Parathyromatosis Following Endoscopic Parathyroid Surgery: A Rare Occurrence

Sir,
Parathyromatosis, a cause for recurrent hyperparathyroidism is common in middle-aged women and chronic kidney disease patients. It involves multiple nodules of benign hyperfunctioning parathyroid tissue scattered throughout the neck and mediastinum. Primary or Type 1 parathyromatosis is the result of hyperplasia of parathyroid rests from embryologic development. Secondary or Type 2 parathyromatosis, a rare complication of parathyroidectomy, first described in 1975 by Palmer et al., is more common which arises due to seeding of parathyroid tissue during surgery. It has been also considered as a low-grade malignancy. Thirty-five cases have been reported so far. Its preoperative diagnosis is rare due to the lack of awareness of this entity. Sonographic imaging provides clues to the diagnosis. Medical and surgical interventions carry high failure rates. Cinacalcet and bisphosphonates are main-stays of medical therapy. Alcohol ablation and several novel calcimimetics have been used in these patients. Repeated neck explorations to remove parathyroid implants are often unsuccessful.

We came across a 55-year-old male with bone pains, pruritus, polyuria, difficulty in getting up from the chair for the last 1 year. He was operated in the past for renal stones. He was diagnosed with primary hyperparathyroidism (PHPT) due to left superior parathyroid adenoma and got operated endoscopically for the same. Postsurgery, discharged medications included oral calcium and Vitamin D supplements. Two years after the surgery, he presented with bone pains and increasing fatigue. Physical examination was unremarkable. Serum calcium was 14 mg/dl, intact parathyroid hormone (PTH) was 1400 pg/ml, and 24-h urinary calcium was 649.28 mg/dl, suggestive of recurrent hyperparathyroidism. Ultrasonography, sestamibi and positron emission tomography scans failed to localize lesion. Differential diagnosis of incomplete removal of adenoma, hyperplasia, multiple/ectopic adenoma, malignancy, and parathyromatosis were considered. On exploration [Figure 1a], multiple nodules (<5 mm) were evident in the left-side neck compartment embedded in strap muscles, sternocleidomastoid, on thyroid surface, and left central compartment. Right parathyroid glands were normal. The patient underwent left hemithyroidectomy, removal of ipsilateral straps, parts of sternocleidomastoid, berry picking of superficial nodules, and clearance of tissue close to the entry of ports. Postoperatively, serum calcium was 9.7 mg/dl and PTH (<2.5 pg/ml) was undetectable. Histopathology revealed multiple, small, hypercellular parathyroid glands along with normal looking thyroid follicles with diagnosis of parathyromatosis [Figure 1b]. On 1-year follow-up, serum calcium was normal.

Parathyromatosis is a rare but clinically relevant disease. It is characterized by ectopic hormone secreting parathyroid tissue scattered throughout the neck and mediastinum. It may be considered a benign malignancy with locally invasive behavior. In our opinion, the cause of secondary hyperparathyroidism, in this case, was most likely due to rupture of capsule leading to spillage of tumor cells during removal of parathyroid adenoma by endoscopic parathyroid surgery. Secondary parathyromatosis following endoscopic parathyroid surgery has not been reported so far. The preoperative diagnosis of parathyromatosis poses great challenges and needle aspiration may be helpful in selected patients. The sonographic appearance of parathyromatosis may mimic that of thyroid nodules, parathyroid adenomas, and metastases. The diagnosis is based on histopathology of the surgical specimen.
of disseminated local malignancy.\(^1\) Parathyroid carcinoma is an important differential diagnosis, but significantly elevated serum calcium (>14 mg/dl) are seen in carcinoma.\(^1\) Histopathology can be diagnostic.

The surgery to control parathyromatosis is often unsuccessful. To identify and remove all the parathyroid nodules is extremely difficult. Regular follow-ups are necessary to diagnose any recurrence. Medical treatment with calcimimetics such as cinacalcet and bisphosphonate may be considered in those with failed surgery or unfit for surgery. To the best of our knowledge, this is the first observation of parathyromatosis following an endoscopic surgery removal of benign parathyroid adenoma which calls for vigilant follow up in such cases.

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**Conflicts of interest**

There are no conflicts of interest.

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