Intra-thyroid parathyroid adenoma: a difficult task to find
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ABSTRACT
We report a case of 66-year-old female, who presented with features of hypercalcemia and developed acute kidney injury (AKI). Parathyroid hormone level was raised up to 740 pg/ml. Ultrasound and sestamibi scan suggested presence of a right sided parathyroid adenoma. A decision of right sided parathyroidectomy was taken. The sample that was sent for frozen section showed only thyroid tissue. No macroscopically visible or palpable nodule suspected to be parathyroid adenoma could be found. So, a decision of right hemithyroidectomy was taken and done accordingly. On routine histopathology a solid gray white nodule was found within thyroid tissue and it was confirmed microscopically as parathyroid adenoma.

Key words: hyperparathyroidism, hypercalcemia, intrathyroidal parathyroid, ectopic parathyroid.

INTRODUCTION
The parathyroid glands, four in number, are located behind the thyroid gland but it is sometimes found in a variety of places, from carotid bifurcation to pericardium.\textsuperscript{1} Ectopic parathyroid gland occurs when the parathyroid gland is not in its proper location which is caused by embryologically abnormal migration.\textsuperscript{2} An intrathyroidal parathyroid adenoma is considered to be an ectopic parathyroid gland by definition. This is rare and its incidence is reported to be about 1.3% to 6.7%.\textsuperscript{3} It is usually diagnosed preoperatively by biochemical changes, ultrasound and nuclear imaging as well as per operatively by frozen section examination and parathyroid hormone levels after excision.\textsuperscript{4} But sometimes parathyroid glands are difficult to recognize macroscopically during operation and turn to be thyroid tissue by frozen section examination. Again, in case of ectopic location, parathyroid gland may not be found even after meticulous search. Infrathyroidal parathyroid is the most likely diagnosis in such situations and excision of thyroid is the protocol of surgery in such cases.\textsuperscript{5} Routine histopathological examination of the thyroidecmy specimen is required for final diagnosis. We report on such a case with suspected preoperative parathyroid adenoma but no parathyroid tissue was found in normal location. Finally, it was confirmed as intrathyroidal parathyroid adenoma during pathological evaluation after hemithyroidectomy.

CASE REPORT
A 66-year-old female with known diabetes, hypertension and chronic kidney disease was presented with complaints of leg swelling and decreased urine output for 3 days. She was previously diagnosed as a case of Wegener’s granulomatosis in 2015. At that time, she presented with shortness of breath, cough, hemoptysis, epistaxis and vomiting. She had microscopic hematuria and cANCA, pANCA, ANA were positive. She was stable after treatment with steroid and cyclophosphamide. Two years later on regular follow up, due to rising serum creatinine and microscopic hematuria, a renal biopsy was done which later showed diabetic nephropathy class II.
On her present admission, she was diagnosed with acute kidney injury and hypercalcemia. After conservative management and injectable calcitonin, calcium level decreased from 11.9 mg/dl to 5.4 mg/dl. Raised parathyroid hormone level of 740 pg/ml (up to 55 pg/ml) was found. Ultrasound and sestamibi scan confirmed the diagnosis of right sided parathyroid adenoma and multinodular goiter. Thyroid hormone was within normal limits. Surgery was planned for excision of adenoma with per operative frozen section for confirmation.

A small 1.6 cm x 0.6 cm x 0.5 cm nodule, which was suspected as parathyroid gland was sent for frozen section. The cut surface was grey brown. Microscopic examination of the submitted sample showed normal thyroid tissue. No grossly detectable enlarged parathyroid gland was found even after vigorous searching. So, a decision of right hemithyroidectomy was taken and done accordingly.

Later on, a 6 cm x 3 cm x 2 cm piece of thyroid tissue was sent for routine histopathological evaluation (Figure 1). At the same time serum parathyroid level was done postoperatively, which were 557, 263, 172 and 91 pg/ml consecutively at an interval of 30 minutes. The PTH level gradually decreased. The cut surface of the thyroid tissue reveals a 1.5 cm solid grey white nodule. The other areas were meaty. Sections were submitted for tissue processing and paraffin embedding. After preparation of tissue slides, microscopic examination of the nodule showed an encapsulated intrathyroidal parathyroid adenoma. It consisted mainly of chief cells having round to oval uniform nuclei and moderate amount of clear cytoplasm (Figure 2). These are arranged diffusely and in clusters. The surrounding thyroid tissue shows colloid filled follicles. Unfortunately patient did not come for follow up visit.

DISCUSSION

Primary hyperparathyroidism is considered as the second most common cause of predominant hypercalcemia in postmenopausal women. The treatment of choice for symptomatic hypercalcemia is parathyroidectomy.6 Usually, there are three to four parathyroid glands including superior and inferior. The superior parathyroid gland derives from the fourth branchial arch. The inferior parathyroid gland, like the thymus, derives from the third branchial arch. It has been hypothesized that an intrathyroidal parathyroid gland is formed when the parathyroid tissue is trapped between the central and lateral lobes of the thyroid gland during their fusion.1,2 Ectopic parathyroid glands occur more often in the inferior gland than in the upper gland because of the differentiation of embryonic development of the parathyroid gland. The inferior parathyroid gland descends through the neck and down the thymus along an embryonic long virtual tract and the superior parathyroid gland descends through a shorter length compared to the inferior parathyroid gland.3 In our case,
it was an intrathyroidal parathyroid adenoma of the superior parathyroid gland.

Ultrasound of the thyroid gland is usually done in patients with primary hyperparathyroidism. Ultrasonogram detects parathyroid hyperplasia, parathyroid adenoma and parathyroid carcinoma as hypoechoic nodules but normal and ectopic parathyroid glands within the mediastinum cannot be seen on ultrasonogram. Computed tomography (CT) scan is a good additional test to complement these limitations. Tc-99m sestamibi scan is also done to confirm the diagnosis of parathyroid adenoma and for its location.

However, if the parathyroid adenoma cannot be found even after careful surgical field exploration, a possibility of intrathyroidal parathyroid gland should be kept in mind. In that case, thyroid lobectomy of the suspected side should be performed. This happened in our case where parathyroid adenoma was lying within the right lobe of thyroid gland and hemithyroidectomy explored the lesion.

Conclusion

Hyperparathyroidism is a common endocrine disorder but intrathyroidal parathyroid adenoma is rare. This case is an intrathyroidal parathyroid adenoma in the superior parathyroid gland, which is rarer. Meticulous search during operation even using the facility of frozen section failed to detect intrathyroidal location of parathyroid gland. Knowing the fact that parathyroid adenoma may remain hidden within thyroid tissue, decision of hemithyroidectomy becomes beneficial to the patient and helpful for a good clinical outcome.

Authors’ contribution: ZT: conceptualization, literature search and writing manuscript, BA: manuscript writing, NA, SS: diagnosis, management, review and editing.

Consent: Informed written consent was taken from the patient for publication of this case report and any accompanying images.

Conflicts of interest: Nothing to declare.

REFERENCES