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Case Report
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Insulinoma Presenting with Recurrent Right Sided Hemiparesis

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Abstract

Insulinomas are a particular type of neuroendocrine tumor that represents the most common cause of hyperinsulinemic hypoglycemia in adults. Women from 40 to 50 years old are the group of patients more frequently affected. Common clinical presentation of this entity includes diplopia, blurred vision, confusion, palpitations, and weakness. In this report of case, we present a 67-year-old woman with an atypical clinical presentation of insulinoma: She arrived at the emergency department with right-sided hemiparesis and mild confusion associated with a 48 mg/dl serum glycemia. All these symptoms in conjunction lead to the suspicion of a probable case of insulinoma in an adult patient.

Due to the inconclusiveness of the images previously obtained from magnetic resonance and the possibility to perform a 18F-DOPA PET-CT, we decided to use this molecular imaging tool to identify a pancreatic tumor with morphomolecular characteristics of insulinoma. Once the tumor was identified, a surgical procedure to enucleate was done and then the insulinoma was confirmed by pathological examination.

As a result of this case, we consider the 18F-DOPA PET-CT is a useful molecular imaging tool to characterize insulinomas when there is an atypical clinical presentation and we lack information from conventional morphological images because they are not conclusive or clear.

Keywords: Insulinoma; PET-CT; 18F-DOPA; Paresis; Hypoglycemia

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Introduction

Insulinomas, the most common cause of hyperinsulinemic hypoglycemia in adults, are pancreatic islet cell tumors with inappropriately high serum insulin production [1], described as a very small and hypervascularized lesion, usually measuring between 1 and 2 cm [2-5].

These are rare tumors, with an incidence of up to 10 cases per million inhabitants [6]. The average age of presentation ranges between 40 to 50 years [1], with women being more frequently affected [6]. 90% of cases are usually sporadic, benign and solitary [1,3-5] masses, whereas only 10% are hereditary, associated with the MEN-1 syndrome [6] representing 30% to 80% of these individuals [5]. Less than 2% of insulinomas are extrapancreatic [1].

We report a case of a 67-year-old female with an atypical clinical presentation of insulinoma, in whom a 18F-DOPA PET-CT scan facilitated the diagnosis.

Case Presentation

A 67-year-old female with no major cardiovascular risk factors presented to the emergency department with right-sided hemiparesis and mild confusion. Her husband noticed she was disoriented, with dysarthric speech and limited movements of her right-side body in the morning when she woke up. Upon arrival, 90 min after symptoms onset, she had fully recovered from the neurological deficit, and was alert and oriented. Her blood pressure was 120/70 mmHg, and her pulse was regular at 88 beats per minute. She felt tired, despite the neurological examination was normal.

She described two similar episodes in the past year. Previous cardiovascular studies revealed mild carotid atherosclerotic disease, sinus rhythm, and no brain lesions were identified on Magnetic Resonance Imaging (MRI). She had no clinical history of dyslipidemia, diabetes, hypertension,

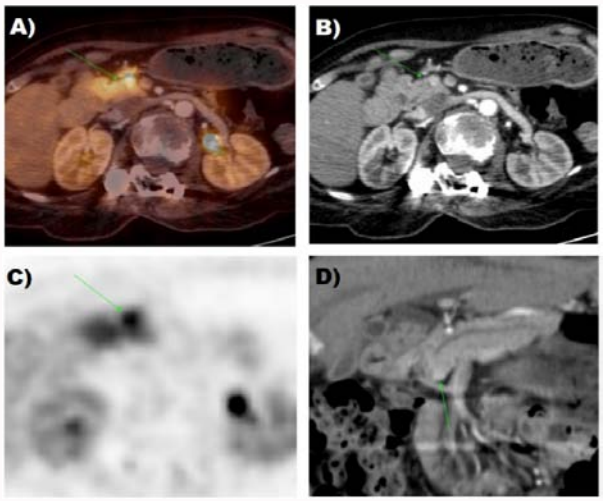

Figure 1: 18F-DOPA PET-CT images of patient's upper abdomen using iodinated IV contrast on axial (A and B) and coronal (C) plane, showing: A) small rounded lesion with abnormal increased radiotracer uptake (SUV max. 7.8) and arterial-phase contrast enhancement. B) PET image without TC fusion with radiotracer concentrated mostly on the pancreatic lesion topography.

smoking, or alcohol abuse. Her family history only highlighted a niece with nesidioblastosis since childhood.

A new brain MRI showed no abnormalities. Her laboratory tests in general were on normal values but her serum glucose levels report 48 mg/dL. She was treated with aspirin, clopidogrel, and intravenous dextrose, and admitted to the emergency ward for further studies.

Once admitted, after a fasting indication of 10 h, her serum glucose level was 41 mg/dL. No symptoms or neurological deficits were observed this time. Given the low blood glucose levels, serum insulin was requested, reporting a value of 4.60 uU/mL (normal range between 1.40-16.80 uU/mL). Because of the unexpected normal value of insulin and in order to explore a probable pancreatic image could be seen, an abdominal MRI was performed and it revealed a poorly defined 10 mm nodular image at pancreatic isthmus, with arterial contrast enhancement and apparently contacting the Wirsung duct (Figure 1).

Given the high suspicion of insulinoma due to the clinical presentation and the poor MRI morphological definition obtained, a 18F-DOPA PET-CT was done (Figure 2). Treatment with diazoxide was started, with good tolerance and no new episodes of hypoglycemia.

The lesion was morphological and molecularly characterized as a small rounded pancreatic lesion with abnormal increased radiotracer uptake, SUV max. 7.8, and with contrast enhancement in arterial phase of CT.

The patient underwent surgery two months later. The nodule was palpated by the surgeon on the topography described in the PET-CT, before successfully enucleating it during laparotomy. She had no neurological symptoms and all serum glucose measurements remained normal after surgery.

Histological sections showed a well-defined tumor proliferation composed of medium and small cells with eosinophilic cytoplasm with rounded or oval nuclei with granular "salt and pepper" chromatin, arranged in nests and cords surrounded by a desmoplastic stroma.

Figure 2: Quirurgical piece (insulinoma).

Immunohistochemistry was performed, with positive chromogranin and synaptophysin, confirming a well-differentiated neuroendocrine tumor.

Discussion

The presence of hypoglycemia raises the awareness of the possibility of insulinoma as a differential diagnosis. The successful localization of the insulinoma depends on sensitivity of abdominal CT and magnetic resonance imaging, representing 64% and 75%, respectively [1].

When conventional imaging modalities are inconclusive or unavailable, PET-CT imaging represents a helpful morphological and metabolic instrument to identify it.

In this case, MRI findings were inconclusive, while PET-CT with 18F-Fluorodihydroxyphenylalanine (18F-FDOPA) was able to find the tumor, this modality possess a 75% of sensitivity and 100% of specificity in different endocrinology scenarios and achieves an accurate morphomolecular assessment [7].

L-DOPA is an amino acid precursor that binds the LAT transporters of neuroendocrine cells and is converted to 18F-FDODA, finding its place in stored vesicles [8].

Other radiotracers can be used instead of 18F-DOPA, such as Somatostatin receptors (SST2) radioligands, 111In-pentetreotide as single photons emitter and 68Ga-labeled somatostatin analogues as positrons emitter. However only 60% of insulinomas express SST2 [9,10].

Conclusion

Insulinoma is an infrequent neoplasm, with fasting hypoglycemia as the most common symptom, in some cases, discrete neuroglycopenic symptoms such as hunger, vertigo, headache, weakness, blurred vision, focusless and confusion could be present. In a few cases, such as the one we are presenting, it can simulate a stroke [1,4,5]. This clinical presentation may or may not be preceded by autonomic symptoms [4].

In some cases, molecular imaging helps to evaluate the morphology and metabolic characteristics of lesions presenting with atypical clinical presentation associated with inconclusive conventional imaging modality. Although 18F-DOPA is not commonly used as an imaging instrument to identify insulinomas, it provides a molecular diagnosis support in order to characterize this type of tumors.

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