Insulinoma: Case Report of a Rare Neuroendocrine Tumor

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Abstract

Background

Insulinomas are neuroendocrine tumours of pancreatic origin causing excessive release of insulin that results in recurring episodes of hypoglycemia. With a prevalence rate of up to 4 people per million, these tumours are quite rare, with no cases reported in Nepal. Therefore, a report on this case can guide clinicians in the future when they come across a similar condition in making a diagnosis and planning the treatment course.

Case Presentation

We report a case of a 34-year-old male with insulinoma presenting with complaints of dizziness, sweating, and palpitation who was managed successfully with surgical enucleation of the tumour resulting in the resolution of the symptoms.

Conclusions

Insulinoma may present with non-specific findings that could result in underdiagnosis. A history of recurrent neuroglycopenic symptoms along with markedly increased levels of insulin should raise suspicion of insulinoma and imaging modalities like Endoscopic Ultrasonography (EUS) in conjunction with Magnetic Resonance Imaging (MRI) should be performed for appropriate management.

Keywords: *Case Report, Endoscopic Ultrasonography, Hypoglycemia, Insulinoma.*

Introduction

Endocrine cell tumours of the gastrointestinal tract can be classified histologically as either carcinoids or endocrine tumors.[1,2] Insulinoma is usually a benign and solitary tumour of the pancreas which can occur both sporadically or in conjunction with Multiple Endocrine Neoplasia Type[1] (MEN1) syndrome.[3] Insulinomas associated with MEN1 syndrome are often multicentric and develop earlier than sporadic insulinomas.[3] They are characterized by features of hypoglycemia including forgetfulness and altered consciousness along with clinical features of sympathetic overstimulation like sweating, tremors, palpitations, and hyperphagia.[3,4] These episodes are exacerbated following exertion, fasting, or even after meals.[5] Herein, we present a case of a 34 years old male diagnosed with insulinoma with good clinical outcome following treatment.

Case Report

We report a case of a 34-year-old male who presented to the Emergency Department of Dhulikhel Hospital in October 2022 with complaints of dizziness, sweating, and palpitation which got resolved shortly after by intake of sweet food like chocolate. The patient added that he experienced similar bouts of symptoms more than 5 times within a month with each event lasting

for nearly an hour after having dinner. The documented value of random blood sugar during each event ranged from 40-50 mg/dl.

On examination, the vitals were stable. However, the lab investigation findings were significant with markedly decreased blood glucose levels as suggested by the random blood sugar (RBS) of 45 mg/dl. The fasting blood sugar was 110 mg/dl, post-prandial blood sugar was 89 mg/dl, fasting insulin was 29.8 µIU/ml, post-prandial insulin was 72.3 µIU/ml, total bilirubin was 3.6mg/dl, and direct bilirubin was 0.2 mg/dl. On overnight fasting, fasting insulin was 100.7 µIU/ml, fasting blood sugar was 100 mg/dl, and HbA1c was 5.1%. All other haematological and biochemical parameters were unremarkable. Endoscopic Ultrasonography (EUS) (Figure 1) showed an 8.4 mm hypoechoic lesion in the pancreatic neck body region suggestive of a tumour. To corroborate this finding, a dynamic MRI study was performed which illustrated a 9.4x7.5 mm size hypointense lesion in the neck of the pancreas (Figure 2).

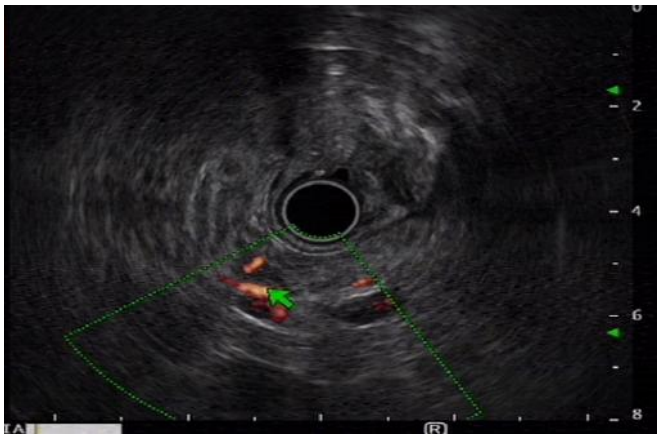

Figure 1. EUS showing an 8.4 mm hypoechoic lesion (green arrow) in the pancreatic neck body

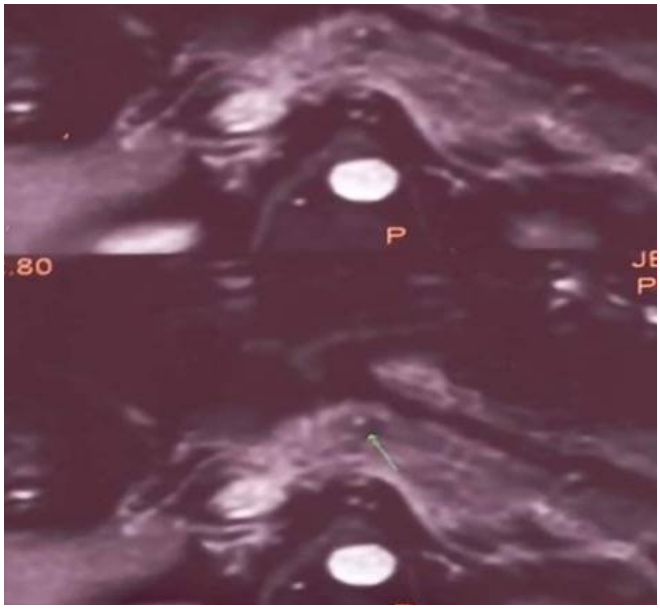

Figure 2. MRI showing a 9.4x7.5 mm size hypointense lesion in the neck of the pancreas

The patient underwent surgical exploration in a tertiary care hospital in Kathmandu. A small tumour was identified at the neck of the pancreas at the surgery. The complete enucleation of the lesion was performed and was sent for histopathological examination which was unremarkable.

Following the surgery, the neuroglycopenic symptoms and palpitation resolved. On a five-week follow-up, the fasting insulin was 27.3 µIU/ml, fasting C-peptide was 5.1 ng/ml, and fasting blood sugar (FBS) was 102 mg/dl. After 3 months, a repeat EUS showed no obvious pathological changes apart from a scar resulting from the prior surgery (Figure 3).

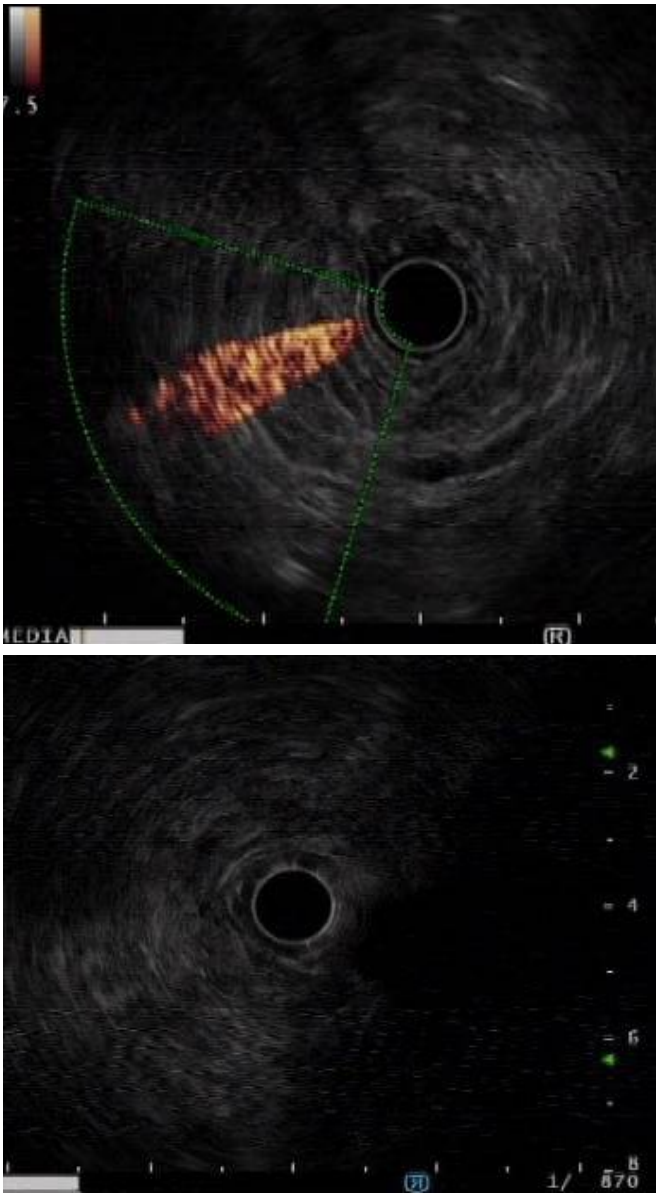

Figure 3. Repeat EUS showing homogenous pancreatic parenchyma with no obvious pathological changes.

Discussion

Insulinomas are pancreatic neuroendocrine tumours that are derived from the pancreatic beta cells.[5] These tumors are the most common endogenous cause of

hyperinsulinemia having a prevalence of up to 4 people per million of the general population.[5]

Whipple's triad is a commonly used screening modality for diagnosing insulinoma and includes (1) hypoglycemia (plasma glucose < 50 mg/dL); (2) symptoms that are consistent with hypoglycemia; and (3) relief of symptoms following the administration of glucose.[6] For the indexed case, the diagnosis of an insulinoma met all three criteria of the triad. Laboratory investigations include the measurement of plasma glucose, insulin, and C-peptide, with the gold standard being the measurement of proinsulin in a 72-hour fast.[5] However, a normal insulin level does not rule out the condition.[7,8] In our case, the patient had a random blood sugar of 45 mg/dl, and preoperatively, fasting insulin was 29.8 µIU/ml, post-prandial insulin was 72.3 µIU/ml and on overnight fasting, the insulin was 100.7 µIU/ml with the fasting blood sugar 100 mg/dl. High proinsulin levels have been proposed as an insulinoma diagnostic marker regardless of concurrent blood glucose levels as the fraction of proinsulin produced by insulinoma cells is often larger than that secreted by normal cells.[7]

Transabdominal ultrasound, abdominal computed tomography (CT), magnetic resonance imaging (MRI), and endoscopic ultrasonography (EUS) are radiological investigations that can be done to localize the lesion.[9] EUS has a higher sensitivity than triple-phase MDCT scan in detecting pancreatic neuroendocrine tumors, notably for insulinomas. For lesions <2 cm, EUS aids in the detection of 91% of insulinomas missed by MDCT.[10] CT and MRI demonstrate characteristic lesions on imaging with MRI being generally better than CT scan.[10,11] Insulinomas exhibit more enhancement under contrast than normal pancreatic parenchyma because of hypervascularity.[11] A notable peculiarity in our case is that CT and MRI yielded normal results whereas EUS demonstrated the presence of a lesion. This goes to show the superiority of invasive modalities like EUS in diagnosing Insulinoma above non-invasive ones like CT and MRI.[3,5]

Conservative management of patients with insulinoma includes frequent meals and glucose infusion.[12] Somatostatin analogs are also shown to be effective as somatostatin receptors are expressed in the neoplastic cells.[13] Since this receptor is not universal, the outcomes may vary.[13] Additionally, long-acting somatostatin analogs are linked to aggravating hypoglycemia due to peripheral glucose utilization.[14] Thus, surgical management remains the mainstay treatment for insulinoma.[5] Owing to the range of techniques available for diagnosing the tumor, blind resection can be avoided.[5] Small, benign tumors that are at least 2-3 mm from the main pancreatic duct should be enucleated.[9] Resection is necessary when the tumor borders a major vascular or pancreatic duct, and in cases of infiltrating tumor puckering the nearby soft tissue.[9] Depending on the location of the insulinoma, resection options include distal pancreatectomy, pylorus-preserving Whipple surgery, or mid-body pancreatectomy with laparoscopic resection gaining popularity.[9] In cases of malignant insulinomas, multiple approaches like resection of the pancreas and liver, liver transplantation, radiofrequency ablation, or chemoembolization may be essential.[5,15] In our case, enucleation was done specifically since we had a single lesion. On follow-up revealed normal pancreatic parenchyma with no pathological changes apart from the fibrosis resulting from the surgery.

Conclusively, any cases of recurrent neuroglycopenic symptoms should be thoroughly evaluated in suspicion of insulinoma to ensure early treatment and decrease the risk of malignant conversion.

Conclusion

Insulinoma is a rare tumor, which presents with non-specific findings resulting in underevaluation. Thus, a history of recurrent neuroglycopenic symptoms along with markedly increased levels of insulin should raise suspicion of this disease. Endoscopic ultrasonography in conjunction with MRI should be performed for establishing the diagnosis and planning the treatment.

Consent

Written informed consent was obtained ensuring patient's anonymity

Declaration of competing interest

There are no conflicts of interest.

Acknowledgement

N/A

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