bone destruction results. Although dental foci can be a source of bacterial endocarditis,4 hematogenous spread of infection apparently is rare in periodontal disease.5 A review of the literature disclosed three previous reported cases of septic pulmonary embolism associated with periodontal disease.5–7 All patients reported, including ours, were middle-aged or elderly men. Blood culture findings were negative in three of four cases in which cultures were obtained. No patient was in serious condition, in contrast to the typical severe illness seen when septic pulmonary embolism originates from bacterial endocarditis or septic thrombophlebitis. None of the patients with periodontal sources were immunocompromised except for case 2, who received oral prednisolone for treatment of bronchial asthma. Three of five patients had a toothache at the time of initial presentation. All were treated successfully with antimicrobial agents in addition to periodontal surgery.

Dental infection should be considered as a possible source of septic pulmonary emboli. In particular, gingival infection should be treated effectively.

REFERENCES

Choriocarcinoma in the Pulmonary Artery Treated With Emergency Pulmonary Embolectomy*

Shun-ichi Watanabe, MD; Shinji Shimokawa, MD; Koh-ichi Sakasegawa, MD; Hiroshi Masuda, MD; Ryuzo Sakata, MD; and Miehito Higashi, MD

A 42-year-old woman with choriocarcinoma required emergency pulmonary embolectomy under cardiopulmonary bypass. After diagnosis of choriocarcinoma was confirmed by examination of tumor emboli specimens, the patient was treated and had complete remission by chemotherapy over a 6-month period. Although rare, choriocarcinoma should be considered in the differential diagnosis of fertile women presenting with pulmonary embolism.

Key words: cardiopulmonary bypass; choriocarcinoma; emergency; pulmonary artery; pulmonary embolectomy; pulmonary embolism

Abbreviations: hCG = human chorionic gonadotropin; MPA = main pulmonary artery; RPA = right pulmonary artery

Due to their rarity, tumors of the pulmonary arteries are often incorrectly diagnosed as more common diseases, such as pulmonary thromboembolism, and are thus seldom diagnosed during a patient’s lifetime. We report a patient with a choriocarcinoma mimicking pulmonary embolism, who required emergency pulmonary embolectomy and had a complete remission with adequate chemotherapy thereafter.

CASE REPORT

A 42-year-old woman presented with exertional dyspnea in August 1999. The patient had three children, the youngest being 4 years old, and she had two previous spontaneous abortions. The chest radiographs showed a bilateral diffuse infiltrative shadow. She was treated for interstitial pneumonia; however, her symptoms became worse. On December 16, pulmonary thromboembolism was suspected and she was urgently admitted to our hospital. On hospital admission, she was orthopneic and could not be placed in a left decubitus position. Arterial blood gas levels under oxygen mask inhalation (5 L/min) were as follows: pH, 7.49; Pco2, 44 mm Hg; and Paco, 26 mm Hg. The chest enhanced CT showed a filling defect from the main pulmonary artery (MPA) to the right pulmonary artery (RPA) [Fig 1]. On the same

*From the Second Department of Surgery (Dr. Higashi) and the Second Department of Pathology (Drs. Watanabe, Shimokawa, Sakasegawa, Masuda, and Sakata), Kagoshima University Faculty of Medicine, Kagoshima, Japan.

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Correspondence to: Shun-ichi Watanabe, MD, The Second Department of Surgery, Kagoshima University Faculty of Medicine, 8–35-1 Sakuragaoka, Kagoshima, 890-8520 Japan

FIGURE 1. The chest enhanced CT showed a filling defect from the pulmonary trunk to the RPA.
day, the patient underwent emergency pulmonary embolectomy under cardiopulmonary bypass. Opening from the MPA to the bilateral pulmonary arteries, a whitish, soft tumor embolus with a papillary surface was found occluding the MPA and extending into the RPA. The tumor embolus was removed as much as possible. Pulmonary arterial pressure dropped from 80 to 50 mm Hg immediately after the operation. However, the patient required ventilator support for subsequent respiratory failure owing to residual tumor emboli.

Microscopically, the pulmonary tumor emboli specimens showed necrotic tissue in the central area and proliferation of atypical cells in the peripheral areas (Fig 2, top). The cells had hyperchromatic nuclei and eosinophilic cytoplasm with indistinct cell borders forming “syncytiotrophoblastic cells” (Fig 2, bottom). After a diagnosis of choriocarcinoma was made, serum human choriionic gonadotropin (hCG) level was measured and a gynecologic examination was performed. As serum hCG values were > 70,000 mIU/mL, chemotherapy was initiated with actinomycin D, 0.1 mg/m² for 5 days. Then, etoposide, 80 mg/m² on days 1 to 5; methotrexate, 100 mg/m² via IV bolus on day 1 and 200 mg/m² via IV infusion over 12 h on day 1; actinomycin D, 0.1 mg/m² for 5 days. Then, etoposide, 80 mg/m² on days 1 to

![Image](http://journal.publications.chestnet.org/pdfaccess.ashx?url=/data/journals/chest/21973/)

**Figure 2.** *Top:* Microscopically, the pulmonary tumor emboli specimens showed necrotic tissue in the central area and proliferation of atypical cells in the peripheral areas (hematoxylin-eosin, original × 40). *Bottom:* The cells had hyperchromatic nuclei and eosinophilic cytoplasm with indistinct cell borders forming “syncytiotrophoblastic cells”. These cells were localized on the peripheral side (hematoxylin-eosin, original × 200).

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**COMMENT**

Bagshawe and Brooks were the first to suggest that pulmonary embolism and pulmonary hypertension could be due to a choriocarcinoma of the pulmonary arteries. Choriocarcinoma is the most aggressive form of gestational trophoblastic disease. Most cases occur following a complete hydatidiform mole. The natural history of untreated choriocarcinoma is characterized by the development of early hematogenous metastases. Some of the vagaries of this tumor have attracted attention, and failure to find a primary growth in the uterus, even on careful microscopic examination, is not infrequent. To the best of our knowledge, only a few cases of primary choriocarcinoma in a patient’s lifetime have been reported. However, this is the first patient who underwent emergency pulmonary embolectomy and was treated with subsequent chemotherapy after a confirming pathologic diagnosis. The patient had been monitored for interstitial pneumonia for > 5 months, and pulmonary thromboembolism was finally diagnosed. Diagnosis of choriocarcinoma in the pulmonary artery is very difficult in the early stage. In the present case, a pulmonary tumor thrombus was examined histologically and diagnosed as choriocarcinoma for the first time.

Many medical textbooks omit choriocarcinoma as a cause of pulmonary emboli and pulmonary artery hypertension. However, Seckl and coworkers reported that although rare, choriocarcinoma should be considered in the differential diagnosis of fertile women presenting with pulmonary embolism or pulmonary artery hypertension. Furthermore, Savage et al emphasized the importance of considering choriocarcinoma and measuring serum hCG in the investigation of fertile women presenting with pulmonary emboli or pulmonary artery hypertension. In our case, choriocarcinoma was not diagnosed in an early stage. However, chemotherapy was initiated as soon as the diagnosis was made and the patient responded positively, with serum hCG falling to within normal levels. Unfortunately, because the hospitalization was long, the cost of treatment became expensive. The problem of the limited health-care resources is a further consideration. Our results indicate that although rare, choriocarcinoma should be considered in the differential diagnosis of fertile women presenting with pulmonary embolism, because complete remission can be achieved with appropriate chemotherapy.
Pulmonary arterial hypertension (PAH) is commonly associated with the CREST (calcinosis, Raynaud phenomenon, esophageal dysmotility, sclerodactyly, telangiectasia) syndrome. Inhaled nitric oxide (iNO) is often used to assess acute vasoresponsiveness in patients with PAH, and reports of adverse reactions have been infrequent. We describe two of nine patients with PAH and CREST syndrome who had pulmonary edema develop during acute iNO testing. This complication was not encountered in the 46 patients with other forms of PAH tested with iNO. We suggest that iNO should be used with caution, if at all, to test acute vasoreactivity in patients with CREST syndrome.

**Key words:** CREST; epoprostenol; nitric oxide; pulmonary edema; pulmonary hypertension; scleroderma; vasodilators

**Abbreviations:** CREST = calcinosis, Raynaud phenomenon, esophageal dysmotility, sclerodactyly, telangiectasia; iNO = inhaled nitric oxide; PAH = pulmonary arterial hypertension

Pulmonary arterial hypertension (PAH) occurs in up to 50% of patients with the limited form of progressive systemic sclerosis, known as the CREST (calcinosis, Raynaud phenomenon, esophageal dysmotility, sclerodactyly, telangiectasia) syndrome, and adversely affects prognosis. Uncontrolled studies and a randomized trial have reported favorable responses to long-term epoprostenol (prostacyclin) infusion in these patients, which led to the approval of epoprostenol by the US Food and Drug Administration in 1999 for use in PAH associated with connective tissue disease. Inhaled nitric oxide (iNO), by virtue of its rapid inactivation by hemoglobin and paucity of systemic side effects, has been considered an ideal agent for testing acute vasoreactivity in patients with PAH of various etiologies. It has been reported to cause pulmonary edema in patients with congestive heart failure, but not in patients with PAH. We report two of a total of nine patients with the CREST syndrome who developed pulmonary edema during acute iNO testing. This complication was not encountered in the 46 patients with other forms of PAH who underwent acute vasodilator trials at our hospital during the same time period.

**RESULTS**

Between 1996 and 2000, 56 patients with PAH underwent acute vasodilator testing at Rhode Island Hospital, a teaching affiliate of Brown Medical School. The study protocol was approved by the Committee for the Protection of Human Subjects at Rhode Island Hospital, and all patients gave written consent. Patients were catheterized if they had progressive symptoms of dyspnea on exertion and an estimated peak pulmonary artery pressure > 40 mm Hg by echocardiography. During right-heart catheterization, iNO was delivered sequentially at concentrations of 5, 10, 20, and 40 ppm together with supplemental oxygen administered by tight-fitting face mask. Nitric oxide levels were measured continuously and were within normal limits. iNO was followed by an epoprostenol infusion starting at 1 ng/kg/min and increased by 1 ng/kg every 15 min until systemic effects such as flushing, nausea, jaw pain, headache, or hypotension occurred. Hemodynamic measurements were made at baseline and after each dose of vasodilator. A favorable acute response was considered to be a ≥ 20% decrease in pulmonary vascular resistance. Ten patients had PAH associated with the CREST syndrome. Of these, seven of eight patients administered epoprostenol and six of nine patients administered iNO had favorable responses. All six responders to iNO also had a favorable response to epoprostenol, whereas one patient responded to epoprostenol but not to iNO. Of the nine patients tested with iNO, acute pulmonary edema developed in two patients. None of the 46 patients with other forms of PAH (primary pulmonary hypertension, PAH associated with cirrhosis, HIV, systemic lupus erythematosus, secondary to thromboembolic disease, sarcoidosis, obstructive sleep apnea, or chronic obstructive lung disease) had pulmonary edema develop during acute vasodilator testing.

**Case 1**

A 70-year-old woman with a 20-year history of CREST syndrome presented with progressive dyspnea on exertion. She had systemic hypertension, glaucoma, a hiatal hernia, and a history of sarcoidosis diagnosed 24 years previously by mediastinoscopy, but with no evidence of active disease. Medications included warfarin, spiranolate, losartan, nifedipine, and supplemental nasal oxygen at 5 L/min. Physical examination showed mild facial and manual telangiectasias, clear lungs, increased P2, and trace ankle edema.