

Intra-Thyroidal Parathyroid Adenoma: A Case Report.

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Abstract: Parathyroid adenomas account for 85% of cases of primary hyperparathyroidism. In approximately 6-16% of cases, one or more hyperfunctioning parathyroid glands are in an ectopic location. The presence of an intrathyroidal parathyroid is a rare condition known for its preoperative and intraoperative diagnostic problems in localization. Hungry bone syndrome is a potentially preventable condition in postparathyroidectomy, which correlates with the weight of the resected adenoma. We report the case of an intrathyroidal parathyroid adenoma in a 51-year-old woman with hypercalcemia requiring adequate preoperative correction before surgery.

Keywords: Adenoma, Ectopic, Parathormone, Thyroid nodules

INTRODUCTION

Primary hyperparathyroidism (PPH) is a common endocrine disorder characterized by hypercalcemia and excessive parathyroid hormone (PTH) secretion that may or may not be symptomatic. It is most commonly caused by a single adenoma of the parathyroid gland. Most parathyroid neoplasms are benign, with the lower glands being relatively more frequently affected than the upper glands.

Intrathyroidal parathyroid adenoma is a rare abnormality explained by abnormalities in the migration of embryonic parathyroid embryos. Preoperative diagnosis of this entity by ultrasound is possible as well as data from Sestamibi scintigraphy; however, it is difficult to distinguish it from a thyroid nodule.

Thyroid lobe hemi-thyroidectomy with intrathyroid adenoma is the preferred surgery. However, coexisting thyroid disease, diagnosed on preoperative imaging, also determines the extent of surgery. Here we report a case of hyperparathyroidism due to an intrathyroidal parathyroid adenoma.

CASE PRESENTATION

A 51-year-old female patient with type 2 diabetes on oral medication presented with a 3-month history of an enlarged mass in the anterior cervical region lateralized to the right. There was no dysphagia, stridor, or symptoms of hyperthyroidism or hypercalcemia. On examination, there was a firm, painless swelling of the right thyroid lobe measuring 4 cm. The cervical lymph node areas were free.

Thyroid function tests were normal. Serum calcium was markedly elevated at 147 mg/L, with hypophosphatemia of 23 mg/L. She was treated with bisphosphonates. Her parathyroid hormone level was more than 25 times the upper limit of normal.

Ultrasound revealed a multi-nodular thyroid, the largest nodule located in the right lower lobar region measuring 45 mm long axis classified as TIRADS IV [Fig.1]. Parathyroid scintigraphy showed a lesion that concentrated the tracer in the right lower pole of the thyroid. The topographic determination of the location was difficult [Fig.2].

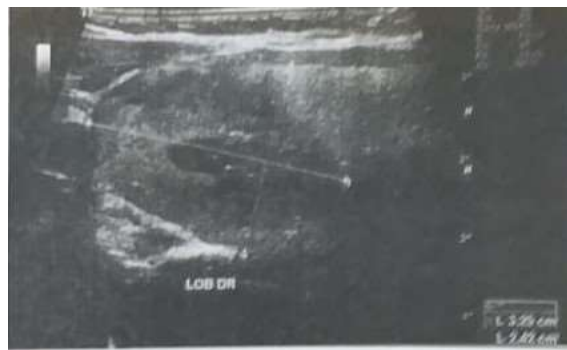


Fig. 1: Thyroid ultrasound showing a 4 cm right intrathyroidal nodule.



Fig.2: Early and late MIBI parathyroid scans reveal focal accumulation of radiotracer uptake in the right lobe of the thyroid.

The patient benefited from a surgical exploration which did not show a parathyroid adenoma. Intraoperative palpation of the right lobe revealed just the intrathyroidal nodule described on ultrasound. An intrathyroidal parathyroid adenoma was suspected. A total thyroidectomy was performed due to the multinodular character of the thyroid [Fig.3]. Anatomopathological examination [Fig.4] of the excisional specimen did not find any sign of malignancy but discovered a parathyroid surrounded by thyroid parenchyma in the right lobe, so it was an intrathyroidal parathyroid.



Fig. 3: Macroscopic image of total thyroidectomy showing the right lower lobar intrathyroidal nodule, which corresponds to the adenoma.

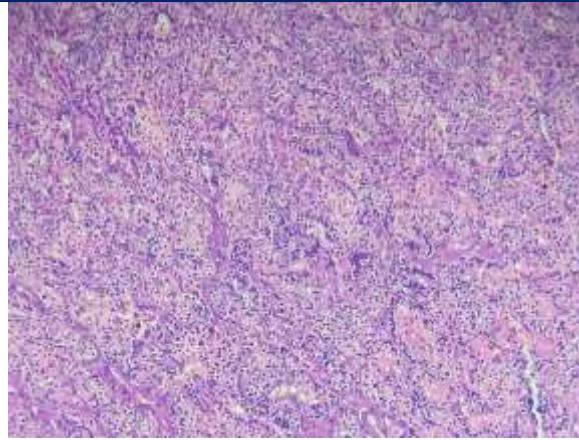


Fig. 4: Histological section of the right lobe of the thyroid showing the intrathyroidal location of the parathyroid adenoma.

On the third postoperative day, a starved bone syndrome developed and intravenous calcium replacement was required. The patient was discharged on the sixth postoperative day on oral calcium and alfacalcidol. A follow-up ultrasound was unremarkable.

DISCUSSION

The most common etiology of primary hyperparathyroidism is single parathyroid adenoma (80-85% of cases), followed by parathyroid hyperplasia (10-15% of cases), double parathyroid adenoma (2-5% of cases) and parathyroid carcinoma (1% of cases) [1]. Most cases occur in women (74%), but the incidence is similar in men and women before 45 years of age [2]. Usually primary hyperparathyroidism is an asymptomatic disease discovered during routine laboratory screening [2].

Intra-thyroidal localization of parathyroids is rare [3]. In surgical series, the estimated prevalence of intra-thyroidal parathyroid adenoma ranges from 1.4 to 6% [4]. Embryologically, the upper parathyroids develop from the fourth pharyngeal pouch and the lower parathyroids from the third pouch [2]. The inferior parathyroids migrate a greater distance, therefore they are more likely to be found in ectopic sites [5]. Parathyroid ectopy can be located from the angle of the mandible to the pericardium, but always in the cervicomediastinal visceral sheath. In the series of Jaskowiak et al, the thyroid location represents the second most common ectopic site after the thymic and mediastinal sites [6]. The location of most adenomas (70.6%) was found to be on the right side and on the lower third of the thyroid lobes [4].

Intrathyroidal parathyroid adenoma can easily be mistaken for a thyroid nodule [7]. The search for parathyroid adenoma can be aided by ultrasound and scintigraphy [3]. In the case of intrathyroidal adenomas, preoperative or intraoperative ultrasound may be the most informative imaging method [2]. Tc-99m sestamibi scintigraphy reports a sensitivity of 88% in the preoperative localization of a parathyroid adenoma [8]. Although ultrasound alone has an overall sensitivity of 60-65% for the detection of parathyroid adenomas and when combined with sestamibi Tc-99m scintigraphy in the evaluation of patients with a single adenoma, the sensitivity can increase to 100% [9].

Histologically, to affirm that a parathyroid is intrathyroidal, it must be surrounded by the thyroid parenchyma and it must not have a capsule [3]. These variations in parathyroid locations explain the difficulty encountered when searching for parathyroid adenomas [3]. Parathyroid adenomas usually measure less than 2 cm and weigh less than 1 g. In case of parathyroid lesions larger than 2 cm with very high PTH levels, the differential diagnosis between parathyroid adenomas and parathyroid carcinomas should be considered [10, 4].

Surgery is a valid option given the high calcium levels and osteoporosis. It is the most appropriate treatment for primary hyperparathyroidism [4]. Parathyroidectomy is associated with a cure rate of 95-98% and a low complication rate (1-3%) [2]. The current therapeutic strategy aims at a preoperative identification of pathological parathyroid glands that combines data from cervical ultrasound, MIBI-Tc99m scintigraphy [11]. During parathyroid surgical exploration, if a missing parathyroid gland cannot be found, any nodule within the thyroid should be considered an intra-thyroid parathyroid adenoma until proven otherwise and thyroid lobectomy is appropriate [12].

Our patient developed Bone Starvation Syndrome on the third postoperative day. This is the name given to the rapid and severe drop in serum calcium due to the abrupt cessation of PTH after parathyroidectomy in patients with severe primary hyperparathyroidism [7]. The lowest serum calcium level is usually observed around the third postoperative day [13]. The volume of the resected gland, high preoperative serum calcium level and mean PTH concentrations greater than or equal to 80 pg/ml correlate with the risk of bone

starvation syndrome [2, 7]. Prophylactic measures such as preoperative bisphosphonate supplementation are currently under investigation [7]. Treatment involves the administration of calcium [7]. In our case, the PTH level collected after total thyroidectomy decreased by approximately 98% compared to the preoperative value [2].

Some studies estimate that concomitant thyroid disease occurs in 20-50% of cases of primary hyperparathyroidism, of which 3-4% are malignancies [14]. The most common thyroid pathology reported in most studies is nodular hyperplasia [14].

CONCLUSION

Intrathyroidal parathyroid adenomas are rare and may be misinterpreted as thyroid nodules on preoperative imaging, especially in the presence of concomitant thyroid disease. In the setting of hyperparathyroidism, an intrathyroidal parathyroid adenoma should be considered in the presence of a thyroid nodule and negative surgical exploration of the neck. This intrathyroidal location of the parathyroids warrants thyroidotomy in case of a palpable thyroid mass intraoperatively. Preoperative use of bisphosphonates may alleviate bone starvation syndrome after parathyroidectomy.

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