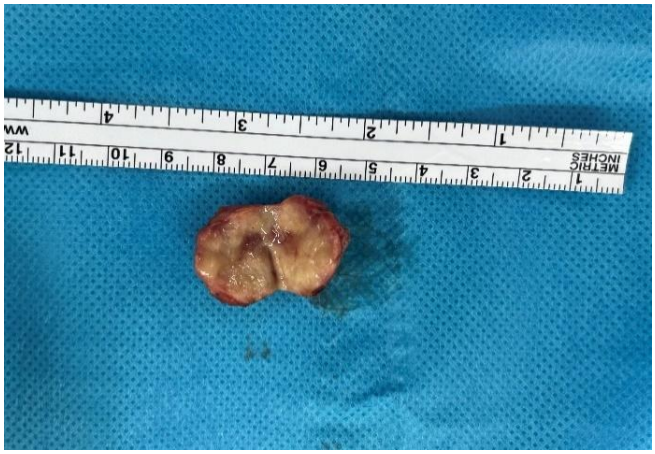

Figure (3) Cut section of the specimen

Immediately after the surgery, her blood glucose level increased. Drainage tube was removed on the third post operative day. The patient was discharged 4 days after surgery. The histopathological examination confirmed the benign insulinoma. The patient remains asymptomatic and her biochemical tests are normal within follow up.

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DISCUSSION

To our knowledge, insulinoma is not a common surgical problem in daily routine. The classical diagnosis of insulinoma depends on the criteria of Whipple's triad which included hypoglycemia (plasma glucose < 50 mg/dL), neuroglycopenic symptoms and prompt relief of symptoms after taking glucose (Rostambeigi, N., et.al 2009). Although the working diagnosis of insulinoma can be obtained by thorough history taking, definite diagnosis may be a great challenge for clinician. Originally, it was considered that symptoms only occurred in the fasting state or following exercise. But it is now known that patients with an insulinoma can also present with postprandial symptoms (Service, F. J.1995). In our case, the definite diagnosis of insulinoma was delayed up to 2 years duration because of lacking the search for medical consultation and effective localization as her symptoms were relieved with administration of glucose injection by local general practitioners. The gold standard biochemical tests for diagnosis of insulinoma are measurement of plasma glucose, insulin, C-peptide, and proinsulin during a 72-hour fasting. In our report, biochemical tests confirmed the cause of hypoglycaemic symptoms as the insulinoma.

Other challenge in a patient with insulinoma is to localize the specific site of tumour. Noninvasive imaging modalities likef USG, CT and MRI can localize and assess the extent of tumour (Tarchouli, M., et.al 2015). Some researchers reported that endoscopic ultrasound scan (EUS) is the most sensitive and reliable method to detect insulinoma although it is operator dependent (Wiese, D., et.al 2023). Furthermore, EUS done by expert may give the benefit for taking fine needle aspiration (FNA) biopsy as in our patient. Although FNA is not essential in the every case of insulinoma, it may exclude the malignancy. The accuracy of CT scans for localizing insulinomas is only 64% in some reports. Some report that localization by intraoperative ultrasound and pancreatic palpation increases the accuracy to 97.1% (Roland, C. L., et.al 2008).

To date, the only chance of cure for insulinoma is surgery and medical treatments are only reserved for the patients who are unfit for surgical operation. Enucleation is the most administered type of surgery (56%). Different types of resections include distal pancreatectomy (32%), Whipple procedure (3%), and subtotal pancreatectomy (<3%). Despite the development of laparoscopy, open approach is the favorite method (90%). The most common surgical complication is postoperative pancreatic duct fistula (Mehrabi, A., et.al 2014).

Current clinical practice for managing neuroendocrine tumors often depends on the surgeon's discretion due to the lack of clear guidelines. Successful surgical management of insulinoma requires significant expertise to minimize the risk of postoperative morbidity (Alogakos, M., et.al 2025). The latest consensus of North American Neuroendocrine Tumor Society (NANETS) suggested that enucleation should be used for smaller tumors, particularly those that are likely benign and located more than 2–3 mm from the main pancreatic duct (Howe, J. R., et.al 2020).

Nowadays, laparoscopic approach is more feasible and becomes increasingly reported with good results in selected patients. Tumor location should be confirmed intra-operatively by laparoscopic ultrasonography (Isla, A., et.al 2009).

Most insulinomas can be identified intraoperatively by an experienced surgeon. However, what should be done in an operating theater when an insulinoma cannot be identified is questionable for surgeons. Some reviews suggested to use various non-invasive and invasive imaging modalities when the tumor cannot be detected using conventional diagnostic procedures (Okabayashi, T., et.al 2013). It is strongly suggested that blind surgical resection of the pancreas should not be undertaken in any patient who suffers from hypoglycemic episodes due to an insulinoma. The precise localization of the insulinoma enables the surgeon to proceed with surgery uninterrupted, minimizes the operating time, reduces the likelihood for re-operation, perioperative complications, and ensures a successful outcome (Rostambeigi, N., et.al 2009).

CONCLUSION

Insulinoma is a rare endocrine tumour which causes diagnostic challenges. Our case report emphasizes the essential of biochemical tests for diagnosis of insulinoma. Moreover, this report highlights the importance of preoperative localization and intraoperative localization with proper mobilization of duodenum and pancreas. Last not the least, meticulous surgical technique with tailored surgical decision making and multimodal perioperative care is crucial for the successful outcome in the management of patient with pancreatic insulinoma.

CONSENT

Written informed consent was obtained from patient for publication of this case report and corresponding photographs.

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