

An unusual cause of recurrent hypoglycemia in a patient with a solitary fibrous tumour of the lung is the doege-potter syndrome.

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Introduction

Only 2% of soft tissue tumours are Solitary Fibrous Tumours (SFT), which are rare tumours of mesenchymal origin. Klempner and Rabin first identified them as pleural tumours in 1931, but in the years that followed, other places were also mentioned. Doege and Potter first documented the occurrence of hypoglycemia caused by a non-islet cell tumour in 1930. Doege Potter Syndrome is the name for non-islet cell tumour hypoglycemia linked to SFTs (DPS). SFTs are considered to be indolent tumours, however the prognosis is uncertain.

A paraneoplastic illness called Doege-Potter Syndrome (DPS) is characterised by hypoglycemia and solitary fibrous tumours [1]. The tumours' production of insulin-like growth factor 2 causes hypoglycemia. Karl Walter Doege (1867-1932), a German-American doctor, and Roy Pilling Potter (1879-1968), an American radiologist, independently characterised the disease for the first time in 1930; the complete phrase Doege-Potter syndrome wasn't commonly used until a 2000 article employing the eponym.

Less than 100 cases of DPS have been described as of 1976, and its malignancy rate is between 12 and 15 percent. In reality, only 4% of solitary fibrous tumours of the lungs induced hypoglycaemia, according to a 1981 study, and these cases are usually connected with large tumours that undergo a lot of mitosis. Typically, symptoms will go away when the tumour is removed [2].

SFTs are uncommon soft tissue neoplasms of mesenchymal origin with fibroblastic differentiation. They can occur in almost any site, intrathoracic location being the most common. While the majority are pleural based, they can also arise from the lung parenchyma, mediastinum and the diaphragm [3]. The peak incidence is in the sixth to seventh decades of life, occurring equally in both genders. SFTs, when occurring intrathoracically, can cause dyspnea, cough, chest pain and rarely haemoptysis. They can also result in pleural effusions. There are no recognised standard treatment guidelines for SFTs due to the dearth of clinical trials. The best curative method for an SFT is surgical resection, which also stops hypoglycaemia from returning. Dextrose injections and glucocorticoids are used as a temporary symptomatic treatment for DPS since they are known to enhance gluconeogenesis and decrease the development of big-IGF-2 [4].

Case report

A 71-year-old woman with no relevant history visited a primary care center for generalized tonic-clonic seizure caused by severe hypoglycemia. According to her medical history, she had been experiencing weight loss, dyspnea, and cough with whitish expectoration for the past 10 months as well as seizures that were treated with dextrose. On admission, her blood pressure was 142/82 mmHg, temperature was 37.2°C, heart rate was 106 beats per min, and respiratory frequency was 32 breaths per minute. She was alert, with increased inspiratory effort. Auscultation findings included decreased breath sounds in both lung fields with right prevalence and asymmetrical thoracic expansion. She presented nail clubbing. Neurological and physical examination results were normal. Laboratory study results are reported in during hospitalization, it was necessary to administer 50% dextrose via central venous catheter, with partial improvement of glycaemia measurements. Thoracic X-ray and Computed Tomography (CT) revealed a large mass with right basal predominance that was encapsulated and non-homogeneous; it occupied 50% to 70% of the right hemi thorax. CT-guided percutaneous biopsy results revealed a malignant SFTP, hemangiopericytoma type. She underwent surgery for tumour resection, and a tumor approximately 30 cm in diameter with a pedicle that was dependent on the right lower lung lobe and with lax adhesions to the thoracic wall, diaphragm, and mediastinum was found [5].

Conclusion

SFTs can remain completely asymptomatic or come to clinical attention because of recurrent hypoglycemia, like our patient. Diagnosis involves imaging and histopathological examination. Intrathoracic SFTs are often seen as a well circumscribed, lobular, solitary nodule or mass in the lung periphery, usually in close approximation to the pleural surface. Under the microscope, SFTs consist of cells with oval to spindle shaped nuclei with minimal cytoplasm with intervening collagen bands in a pattern less distribution.

References

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Received: 01-Jul-2022, Manuscript No. AADY-22-71115; Editor assigned: 04-Jul-2022, PreQC No. AADY-22-71115(PQ); Reviewed: 18-Jul-2022, QC No AADY-22-71115; Revised: 21-Jul-2022, Manuscript No. AADY-22-71115(R); Published: 28-Jul-2022, DOI: 10.35841/aady-6.4.117

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Citation: Perinkulam S. An unusual cause of recurrent hypoglycemia in a patient with a solitary fibrous tumour of the lung is the doege-potter syndrome. *J Diabetol*. 2022;6(4):117