Insulinoma camouflaged by vague neuroglycopenic symptoms: A case report

Kiran Sunil Mahapure[1], Shrishail Chandrakant Metgud[2]

From [1]Senior Resident, [2]Professor, Department of General Surgery, KAHER J. N. Medical College, Belgaum, Karnataka, India

Correspondence to: Dr. Kiran Sunil Mahapure, Mahapure Hospital, Shivaji Nagar, Peth Vadgaon, Kolhapur, Maharashtra - 416 112, India. E-mail: drkiranmahapure@gmail.com

Received - 21 March 2020 Initial Review - 06 April 2020 Accepted - 23 April 2020

ABSTRACT

Pancreatic endocrine tumors are rare lesions, with a reported incidence of four cases per 1 million patients per year. Of them, insulinomas are the most common. These are usually benign, solitary, and small size (<2 cm in diameter). Most are sporadic; however, 10% are multiple and occur as part of multiple endocrine neoplasia syndrome type I. Chronic hypoglycemia due to an insulinoma often presents with neuroglycopenic symptoms that can easily be mistaken for neurological or psychiatric symptoms, especially in developing countries. This may result in delayed diagnosis with associated complications. We hereby present the case of a 45-year-old female from the rural background that initially sought treatment by a psychiatrist for her vague neurological symptoms and were later diagnosed to have insulinoma.

Key words: *Hypoglycemia, Insulinoma, Neuroglycopenic symptoms*

Pancreatic endocrine tumors are rare lesions, with a reported incidence of four cases per 1 million patients per year. Of these lesions, insulinomas are the most common with estimates of 1 in 250,000 people per year. The majority of patients are between 30 and 60 years of age, with women accounting for 59% [1]. Usual clinical presentation is benign, with a solitary and small size (<2 cm in diameter). Almost all insulinomas are located within the pancreas, even though aberrant cases have been described in the duodenum, ileum, lung, and cervix [1]. Most are sporadic; however, 10% of insulinomas are multiple and occur as part of multiple endocrine neoplasia syndrome type I (MEN-I) [2].

The clinical manifestations are non-specific and varied. Normal health in between the episodes of hypoglycemia delays the diagnosis. Patients with insulinoma have symptoms of hypoglycemia resulting from neuroglycopenia and increased catecholamine release. Surgical excision is the treatment of choice and is curative in most cases [1]. Chronic hypoglycemia due to an insulinoma often presents with neuroglycopenic symptoms that can easily be mistaken for neurological or psychiatric symptoms, especially in developing countries, where awareness and availability of health services are comparatively less. This may result in delayed diagnosis with prolonged periods of untreated hypoglycemia and associated complications [3].

We hereby present the case of a 45-year-old female from the rural background that initially sought treatment by a psychiatrist for her vague neurological symptoms and were later referred to our surgical team and diagnosed to have insulinoma.

CASE REPORT

A 45-year-old female suffered from intermittent episodes of altered mental status including fatigue, irritability, poor concentration, and tremors over a period of 3 years. She also had recurrent attacks of giddiness with sweating and palpitations accompanied by diffuse pain in the abdomen and tingling and numbness over both the upper and lower limbs. The symptoms used to relieve after eating, especially sweet. She had a voracious appetite and had gained a tremendous amount of weight (>10 kg). The patient did not seek medical attention due to the ambiguity of her symptoms and normal health in between the attacks.

The patient was first taken for the psychiatric evaluation by her relatives misinterpreting her symptoms who subsequently referred her to the physician for the treatment of recurrent hypoglycemia. The physician referred her to our surgical team for further evaluation after putting her on dextrose tablets. On examination, the patient was a plump middle-aged female with vitals within normal limits. Per abdomen examination did not rule out any abnormal finding.

Her serum insulin levels were 60.4 mcU/ml on admission (normal range 3–28 mcU/ml), blood sugar levels 80 mg/dl, and C-peptide levels were 3.3 ng/ml (normal: 1.0–5.4). Her HbA1c levels were 5.0%. On the 3rd day, her serum insulin levels were 52.6 mcU/ml and blood sugar levels 49 mg/dl. A computed tomography scan abdomen showed homogeneously enhancing lesion in the tail of the pancreas measuring 1.2 × 1.3 cm showing enhancement in the arterial phase suggestive of insulinoma in the tail of the pancreas (Fig. 1).

After due preparation, the patient underwent laparotomy by the left subcostal incision under general anesthesia. Complete mobilization of the pancreas discovered a firm and unifocal nodule of size 1.2 × 1.3 which was localized in the tail of the pancreas (Fig. 2). Enucleation of the tumor was done followed by the placement of the drain. Postoperatively, the patient's blood sugar levels were raised for 3 days (day 1 – 350 mg/dL and day 3 – 260 mg/dL) which normalized gradually (day 5 – 124 mg/dL). Insulin levels came back to normal limits postoperatively, immediately (post-operative day 5 – 7.55 mcU/ml).

Histopathology report showed insulinoma with normal pancreatic elements. The patient was discharged on post-operative day 7th and followed up after 15 days with complete resolution of symptoms. The delay in seeking medical attention resulted in curative surgery more than 3 years after symptom onset.

DISCUSSION

Insulinomas are rare pancreatic tumors comprised pancreatic beta islet cells that produce excess amounts of endogenous insulin. Common age of presentation is between 30 to 60 years with 45 years being the average. These are located in the pancreas with equal distribution in the head, body, and tail with a rare occurrence at the ectopic tissues such as duodenum, splenic hilum, or gastrocolic ligament. Approximately 90% of insulinomas are <2 cm in size with morphological features of an encapsulated, firm, yellow-brown nodule, often solitary and the remaining 10% are multicentric. Microscopic findings are clusters of beta cells, stain positive for insulin [1,2].

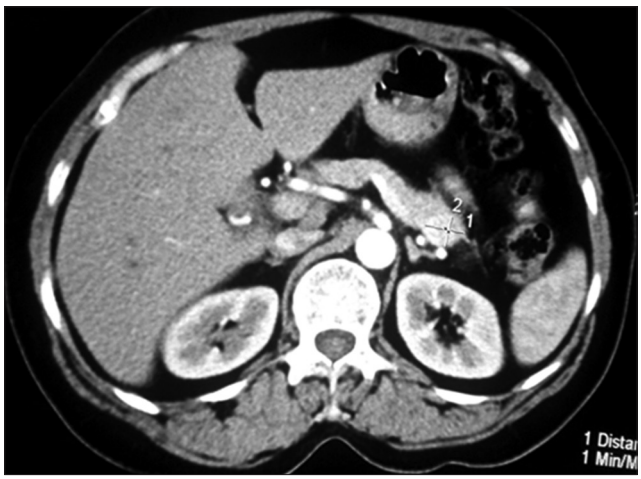

Figure 1: Computed tomography abdomen showing insulinoma

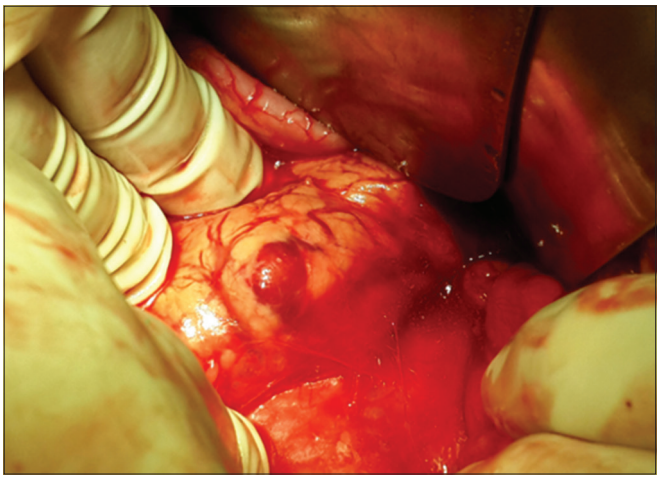

Figure 2: Intraoperative picture of insulinoma

This inappropriate insulin secretion results in hypoglycemia with its associated neurogenic and neuroglycopenic symptoms. With prolonged and repeated hypoglycemia, neurogenic symptoms are minimized and non-specific neuroglycopenic symptoms predominate [3]. The patients with insulinoma have symptoms of hypoglycemia resulting from neuroglycopenia and increased catecholamine release. Neuroglycopenic symptoms are most common, including anxiety, dizziness, lightheadedness, personality changes, unusual behavior, confusion, incoherence, blurred vision, seizures, and coma. Sympathoadrenal signs and symptoms, such as palpitations, tremulousness, diaphoresis, and tachycardia, may also be present and are due to catecholamine release in response to low serum glucose levels [4]. Other symptoms are vague abdominal pain and discomfort.

Whipple's triad provides diagnostic criteria which include blood sugar levels <45 mg/dl, symptoms of hypoglycemia on exertion, and relief of symptoms after glucose intake. Laboratory assessment should include serum glucose, insulin, and C-peptide or proinsulin levels which should demonstrate hypoglycemia with inappropriately normal or elevated insulin concentrations. Elevated C-peptide or proinsulin levels support endogenous insulin production, whereas these values would be suppressed with exogenous insulin administration (factitious hypoglycemia).

Considering this a case of sporadic insulinoma, investigations to rule out MEN should have been done which were not done in our case. Failing to have restoration of blood glucose to fasting levels as rapidly as normal when made hypoglycemic with insulin is characteristic of insulinoma. This principle was the basis of the insulin tolerance test which made the diagnosis easier with the availability of plasma insulin assay [5].

The differential diagnosis could be the chronic liver disease, tumor-related hypoglycemia, and endocrinopathy. Surgical excision is the treatment of choice and is curative in most cases [6]. Long-term survival of patients with insulinoma is generally excellent; it is considered that approximately 90–95% of insulinomas have benign histological behavior, so healing with the disappearance of symptomatology after complete resection is the rule [7].

CONCLUSION

We demonstrate the importance of being vigilant for fasting hypoglycemia secondary to insulinoma even when the patient presents with vague neurological or neuroglycopenic symptoms. If these neuroglycopenic complaints are unnoticed or misdiagnosed, patients with a potentially curable disease are put at risk of neurologic injury, or even death, due to untreated severe hypoglycemia. Early diagnosis by investigating for insulinoma

in cases presenting with vague symptoms of hypoglycemia and neuroglycopenia along with multidisciplinary management is the key.

REFERENCES

1. Patel S, Narvaria M, Parekh D, Shah V. Insulinoma: Case Report and Review of Diagnostic and treatment Modalities. J Assoc Physicians India 2013;61:423-6.
2. Tarchouli M, Ali A, Ratbi M, Belhamidi M, Essarghini M, Aboulfeth E, *et al*. Long standing insulinoma: Two case reports and review of the literature. BMC Res Notes 2015;8:444.
3. Halpin K, McDonough R, Alba P, Halphin J, Singh V, Yan Y. Vague neuroglycopenic complaints camouflage diagnosis of adolescent insulinoma: A case report. Int J Pediatr Endocrinol 2016;2016:14.
4. Dajbong E, Lefter LP, Scripcariu V, Ferariu D, Dragomir C. Diagnostic features of benign pancreatic insulinomas. An analysis of three cases. J Gastrointestin Liver Dis 2006;15:61-5.
5. Marks V. Diagnosis and differential diagnosis of insulinoma. Mayo Clin Proc 1989;64:1558-61.
6. Mittendorf EA, Liu Y, McHenry C. Giant insulinoma: Case report and review of literature. J Clin Endocrinol Metab 2005;90:575-80.
7. Negrean V, Tudir A, Aioanei O, Domsa I. Pancreatic insulinoma. Case report and review of the literature. Clujul Med 2013;86:377-9.

Funding: None; Conflict of Interest: None Stated.

How to cite this article: Mahapure KS, Metgud SC. Insulinoma camouflaged by vague neuroglycopenic symptoms: A case report. Indian J Case Reports. 2020;6(5):241-243.

Doi: 10.32677/IJCR.2020.v06.i05.008