CASE REPORT

A Case of Thoracic Hemorrhage due to Ectopic Parathyroid Hyperplasia With Chronic Renal Failure

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• **We report a case of secondary hyperparathyroidism in a 53-year-old man who had thoracic hemorrhage originating from an ectopic thymic parathyroid gland tumor. He was on long-term hemodialysis treatment and had persistent secondary hyperparathyroidism caused by 3 swollen parathyroid glands around the thyroid gland and a mediastinal ectopic parathyroid gland tumor. Parathyroidectomy and removal of the mediastinal tumor were planned. Preoperative chest x-ray and a computed tomographic scan obtained just before surgery showed left pleural effusion. Furthermore, diagnostic thoracentesis yielded hemorrhagic exudative fluid. Thoracoscopy confirmed the presence of a mediastinal tumor lesion that was bleeding into the thoracic cavity. The tumor was resected using thoracoscopic surgery, and ectopic thymic parathyroid gland hyperplasia associated with hemorrhage was pathologically diagnosed. A postoperative diagnostic chest computed tomographic scan and technetium 99m 2-methoxyisobutyl isonitrile scintigraphy showed successful removal of the ectopic parathyroid gland tumor. Subsequently, the secondary hyperparathyroidism could be clinically controlled by medical treatment, and total parathyroidectomy has been postponed indefinitely. We consider that hemorrhage from the ectopic parathyroid gland tumor resulted in thoracic bleeding. To our knowledge, this is the first case report of secondary hyperparathyroidism with thoracic hemorrhage originating from an ectopic mediastinal parathyroid gland. *Am J Kidney Dis* 45:E109-E114.**

INDEX WORDS: Secondary hyperparathyroidism; ectopic parathyroid; thoracic hemorrhage; mediastinal tumor; pleural effusion.

PARATHYROID GLANDS have an important role in calcium metabolism. There are 4 parathyroid glands in the majority of humans.[1] A pair of superior parathyroid glands develop as a proliferation of entodermal cells in the fourth pharyngeal pouch on each side and take up their final position on the posterior aspect of the lateral lobe of the thyroid gland on each side, at about the level of the isthmus.[2] Conversely, a pair of inferior parathyroid glands, which share a common embryonic origin with the thymus, develop from the third pharyngeal pouch. Although the inferior parathyroid glands primordia initially comigrate with thymic primordia caudally from their site of origin, they ultimately continue to migrate toward the lateral lobe of the thyroid gland near its lower pole and become completely separate from the thymus.[2] It therefore is not surprising that ectopic parathyroid hyperplasia occasionally occurs in the mediastinum, embedded within the thymus.[3,4] We present a case of thoracic hemorrhage caused by upper anterior mediastinal ectopic parathyroid hyperplasia within the thymus in a patient with chronic renal failure.

CASE REPORT

A 53-year-old man had been receiving thrice-weekly hemodialysis since 1994. In 2003, the patient was found to have a high serum intact parathyroid hormone (iPTH) level. A diagnosis of secondary hyperparathyroidism was made, and intravenous maxacalcitol pulse therapy was started. However, secondary hyperparathyroidism was refractory to medical treatment, and there was no remarkable decrease in serum iPTH levels. Ultrasonography and a computed tomographic (CT) scan of the neck, performed in July 2004, showed 3 swollen parathyroid glands in the posterior region of the thyroid gland. Because abnormal uptake of technetium Tc 99m (^{99m}Tc)-2-methoxyisobutyl isonitrile (MIBI) also was detected by using scintigraphy in the upper mediastinum, we performed CT focusing on the area. An isodense well-delineated tumor lesion of 2 cm in diameter was detected in the upper anterior mediastinum. These findings suggested an ectopic mediastinal parathyroid gland within the thymus.

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Table 1. Laboratory Findings in October 2004

Peripheral blood		Total protein	8.0 g/dL
White blood cells	7,300/μL	Albumin	3.9 g/dL
Red blood cells	334 $\times$ 10^4/μL	Alkaline phosphatase	363 IU/L
Hemoglobin	10.7 g/dL	Lactate dehydrogenase	139 IU/L
Hematocrit	33.1%	Blood gas analysis	
Platelets	22.9 $\times$ 10^4/μiL	pH	7.342
Blood chemistry		Pco_2	37.5 mm Hg
Blood urea nitrogen	46 mg/dL	Po_2	91.2 mm Hg
Creatinine	11.3 mg/dL	Bicarbonate	20.1 mEq//L
Sodium	137 mEq/L	Anion gap	24.0 mEq/L
Potassium	4.3 mEq/L	Hormonal data	
Chloride	101 mEq/L	iPTH	842 pg/mL
Calcium	10.2 mg/dL	Serological test	
Phosphate	5.0 mg/dL	C-Reactive protein	4.81 mg/mL

NOTE. To convert hemoglobin, protein, and albumin in g/dL to g/L, multiply by 10; urea nitrogen in mg/dL to mmol/L, multiply by 0.357; serum creatinine in mg/dL to μmol/L, multiply by 88.4; sodium, potassium, chloride, bicarbonate, and anion gap in mEq/L to mmol/L, multiply by 1; calcium in mg/dL to mmol/L, multiply by 0.2495; iPTH in pg/mL to ng/L, multiply by 1.

Parathyroidectomy, including removal of the ectopic parathyroid gland, was planned for October 2004. In late September 2004, while the patient was waiting for surgery, he experienced mild cough with sputum, and these symptoms persisted until he was admitted to our unit.

On admission (October 21, 2004), the patient was afebrile. Blood pressure was 128/76 mm Hg, and physical examination showed no notable findings, except for anemic palpebral conjunctiva and moist rales in the left chest. Among laboratory findings on admission, C-reactive protein was positive (Table 1). A chest x-ray and ultrasonogram on admission showed an accumulation of fluid in the left thorax (Fig 1). A diagnostic thoracentesis was performed and yielded hemorrhagic exudative fluid. An urgent CT scan showed not only pleural effusion of the left thoracic cavity, but also passive atelectasis of the left lower lobe of the lung. Moreover, the size of the tumor lesion in the anterior mediastinum seemed to have decreased, and the low-intensity area within the mass lesion seemed to have expanded.

To explore the source of bloody effusion and confirm the diagnosis of the mass lesion, video-assisted thoracoscopic surgery was performed. Macroscopically, there was approximately 1,400 mL of bloody pleural effusion in the left thoracic cavity. After the effusion had been removed, a well-circumscribed tumor lesion corresponding to the diagnostic CT scan appeared within the thymus. The tumor was removed carefully from the mediastinum. The parenchyma of the tumor was dark red, indicating hemorrhage. Histological examination confirmed the diagnosis of mediastinal intrathymic ectopic parathyroid hyperplasia with hemorrhage (Fig 2). Just after surgery, PTH level was 500 pg/mL (ng/L), which then decreased to 210 pg/mL (ng/L; Fig 3). Also, the persistent cough and sputum disappeared, and C-reactive protein, which was positive on admission, became negative. Postoperative diagnostic CT and ^{99m}Tc-MIBI scintigraphy showed successful removal of the ectopic parathyroid hyperplastic tissue (Fig 4). Pleural effusion and atelectasis of the left lung, which were observed preoperatively, were no longer detected (Fig 4). Finally, the secondary hyperparathyroidism could be controlled clinically by means of medical treatment, and total parathyroidectomy has been postponed indefinitely.

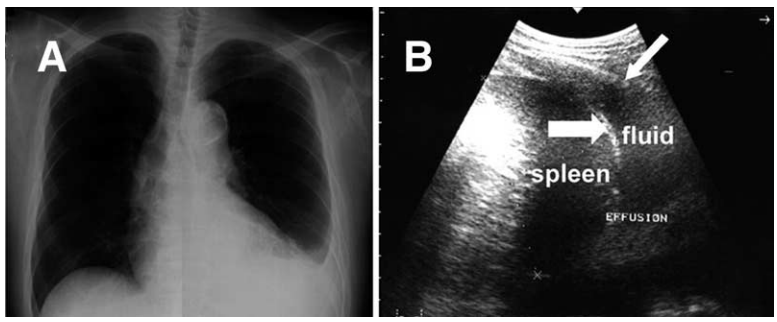

Fig 1. (A) Chest x-ray and (B) ultrasonogram of the left thorax on admission. (A) Upright posteroanterior chest x-ray on the first day of admission shows a blurred left costophrenic angle and obscured diaphragm, suggesting pleural effusion of the left thoracic cavity. (B) Transverse lateral chest ultrasound image obtained with the patient in the supine position and at maximum expiration shows anechoic fluid surrounded by the diaphragm (wide arrow) and parietal pleura (narrow arrow).