



Research Article

DOI: 10.36959/608/465

Recurrent Hypoglycemia as a Presenting Symptom for Rare IGF-2 Secreting Retroperitoneal Spindle Cell Neoplasm

David Thornton, MD¹, Lux Shah, MPH^{2*}¹Rutgers St. Barnabas Medical Center, USA²St. George's University School of Medicine, True Blue, Grenada

Introduction

Spindle cell neoplasms are a broad group of connective tissue cancers that have spindle-shaped cells under microscopy [1]. Solitary Fibrous Tumors (SFT) are a rapidly growing and rare spindle cell neoplasm that generally occur in the thorax and comprise roughly 2% of all soft-tissue masses [2]. Due to their rarity, it is difficult to ascertain a true incidence rate. Memorial Sloan-Kettering Cancer Center has studied their cases of SFTs from 1982-2000, inclusively. This data can shine light on the rarity of this malignancy along with providing a good look into the varied presentations and patterns of SFTs. In the 18 year study period, 134 cases of SFTs were identified, 55 cases were excluded, and 79 were studied further [3]. Out of the 79 confirmed cases, 1 was retroperitoneal and 54 were thoracic masses. The remaining 24 were varied between the head/neck, pelvis, and extremities. The most common characteristics for the tumors were that they were non-necrotic (77%), had no malignant component (80%), lacked macroscopic surgical margins (95%), and lacked microscopic surgical margins (82%). A statistically significant amount of patients with extra thoracic SFTs were symptomatic (83%). An unexpected symptom for someone with a solid tumor is recurrent hypoglycemia.

Non-islet cell tumor hypoglycemia is a rare and dangerous disease for many reasons. Particularly, the hypoglycemic events that accompany these tumors can surprise patients and put them in a dangerous position. A common cause of these phenomena is a solid tumor secreting IGF-2 and increasing the levels of this insulin analog in the bloodstream [4]. In fact, the name Insulin-like Growth Factor-2 is corroborated by the NLM's Medical Subject Heading definition for IGF-2 which includes stating that, "it has growth-regulating, insulin-like, mitogenic activities" [5]. Many patients with non-islet cell tumor hypoglycemia will present with new onset of hypoglycemic events and signs of hyperinsulinemia. On blood tests, insulin and C-peptide will be low [6]. This finding can clue in to the fact that a non-islet cell tumor that secretes IGF-2 should be explored as a differential diagnosis. The next step is to check for blood IGF-2 levels and see how they coincide with hypoglycemic events. Surgical resection of the mass is

the standard accepted treatment for localized SFTs [7]. It is also prudent to check IGF-2 levels postoperatively along with monitoring blood sugar for any further hypoglycemic events.

Lastly, it is important to emphasize the rarity and dangers involved with a retroperitoneal solitary fibrous tumor that secretes IGF-2. The rarity of this disorder is so pronounced that it is hard to find adequate data to estimate the incidence of this presentation. The dangers of this pathology stem from the fact that the patient likely has no reason to know or prepare for recurrent hypoglycemic events. Up to 10% of deaths in Type 1 diabetics are from hypoglycemic events [8]. These are people who are warned about these events and taught how to combat them. A non-diabetic middle aged man, who has a rapidly-growing IGF-2 secreting retroperitoneal mass, has little to no way to prepare for hypoglycemic events. We present this exact case of recurrent hypoglycemia and enlarging abdominal mass with subsequent diagnosis, treatment, and recovery.

Case Report

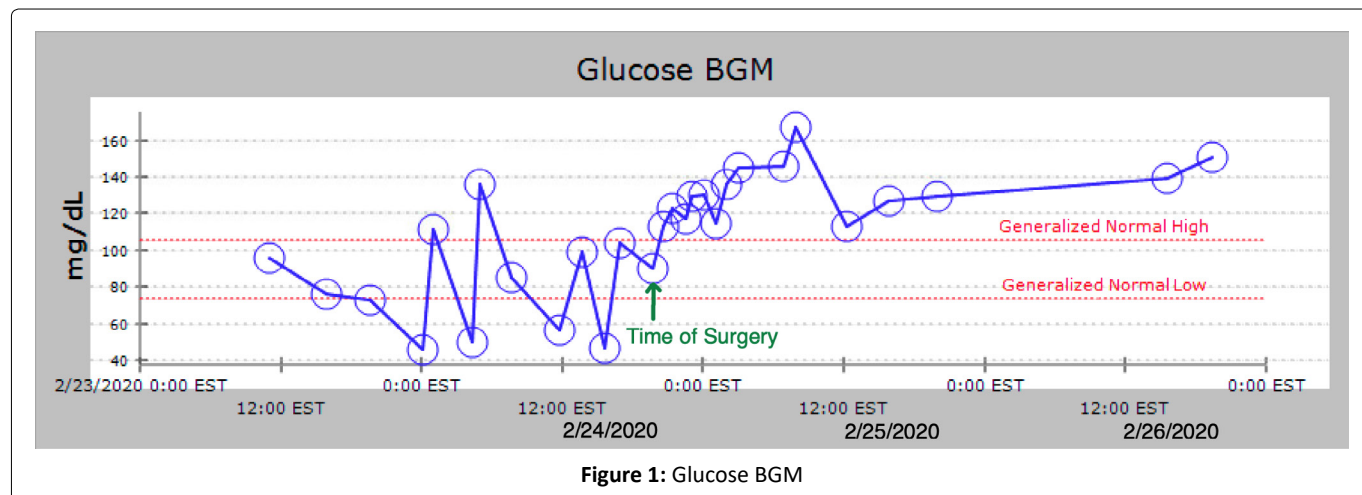
A 53-year old Asian male presents to the ED with his son who states that the patient had an episode of confusion that lasted 15-20 minutes. The son states that this kind of event has happened 2-3 times in the past week. These episodes were described as a "blank" appearance with confusion and inability to answer questions appropriately. The family states there have not been any motor symptoms or headaches. For the past 3 weeks, the patient's wife noticed multiple similar episodes with no loss of consciousness, falls, or trauma.

***Corresponding author:** Lux Shah, MPH, 4th Year medical student, St. George's University School of Medicine, True Blue, Grenada, USA, Tel: +473-444-4175

Accepted: April 19, 2022

Published online: April 21, 2022

Citation: Thornton D, Shah L (2022) Recurrent Hypoglycemia as a Presenting Symptom for Rare IGF-2 Secreting Retroperitoneal Spindle Cell Neoplasm. *J Surgical Endocrinol* 4(1):146-147



Concurrently, the patient and family complain of a growing abdomen that has been getting more firm. The patient denies any severe abdominal pain, constipation, diarrhea, or bloody stool. The patient associates his abdominal tenderness with his new exercise regimen revolving around core workouts. The patient is a caregiver who lives with his wife. There is no medical history per the patient and he does not take any medications at all. No alcohol since high school and has never smoked or used illicit drugs. There is no family history aside from his mother having a stroke.

On physical examination, vitals are within normal range, height is 160 cm and weight is 70.5 kg (BMI 27.5). Physical examination shows no abnormalities on abdominal examination and neurologic examination. Of note, lab work showed a blood glucose of 40 upon arrival to the ED. Due to abdominal pain, abdominal CT scan was ordered and showed, "a large heterogeneous abdominopelvic mass eccentric to the right" that measured approximately 15 x 23 x 26 (AP x transverse x CC). The mass included small calcifications and demonstrated areas of hypervascularity.

The mass was biopsied and returned from pathology with a diagnosis of, "spindle cell tumor, low grade; pending outside consultation for definite diagnosis." The sample was sent to Memorial Sloan-Kettering Cancer Center for consultation and was returned with a diagnosis of, "Solitary Fibrous Tumor." The patient had recurrent blood sugar drops during his preoperative hospital stay, this is shown in the graph below.

Exploratory laparotomy was performed with resection of retroperitoneal sarcoma. The mass was retroperitoneal

and had attachments to the bladder and colon. Images of the mass are included below which show the vascularity of this large mass. Postoperatively, the patient did not have any hypoglycemic events. The recovery for the patient was uneventful and proper glucose control seemed to occur without the need for blood sugar interventions. Once again, the graph below portrays the change that occurred between preoperative sugar control and postoperative sugar control.

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