A parathyroid cyst associated with an adenoma causing primary hyperparathyroidism (PHPT): Gamma camera localization for case with MIBI scan-negative PHPT

Takaaki Fujii, Reina Yajima, Hironori Tatsuki, Hiroyuki Kuwano

ABSTRACT

Introduction: A cystic lesion of the parathyroid gland causing hyperparathyroidism is an uncommon finding. Tc-99m MIBI scintigraphy is a useful preoperative diagnostic tool in primary hyperparathyroidism (PHPT). We report a rare case of a patient with PHPT with a functional parathyroid cyst in whom MIBI scintigraphy failed to detect a parathyroid tumor.

Case Report: A 70-year-old Japanese man was referred to our hospital for the examination and treatment of hypercalcemia and a high level of intact PTH. Computed tomography scan, Magnetic resonance imaging scan, and ultrasonography of his neck showed a cystic mass in the lower right side of the neck measuring approximately 3.0×2.0 cm, whereas MIBI scintigraphy failed to detect a parathyroid tumor. Resection of the right cystic mass was performed. Scintigraphy images of the neck were acquired by an eZ-SCOPE hand-held gamma camera before the skin incision, and ex vivo imaging of the specimen was performed, which was useful for the navigation surgery and minimally invasive parathyroidectomy. Histopathology showed a parathyroid adenoma with prominent cystic degeneration.

Conclusion: False-negative diagnostic results have been reported in cystic parathyroid adenomas. In cases of cystic hyperfunctioning scan-negative parathyroid tumors, the eZ-SCOPE may be useful for the localization and navigation surgery of primary hyperparathyroidism due to a cystic parathyroid tumor.
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Keywords: Gamma camera, Navigation surgery, Primary hyperparathyroidism, Parathyroid cyst, Sestamibi scintigraphy

INTRODUCTION

Primary hyperparathyroidism (PHPT) is a condition characterized by an excess secretion of parathyroid hormone by adenomatous or hyperplastic glands [1, 2]. The accurate preoperative localization of parathyroid adenomas is important to reduce the operative failure rate [2, 3]. Tc-99m sestamibi (MIBI) scintigraphy is a useful preoperative diagnostic tool for PHPT [4, 5].
The association of a functional parathyroid cyst with a parathyroid adenoma is an uncommon finding [6–9]. It was reported that in some patients with PHPT and a functioning parathyroid cyst, Tc-99m MIBI scintigraphy failed to detect a parathyroid tumor [6, 7]. There have also been some reports of radio-guided surgery for PHPT [10–19]. We recently demonstrated that in scan-positive patients identified by preoperative Tc-MIBI, Tc-MIBI scintigraphy with the use of a hand-held gamma camera, the eZ-SCOPE AN (Anzai Medical Co. Ltd., JAPAN), is useful for navigation surgery for PHPT and minimally invasive parathyroidectomies [18, 19].

The eZ-SCOPE AN is designed to be used as a hand-held, regional diagnostic imaging device, which is a new compact-type semiconductor gamma camera based on the use of a cadmium zinc telluride (CdZnTe) [18–21]. We report here a rare case of a patient with a functioning parathyroid cyst in whom MIBI scintigraphy failed, but in whom a gamma camera could detect a parathyroid tumor.

**DEVICE DESCRIPTION**

The eZ-SCOPE AN is designed to be used as a hand-held, regional diagnostic imaging device [17–20]. This compact-type semiconductor gamma camera made of CdZnTe has 256 semiconductors representing the same number of pixels. Each semiconductor is a 2-mm square and is located in 16 lines and rows on the surface of the detector. The outer dimensions of the camera are 74×72×210 mm, and it weighs 820 g. The most significant differences between it and previous cameras include the spatial resolution, sensitivity, high count rate characteristics, and energy resolution [18–21].

**CASE REPORT**

The patient was a 70-year-old Japanese man with a history of hepatocellular carcinoma with chronic hepatitis C. Hypercalcemia and a high level of intact parathyroid hormone (PTH) had been detected 27 months prior to his admission to our hospital. Neck ultrasonography and computed tomography (CT) scan revealed a right parathyroid tumor, but a Tc-99m MIBI scintigraphy examination failed to detect a parathyroid tumor. He had been followed up because he was also suffering from the hepatocellular carcinoma with liver cirrhosis. His serum calcium level had been significantly elevated, and thus he was referred to our hospital for further examination and treatment of hypercalcemia and the high level of intact PTH. On admission, his serum calcium was 14.4 mg/dL, albumin was 3.7 g/dL, and the intact PTH level was 423 pg/mL. Computed tomography scan of his neck showed a hypodense mass in the right side of the neck measuring approximately 3.0×2.0 cm (Figure 1).

Magnetic resonance imaging (MRI) scan showed a mass in the right neck, which showed low to intermediate signal intensity on T1-weighted images and high signal intensity on T2 images compared to the surrounding tissues (Figure 2). Neck ultrasonography (US) revealed a cystic mass measuring 3.0×1.5 cm in the right neck (Figure 3). Tc-99m MIBI scintigraphy was performed to diagnose primary hyperparathyroidism and examine other parathyroid glands, but it failed to detect a parathyroid tumor (Figure 4). As there was suspicion of hyperparathyroidism due to the functioning parathyroid
cyst, we performed resection of the right cystic mass. Scintigraphy images of the neck were acquired with the eZ-SCOPE AN before the skin incision, and ex vivo imaging of the specimen was performed (Figure 5). This gamma camera was useful for the navigation surgery and minimally invasive parathyroidectomy.

The intact PTH level was measured 10 min after the removal of the cystic mass, and we confirmed a decrease in the intact PTH level. Histopathology showed a parathyroid adenoma with prominent cystic degeneration (Figure 6). Our follow-up of the patient has remained uneventful, with no sign of recurrent disease.

**DISCUSSION**

Tc-99m MIBI scintigraphy has been widely used with high accuracy for the detection and localization of parathyroid tumors [4, 5]. Here we encountered a rare case of a patient with PHPT with a functioning parathyroid cyst in whom Tc-99m MIBI scintigraphy failed to detect a parathyroid tumor. Cystic lesions of the parathyroid gland are very uncommon, and the association of a functional parathyroid cyst with a parathyroid adenoma is an uncommon finding [6–9]; the most common cause is cystic degeneration of the adenomas, which is consistent with our case. Computed tomography scan, magnetic resonance imaging scan and ultrasonography could demonstrate our patient’s cystic parathyroid adenoma, whereas the Tc-99m MIBI scintigraphy failed to detect the cystic adenoma.

Although the exact mechanisms of the visualization of abnormal and hyperfunctioning parathyroid tissue by MIBI scintigraphy are not clear, false-negative diagnostic results have been reported in cystic parathyroid adenoma [6, 7]. In our previous study, we found that for single adenomas and cases with positive Tc-MIBI scans, radio-guided surgery is an effective tool in the surgical management of primary hyperparathyroidism [18, 19]; however, in cases that are scan-negative due to cystic disease, radio-guided surgery is also thought to be useful.
CONCLUSION

We have reported a rare case of a patient with a cystic parathyroid adenoma presenting hyperparathyroidism in whom a gamma camera could detect the parathyroid tumor. Preoperative diagnoses can be challenging for cystic parathyroid adenomas. In some cystic hyperfunctioning scan-negative parathyroid tumor cases, the eZ-SCOPE AN may be useful for the localization and the navigation surgery of primary hyperparathyroidism due to a cystic parathyroid tumor.

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Author Contributions

Takaaki Fujii – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Reina Yajima – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Hirnori Tatsuki – Substantial contributions to conception and design, Drafting the article, Final approval of the version to be published
Hiroyuki Kuwano – Substantial contributions to conception and design, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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