CASE REPORT

Hyperparathyroidism caused by a functional parathyroid cyst

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SUMMARY

A 67-year-old Japanese woman was admitted to our hospital for malaise and loss of appetite. Relevant biochemical examinations showed definite hypercalcaemia and elevated serum levels of intact parathyroid hormone (PTH). We performed thyroid ultrasonography and CT of the neck, which revealed a cystic lesion in the right lower lobe of the thyroid glands. Ultrasound-guided fine-needle aspiration was performed, and PTH level of the cystic fluid was markedly elevated. Technetium-99m-hexakis 2-methoxyisobutyl isonitrile sesta ($^{99m}\text{Tc-MIBI}$) scintigraphy showed intense ring-shaped accumulation of radioactivity in the wall of the cyst. The patient underwent a right lobectomy to resect the cystic parathyroid adenoma. After surgery, her serum calcium and PTH level returned to normal ranges.

BACKGROUND

Primary hyperparathyroidism has become a relatively frequent endocrine disorder, its incidence ranging between 1:3000 and 1:5000. In clinical practice, functional parathyroid cysts are a very rare cause of primary hyperparathyroidism, representing only 1–2% of patients with primary hyperparathyroidism.¹ We report a rare case of a functional cystic parathyroid adenoma as a cause of primary hyperparathyroidism.

CASE PRESENTATION

A 67-year-old Japanese woman was admitted to our hospital for evaluation of malaise and appetite loss. Physical examinations showed no significant findings of hypercalcaemia. She had no history of disease, cigarette smoking or alcohol intake. She has not been taking drugs which influence parathyroid hormone levels, such as thiazide diuretic and lithium. Family history was unremarkable.

INVESTIGATIONS

Laboratory data showed definite hypercalcaemia of 2.92 mmol/L (serum albumin 4.0 g/dL), hypophosphataemia of 0.84 mmol/L, elevated serum levels of intact parathyroid hormone (PTH) of 270.9 pg/mL, 1α -hydroxylase 25-hydroxy vitamin D of 83 pg/mL and elevated markers of bone metabolism, NTx of 162 nmol/BCE/mmolCr, TRACP5b of 1110 mU/dL, respectively. Although renal function was unremarkable, high levels of calcium excretion were found in a 24-hour specimen of urine (U-Ca/day: 12.0 mmol/L). Bone mineral density was also decreased to -3.2SD of T-score. We performed thyroid ultrasonography (figure 1A) and CT of the

neck (figure 1B), which revealed a cystic lesion of $16.8\times 16.2\times 20.1$ mm in size on the right lower lobe of the thyroid gland. Technetium-99m-hexakis 2-methoxyisobutyl isonitrile sesta ($^{99m}\text{Tc-MIBI}$) scintigraphy showed a marginal photopenic lesion in the cystic lesion (figure 1C). In addition, ultrasound-guided fine-needle aspiration was performed, and intact PTH level in the cystic fluid was markedly elevated at 2 970 000 pg/mL (3). The fluid of this parathyroid cyst was haemorrhagic colour, and histopathological examination of the cyst showed a proliferation of the chief cells of parathyroid tissue with infiltration of eosinophilic cells (figure 2). This is the rare case report of cystic parathyroid adenoma. The assay for PTH content in the cyst fluid is very useful to make the diagnosis of cystic parathyroid adenoma.

DIFFERENTIAL DIAGNOSIS

None.

TREATMENT

Her biochemical profiles indicated severe reduction in bone mineral density.

According to consensus guideline for the hyperparathyroidism,² indication for surgery included serum calcium (>2.5 mmol/L upper limit of normal), Creatine clearance (reduced to <60 mL/min) and reduction in bone mineral density (T-score <-2.5). Therefore, we selected surgical intervention; the patient underwent a right lobectomy to resect the cystic parathyroid adenoma.

Unfortunately, an intraoperative intact PTH was not measured in this case.

OUTCOME AND FOLLOW-UP

Postoperatively, her symptoms improved, and her serum calcium and PTH level returned to normal ranges. Histopathological examination of the cystic lesion showed a proliferation of the chief cells of parathyroid gland with infiltration of eosinophilic cells (figure 2). She did well at her 1 year review and her serum calcium and PTH levels remained within normal limits.

DISCUSSION

The first case of parathyroid cyst was reported in 1905. In 1953, Crile Jr and Perryman reported the first case of parathyroid cyst with the use of fine-needle aspiration for the preoperative diagnosis of cystic masses in the neck.³

Functional parathyroid cysts are an extremely rare cause of primary hyperparathyroidism. The criteria for a functional parathyroid cyst include the

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Figure 1 (A) Thyroid ultrasonography showing a cystic parathyroid adenoma. (B) CT scan of the neck showing a cystic parathyroid adenoma. (C) Technetium-99m-hexakis 2-methoxyisobutyl isonitrile sesta showing intense marginal uptake in the right inferior side of the thyroid gland (arrow).

following definitions; preoperative clinical and biochemical evidence of hyperparathyroidism, intraoperative identification of normal remaining parathyroid glands, histological identification of parathyroid tissue within cysts and postoperative correction of hypercalcaemia.⁴ Our case fulfilled with all of these criteria.

In general, parathyroid cysts are divided into two entities; functional and non-functional cysts. Functional cysts result from a cystic degeneration of a pre-existing parathyroid adenoma. Moreover, the fluid of a functional cyst may be not clear but haemorrhagic or dark brown in colour.^{5–6} Interestingly, the presence of haemorrhage in a parathyroid cyst has been associated with hypercalcaemia in previous reports.⁷ In our case, the fluid of the parathyroid cyst was also dark brown in colour.

Neck ultrasonography is a useful diagnostic tool for detecting parathyroid cysts. Ultrasound-guided fine-needle aspiration of cystic fluid with assay for PTH level is an accurate method to confirm parathyroid tissue origin.^{8–9} However, it has been reported significant overlap of the cytomorphological features of thyroid and parathyroid.⁷ In our case, ^{99m}Tc-MIBI scintigraphy showed intense ring-shaped accumulation of radioactivity in the wall of the cyst, suggesting that the cyst wall originated from the large part of parathyroid tissue. Cystic adenomas of the parathyroid glands are often misdiagnosed as thyroid cysts.^{10–11} Therefore, we speculate the combination of neck ultrasonography and scintigraphy (usually ^{99m}Tc-MIBI) is the most efficacious modality for diagnosis and localisation of the cystic parathyroid adenoma as a cause of hyperparathyroidism.

Figure 2 (A) Histological aspect H&E staining. (B) Showing the chief cells of parathyroid gland with infiltration of eosinophilic cells.

Learning points

- ▶ Functional parathyroid cysts are a rare cause of primary hyperparathyroidism, representing only 1–2% of patients with primary hyperparathyroidism.
- ▶ Cystic adenomas of the parathyroid glands are often misdiagnosed as thyroid cysts.
- ▶ The combination of neck ultrasonography and scintigraphy (usually technetium-99m-hexakis 2-methoxyisobutyl isonitrile sesta) is the most efficacious modality for diagnosis and localisation of functional parathyroid cysts in hyperparathyroidism.
- ▶ The assay for parathyroid hormone content in the cyst fluid is very useful to make the diagnosis of cystic parathyroid adenoma.

Contributors KS, AS, and CA took care of this patient and conducted the examination and treatment. KS, AS, and YA acquired and analysis data. KS, and YA wrote the manuscript.

Competing interests None.

Patient consent Obtained.

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