

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

Casella C, Alfano MS, Russo G*, Molfino S, Cappelli, Simbeni

University of Brescia, Brescia BS, Italy

*Corresponding Author: Russo G, University of Brescia, Brescia BS, Italy. Tel: 3662358076, Email: gaetanorussomd@gmail.com

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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, Cancer

Introduction

Plasma cells are an unusual finding in the thyroid. The malignant proliferation of these 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 tumors usually mimic other malignant lesions such as papillary thyroid carcinoma and anaplastic thyroid carcinoma.

The peak of incidence of this type of neoplasm has 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 fibro-optic-laryngoscopy was performed, showing an anterior left-convex septal deviation, free nasopharynx, normal conformed pharyngeal axis in the absence of suspicious lesions. Normal larynx. Arytenoids' motility in the normal bilaterally. At the anesthesiologic evaluation no contraindications to surgical intervention were detected.

The patient was carefully informed of the risks (hemorrhages, vocal cord paralysis, infections, temporary/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 consideration of the size of the multinodular goiter, a bilateral paralysis was likely caused by "stupor" of the vocal cords.

In the subsequent post-operative period, the patient was evaluated several times by otolaryngologists and speech therapists for 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.

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 (Figures 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.

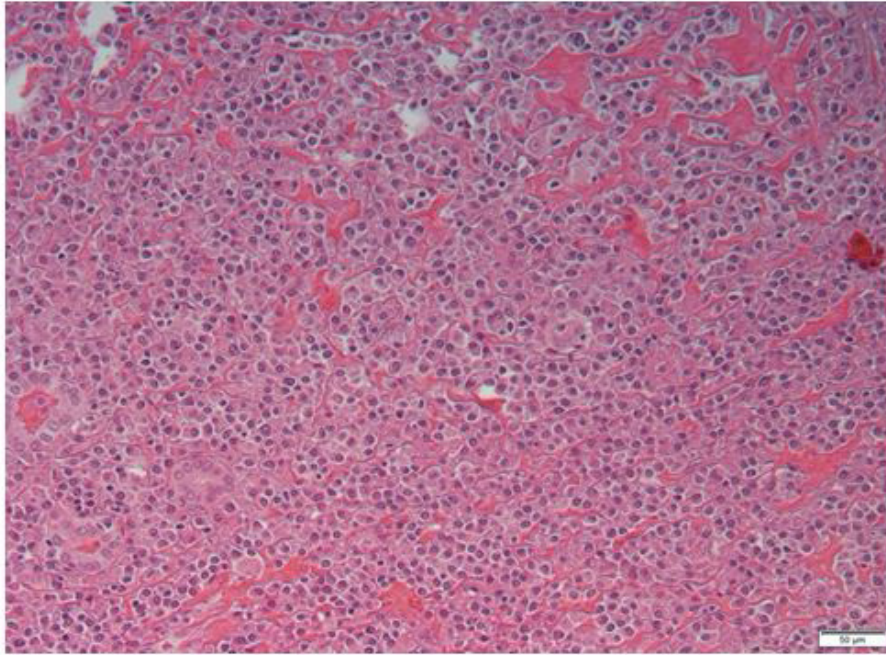


Figure 1: Thyroid parenchyma replaced by neoplastic proliferation consisting of a population of mature plasma cells

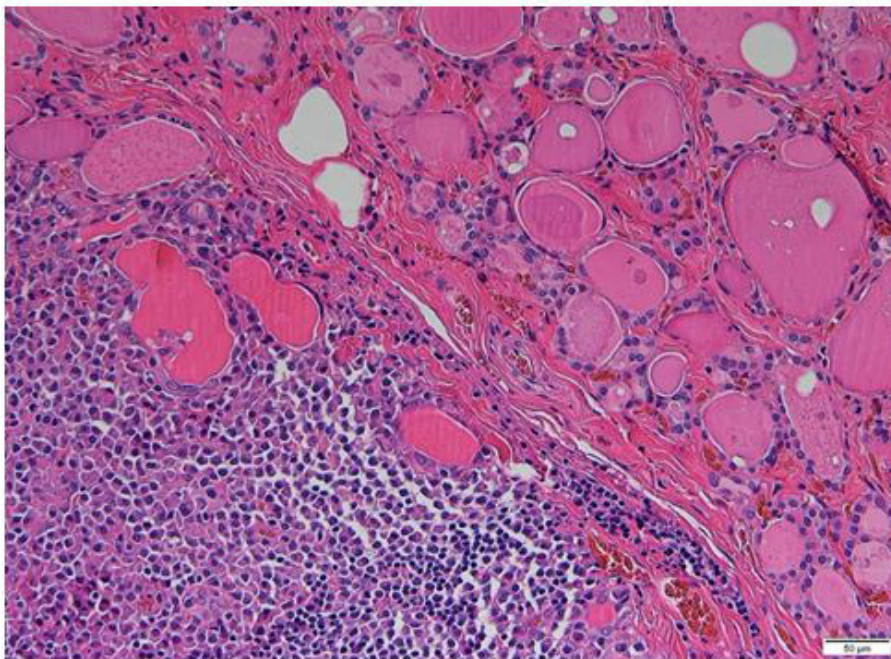


Figure 2: In the lower left corner there is neoplastic plasmacellular proliferation; in the upper right corner on the right there is thyroid parenchyma, consisting of flat cubic thyrocytes, with micro and medium follicular architecture

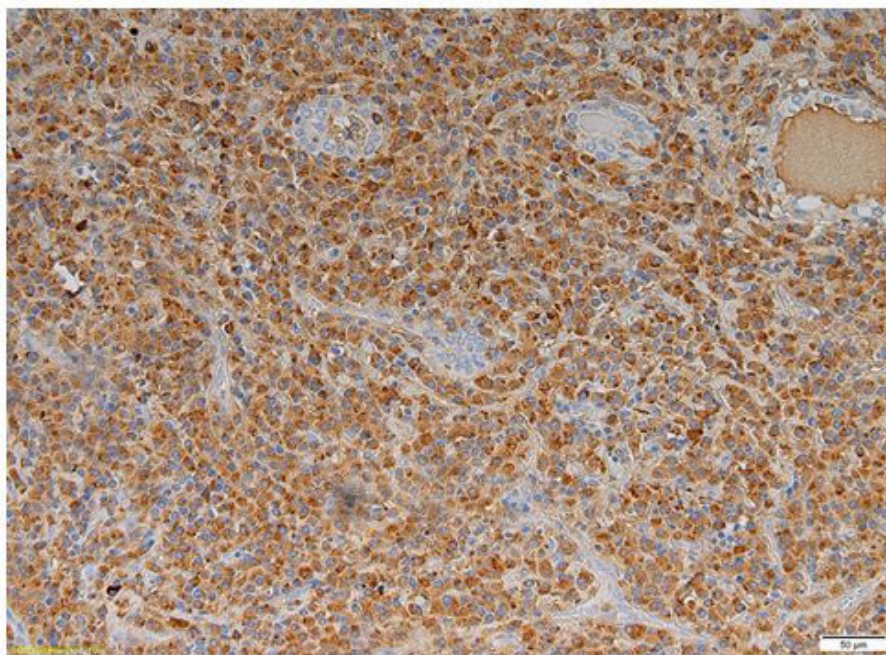


Figure 3: Immunohistochemical investigation showing monotypic restriction for Kappa light chains

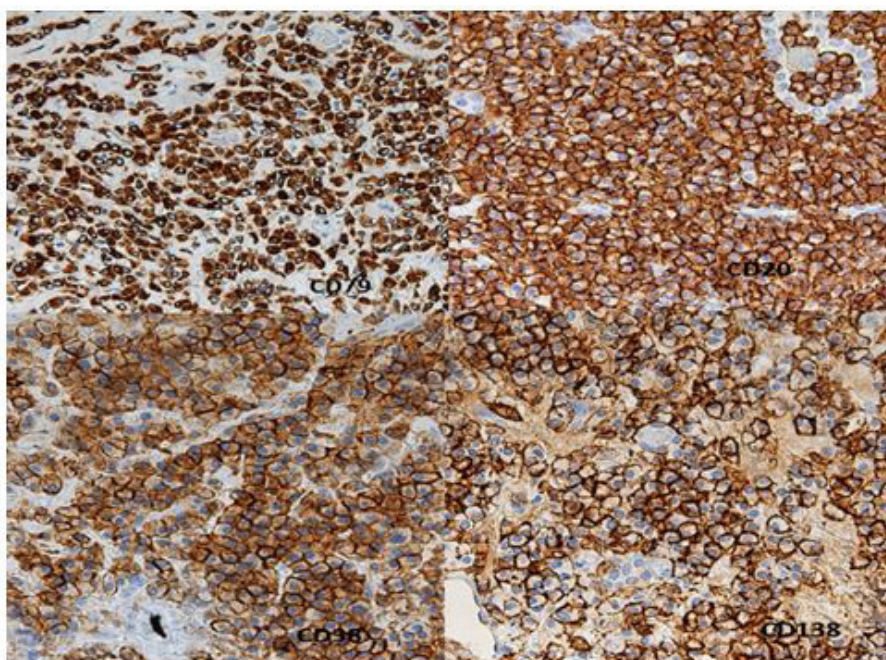


Figure 4: Cd38,138,20,79 EMA: Immunohistochemical survey positive for Cd38,138,20,79 EMA

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.

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]. 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 are 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 favors 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 plasmacytic 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.

References

1. Kovacs CS, Mant MJ, Nguyen GK, Ginsberg J (1994) Plasma cell lesions of the thyroid: Report of a case of solitary plasmacytoma and a review of the literature. *Thyroid*. 4:65-71.
2. McKenna R, Kyle R, Kuehl W, Grogan T, Harris N, Coupland R, et al. (2008) Lyon: IARC. WHO Classification of Tumours of Haematopoietic and Lymphoid Tissues Plasma Cell Neoplasms. 200-13.
3. Alexiou C, Kau RJ, Dietzfelbinger H, Kremer M, Spiess JC, Schratzenstaller B, et al. (1999) Extramedullary plasmacytoma: Tumor occurrence and therapeutic concepts. *Cancer*. 85:2305-14.
4. Kapadia SB, Desai U, Cheng VS (1982) Extramedullary Plasmacytoma of the head and neck. *Medicine (Baltimore)*. 61:317-329.
5. Aozasa K, Inoue A, Yoshimura H, Miyauchi A, Matsuzuka, KumaK, et al. (1986) Plasmacytoma of the thyroid gland. *Cancer*. 58:105-10.
6. Tandon A, Pauk TR, Singh R, Narendra AM (2015) Synchronous thyroid involvement in plasma cell leukemia masquerading as Hashimoto's thyroiditis: Role of ancillary cytology techniques in diagnostic workup. *Endocr Pathol*. 26:324-7.
7. More JR, Dawson DW, Ralston AJ (1968) Plasmacytoma of the thyroid. *J.clin.Path.* 21:661-667.
8. Bhat V, Shariff S, Reddy RAN (2014) Extramedullary plasmacytoma of thyroid - a mimicker of medullary carcinoma at fine needle aspiration cytology: A case report. *J Cytol*. 53-56.
9. Patten DK, Fazel M, Dina R, Tolley N (2011) Solitary extramedullary plasmacytoma of the thyroid involved by papillary carcinoma: a case report and review of the literature. *Endocr Pathol* 22: 155-158.
10. Ridal M, Ouattassi N, Harmouch T, Amarti A, Alami MN (2012) Solitary extramedullary plasmacytoma of the thyroid gland. *Case Rep Otolaryngol* 2012: 282784.
11. Cheslyn-Curtis S, Akosat AB (1990) Primary plasmacytoma of the thyroid. *Postgrad Med J*. 66:477-478
12. Boursos EP, Bedrossian CW, De Frias DV, Nayar R (2000) Thyroid plasmacytoma mimicking medullary carcinoma: A potential pitfall in aspiration cytology. *Diagn Cytopathol*. 23:354-8.
13. Bhat V, Shariff S, Reddy RA (2014) Extramedullary plasmacytoma of thyroid-a mimicker of medullary carcinoma at fine needle aspiration cytology: A case report. *J Cytol*. 31:53-6.
14. Puliga G, Olla L, Bellisano G, Di Naro N, Ganau M, Lai ML, et al. (2011) Solitary extramedullary plasmacytoma of the thyroid gland associated with multinodular goiter: Case report and review of the literature. *Pathologica*. 103:61-3.
15. Lee CH, Jung YY, Chung YR, Ryu HS (2014) Liquid-based cytologic findings of solitary extramedullary plasmacytoma in thyroid: A case report identified with fine-needle aspiration cytology. *Diagn Cytopathol*. 42:964-9.
16. De Schrijver I, Smeets P (2004) Thyroid enlargement due to extramedullary plasmacytoma. *JBR-BTR*. 87:73-5.

17. Patten DK, Fazel M, Dina R, Tolley N (2011) Solitary extramedullary plasmacytoma of the thyroid involved by papillary carcinoma: A case report and review of the literature. *Endocr Pathol.* 22:155-8.
18. Gochhait D, Govindarajalou R, Kar R, Rangarajan V, Dehuri P, Dubashi B (2019) Plasmacytoma of thyroid clinically and morphologically mimicking primary thyroid carcinoma. *Cytopathology.* 30:113-6.
19. Sahu KK, Singh P, Malhotra P, Srinivasan R (2019) Thyroid plasmacytoma: A rare cause of hoarseness of voice. *Indian J Nucl Med.* 34:78-80.
20. Shahani S, Ahmad A, Barakat FH, Chuang HH, Fowler NH, Myers JN, et al. (2011) F-18 FDG PET/CT detecting thyroid plasmacytoma after the successful treatment of gastric large B-cell lymphoma. *Clin Nucl Med.* 36:317-9.
21. Abdel Khalek MS, Ibrahim WG, Crawford BE, Kandil EH (2010) Euthyroid enlargement of the thyroid gland. Plasmacytoma in thyroid. *Neth J Med.* 68:424-429.
22. Ozkan E, Tokmak E, Kir KM (2008) Detection of thyroid plasmacytoma by F-18 FDG PET/CT imaging. *Clin Nucl Med.* 33:292-3.
23. Chaganti S, Gurunathan R, McNaboe E (2007) Solitary extramedullary plasmacytoma of the thyroid gland: A case report and review of literature. *Internet J Head Neck Surg.* 2:1-4.
24. Kuo SF, Chang HY, Hsueh C, Lin JD (2006) Extramedullary plasmacytoma of the thyroid. *N Z Med J.* 119: U2005.
25. Hasegawa Y, Itoh T, Tamagawa Y, Komeno T, Kojima H, Ninomiya H, et al. (1999) Non-Hodgkin's lymphoma followed by plasmacytoma, both arising in A thyroid gland with Hashimoto's disease. *Leuk Lymphoma.* 35:613-8.
26. Avila A, Montoya A, Luna M (2009) Clinical features and differential diagnoses of solitary extramedullary plasmacytoma of the thyroid. *Annals of Diagnostic Pathology* 13:119-123.
27. Galieni P, Cavo M, Pulsoni A, Avvisati G, Bigazzi C, Neri S et al. (2000) Clinical outcome of extramedullary plasmacytoma. *Haematologica.* 85:47-51.

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