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Clinical value of 11C-methionine positron emission tomography in persistent primary hyperparathyroidism—A case report with a mediastinal parathyroid adenoma

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ABSTRACT

INTRODUCTION: Primary hyperparathyroidism (PHPT) is a common endocrine disorder caused by pathologic growth of one or more of the parathyroid glands. Parathyroidectomies (PTX) in patients with PHPT are procedures with low morbidity, few complications, and a high cure rate. However, the parathyroid glands may be found at various anatomical locations and occasionally they are intrathoracic.
CASE PRESENTATION: We present a 57-year-old patient with PHPT. Before the first and second operation, the preoperative imaging indicated pathologic parathyroid tissue in the neck. Due to postoperative persistent hypercalcemia we performed a 11C-methionine positron emission tomography (11C-MET-PET/CT). The scan showed a focus with increased activity in the mediastinum. Due to persistent disease, an ectopic parathyroid gland in the mediastinum was suspected. At a third operation, the parathyroid adenoma was resected through an anterolateral thoracotomy. Biochemical values normalized and bone mineral density improved postoperatively. Hence, an ectopic localization of a parathyroid gland should be considered during the preoperative planning of a PTX, especially in the re-operative setting. A multidisciplinary effort is necessary to address an intrathoracic adenoma.
CONCLUSION: Ectopic parathyroid glands should be suspected when positive sestaMIBI uptake is seen in the mediastinum and other types of imaging (e.g. contrast enhanced CT scan or PET-CT) may confirm the finding of an ectopic parathyroid adenoma. From the present case and previous studies we found 11C-MET-PET/CT valuable in difficult PHPT cases.

1. Introduction

Primary hyperparathyroidism (PHPT) is a common endocrine disorder caused by pathologic growth of one or more of the parathyroid glands. The pathologic tissue is a single adenoma in approximately 85–90% of the cases whereas 10–15% have multiple adenomas or hyperplasia [1]. PHPT is diagnosed by elevated plasma calcium levels with concomitant elevated or inappropriately high-normal PTH levels, after excluding other causes of hyperparathyroid hypercalcemia.

The only curative treatment for PHPT is parathyroidectomy (PTX), with a cure rate of approximately 93%–98% [1,2]. There are a few different causes of persistent disease. First, it can be impossible to find all the glands in the neck due to an ectopic pathologic gland or a very small missed gland in the neck. Secondly, in the case of 4 hyperplastic glands it may be advisable to keep one of them to avoid hypoparathyroidism. Due to increasing diagnosis of new PHPT patients, a low rate of surgical complications, and overall good surgical results, the number of patients undergoing PTX is increasing.

The work has been reported in line with the SCARE criteria [11].

2. Case presentation

A 57 year old woman was referred to our Department of Endocrinology and Internal Medicine due to hyperparathyroid hypercalcemia. The biochemical abnormalities were accidentally found during hospitalization for a urinary tract infection.

Laboratory findings revealed elevated plasma ionized calcium (Ca^{2+}) of 1.56 mmol/L (reference interval 1.18–1.32 mmol/L) and plasma parathyroid hormone (PTH) of 10.6 pmol/L (1.6–6.9 pmol/L). She had no clinical symptoms of hypercalcemia, but was diagnosed with osteoporosis by dual energy X-ray

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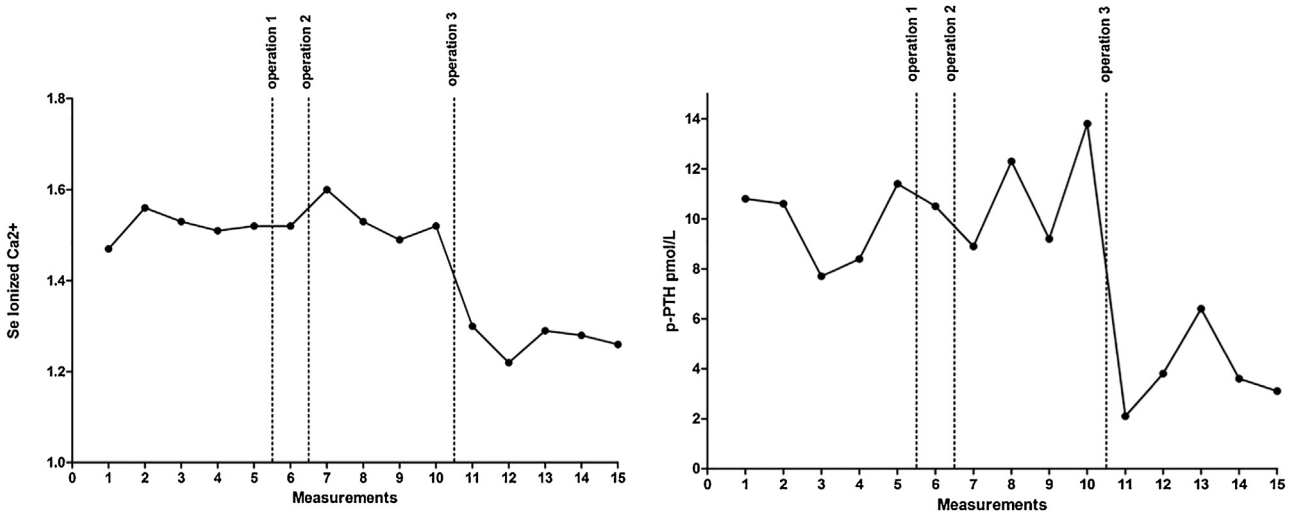

Fig. 1. Measures of ionized Ca^{2+} and PTH at the preoperative work-up (1–5), between the first and second operation (6), between the second and third operation (7–10), and after the third operation (11–15).

absorptiometry (DXA) in the lumbar spine and forearm with T-scores of −2.7 and −2.8, respectively. The T-score was −1.8 at the total hip. There were no kidney stones on a computed tomography scan (CT-scan) and a 24-h urine sample was with normal calcium creatinine clearance ratio (>0.02). In summary, the indication for surgery was based on moderate hypercalcemia and presence of osteoporosis.

Preoperatively, a ^{99m}Tc-methoxy-isobutyl-isonitrile (MIBI) scintigraphy showed possible parathyroid adenoma near the lower left part of the thyroid gland. In addition, there was a focus with increased uptake near the ascending aorta, but this was in first instance interpreted as an inflammatory lymph node. Further, ultrasonography of the neck showed a multinodular goiter and was without visible parathyroid glands.

A bilateral neck exploration was performed, and three parathyroid glands were identified and two and a half parathyroid glands were removed. The fourth parathyroid gland at the inferior position on the right side was not identified. Intraoperative PTH levels decreased insufficiently by only 44% (from 18.6 pmol/L to 10.4 pmol/L) after resection of 2.5 glands. Due to postoperative persistent hypercalcemia we performed a 11C-methionine positron emission tomography (11C-MET-PET/CT). The scan showed a focus with increased activity inside the right thyroid lobe, correspondent to the same area where no parathyroid gland was identified during the first operation. In addition the previous focus with increased activity in the mediastinum was also present on the 11C-MET-PET/CT scan, and interpreted as an inflammatory lymph node.

Consequently, it was decided to perform a right-sided hemithyroidectomy. Again the intraoperative PTH levels did not decrease significantly (Fig. 1) and hypercalcemia persisted with Ca^{2+} levels of 1.50 mmol/L after the second operation. Histopathological examination showed no parathyroid tissue in the specimen removed.

After the second operation, the patient went through a wide endocrine examination to exclude rare causes of hypercalcemia. We performed genetic tests to exclude Multiple Endocrine Neoplasia, mutations in the gene encoding the Calcium-sensing Receptor, and Cell Division Cycle 73 gene. A control DXA scan revealed a sustained low T-score of −3.0 at the lumbar spine and −3.1 at the forearm.

Due to persistent hypercalcemia, an ectopic localized parathyroid gland in the mediastinum was considered. The interdisciplinary team advised to perform a third operation to explore the upper mediastinum. Prior to the operation, a contrast enhanced CT scan was performed, to identify the precise localization of the process (Fig. 2). The gland was located directly behind the ascending aorta, in the aorta-pulmonary window. An anterolateral thoracotomy was performed and a parathyroid adenoma was resected.

Postoperatively PTH decreased to 1.4 pmol/L (Fig. 1) and ionized calcium normalized at 1.30 mmol/L. There were no postoperative complications, especially no palsy of the recurrent laryngeal nerve. One year after operation the T-score increased by 7.8% in the lumbar spine to −2.5, whereas T-scores in the hip and forearm were unchanged.

3. Discussion

Hyper functioning adenomas are found in an ectopic location in 6% to 16% [3]. These ectopic glands can be found in numerous places, including high cervical positions, the carotid bifurcation, within the thyroid tissue, para-aortic, retro- and para-esophageal and within the thymus [3]. Due to these frequent and various locations there are many ways to identify the precise location of the pathologic gland in the preoperative phase. Further, the preoperative scans are used to evaluate whether the patient is likely to have single- or multigland disease. In the case of negative imaging, the indication for surgery is rarely affected since the operative indication is based on age, symptoms, severity of disease, and presence of organ manifestations to PHPT [4,5]. Hence, the experience of the parathyroid surgeon is crucial when there is no evidence of the number of affected glands or where to find the pathologic gland(s). In the case of negative preoperative imaging, there is a higher risk of persistent postoperative PHPT and the use of intraoperative PTH analysis is crucial [6]. Further, if there is hyperplasia in all four glands, there is a risk of both persistent PHPT after removal of 3 glands but also a higher risk of postoperative hypoparathyroidism.

Within recent years, improvements in the field of imaging have allowed a more targeted surgical approach. Commonly used imaging techniques are ultrasonography (US) and MIBI scintigraphy, usually performed as SPECT with low-dose CT. US is non-invasive, inexpensive and a relatively simple method of imaging. MIBI scintigraphy can be formed using different protocols, usually in goiter rich regions, as ours, the subtraction protocol with petechnetate or I-123 for thyroid imaging is performed together with the sestamibi scan. There is also uptake of sestamibi in thyroid nodules and lymph nodes, which can lead to false-positive scans [4]. There are various ways of performing the scintigraphy. Different