Cavernous hemangiomas are characterized by sharply defined yet unencapsulated masses, which are composed of large dilated vascular spaces lined by a single layer of endothelial cells and filled with blood. They are benign vascular tumors that are frequently located in the skin, subcutaneous tissues, and liver. Cavernous hemangiomas can occur in various internal organs, but they rarely occur in the heart and lungs. We report a rare case of a co-occurring cardiac cavernous hemangioma and multiple pulmonary cavernous hemangiomas (PCHs) and review the literature.

A 36-year-old man with a 1-month history of pericardial effusion was admitted to our hospital. Imaging with ultrasonography and chest roentgenography (Fig 1A) showed bilateral pleural effusion and pericardial effusion. Computed tomography (CT) of the chest and heart (Figs 1B, 1C) showed bilateral pulmonary nodules, left pleural effusion, and pericardial effusion. Positron emission tomography showed a pericardial neoplasm. After extensive workup, a thoracotomy, pericardial tumor closure of a benign oesophagobronchial fistula pre-
includes capillary, cavernous, and arteriovenous hemangiomas, but the cause is not yet very clear. Cardiac hemangiomas are exceedingly rare, representing only 1% to 2% of all detected benign heart neoplasms [1]. They can occur at any age and in any layer of the heart, including the epicardium, myocardium, and endocardium [2], although the epicardium is the most common location [3]. Cardiac hemangiomas can also develop in all the cardiac chambers, but they mainly occur on the right side of the heart and the left atrium [2]. Pulmonary cavernous hemangiomas (PCHs) are also exceptionally rare. Generally, PCHs are solitary lesions and can present in all lobes, involving both lung parenchyma and subpleural tissue [4]. The diagnosis in our patient was cardiac cavernous hemangioma and multiple PCHs. To our knowledge, no such case has ever been reported previously.

Most hemangiomas are found incidentally. The symptoms of cardiac hemangiomas depend on the location and size of the tumors. Patients may have no symptoms, or they may present with dyspnea, heart palpitations, atypical chest pain, or arrhythmia or pericardial effusion [2]. Similarly, some patients with PCHs have no obvious symptoms.
symptoms, whereas others display a series of clinical processes such as hemoptysis and respiratory distress until heart failure occurs [4].

Echocardiography is usually the initial imaging examination for the diagnosis of cardiac tumors, although computed tomography and magnetic resonance imaging actually have a higher accuracy rate in exposing this condition. Usually, these modalities have been used in preoperative diagnosis [5]. However, the nature of the tumors cannot be cleared by preoperative diagnosis in many cases. Preoperative diagnosis of PCHs is also difficult. All cases that have been reported were diagnosed by intraoperative or postoperative examination or at autopsy.

The histologic appearances of cardiac cavernous hemangiomas and PCHs are no different from the appearances of other parts. The diagnosis in our patient was not difficult based on the histologic appearance and immunohistochemical reactivity. However, other entities, such as mesothelioma, angiosarcoma, and myxoma, must sometimes be considered in the differential diagnosis. Microscopically, most epithelioid mesotheliomas show bland cells and little mitosis. Epithelioid tumor cell nests can be seen within a bland fibrous stroma [6]. Calretinin, WT-1, and D2-40 should be positive by immunohistochemical examination. Mitotic activity, cellular pleomorphism, necrosis, and cellularity can distinguish hemangiomas from angiosarcomas. Vascular endothelial markers such as FVIII, CD31, and CD34 all have a strong reaction in angiosarcomas [7]. Myxomas have a typical appearance, with spindle cells widely spaced by abundant myxoid matrix, which can differentiate them from cavernous hemangiomas.

Hemangiomas may have a good prognosis and a low recurrence rate. So far, no malignant change has been reported to our knowledge. Total resection should be the standard treatment for symptomatic cavernous hemangiomas whenever possible [8]. For most cardiac cavernous hemangiomas, complete excision is possible. However, some lesions that have extensive infiltration are appropriate only for incomplete resection or simple biopsy, and the prognosis is usually poor [2]. Similarly, solitary PCHs should be treated with surgical resection, usually wedge resection or enucleation [4]. However, no intervention was undertaken in the present reported case of multiple PCHs. In this case, cavernous hemangiomas were found in both the heart and the lungs, which was exceedingly rare and complicated. Embolizing the remaining cavernous hemangiomas was considered; however, the patient’s condition did not allow it. Close radiographic and clinical follow-up was needed. Unfortunately, as a result of pericardial effusion and pleural effusion, the patient experienced a series of processes such as heart failure, respiratory failure, causally insufficient blood supply, and ischemia and anoxia of the vital organs, and finally died of multiple organ dysfunction syndrome 2 months later.

References