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.
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.

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 with peripherally enhancing walls measuring approximately 3.0 × 2.0 cm (Figure 1).

Magnetic response 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

Figure 1: Computed tomography of neck showing a hypodense mass in the right side of the neck with peripherally enhancing walls measuring approximately 3.0 × 2.0 cm.

Figure 2: Magnetic resonance imaging showing a mass in the right neck which showed low to intermediate signal intensity on T1-weighted images (a) and high signal intensity on T2 images (b) compared to the surrounding tissues.
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.

**********

Acknowledgements
We would like to thank Saitoh Y, Yano T, Matsui Y, Ishida A, Ishikubo A for their secretarial assistance.

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.

Copyright
© 2016 Takaaki Fujii et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES
15. Ortega J, Ferrer-Rebolleda J, Cassinello N, Lledo S. Potential role of a new hand-held miniature gamma camera in performing minimally invasive


Edorium Journals: An introduction

Edorium Journals Team

About Edorium Journals
Edorium Journals is a publisher of high-quality, open access, international scholarly journals covering subjects in basic sciences and clinical specialties and subspecialties.

Invitation for article submission
We sincerely invite you to submit your valuable research for publication to Edorium Journals.

But why should you publish with Edorium Journals?
In less than 10 words - we give you what no one does.

Vision of being the best
We have the vision of making our journals the best and the most authoritative journals in their respective specialties. We are working towards this goal every day of every week of every month of every year.

Exceptional services
We care for you, your work and your time. Our efficient, personalized and courteous services are a testimony to this.

Editorial Review
All manuscripts submitted to Edorium Journals undergo pre-processing review, first editorial review, peer review, second editorial review and finally third editorial review.

Peer Review
All manuscripts submitted to Edorium Journals undergo anonymous, double-blind, external peer review.

Early View version
Early View version of your manuscript will be published in the journal within 72 hours of final acceptance.

Manuscript status
From submission to publication of your article you will get regular updates (minimum six times) about status of your manuscripts directly in your email.

Our Commitment

Six weeks
You will get first decision on your manuscript within six weeks (42 days) of submission. If we fail to honor this by even one day, we will publish your manuscript free of charge.*

Four weeks
After we receive page proofs, your manuscript will be published in the journal within four weeks (31 days). If we fail to honor this by even one day, we will publish your manuscript free of charge and refund you the full article publication charges you paid for your manuscript.*

Favored Author program
One email is all it takes to become our favored author. You will not only get fee waivers but also get information and insights about scholarly publishing.

Institutional Membership program
Join our Institutional Memberships program and help scholars from your institute make their research accessible to all and save thousands of dollars in fees make their research accessible to all.

Our presence
We have some of the best designed publication formats. Our websites are very user friendly and enable you to do your work very easily with no hassle.

Something more...
We request you to have a look at our website to know more about us and our services.

* Terms and condition apply. Please see Edorium Journals website for more information.

We welcome you to interact with us, share with us, join us and of course publish with us.