


Secondary Mediastinal Bleeding Caused by Parathyroid Adenocarcinoma: A Case Report

Hiep Hoang Phan¹, Cong Thanh Do¹, Luong Ngoc Tran¹, Son Giang Nguyen¹, Diep Ngoc Tran¹, Son Tien Nguyen² and Tuan Dinh Le² 

¹Department of High-Tech Medical Treatment, Vietnam National Endocrinology Hospital, Hanoi Vietnam. ²Department of Rheumatology and Endocrinology, Vietnam Military Hospital 103, Military Medical University, Hanoi, Vietnam.

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ABSTRACT

INTRODUCTION: The clinical picture of parathyroid tumors is mainly related to hypercalcemia such as kidney stones and bone and muscle pain. However, spontaneous cervical hemorrhage due to parathyroidoma bleeding is rare with clinical manifestations of the painful swelling and bruising of the neck accompanied by dysphagia and dyspnea.

CASE PRESENTATION: We report a case of a 71-year-old female patient who presented with acute cervical swelling and extensive bleeding spreading from the neck to the abdomen and 2 flanks. Investigation of patients revealed increased parathyroid hormone levels and hypercalcemia. The neck ultrasound showed the thyroid nodules in 2 lobes, and goiter plongeant on the right. Computed tomography scan images showed a hematoma spreading from the right side of the neck to the mediastinum.

RESULT: The patient required emergency surgery due to dyspnea and hemodynamic instability. The preoperative diagnosis was cervical bleeding with the likely cause being thyroid nodule rupture. However, during the surgery, the bleeding source was determined to be the right parathyroid tumor located deeply below the superior mediastinum. The patient's histopathological result of the tumor is parathyroid adenocarcinoma.

CONCLUSION: From our experience, the hemorrhage from parathyroid tumor should be considered as a cause of acute neck bleeding when no history of trauma or surgery is identified. Post-surgery histopathological analyses of the tumor are very important to detect parathyroid adenocarcinoma.

KEYWORDS: Cervical hemorrhage, parathyroid tumor, parathyroid adenocarcinoma

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CORRESPONDING AUTHOR: Tuan Dinh Le, Department of Rheumatology and Endocrinology, Vietnam Military Hospital 103, Military Medical University, Hanoi 10000, Vietnam. Email: letuan985@gmail.com

Introduction

A parathyroid tumor is a tumor of the parathyroid gland located in the neck, near or attached to the back side of the thyroid gland. Hemorrhage outside the parathyroid glands without risk factors is very rare.¹ The clinical presentation of cervical hemorrhage caused by the rupture of parathyroid tumors was first described by Capps in 1934 after one death due to this cause.² Although rare, the disease progresses seriously and could cause death easily.³ Large hematoma in the cervicothoracic area can compress the trachea, causing dyspnea which leads to respiratory failure; severe bleeding also affects the hemodynamic stability of the patient.^{3,4} Also, the patient may have other symptoms such as dysphagia caused by a hematoma compressing the esophagus or hoarseness due to the compression of the recurrent laryngeal nerve. Localized symptoms of parathyroid hemorrhage include ecchymosis, diffuse neck swelling and tenderness.²

The reason why many authors reach a consensus on the pathogenesis is an imbalance between tumor growth velocity and its blood supply. Parathyroid cell death is more likely to

result in bleeding points, which is similar to the hemorrhage in other cysts like the thyroid cyst. However, the cell wall and fibrous capsule of the thyroid gland are relatively thicker than those of parathyroid tumors,⁵ so the hemorrhage of the thyroid tumor is mainly intracapsular. In contrast, extracapsular hemorrhage from the parathyroid tumor is more common.⁶ Due to the diverse and nonspecific clinical symptoms, the disease is often diagnosed late. Moreover, the clinical picture can be confused with a ruptured thoracic aortic aneurysm, necrotizing mediastinitis, and hemorrhage from atypical thyroid nodules.⁴

Case Presentation

A 71-year-old female patient was admitted to the Department of Intensive Care of the National Hospital of Endocrinology on June 18, 2020, due to swelling and pain in the neck area with dermal hemorrhage spreading from the neck to the abdomen and 2 flanks. Additionally, the patient presented with mild dyspnea, no chest pain, cough or fever. She had no history of neck surgery; however, 5 years ago she was diagnosed with a solitary nodular goiter on the right lobe of the thyroid gland





Figure 1. Extensive subcutaneous bleeding in neck, chest, and abdomen area in the patient.

growing to the mediastinum and a colloid cyst on the left lobe in a hospital, which has not received any treatment yet. In addition to this, her past medical history included osteoarthritis pain for many years, untreated osteoporosis, kidney stone surgery 8 years ago, and stone dissipation 5 years ago. On admission, a physical examination revealed a thin and weak patient with the presence of scoliosis and pale skin mucosa, heart rate of 90 bpm, blood pressure of 100/60 mmHg, temperature 37°C, and SpO₂ of 98%.

The patient had signs of bleeding spreading from neck to abdomen and flanks on both sides and swollen neck on both sides with soft density; impalpable thyroid (Figures 1 and 2).

Further investigation has been made for hematology: hemoglobin of 68 g/L (125-160 g/L), hematocrit of 21.8% (37%-47%), white blood cells of 9.2 G/L (4-10 G/L), % neutrophil of 92% (55%-70%), and platelet of 170 G/L (150-450 G/L); and biochemistry: parathyroid hormone (PTH) of 85 pmol/L (1.6-6.9 pmol/L), total calcium of 3.23 mmol/L (2.20-2.65 mmol/L), ionized calcium of 1.74 mmol/L (1.17-1.30 mmol/L), free T3 of 3.47 pmol/L (0.54-2.96 nmol/L), free T4 of 14.06 pmol/L (12-22 pmol/L), and thyroid-stimulating hormone (TSH) of 0.78 μ UI/mL (0.35-4.94 μ IU/mL).

Neck ultrasound showed the thyroid nodules well-demarcated, mixed tumor, hypoechoic, and uncalcified in 2 lobes (TIRADS 3)/goiter plongeant, right lobe nodule size of 27 \times 46 mm, spreading deep below the muzzle, left lobe nucleus size of 21 \times 27 mm while abdominal ultrasound showed kidney stones on both sides (Figure 3).

CT Scanner: a high-density hematoma (fluid-blood) spreading vertically from the right neck to the median then wriggling into the great blood vessels in the mediastinum, reaching far below the tracheal bifurcation (Figure 4).

The patient was admitted to the hospital with the clinical presentation of a painful and swollen neck accompanied by significant dermal hemorrhage at the neck-abdomen-chest. This patient had anemia and was assigned to receive 1 unit of red blood cells after 12 hours of hospital admission. On the CT



Figure 2. Image of scoliosis and hemorrhage on the back of the patient.

Scanner images, it can be seen the mediastinal bleeding, so we were considering the cause of the cervical tumor.

The patient was indicated for emergency surgery after 20 hours of hospital admission due to excessive anemia and beginning to have symptoms of dyspnea. At the beginning of hospitalization, which are 11 hours and 15 hours after admission, the patient's hemoglobin concentrations were 87, 76, and 68 g/L respectively; hematocrit concentrations were 28.6%, 24.3%, and 21.3% respectively. Bleeding from the neck and chest area was still going on.

The pre-operative likely diagnosis for the patient was neck bleeding caused by thyroid nodule or rupture of parathyroid tumors. At the beginning of the thyroidectomy surgery, the entire thyroid gland was found to be soaked in maroon blood. Many nodules in the right lobe of the thyroid gland were palpable, but the bleeding point was not clear yet. We performed a right-lobe thyroidectomy. However, in the process of dissection to the right sternum, we found that the right lower parathyroid tumor was hypertrophic and located deeply in the upper mediastinum. A black tumor of 2.5 \times 6 \times 2.5 cm was found to be the bleeding source (Figure 5). Diagnosis of parathyroid tumors was established by the rapid reduction of PTH upon the surgery (from 85 to 6.5 pmol/L).

The patient's histopathological result of the tumor is consistent with parathyroid adenocarcinoma: Tumor tissue is composed of clusters, bands of cells, connected together to form a reticular net. Tumor cells are polyhedral, rather monomorphic, with round nuclei, scattered chromatin, blurry nuclei, and huge cytoplasm. Vascularized interstitial stroma lined with raft and

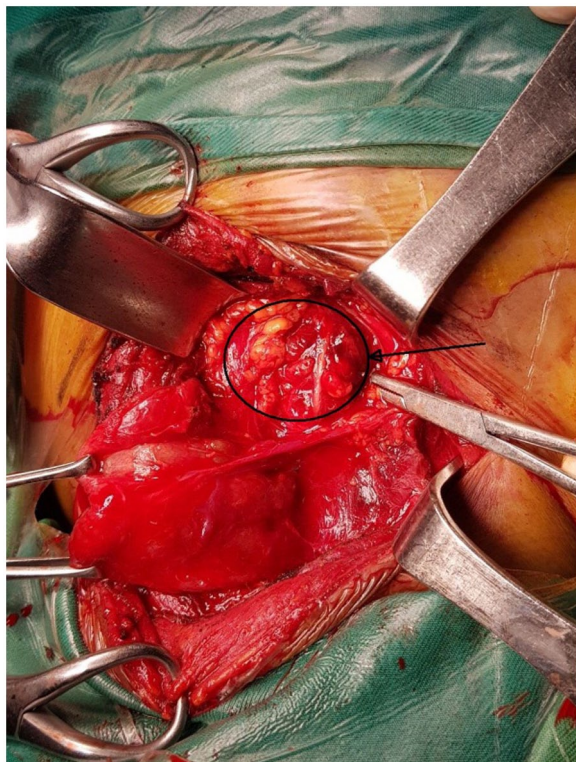


Figure 5. The tumor found in the surgery. Black arrow indicates the tumor. Black oval indicates the area of the tumor.

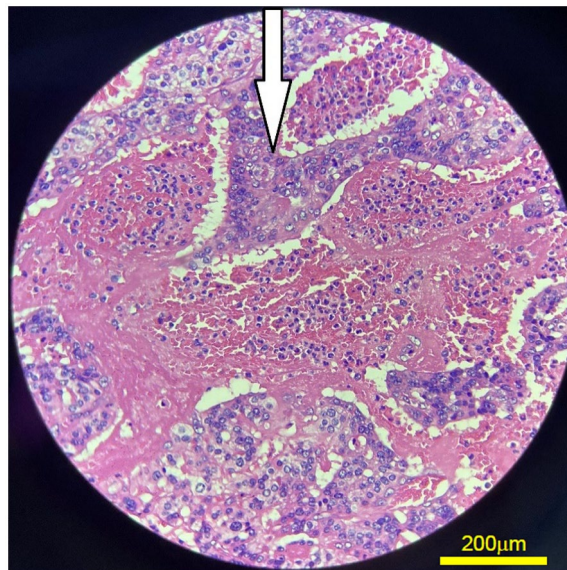


Figure 6. Hematoxyline-Oesine staining images of the tumor under light microscope (40 \times). The image is consistent with parathyroid carcinoma. White arrow indicates Tumor tissue is composed of clusters, bands of cells, connected together to form a reticular net. Tumor cells are polyhedral, rather monomorphic, with round nuclei, scattered chromatin, blurry nuclei, and huge cytoplasm. Vascularized interstitial stroma lined with raft and glandular structures, surrounded by hemorrhages and dilated vascular sinuses. Scale bar, 200 μ m.

following up on some cases of primary hyperparathyroidism that have recovered. To explain this phenomenon, these authors suggest that the rupture of the gland causes damage to the nourishing vessels, thereby causing malnutrition to the parathyroid tumor before dying on its own.³ However, one case also has been recently reported in Ireland that acute cervical hemorrhage due to parathyroid tumors bleeding caused large airway compression leading to respiratory failure.¹² The patient who was treated in the intensive care unit of Beaumont Hospital (Dublin, Ireland) was intubated to maintain respiration and indicated an emergency surgery the day after to resolve the trachea and parathyroid tumor compressions due to hematoma.

Histology was consistent with parathyroid adenocarcinoma. Parathyroid carcinoma is a rare endocrine malignancy, that may begin with parathyroid adenoma, accounting for less than 1% of cases of primary hyperparathyroidism.^{13,14} The clinical manifestations of parathyroid carcinoma are mainly due to the effects of markedly elevated serum PTH levels and hypercalcemia rather than local infiltrates or distant metastases. suggests the progression and transformation of parathyroid adenoma into carcinoma.^{15,16} Histology is the optimal method to diagnose the tumor parathyroid adenocarcinoma.¹⁷ Surgery is the treatment for parathyroid carcinoma, which involves complete resection of the primary lesion. For this reason, both preoperative suspicion and intraoperative awareness are important because treatment with chemotherapy and radiotherapy is not helpful.¹⁸

Conclusions

When the patient presents with symptoms of swelling, pain, and an acute swollen neck with extensive hemorrhage in the thoracic region without a history of trauma or thoracic surgery, parathyroid tumors hemorrhage should be considered. The surgical treatment for the patient is to remove the bleeding parathyroid tumor, treat the hematoma that compresses the airways, and remove the tumor for pathology. Surgery is the optimal treatment when the pathological findings are consistent with parathyroid adenocarcinoma.

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Author Contributions

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

Ethical Statement

Participant provided written informed consent and agreed to join our study. The study was conducted in accordance with the Declaration of Helsinki, and approved by the Ethical Review Committee of Military Hospital 103 (Reference No.188/2022/HĐĐĐ)

ORCID iD

Tuan Dinh Le  <https://orcid.org/0000-0003-2633-583X>

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