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.[1,2] Primary or Type 1 parathyromatosis is the result of hyperplasia of parathyroid rests from embryologic development.[3] Secondary or Type 2 parathyromatosis, a rare complication of parathyroidectomy, first described in 1975 by Palmer *et al*.,[4] is more common which arises due to seeding of parathyroid tissue during surgery. It has been also considered as a low-grade malignancy.[5] Thirty-five cases have been reported so far.[6] Its preoperative diagnosis is rare due to the lack of awareness of this entity. Sonographic imaging provides clues to the diagnosis.[7] 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.[8] 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.

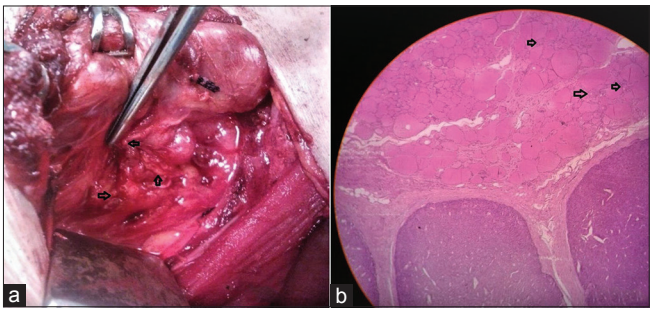

Figure 1: Gross (a) and histopathological (b) findings in parathyromatosis

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.[9] The sonographic appearance of parathyromatosis may mimic that

of disseminated local malignancy.[7] 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.

Acknowledgment

We would like to thank Dr. Vijay Katekhaye for his assistance in editing this letter.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

Ajay Aggarwal, Roopak Wadhwa, Vivek Aggarwal[1]

Departments of Endocrinology and [1]Endocrine Surgery, Fortis Hospital, Shalimar Bagh, New Delhi, India

Address for correspondence:
Dr. Ajay Aggarwal,
Department of Endocrinology, Fortis Hospital, Shalimar Bagh,
New Delhi - 110 088, India.
E-mail: endocrinologist39@yahoo.co.in

References

1. Fernandez-Ranvier GG, Khanafshar E, Jensen K, Zarnegar R, Lee J, Kebebew E, *et al.* Parathyroid carcinoma, atypical parathyroid adenoma, or parathyromatosis? Cancer 2007;110:255-64.
2. Meakins JL, Milne CA, Hollomby DJ, Goltzman D. Total parathyroidectomy: Parathyroid hormone levels and supernumerary glands in hemodialysis patients. Clin Invest Med 1984;7:21-5.
3. Reddick RL, Costa JC, Marx SJ. Parathyroid hyperplasia and parathyromatosis. Lancet 1977;1:549.
4. Palmer JA, Brown WA, Kerr WH, Rosen IB, Watters NA. The surgical aspects of hyperparathyroidism. Arch Surg 1975;110:1004-7.
5. Barnes BA, Cope O. Carcinoma of the parathyroid glands, report of 10 cases with endocrine function. JAMA 1961;178:556-9.
6. Hage MP, Salti I, El-Hajj Fuleihan G. Parathyromatosis: A rare yet problematic etiology of recurrent and persistent hyperparathyroidism. Metabolism 2012;61:762-75.
7. Tublin ME, Yim JH, Carty SE. Recurrent hyperparathyroidism secondary to parathyromatosis: Clinical and imaging findings. J Ultrasound Med 2007;26:847-51.
8. Daphnis E, Stylianou K, Katsipi I, Stratigis S, Karamitopoulou E, Karkavitsas N, *et al.* Parathyromatosis and the challenge of treatment. Am J Kidney Dis 2006;48:502-5.
9. Baloch ZW, Fraker D, LiVolsi VA. Parathyromatosis as cause of recurrent secondary hyperparathyroidism: A cytologic diagnosis. Diagn Cytopathol 2001;25:403-5.

Access this article online

Quick Response Code:

Website: www.ijem.in

DOI: 10.4103/ijem.IJEM_61_17

How to cite this article: Aggarwal A, Wadhwa R, Aggarwal V. Parathyromatosis following endoscopic parathyroid surgery: A rare occurrence. Indian J Endocr Metab 2017;21:641-2.