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**Case Series**

# Intrathyroid parathyroid adenoma: report of two cases and literature review

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## ABSTRACT

Approximately 80% of patients with primary hyperparathyroidism have a parathyroid adenoma, with surgery being the only definitive treatment. Sometimes in surgery there is difficulty in identifying the pathological parathyroid gland at which time the possibility of ectopic parathyroid should be considered. We present two cases of patients which after hemithyroidectomy histopathological report, reported intrathyroid parathyroid adenoma. Intrathyroid parathyroid adenomas are an infrequent presentation of parathyroid adenomas that require high clinical suspicion if they are not detected by imaging studies in the preoperative period.

**Keywords:** Intrathyroid parathyroid adenoma, Hemithyroidectomy, Hyperparathyroidism

## INTRODUCTION

Primary hyperparathyroidism is an endocrine disorder characterized by mild or moderate hypercalcemia and normal or elevated parathyroid hormone (PTH). Approximately 80% of patients with primary hyperparathyroidism have a parathyroid adenoma, with surgery being the only definitive treatment.[1]Sometimes in surgery there is difficulty in identifying the pathological parathyroid gland at which time the possibility of ectopic parathyroid should be considered.[2] Intrathyroid parathyroid adenoma is a rare entity, occurring in 2.2 to 4% of all intrathyroid adenomas.[3] We present the case of a 48-year-old woman with primary hyperparathyroidism due to an adenoma with an intrathyroid location.

## CASE SERIES

### *Case 1*

48 year old woman, with a history of recurrent kidney stones under urology control. During his follow-up in December 2019, elevated serum levels of calcium and parathyroid hormone were evidenced (11.1 mg/dl and 321 pg/ml).

A scintigraphy was performed detecting cellular activity in the lower right parathyroid gland, so it was decided to carry out a parathyroidectomy by the oncological surgery service.

During the procedure, it was not possible to locate the lower right parathyroid gland in its usual anatomical position, however, a 5×5 mm nodular lesion was palpated, mobile with regular edges in the lower pole of the right thyroid lobe, for which a right hemithyroidectomy was performed and the tissue obtained was sent for pathological study.

Patient is discharged home 24 hours after surgery without complications.

The histopathological report reported 2.3 cm intrathyroid parathyroid adenoma without evidence of necrosis.

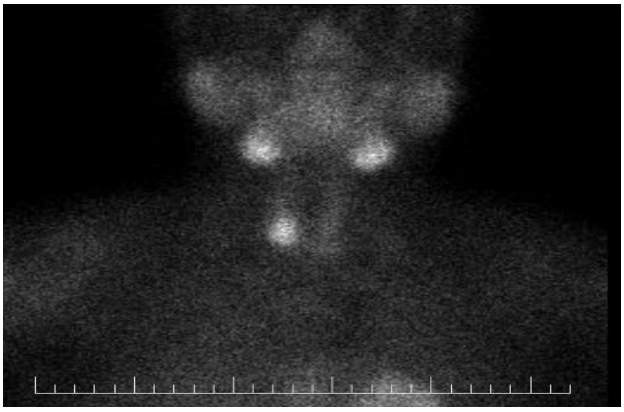

**Figure 1: Scintigraphy case 1 (arrow: cellular activity in the lower right parathyroid).**

***Case 2***

A 74-year-old woman with a history of diabetes mellitus and systemic arterial hypertension under management with metformin and enalapril. Started with a mass on the anterior face of the neck 2 years previously associated with dysphagia for 4 months previously.

Tomography of the neck and thorax was performed where a lesion in the left thyroid lobe was observed, for which an ultrasound-guided fine needle aspiration biopsy was performed, reported by Bethesda 1; oncological surgery carry out a resection with study intraoperative.

During the procedure, hemithyroidectomy was performed, sending a surgical specimen for intraoperative study that reports intrathyroid intrathyroid adenoma, which is why the procedure is completed.

Patient was discharged home 24 hours after surgery without complications.

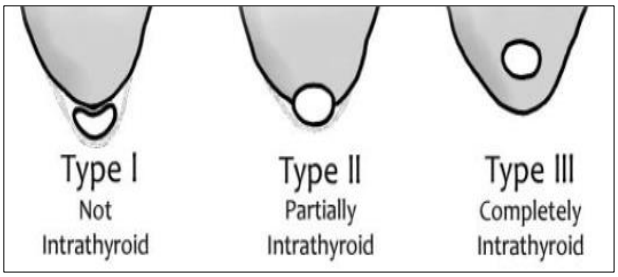


**Figure 2: Classification of intrathrodeal parathyroid adenoma.**

## DISCUSSION

Primary hyperparathyroidism, as previously mentioned, has elevated serum calcium levels without suppression of PTH levels, which increases the risk of cortical bone loss, fractures and, as observed in our case, kidney stones. Surgery as treatment is recommended for patients younger than 50 years of age with significant hypercalcemia, osteoporosis or fragility fractures, kidney stones, hypercalciuria and impaired renal function.[1]

The intrathyroid parathyroid glands are classified into three types according to their location in relation to the thyroid gland. Type I in which the parathyroid is very attached to the thyroid and within the tissues that surround it. It is outside the thyroid capsule. Type II basically contained within the thyroid but still visible on the thyroid surface. Type III in which the parathyroid is completely within the thyroid parenchyma corresponding to the true intrathyroid location, which in the case of a parathyroid adenoma has an incidence of only 0.7%.[4]

During the 1980s, there was controversy over the embryological origin of intrathyroid parathyroid glandular tissue, however, several current publications have shown that IPAs can correspond to both the III and IV branchial arches and sometimes to supernumerary glands that are trapped during the descent of the primordium. Central thyroid and its fusion with the lateral portions, although another theory postulates an excessive descent in the inferior migration of the IV brachial arch, being the most frequent location being the lower right.[4,5]

Intrathyroid parathyroid adenomas are the second leading cause of persistent hyperparathyroidism after intrathymic adenomas, with a reported incidence between 1.3 to 6.7.[4,5]

Diagnostic precision and preoperative location are important because they affect surgical behavior. Scintigraphy technetium (TC) 99m scan with single proton emission tomography (sestamibi SPECT) has a high sensitivity for the diagnosis of intrathyroid parathyroid lesions and is the main method of localization, however it does not have adequate specificity and its ability to identify anatomy is not the ideal. Ultrasound can be used as the first line since it is inexpensive, non-ionizing and can differentiate anatomical structures very well, however, the differential diagnosis between intrathyroid parathyroid lesions and thyroid nodules can be difficult.[6-8]

There is no defined standard or surgical strategy for patients with intrathyroid parathyroid adenomas, considering that preoperative localization studies for adenomas are usually not reliable. Few publications have focused on the surgical procedure for IPA. Hurden et al proposed a sequential surgical approach for ectopic adenomas comprising: systematic cervical parathyroid exploration beginning at the side with the higher probability of adenoma; in cases where the parathyroid adenoma was not found initially or when the resection of the adenoma showed not decrease more of 50% in the parathyroid hormone (PTH) was should performed extended cervical exploration centripetally from the potential parathyroid localization of the parathyroid glands along the thyreothymic ligament, the esophagotracheal sulcus, the carotid sheath, the retropharyngeal/esophageal region, and of the cranial ventral and dorsal mediastinum; if no adenoma is found, continue with hemithyroidectomy

on the side with higher suspicion for intrathyroid adenoma based on preoperative testing or higher clinical suspicion.[3]

In the pathological study, the intrathyroid parathyroid adenomas do not differ from the usual parathyroid adenomas, observing under the microscope main cells organized in nests, cords or vesicles, in addition to papillary or pseudopapillary appearance. The light from the vesicles and blood may contain eosinophilic material. Adipocytes are absent, interstitial tissue is well vascularized and not abundant, usually with edematous or fibrous changes with or without hemosiderin deposits. The main cells of the parathyroid adenoma are larger than those that form the normal parathyroid and they also have weakly eosinophilic or clear cytoplasms. Their cores are rounded with dense chromatin without nucleolus. Regarding immunohistochemistry, parathyroid adenoma expresses parahormone and chromogranin A.[9]

Parathyroid adenomas usually measure less than 2 cm and weigh less than 1 gram. In lesions larger than 2 cm, a differential diagnosis between giant parathyroid adenoma and parathyroid carcinoma should be considered. The main signs of malignancy are capsular invasion, angioinvasion and invasion of adjacent structures. There is no correlation between adenoma size with symptoms and functional status.[10]

## CONCLUSION

In conclusion, intrathyroid parathyroid adenomas are an infrequent presentation of parathyroid adenomas that require high clinical suspicion if they are not detected by imaging studies in the preoperative period.

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