International Psychogeriatrics: page 1 of 3 © International Psychogeriatric Association 2013
doi:10.1017/S104161021300104X

CASE REPORT

An insulinoma presenting with hypochondriac delusions and food refusal

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ABSTRACT

The authors report a case of a 68-year-old man with an unrecognized insulinoma manifesting with neuropsychiatric symptoms.

For two years, he presented with unspecified behavior changes, autonomic and neuroglycopenic symptoms, which led him to be misdiagnosed with a neurologic and psychiatric disorder before the insulinoma was recognized.

Following neurological alterations in context of hypoglycemia, subsequent to longstanding food refusal, he was admitted in the psychiatric ward. Despite good global response and normal food intake, hypoglycemic episodes were still occurring and led to a careful evaluation which permitted the definitive diagnostic.

This case highlights the diagnostic difficulties of medical disorders presenting with clinical features overlapping neurological and psychiatric syndromes. It also reflects the diagnostic difficulties in rare clinical entities, particularly in patients previously followed in psychiatry and underlines the need for a constant dialogue and updating of clinicians.

Key words: endocrinology, oncological disorders, physical illness, neuropsychiatry

Introduction

Although rare, insulinomas are the most common pancreatic endocrine tumors with an estimated incidence of four cases per million persons/year (Service *et al.*, 1991; Mathur *et al.* 2009).

Common clinical manifestations include a wide range of neuropsychiatric symptoms secondary to hypoglycemic episodes resulting from endogenous hyperinsulinism (Dizon *et al.*, 1999).

Evidence from previous reports suggests that a significant proportion of cases are misdiagnosed with a neurologic or psychiatric disorder before the insulinoma is detected (Harrington *et al.*, 1983; Dizon *et al.*, 1999; Alemdar *et al.*, 2006). We report a case of an unrecognized insulinoma manifesting with hypochondriac delusions and food refusal.

Case report

A 68-year-old male admitted to the emergency department with acute seizures and right hemiparesis, associated with hypoglycemia (plasma glucose: 47 mg/dL), was referred to psychiatric evaluation as longstanding food refusal was considered to be the primary cause of the symptoms.

The patient had been presenting, for the last eight months, hypochondriac delusions of having neck cancer obstructing his throat together with clogged bowels despite all the refuting evidence obtained by clinical and ancillary examinations. These ideas lead to persistent food avoidance and self-induced fasting with frequent and symptomatic hypoglycemic episodes.

His past psychiatric history was remarkable only for a successfully treated depressive episode eight years before. However, over the last two years, behavioral changes were reported consisting in hiding food and refusing meals associated with frequent episodes of sweating, slurred speech, and tremulousness that were relieved by forced eating. Associated clinical manifestations included self-neglect, anhedonia, lethargy, isolation, periods of confusion with disorganized speech, episodes of unresponsiveness and attention impairment with significant difficulties in performing daily tasks.

The patient was admitted to the acute psychiatric ward with the provisional diagnosis of "depressive episode with hypochondriac features" and treated with Venlafaxine 75 mg po id, Risperidone 1 mg

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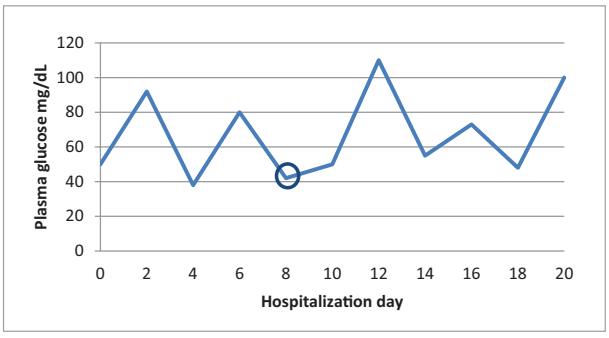

Figure 1. (Colour online) Episode with symptoms of hypoglycemia (Plasma glucose: 42 mg/dL ○ [<55 mg/dL]). Additional workout was performed.

po 2id, and Alprazolam 0.5 mg po 3id. Within a week there was a significant improvement in Brief Psychiatric Rating Scale (BPRS) scores from baseline particularly in somatic concern (6 to 3), anxiety (6 to 2), depression (6 to 3), and unusual thought content (5 to 2) as well as an overall improvement in the Hamilton Rating Scale for Depression (HRSD) (21 to 12). At this time, the eating behavior was normalized as the food intake significantly increased from baseline (900 Kcal/day) to about 1,600 Kcal/day without the need of further interventions. Despite the good clinical response, unremitting hypoglycemic episodes were still occurring even with normal food intake (Figure 1). Additional workout revealed inappropriately high serum insulin (9.5 μU/mL [$\leq$3 μU/mL]), C-peptide (0.4 nmol/L [$\leq$0.2 nmol/L]), and proinsulin concentrations (9 pmol/L [$\leq$5 pmol/L]) associated with low serum glucose (42 mg/dL [$\geq$55 mg/dL]) together with a negative test for insulin secretagogues. These results obviated the need of a provocative test (Service, 1999).

A pancreatic head mass with 1.8 cm was evidenced by abdominal computerized tomography and transendoscopic ultrasound. After surgical enucleation, it was histopathologically confirmed to be an insulinoma showing a distinct immunostaining pattern for proinsulin and insulin compared with adjacent normal B cells of pancreatic islets. Six months after surgery, the patient showed an improvement in BPRS and HRSD scores in relation to baseline (68 to 18 and 21 to 8, respectively) with a better personal and social functioning. During follow-up no hypoglycemic episodes were detected. As the patient maintained mild depressive symptoms, the antidepressant was prescribed for one more year.

Discussion

Insulinomas impose a diagnostic challenge as they present with clinical features overlapping more common psychiatric syndromes.

These tumors produce high levels of insulin, even in the presence of hypoglycemia, resulting in a decrease in circulating blood glucose. Not surprisingly, since glucose is the major energy substrate in the adult brain supporting a complex metabolic processes essential for neuronal activity (McCrimmon, 2012) hypoglycemic episodes may induce neuropsychiatric symptoms and repeated periods of hypoglycemia can mimetize certain psychiatric states.

As reported in other cases (Harrington *et al.*, 1983; Service *et al.*, 1991; Dizon *et al.*, 1999), autonomic symptoms were overlooked and taken as consequent to hypoglycemia. However, this case is peculiar because the hypoglycemic episodes were initially judged as secondary to behavioral disturbances with food refusal and prolonged fasting. The adequate treatment of psychiatric symptoms associated with persistent episodic hypoglycemia was crucial to clarify the pathophysiology. A delay of two years occurred between the onset of symptoms and the correct diagnosis of insulinoma, which is in accordance with the literature revealing that these patients are commonly misdiagnosed with psychiatric and neurologic illnesses (Service *et al.*, 1991; Dizon *et al.*, 1999; Vig *et al.*, 2001; Ding *et al.*, 2010). This case highlights the difficulty in accurately recognizing an insulinoma.

Conflict of interest

None.

Description of authors' roles

All authors made substantial contributions to the conception and description of this case report. All authors contributed to drafting the paper and revising it critically for intellectual content. All authors approved of the final version of the paper.

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