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Parathyroid adenoma associated with thymoma in a female adult with primary hyperparathyroidism

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ABSTRACT

INTRODUCTION: Parathyroid adenoma associated with a thymoma is a rare clinical entity and few cases have been reported in the literature. This association can be explained by the common embryologic origin of the parathyroid glands and the thymus. These patients may present only with clinical signs and symptoms of primary hyperparathyroidism making it difficult to suspect the coexistence of thymoma.

PRESENTATION OF CASE: A 68-year-old female with medical history of primary hyperparathyroidism underwent surgical excision of a single parathyroid adenoma. Intra-operatively a small mass was detected in the proximity of the parathyroid adenoma and was also removed. Pathologic examination revealed parathyroid adenoma as suspected pre-operatively whereas the small mass excised was identified as a type A thymoma. The postoperative course of the patient was favourable, with normal serum calcium levels and in six months follow-up the patient is symptom-free and with no recurrence.

DISCUSSION: Objective of this study is to report the rare case of a female adult with a parathyroid adenoma and a coexisting thymoma in order to underline the importance of these two pathologies and the ideal treatment that should be followed according to the latest records.

CONCLUSION: Surgical removal of parathyroid adenoma is the treatment of choice for primary hyperparathyroidism, with complete recovery and no postoperative complications. Thymoma may be associated with primary hyperparathyroidism, with or without clinical signs and symptoms of myasthenia gravis and this is a rare clinical entity as very few cases have been described worldwide. To our knowledge this is the first case reported in Greece.

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1. Introduction

Parathyroid adenoma associated with a thymoma is a rare clinical entity and very few cases have been reported in the literature. In order to explain this possible association it is essential to refer that embryologically, the inferior parathyroid glands and thymus are derived from the third pharyngeal pouch, while the superior parathyroid glands originate from the fourth pharyngeal pouch. Later on, the inferior parathyroid glands separate from the thymus and come to lie posterior to the thyroid glands.¹

Patients diagnosed with parathyroid adenoma usually present with clinical signs and symptoms of primary hyperparathyroidism since parathyroid neoplasms are responsible for approximately 85% of all cases of primary hyperparathyroidism and the vast majority are benign (95%).² On the other hand, thymoma may not always be clinically evident. It is exceedingly uncommon in children and young adults, rises in incidence in middle age, and peaks in the seventh decade of life while its' overall incidence in the US is 0.13 per 100,000 person-years.³

Given this uncommon clinical entity, a review of the literature followed in the international electronic database (Pubmed).

Byrne et al. describe the case of a 65-year-old female patient with a history of myasthenia gravis, not medically controlled, who underwent thymectomy and simultaneous excision of three and a half parathyroid glands. Pathologic examination posed the diagnosis of lymphocytic thymoma and nodular hyperplasia of the parathyroid glands. Post-operatively the patient was eucalcemic and the symptoms from myasthenia gravis were reduced.⁴

Palmer et al. report two cases of hyperparathyroidism in which one patient had an associated chemodectoma and the other had a thymoma and myasthenia gravis.⁵

Palin et al. describe the case of a 67-year-old female patient with hyperparathyroidism, who underwent surgical removal of parathyroid adenoma.

Post-operatively the patient presented symptoms that raised the suspicion of myasthenia gravis, later diagnosed by the detection of high levels of acetylcholine receptor antibodies. Further investigation with a normal chest radiograph and thoracic CT scan failed to show the presence of a thymoma, although antibodies to striated muscle were detected. The patient was medically controlled and was symptom-free, yet as a conclusion there was no certainty that myasthenia gravis was associated with hyperparathyroidism, since there was a possibility that the syndrome could have been

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Fig. 1. Left inferior parathyroid gland.



Fig. 2. Parathyroid adenoma up on the left and thymoma down on the right.

precipitated by administration of pamidronate, as a treatment for hypercalcemia.⁶

According to Suzuki et al. a 50-year-old female with hyperparathyroidism and parathyroid adenoma presented a non-invasive thymoma and was treated with concomitant extended thymectomy and resection of the right superior parathyroid gland. The patient did not evidence myasthenia gravis and in a two years follow-up she had no recurrence.⁷

Triggiani et al. describe a patient with myasthenia gravis due to a wide invasive malignant thymoma associated with hyperparathyroidism caused by parathyroid adenoma.⁸

Most recent report of coexisting thymoma and parathyroid adenoma is described by Ceriani et al. and was discovered by sestamibi parathyroid scintigraphy.⁹

Objective of this study is to present the case of a female adult who was surgically treated for primary hyperparathyroidism due to a parathyroid adenoma and who was also diagnosed post-operatively with thymoma, until then asymptomatic. According to our research this is the first case reported in Greece.

2. Presentation of case

A 68-year-old female was diagnosed with primary hyperparathyroidism and underwent surgical excision of a single parathyroid adenoma. According to the patient's history she was under treatment for diabetes and hypertension, while she was prescribed to take Cinacalcet 30 mg daily until the operation day, in order to normalise the high serum calcium levels. During the pre-operative control, thyroid ultrasonography demonstrated an enlarged hypo-echoic left inferior parathyroid gland suggestive for a parathyroid adenoma. The left superior parathyroid gland and the right superior and inferior parathyroid glands were found to be eutopic and of normal size. Pre-operatively serum calcium was 10.2 mg/dL (normal: 8.5–10.4 mg/dL) and parathyroid hormone (PTH) level of 124.2 pg/ml (normal: 10–65 pg/ml).

Neck exploration was performed through a transverse cervical incision. In the inferior pole of the left thyroid lobe an enlarged parathyroid gland was found (Fig. 1) and excised and an intra-operative parathyroid hormone level was determined by a blood sample taken from a peripheral vein catheter, fifteen minutes after the removal of the parathyroid adenoma, that demonstrated a reduction of the PTH level of 58%. In the proximity of the

excised parathyroid gland a small oval mass was identified and also removed (Fig. 2). The right superior and inferior parathyroid glands and the left superior parathyroid gland were identified and found normal.

Pathologic examination of the first specimen revealed a parathyroid adenoma, size 3 cm × 1.5 cm × 1 cm while the second specimen of size 1.3 cm × 1 cm × 0.6 cm was identified as type A thymoma (Fig. 3).

The patient had an uneventful postoperative course and was discharged in the 3^d postoperative day. In a six months follow-up the patient is eucalcemic and with no symptoms of any kind.

3. Discussion

There are few cases documented worldwide of parathyroid adenoma associated with thymoma. To our knowledge this is the first case reported in Greece.

Objective of this study is to present a case of concomitant presence of these two important neoplasms in a 68-year-old female patient who was surgically treated for primary hyperparathyroidism due to parathyroid adenoma and was also diagnosed post-operatively with thymoma identified intra-operatively as a small mass in the proximity of the excised parathyroid adenoma.

In the case of parathyroid adenoma, the only definitive treatment for primary hyperparathyroidism is curative parathyroidectomy, which is defined by normocalcemia after surgery.¹³

The patient was also diagnosed with a type A thymoma. Type A thymoma (also known as spindle cell thymoma and medullary thymoma) accounts for approximately 4–7% of all thymomas.^{10,11} Approximately 17% of this type may be associated with myasthenia gravis.¹⁰ Morphologically the tumour is composed of neoplastic thymic epithelial cells that have spindle/oval shape, lack nuclear atypia, and are accompanied by few, if any non neoplastic

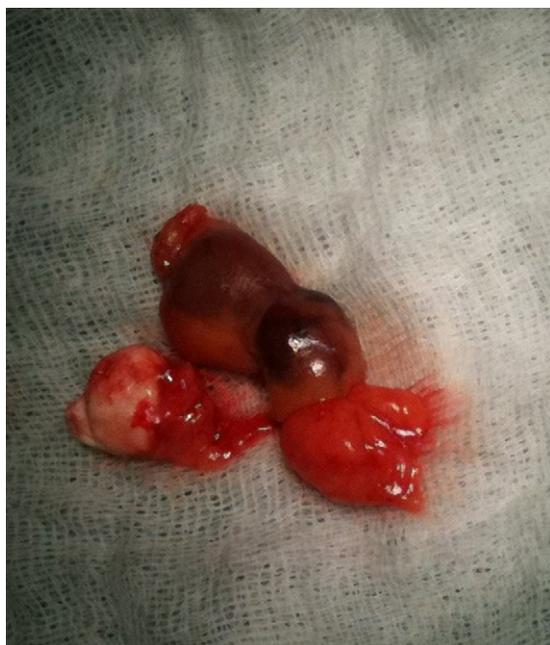


Fig. 3. Surgical specimens of parathyroid adenoma on the right and thymoma on the left.

lymphocytes.¹² The prognosis of this tumour type is excellent and has long-term survival rates (15 years or more) that are reported to be close to 100% in retrospective studies.^{10,11}

In our case the patient in a six months follow-up is symptom-free.

When thymoma is associated with myasthenia gravis surgical removal is the treatment of choice, in case of encapsulated thymomas, whereas in case of invasive thymomas postoperative radiotherapy is advisable regardless of the extent of the resection.¹⁴

In our study the patient had no medical record for myasthenia gravis, no clinical signs or symptoms that could be indicative of the disease and thymectomy was considered to be the curative treatment.

A recent study of Welch et al. proposes transcervical thymectomy, even in absence of thymic lesions because according to the conclusions there is a high yield of parathyroid tissue within the thymus gland and this results essential for the cure of selected patients with hyperparathyroidism.¹⁵

In this case report a 68-year-old female patient treated for primary hyperparathyroidism with parathyroidectomy, due to parathyroid adenoma was also diagnosed with thymoma.

Thymus gland and parathyroid glands have common embryologic origin and therefore, during the surgical procedure of parathyroidectomy for parathyroid adenoma, it is essential a cautious and scrupulous exploration of the surgical field in order to identify the adenoma that may be located in the proximity of the thymus gland, which should be removed in these cases, since it may veil malignant alterations.

4. Conclusion

Due to the intimate developmental relationship of parathyroid gland and thymus gland it is essential to seek for the possible

presence of thymic lesions when treating patients with primary hyperparathyroidism, especially in those cases whose patient history is negative for myasthenia gravis and the suspicion of a possible coexisting thymoma may not be evident.

Conflict of interest

None.

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None.

Ethical approval

“Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request”.

Authors' contributions

Verrioutou Maria is responsible for data collections, data analysis and writing.

Al Mogrampi Saad was involved in revision of the article.

Fardellas Ioannis was involved in final approval of the article to be submitted.

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