

## Recurred Parathyroid Carcinoma: A Case of Cervical Recurrence Presenting Discrepancy between Image Findings and Operative Findings

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Parathyroid carcinoma is a rare malignancy presenting hyperparathyroidism. At times, diagnosis and localization are difficult. The optimum treatment for parathyroid carcinoma is en bloc resection when malignancy is highly suspicious or diagnosed. However, even after the adequate surgical treatment, persistent or recurrent disease is well encountered. Here we report a case with recurred parathyroid carcinoma presenting discrepancy between image findings and operative findings. (*Korean J Endocrine Surg* 2011;11:35-37)

**Key Words:** Recurred parathyroid carcinoma, Ultrasound, En bloc resection

### INTRODUCTION

Parathyroid carcinoma is a rare malignancy accounting for 0.5% to 5.0% of primary hyperparathyroidism.(1) The clinical feature of parathyroid carcinoma is usually not very distinctive. At times, preoperative diagnosis and localization are difficult.(1-3) The optimum treatment for parathyroid carcinoma is en bloc resection with ipsilateral thyroid lobectomy when malignancy is highly suspicious or diagnosed.(1,2,4) Unfortunately persistent or recurrent disease is well encountered even after the adequate surgical treatment.(5,6)

Localization and surgical resection for suspected recurrence is a challenge for surgeons and such prevalence of persistency or recurrence requires close and life-long observation.(6) Here we report a case with recurred parathyroid carcinoma presenting an unusual cervical recurrence pattern which did not correlate with the report of image findings and operative findings.

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### CASE REPORT

A 41-year-old male was hospitalized in April 2008 due to abdominal pain, both knee pain and high serum calcium level. The patient had history of acute abdominal pain associated with acute pancreatitis for a year prior to hospitalization. He experienced at least 4 times of acute pancreatitis before being referred to our endocrinology for high serum calcium level. The patient had no other history of medication or surgery.

Laboratory and image studies were carried out under the suspicion of primary hyperparathyroidism due to hypercalcemic symptoms and signs. At admission, laboratory examination showed serum calcium level of 15.3 mg/dL (normal 8.3~10.0 mg/dL), ionized calcium level of 8.2 mg/dL (normal 3.9~4.5 mg/dL), and intact parathyroid hormone (iPTH) level of 2,281.0 pg/mL (normal 7.6~75.0 pg/ml).

Abdomen pelvis computed tomography (CT) was taken revealing multiple renal stones in both kidneys, diffuse osteoporosis with multiple brown tumors, but without definite evidence of acute pancreatitis, which correlated with finding of hyperparathyroidism. Series of X-ray taken to evaluate the bone defect in skull, both extremities, and spine showed diffuse osteopenia and multiple osteoblastic lesions.

Neck ultrasound (US) revealed 3.7x3.5 cm size solid and irregular mass around right mid pole of thyroid. No cervical lymphadenopathy was found. Fine needle aspiration biopsy (FNAB) revealed polygonal cell neoplasm with abundant cytoplasm and a few oncocytic cells, suggestive of parathyroid neoplasm. Preoperative 99 m Tc-MIBI parathyroid scan showed focal slightly increased uptake in right thyroid lobe posterior portion suggesting parathyroid adenoma.

At surgery, a hard nodular mass about 4.0x3.5 cm size was found slightly posterior to right thyroid. It was attached to surrounding soft tissue, thyroid and right recurrent laryngeal nerve.

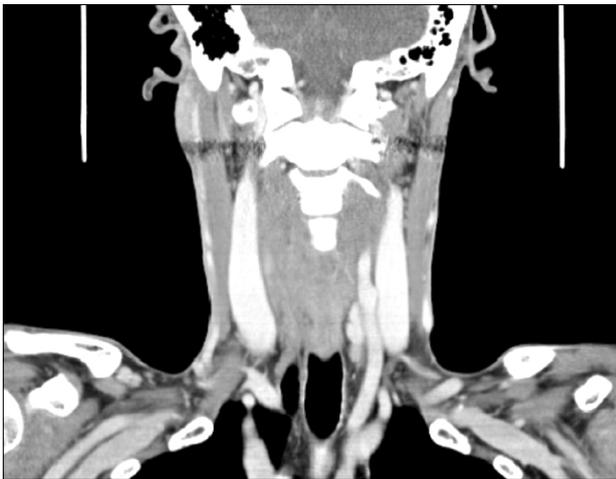
Under the impression of parathyroid malignancy, en bloc resection was performed including parathyroid resection, ipsilateral right thyroid lobectomy, recurrent laryngeal nerve resection and central compartment lymph node dissection. Histopathology revealed 4.5x3.7 cm parathyroid carcinoma with periparathyroidal soft tissue extension and lymphovascular tumor invasion. There was no tumor present in 15 dissected lymph nodes.

Postoperative laboratory examination was performed showing remarkable decrease in serum calcium level to 9.0 mg/dL and iPTH level to 21.8 pg/mL. The patient showed serum calcium level around the normal range and minimal fluctuation of iPTH level around upper normal limit during the close short-term follow-up.

On July 2009, image studies were taken for postoperative observation. Neck CT revealed no evidence of abnormality (Fig. 1) and parathyroid scan was negative. However, neck US detected abnormal lesion at right level IV area (Fig. 2) and FNAB came out as clusters of polygonal oncocytic cells with bland looking round nuclei and clear cytoplasm, suggestive of parathyroid neoplasm.

Reoperation was performed on September 2009 for recurred parathyroid carcinoma. During the exploration of right lateral cervical area, we were not able to find any suspicious lymphadenopathy or metastatic lesions at first even though examination was performed based on the neck US findings. After thorough examination, we detected highly suspicious metastatic congregated lesions about 3.5 cm in length located at posterior medial to common carotid artery. Right modified radical neck dissection was performed for second operation.

Histopathology revealed that out of 36 dissected lymph nodes,



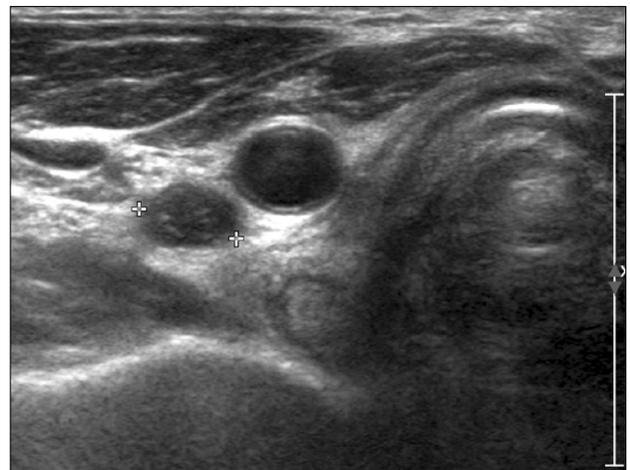
**Fig. 1.** Neck computed tomography showing no evidence of abnormality at area around posterior to right common carotid artery.

10 came out as metastatic parathyroid carcinoma. However, there was no metastasis found at routinely dissected lateral neck lymph nodes. All 10 metastatic lymph nodes were located at lesions found at posterior medial to common carotid artery with maximum diameter of metastatic tumor size of 1.6 cm. Postoperative serum calcium level and iPTH level after reoperation were 9.6 mg/dL and 26.2 pg/mL, respectively.

## DISCUSSION

Parathyroid carcinoma is a rare malignancy causing hyperparathyroidism. Markedly elevated serum calcium level and iPTH level with associated obvious hypercalcemic symptoms suggest potential malignancy.<sup>(1)</sup> When hyperparathyroidism is detected, image study is performed including neck US to detect the location of the parathyroid lesion. However the clinical feature of parathyroid carcinoma is usually not very distinctive compared to benign parathyroid diseases. At times, preoperative diagnosis and localization are difficult to perform.<sup>(3)</sup> Commonly used imaging tools are neck CT, neck US and parathyroid scan to located the parathyroid lesions. However, these image findings do not always correlate with each other and preoperative clinicopathological diagnosis of parathyroid carcinoma is very unlikely.

Due to such circumstances, the surgeons have to decide the extent of surgery for suspected malignancy under the collaboration of preoperative image findings and intra-operative gross findings. The treatment of choice for suspected parathyroid carcinoma is en bloc resection of the involved parathyroid with ipsilateral thyroid lobectomy and central compartment lymph node dissection.<sup>(1,3,7)</sup> Sandelin et al<sup>(4)</sup> reported that with such ex-



**Fig. 2.** Neck ultrasound showing 0.7x0.6 cm suspicious metastatic lymphadenopathy at right level IV area.

tensive surgery, the patients had longer survival and a longer relapse-free period. However, recurrence is commonly encountered even after such surgical treatment. When local recurrence and regional lymph node metastasis occurs, reoperation is highly recommended with a wide resection of the involved area including aggressive resection of contiguous structures such as the trachea and muscle layer of esophagus.(1,2,5,7) To perform reoperation for recurred parathyroid carcinoma, localization and surgical resection is the challenging task for surgeons. In our case, recurrence was only detected in neck US. Neck CT and parathyroid scan revealed negative findings. To our knowledge, neck US seems beneficial in detecting small nodular recurrence but precise localization was the limitation. In our presented case, the recurred metastatic lymph nodes were located posterior medial to common carotid artery which did not correlate with the US findings. Even though neck US findings suggested metastatic lateral neck lymph nodes in right level IV area, actual intra-operative findings revealed congregated metastatic lesions located at posterior medial to common carotid artery which could be stated as central compartment area. One of the reasons for this discrepancy could be due to anatomic structural distortion after thyroid lobectomy and central compartment node dissection. The common carotid artery could have been displaced more medial than normally detected position after surgery giving misconception of lateral neck lymph node area.

We conclude that when parathyroid carcinoma is diagnosed and surgically treated, recurrence is a common encounter. And, when recurrence is detected, reoperation is the treatment of choice. However, there is limitation and difficulty in localization of the recurred lesion. Therefore, even though the reoperation is performed based on the image findings such as neck US and CT, additional thorough examination of the surrounding structures including previous operative field is essential.

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