**Introduction**

A 68-year-old man presented with persistent hypercalcemia (12.8 mg/dL) and hyperparathyroidism (PTH 1087 pg/ml) after four outside attempts at parathyroidectomy (in 2006, 2013, 2015). He had initially presented with a pathologic fracture from a brown tumor in his hand. Of not, he also had a history of radiation therapy for acne as a child.

The records indicated that the original right superior adenoma removed in 2006 was 6.6 grams. In 2013 he had a left inferior parathyroid adenoma removed after recurrence of his hyperparathyroidism. His 3rd (2013) and 4th (2015) operations reported removal of parathyroid adenomas from a right superior location in an attempt to treat his recurrent, persistent hyperparathyroidism. In the pathology report from 2013, there was specimen submitted as “right neck soft tissue” that was found to be fragmented parathyroid adenoma on microscopic assessment. In 2015 the pathologic assessment demonstrated some possible muscular invasion, queried as an implant at the time of surgery.

**Imaging**

A pre-operative MIBI/SPECT-CT scan demonstrated nodular hypodense tissue with radiotracer uptake in the right thyroid bed. Surgeon performed pre-operative ultrasound demonstrated numerous heterogenous nodules within the right thyroid bed. There were also numerous hypodense nodules superficial to the strap musculature.

**Operation**

In June 2016, the patient underwent removal of the multifocal disease with frozen section confirmation of parathyroid tissue in multiple locations including superficial to the strap muscles, within the right central neck contents, and deep to the recurrent laryngeal nerve at the cricothyroid joint. The disease burden extended into the right cricothyroid muscle (requiring resection of the muscle) and was immediately adjacent to the esophagus posterior to the trachea. Rapid intra-operative PTH assessment confirmed a drop in PTH to 46 pg/mL (pre-operative value of 817 pg/mL) 10 minutes after completion of the right central neck dissection.

The integrity of the recurrent laryngeal nerve was intact at conclusion of the case and verified by stimulation of the vagus nerve within the carotid sheath. The patient had normal vocal fold function demonstrated on post-op laryngoscopy at one week. The patient developed hungry bone syndrome postoperatively with symptomatic hypocalcemia (calcium was 8.4) treated with oral supplementation post-operatively.

**Surgical Pathology**

The pathologic assessment revealed atypical multinodular parathyroid proliferation within the right central neck and in the soft tissue superficial to the strap muscles.

This institution’s pathology department reviewed the patient’s case and concluded that it was favored to represent an atypical benign multifocal proliferation of hypercellular parathyroid. It was agreed that the atypical appearance could be consistent with malignancy, however, this was considered less likely given the history of multiple previous parathyroid surgeries, a calcium level <15 mg/dL and an absence of mitotic figures or a single large palpable mass.

**Conclusions**

We present a highly unusual cause of recidivistic hyperparathyroidism. It is difficult to determine the underlying cause of the patient’s disease process, which pathologically is consistent with a benign process, but demonstrated uncharacteristic clinical aggression.

Malignancy of the parathyroid gland is a very rare cause of hyperparathyroidism, accounting for <1% of hyperparathyroidism. The 10 year overall survival rate is relatively high, reported as 67.8%, and most of the morbidity and mortality is attributed to the metabolic complications. The case described here lacked the aggressive pathologic characteristics of parathyroid carcinoma, though the indolent, recurrent nature of this patient’s disease could be consistent with malignancy.

There are a few reports in the literature of what previous authors coined “parathyromatosis”, the seeding of parathyroid tissue throughout the neck after intra-operative rupture, resulting in multifocal recurrence. The abnormal location of parathyroid tissue (superficial to strap muscle), and multifocal nature of the disease, make this the most likely diagnosis. Additionally, the pathology report from the patient’s previous surgery reported that fragments of parathyroid tissue were submitted, suggesting possible intra-operative rupture.

The patient was discussed at this institution’s multidisciplinary tumor board and recommended for post-operative radiation to the right neck, given the recurrent and multifocal nature of the disease. He completed this to 66 Gy. His most recent PTH and calcium were 81.5 pg/mL and 10.1 mg/dL, respectively, at follow up in November 2016, three months after completion of radiation therapy.

**References**


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