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Moans, Bones, Groans, and a Thyroid Mass

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Parathyroid carcinoma accounts for less than 1 per cent of cases of primary hyperparathyroidism.^{1, 2} Its synchronous occurrence with thyroid carcinoma is rarer still, with only seven such patients previously reported in the literature.^{3, 4} We report another case, unique not only because the parathyroid carcinoma was found within the left thyroid lobe but also because of the presence of a synchronous papillary thyroid carcinoma within the contralateral thyroid lobe. The case illustrates the need for surgeons to be aware of the possibility of concomitant thyroid pathology and the potential for the synchronous occurrence of other endocrine neoplasms.

Our case is a 56-year-old female who presented with hypercalcemia, complaints of pain in her left shoulder and hip and a history of recurrent kidney stones over a period of several months. One year prior to the present admission she had undergone an extensive workup for an intrathyroidal lesion located within the central aspect of the left thyroid lobe and for bilateral adrenal incidentalomas measuring 4.3 cm on the right and 1.7 cm on the left. Fine-needle aspiration biopsy of the thyroid nodule revealed parathyroid tissue. Biopsy of the larger of the two adrenal lesions was negative for malignancy. At that time she had bilateral ureteral stents placed for obstructive uropathy. She was discharged with instructions to follow up after recovery from her ureteral stenting to undergo resection of her presumed intrathyroidal parathyroid adenoma.

She subsequently was readmitted because of worsening bone pain. On her subsequent presentation, physical examination her head, neck, and cervical nodal basins were normal. Laboratory evaluations revealed hypercalcemia with a serum level of 12.2 mg/dL and a parathyroid hormone (PTH) level of 1246 pg/mL. Radiological imaging confirmed pyelocaliectasis from an obstructed stent in her left ureter and areas of osteitis fibrosa cystica in her left iliac crest and left humeral head. She was treated with intravenous saline infusion, diuresis, and cinacalcet (Sensipar™, Amgen™, Thousand Oaks, CA) to acutely control her hypercalcemia. A percutaneous nephrostomy tube was placed within the left renal pelvis and replacement of both ureteral stents were performed to relieve her recurrent obstructive uropathy.

Ultrasound demonstrated two lesions in the thyroid: a solid 5.6 × 3 × 3.4 cm mass in the left lobe and a 1.4-cm hypoechoic lesion in the central aspect of the right lobe. A Single-photon

emission computed tomography (SPECT-CT) study demonstrated the presence of an intrathyroidal mass within the left lobe with imaging characteristics consistent with parathyroid tissue (Fig. 1A and B). A 24-hour urine for metanephrines and vanillylmandelic acid were within normal limits.

She underwent total thyroidectomy and biopsy of the remaining three parathyroid glands. The right thyroid lobe contained a firm mass surrounded by a sclerotic capsule and frozen section confirmed the mass in the right lobe to be a follicular variant of papillary thyroid carcinoma. Pathology also confirmed intrathyroidal parathyroid carcinoma in the left lobe. A bilateral central compartment neck dissection was then performed. Final pathological examination confirmed the presence of both carcinomas, and demonstrated nodal metastases of papillary carcinoma to the right paratracheal lymph nodes. Intraoperative PTH determinations were obtained and demonstrated a drop from a preexcision level of 3300 pg/mL to a steady state value of 259 pg/mL at 60 minutes.

The patient was scheduled to receive radioactive I^{131} ablation and was given levothyroxine at a dose sufficient to achieve thyroid-stimulating hormone suppression due to her nodal metastases. She underwent periodic measurements of serum calcium and PTH for follow-up of her parathyroid carcinoma. In addition, laparoscopic resection of her 4.3-cm nonfunctioning adrenal incidentaloma is planned, as is close follow-up of the contralateral smaller adrenal lesion to ensure benign biologic behavior.

Parathyroid carcinoma, itself a rare tumor may be very rarely accompanied by synchronous thyroid cancer. More commonly parathyroid carcinoma may coexist with another parathyroid disease process, such as a parathyroid adenoma, hyperplasia, or a second parathyroid carcinoma.¹⁻⁴ Our case of synchronous thyroid and parathyroid carcinomas is only the eighth such case reported³⁻⁴ (Table 1), and the first to coexist with bilateral adrenal incidentalomas. The spectrum of endocrine neoplasms encountered in this patient suggests the possibility that she has a genetic predilection for the development of endocrine neoplasia, albeit not the classically described syndromes of multiple endocrine neoplasia 1 or 2.

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Fig. 1. SPECT-CT Scan, (A) Nuclear parathyroid imaging. (B) Tracer accumulation in left thyroid bed.

Table 1

Previous Reported Cases of Synchronous Thyroid and Parathyroid Carcinoma

References	Sex	Age	Calcium (mg/dL)	PTH (pg/ml)	Parathyroid Size (cm)	Thyroid Cancer	Associated Parathyroid Disease	Parathyroid Carcinoma Metastases at Time of Operation	Thyroid Carcinoma Metastases at Time of Operation	Surgery	Outcome
Kurita et al. 1979	F	68	12.2	63,000	4.2 × 3.2 × 2.4	Left lower	Papillary	None	None	En bloc resection	Postop normocalcemia
Christmas et al. 1988	F	62	Increase	Unknown	Unknown	Follicular-posttradation	None	None	None	Unknown	Died from metastatic parathyroid carcinoma
Savli et al. 2001	F	47	Normal	N.D.	Unkn left	Papillary	Hyperplasia	None	None	Total thyroidectomy with parathyroidectomy ×2	Normocalcemia (one year)
Schoretsanitis et al. 2002	F	55	14.2	>1,000	3 × 3	Left lower	Papillary (follicular variant)	Small nests of carcinoma cells were present in the surrounding thyroid tissue.	None	Near total thyroidectomy with en bloc resection of the parathyroid tumor	Normocalcemia (two years)
Lin et al. 2005	M	38	16.5	351	4×3×3	Left lower	Papillary	Two enlarged parathyroid glands on contralateral side (?)	None	Total thyroidectomy left parathyroidectomy	Normocalcemia (six years)
Goldfarb et al. 2010	M	58	14.4	2,023	3.4 × 3.3 × 2.2	Left lower	Papillary	Contralateral parathyroid adenoma Right parathyroid microcarcinoma	None	En bloc resection	Persistent hypercalcemia after resection of parathyroid carcinoma. Normocalcemia after excision of CL parathyroid adenoma (one year)
Chaychi et al. 2010	F	79	10.4	89	5	Left middle and superior	Multifocal papillary	None	None	Total thyroidectomy and left-sided parathyroidectomy	Normocalcemic six months postoperatively (calcium 8.8 mg/dL)
Present case	F	56	12.2	>3,300	4.6 × 4.5 × 2.5	Left lower	Papillary (follicular variant)	Abnormal parathyroid tissue on CL side	None	Total thyroidectomy left parathyroidectomy right trachea-esophageal groove lymph node dissection	

Mets, Metastasis; CL, Contralateral; LN, lymph node.