

## ■ CASE REPORTS

# Use of a Technetium99m-Sestamibi Scan to Detect Ipsilateral Double Adenoma in a Patient with Primary Hyperparathyroidism: A Case Report

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### ABSTRACT

**Introduction:** Patients with primary hyperparathyroidism generally have a single parathyroid adenoma that causes excessive excretion of parathyroid hormone. For about 2% to 15% of these patients, a double adenoma is present that involves one lesion on each side of the neck.

**Case Presentation:** We describe a case of double parathyroid adenoma causing asymptomatic hypercalcemia. A presurgical technetium99m (Tc99m) sestamibi scan suggested an ipsilateral double adenoma in the left thyroid lobe. An intraoperative parathyroid hormone assay confirmed its successful removal.

**Discussion:** Although double adenomas are not yet widely acknowledged, presurgical imaging and nuclear scans can help to localize multiple lesions, and intraoperative parathyroid hormone assays can confirm the diagnosis and cure.

### INTRODUCTION

Primary hyperparathyroidism (PHPT) is the most common cause of hypercalcemia in the US.<sup>1</sup> Most cases can be explained by a single parathyroid adenoma, defined as a solitary neoplastic lesion bordered by normal parathyroid tissue, which is followed by 4-gland hyperplasia and, rarely, parathyroid carcinoma.<sup>2</sup> Small subsets (2%-15%) of PHPT contain 2 solitary adenomas as seen during explorative parathyroidectomy, however. The double adenoma (DA) remains controversial among investigators, some of whom deny its existence. This report looks at a patient with asymptomatic hypercalcemia that was caused by an elusive DA. The DA was discovered with use of a technetium99m (Tc99m) sestamibi scan and successfully removed as seen by a decline in intraoperative parathyroid hormone (IOPTH) levels.

### CASE PRESENTATION

A 63-year-old white man was referred to the surgical team for asymptomatic hypercalcemia of 12.9 mg/dL (reference range [rr], 8.4-10.2 mg/dL). His primary care physician first recognized his elevated calcium level 2 years before this referral. During previous visits to his primary care physician, the patient did not have signs and symptoms of hypercalcemia, and, aside from his hypertension, was otherwise healthy. Presurgical ultrasonography imaging studies suggested a normal-appearing thyroid gland with 2 hypochoic well-defined masses seen posterior to the left lobe, the larger

of which measured 2.5 cm. The patient underwent a standard Tc99m-sestamibi scan that revealed heterogenous delayed persistent tracer localization in the left upper and lower parathyroid regions, which could indicate a multifocal parathyroid adenoma (Figure 1). Of note, the patient's presurgical serum calcium and intact parathyroid hormone (iPTH) levels were 13.3 mg/dL and 354 pg/mL (rr, 8.4-10.2 mg/dL and 15-65 pg/mL), respectively.

During an elective parathyroidectomy, the thyroid gland was found to be grossly normal. An inferior adenoma (0.85 g) in the left lobe was removed for frozen section analysis.

A left-sided superior parathyroid adenoma (2.5 g) was also discovered and sent for confirmation. Frozen sections of both specimens confirmed hypercellular parathyroid tissues. Electrochemiluminescence immunoassay was used to measure IOPTH levels. A drop in iPTH from 397 pg/mL to 169 pg/mL was seen 13 minutes after the second adenoma was removed. A subsequent drop to 118 pg/mL occurred 5 minutes later, representing a 70% decline and appropriate treatment of the disease. The patient was discharged to home the next day. No further evidence of hypercalcemia was noted at 3-month follow-up, and his serum calcium levels remained stable at 9.6 mg/dL. Long-term follow-up included a measured iPTH of 42 pg/mL approximately 1 year later, which remained within defined limits.

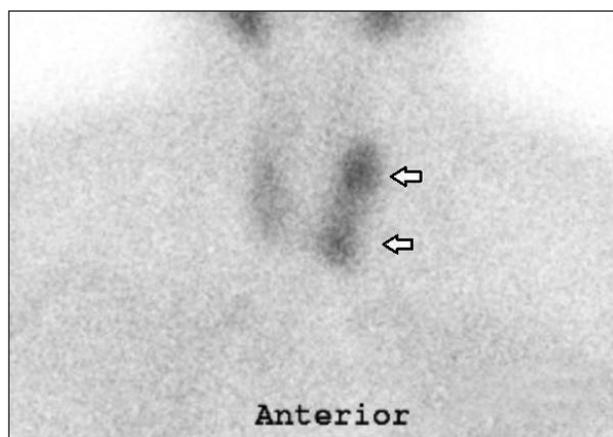


Figure 1. An early 26mCi Tc99m-sestamibi scan showing a left-sided multifocal parathyroid adenoma (arrows).

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## DISCUSSION

PHPT is the most common cause of hypercalcemia.<sup>1</sup> This is especially relevant when a patient's elevated calcium level is an incidental finding. With the addition of serum calcium level on standard chemistry panels in 1974, there was an increase in documented US PHPT incidence; approximately 22 cases per 100,000 people per year were identified between 1993 and 2001.<sup>1</sup>

For 87% to 91% of patients with PHPT, a single adenoma is the culprit.<sup>2</sup> The remaining cases are caused by 4-gland hyperplasia (10% to 15%) and the controversial DA.<sup>1,2</sup> Some researchers continue to doubt the existence of DAs and regard them as uneven multiglandular hyperplasia.<sup>2</sup> However, studies confirm the presence of DAs in 2% to 15% of patients undergoing parathyroid explorations; often, one is located on each side of the neck.<sup>2,3</sup> A 2009 retrospective review<sup>2</sup> described the occurrence of DAs in 47 of 552 patients (8.5%), whereas a 2002 article<sup>4</sup> described 44 of 401 patients (11%) with DAs who were undergoing consecutive conventional parathyroid explorations.

Of note, the standard surgical approach is to investigate the neck, remove abnormal parathyroid gland tissue, and send the sample for intraoperative frozen section.<sup>5</sup> This method, however, fails to detect all abnormal tissue in some cases of multiglandular disease, with discrepancies between the frozen section and the definitive histology in up to 10% of cases.<sup>3,5</sup> As a result, the use of minimally invasive parathyroidectomy is increasing. Presurgical localization of adenomas with ultrasound imaging and Tc99m-sestamibi scans is used with consideration of IOPTH level to confirm removal of these lesions.<sup>3</sup> A decrease of at least 50% in perioperative iPTH suggests likely biochemical cure.<sup>2</sup> In a 2010 clinical study,<sup>3</sup> use of IOPTH level helped physicians identify a DA that was missed by presurgical scans; IOPTH level was associated with 100% sensitivity in detection compared with ultrasound or nuclear imaging alone (15% of DAs are missed).<sup>3</sup> Case reports appear to confirm that IOPTH level can serve as an accurate predictor of cure in combination with presurgical scanning to help physicians prevent unsuccessful outcomes, surgical reexploration, and unnecessary patient costs.<sup>2,3</sup> Despite these advantages, the use of IOPTH level is not yet widely accepted in the management of PHPT.<sup>3</sup> Concerns include increases in minimally invasive parathyroidectomy surgery time, cost, and reports of false-negative rates in other studies. Nevertheless, IOPTH level assessment should remain part of the arsenal in PHPT treatment.<sup>3,6</sup>

## CONCLUSION

This report describes a case of PHPT that was caused by a DA and identified with a presurgical Tc99m-sestamibi scan. The result was a 70% decline in IOPTH levels after excision. Although DAs are not universally recognized, physicians should be cognizant about the possibility that DAs can (rarely) cause PHPT. Presurgical testing that includes a Tc99m-sestamibi scan and IOPTH level is necessary to maximize successful outcomes. ♦

## Disclosure Statement

*The author(s) have no conflicts of interest to disclose.*

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## How to Cite this Article

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