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Case Report

Concurrent Intrathyroidal Parathyroid Adenoma and Papillary Thyroid Microcarcinoma: A Case Report

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Abstract

Papillary thyroid carcinoma (PTC) is the commonest endocrine thyroid malignancy. The concurrent incidence of intrathyroidal parathyroid adenoma and papillary thyroid carcinoma is an infrequent finding. Surgical treatment via excision is the only definitive management approach. Here in we present a case report of intrathyroid parathyroid adenoma with incidental papillary micro carcinoma in a 36 year-old-female who initially presented with osteoporosis.

Keywords: Parathyroid adenoma, osteoporosis, papillary thyroid carcinoma, surgical excision.

INTRODUCTION

The frequency of parathyroid adenomas placed completely inside the thyroid is 0.7%-6%. They are often problematic to confine preoperatively and intraoperatively, making documentation and management are both challenging. To our knowledge, this is the second case of synchronized parathyroid adenoma and micropapillary thyroid carcinoma reported in the Kingdom of Saudi Arabia.

CASE REPORT

A 36 year-old- female has been referred to Endocrinology service at king Fahad specialist hospital-Dammam in October 2022. The patient was discovered to have high serum calcium level during her pregnancy in 2021. A dual-energy X-ray absorptiometry scan showed a lumbar spine L4 Z-score of -3.3, total of -2.8 and a T score of -2.8, confirming the diagnosis of osteoporosis. The patient reported no history of renal stones, abdominal pain, fracture or family history of similar disorders. The patient had hypercalcemia and persistent hyperparathyroidism. Her lab tests revealed Serum Calcium level of 2.66 mmol/l, parathyroid hormone (PTH) level is 677 pg/ml and thyroid stimulating hormone (TSH) level 1.05 uIU/ml. Based on the above laboratory results a primary diagnosis of primary hyperparathyroidism is established for definitive localization. Ultrasound imaging report of the neck revealed the following; thyroid gland demonstrates normal size and echogenicity, however, a single hypoechoic solid nodule in the right lobe measuring 2.6 x 1.4 x 0.9 cm is seen compatible with *TR4 nodule*, or "moderately suspicious". No definite mass in the left thyroid lobe identified. No definite mass at the expected location of parathyroid gland. Her sestamibi parathyroid scan revealed prominent hypodense lesion overlying the right thyroid lobe measuring 2.6 cm in maximum dimension. Otherwise, no abnormal focal uptake to suggest the presence of hyper functioning thyroid tissues. The report concluded that there is no scintigraphic evidence of hyper functioning parathyroid adenoma, however, Sestamibi avid right hyperfunctioning thyroid nodule is noted. The patient underwent ultrasound guided fine needle aspiration (FNAC) of the right thyroid nodule on April 2023, the final cytological examination reported as parathyroid epithelial lesion/neoplasm. After five months of establishment of the final diagnosis, the patient had right hemithyroidectomy with right parathyroidectomy. Grossly, we received the specimen as fragmented pieces of para thyroid tissue measuring in aggregate 3.0 x 2.0 x 0.3 cm and weighing 3 gm along with another tissue piece of right thyroid lobectomy measuring 3.5 x 2.0 x 1.3 cm, with an iatrogenic surgical cut through the surface, probably related to the resected intrathyroidal parathyroid lesion. the histopathological examination confirmed the diagnosis of unifocal papillary thyroid microcarcinoma. the right fragmented lesion was confirmed to be IPA, which consisted of chief cells and was completely embedded within thyroid tissue. the immunohistochemical staining was positive for

chromogranin A and synaptophysin in both parathyroid tissues (Fig. 4c, d). The serum calcium levels returned to normal, and iPTH decreased from 732.7 before surgery to 13.1 pg/mL after surgery. Her corrected calcium level is 2.41 mmol/liter, so the patient was discharged home without medication. At 1-month postoperative, she was asymptomatic and calcium level was normal. At 3 months follow-up, the serum Parathyroid Hormone (PTH) measures 78.3 pg/mL, and the patient's overall status remained stable.

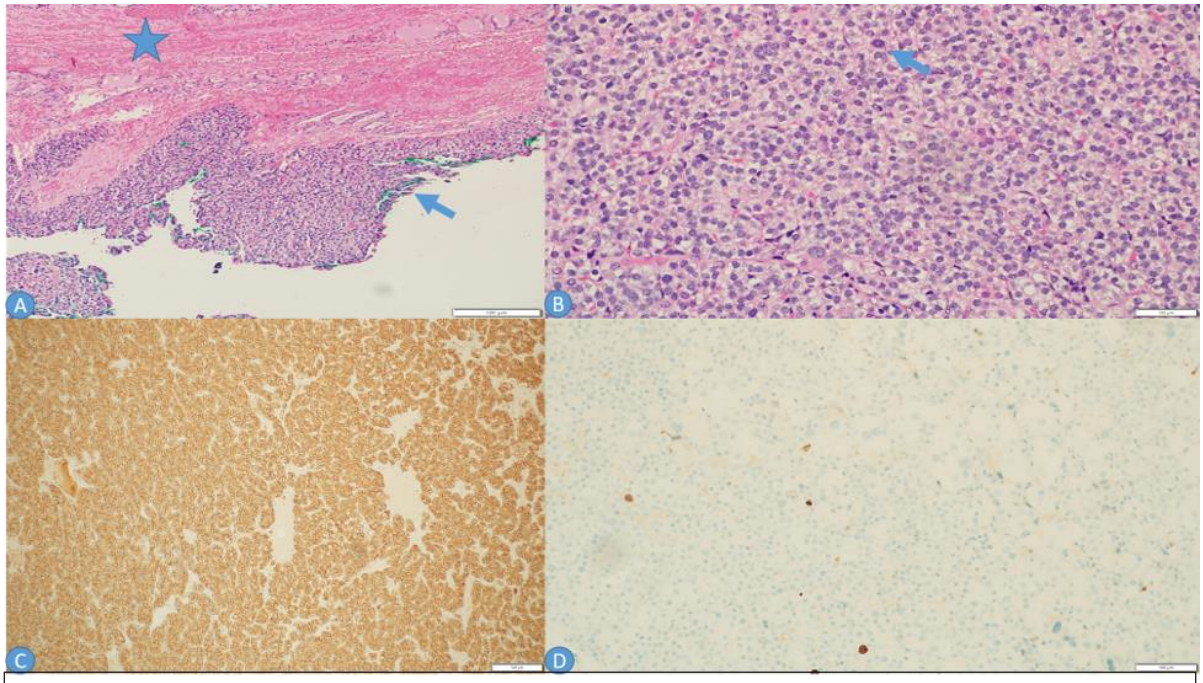

Figure 1: Microscopic pictures of the intrathyroidal parathyroid adenoma; (A) shows thyroid parnechyms (marked with a star), encasing parathyroid adenoma (marked with a blue arrow)(10x, H&E). (B) shows higher magnification of the adenoma with focal nuclear hyperchromatic nuclei and enlargement (marked with a blue arrow) (40x, H&E). (C) Immunohistochemical stain of chromogranin, features diffuse positive membranous expression (10x, IHC). (D) Ki-67 proliferation index of the lesion is not exceeding 3 % (10x, IHC).

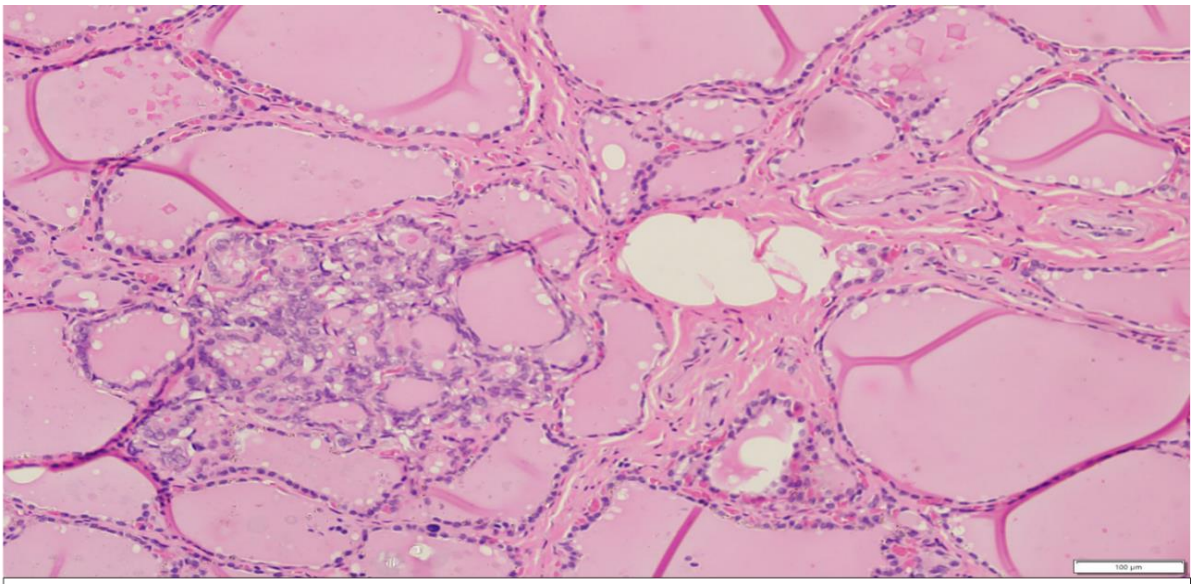

Figure2: Microscopic Picture of the unifocal microscopic papillary thyroid carcinoma (20x, H&E)

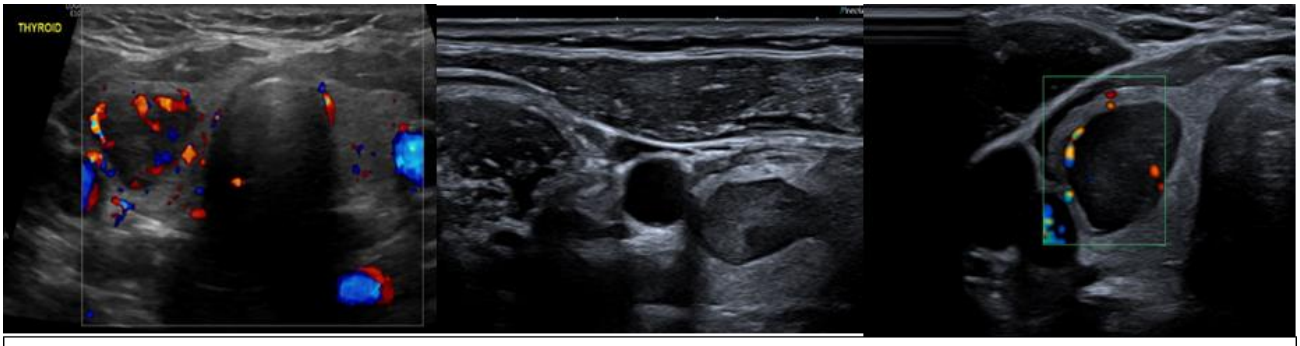

Figure 3: Ultrasonography image of hypoechoic nodule with well-defined margin, measuring 2.6 x 1.4 x 0.9 cm located in the right lobe of the thyroid gland.

DISCUSSION

Primary hyperparathyroidism (PHPT) is a common endocrine disorder caused by excess parathyroid hormone (PTH) production. The disorder is initiated by a single parathyroid adenoma up to 85% of the time [1]. Typically, there are four separate parathyroid glands [1]. Two glands are usually located posterior to the upper poles of the thyroid and are termed "superior" parathyroid glands; the remaining two glands have a more variable location but are typically located near the inferior thyroid poles and are termed "inferior" parathyroid glands. Though, parathyroid glands can similarly be located at "ectopic" sites, due to deviations in migration during embryological growth [2].

Due to difficulties in appropriately identifying an IPA, patients can present with advanced symptoms of primary hyperparathyroidism over a extended period. Severe bone disease seems to be more dominant with ectopic parathyroid adenomas, including IPAs [3]. This may reflect postponements in diagnosis and conclusive management in patients with ectopic parathyroid adenomas.

Biochemical results typically demonstrate hypercalcemia and elevated PTH levels, even though patients can be normocalcaemic. A substantial mass on neck examination is rare for parathyroid adenomas, including IPAs, and more indicative of thyroid pathology or parathyroid malignancy [3].

Primary hyperparathyroidism can be associated with osteoporosis, however, our report presents multiple challenges with respect to identifying the unique imaging features that clinicians should be aware of while managing patients with simultaneous hyperparathyroidism and thyroid nodules, especially patients with features of osteoporosis. For instance, on imaging, they can appear similar to other structures in the thyroid, including benign thyroid nodules, making differentiation difficult [4].

Ultrasonography and $^{99m}TcO^4$-sestamibi SPEC/CT are the most common imaging modalities used to locate abnormal parathyroid glands. An important clue for diagnosing IPA is the presence of sonographic hyperechoic line on the ventral surface of the nodule and Polar vessel supplying adenoma, however, our patient had no characteristic imaging features. Therefore, additional assessment was mandatory with $^{99m}TcO^4$-sestamibi SPEC/CT, which has high sensitivity (84%) and positive predictive value (95%) and improves upon the detection rate of ultrasonography for ectopic lesions [5].

It is well known that ultrasonography can enable an exceptional assessment of parathyroid adenomas with a sensitivity of 76% and a positive predictive value of 93% [16]. In our case, ultrasonography only found one right thyroid nodule without tracer uptake which was confirmed to be a parathyroid neoplasm by Fine needle Aspiration (FNAB) [6].

In a patient with osteoporosis due to primary hyperparathyroidism, the management of the osteoporosis includes not only its medical treatment but also a parathyroidectomy. This surgical approach can usually reverse the bone damage after one to two years [6].

The prevalence of incidental papillary thyroid micro-carcinoma increased from *1.03% of all thyroid tumors in 2008 to 2.31% in 2016 (7)*. In our report, we present a case of unifocal microscopic papillary thyroid carcinoma diagnosed with concurrent evidence of osteoporosis. Regarding definitive management, there are no consensus guidelines exist for the best practices. Therefore, the decisions depend on the individual patient characteristics and the surgeons' preferences, and the treatment should maintain normal calcium levels [7].

Due to difficulties in correctly diagnosing an IPA, patients can present with progressive symptoms of primary hyperparathyroidism over a prolonged period. Symptoms of hypercalcemia are varied and can include bone pain, polyuria, renal colic, constipation, and depression. Severe bone disease appears to be more

prevalent with ectopic parathyroid adenomas, including IPAs. [4]This might reflect delays in diagnosis and definitive management in patients with ectopic parathyroid adenomas [8].

For effective parathyroidectomy, a precise preoperative localization of the parathyroid tissues is precarious. The adenoma is either partly (>50%) or completely enveloped by the thyroid gland. Thyroid lobectomy (partial or full), thyroidotomy and enucleation are all considered available techniques for removing IPAs with higher success rates have been reported for lobectomy [8, 9].

Although the precise localization and differential diagnosis of these conditions are challenging, the combination of ultrasonography, SPECT/CT and careful interpretation could help clinicians confirm the presence of IPA and papillary thyroid carcinoma [8, 9].

To our knowledge, this is the second case of concurrent occurrence of parathyroid adenoma and micropapillary thyroid carcinoma reported in the Kingdom of Saudi Arabia. Intraoperatively, excision and confirmation of the parathyroid localization was done through intraoperative parathyroid hormone level measurement. We advise that the possibility of concurrent occurrence of these two tumors has to be studied [10].

CONCLUSION

The intrathyroidal region is infrequent ectopic site of the parathyroid adenoma. Imaging method is essential to detect accurate localization of ITPAs in the presence of negative or discordant examinations. Subsequently the pre-operative localization studies may disclose the ITPAs in the bulk of patients in qualified centers, surgical achievement is high and blind thyroidectomy must be avoided.

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