Hyperfunctioning insular thyroid carcinoma: A rare case report

Abdulwahid M. Salih a, F.H. Kakamad b,*, Han Aube c

a Faculty of Medical Sciences, School of Medicine, Department Surgery, University of Sulaimani, François Mitterrand Street, Sulaymaniya, Iraq
b Faculty of Medical Sciences, School of Medicine, Department of Cardiothoracic and Vascular Surgery, University of Sulaimani, François Mitterrand Street, Sulaymaniya, Iraq
c Faculty of Medical Sciences, School of Medicine, Department Pathology, University of Sulaimani, François Mitterrand Street, Sulaymaniya, Iraq

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A B S T R A C T
INTRODUCTION: Insular carcinoma of thyroid (ICT) is a rare pattern of thyroid tumor. Being hyperfunctioning makes the condition rarer. Here we reported a case of ICT associated with hyperthyroidism.
CASE REPORT: A 65-year-old female presented with neck mass for 4 years. She had symptoms of thyrotoxicosis and received antithyroid treatment. She was referred for surgical management. She had multiple, non tender, mobile masses in the neck. Ultrasound showed midline multiple heterogeneous mass lesions with multiple lymph nodes in right side of the neck largest one measuring 2 × 2 cm. The patient was prepared for total thyroidectomy with radical neck dissection. The result of histopathology confirmed unifocal, poorly differentiated ICT. The post operative course was uneventful.
CONCLUSION: Insular carcinoma is a rare disease. It may present with long history of signs and symptoms of hyperthyroidism. Total thyroidectomy is the main line of treatment.

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1. Introduction

In 1984, a thyroid neoplasm was described by Carcangi and his colleagues whose characteristics were between the anaplastic and well differentiated thyroid carcinoma [1]. Subsequently, its well-defined nests, made the name to be ‘insular’ [2]. Some studies have recommended insular carcinoma of thyroid (ICT) as an aggressive variant of a well differentiated cancer while others have suggested as a poorly differentiated thyroid carcinoma which is less aggressive [1]. It is a rare pattern of thyroid tumors accounting for about 5% [2]. According to the literature, carcinoma of the thyroid are rarely associated with hyperthyroidism. The incidence of this co-incidence is very rare reaching about 0.15% [3].

In daily practice, thyroid mass detected either by physical examination or by ultrasound in an individual with high thyroid function tests is suggestive for a benign condition [4]. In line with the CARE criteria, we reported a rare case of ICT associated with hyperthyroidism [5]. Reporting this kind of case is necessary to highlight the clinical course of the disease and its prognosis as there is still too much controversy in literature [1,2].

1.1. Patient information

A 65-year-old house wife presented with slow growing anterior neck mass for 4 year duration. She had symptoms of thyrotoxicosis (weight loss, palpitations, nervousness, tremor and fatigue) and received antithyroid treatment with poor control of her thyrotoxicosis. She was referred to us from physician for surgical management.

1.2. Clinical findings

On examination, there were multiple variable size midline masses, largest one about 3 × 4 cm, mobile, firm, not tender, moving with the swallowing with multiple, lymph node enlargement in the right side of the neck. Neither axillary or inguinal lymphadenopathy.

1.3. Diagnostic assessment

Free T3: 10.49, pmol/L, freeT4: 35.41 pmol/L, TFT: less than 0.006 mU/ml. Ultrasound showed midline multiple heterogenous mass lesions, largest one measuring about 43 × 38 × 26 cm with multiple lymph nodes in right side of the neck largest one measuring 2 × 2 cm.
1.4. Therapeutic intervention

The patient prepared for total thyroidectomy with radical neck dissection. The result of histopathology confirmed unifocal, poorly differentiated ICT with capsular invasion and additional pathology of multinodular colloid goiter (Figs. 1 and 2).

1.5. Follow-up and outcomes

The post operative course was uneventful. The patient was put on lifelong thyroxin (100 μg × 1).

2. Discussion

Thyroid malignant tumors cause hyperthyroidism when they occur with Graves' disease, when thyroid-stimulating antibodies induce metastases, or when it presents as a hyperfunctioning autonomously functioning thyroid nodule [6]. According to our knowledge, this is the second reported case of ICT presenting with hyperthyroidism [6]. ICTs are described as poorly differentiated type of thyroid malignant tumor that have much more aggressive clinical outcome than follicular and papillary thyroid carcinomas, with a higher incidence rate of local recurrence, early neighboring lymph node involvement, and metastasis [7]. In this case, right side cervical lymph nodes were positive for malignancy [7]. In terms of clinical behavior and morphology, ICTs have an intermediate position between the undifferentiated or anaplastic thyroid carcinomas and the well differentiated thyroid carcinomas [8]. Presence of this condition for 4 years in our case indicates relativity benign nature of this disease at least in our case. Histopathologically, ICTs compose of solid islands of somewhat uniform neoplastic cells which are separated by artifactual clefts [8]. Although the histologic diagnostic criteria are well defined, the cytologic diagnosis of this entity has not received much attention. Fine Needle Aspiration Cytology (FNAC) is widely used for diagnosis of thyroid gland lesions. However there is little information available in literature about cytologic characteristic of this entity [9]. The differential diagnosis of ICT in FNAC are papillary carcinoma (follicular variant), follicular neoplasm and anaplastic carcinoma [10]. In this case, FNAC was omitted from the diagnostic workup as the patient presented with hyperthyroidism and diagnosed as multinodular goiter.

In conclusion, insular carcinoma is a rare disease with not well recognized clinical course in literatures. It may present with long history of signs and symptoms of hyperthyroidism. Total thyroideectomy is the main line of management.

Informed consent

Informed consent has been taken from the patient for publication of this work.

Conflict of interest

There is no conflict of interest to be declared.

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Consent

Consent has been taken.

Author contribution

Abdulwahid M. Salih: Surgeon performed the operation and follow up. Fahmi H. Kakamad: writing the manuscript and follow up. Han Aube: Examining the specimen, follow up.

Guarantor

Fahmi Hussein kakamad.

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