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Case Report

Munchausen Syndrome-Induced Hypoglycemia Following Successful Partial Pancreatectomy for Insulinoma: Diagnostic Challenges in Hypoglycemia Management

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Abstract

Introduction: Hypoglycemia is rare in individuals without diabetes. One cause of hyperinsulinemic hypoglycemia is insulinoma, a rare but serious condition. Careful evaluation is essential to exclude other causes, including factitious hypoglycemia, which involves the intentional induction of low blood glucose levels without any apparent external gain, primarily to seek medical attention. Munchausen syndrome is a severe form of factitious disorder and presents a diagnostic challenge.

Case Presentation: We report a case of a female patient who, at age 14, began experiencing signs and symptoms of hypoglycemia that worsened over time. At age 27, she was admitted to the emergency room following a seizure due to a hypoglycemic episode and was initially evaluated for hyperinsulinemic hypoglycemia caused by insulinoma. Surgical intervention with distal pancreatectomy improved her symptoms. However, 2-3 weeks post-surgery, she experienced recurrent hypoglycemic episodes. After an extensive investigation over ten months as an inpatient receiving continuous intravenous glucose infusion, factitious hypoglycemia was suspected and ultimately confirmed.

Conclusion: This case highlights the importance of a thorough diagnostic evaluation to prevent unnecessary invasive procedures. To our knowledge, no similar case has been reported. When factitious disorder is suspected, a psychiatric evaluation is crucial. Long-term follow-up with family support and a multidisciplinary team is essential for effective management.

Keywords: Insulinoma; Factitious hypoglycemia; Munchausen syndrome.

Introduction

Hypoglycemia is a rare condition in individuals not using medications that alter glucose levels. Therefore, its diagnosis should be confirmed using Whipple's triad (low glucose levels, accompanied by compatible symptoms that improve with glucose correction), followed by further investigation with the fasting test [1], which allows for the classification of hypoglycemia as either hyperinsulinemic or non-hyperinsulinemic.

One cause of hyperinsulinemic hypoglycemia is insulinoma, a rare, insulin-secreting pancreatic neuroendocrine tumor that is most often benign, solitary, sporadic, well-differentiated, and exhibits a slight female predominance [2,3]. However, careful evaluation of additional fasting test parameters, beyond insulin levels, is essential to exclude, among other causes, factitious hypoglycemia.

Factitious hypoglycemia refers to the intentional induction of low blood glucose levels to assume the role of a sick individual, done consciously and deliberately, and may occur even without any apparent external gain, with the primary aim of seeking medical attention. Munchausen syndrome is a severe, chronic form of factitious disorder, characterized by repeated hospitalizations and an extensive medical history, including unnecessary diagnostic and therapeutic procedures [4]. This disorder differs from malingering, where the motivation is financial gain or the avoidance of legal responsibilities, and individuals typically seek to avoid invasive procedures or therapies [5].

We present a case of factitious hypoglycemia that occurred following successful surgical treatment of an insulinoma, emphasizing key aspects of the diagnostic evaluation to minimize or prevent harm from unnecessary invasive medical interventions. To the best of our knowledge, no similar case has been reported to date.

Case Presentation

A female patient with a past history of polycystic ovary syndrome (PCOS), without the use of medications, experienced recurrent episodes of adrenergic and neuroglycopenic symptoms starting at the age of 14, occurring primarily during fasting, particularly in the morning, with improvement after eating. These episodes were not investigated at that time.

At the age of 27, she was admitted to the emergency room following a seizure due to a hypoglycemic episode (capillary blood glucose <

55 mg/dL). Upon admission, Whipple's triad was confirmed.

Seen the long history of hypoglycemic episodes, the hypothesis of insulinoma was raised and an investigation was carried out with the fasting test, which revealed hyperinsulinemic hypoglycemia (glucose 53 mg/dL, normal value > 55 mg/dL; insulin 11.5 µU/mL, normal value < 3 µU/mL) and elevated C-peptide (3.83 ng/mL, normal value < 0.6 ng/mL) (Table 1).

The patient had normal calcium and pituitary hormone levels, with no dermal lesions such as trunk collagenoma or nasal angiofibroma. Her personal and family history was negative for tumors associated with genetic syndromes, including multiple endocrine neoplasia type 1 and tuberous sclerosis.

During hospitalization, the patient developed Raynaud's phenomenon, leading to the diagnosis of Raynaud's disease, with no additional symptoms and signs suggestive of autoimmune disease.

Imaging studies were conducted to investigate a pancreatic tumor, and abdominal magnetic resonance imaging (MRI) revealed a 1.4 cm nodule in the pancreatic tail (Figure 1). Laparoscopic distal pancreatectomy was performed, confirming the diagnosis of insulinoma via immunohistochemistry (IHC) (Figures 1, 2, 3).

She was discharged four days after surgery, as there was complete remission of hypoglycemia during the perioperative period, without complications. Hypoglycemic episodes recurred two weeks later, and the patient was admitted for further investigation.

Imaging studies did not reveal any new pancreatic lesions, although 68Gallium-DOTATOC PET/CT suggested a lesion in the head of the pancreas, located near a region of physiological capture, without anatomic correspondence by aggregated computed tomography (Figure 4).

Somatostatin analog therapy was initiated, but there was no significant improvement after several weeks, leading to discontinuation two months later. Due to recurrent hypoglycemia and new seizures, the patient required continuous intravenous glucose infusion through central venous access and prolonged hospitalization.

Due to Raynaud's disease, autoimmune hypoglycemia was excluded based on negative anti-insulin and antinuclear antibody tests. New insulin and C-peptide samples were collected during hypoglycemic episodes, but the results were inconclusive, with a borderline C-peptide value during hyperinsulinemic hypoglycemia, both with and without somatostatin analog therapy (Table 1). Factitious hypoglycemia was then suspected, and the patient was kept in an isolated bed for observation, continuing to experience hypoglycemic episodes.

Due to the inconclusive diagnosis, the patient was referred to our tertiary center for further investigation of insulinomatosis, nesidioblastosis, occult malignant insulinoma, or multiple insulinomas associated with inherited disorders. A new fasting test was again inconclusive (Table 1). Other attempts to localize a potential occult tumor using MRI and endoscopic ultrasound were unsuccessful. While further fasting test, measurements were being conducted and the pathology of the surgical specimen was reviewed, the patient experienced recurrent hypoglycemic crises during hospitalization, requiring intravenous glucose infusion through central venous access. As a result, pancreatic arteriography was performed, followed by a selective arterial calcium stimulation test, which returned negative results.

Shortly thereafter, the complete results of the fasting test revealed hyperinsulinemic hypoglycemia (glucose 42 mg/dL, insulin 43.3 mcU/mL) with inappropriately normal values for C-peptide (0.5 ng/mL, normal value < 0.6 ng/mL) and proinsulin (2.39 pmol/L, normal value < 5 pmol/L), negative ketonemia, and a positive response to glucagon, suggesting malicious and surreptitious insulin administration (Table 1). In parallel, insulinomatosis and nesidioblastosis, which had also been considered potential etiologies, were definitively excluded after a thorough review of the surgical specimen (Figure 3).

The patient remained hospitalized for 10 months across various hospital services, displaying a strong inclination to assist the nursing team with organizing supplies, independently adjusting the glucose infusion rate, refusing medication, and engaging in conflicts with other patients. Both the psychology and psychiatry teams were involved in her care.

Due to suspicious behaviour, a review of her personal belongings revealed vials of human insulin and injection needles. The patient admitted to injecting insulin to simulate symptoms following pancreatectomy, also disclosing a history of childhood trauma, sexual abuse, and previous suicide attempts.

Following psychiatric evaluation, a diagnosis of borderline personality disorder was made, and Munchausen syndrome was diagnosed. She initiated treatment with fluoxetine and quetiapine. The patient was discharged after securing a support group and arranging psychiatric follow-up. At the one-year follow-up, she missed several scheduled appointments and was readmitted to the emergency department six months ago due to a new hypoglycemic episode. Since then, no further hypoglycemic events have occurred. In her most recent consultation, she reported that she was enrolled in a nursing technical course and was performing excellently. Genetic testing for *MEN1* was performed, and no pathogenic variants in the *MEN1* gene were identified by Sanger sequencing or Multiplex Ligation-dependent Probe Amplification (MLPA) assay.

Fasting Test						
Date	**Glycemia (> 55 mg/dL)**	**Ketonemia (< 1 mmol/L)**	**Insulin (< 3 µU/mL)**	**C-Peptide (< 0,6 ng/mL)**	**Proinsulin (< 5 pmol/L)**	**Glucagon Test Response**
Before partial pancreatectomy	53	-	11.5	3.83	-	-
7 months after partial pancreatectomy	41	-	54	0.6	-	-
11 months after partial pancreatectomy	23	0.2	30.3	0.45	-	Positive (glycemia: 23 -> 86 (Δ > 25)
12 months after partial pancreatectomy	45	0.1	43.3	0.5	2.39	Positive (glycemia: 45 -> 96 (Δ > 25)

Table 1: Typical biochemical pattern of endogenous hyperinsulinemic hypoglycemia documented by the fasting test performed prior to partial pancreatectomy for insulinoma, followed by an erratic and inconclusive biochemical profile due to early recurrent postoperative hypoglycemia, ultimately leading to laboratory findings strongly suggestive of factitious hypoglycemia, likely resulting from surreptitious exogenous insulin use.

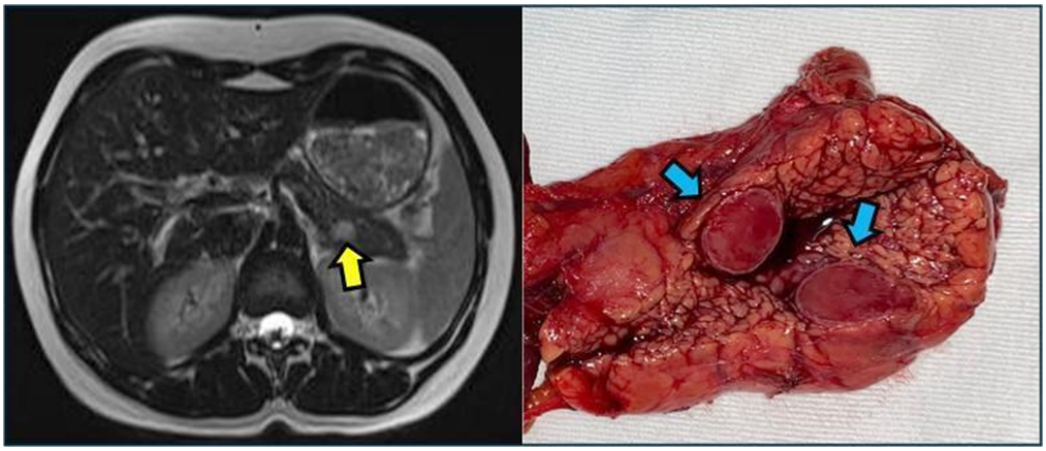

Figure 1: Preoperative radiological image and macroscopic view of the pancreatic tumor resected during subtotal pancreatectomy. A. Abdominal magnetic resonance imaging: A nodular formation (yellow arrow) at the body-tail junction of the pancreas, measuring 1.4 x 0.9 cm; B. Surgical specimen following partial pancreatectomy, showing a solitary pancreatic nodule at the body-tail junction. The nodule, sectioned in half (blue arrows), is wine-colored with well-defined borders, and measures 1.5 x 1.3 x 1.0 cm.

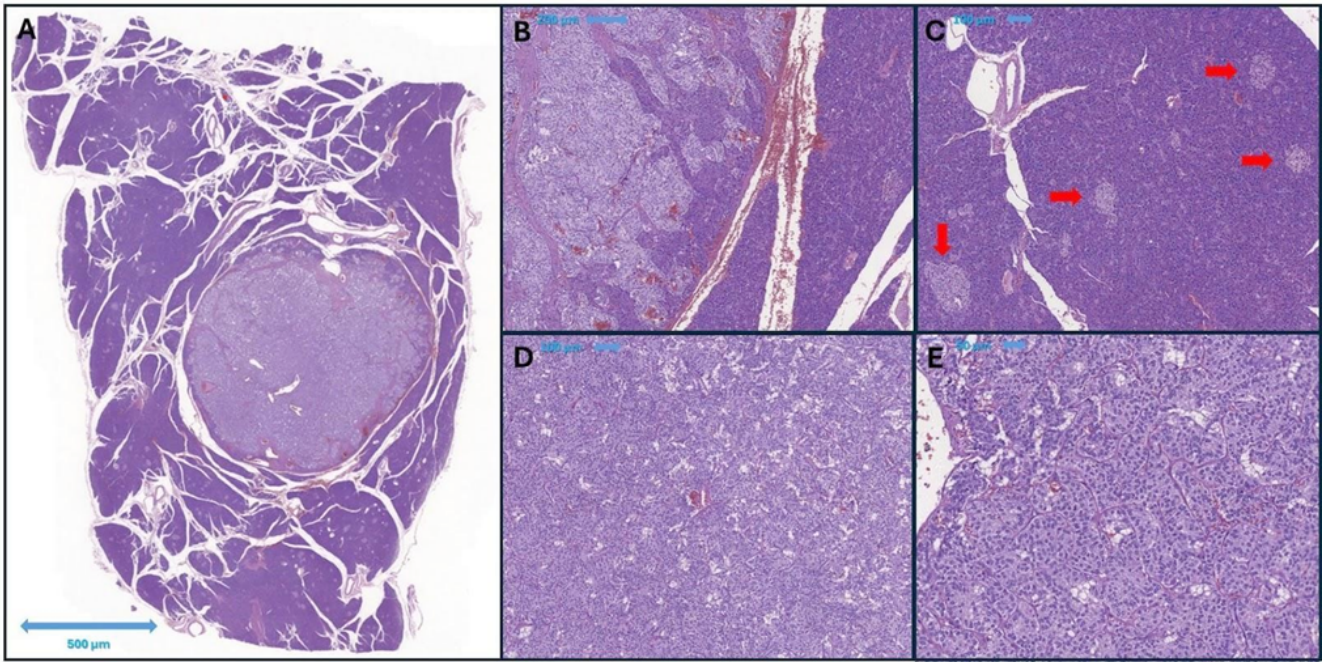

Figure 2: Histology of the pancreatic neuroendocrine tumor (NET) located at the body-tail junction. A-E: Stained with hematoxylin and eosin. A. Microscopic panoramic view of the NET measuring 1.5 x 1.3 x 1.0 cm, unifocal, well-demarcated, with clear borders free of neoplasia, and no evidence of angiolymphatic or perineural invasion, nor lymph node involvement (0/7) (scale: 500 µm). B. Transition between tumor and normal pancreatic tissue, with the NET on the left and normal tissue on the right (scale: 200 µm). C. Normal pancreatic tissue, including multiple endocrine cell islets (red arrows) (scale: 100 µm). D. Tumoral tissue (scale: 100 µm). E. Tumoral tissue at higher magnification (scale: 50 µm).

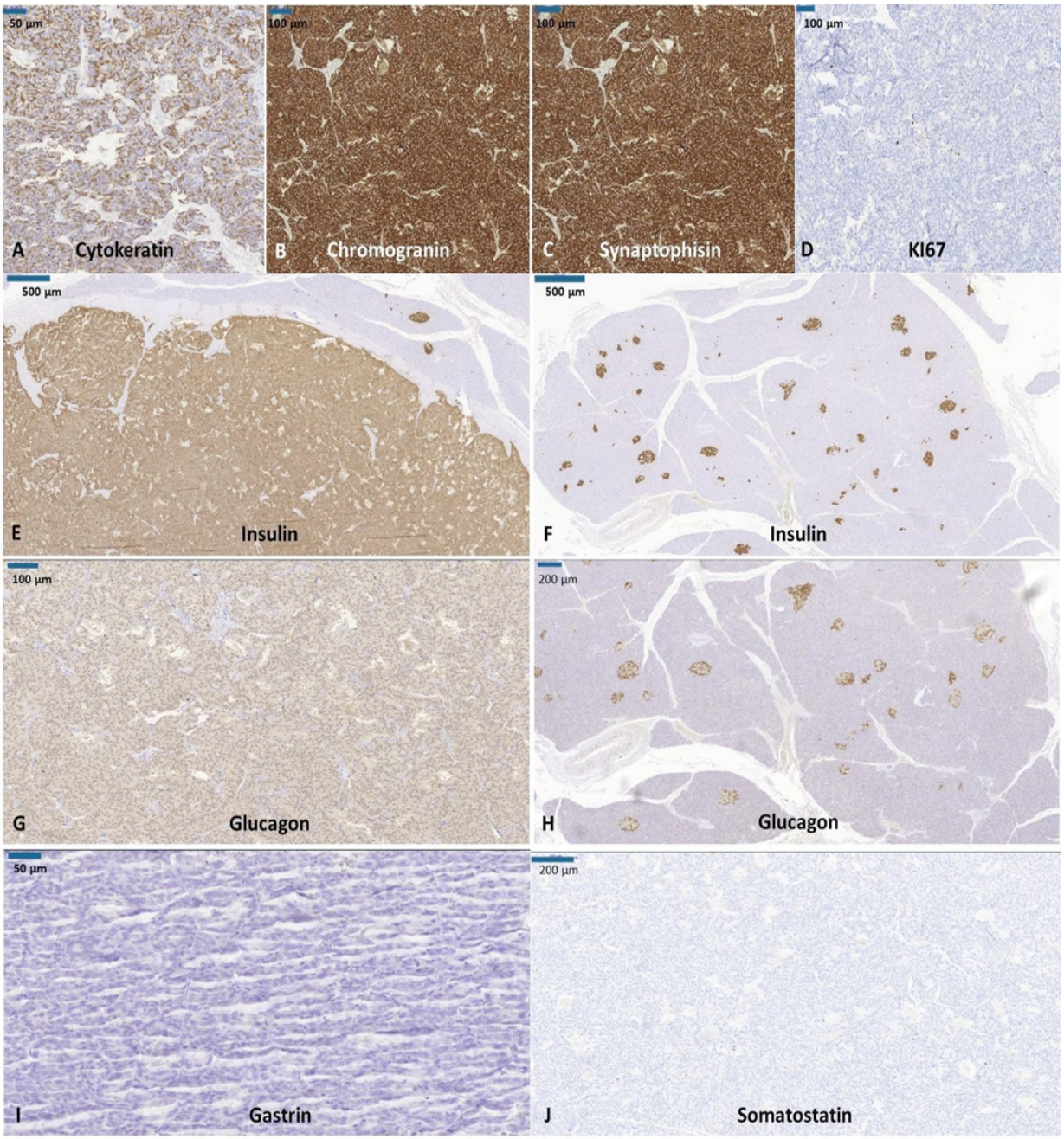

Figure 3: Immunohistochemistry (IHC) demonstrating an insulin-secreting grade 1 (G1) well-differentiated neuroendocrine tumor (WDNET). A-C. Positive IHC for cytokeratin (CK8/18) (A), chromogranin (B), and synaptophysin (C), classifying the tumor as neuroendocrine (positive for AE1/AE3, not shown). D. Ki67 labelling index < 3%, classifying this WDNET as G1. E-F. Positive IHC for insulin in the neuroendocrine tumor (E) and in normal islet cells (F). G-H. Negative IHC for glucagon in the insulin-secreting NET (G) and positive in normal islet cells (H). I-J. Negative IHC for gastrin (I) and somatostatin (J). Scales in µm.

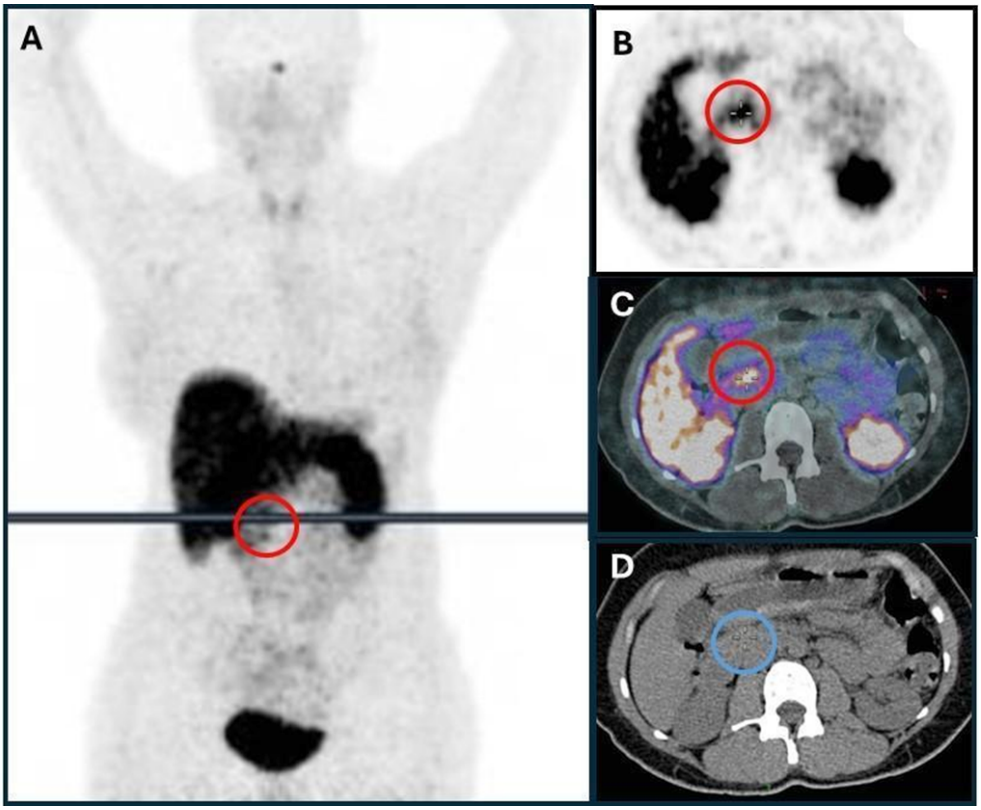

Figure 4: False-positive results suggesting a pancreatic tumoral lesion despite an inappropriately normal C-peptide value, which was borderline for the laboratory diagnosis of hyperinsulinemic hypoglycemia (see Table 1). A-C. 68Gallium-DOTATOC PET/CT showed a focal area of uptake (SUV max = 6.9) in the uncinate process/head of the pancreas (red circle); D. with no corresponding anatomical findings on computed tomography (blue circle) and another area of uptake near the site of previous surgical manipulation (SUV max = 4.9) (not shown). MRI suggested a possible 3-mm lesion in the same location within the uncinate process (not shown). Endoscopic ultrasound was normal in this region, and biopsy identified an inflammatory lymph node near the area of prior surgery (not shown). Given the inconclusive results of subsequent biochemical tests, pancreatic arteriography was performed with a selective arterial calcium stimulation test, which yielded a negative result (not shown).

Discussion

We reported a case diagnosed with failure of the resolution of hypoglycemia after distal pancreatectomy for insulinoma in a young adult woman. Surgery is the cornerstone of treatment of insulinoma and scenarios of unsuccessful surgical treatment occur exceptionally. In this context, the main causal events rememorated by specialists associated with primary surgical failure are represented for inherited syndromic forms of insulinoma, especially by multiple endocrine neoplasia type 1 and exceptionally by tuberous sclerosis, by insulinomatosis and by nesidioblastosis, malign insulinoma or by initial failure of radiological localization of solitary insulinoma. Ultimately, in a case with a previous background of proved organic, the hypothesis of factitious hypoglycemia may be underestimated. Thus, this case teaches us that the basic rules of investigation in cases with recurrent hypoglycemia must be fully remade step by step.

Occult insulinoma was the first differential diagnosis to be excluded in the present case as insulinoma was found with complete remission of hypoglycemia up to 2-3 weeks after surgery. Thus, inherited disorders frequently related with multifocal insulinoma were investigated and appropriately excluded. Around 5% to 10% of insulinomas are associated with multiple endocrine neoplasia type 1 (*MEN1*) syndrome [3,6,7]. The youth and recurrence of hypoglycemia were points favouring this hypothesis [6,7]. However, the absence of primary hyperparathyroidism observed in at least 80% of *MEN1* cases at 30y, the absence of combination of other *MEN1*-related tumors, especially the frequent combination of non-functioning pancreatic neuroendocrine tumors with insulinoma, absence of *MEN1*-related dermic lesions, negative familial history and, absence of germline *MEN1* mutation excluded MEN1 [8,9].

In parallel, tuberous sclerosis was clinically excluded and reinforced by exceptionality of the functioning pancreatic NETs reported in this disorder [10,11]. Insulinomatosis, another extremely rare hypoglycemic disorder, was suspected [12], however, careful microscopic review of the surgical specimen excluded occurrence of micro-tumor and smaller clusters of neuroendocrine cells scattered within the macroscopically normal pancreas typically found in this disorder, corroborated by positive IHC for glucagon in normal islets and negative within tumoral bed. In addition, absence of nodular or diffuse islet cells hyperplasia/hypertrophy in adjacent area of the insulinoma excluded other rarer forms of hyperinsulinemic hypoglycemia as adult-onset non-insulinoma persistent hyperinsulinemic hypoglycemia syndrome (NIPHS). Yet, by the presence of Raynaud's phenomenon, negative antibody measurements tried against autoimmune hypoglycemia.

There is no precise estimate of the prevalence of factitious hypoglycemia in the literature. In addition to case reports, we identified a review of 31 cases of adult diabetic and non-diabetic patients with factitious hypoglycemia. The mean age of the patients was 33.7 (±13.5) years, with a predominance of women (female: male ratio 4.3:1). Among them, 38% had medical occupations or prior medical training, 53% had diabetes mellitus, and 41% had a positive psychiatric history [13]. Psychiatric literature supports this finding, indicating that patients with factitious disorders often have a history of extensive healthcare utilization, provide inconsistent, selective, or misleading biographical information, exhibit evasive behaviour during history-taking, present with atypical disease manifestations, show normal or inconclusive diagnostic results, demonstrate a strong desire for medical procedures, and display aggression or defensiveness toward healthcare staff [8,13].

In this case, upon admission to our service, somatostatin analogs had been discontinued for three months. This medication is known to induce or worsen hyperglycemia in patients with metastatic NET. Radiological exams, including endoscopic ultrasound, remained negative. Hyperinsulinemic hypoglycemia was confirmed, but C-peptide was inappropriately normal, and proinsulin was unavailable. Due to severe hypoglycemia requiring continuous intravenous glucose, pancreatic arteriography was performed while awaiting proinsulin results. The selective arterial calcium stimulation test failed to localize the insulinoma.

Subsequent biochemical tests documented symptomatic hypoglycemia with hyperinsulinemia and inappropriately normal C-peptide and proinsulin, suggesting exogenous insulin use. Meanwhile, the surgical specimen review excluded insulinomatosis and NIPHS, and genetic testing ruled out *MEN1*. A final review of the patient's belongings uncovered vials of human insulin and injection needles.

Excessive and redundant testing incurs significant financial costs and wastes valuable time for both patients and the healthcare system. Furthermore, the risks associated with unnecessary hospital admissions and invasive procedures, such as pancreatic catheterization and, in some cases, even pancreatectomy, must be carefully evaluated [14]. Factitious hypoglycemia can occur in both adults and children, with Munchausen syndrome by proxy being a related condition, primarily observed in the pediatric population [14–16].

This case highlights the importance of thoroughly considering all diagnostic steps, beyond the assessment of C-peptide levels and the insulin/C-peptide ratio. Yet, proinsulin values higher than 22 pmol/l during the fasting test shows high sensitivity and specificity for the diagnosis of insulinoma [12,17]. In particular, the inappropriately normal proinsulin measurement observed in this case was crucial in establishing the diagnosis, as proinsulin is typically normal or low only in cases of factitious hypoglycemia due to surreptitious exogenous insulin use. However, proinsulin may also be normal or low in the rare Doege-Potter syndrome, a condition that was easily excluded in this case due to its association with hypoinsulinemic hypoglycemia and a distinct clinical presentation, often caused by IGF2-secreting giant mesenchymal tumors [18,19].

A similar case reported by Texeira et al. confirmed factitious hypoglycemia due to exogenous insulin administration in a patient, with pancreatic histological evaluation revealing nesidioblastosis [20]. As expected, in most of these cases, C-peptide levels were undetectable in the presence of hyperinsulinemia and hypoglycemia, in contrast to our patient, who exhibited an inappropriately normal C-peptide value. Interestingly, our patient's behaviour mirrored that described by Teixeira et al., characterized by rudimentary social interactions that were resolved through assuming the role of a chronically ill person. As a result, unnecessary prolonged hospitalizations enabled her to establish a beneficial social network with healthcare professionals [20]. Likely, the inadvertent use of insulin, by perpetuating the symptoms of hypoglycemia, became a potent mechanism for the patient to protect herself from the psychological distress associated with the abrupt disruption of her social network following her first hospital discharge.

The use of various insulin assays can be a valuable tool for excluding or confirming factitious hypoglycemia due to exogenous insulin analogs use. Human insulin consists of two polypeptide chains stabilized by disulfide bonds. Insulin analogs, which are produced through amino acid substitutions or additions in these chains, exhibit distinct immunoreactivity profiles when compared to human insulin [21,22]. Different commercial assays exhibit varying cross-reactivity profiles, which can be leveraged to assist in diagnostic investigations [23–25]. In studies comparing different assays, the Roche Elecsys® E170 assay demonstrated the best performance, reacting only to human insulin and insulin glargine

[26]. Additionally, the Mayo Clinic has validated an immunoassay for detecting insulin analogs [27]. In our case, the patient was using human insulin, which limited the use of alternative assays to aid in the diagnosis.

In addition to exogenous insulin use, it is important to consider other forms of factitious hypoglycemia, such as the use of hypoglycemic medications. A French review of 129 patients without a diabetes diagnosis and a history of hypoglycemia, conducted between 1997 and 1999, found a 17% prevalence of sulfonylurea use, as determined by serum medication levels [28]. There are also reports of patients using both oral medications and insulin, which can make diagnosis even more challenging, potentially leading to variable insulin and C-peptide laboratory patterns [29].

Conclusion

Investigating hypoglycemia can be challenging and requires the implementation of appropriate diagnostic protocols. Primary surgical failure in insulinoma treatment is rare and typically associated with hereditary syndromic forms, tuberous sclerosis, insulinomatosis, nesidioblastosis, malignant insulinoma, or initial failure in radiological tumor localization. Even in patients with a seemingly well-defined diagnosis, such as our case with anatomopathological confirmation of insulinoma, it is crucial to exclude factitious hypoglycemia by reviewing the investigation step by step. This should include assessing proinsulin and C-peptide levels, measuring serum sulfonylurea levels, and considering the use of different insulin assays. An incorrect diagnosis may lead to inappropriate invasive and surgical procedures with lasting consequences, such as exogenous pancreatic insufficiency and insulin-dependent diabetes mellitus. When factitious disorder is suspected, psychiatric evaluation is essential. Long-term management of such patients requires a multidisciplinary team, including psychiatric and psychological support, general practitioners, social workers, and family involvement.

Disclosure statement: The authors report there are no competing interests to declare.

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