Case Report

Single photon emission computed tomography-CT in ectopic parathyroid adenoma

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ABSTRACT

Primary hyperparathyroidism often presents with protean manifestations, resulting in delayed diagnosis. At times, aberrant development and migration of the gland leads to ectopic location leading to problems in localization. Judicious use of combination methods of localization is recommended in treatment failure or recurrent disease. We report the use of single photon emission computed tomography-CT in precise localization of parathyroid adenoma in a patient with failed initial surgery.

Key words: Ectopic parathyroid, hyperparathyroidism, emission computed tomography-CT

INTRODUCTION

Primary hyperparathyroidism is commonly due to an adenoma of the parathyroid glands. The adenoma is solitary in 85–90% of patients, while others have multiple adenomas or parathyroid hyperplasia.[1] Accurate preoperative localization is essential for good surgical outcome, and inability to locate the adenoma in an ectopic gland may delay the diagnosis.[2] Nuclear imaging accurately localizes the tumor in more than 90% of cases, obviating the need for advanced imaging modalities.[3] Rarely, patients present with localization failure posing a great challenge to the treating endocrinologist and operating surgeon. We report the use of a novel imaging method leading to successful outcome in a patient of primary hyperparathyroidism with failed first surgery.

CASE REPORT

A 54-year-old lady presented with body pains and muscle aches for 1-year duration to a peripheral hospital. Investigations revealed high serum calcium (11.6 mg/dL), low phosphorus (2.6 mg/dL), elevated alkaline phosphatase (677 U/L) and intact parathyroid hormone (iPTH) of 116 pg/mL (Normal 10-65 pg/mL). Sestamibi scan revealed right inferior parathyroid adenoma, and she was diagnosed as a case of primary hyperparathyroidism. She underwent adenomectomy along with thyroidectomy and showed no features of hungry bone syndrome postoperatively. There was no confirmation of parathyroid adenomectomy by using intraoperative PTH levels or by frozen section of the removed tissue.

Her clinical symptoms persisted after surgery and histopathological examination of the specimen removed showed thyroid tissue with no evidence of parathyroid adenoma. She reported to us after 6 months of initial surgery with persisting complaints of body aches and myalgia. Her clinical examination was unremarkable, with a normotensive blood pressure, and well-healed scar in the neck. Serum biochemistry revealed elevated calcium (10.8 mg/dL), low phosphorus (2.8 mg/dL) and elevated alkaline phosphatase (280 IU/L). Bone mineral density

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estimation revealed a T-score of -2.2 at hip joint and Z-score of -2.3. Serum 25 hydroxy vitamin D level was 22 ng/L and parathyroid hormone was elevated (iPTH-140 pg/mL). Skeletal survey revealed lytic lesions in the metacarpal bones. Localization with sestamibi scan revealed right inferior parathyroid adenoma with no tracer uptake in thyroid bed [Figure 1]. Abdominal sonography showed normal renal parenchyma and ultrasonography neck and plain CT neck did not show parathyroid adenoma.

In view of past history of failed surgery, Tc 99m Sestamibi Single Photon Emission Computed Tomography CT (SPECT) was done for precise localization of the adenoma prior to re-exploration. It revealed an ectopic parathyroid adenoma, located suprasternally in the pretracheal region on right side [Figure 2]. Repeat surgery was uneventful and the postoperative course was complicated by hypocalcemia. Histopathological examination of the specimen confirmed the parathyroid adenoma. During last follow-up, 1 year after second surgery the patient is free of all symptoms and had normal serum calcium, phosphorus and alkaline phosphatase values.

**DISCUSSION**

Primary hyperparathyroidism is a common endocrine disorder often missed due to varied presentation. The disease is detected during asymptomatic stage in developed countries, while we encounter the advanced spectrum of the disease with severe metabolic bone disease.[4] Precise localization is important to prevent further delay in definitive therapy after biochemical confirmation of the diagnosis.

Parathyroid glands are derived from pharyngeal pouches (superior parathyroid glands from 4th and inferior from 3rd pouch) with subsequent caudal migration. This explains their tendency for ectopic localization.[5]

The modalities available for precise localization of a parathyroid adenoma are palpation, ultrasonography (USG), CT, MRI, nuclear scintigraphy, and combination of these tests. Palpable neck mass anteriorly is commonly seen in parathyroid tumor but not in adenoma. Ultrasonography is useful for its wide availability, convenience, cost and a guiding tool for the surgeon before surgery. However, the sensitivity and specificity of USG reported was 73% and 100%, respectively.[6] CT and MRI scans provide excellent spatial resolution but often miss a small parathyroid adenoma.[7]

Parathyroid scintigraphy remains an important tool for precise localization prior to surgery.[8] This is recommended mostly in cases of failed surgery, recurrent disease and when planned for a limited surgical exploration. Immediate imaging reveals the tracer uptake in both thyroid and parathyroid gland along with adenoma but the adenomatous tissue shows retention of the tracer in delayed images. SPECT scan is an advance in radionuclide studies with a three-dimensional (3-D) reconstruction, further increasing the sensitivity for adenoma localization. SPECT scan, with its 3-D capability, combined with CT images, is very helpful in directing the surgeon particularly in recurrent or residual hyperparathyroidism. Recent reports suggest that SPECT/CT is superior to SPECT scan alone for localization of parathyroid adenoma with nodular goiter, distorted neck anatomy and those with ectopic parathyroid glands.[9,10]

**Conclusions**

To conclude, our patient had an ectopic parathyroid adenoma resulting in failed initial surgery. Use of a novel
imaging modality like SPECT helped in accurate localization of the adenoma prior to repeat surgical exploration.

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