BACKGROUND

Solitary fibrous tumors are an uncommon group of spindle cell neoplasm of submesothelial origin with malignant potential. The majority of these tumors are found to be originating from the thoracic cavity, but sometimes they do occur at different sites including the pelvis, muscles, and soft tissue. Malignant SFT are hypercellular, mitotically active, and associated with larger tumors. This tumor spread hematogenously, most commonly to the lungs. Incidence of SFT ranges from about 0.2/100,000/years, mostly present in sixth to eighth decades of life with slightly male predominance. On rare occasions, such SFT have been found to be associated with many paraneoplastic syndrome and secret insulin-like growth factor II (IGF-II), which causes hypoglycemia. Surgical excision has been found to be the treatment of choice for hypoglycemia in patients with solitary fibrous tumors. We hereby report a case of a patient with malignant fibroma who presented with a recurrent episode of hypoglycemia spells, and later on, it was diagnosed that hypoglycemic spells were secondary to fibroma.

KEYWORDS
fibroma, hypoglycemia, insulin-like growth factor II

CASE DESCRIPTION

A 45-year-old male patient with a past medical history of biopsy-proven malignant fibroma successfully removed...
by exploratory laparotomy presented to the emergency department with recurrent episodes of hypoglycemia and loss of consciousness from the last three months and one episode of a tonic–clonic seizure this morning. The seizure episode lasted for 2 min and was associated with tongue biting and urinary incontinence. Examination findings were unremarkable except for tongue laceration and pulse rate of 105 bpm. Fingerstick glucose levels were low, and he was given intravenous bolus dextrose. He was admitted for further evaluation, and baseline investigations, complete metabolic profile, urine, and blood toxicology were sent for analysis, the results of which were unremarkable. U/S abdomen showed b/l hydronephrosis and computed tomography (CT) abdomen and pelvis revealed huge lobulated soft tissue enhancing mass in the small bowel mesentery and right paracolic gutter measuring 11.8*12.9*11.2 cm (Figure 1). Another such lobulated soft tissue mass was seen in the right pararenal area causing compression of the right ureter, while another similar mass was noted adjacent to the left external iliac vessel causing compression of the left lateral urinary bladder wall. He underwent two episodes of hypoglycemia and was reverted by giving an i/v bolus of dextrose. To evaluate the cause of recurrent hypoglycemia with loss of consciousness, insulin-like growth factor I (IGF-I), insulin-like growth factor II (IGF-II), and C-peptide levels were sent for analysis, the results of which showed high level of IGF-II and IGF-II/IGF-I ratio and low level of IGF-I and C-peptide levels (Table 1).

The patient was scheduled for elective laparotomy, and the pelvic masses were removed. Post-op IGF-I, IGF-II, and C-peptide levels were normal, and he had no episode of hypoglycemia thereafter. He was discharged vitally stable and was advised to follow up after 2 weeks.

3 | DISCUSSION

Solitary fibrous tumors (SFTs) are rare growths of soft tissue, either benign or malignant and only 10%–15% of the tumor can convert into malignant behavior (MFT). Usually, this kind of tumor grows very slowly and may not cause any signs and symptoms until it becomes very large. They can arise from different locations including the breast, kidney, prostate, spinal cord, head, neck, oral cavity, pleural cavity, abdominal cavity, and retroperitoneum even though the liver is not often involved. Morphologically, this tumor resembles brown, partially encapsulated, pleasantly circled masses, with a rubber-like consistency. Microscopically, these tumors are patternless and composed of round to oval epithelial-like cells with pleomorphic nuclei and collagen strands in between. The tumor antigenic profile of MFT is very similar to gastrointestinal stromal tumor (GIST) and that is why can be misdiagnosed with GIST. It is important to confirm CD117, which is a basic marker that distinguishes MFT and GIST, to avoid such an error. The pathological findings seen in malignant fibrous tumors are as follows: tissue necrosis, high cellularity, high mitotic activity, and hemorrhagic change which differentiate it from benign SFTs.

These fibrous tumors also secrete insulin-like growth factors (IGF), for example, IGF-I, and IGF-II, and these IGFs have various effects on growth and differentiation. IGF-I binds to a group of six homologous proteins, widely distributed in the body and mediates the actions of GH, and helps to coordinate the growth and differentiation of various tissues and organs, while IGF-II is different than IGF-I which is produced in an autocrine or paracrine fashion which acts via non-signaling IGFII receptor. Various endocrine symptoms in MFTs, that is hypoglycemias, are due to abnormal paraneoplastic secretion of mature IGF-II, and the signs and symptoms of hypoglycemia help in the early diagnosis of tumor. Mature IGF-II is a 7.5 Kilodalton, 67

![FIGURE 1 CT abdomen with oral contrast shows a well-defined oval hypoattenuating mass in the region of descending colon.](image)

| Tests         | Results       | Reference Level |
|---------------|---------------|-----------------|
| IGF-I         | 65 ng/ml      | 90–278 ng/ml    |
| IGF-II        | 1325 ng/ml    | 267–616 ng/ml   |
| IGFII:IGFI ratio | 20.38         | <10             |
| C-peptide level | 0.687 ng/ml  | 1.1–1.4 ng/ml   |

TABLE 1 Laboratory findings of the patients
amino acid peptide vs immature 11–18 Kilodalton, >67 amino acid, and it can bind to IR or IGF-IR and will increase the intake of glucose through autocrine and paracrine activity, while big immature IGF-II is abnormally produced through abnormal post-translational modification of a Pro-IGF-II precursor in tumors. This big immature IGF-II can easily cross the endothelium and get access to the insulin receptors (IR) of parenchymal cells and cause refractory hypoglycemia with the help of binary complex formation with IGF-binding proteins (IGFBP). 13 This case gives the significance of unexplained hypoglycemia symptoms associated with the extrapleural SFT, particularly when the pro-IGF-II/IGF-II and IGF-II/IGF-I ratio is high. In some cases, the total serum IGF-II level can be normal that is why the pro-IGFII/IGFII and IGFII/IGFI ratios are helpful to diagnose the tumor. The IGF-II/IGF-I ratio ~3:1 is considered normal. Most cases that produce high numbers of IGF-II have a ratio of >10:1. However, in our case, it indicates ~20:1. The treatment of SFT should aim at both symptomatic controls of neuroglycopenic signs and symptoms and tumor treatment itself. Hypoglycemia will be reversible after the successful removal of the tumor by surgery. 14 This case illustrates a biological transformation of an SFT into a malignant fibrous tumor to secrete a large number of IGF-II after a long period of dormancy.

4 | CONCLUSION

Fibroma causing recurrent hypoglycemia has rarely been reported in the literature. The physicians should keep in mind this rare possibility while encountering any patients with hypoglycemia and should rule out this possibility in addition to others. Recurrent hypoglycemia if prolonged can cause permanent changes in the brain.

AUTHOR CONTRIBUTIONS

Muhammad Hanif: Conceptualization; project administration; supervision; writing – original draft; writing – review and editing. Vikash Jaiswal: Data curation; project administration; validation; visualization; writing – original draft; writing – review and editing. Sidra Naz: Writing – original draft; writing – review and editing. Nirmit Patel: Writing – original draft; writing – review and editing. Nishan Babu Pokhrel: Writing – review and editing. Mounika Reddy Vadiyala: Writing – review and editing.

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DATA AVAILABILITY STATEMENT

All relevant data is available online

ETHICAL APPROVAL

Need for ethical approval waived. Consent from the patient is deemed to be enough.

CONSENT

Written informed consent has been taken from the patient which would be available upon the Editor-in-Chief's request.

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