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Primary and isolated thyroid Hodgkin's lymphoma: A case report



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ABSTRACT

INTRODUCTION: Hodgkin's lymphoma rarely involves the thyroid gland. It is typically presented as a fast growing neck mass that is sometimes accompanied by respiratory compression symptoms.

CASE REPORT: We report one of the few (the seventeenth) case of primary and isolated Hodgkin's thyroid lymphoma presented by a 65 years old man, consulting for a fast growing neck mass with Hodgkin's symptoms. The patient had total thyroidectomy and short courses of chemotherapy, then total resolution of symptomatology.

CONCLUSION: Most thyroid Hodgkin's lymphoma are presented by women, rarely man, isolated and primary. Since 1962, we only found sixteen cases described in the literature. Hodgkin's lymphoma should be considered in the differential diagnosis of patients with a thyroid mass for rapid management.

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1. Introduction

Primary lymphoma of the thyroid gland is a rare tumour, with no clinical and paraclinical specificities, accounting for only 5% of thyroid malignancies and 2% of extranodal lymphomas [1]. Hodgkin's lymphoma rarely involves the thyroid gland.

The diagnosis is histological. Treatment is based on chemotherapy, monoclonal antibody and radiotherapy. The surgery must be avoided when the diagnosis can be obtained before or during the intervention, but thyroidectomy must be done and it is the main way to get healing in association with chemotherapy with or without radiotherapy. Only rare cases of Hodgkin's lymphoma presenting in the thyroid have been reported in the literature [2].

We report the case of a 65 years old man consulting in our ENT department, 20 August 1953 Hospital, Casablanca Morocco for primary and isolated thyroid Hodgkin's lymphoma.

This work has been written in accordance with the SCARE criteria [3].

2. Case report

We report the case of a (It is about) 65 years old man, living in Casablanca, Morocco, with no medical or surgical history, admitted in our ENT department for an anterior and medial cervical tumefaction,

which started growing 8 months ago, rapidly increasing in volume without pain. The other symptoms were general pruritus, night sweats and fatigue, with no fever, no emaciation, no dyspnea, no dysphonia or thyroid gland disorder. There were no similar cases reported in the patient's family. The palpation found a hard tumefaction, and no palpable cervical lymph nodes. The general physical examination didn't find any hepatomegaly, or splenomegaly or other clinically palpable lymph nodes in the body. Blood count cells showed a disorder of lymphocytes that were slightly increased. Accelerated sedimentation rate. Cervical and thoracic CT scans were done showing a tissue mass of the right thyroid lobe dipping to the anterior and middle mediastinum. Thyroid fine needle aspiration was performed before thyroidectomy. It contained some atypical cells, raising the possibility of Hodgkin's lymphoma. A total thyroidectomy was decided. The patient was operated without incident, with a good postoperative warning, without dysphonia or dyspnea by trauma of the laryngeal recurrent nerves. The patient is under Levothyroxine sodium 100 µg per day. All the symptoms of the patient have completely disappeared after thyroidectomy. The histological study showed a scleronodular Hodgkin's lymphoma confirmed by the immunohistochemical study which bring out a strong and diffuse positivity of the tumor cells to the anti-CD-15 and anti-CD20 antibodies. It is classified I B (I for the involvement of a single lymph node region, so thyroid gland, and B for the presence of systemic symptoms).

The patient was referred to the haematology department for further treatments. The postoperative course was uneventful and the patient began chemotherapy treatment including four cycles of combined cyclophosphamide, doxorubicin, prednisone and vincristine. Finally, surgery and chemotherapy realized the stable cure

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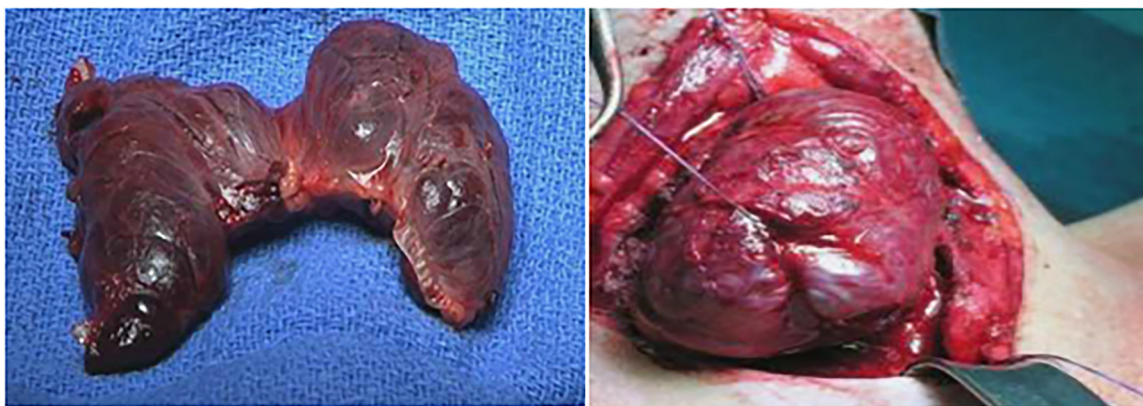


Fig. 1. XXX.

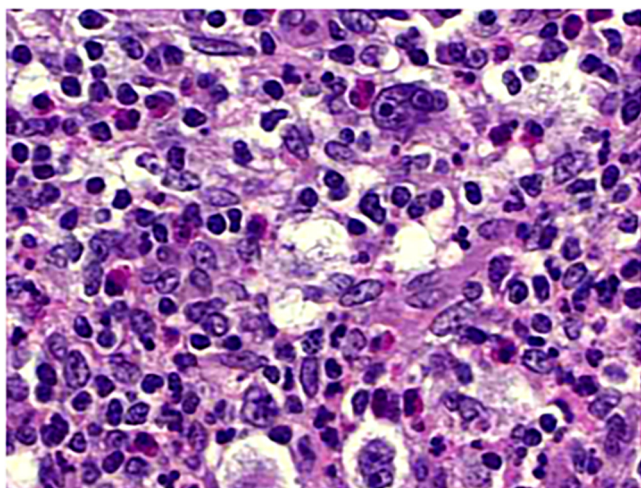


Fig. 2. XXX.

of the disease and the patient is alive after two years without recurrence or metastases (Figs. 1 and 2).

The ABVD (Dacarbazine, Bleomycin, Vinblastine and Doxorubicin) protocol was established, four courses were done. PET scan, cervicothoracoabdominopelvic scan is completely normal in postoperative, and the blood count is balanced with a normal lymphocyte rate.

3. Discussion

Thyroid carcinoma is the most common endocrine malignancy however primary thyroid lymphoma (PTL) accounts from only 1–5% of all thyroid malignancies. B-cell type non-Hodgkin lymphoma (NHL) is a frequently described type of PTL, while Hodgkin's and T-cell lymphoma are rare [1]. Thyroid lymphoma typically presents with a rapidly enlarging neck mass leading to compressive symptoms [4]. However, primary thyroid lymphoma develops in only 0.5% of all cases of Hashimoto's thyroiditis [5]. Due to this underlying risk factor, primary thyroid lymphoma typically occurs more often in women than men (8:1) and usually later in life (sixth or seventh decade) [6].

In this study, it is about a man, which is making the case more interesting and rare.

And this patient present scleronodular Hodgkin's lymphoma and it is rarely described in the literature.

The lesion having an eventual history of Hashimoto's thyroiditis (HT) appears as a more or less rapidly enlarging anterior cervi-

cal mass associated or not with lymphadenopathy which in time add symptoms related to compression such as hoarseness, dyspnea and dysphagia. In our case, no known antecedent of thyroiditis. Patients with a background history of chronic thyroiditis has a 67- to 80-fold greater risk factor to develop PTL than those without this inflammatory process [7].

Similar to other lymphomas, subtypes in thyroid lymphoma are classified according to histological and immunological features. The thyroid gland contains no native lymphoid tissue; intrathyroidal lymphoid tissue can develop in various pathological conditions, but most commonly occurs in the setting of autoimmune thyroiditis. This acquired lymphoid tissue bears a close resemblance to mucosa-associated lymphoid tissue and can evolve to an extranodal marginal zone B-cell lymphoma [8]. The development of extranodal marginal zone B-cell lymphoma in the thyroid gland is often characterized by an indolent course, but transformation to an aggressive lymphoma can also occur [8]. In contrast, any association between Hodgkin's lymphoma and underlying thyroiditis has been difficult to document because of the small number of cases.

A review of the English literature between 1962 and 2005 revealed 16 cases of thyroid Hodgkin's lymphoma, with a female preponderance and generally favourable outcome similar to our case [9], patients with Hodgkin's lymphoma commonly presented with a rapidly enlarging thyroid gland as our case, or a thyroid mass, similar to the presentation of non-Hodgkin's lymphoma of the thyroid. The mass may cause symptoms related to compression or infiltration of the surrounding neck organs. Symptoms reflecting airway or esophageal obstruction occurred in 9/16 of the previously reported cases, but this signs were not reported by the patient.

On physical examination, the thyroid mass was commonly described as being hard upon palpation (Table 1).

Ultrasonography is usually the first imaging modality performed in the evaluation of a thyroid mass. Previous studies have categorized ultrasound findings into three categories: diffuse, nodular or mixed. It has significant overlap with anaplastic thyroid cancer [10–12]. The presence of significant internal vascularity and absence of calcifications may be distinguishing features between thyroid lymphoma and anaplastic thyroid cancer on ultrasonography [13].

The diagnosis of Hodgkin thyroid lymphoma is often postponed by its prolonged indolent evolution which does not always appear clear. Although FNA has become the procedure of choice for the diagnosis of any thyroid tumor it has yielded mixed results asserting the presence of MALT so that core or open biopsy and even surgical excision (as in our case) is decisive for diagnosis [14].

Combined pathology and immunohistochemistry may specify microscopy of these lesions orienting therapeutic planning and predicting prognosis of the patients. [15]

Table 1
Clinicopathologic features of previously reported cases of thyroid Hodgkin's lymphoma.

Authors	Age (yrs)/sex	Presenting symptoms	Imaging/Intraoperative findings	Pathologic findings			Stage	Treatment/Outcomes
				Evidence of HL	HL subtypes	Uninvolved thyroid		
Rupp et al.	64/M	Neck pain, dysphagia and B symptoms	Diffuse firmly enlarged thyroid	Image, description, LN Biopsy	Not reported	Lymphocytic Thyroiditis	IIE	Unknown
Roberts et al.	61/M	Painful enlarging neck mass	Firm tumor mass extending into mediastinum and invading trachea and carotid sheath	Image, description	NS	Hashimoto's Thyroiditis	IIE	Subtotal neck mass
Gibson et al.	59/F	6 years history of painless progressive neck enlargement	Soft tissue swelling with tracheal deviation	Image, description, LN Biopsy	NS	Lymphocytic Thyroiditis	IIE	thyroidectomy then XRT, NED at 6 mo.
De Beats et al.	57/F	6 years history of goiters, with rapid painful enlargement of thyroid	Soft-tissue swelling	Image, description	NS	Not described	IIE	Subtotal thyroidectomy, no other therapy, Ned at 6 mo.
Feigin et al.	64/F	2 years history of small thyroid nodule and hypo thyroidism, hoarseness and weight loss	No mediastinal lymphadenopathy or lung disease	Image, description	NS	Lymphocytic thyroiditis	IIE	Thyroid lobectomy, chemo and XRT, NED at 3 years.
Kugler et al.	27/F	Slow thyroid enlargement over 1 yr, stridor, dysphagia and hoarseness over 3 yr.	Thyroid mass contiguous with mediastinal mass. Tracheal deviation and narrowing	Image, description	NS	Not described	IIIE	Subtotal resection and chemo, no FU
Mate et al.	60/F	1 yr history of hypothyroidism, then dysphagia and hoarseness	Mass involving cricoid cartilage, oesophagus and mediastinum.	LN biopsy	MC	Not described	IIE	XRT alone, recurred in lung after 6 mo, treated with chemo
Mate et al.	25/F	Enlarging goiter and dysphagia	Thyroid mass compressing trachea and displacing oesophagus	LN biopsy	NS	Not described	IIE	XRT alone, recurred 2 yrs later in abdomen, treated with chemo, NED at 7 yr.
Smith et al.	19/F	5 yr history of goiter, then dysphagia, dyspnea and rapidly enlarging thyroid	Firm thyroid adherent to trachea and oesophagus	Description	NS	Not described	IIE	Unknown
Granados et al.	36/F	1 yr history of goiter, hypothyroidism. Increase in size and firmness despite	Bulky mediastinal mass, infiltrating thyroid with tracheal narrowing.	RS cells CD15+	NS	Not described	IIE	XRT alone, NED at 1 yr.
Vailati et al.	29/F	2 mo history of thyroid enlargement, stridor, low-grade fever, fatigue and pruritus.	Homogeneous mass involving isthmus and left lobe, with tracheal displacement	Image, description	NS	Not described	IE	Subtotal thyroidectomy, and XRT NED at 2 yr.

Jayaram	53/F	Neck swelling for 2 mo, then cervical lymphadeno-pathy, and hepatospleno-megalie	6 cm firm nodule in left lobe, no mediastinal or hilar lymphadenopathy	description, LN biopsy	Not reported	Not described	IV	thyroid lobectomy, lost to FU.
Hardoff et al.	20/F	1 yr history of solitary painless thyroid nodule, then developed cervical lymphadenopathy.	cervical, axillary and mediastinal lymphadenopathy.	RS cells CD15+, CD30+.	NS	Not described	II E	Chemo and XRT, NED at 1 yr.
Hardoff et al.	18/F	Painless throat fullness, dysphagia, fever.	Solitary left thyroid nodule, mediastinal and cervical lymphadenopathy.	RS cells CD15+, CD30+.	NS	Not described	II E	Chemo and XRT, NED at 18 mo.
Luboshitzky et al.	19/F	Single nodule in left lobe of thyroid.	5 cm mediastinal mass and an enlarged cervical lymph node	RS cells CD15+, CD30+.	NS	Not described	II E	NED at 2 yr.
Nakamura et al.	18/M	Progressively enlarging neck mass and dyspnea	Large mass involving entire thyroid extending into mediastinum	RS cells CD15+, CD30+.	NS	Not described	II E	XRT alone, NED at 4 yr.
Tatari et al.	65/M	8 mo rapidly cervical mass increasing, painless, general pruritis, night sweats, fatigue	tissue mass of the right thyroid lobe, dipping to the anterior et middle mediastinum	Image, description	NS	Not described	II E	Totale thyroidectomie and chemo.

Yr = year; mo = month; wk = week; HL = Hodgkin's lymphoma; LN = lymph node; RS = Reed–Sternberg; NS = nodular sclerosis subtype; MC = mixed cellularity subtype; XRT = radiation therapy; NED = no evidence of disease; FU = follow-up; chemo = chemotherapy.

Hodgkin's lymphoma is characterized by the presence of Reed–Sternberg cells. This is consistent with the well-recognized tendency for scleronodular subtype to occur in the mediastinum and head and neck region [16]. In many cases, the associated fibrosis and sclerosis were more pronounced within the thyroid gland in comparison to the adjacent nodal areas, mimicking the fibrosclerosing variant of Hashimoto's thyroiditis or in some cases Reidel's thyroiditis, and requiring careful histologic examination and immunohistochemical analysis to make the diagnosis of Hodgkin's lymphoma.

Most of the thyroid Hodgkin's lymphoma patients presented with low-stage disease responded to chemotherapy with or without radiation therapy and had a favorable clinical outcome. Surgical intervention is rarely required in the treatment of nodal Hodgkin's lymphoma. However, in thyroid Hodgkin's lymphoma, some patients presented with symptoms that compromised their airway or caused severe pain, requiring surgical therapy to palliate their symptoms. Surgical intervention did not appear to be associated with adverse outcome in thyroid Hodgkin's lymphoma patients, similar to thyroid non-Hodgkin's lymphoma patients [17].

Our case highlights the value of total thyroidectomy in thyroid Hodgkin's lymphoma for clinical and haematological resolution.

4. Conclusion

Since 1962, only sixteen cases appear to have been described in the literature, that show the interest of the presented case.

The particularity of this case is the fact that it is about a man, and the isolated and primary localisation in the thyroid gland. All symptoms have disappeared after total thyroidectomy and few cures of chemotherapy. No recurrence was noted after a two years follow-up.

Conflicts of interest

The authors declare having no conflicts of interest for this article.

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Ethical approval

Written informed consent for publication of their clinical details and/or clinical images was obtained from the patient.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying image.

Author contribution

Mohammed Moutaa Tatari: Corresponding author writing the paper

Said Anajar: writing the paper

Reda Abada: study concept

Sami Rouadi: study concept

Mohammed Roubal: correction of the paper

Mohammed Mahtar: correction of the paper

Registration of research studies

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Guarantor

DR Tatari Mohammed Moutaa.

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