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Case Report

## A Rare Case of Primary Thyroid Lymphoma: Case Report and Literature Review

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### ABSTRACT

**Introduction:** Primary lymphomas of the thyroid gland are rare and very uncommon, representing less than 5% of thyroid neoplasia and occur more frequently in women than in men, it develops in most cases on a pre-existing thyroid condition, in particular Hashimoto's thyroiditis.

Case presentation: we report the case of a 65-year-old patient, with no history of chronic thyroiditis, admitted for a rapidly progressive, hard and fixed anterior cervical mass, accompanied by signs of compression. Once in our structure after a radiological examination (ultra sound and CT scan), the patient was scheduled for a surgical management. However, during surgery, invasion of adjacent thyroid tissue was noted, justifying partial tumor reduction. Definitive pathological examination, supported by immunohistochemistry, confirmed the diagnosis of thyroid lymphoma. Treatment consisted of exclusive chemotherapy. The clinical evolution was very favorable.

**Conclusion:** The key messages to remember are that thyroid lymphoma does not occur exclusively in patients with Hashimoto's disease, and if adjacent tissue invasion is observed during surgery, a tumor reduction or a simple biopsy is sufficient for diagnosis. Additionally, exclusive chemotherapy yields highly impressive results.

Keywords: Primary thyroid lymphoma; Chemotherapy; Immunohistochemistry; Chronic thyroiditis

#### Introduction

Primary lymphomas of the thyroid gland are unusual and very uncommon. They account for around 5% of thyroid tumors and 2% of extra-nodal lymphomas<sup>1-3</sup>.

Its annual incidence is estimated at 2 per 1 million inhabitants, most often affecting patients with a median age of 60, with a predominance of females, the sex ratio being  $3:1^4$ .

Patients with Hashimoto's thyroiditis have an increased risk of developing this disease compared to patients without thyroiditis, with a relative risk of  $67^5$ .

There are 70% cases of diffuse large B-cell lymphoma, followed by 10 to 23% of MALT (mucosa-associated lymphoid tissue) lymphoma<sup>7</sup>. The most typical form is a rapidly growing cervical mass, often adherent to adjacent tissues, which becomes compressive in about a third of cases<sup>6</sup>.

The diagnosis of certainty is based on histology, and the therapeutic course of action depends essentially on the histological type and stage of the tumor.

Surgical indications, previously predominant, have been restricted with the advent of new chemotherapy protocols which have become the standard treatment for thyroid lymphoma.

#### Observation

Its A 65-years-old female patient, with no significant medical history or known thyroid issues, who was referred to us for a lower anterior cervical swelling rapidly increasing in volume evolving since 4 months, associated to a dyspnea to the effort and dysphagia to solids, without dysphonia or any other associated signs, all evolving in a context of alteration of the general state.

The physical examination revealed a large goiter, painless, with a non-palpable lower edge, hard and fixed to the deep plane, with an irregular surface, but without any inflammatory-looking or notion of lymphadenopathy.

The Cervical ultrasound performed showed a heterogeneous goiter depending on the right lobe and isthmus without any visible nodule.

A cervicothoracic CT scan was ordered (because of the strong suspicion of anaplastic carcinoma), which showed a general increase in the size of the thyroid with multiple nodules, especially in the right lobe slightly plunging towards the upper mediastinum and pushing the adjacent tissue structures, moreover, the examination did not reveal any pulmonary involvement or cervico-mediastinal lymphadenopathy (**Figure 1**).





**Figure 1:** Axial CT images showing: an enlarged thyroid gland extending beyond the cervicothoracic contour

Biological tests were normal, in particular the TSH us level, which was 5 mUI/l.

Intraoperatively, the thyroid was clearly suspicious of malignancy, with massive invasion of the trachea and larynx. The 2 recurrent nerves were engulfed by the tumour, and surgery was stopped after removing a part of the right lobe of the thyroid (**Figures 2,3**).

Postoperatively, the patient presented respiratory distress for which she benefited from a tracheotomy on day 4 (Figure 4).

The definitive histopathological examination showed the presence of a diffuse cellular infiltrate with round cells (lymphoma?) and the complementary immunohistochemical study revealed a diffuse large cell B lymphoma.

Two months after surgery, the patient was referred to the clinical hematology department with the diagnosis of HL where she benefited from treatment with chemotherapy according to the CHOP protocol (Cyclophosphamide, Doxorubicin, Vincristine and Prednisone). The clinical evolution was favorable from the first course of chemotherapy, with obvious regression of the thyroid tumor volume. The patient's control nasofibroscopy showed that both voice cords were mobile enough and had a normal appearance, allowing for a successful decannulation.



Figure 2: Image the thyroid gland intraoperatively (after tumor reduction).



Figure 3: Operative piece of the thyroid gland.



Figure 4: Image of the neck after tracheotomy.

#### Discussion

Thyroid lymphomas are extremely rare, whether primary or secondary<sup>8</sup>.

They represent approximately 5% of all thyroid cancers9.

Their incidence is estimated at 1.1 to 2.06 per million inhabitants per year, this incidence has seen a clear increase in recent years<sup>10,11</sup>. However, the incidence of this disease is not yet documented in Morocco.

Patients are often female and generally consult during the sixth or seventh decade, presenting compressive symptoms such as dyspnea, and dysphagia associated with general symptoms such as weight loss, sweating or fever in 10% of cases<sup>12</sup>.

Apart from dysphonia, all these symptoms were reported by our patient during his first consultation. The consultation time can vary from a few days to 36 months<sup>13</sup>.

This disease is closely associated with Hashimoto's thyroiditis<sup>5</sup>, this association was observed in 25 to 100% of cases according to published studies<sup>14-16</sup>.

Different theories have been put forward to explain the HT/PTL association, it has been proposed that chronic and continuous stimulation of lymphocytes by antigens could lead to lymphocyte proliferation whose mutations lead to malignant differentiation causing lymphoma in the thyroid gland. which is normally a gland devoid of lymphoid tissue<sup>17,18</sup>.

The clinical picture is characterized by a rapidly growing thyroid tumor, leading to compressive signs in 20 to 25% of cases<sup>19</sup>.

This goiter can sometimes be adherent to surrounding tissues, and the presence of pain would strongly support the diagnosis; satellite cervical lymphadenopathy is observed in 20% of cases<sup>20,21</sup>.

Biological hypothyroidism is present in almost 40% of cases, although its clinical expression is rare<sup>20</sup>.

The diagnosis is confirmed in 61% of cases by fine aspiration<sup>22,23</sup>, it helps to differentiate lymphoid proliferation from epithelial tumor<sup>6</sup>. Ultrasound-guided biopsy has also demonstrated higher diagnostic accuracy because it can obtain more tissue than fine-needle aspiration cytology and thus distinguish between Hashimoto's thyroiditis, thyroid lymphoma and anaplastic carcinoma<sup>24</sup>.

Historically, surgery and radiation therapy were the standard treatments for primary thyroid lymphoma. Before retrospective studies demonstrated that LT is sensitive to chemotherapy and radiotherapy.

The interrogation, the clinical examination, the biological and radiological explorations in our patient all pointed towards a strong suspicion of anaplastic carcinoma so the cytopuncture was not carried out in our patient and the discovery of tumor invasion of adjacent structures was only noted intraoperatively, necessitating interruption of the surgical procedure after achieving tumor reduction while preserving essential structures such as recurrent nerves, while awaiting the results of the definitive histology.

#### Conclusion

Thyroid lymphoma is a rare disease often neglected and

underdiagnosed, requiring increased vigilance when observing any goiter or thyroid nodule rapidly increasing in size with signs of compression. This will enable early suspicion of the diagnosis and potentially avoid excessive surgery for the patient.

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