



Case Report

A case report of a functioning mediastinal parathyroid cyst with no ^{99m}Tc -MIBI uptakeNaotaka Uchida ^{a,*}, Manabu Yoshida ^b, Yasuki Matsui ^a, Takeshi Notsu ^a^a Matsue City Hospital, Department of Breast, Endocrine, Vascular and Thoracic Surgery, 32-1, Noshira-cho, Matsue City, Shimane 690-8509, Japan^b Matsue City Hospital, Department of Pathology, 32-1, Noshira-cho, Matsue City, Shimane 690-8509, Japan

ARTICLE INFO

Article history:

Received 7 February 2018

Received in revised form

1 May 2018

Accepted 7 May 2018

Available online 8 May 2018

Keywords:

Parathyroid cyst

Mediastinal mass

Hyperparathyroidism

 ^{99m}Tc -MIBI scintigraphy

Case report

ABSTRACT

Introduction: Functioning mediastinal parathyroid cysts are extremely rare. Technetium-99m-methoxyisobutylisonitrile (^{99m}Tc -MIBI) scintigrams usually helps to localize parathyroid lesions, but we report a case with no MIBI uptake.

Presentation of case: A 79-year-old woman with lower extremity edema and dysphagia was referred to our hospital. Her blood calcium (Ca) level was 11.1 mg/dl, and the intact parathyroid hormone (PTH) level was 132 pg/ml. Computed tomography showed a 3.5 cm solid nodule in the right thyroid lobe and a 5.4 cm cystic mass extending to the mediastinum. The ^{99m}Tc -MIBI scintigram showed abnormal uptake in the thyroid nodule but no uptake in the mediastinal mass. The diagnosis was a thyroid nodule in the right lobe, primary hyperparathyroidism and a mediastinal mass. Under general anesthesia, the thyroid nodule and the mediastinal mass were resected via a cervical incision. The right lower parathyroid gland was resected because it measured about 1 cm and had the dark red color of a parathyroid adenoma. The pathological diagnoses were adenomatous goiter, normal parathyroid gland and mediastinal parathyroid cystic adenoma. The histopathological examination of the mediastinal mass showed a uniform population of parathyroid cells. The postoperative serum Ca and intact-PTH levels were normal. There has been no relapse.

Discussion and Conclusion: Patients with parathyroid cysts sometimes have negative ^{99m}Tc -MIBI scans. We should consider a parathyroid cyst when a mediastinal cystic mass is demonstrated. Surgical resection is thought to be the optimal therapeutic choice for mediastinal parathyroid cysts irrespective of functioning because mediastinal parathyroid cysts are large and produce compressive symptoms.

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1. Introduction

Parathyroid cysts are rare lesions of the neck or mediastinum. Among 6621 patients who underwent neck ultrasonography mainly for evaluation of thyroid disease, a parathyroid cyst was diagnosed in 5 cases (0.075%) [1]. While 85% of cases parathyroid cyst are located in the neck, the rest can occur in the mandible or mediastinum [2].

Technetium-99m-methoxyisobutylisonitrile (^{99m}Tc -MIBI) scintigraphy helps in the localization of parathyroid lesions. However, to our knowledge, some cases of ^{99m}Tc -MIBI scans showing

negative ^{99m}Tc -MIBI uptake in parathyroid cysts have been reported previously (Table 1) [3–5].

We report the case of a mediastinal parathyroid cyst that had a negative ^{99m}Tc -MIBI scan. We discuss the difficulties of its preoperative diagnosis with fine-needle aspiration for mediastinal parathyroid cyst. We also discuss treatment for it.

We have reported our case in line with SCARE criteria [6].

2. Case presentation

A 79-year-old woman with a 5-month history of edema in the lower extremities was referred to our hospital for evaluation of possible deep venous thrombosis. She also had dysphagia. Her past medical history included a first lumbar stress fracture, osteoporosis, osteoarthritis and hypertension. She had taken the following drugs: alfacalcidol (vitamin D), etodolac (a non-steroidal anti-inflammatory drug [NSAID]), irbesartan (an angiotensin II

Abbreviation: Technetium-99m-methoxyisobutylisonitrile, ^{99m}Tc -MIBI; calcium, Ca; phosphate, P; parathyroid hormone, PTH.

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Table 1
Reported cases of parathyroid cyst in which ^{99m}Tc -MIBI uptake was negative.

Ref. No.	First author Year	Age sex	Size (cm)	Ca (mg/dl)	Intact-PTH (pg/ml)	function	Place
3	Gough IR 1999	61 male	2	11.0	85	functioning	neck
4	Wirowski D 2008	70 female	2.5	11.7	269	functioning	neck
4	Wirowski D 2008	66 female	2.2	9.8	n.a.	non-functioning	mediastinum
4	Wirowski D 2008	56 Male	5	11.2	151	functioning	neck
5	Hoi WH 2011	56 female	10	15.6	437	functioning	mediastinum
Our case	Uchida N 2017	79 female	5.4	11.1	132	functioning	mediastinum

n.a.: not assessed.

receptor blocker [ARB]). She had no family history of endocrine disease. Physical examination revealed bilateral crural edema and a palpable thyroid mass.

She did not have deep venous thrombosis. Laboratory findings revealed an elevated calcium (Ca) concentration (11.1 mg/dl, normal range: 8.5–10.2 mg/dl), a normal phosphate (P) concentration (2.9 mg/dl, normal range: 2.4–4.4 mg/dl), and an elevated intact-parathyroid-hormone (PTH) concentration (132 pg/ml, normal range: 10–65 pg/ml). Her thyroid function, thyroglobulin level, antithyroglobulin level and kidney function were within normal limits. Ultrasonography detected a 3.8 cm, well-defined, follicular mass of the right thyroid lobe and a mass extending from the lower end of the right thyroid lobe to the mediastinum. Computed tomography showed a 3.5 cm solid nodule in the right lobe of the thyroid and a 5.4 cm cystic mass extending to the mediastinum (Fig. 1). ^{99m}Tc -MIBI scintigram showed high uptake in the thyroid nodule but no uptake in the mediastinal mass (Fig. 2A/B). The reconstructed ^{99m}Tc -MIBI single photon emission computed tomography (SPECT) images were graded as negative, as no localization that corresponded in anatomic location to the mediastinal mass was observed (Fig. 2C/D). Fine-needle aspiration showed no malignant cells in the thyroid nodule. The preoperative diagnosis was primary hyperparathyroidism, a thyroid nodule, and a mediastinal mass.

Under general anesthesia, resection of the thyroid nodule and the mediastinal mass were performed via a cervical incision. Parathyroidectomy of the right lower parathyroid gland was done because it was about 1 cm in size and had a dark red color

reminiscent of a parathyroid adenoma. The intact PTH level was measured 15 minutes after excision of the suspected parathyroid adenoma. The intraoperative intact PTH level was 75 pg/ml, which was 56% of the preoperative value. The patient's serum Ca and intact PTH concentrations decreased to normal levels (Ca: 8.8 mg/dl, intact PTH: 17 pg/ml) on postoperative day 1. The histopathological diagnosis of the thyroid nodule, parathyroid gland and mediastinal mass were adenomatous goiter, normal parathyroid tissue and parathyroid cystic adenoma, respectively. Histopathological examination of the mediastinal mass showed a uniform cell population consisting of parathyroid cells secreting PTH (Fig. 3). No findings of malignancy were detected.

The postoperative serum Ca and intact PTH levels remained within the normal range. There was no recurrence of the mediastinal mass.

3. Discussion

An issue with our case was the negative ^{99m}Tc -MIBI results regarding detection of the parathyroid cyst. ^{99m}Tc -MIBI scan is a useful tool to detect ectopic or multiple parathyroid lesions. A meta-analysis of ^{99m}Tc -MIBI planar scintigraphy used to detect parathyroid adenomas in primary hyperparathyroidism showed a sensitivity of 84% and specificity of 80% [7]. ^{99m}Tc -MIBI SPECT imaging increased the sensitivity, which ranged from 68% to 95% [8,9]. However, little is known about the diagnostic accuracy of ^{99m}Tc -MIBI planar or SPECT imaging for evaluation of cystic parathyroid adenoma. The predominance of mitochondrial-rich oxyphil

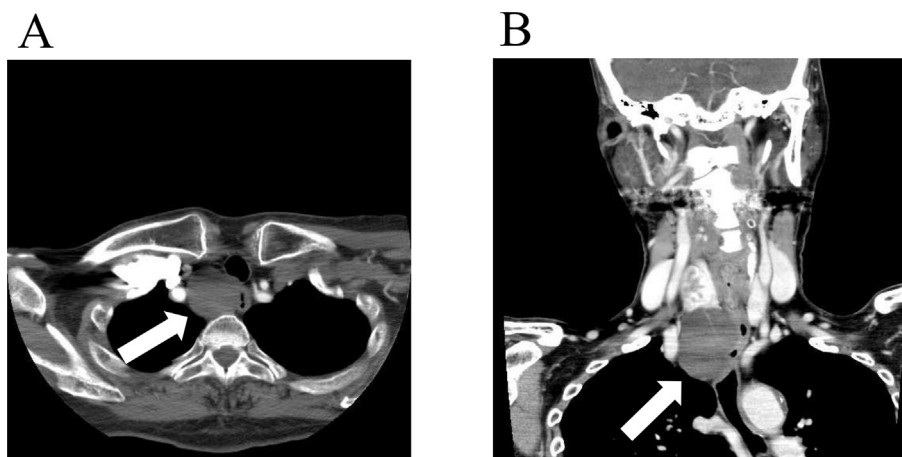


Fig. 1. Enhanced computed tomography scan. (A) Horizontal plane and (B) Coronal plane. Arrow indicates the cystic lesion that extended from the lower end of the right thyroid lobe to the mediastinum.

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