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CASE REPORT

Late recurrent papillary thyroid carcinoma presenting as thymoma

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DOI: 10.21040/eom/2018.4.4.1

Recived: August 8th 2018
Accepted: October 16th 2018
Published: November 12th 2018

Funding: None.

Conflict of interest statement: The authors declare that they have no conflict of interest.

Data Availability Statement: All relevant data are within the paper.

Abstract

Papillary thyroid carcinoma is the most common thyroid tumor, although generally rare. It has a good outcome because it rarely metastasizes, and if it does, it is characterized by local spread. Recurrence is also infrequent and usually occurs within three to five years after primary tumor treatment, although late relapses have been reported sporadically. Thymoma is another rare neoplasm. As it has a typical radiological presentation in the upper anterior mediastinum, biopsies are unnecessary because it is unlikely that the postoperative pathohistological finding will be surprising. We describe a rare case of a late recurrent papillary thyroid carcinoma presenting as a mediastinal mass, twenty-one years after the primary carcinoma. Because of the long interval between thyroid carcinoma treatment and appearance of the new tumor mass, with characteristic features, thymoma was suspected. However, the postoperative pathohistological findings revealed that the tumor was a papillary thyroid carcinoma metastasis, probably originating from a mediastinal lymph node. To our best knowledge, this is the second reported case of this kind.

Key words: papillary thyroid carcinoma, thymoma, mediastinal cyst, late recurrent

1. Introduction

Papillary thyroid carcinoma is an indolent tumor with a favorable prognosis and a 5-year survival of 98% [1]. Histologically, it is the most common type of thyroid tumor, although thyroid tumors are generally relatively rare [1]. Papillary thyroid carcinoma rarely metastasizes to distant sites [1], mostly spreading into the central and lateral lymph nodes of the neck, which are usually already involved at the time of diagnosis [2]. The mediastinal lymph nodes may be involved as well, although this occurs in less than 3% of affected patients [2]. Within three to five years of primary tumor treatment, there is a relapse in only 1.4% of patients [3]. The tumor usually recurs locally, in the thyroid fossa or the cervical lymph nodes.

The most common tumors of the upper anterior mediastinum are thymomas, followed by lymphomas, and rarely, retrosternal thyroid goiters, locally invasive thyroid tumors, and germ cell tumors [4]. In most cases, tumors of the thymus gland are thymomas, and rarely, carcinomas or neuroendocrine tumors [4]. Other tumors, such as primary thymic lymphomas, lipomas, liposarcomas, and others, occur only sporadically [5].

Although thymomas are the most frequent thymic tumors, in general, they are rare. It is estimated that they make up less than 1% of all tumors in adults, and are even rarer in children [4,5]. They may have a solid or solid-cystic structure. In some cases, they are entirely cystic, which may be a problem in the differential diagnosis of mediastinal cysts, since cysts, whether congenital or acquired, frequently develop in this location [6]. As a rule, they do not cause any problems, except in cases when they grow enough to exert pressure on the surrounding structures. If they are asymptomatic, they need not be surgically removed; this is why it is essential to distinguish a cyst from a cystic neoplasm [6].

Thymomas rarely metastasize; however, they have a strong tendency to spread locally [7]. Currently, it is believed that tumors of the upper anterior mediastinum should not be biopsied if they present as typical thymomas on radiological examination [4]. Only in the case of an inoperable tumor or a diagnostic dilemma should a preoperative tumor biopsy be performed [4].

Here we present a very rare case of a late recurrent papillary thyroid carcinoma which developed twenty-one years after surgical treatment of the primary carcinoma, and presented as a cystic-solid mediastinal tumor on radiology. Due to the long time lapse after treatment of the thyroid carcinoma as well as the typical radiological presentation, thymoma was suspected.

2. Case Report

A 68-year-old male was admitted to our department due to a newly detected expansive mediastinal growth. At the age of 47 he underwent surgery for a papillary thyroid carcinoma with cervical metastases. A total thyroidectomy with paraglandular neck dissection was performed, followed by postoperative radioiodine ablation. The patient was followed up for three years and there were no signs of disease recurrence. He then stopped attending follow-up visits, but continued taking thyroid hormone replacement therapy and vitamin D. Two years after his thyroid surgery, he operated varicose veins in his left lower leg. Two to three years prior to his first admittance to our department, he underwent routine colonoscopy during which a benign polyp was removed. At the same time, he was diagnosed with atrial fibrillation and was started on anticoagulant therapy. Prior to his admission in our hospital, he was surgically treated for chronic bilateral venous insufficiency in his legs.

Three weeks prior to being admitted to our department, he was examined by a nephrologist due to frequent urination, and was diagnosed with renal insufficiency and hypercalcemia. An ultrasound examination confirmed normal kidney size. It was concluded that the hypercalcemia was iatrogenic, caused by uncontrolled vitamin D intake, which also resulted in renal insufficiency. Vitamin D was discontinued and the patient was treated symptomatically. Subsequently, a chest radiograph revealed mediastinal infiltration (figure 1a and 1b), and a computed tomography (CT) scan of the mediastinum and upper abdomen was performed, which showed an 80 mm neoplastic process in the upper anterior mediastinum (figure 2). The infiltration had a heterogeneous structure, composed of solid and cystic parts, was well differentiated from the surrounding structures, and highly suspicious for a tumor of the thymus. Additionally, a number of small lung nodes of

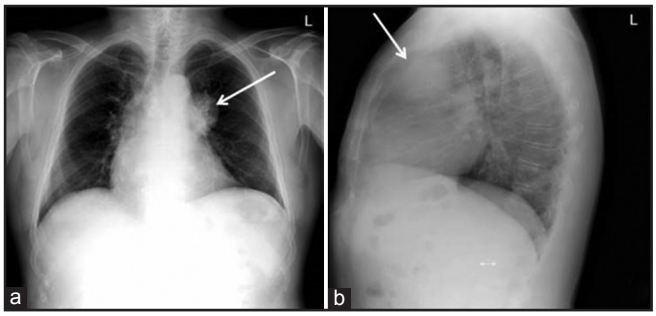

Figure 1. a. Chest radiograph in the anterior posterior (AP) view: infiltration in the upper anterior mediastinum (marked by the arrow) b. Chest radiograph in the profile view: infiltration in the upper anterior mediastinum (marked by the arrow)

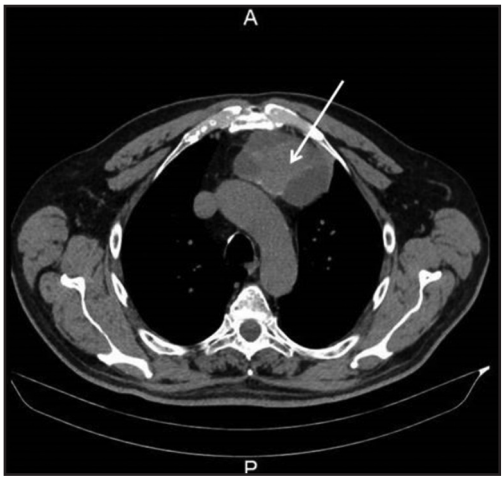

Figure 2. Computed tomography (CT) scan of the thorax: cystic-solid infiltration in the upper anterior mediastinum (marked by the arrow)

several millimetres in size were found, but they were too small for further characterization. No other pathological findings were revealed. The patient was transferred to the Department of Respiratory Diseases for further workup.

At the time of admission, the patient was asymptomatic, since his only symptom, urinary frequency had resolved. He was in good condition and his physical examination was unremarkable. A routine laboratory workup revealed anemia of chronic disease, renal insufficiency, and hypercalcemia. During his stay at our department his serum calcium returned to normal, creatinine levels decreased to almost normal, and the anemia was partially corrected. In consultation with a thoracic surgeon and a radiologist, it was concluded that the patient most probably had a thymoma. Following preoperative workup, the patient was operated several days after his transfer.

The postoperative pathohistological finding revealed that the mass was a papillary thyroid carcinoma metastasis manifested as a multilocular blood-filled cyst. The metastasis site was probably a lymph node, because borderline remnants of tissue resembling a lymph node were found. The patient's postoperative recovery was uneventful, and his performance status was still good. The 2-month follow-up CT scan of the thorax revealed no new infiltrations, however, an enlarged neck lymph node was found on ultrasound. At that time, the patient did not agree to the suggested lymph node fine needle aspiration (FNA), but agreed to ultrasound surveillance and follow-up. Although scintigraphy did not show any pathological radioiodine accumulation, due to the small nodes that had been found in the lung, another course of radioiodine therapy was recommended, which the patient received seven months after his surgery. Since follow-up ultrasound revealed further neck lymph node enlargement, a right selective dissection of the neck was performed and lymph node metastases were confirmed in three out of the twenty-eight removed nodes. Upon treatment completion, the patient has been undergoing regular follow-up examinations and so far, has had no signs of recurrent disease.

3. Discussion

Papillary thyroid carcinoma generally does not metastasize to distant sites. Due to its slow growth, late recurrent disease has been reported after two or more decades, the longest recorded period between primary tumor surgery and recurrent disease being forty-three years [8]; however, these are rare cases. Retrospective analyses of large patient groups followed for forty years, reveal that in most cases relapses occur in the first few years after treatment, and most of the recurrent disease affects the neck area [9]. What makes this case interesting is not only the fact that the disease recurred after more than twenty years, but also that it manifested with a very unusual radiological picture.

A literature search yielded one similar case, where the disease recurred in the mediastinum six years after surgical removal of the primary thyroid tumor, and the radiological picture suggested a thymoma [10]. In contrast to that case, where the mediastinal metastasis developed several years after primary tumor treatment, in our patient

the late recurrent disease presented as another malignant disease. Although there is no consensus on the need for long-term follow-up of papillary thyroid carcinoma patients, it has been suggested that for some patients it is necessary [3].

4. Conclusion

In spite of the advancements in medicine and numerous novel diagnostic procedures, information obtained from the patients themselves is still essential. The fact that a patient was successfully treated for a malignant disease in the past is important not only because late recurrent disease is always possible, but also because the incidence of secondary neoplasms in patients who had a malignant disease is significantly higher.

5. Author's Contributions

GD gave the idea for the case, collected the data concerning patient medical history and radiological images and wrote the manuscript. BB searched the literature and participated in manuscript drafting. SKK collected the data concerning patient medical history and participated in patient follow-up. SK was involved in the conception of the case report, revised the draft of the article and critically reviewed the manuscript. All authors provided critical feedback and approved the final article.

6. Acknowledgement

We wish to thank Aleksandra Zmegac Horvat, University of Zagreb School of Medicine, for language editing the text.

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