

Solitary Fibrous Tumor of the Pleura: A Case Report

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DOI: [10.36347/sasjm.2023.v09i10.014](https://doi.org/10.36347/sasjm.2023.v09i10.014)

| Received: 13.09.2023 | Accepted: 19.10.2023 | Published: 24.10.2023

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Abstract

Case Report

Solitary fibrous tumors of the pleura are rare malignancies, with fewer than 1000 reported cases. They are either discovered incidentally, or in association of non-specific symptoms such as hypoglycemia. The association of paraneoplastic hypoglycemia and solitary fibrous pleural tumors is rare. Hypoglycemia occurs in large tumors and normalizes after surgery, the treatment of choice for pleural solitary fibrous tumors (PSTF). The authors discuss the management of this rare tumor and the association of solitary fibrous tumor of the pleura (SFTP) with hypoglycemia.

Keywords: Fibrous tumors, hypoglycemia, surgery, imaging.

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INTRODUCTION

Solitary fibrous tumors of the pleura are rare pleural neoplasms, the majority of which are benign. They occur infrequently and are often found incidentally or in association with non-specific respiratory symptoms. When associated with hypoglycemia, they are defined as Doege-Potter syndrome.

OBSERVATION

A 49-year-old patient, smoker at 22 pack-years, admitted for recurrent brief loss of consciousness with hypoglycemia. Clinical exam was unremarkable.

Glycemic monitoring for five days was diminished despite intravenous treatment with 10% glucose. His serum insulin was low at 2.9 ng/ml.

Thoracic CT revealed a well-limited homogeneous left basithoracic mass (Figure 1 & 2). Bronchoscopy was normal.

Scannoguided biopsy of the mass revealed a typical spindle-cell tumour.

Surgical exploration revealed a large, well-limited, pedunculated tissue mass with a broad implantation base at the expense of the visceral pleura opposite the middle lobe. The tumour was completely resected, measuring 61×52×38 mm.

Postoperatively, there was a marked improvement in hypoglycemia and disappearance of recurrent episodes of loss of consciousness.

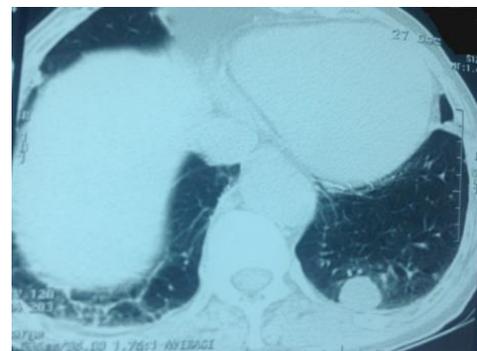


Figure 1: Chest CT (lung window and soft tissue window)



Figure 2: Chest CT (lung window and soft tissue window)

DISCUSSION

Solitary fibrous tumors of the pleura (SFTP) are rare malignancies, with fewer than 1000 cases reported in the literature [1]. They were first reported by Klemperer and Rabin in 1931 [2], accounting for less than 5% of all pleural tumors [3].

The association between hypoglycemia in Doege-Potter syndrome and a solitary fibrous pleural tumor remains rare. Briselli *et al.*, have shown that 4% of solitary fibrous tumors are associated with hypoglycemia and that this is more frequent in large tumors with a high mitosis rate [4]. In our case, the tumor was smaller than 10 cm.

In paraneoplastic syndrome, hypoglycemia may be caused by the production of an insulin-like growth factor [5].

This growth factor can stimulate membrane insulin receptors, stimulating peripheral glucose utilization and blocking glycogenolysis and gluconeogenesis, thus avoiding simultaneous endogenous and exogenous glucose supply and the hyperglycemia that could result [6]. It can also act on the pituitary gland, reducing the production of growth hormone, which in turn reduces insulin secretion by the pancreas [7]. The thoracic CT scan is the reference examination, providing an objective view of the location, size and description of the tumour and guiding the surgical procedure. The mass is generally of variable size and regular contour.

The diagnoses to be evoked on imaging are broad, ranging from thoracic, mediastinal, pulmonary and pleural malignancies to tumors of neurological origin.

Percutaneous echo- or scannoguided biopsy of the tumour can sometimes help evoke the diagnosis.

Total surgical removal is the treatment of choice for solitary fibrous tumors of the pleura. Neoadjuvant supplementary treatment is controversial, given the risk of malignant transformation and recurrence, and long-term follow-up is necessary in all patients [8].

CONCLUSION

Solitary fibrous tumors of the pleura are rare lesions, most often discovered by chance. Hypoglycemia

can be a revealing symptom. Their course is generally benign, and surgery is the mainstay of treatment.

Competing interests: The authors declare that they have no competing interests.

Authors' Contributions: All authors contributed to the manuscript, read and approved the final manuscript.

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