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Case Report / Olgu Sunumu

Giant solitary fibrous tumor of pleura revealed by epileptic seizures: A case report

Epileptik nöbetlerle ortaya çıkan plevranın dev bir soliter fibröz tümörü: Bir olgu sunumu

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ABSTRACT

This study presents the case of a 56-year-old female patient suffering from repeated epileptic seizures. No abnormality was observed following cranial magnetic resonance imaging and electroencephalogram, and treatment with antiepileptic drugs had no obvious effects. Subsequently, an abnormal blood glucose level was observed during epileptic seizures. Severe hypoglycemia was highly suspected to be the cause of seizures. Absent respiration on the right middle and lower chest was observed through further physical examination. A thoracic computed tomography scan revealed a giant heterogamous mass in the patient's right thoracic cavity. This massive tumor was completely resected, and blood glucose level returned to normal shortly after the excision of the tumor. Immunohistochemical staining of the resected mass confirmed it to be a solitary fibrous tumor of the pleura. The patient remained in a favorable condition and did not suffer the recurrence of epileptic seizures over a 12-month follow-up period. To our knowledge, this is the first report on a giant solitary fibrous tumor of the pleura complicated with epileptic seizures caused by severe tumor-induced hypoglycemia.

Keywords: Epileptic seizures; hypoglycemia; solitary fibrous tumor.

ÖZ

Bu çalışmada tekrar eden epileptik nöbetler geçiren 56 yaşında bir kadın hasta olgusu sunuldu. Kraniyal manyetik rezonans görüntüleme ve elektroensefalogramı takiben anormallik gözlemlenmedi ve antiepileptik ilaçlarla tedavinin görünür etkileri olmadı. Daha sonra, epileptik nöbetler sırasında anormal bir kan şekeri düzeyi gözlemlendi. Nöbetlerin nedeninin büyük olasılıkla şiddetli hipoglisemi olduğundan şüphelenildi. İleri fiziksel muayenede sağ orta ve alt göğüste solunum olmadığı gözlemlendi. Torasik bilgisayarlı tomografi taramasında hastanın sağ torasik kavitesinde dev bir heterogam kitle görüldü. Bu büyük tümör tamamen kesip çıkartıldı ve kan şekeri düzeyi tümörün kesilip çıkartılmasından kısa süre sonra normale döndü. Kesilip çıkartılan kitlenin immünohistokimyasal boyamasında kitlenin plevranın bir soliter fibröz tümörü olduğu doğrulandı. Hastanın durumu olumlu seyretti ve hasta 12 aylık takip dönemi boyunca epileptik nöbet tekrarı yaşamadı. Bildiğimiz kadarı ile bu rapor, tümöre bağlı şiddetli hipogliseminin neden olduğu epileptik nöbetler ile komplike, plevranın dev bir soliter fibröz tümörü hakkında ilk rapordur.

Anahtar sözcükler: Epileptik nöbetler; hipoglisemi; soliter fibröz tümör.

Solitary fibrous tumor of the pleura (SFTP) is a rare intrathoracic tumor that arises from mesenchymal tissue directly subjacent to the mesothelial-lined pleura, and accounts for less than 5% of primary pleural tumors.[1] Solitary fibrous tumor of the pleura can manifest with intrathoracic or systemic symptoms.[2] However, the

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majority of patients with SFTP are asymptomatic, and in many cases, its preoperative diagnosis is relatively difficult. The presence of secondary hypoglycemia with an intrathoracic fibrous tumor was first reported in 1930 by Doege.[3] Hypoglycemia was observed to be associated with large SFTP (>10 cm), but had no obvious relevance with histological features of malignancy.[4] In this article, we present a giant SFTP complicated with epileptic seizures caused by severe tumor-induced hypoglycemia.

CASE REPORT

A 56-year-old female patient was admitted to our hospital due to recurrent episodes of epileptic seizures over the preceding two months. At the onset of attack, she suffered from epileptic seizures in the morning and spontaneously remitted within 30 minutes to one hour. At the initial hospital, she was admitted to the Department of Neurology and diagnosed with seizures because no other systemic symptoms were observed. Unfortunately, antiepileptic drug therapy was invalid. Two months later, epileptic seizures occurred once a day for two weeks, specifically in the morning.

The patient was transferred to our hospital for further diagnosis and treatment. On admission, peripheral blood analysis and biochemistry data were both within normal limits, and no abnormality was observed in brain magnetic resonance imaging (MRI) or electroencephalogram (EEG). An abnormal blood glucose level of 0.89 mmol/L (normal range: 3.9-6.1 mmol/L) was detected during epileptic seizures two days later, and the epileptic seizures were mitigated after the administration of 50% glucose intravenous (100-200 mL). As such, severe hypoglycemia was suspected to be the cause of epileptic seizures. Further detection revealed that the following related indices were extremely lower than normal values: insulin, <0.2 μIU/mL (normal range: 8-11 μIU/mL); C-peptide, 0.025 nmol/L (normal range: 0.48-0.78 nmol/L); and insulin-like growth factor-I (IGF-I), 6.4 μg/mL (normal range: 7.0-46.2 μg/mL). However, no abnormality was observed during the ultrasound scanning of the pancreas. Further physical examination revealed absent breathing sounds in her right hemithorax. Chest X-ray revealed a sizeable mass in the right pleural space (Figure 1a). A thoracic computed tomography (CT) scan revealed a giant, heterogeneous, solid-appearing mass lesion in the right thoracic cavity (Figure 1b). No metastases were found. A CT-guided biopsy confirmed a benign SFTP. A written informed consent was obtained from the patient.

The patient underwent standard open thoracotomy via the right fifth intercostal space. A well-capsulated tumor originating from the visceral pleura of the right middle lobe of the lung was discovered (Figure 2a). Two vascular pedicles were found arising from the visceral pleura, while another pedicle without blood vessels was traced to the diaphragmatic surface. The tumor was completely excised without pulmonary resection. The gross appearance of the cut surface of the tumor displayed a yellowish white and firm mass without hemorrhage or necrosis (Figure 2b). The mass was measured as 39×22×9 cm and weighed 2,900 g.

A final diagnosis of benign SFTP was established by hematoxylin-eosin staining (Figure 3a) and immunohistochemistry, which revealed positivity for cluster of differentiation 34 (CD34) (Figure 3b), CD99 and ki67, and negativity for smooth muscle actin,

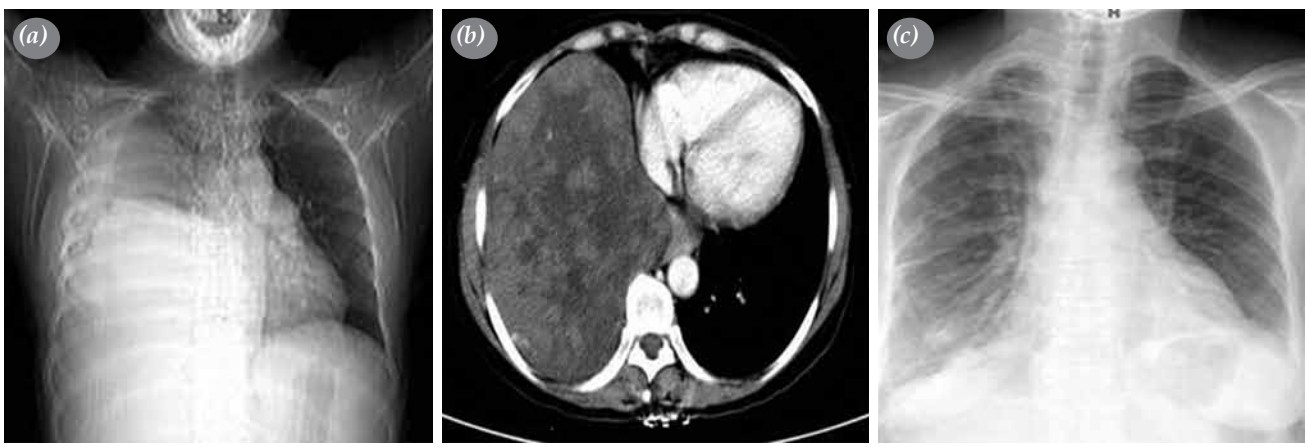

Figure 1. (a) Chest roentgenogram shows lesion in right pleural space. **(b)** Thoracic computed tomography scan demonstrates a giant, heterogeneous, solid-appearing mass lesion in right thoracic cavity. **(c)** Right lung that expanded postoperatively.

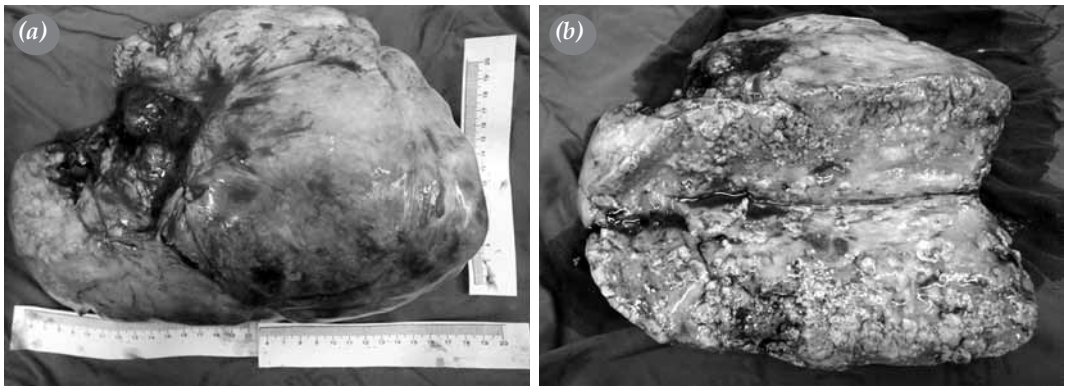

Figure 2. (a) Well-capsulated tumor originated from visceral pleura of right middle lobe of lung (39×22×9 cm). **(b)** Cut surface of tumor showed a yellowish white, firm mass without hemorrhage or necrosis.

epithelial membrane antigen, cytokeratin (CK), CK19 and S-100. The lung expanded on the right side postoperatively (Figure 1c), while serum levels of blood glucose, insulin, C-peptide and IGF-I returned to normal.

The patient experienced an uneventful recovery and was discharged on postoperative ninth day. She did not receive postoperative chemotherapy or radiotherapy. A 12-month follow-up in the outpatient clinic yielded no recurrence of tumor or epileptic seizures attack. The plasma glucose level remained normal.

DISCUSSION

Epileptic seizures occurring in patients with tumors may be traced to a variety of causes, such as primary brain tumors, brain metastasis, adverse reaction to cytotoxic chemotherapy, or other conditions that disturb the normal pattern of neuron activity. Hypoglycemia is one of the culprits behind epileptic seizures as it interferes with the cerebral metabolism by reducing the brain's energy supply. Hypoglycemia can lead to metabolic brain insult when the glucose level falls below 1 mmol/L, which may lead to seizures.[5]

In this case, no abnormality was observed in the brain MRI and EEG. Meanwhile, severe hypoglycemia was observed during the epileptic seizures. After tumor removal, the glucose serum level returned to normal value and no epileptic seizures reoccurred. Thus, it was further speculated that epileptic seizures may be caused by severe hypoglycemia.

The presence of secondary hypoglycemia with intrathoracic fibrous tumor was first reported in 1930 by Doege.[3] Solitary fibrous tumor of the pleura associated with severe hypoglycemia is rare.[4] The cause of hypoglycemia is believed to be related to IGFs produced by the tumor. Insulin-like growth factor-II is among the insulin-like peptides commonly believed to be responsible for non-islet cell tumor hypoglycemia. Insulin and IGF-I will be suppressed by negative feedback when IGF-II is overproduced.[6] Therefore, abnormal levels of insulin and IGF-I may indicate an IGF-II secreting tumor.

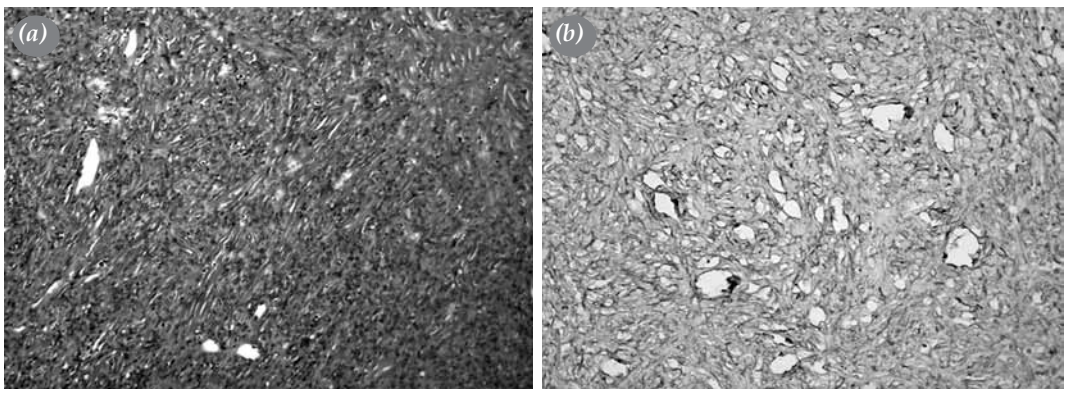

Figure 3. (a) Hematoxylin-eosin staining (H-E × 200) showed that tumor composed of fibroblast-like cells, accompanying deposition of abundant collagen and mucinous degeneration. **(b)** Immunohistochemical staining was positive for cluster of differentiation 34 (immunohistochemistry, × 200)

In this case study, the levels of insulin and IGF-I were extremely lower than normal values preoperatively, and became normal postoperatively. This indicates that the current case involves an IGF-II secreting tumor, subsequently explaining that the epileptic seizures were caused by hypoglycemia.

In conclusion, tumor-induced hypoglycemia must be considered among patients suffering from epileptic seizures without brain tumors or traumas. The cause of tumor-induced hypoglycemia is related to insulin-like growth factors produced by these tumors. Complete surgical resection is the preferred therapy. Close follow-up with chest roentgenogram and computed tomography scan for postoperative years are highly recommended.

Declaration of conflicting interests

The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

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