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An Unexpected Diagnosis of Plasmacytoma of The Thyroid: A Case Report and Review of The Literature

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Abstract

Plasmacytoma is a malignant proliferation of plasma cells that can arise in the bone marrow (osseous) and in the soft tissues. Extramedullary plasmacytomas most commonly occur in the upper respiratory tract. Thyroid involvement is rare. In this report, we describe a new case plasmacytoma of the thyroid gland in a 60-year-old man with a history of multinodular goiter present for several years and an unexpected rapid increase of the mass associated to dysphagia, dyspnea, and dysphonia. Histopathological examination of surgical specimen, along with immunohistochemical studies, unexpectedly led to the diagnosis of plasmacytoma.

We also briefly summarize the last 30 years of literature on thyroid plasmacytoma and review the salient characteristics of this rare condition.

Keywords: Plasmacytoma, Thyroid

Introduction

Plasma cells are an unusual finding in the thyroid. The malignant proliferation of this cells usually arises in the bone marrow (osseous) or in the soft tissue (extramedullary).

Plasmacytomas can occur in the thyroid gland either as primary lesions (Solitary extramedullary plasmacytoma- SEP) or secondary to systemic spread of multiple myeloma [1]. These lesions have often been associated with Hashimoto's thyroiditis and with chronic lymphocytic thyroiditis. These tumours usually mimic other malignant lesions such as papillary thyroid carcinoma and anaplastic thyroid carcinoma.

The peak of incidence of this type of neoplasm have been reported mostly in men in the sixth decade of life.

In this case report, we describe a case of an unexpected diagnosis of solitary extramedullary plasmacytoma of the thyroid.

Case Report

A 60-year-old man was referred to the Spedali Civili of Brescia, Italy in March 2021 with a history of multinodular goiter present for several years and a rapid increase of the mass in the last three months. This enlargement of the mass was associated to dysphagia, dyspnea, and dysphonia. No cervical lymphadenopathy was reported. Clinically and biochemically, the patient was euthyroid.

A thyroid ultrasound examination revealed a nodule affecting the right thyroid lobe (37x20x37 mm) previously reported as TIR3a in a multinodular goiter.

Due to the severe symptomatology, the patient was prepared for surgery. All routine investigations, including complete blood count, liver function test and renal function test, were within normal limits.

Trachea x-ray showed tracheal aerial lumen substantially in axis, with slight reduction of caliber to the middle third for imprint on the side and rear profiles. For this reason, a fibre-optic-laryngoscopy was performed, showing an anterior left-convex septal deviation, free nasopharynx, norm conformed pharyngeal axis in the absence of suspicious lesions. Normal larynx. Arytenoids' motility in the norm bilaterally. At the anaesthesiologic evaluation no contraindications to surgical intervention were detected. The patient was carefully informed of the risks (haemorrhages, vocal cord paralysis, infections, temporaneous/definitive tracheostomy, hypoparathyroidism).

Therefore, the patient was submitted to total thyroidectomy and no enlarged lymph nodes were noted intraoperatively. The intraoperative neuromonitoring of recurrent laryngeal nerves recorded normal function of the laryngeal nerves.

At the end of the surgery, immediately after extubating, the patient developed respiratory distress with stridor, requiring reintubation with an urgent ENT assistance. A closed glottis with bilateral chordal paralysis was detected at the fibro-optic-laryngoscopy.

Surgical wound was reopened, and the interior face of the trachea exposed. A Bjork flap tracheostomy was performed with an intubation through the stoma with a cannula Rusch [7,5].

In the subsequent post-operative period, the patient was evaluated several times by otolaryngologists and speech therapists for post-operative dysphonia and the management of the tracheostomy, re-education to phonation and re-feed. The last otolaryngological evaluation documented a regained motility of both the vocal cords with a subsequent closure of the tracheostomy. In consideration of the size of the multinodular goiter, the bilateral paralysis was likely caused by "stupor" of the vocal cords.

The histopathological examination of the surgical specimen revealed an irregularly enlarged thyroid gland, a nodular formation of 6x3.5x2.8 cm in the right lobe. The parenchyma of both lobes shows numerous confluent nodular formations, most of adenomatous appearance, the greater localized at the level of the left upper pole and upper-middle third, measuring 5.2x4.2x3.5 cm.

Microscopic examination of histological sections showed a parenchyma largely replaced by neoplastic proliferation of a population of mature plasma cells that, at immunophenotypic investigations showed the following immunophenotype: positivity for CD38, CD138, MUM1, CD20, EMA and negativity for CD3, CD5, Cyclin D1 and a monotypic restriction for K light chains (Figure 1-4). Amyloid substance deposits positive for histochemical coloration of Congo Red were also found. The residual thyroid parenchyma showed atrophy of the epithelial component. Parathyroid glands were not found.

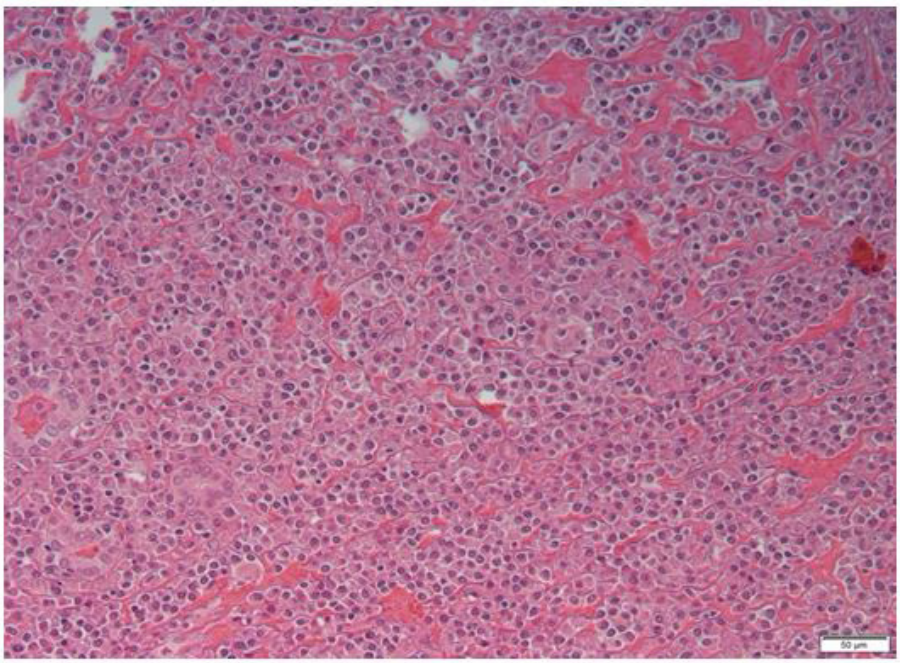

Figure 1: Thyroid parenchyma replaced by neoplastic proliferationconsisting of a population of mature plasma cells

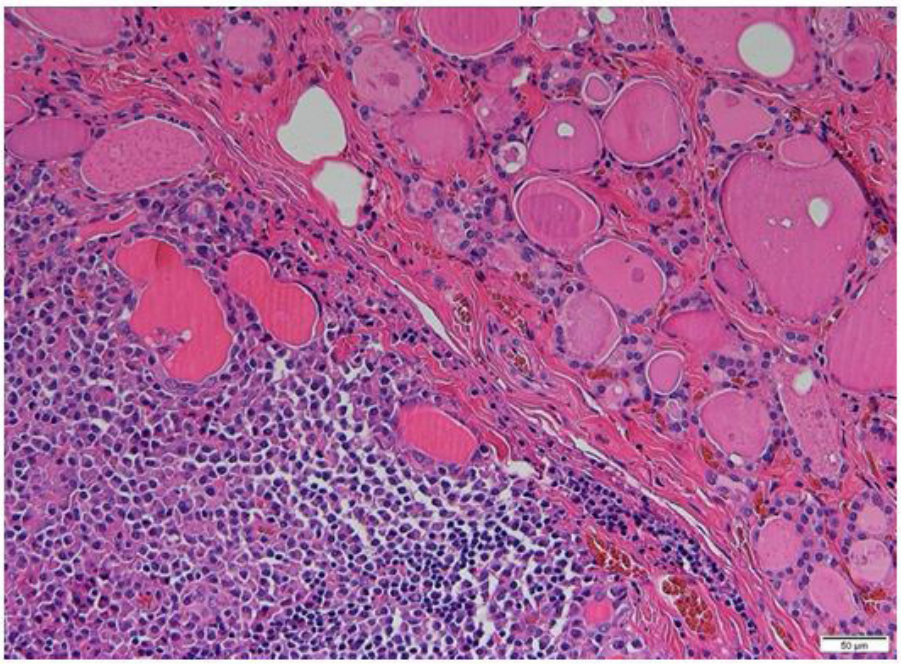

Figure 2: In the lower left corner there is neoplastic plasma cellular proliferation; in the upper right corner on the right there is thyroid parenchyma, consisting of flat cubic thyrocytes, with micro and mediumfollicular architecture

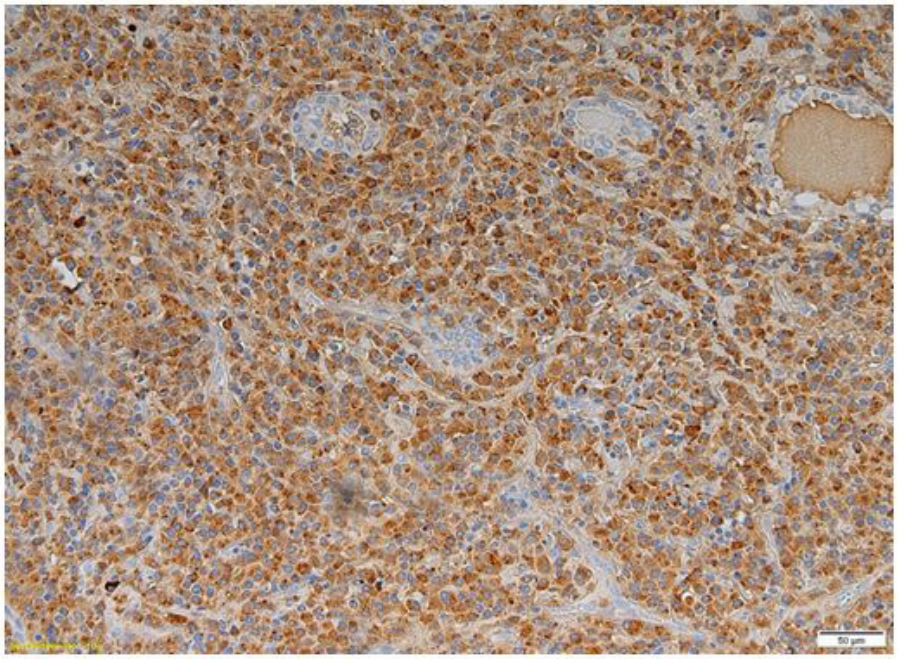

Figure 3: Immunohistochemical investigation showing monotypicrestriction for Kappa light chains

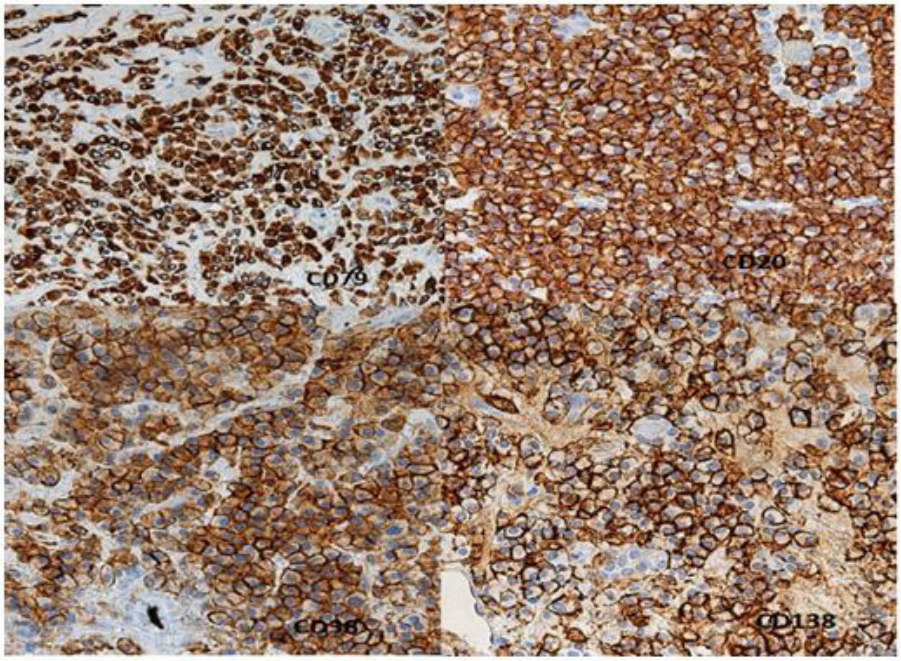

Figure 4: Cd38,138,20,79 EMA: Immunohistochemical survey positive forCd38,138,20,79 EMA

The morphological and immunophenotypic picture are indicative of mature plasma cell neoplasia with monotypic restriction K. Differential diagnosis between plasmacytoma and multiple myeloma was subject to clinical-instrumental evaluation of the patient. Ten months after discharge, the patient doesn't report sequelae of the surgery. Thyroid gland scintigraphy, Positron emition tomography (PET) and thoraco- abdominal CT scan was performed to confirm the involvement only of the thyroid. Therefore, the negativity of all these exams, the patient had no indication for further treatments.

Discussion

Thyroid gland is one of the rarest sites to be affected by Plasmacytoma. Plasma cell dyscrasias are a group of disorders that are characterized by expansion of a single clone of immunoglobulin-secreting plasma cells, with resultant increase in serum levels of single

complete or partial immunoglobulin. Those disorders are subdivided in different variants such as multiple myeloma, lymphoplasmacytic lymphoma, heavy-chain disease, monoclonal gammopathy and localized plasmacytoma (table 1). Localized plasmacytoma are further sub-classified into two groups: solitary skeletal plasmacytoma and extramedullary plasmacytoma (EMP) that involves soft tissue such as the thyroid. EMP represents less than 5% of all plasma cell neoplasms [2,3]. The most common location for EMP is upper respiratory tract and oral cavity. Most of their symptomatology is related to their specific location in the head and the neck. In Kapadia et al. series [4]; 80% of head and neck plasmacytoma presented with a mass, 35% complained airway compression. On the other hand, thyroid plasmacytoma usually presents with painless, firm, non-tender, mobile, multinodular mass with no associated cervical lymphadenopathy. One of the symptoms of presentation of the disease may be hoarseness of the voice [5,6]. Rapidly growing thyroid mass that brought the patient to seek medical advice such as in this case were already reported in different series. Patients usually are euthyroid or hypothyroid. The antithyroid antibodies may be elevated. Primary plasmacytoma of the thyroid, like malignant lymphoma, is often accompanied by evidence of autoimmune thyroiditis in the residual portion of the gland [7].

YEAR	AUTHOR	DIFFERENTIAL DIAGNOSIS AND BACKGROUND
1990	Chesyln-Curtis S, Akosa AB et al.	Multiple Myeloma
1994	Ohshima et al.	Primary thyroid plasmacytoma with Hashimoto's disease.
1994	Kovacs CS et al.	Solitary plasmacytoma arising from a thyroid with Hashimoto's disease.
1999	Hasegawa et al.	Malignant lymphoma followed by plasmacytoma in Hashimoto'sthyroiditis.
2000	Bourtsos EP etal.	Medullary carcinoma
2004	De Schrijver etal.	Rapidly enlarging thyroid mass in a patient with known Hashimoto'sdisease.
2006	Kuo et al.	Mass in the right thyroid lobe.
2007	Chaganti et al.	Rapidly enlarging palpable thyroid mass with Hashimoto's thyroiditis
2008	Ozkan et al.	Multiple Myeloma had a rapidly enlarging mass in the right thyroid lobe
2009	Avila et al.	Rapidly enlarging palpable thyroid mass.
2010	Fraser et al.	Hashimoto's thyroiditis
2010	Meccawy et al.	Hashimoto's thyroiditis with progresssively enlargin mass with pressure syntoms including change in voice dyspenea and dysphasia.
2010	Abdel Khalek etal.	Hoarseness, dyspnea, dysphagia and anterior neck discomfort.
2010	Kandil et al.	Plasmacytoma is a cause of diffuse thyromegaly
2011	Patten et al.	Hashimoto's thyroiditis in a Micropapillary carcinoma.
2011	Puliga et al.	Multinodular goiter
2011	Shahani et al.	Previous Diffuse large B cell lymphoma followed by a Plasmacytoma
2012	Yao CM et al.	this report discuss a case of Total thyroidectomy and neck lymph node dissection
2014	Hassan et al.	A 53-year-old male patient presented to the surgery OPD with history of swelling of the left side of his neck for the last six months.
2014	Lee et al.	Hashimoto thyroiditis presented with a growing nodule in the thyroid.
2014	Bhat et al.	Hashimoto's thyroiditis
2015	Mertens de Wilmars et al.	Rapidly enlarging palpable thyroid mass.
2019	Gochhait et al.,	Rapidly increasing diffuse thyroid mass
2019	Sahu KK et al.	Hoarseness of voice
2019	Fahd Refai et al.	Hashimoto's thyroiditis

Table 1: Cases of Plasmacytoma since 1990 with differential diagnosis and background

Due to the rarity of the neoplasm the diagnosis is often challenging although FNAC is frequently used in the diagnostic path of thyroid nodular lesions, there is limited experience for the preoperative diagnosis of thyroid plasmacytomas. A thyroid plasmacytoma can be often mistaken as thyroid lymphoma and even medullary carcinoma. Very few cases of FNAC of a plasmacytoma have been reported in literature: EMP may mimic a medullary carcinoma or can be confused with Hürthle cell neoplasm or poorly differentiated carcinoma [12]. Furthermore, EMP should also be placed in differential diagnosis with plasma cell granuloma (PCG) of thyroid, which is a polyclonal proliferation of plasma cells. Immunocytochemistry for CD38 and CD138 along with κ and λ for stamping the clonality may be useful for the diagnosis of this rare entity. On histological examination the main differential diagnosis is mucosa associated lymphoid tissue (MALT) lymphoma, large B cell lymphoma and cyclin D1+ B-cell neoplasm like mantle cell lymphoma and hairy cell leukemia. Expression of CD20 by plasmacytoid cells or lymphocytes within the lesion and expression of mu heavy chain rather than alpha or gamma favours a diagnosis of lymphoma over plasmacytoma. The immunohistochemical demonstration of a cytoplasmatic light-chain restriction may support a neoplastic plasma cell proliferation even if a monotypic plasmocytic component may also be seen in MALT lymphoma [12,13].

Unlike plasmacytoma of the bone, EMP usually remains localized and does not often convert into multiple myeloma (only 3 cases progressing to multiple myeloma in a study of 50 patient).

Although globally no guidelines have been defined for the treatment of this rare disease, the main therapeutic options used are surgery and radiotherapy or their combination. Unlike the MM-associated forms of Plasmacytoma in which the mean survival is 9.7 ± 15.7 months, the literature review showed that none of the 19 patients with solitary EMP of the thyroid died, albeit during the short follow-up period [1,27].

An accurate integration of clinical, laboratory and radiological data to the pathological characteristics is fundamental to determine the nature, solitary or isolated, of the lesion and direct the patient towards the correct therapeutic pathway.

Declarations

• Ethics approval and consent to participate: the report was approved by the appropriate ethic review board.
• Consent for publication : the participant provided informed consent.
• Availability of data and materials : the datasets used for the report are available from the corresponding author on reasonable request.
• Competing interests : the authors declare that they have no competing interests.
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