

Intra-thoracic Parathyroid Adenomatosis: A Case Report

Wan Kee Kim, M.D., Dong Kwan Kim, M.D., Se Hoon Choi, M.D., Hyeong Ryul Kim, M.D.,
Yong Hee Kim, M.D., Seung-Il Park, M.D.

Ectopic mediastinal parathyroid adenomas are rare, but can be life-threatening. Resection is indicated in those cases accompanied by hypercalcemia, especially in young patients. Although most mediastinal parathyroid adenomas can be removed by a cervical approach, a transthoracic approach is needed when the adenoid tissues are located deep within the thoracic cavity. We describe the case of a 37-year-old female who underwent excision of an intra-thoracic ectopic parathyroid adenoma after parathyroidectomy four months earlier.

Key words: 1. Parathyroid
2. Adenomatosis
3. Operation

CASE REPORT

A 37-year-old female who had undergone a total parathyroidectomy 4 months previously for a parathyroid adenoma presented in our clinic with high serum calcium (10.3 mg/dL) and high intact parathyroid hormone levels (iPTH; 1,378 pg/mL). A technetium-99m sestamibi scan was performed to determine whether there was any remnant parathyroid tissue or whether there was a satellite adenoma. A focal nodule with increased uptake of technetium was detected in the left lower paratracheal area and suspected to be an ectopic parathyroid adenoma (Fig. 1). The mass was located on the left side of the pulmonary trunk, and we decided to perform a surgical biopsy. Preoperatively, we performed a chest computed tomography (CT) scan to localize the mass more accurately without enhancement because of her impaired renal function. She had been diagnosed with chronic rejection following transplantation of both kidneys, and was on hemo-

dialysis. The mass was located between the aortic arch and the pulmonary trunk, apparently adhering to the inferior side of the aortic arch in the CT scan (Fig. 2).

Because the nodule was located deep inside the lower paratracheal area and was thought to adhere to the aortic arch, and a recurrent laryngeal nerve was nearby, excision through a posterolateral thoracotomy was planned. After opening the left 5th intercostal space with one lung ventilation, the mediastinal pleura was opened in the aorto-pulmonary window. A firm and brownish mass about 2 cm in diameter was seen to be in contact with the lesser curvature of the aortic arch. During the operation, the recurrent laryngeal nerve was saved by snaring. The mass was carefully dissected free from the aorta, and it was sent for pathological examination. A frozen biopsy revealed a parathyromatosis. A 24 Fr chest tube was inserted and the wound was closed layer by layer. She was then transferred to a general ward with tolerable vital signs.

The immediately postoperative serum iPTH level was 32.9

Department of Thoracic and Cardiovascular Surgery, Asan Medical Center, University of Ulsan College of Medicine

Received: December 4, 2012, Revised: January 5, 2013, Accepted: January 10, 2013

Corresponding author: Dong Kwan Kim, Department of Thoracic and Cardiovascular Surgery, Asan Medical Center, University of Ulsan College of Medicine, 88 Olympic-ro 43-gil, Songpa-gu, Seoul 138-736, Korea
(Tel) 82-2-3010-3586 (Fax) 82-2-3010-6966 (E-mail) dkkim@amc.seoul.kr

© The Korean Society for Thoracic and Cardiovascular Surgery. 2013. All right reserved.

© This is an open access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/3.0>) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

pg/mL, and it had decreased to 5.1 pg/mL by the time of discharge. The postoperative course was uneventful and the patient was discharged on postoperative day 6 without any problems. The last follow-up took place 4 months after the excision, and the patient had been doing well, without any complications.

DISCUSSION

Ectopic hyperfunctioning mediastinal parathyroid tissues are found in 11% to 25% of patients with hyperparathyroidism. Most of these patients are asymptomatic. In rare cases, however, the patient's status can be life-threatening, such as with metabolic encephalopathy, cardiac arrhythmia, or renal failure

following a sudden calcium surge [1,2]. In general, surgical resection is indicated for symptomatic ectopic parathyroid adenomas. Asymptomatic patients, however, also require surgical resection when their serum calcium concentration exceeds 1.0 mg/dL, their creatinine clearance is reduced, their bone density is low, and they are under 50 years of age [3]. Though 98% of such mediastinal parathyroid adenomas are resectable via a neck collar incision, the other 2% require a different approach [4]. In such cases, planning the operation is important in order to achieve complete surgical resection of ectopic parathyroid adenomas.

This case taught us two crucial lessons about the surgical treatment of hyperparathyroidism due to mediastinal parathyroid tissue. First, the correct choice of opening technique is most important. The favored approaches for resecting parathyroid adenomas are the trans-sternal approach with sternal splitting, the trans-thoracic approach, the mediastinoscopic approach, and angiographic ablation. It has been established that the trans-thoracic approach is superior to the trans-sternal approach in terms of reducing complication rates [5]. The trans-thoracic approaches include open thoracotomy and video-assisted thoracic surgery (VATS). The removal of ectopic mediastinal parathyroid glands using VATS has been described in several case reports and case series [4] and is known to be less harmful to adjacent tissues than angiographic ablation. Because a minimally invasive modality such as VATS and a subxiphoidal laparoscopic approach using a mediastinoscope confer several benefits, such as shorter hospital stays and less pain, they have been described as alternatives to sternotomy or open thoracotomy in recent studies [6]. In our case, however, we chose open thoracotomy for

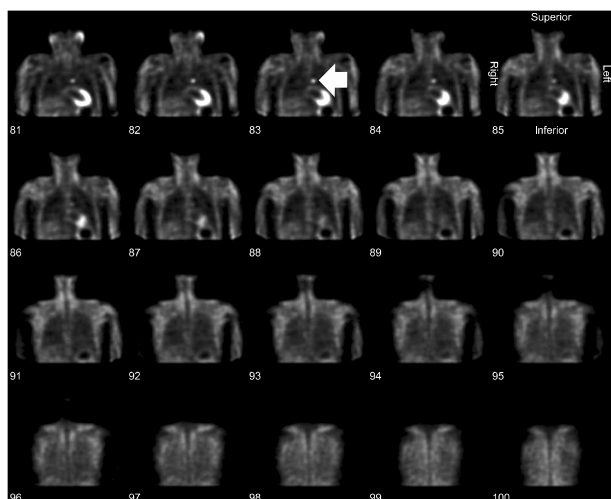


Fig. 1. Preoperative technetium 99m sestamibi scan. Preoperative sestamibi scan shows an increased technetium uptake at the left lower paratracheal area (arrow: the ectopic mediastinal parathyroid adenomatosis).

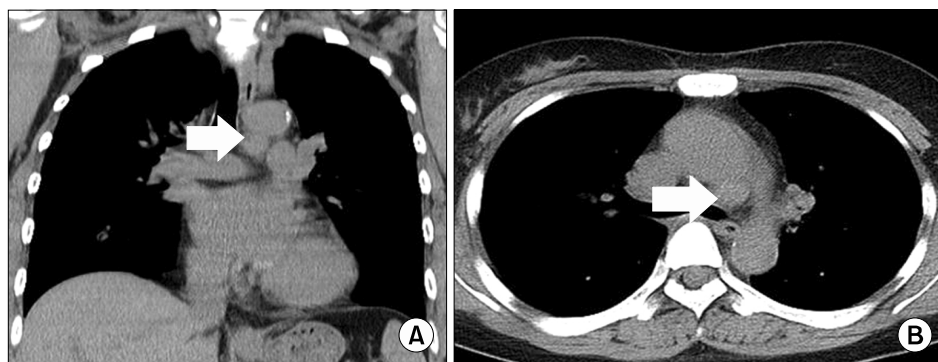


Fig. 2. Preoperative computed tomography (CT). Preoperative CT findings show an ectopic parathyroid adenoma about 1.5 cm located in the aorto-pulmonary window, and it is suspected to be adhered to the aortic arch. (A) Coronal view of mediastinum. (B) Horizontal view of mediastinum (arrow: the ectopic mediastinal parathyroid adenomatosis).

several reasons. The patient had already been diagnosed with chronic rejection following a bilateral kidney transplantation; sufficient localizing exams were not possible. Therefore, the open thoracotomy technique was used to reduce the possibility of another recurrence and address the adhesion of the aortic arch. A nearby recurrent laryngeal nerve was the other reason, in terms of lowering the risk of complications.

Second, preoperative localization by imaging is essential. Older studies have reported failure rates of about 40% for detection without imaging [7]. Ultrasonography of the neck is the first line approach to localization. The mediastinal area, however, cannot be visualized by ultrasonography due to interference by the clavicles and sternum. For this reason, sestamibi scans are used to detect ectopic parathyroid adenomas. The sensitivity of sestamibi scans for detecting mediastinal parathyroid glands is 80% [4], and that for ectopic parathyroid glands is 70% to 80% [8]. CT scans can also be used. Although their specificity is lower than that of sestamibi scans, their accuracy is reported to reach 84% [4]. Hence, either a sestamibi or a CT scan is essential prior to resection of a parathyroid adenoma. In our case, the adenoma was detected and localized with sestamibi scan, and the CT scan also revealed adhesion between the aorta and adenoma.

In the present case, although the first sestamibi scan was negative, the second as well as a CT scan revealed the ectopic parathyroid adenoma. These two types of scan permit accurate localization of adenomas and their precise resection. In addition, intraoperative methods for assessing successful resection such as intraoperative iPTH monitoring [9] and selective venous sampling could be helpful.

In conclusion, we report a case of ectopic parathyroid adenoma that was successfully resected by open thoracotomy following its localization by sestamibi and CT scans.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

REFERENCES

1. Fitzpatrick LA, Bilezikian JP. *Acute primary hyperparathyroidism*. Am J Med 1987;82:275-82.
2. Chung JI, Kim JW, Kim SW, Ku BI, Lee HK. *Mediastinal parathyroid cyst: 1 case report*. Korean J Thorac Cardiovasc Surg 2003;36:59-62.
3. Bilezikian JP, Khan AA, Potts JT Jr; Third International Workshop on the Management of Asymptomatic Primary Hyperthyroidism. *Guidelines for the management of asymptomatic primary hyperparathyroidism: summary statement from the third international workshop*. J Clin Endocrinol Metab 2009;94:335-9.
4. Wei B, Inabnet W, Lee JA, Sonett JR. *Optimizing the minimally invasive approach to mediastinal parathyroid adenomas*. Ann Thorac Surg 2011;92:1012-7.
5. Medrano C, Hazelrigg SR, Landreneau RJ, Boley TM, Shawgo T, Grasch A. *Thoracoscopic resection of ectopic parathyroid glands*. Ann Thorac Surg 2000;69:221-3.
6. Liu RC, Hill ME, Ryan JA Jr. *One-gland exploration for mediastinal parathyroid adenomas: cervical and thoracoscopic approaches*. Am J Surg 2005;189:601-4.
7. Edis AJ, Sheedy PF, Beahrs OH, van Heerden JA. *Results of reoperation for hyperparathyroidism, with evaluation of preoperative localization studies*. Surgery 1978;84:384-93.
8. Phitayakorn R, McHenry CR. *Incidence and location of ectopic abnormal parathyroid glands*. Am J Surg 2006;191:418-23.
9. Sagan D, Gozdzik K. *Surgical treatment of mediastinal parathyroid adenoma: rationale for intraoperative parathyroid hormone monitoring*. Ann Thorac Surg 2010;89:1750-5.