CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 40 (2017) 120–123

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Management of mediastinal parathyroid adenoma via minimally invasive thoracoscopic surgery: Case report

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ARTICLE INFO

Article history:
Received 22 May 2017
Received in revised form 27 August 2017
Accepted 28 August 2017
Available online 8 September 2017

Keywords:
Case report
VATS
Ectopic parathyroid adenoma
Idiopathic hypercalcemia

ABSTRACT

INTRODUCTION: The most common cause of chronic hypercalcemia is primary hyperparathyroidism (PHPT). However, owing to the diverse presentation of hypercalcemia, the diagnosis often goes unnoticed culminating as a continuum of recurrence of symptoms. Nephrolithiasis, decreased bone mineral density and peptic ulcer disease are the main clinical sequelae. Among the causes of PHPT 80% are caused by parathyroid adenomas (PA). However, only rarely, these adenomas are found ectopically.
PRESENTATION OF CASE: We present the case of a 66-year-old female with a history of recurrent renal stones and peptic ulcer disease. She was found to have elevated serum calcium and PTH levels. However, subsequent high resolution CT scan of chest and neck failed to demonstrate any abnormality. Therefore, an anterior planar Technetium-99m-sestamibi (MIBI) scintigraphy scan using a single-tracer was done and it identified ectopic anterior mediastinal parathyroid adenoma. The patient was successfully managed with video-assisted thoracoscopic surgery and excision of the mass with follow up calcium level monitoring.
DISSCUSSION: An elevated calcium level should prompt a thorough workup, as sometimes it's the only clue to the unrelated and diversified systemic manifestations of hypercalcemia. Hyperparathyroidism due to ectopic adenoma is quite rare and possess a diagnostic and management challenge.
CONCLUSION: Symptomatic hypercalcemia and high level of PTH without local PA should alert physicians to search for ectopic locations through imaging. VATS is a safe and effective minimally invasive procedure for the resection of ectopic mediastinal PA and it should be considered as the first line approach for resection of these ectopic tumors.

1. Introduction

The following case report has been reported from Aga Khan University Hospital which is an internationally recognized teaching hospital and a tertiary care centre based in Pakistan, in accordance with the SCARE guidelines for case reports [1]. Parathyroid adenomas are the most common cause of PTHP [2]. However, the prevalence of ectopic PA is about 2%–43% in anatomical series and reaches upto 16% and 14% in primary and secondary hyperparathyroidism, respectively [3] Among the various ectopic locations, mediastinal ectopic PAs are extremely rare tumors, constituting 1–2% of all PAs [4]. Due to the rare possibility of ectopic PA, there is a diagnostic and management improbability, which may delay the diagnosis and subsequent surgery due to difficulties in locating the ectopic PAs. Therefore, it is essential for physicians to keep a high index of suspicion while coming across symptomatic malignant hypercalcemia with raised PTH levels. An accurate pre-operative localization is the key to successful surgery for an ectopic PA. The two most commonly employed imaging modalities to locate PAs include ultrasonography (US) and 99mTc-MIBI scintigraphy scan having a combined sensitivity of 88%–95% [3]. CT and MRI are alternative options with sensitivities of 65% and 78% respectively. The resection of mediastinal ectopic PAs has been traditionally done via median sternotomy and thoracotomy. However, Video assisted thoracoscopic surgery (VATS) has proved to be a safe and successful approach for management of mediastinal ectopic PA in our experience.

2. Case report

A 66-year old female housewife (BMI: 24.1 kg/m^2), from a remote city of Pakistan, presented to the surgical OPD with the presenting complaint of intermittent flank pain for the past two years with a history of recurrent renal stones. She had undergone a percutaneous nephrolithotmy (PCNL) 14 years back for nephrolithiasis secondary to calcium oxalate stones. She later had a right extra-

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http://dx.doi.org/10.1016/j.ijscr.2017.08.056

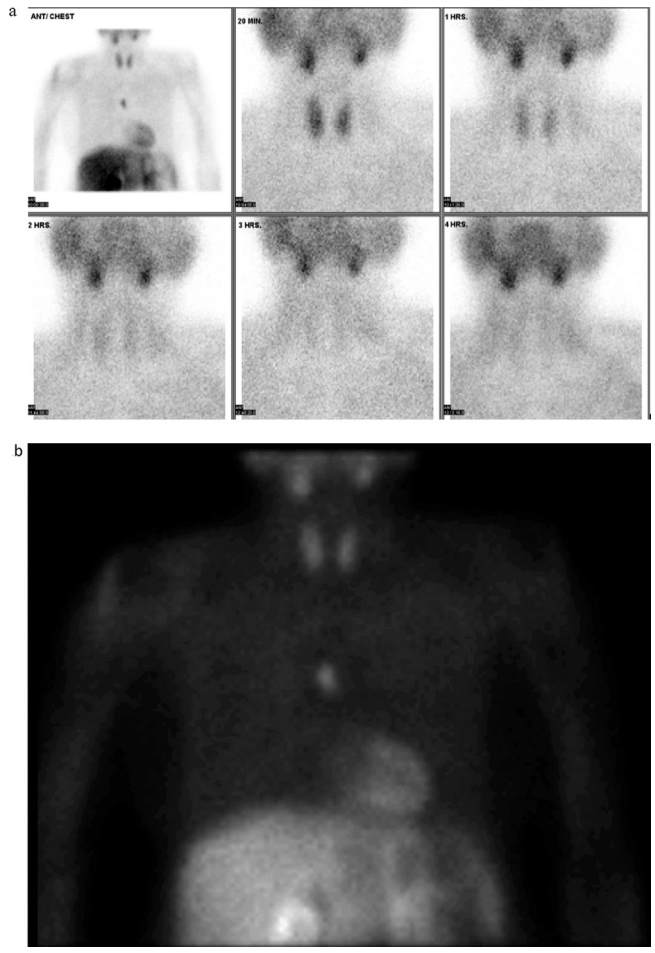

Fig. 1. a. 99mTc-MIBI scintigraphy scan demonstrating PTH adenomas. b. 99mTc-MIBI scintigraphy scan deomonstrating ectopic PTH adenomas in anterior mediastinum.

corporeal shock wave lithotripsy (ESWL) for a residual stones 2 years ago. CT scan of the kidneys showed non-obstructing bilateral renal calculi along with cholelithiasis in the gall bladder. Patient also had multiple gastroenterology clinic visits with the complaints of postprandial bloating and recurrent oral ulcers. Bone densitometry showed marked osteopenia. Her serum calcium level was found elevated at 12.6 mg/dl. The constellation of her symptoms and the elevated calcium levels warranted further workup. Serum PTH was 403 pg/ml (16–87) and serum phosphorus was 2.2 mg/dl (2.5–4.5) respectively; she was diagnosed with primary hyperparathyroidism, aka, PTH dependent hyperparathyroidism causing malignant hypercalcemia. In order, to rule out the possibility of parathyroid adenoma, a high resolution CT scan of chest and neck where carried out, but, there was no abnormal enlargement of the parathyroid gland present, however, 99mTc-MIBI scintigraphy scan was done that showed a well defined area of abnormal tracer deposition in anterior mediastinum just below the manubriosternal junction confirming the diagnosis of mediastinal ectopic PA, furthermore, parathyroid adenomas were outlined over lower pole of right thyroid in anterior mediastinum as well. (Fig. 1a and b).

Pre-operatively, injection pamidronate was administered three week prior to surgery. Repeat chemistry showed a serum calcium level of 10.2 mg/dl. She, subsequently, underwent VATS where right lower parathyroid tissue was identified and excised. The histopathology report established parathyroid parenchyma composed of chief and oxyphil cells with intervening fibroadipose tissue. There was no evidence of malignancy. The ectopic PA was excised as a whole from anterior mediastinum just behind the sternum and right inferior parathyroidectomy was also carried out. Postoperatively, serum calcium level was repeated every 12 hourly and calcium with vitamin D supplements were administered. Follow up chemistry panel showed a serum calcium in decreasing trend from 9.2–8.2 mg/dl. Her PTH levels also remarkably dropped to 23 pg/dl. She had a complete recovery. Upon return to clinic her serum calcium level and PTH level were found within the normal ranges; 8.9 mg/dl and 86.90 pg/dl respectively. Post operative MR imaging also showed no residual tumor. Patient remained asymptomatic at 6 month follow up as well.

3. Discussion

Symptomatic hypercalcemia can present with several different clinical manifestations and possess a difficulty in diagnosis. Elevated calcium levels should always alert the physician and it should be followed by a thorough endocrinology workup, especially PTH levels. Our patient had complaints involving different systems including GI, renal and musculoskeletal. However, the only clue to the endocrine issue was the elevated serum calcium level, which unfortunately remained unnoticed for a long time, leading to the development of symptoms involving multiple systems. The link between malignant hypercalcemia and PHPT is now completely established as per the recent literature available [22].

The incidence of PHPT is approximately 25 per 100,000 in the general population that increases with age [5]. PHPT is classified as being caused by a single adenoma (75%–85%), hyperplasia (10%–20%), multiple adenomas (4%–5%) or carcinoma (1%) giving the histopathological characteristics and the number of involved glands [5]. The superior and inferior parathyroid glands are derivatives of the third and fourth pharyngeal pouches, respectively. During embryogenesis, these glands descend along with the thyroid gland, sharing the same route, while detaching from the pharyngeal apparatus. However, if they fail to descend, they are found high in the neck under the mandible as undescended parathyroids. On the other hand, if they descend too far, they are found within the mediastinum as ectopic parathyroids [4]. The base of the tongue upto the mediastinum can be regarded as potential positions for ectopic PAs [6] Ectopic parathyroid gland adenoma is a rare tumor, particularly causing PHPT. The clinical and laboratory features of patients with primary hyperparathyroidism due to ectopic adenomas have been reported to be more severe than those with localized parathyroid adenomas, since the former remained undetected and may manifest with higher calcium levels and more frequent primary hyperparathyroidism related bone disease more frequently [7].

The localization and subsequent treatment of an ectopic PA can usually be challenging. There are various imaging modalities available for the diagnosis include ultrasonography, CT scan, magnetic resonance imaging (MRI), positron emission tomography (PET), single photon emission tomography (SPECT) and Technetium (Tc99m-Sestamibi) scintigraphy scan. Among these, U/S is the most widely utilized modality due to its low cost and easy availability. High-resolution ultrasound can accurately localize adenomas relative to the thyroid gland. However, its ability to detect abnormalities depends on the experience and skill of the operator, and therefore, its sensitivity in localization of enlarged parathyroid glands varies greatly (44%–87%) [3]. Recently, scintigraphy scan is increasingly being utilized for the localization of ectopic PA with reported sensitivity of up to 90% [8]. High-resolution ultrasound and parathyroid scintigraphy both have their own advantages. The former is good at localizing adenomas relative to the thyroid gland, whereas the latter is usually reserved for detecting adenomas in