Case Report

DOI: http://dx.doi.org/10.18203/2349-2902.isj20164489

Insulinoma: is it still a diagnosis of exclusion? A case report

Vipan Kumar*, Vikrant Sharma, Ankur Sharma, Amit Rattan

Department of Surgery, Dr. RPGMC Kangra, Tanda, Himachal Pradesh, India

Received: 13 September 2016 Revised: 26 October 2016 Accepted: 04 November 2016

*Correspondence: Dr. Vipan Kumar, E-mail: drvipan@aol.in

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ABSTRACT

Few cases of hypoglycaemic peripheral neuropathy due to insulinoma have been reported in literature. We are reporting a case of insulinoma manifesting with chronic progressive encephalopathy, intractable seizures, and neuropathy-a triad of neurological features in insulinoma. The diagnosis was delayed as the repeated investigations including dual phase CECT abdomen was normal. This case report highlights the importance of evaluation of patients for insulinoma with high resolution CECT abdomen and FDG PET scan. A keen suspicion and investigation with appropriate modality can prevent the irreversible neuroglycopenic damage, as reported in this case.

Keywords: Insulinoma, Whipples' triad

INTRODUCTION

Insulinoma is the most common endocrine tumor of pancreas, which constitutes 50% of the islet cell tumors and affects 1 in 100,000 population per year. Symptoms are due to the effects of hypoglycemia manifesting as adrenergic response as well as neuroglycopenic symptoms. The clinical diagnosis of insulinoma is based on the Whipple triad which is present in 75% of the patients. Up to 50% of patients are diagnosed 5 years after onset of symptoms. We are reporting a case of insulinoma manifesting with chronic progressive encephalopathy, intractable seizures, and neuropathy-a triad of neurological features in insulinoma. The diagnosis was delayed as the repeated investigations including dual phase CECT abdomen was normal. The role of FDG PET CT scan in diagnosis is highlighted.

CASE REPORT

54 years old lady, resident of Uttarakhand, no known comorbidities, presented with recurrent episodes of unconsciousness and refractory complex partial seizures

with secondary generalization for last 04 years. Patient also had behavioral, personality changes, severe depression, and fluctuating altered sensorium. There was associated history of difficulty in speech and walking with urinary incontinence and the symptoms worsened over due course of time. The patient was bed bound at the time of presentation to our institute. Relatives reported the alleviation of symptoms with sweets. The patient had suffered left radius fracture 02 years back because of unsteady gait and tendency to fall. On examination she was preserved nutritionally with BMI of 26 kg/m². Neurological examination revealed sub-cortical motor dysarthria, trunkal and sensory ataxia. The rest of systemic and general examination was unremarkable except for dinner fork deformity of left hand (sequel of Colle's fracture left). Patient had been extensively evaluated in peripheral hospitals, including psychiatric evaluation for abnormal behaviour. Her workup included routine investigation, USG abdomen and neck, MRI brain and cervical spine, repeated dual phase CT scan abdomen, EEG and nerve conduction studies of all four limbs. All the investigations were essentially normal except for low blood glucose levels on multiple occasions (26-36 mg/dl) and mild distal sensory-motor neuropathy of all limbs. The diagnosis of insulinoma was considered based upon Whipple's triad and patient was evaluated in consultation with endocrinologist and neurophysician. The fasting insulin level was 9.80 mcU/ml (normal < 03 mcU/ml) and fasting insulin-to-glucose ratio was higher.

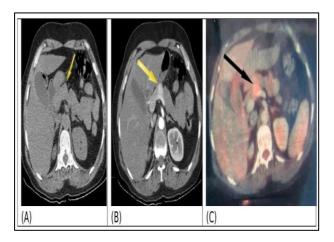


Figure 1: (A), (B) Triple phase CECT abdomen (32 slice) MDCT scanner) showed well circumscribed homogenously enhancing, nodular lesion in neck of pancreas. On non-enhanced section, lesion is predominantly isodense/mildly hypodense relative to normal pancreas. In arterial phase, it enhances homogenously. In portal venous phase also the lesion was hyper attenuating relative to normal pancreas. (C) Whole body FDG PET CT showed well defining mildly FDG avid lesion in neck of pancreas, abutting the splenic vein, no other lesion detected.

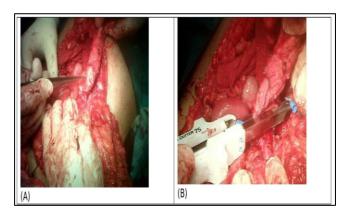


Figure 2: (A) Intra operatively, the tumor was located at the neck of pancreas (pointed by the tip of forceps.
(B) As enoculation was not feasible in this case because the tumor was abutting the splenic artery and vein and was intraparenchymal in location, distal pancreatectomy was done using liner cutter staples.

The C-peptide levels were 2.46 (1.0 - 5.0 mg/ml). She had normal calcium of 2.16 mmol/L (reference value: 2.08 - 2.60 mmol/L), parathyroid hormone (PTH) 54.94 pg/ml(reference value: 15 - 65 pg/ml). Triple phase

CECT abdomen showed well circumscribed homogenously enhancing, nodular lesion in neck of pancreas (Figure 1).

FDG PET CT confirmed the above findings and no other lesion was detected. The above imaging was suggestive of hyper vascular tumor with loss of planes with splenic artery and vein. Patient was taken up for definitive surgery. Intra operatively tumor was intraparenchymal and located at neck of pancreas (Figure 2). As the enucleation was not feasible in this (intraparenchymal, abutting splenic artery and vein), distal pancreactomy was done (Figure 2). Patient had uneventful recovery and her blood sugar levels became normal. The histopathological examination showed well differentiated, encapsulated pancreatic tumor of uncertain behavior because of angioinvasion (Figure 3).

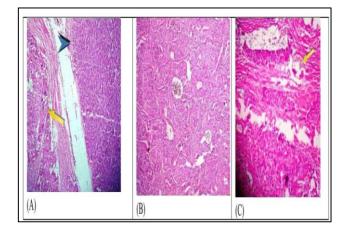


Figure 3: HPE-HE stain 40 X. (A) Normal process (arrow head) and encapsulated tumour (arrow). (B) Insular, gyriform and glandular pattern suggestive of endocrine tumor. (C) Angioinvasion (arrow), suggestive of uncertain behaviour of tumour.

DISCUSSION

Few cases of hypoglycemic peripheral neuropathy due to insulinoma have been reported in literature.⁵ The main clinical sign of insulinoma is the impossibility to suppress the insulin endogen secretion in the presence of hypoglycemia. Successful localization of pancreatic insulinoma aids the planning of surgery which has a cure rate over 90%. However, most (80%) of the insulinomas are smaller than 2 cm and cannot be detected by routine imaging tests including CT, ultrasonography, angiography and nuclear magnetic resonance imaging (MRI).6 Insulinoma is occasionally associated with MEN1. In the present case, we considered that she did not have the association of MEN1, because cervical ultrasonography and brain MRI showed no abnormalities including parathyroid tumour and pituitary tumour. Repeated dual phase CECT abdomen in past failed to demonstrate the lesions probably because of low resolution or mismatch in timing after intravenous contrast injection and image acquisition. Triple phase

CECT abdomen using 32 slice MDCT showed the tumor in neck of pancreas and FDG

PET CT confirmed the diagnosis. The role of PET CT in insulinoma remains yet to be well defined. In summary, it is important to consider the diagnosis of an insulinoma depending on the combined results of clinical, radiological and intervention tests. This case report highlights the importance of evaluation of patients for insulinoma with high resolution CECT abdomen and FDG PET scan. We wish to submit that the insulinoma patients have a considerable diagnostic delay. A keen suspicion and investigation with appropriate modality can prevent the irreversible neuroglycopenic damage, as reported in this case.

Funding: No funding sources Conflict of interest: None declared Ethical approval: Not required

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Cite this article as: Kumar V, Sharma V, Sharma A, Rattan A. Insulinoma: is it still a diagnosis of exclusion? A case report. Int Surg J 2017;4:440-2.