

## Scientific Letter

### Hypoglycemia as an Atypical Presentation of a Pleural Tumor<sup>☆</sup>



#### *Hipoglucemias como presentación atípica de un tumor pleural*

Dear Editor:

Solitary fibrous tumors of the pleura are rare neoplasms (2.8 cases per 100,000 inhabitants)<sup>1</sup> that comprise a heterogeneous group of primary pleural tumors derived from submesothelial cells with blastic and mesenchymal characteristics. A small percentage of patients develop hypoglycemia that is usually poorly controlled and persistent, due in most cases to the secretion of insulin-like growth factors by the fibrous cells of the tumor. This entity is known as Doege-Potter syndrome.<sup>2,3</sup>

We report the case of a 61-year-old man, former smoker of 60 pack-years with no other significant history, who presented in the emergency department with a 3-day history of sleepiness. On arrival in the emergency department, his blood glucose was 20 mg/dl, so an infusion of 5% dextrose solution was administered, after which the patient became asymptomatic. On examination, the only finding was decreased breath sounds in the left lung field. Clinical laboratory and radiological studies were performed. Biochemistry revealed a blood glucose level of 30 mg/dl, while other test results were within normal limits. Chest X-ray revealed opacities in 90% of the left hemithorax with midline shift. Given the suspicion of a chest tumor, the patient was referred for admission to the respiratory medicine department.

As part of the diagnostic procedure, we performed a computed tomography (CT) scan of the chest, which showed evidence of a left pleural mass measuring 21.5 × 12 × 23 cm (Fig. 1). Extended clinical laboratory tests were conducted, including liver function tests, electrolytes, thyroid function, and proteins, with results within normal limits. However, the patient showed strikingly poor glycemic control despite a diet high in slow-absorbing carbohydrates and intermittent glucose supplements; for this reason, with the agreement of the endocrinology department, we decided to start treatment with corticosteroids at a dose of 1 mg/kg/day. Glucose levels improved, although episodes of severe hypoglycemia persisted, preventing patient discharge. A CT-guided biopsy was performed in the radiology department, with a pathology study result consistent with pleural fibroma markers with positive immunohistochemical markers bcl-2 and CD34 and a proliferation index (Ki-67) of 3–7%. Paraneoplastic syndrome due to the fibrous tumor was suspected, so insulin-like growth factor II (IGF-II) determination was requested, with a result of 621 ng/mL (reference values 350–1000 ng/mL).

During admission, the patient developed incapacitating pain in the left hemithorax requiring progressively increasing doses of analgesia. Given these symptoms, the thoracic surgery department was contacted and preoperative studies were performed (PET-CT), identifying a pleural mass with a maximum SUV of 5.3 g/ml with no uptake at other levels, lung scintigraphy with differential count showing 82.3% perfusion corresponding to the right lung and the remaining 17.7% corresponding to the left lung. In view of persistent hypoglycemia and chest pain, the patient was referred for thoracic surgery and underwent complete resection of the pleural tumor by thoracotomy, preserving the pulmonary parenchyma in its entirety. After the operation, the patient's steroid treatment was progressively tapered, with normalization of blood glucose levels and disappearance of pain.

Doege-Potter syndrome, first described in 1930, is a rare entity.<sup>2–4</sup> It is characterized by constant hypoglycemia, suppressed serum insulin, C-peptide, growth hormone, and low IGF-I in serum, with altered levels of IGF-II. High IGF-II levels guide diagnosis, although in some cases values are normal. In these, the IGF-II/IGF-I ratio is usually greater than 3:1 and the high molecular weight IGF-II subtype is usually increased.<sup>4</sup>

This syndrome is associated with solitary fibrous tumors of the pleura, other intrathoracic tumors, and fibrous tumors in other sites.<sup>5</sup> Tumors are usually slow-growing with initially mild respiratory symptoms. Hypoglycemia may be the only symptom, so the importance of chest imaging studies in cases of persistent hypoglycemia must be stressed. These tumors are most common between the sixth and eighth decade of life, and in men in some series. They are also more prevalent in the right hemithorax.<sup>4–6</sup>

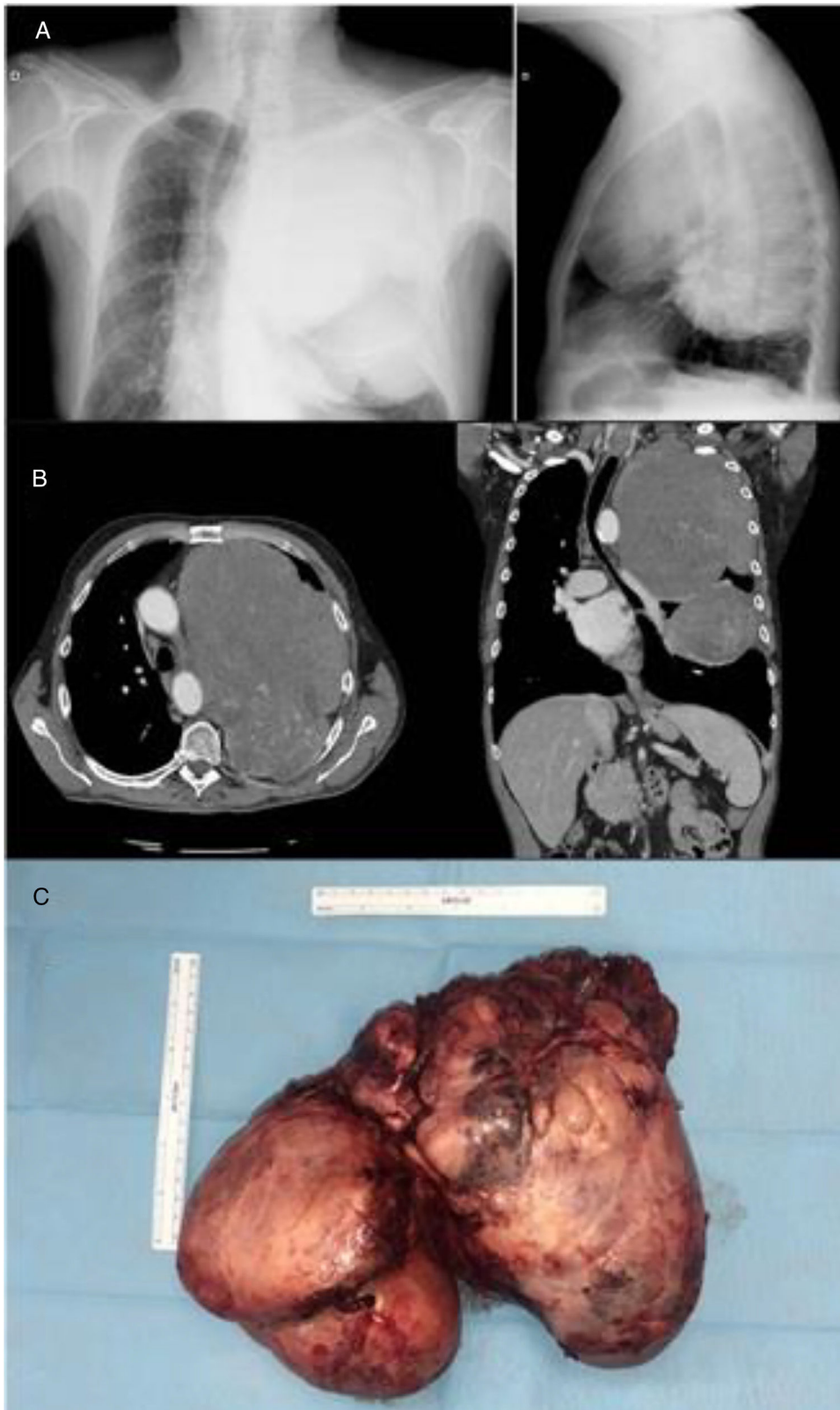
To date there has been no link to environmental or toxic factors. In most cases, these tumors behave benignly and rarely produce metastasis, but malignant behavior has been reported in up to 12%.<sup>7</sup>

Fibrous tumors appear in imaging studies as a mass that is typically homogeneous, clearly defined, and non-invasive. For histological diagnosis, core needle or surgical biopsy is recommended, since these techniques yield a greater amount of tissue.

From a pathological point of view, the tumor presents a spindle cell morphology with alternating areas of greater and lower cellular density; it expresses CD34, which is useful for differential diagnosis with other neoplasms. This marker has a sensitivity of 100% and a specificity of 70.9%.<sup>8</sup> More aggressive tumors sometimes express the proto-oncogene bcl-2.<sup>9</sup>

Hypoglycemia, as a paraneoplastic syndrome of this tumor, is most frequently associated with right-sided tumors measuring more than 20 cm, and with a high mitotic index; studies have shown that hypoglycemia is associated with a poorer prognosis and a risk of recurrence or metastasis.<sup>4</sup> Glucocorticoids have been shown to be useful in the control of blood glucose by suppressing high molecular weight IGF-II.<sup>10</sup> Our patient was receiving glucocorticoid treatment at the time of obtaining blood for the determination

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**Figure 1.** (A) Posteroanterior and lateral chest X-ray. (B) Chest CT with intravenous contrast, showing a large pleural mass with shift of mediastinal structures. (C) Tumor mass after surgical removal.

of this parameter, which may have altered the result obtained. The only curative treatment is surgical resection,<sup>11</sup> although recurrence rates of up to 15% are estimated, according to published series.<sup>4-12</sup>

## References

- Wilson RW, Gallateau-Salle F, Moran CA. Desmoid tumors of the pleura: a clinicopathologic mimic of localized fibrous tumor. *Mod Pathol*. 1999;12:9-14. PMID: 9950156.
- Doerge KW. Fibro-sarcoma of the mediastinum. *Ann Surg*. 1930;92:955-60. PMID: 17866430.
- Hohenforst-Schmidt W, Grapatsas K, Dahm M, Zarogoulidis P, Leivaditis V, Kotoulas C, et al. Solitary fibrous tumor: a center's experience and an overview of the symptomatology, the diagnostic and therapeutic procedures of this rare tumor. *Respir Med Case Rep*. 2017;21:99-104. <http://dx.doi.org/10.1016/j.rmcr.2017.04.007>.
- Han G, Zhang Z, Shen X, Wang K, Zhao Y, He J, et al. Doege-Potter syndrome. *Medicine (Baltimore)*. 2017;96:e7417. <http://dx.doi.org/10.1097/md.00000000000007417>.
- Takizawa I, Saito T, Kitamura Y, Arai K, Kawaguchi M, Takahashi K, et al. Primary solitary fibrous tumor (SFT) in the retroperitoneum. *Urol Oncol Semin Orig Investig*. 2008;26:254-9. <http://dx.doi.org/10.1016/j.urolonc.2007.03.024>.
- Avramenko A, Lahjibi-Paulet H, Gibault L, Roussel A, Grand B, le Pimpec-Barthes F. Doege-Potter syndrome: hypoglycaemic coma in a 90-year old due to a solitary fibrous tumour. *Age Ageing*. 2017;46:527-9. <http://dx.doi.org/10.1093/ageing/afw221>.
- Brozzetti S, D'Andrea N, Limiti MR, Pisanelli MC, de Angelis R, Cavallaro A. Clinical behavior of solitary fibrous tumors of the pleura. An immunohistochemical study. *Anticancer Res*. 2000;20:4701-6. PMID: 11205204.
- Gold JS, Antonescu CR, Hajdu C, Ferrone CR, Hussain M, Lewis JJ, et al. Clinicopathologic correlates of solitary fibrous tumors. *Cancer*. 2002;94:1057-68. <http://dx.doi.org/10.1002/cncr.10328>.
- Kim DW, Na KJ, Yun JS, Song SY. Doege-potter syndrome: a report of a histologically benign but clinically malignant case. *J Cardiothorac Surg*. 2017;12:64. <http://dx.doi.org/10.1186/s13019-017-0630-4>.
- Hosaka S, Katagiri H, Wasa J, Murata H, Takahashi M. Solitary fibrous tumor in the pelvis: Induced hypoglycemia associated with insulin-like growth factor II. *J Orthop Sci*. 2015;20:439-43. <http://dx.doi.org/10.1007/s00776-013-0462-6>.
- Kalebi AY, Hale MJ, Wong ML, Hoffman T, Murray J. Surgically cured hypoglycemia secondary to pleural solitary fibrous tumour: case report and update review on the Doege-Potter syndrome. *J Cardiothorac Surg*. 2009;4:45. <http://dx.doi.org/10.1186/1749-8090-4-45>.
- Forster C, Roumy A, Gonzalez M. Solitary fibrous tumor of the pleura with Doege-Potter syndrome: second recurrence in a 93-year-old female. *SAGE Open Med Case Rep*. 2019;7. <http://dx.doi.org/10.1177/2050313X18823468>, 2050313X18823468.

Juan Diego Alvarez Mavarez,<sup>a,\*</sup> Manuel Angel Villanueva Montes,<sup>a</sup> Manuel Ricardo Rodríguez Seoane,<sup>a</sup> Michelle Leung Shao<sup>b</sup>

<sup>a</sup> Servicio de Neumología, Hospital Universitario San Agustín, Avilés, Asturias, Spain

<sup>b</sup> Servicio de Cirugía torácica, Hospital Universitario Central de Asturias, Oviedo, Asturias, Spain

\* Corresponding author.

E-mail address: [juandiego06@hotmail.com](mailto:juandiego06@hotmail.com) (J.D. Alvarez Mavarez).

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## When should we measure self-efficacy as an aid to smoking cessation?\*



### ¿En qué momento deberíamos medir la autoeficacia para ayudar a dejar de fumar?

To the Editor,

In order to improve interventions to assist in smoking cessation (SC), the multiple facets that make up this addiction and the factors behind relapses must be better understood.<sup>1,2</sup> Self-efficacy (SE), understood as a person's belief in their ability to succeed in a particular situation, has been reliably associated with smoking abstinence and relapse, and is therefore an important target in anti-smoking interventions.<sup>3,4</sup> The objective of our study was to assess the association between SE, measured at the baseline visit, and abstinence at 12 months.

To this end, we performed a multicenter observational study of consecutive patients who attended SC clinics between October 2014 and October 2015. Patient demographic variables and smoking status were collected. SE was assessed from 2 questions included in the Richmond and Khimji-Watts questionnaires for measuring SC motivation<sup>5</sup> (Table 1). The statistical analysis was performed using the IBM SPSS 20.0 package for Windows. Logistic regression models were constructed to assess the association between SE and the outcome variable (SC success or failure), using the outcome variable as the dependent variable, the SE variable as the independent variable, and the remaining variables as control variables and adjustment. The level of statistical significance was set at  $p < 0.05$ .

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The study included 275 subjects, 130 men (47.3%), with an overall average age of 51.2 years (SD 10.7). In total, 53.5% (147 subjects) were successful in SC. The mean values of the different motivation tests were: visual analogue scale, 8 (1.9); Richmond questionnaire, 7.9 (1.5); the Henry Mondor Hospital test, 13 (2.7); and the Khimji-Watts questionnaire, 11.4 (2.5). The average overall degree of dependency, as measured by the Fagerström test (FTCD) was 5.9 (2.2), with no differences in the degree of dependency between those who quit smoking (mean 5.9, SD 2.2), and those who failed to quit (mean 5.9, SD 2.4). We found no significant differences in the analyzed variables between those who were successful in SC compared to those who were not, whether quantitative (age, age of onset, daily consumption of cigarettes, number of years smoked, accumulated consumption in pack-years, number of previous SC attempts, motivation, and FTCD questionnaires) or qualitative (sex, marital status, level of education, and employment status), except in educational level, where subjects with a secondary education had a 17.1% (95% CI: 4.8%-29.4%;  $p = 0.004$ ) greater rate of success, while those with university studies had a 12.4% (95% CI: 0.7%-24.0%;  $p = 0.026$ ) greater rate of failure. We did not find a statistically significant association between SE and the outcome variable (Table 1) in any of the possible logistic regression models constructed when the SE variable was examined both qualitatively and quantitatively, and or when controlling for sex, age, daily consumption of cigarettes, number of years smoked, number of previous attempts to quit, FTCD, and educational level. Nor did we find any differences in the multivariate analysis between men and women in the association between SE and the outcome variable. Cigarette dependence as measured by the FTCD score was not associated with the probability of SC. We conclude that neither SE nor the degree of dependence were predictive of the likelihood of SC success or failure, and only the educational level of participants showed a statistically significant association with the SC outcome.