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Case report

Co-occurrence of bilateral intrathyroidal parathyroid gland and papillary thyroid carcinoma; a case report

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ABSTRACT

Introduction: Intrathyroidal parathyroid is a parathyroid completely embedded within the thyroid parenchyma. The aim of this study is to present a case with co-occurring bilateral normal intrathyroidal parathyroid glands and papillary thyroid carcinoma (PTC).

Case presentation: A 35-year-old female presented with anterior neck swelling for a duration of a week. The patient was generally asymptomatic. Ultrasound showed mild enlargement of the thyroid gland, and a well-defined solid nodule measuring 9 * 8 * 7 mm in the left lobe with malignant characteristics. Laboratory findings were within normal limits. Fine needle aspiration of the nodule resulted in the diagnosis of PTC. The patient underwent total thyroidectomy. Histological examination confirmed the diagnosis of well differentiated multifocal bilateral PTC alongside two bilaterally located intrathyroidal parathyroid glands.

Discussion: The occurrence of normal ITP is exceedingly rare, and even rarer when simultaneous with other thyroidal lesions. It is theorized that inferior parathyroid glands arise from the third pharyngeal pouch, and during embryogenesis they might migrate to other anatomic locations. In this case, two bilateral normal ITP were present with PTC.

Conclusion: Normal ITP presents a diagnostic challenge due to their high insensitivity to current preoperative diagnostic techniques; hence, surgeons are required to carefully examine thyroid tissue during thyroidectomy when missing parathyroid glands are observed.

1. Introduction

Intrathyroidal parathyroid (ITP) whether normal or abnormal is a parathyroid gland which is completely embedded within the thyroid parenchyma [1]. ITP has been classified into complete (entirely within thyroid tissue) and incomplete (partially embedded in thyroid tissue ≥50%) types [2]. The ectopic localization of parathyroid inside the thyroid is uncommon, and it is even rarer when synchronous with another thyroidal tumor, especially non-medullary types [3]. The bilateral presence of ITP is a phenomenal finding with only a few reported cases in the literature [4].

The aim of this study is to present a case with co-occurring bilateral normal ITP and papillary thyroid carcinoma (PTC). This report has been written in accordance with SCARE guidelines [5].

1.1. Patient information

A 35-year-old female presented with anterior neck swelling for one week duration. Past medical, surgical, family and drug history was unremarkable.

1.2. Clinical findings

Uniform, non-tender, ill-defined soft swelling at the anterior part of

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the neck.

1.3. Diagnostic assessment

Thyroid ultrasound (US) showed mild enlargement of the thyroid gland, suggesting chronic thyroiditis and a well-defined solid nodule of about 9 * 8 * 7 mm in the left lobe with features that make it suspicious for malignancy, there was no cervical lymph node enlargement. Laboratory findings revealed normal thyroid function, and a serum calcium level of 9.09 mg/dl. Fine needle aspiration (FNA) resulted in the diagnosis of PTC in the background of chronic lymphocytic thyroiditis Bethesda VI.

1.4. Therapeutic intervention

Under general anesthesia, the patient underwent total thyroidectomy. Histological examination of the specimen confirmed the diagnosis of well differentiated multifocal bilateral PTC of conventional type. Two intra thyroidal parathyroid glands were also observed, one in each side.

1.5. Follow-up and outcomes

After the operation, the patient's laboratory results showed hypocalcemia of 5.7 mg/dl. Follow up of the patient after 5 months revealed low serum calcium level (5 mg/dl).

2. Discussion

It is well known that at least half of the global adult population are associated with the presence of thyroid tumors, most frequently PTC which can be either unifocal or multifocal [6]. Occasionally, non-thyroidal tissues might accompany these tumors. Such as parathyroid, thymus and lymph nodes [7]. Even though parathyroid adenomas or carcinomas inside the thyroid is far from uncommon [8,9], the incidence of normal parathyroid gland embedded in the thyroid parenchyma is much scarcer with only a handful of cases [10,11]. Usually, in both of normal and tumorous ITP cases a solitary parathyroid nodule is imbedded inside the thyroid tissue, and so far, only 2 cases in the English literature have reported in which bilateral presence of parathyroid nodules on both sides of the thyroid gland was observed [4,12]. The current patient showed co-occurrence of bilateral ITP gland with PTC.

Regarding the formation of ITP, it has been reported that inferior parathyroids more frequently occur in ectopic sites when compared to superior parathyroids. It is theorized that inferior parathyroid glands arise from the third pharyngeal pouch, and during embryogenesis they might migrate to other anatomic locations, most commonly in the thymus (30%), followed by the thyroid (22%) [13,14].

Previous reports of ITP have demonstrated that ITP is usually asymptomatic, and any symptom or clinical findings are linked to associated thyroidal lesion that might be present [15,16]. Chen and colleagues reported a 27-year-old asymptomatic female case of ectopic ITP and PTC with normal family and medical history [15]. Velimezis et al. also reported a 26-year-old asymptomatic case which demonstrated the concurrence of ectopic ITP, PTC, and ectopic intrathyroidal thymus with a past medical history of genitourinary system disorders [16]. The only 2 cases of bilateral ITP were of abnormal hyper parathyroids; hence, symptoms of hyperparathyroidism were associated with these cases [4,12]. The patient in the current study was an asymptomatic 35-year-old female presented with neck swelling with no significant medical or family history.

ITP lesions, such as adenomas and carcinomas are much easier to diagnosis through US and SPEC/CT when compared to normal ITP gland. This might be because normal ITP is asymptomatic and US is insensitive in detecting them, while abnormal ITP lesions are associated with hyperparathyroidism and can more easily be distinguished through imaging techniques and FNA [17,18]. Both Chen et al. and Velimezis et al. in their studies reported that ectopic ITP was not detected through US and was accidently found during postoperative histological examination of the other present thyroid lesion, reportedly PTC [15,16]. The case in the current study was diagnosed for PTC through FNA without any detection of an ITP on US, and patient laboratory results were in normal limits.

The presence of ITP when co-occurring with other thyroidal lesions present a challenge to physicians as due to their rarity, insensitivity to imaging techniques, and their invisibility during thyroidectomy as the result of the lack of a distinguishable cleavage plane with the thyroid parenchyma, surgeons are unaware of their presence inside the thyroid, even if they are found, they simply cannot be easily freed from the thyroidal tissue [4,15]. Chen and associates in their study suggested that surgeons should carefully examine the thyroidal tissue to discover any possible normal ITP, so that they can later perform auto-transplantation of the ITP in order to prevent hypocalcemia due to postoperative hyperthyroidism [15].

In conclusion; the occurrence of normal ITP gland is exceedingly rare, and even rarer when simultaneous with other thyroidal lesions, but phenomenal when it is present bilaterally with other thyroidal lesions. Normal ITP is asymptomatic and present a diagnostic challenge due to their high insensitivity to current preoperative diagnostic techniques; hence, surgeons are required to carefully examine thyroid tissue during thyroidectomy when missing parathyroid glands are observed.

Ethical approval

Approval is not necessary for case report in our locality.

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CRediT authorship contribution statement

Ari M. Abdullah, Hiwa O. Baba, Abdulwahid M. Salih, Aras J. Qaradakhy: Doctors managing the case, follow up the patient, and final approval of the manuscript. Fahmi H. Kakamad, Snur Othman, Berwn A. Abdulla, Shvan H. Mohammed Fahmi H. Kakamad, Berwn A. Abdulla, Shvan H. Mohammed, writing the manuscript, final approval of the manuscript.

Guarantor

Fahmi Hussein Kakamad is the Guarantor of submission

Registration of research studies

According to the previous recommendation, registration is not required for case report.

Consent

Consent has been taken from the patient and the family of the patient.

Declaration of competing interest

There is no conflict to be declared.

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