Solitary Extramedullary Plasmocytoma of the Thyroid Gland: A **Case Report**

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Abstract: Solitary extramedullary plasmocytoma (SEP) of the thyroid gland is a very rare disease. The diagnosis of SEP can be made after ruling out multiple myeloma. Histological examinations, immunohistochemical analysis with an overexpression of CD 138, CD 38 and kappa light chain reaction confirmed this uncommon condition in our case. Medullary carcinoma, MALT-Lymphoma and Non-Hodgkin Lymphoma of the thyroid should be excluded in the diagnosis.

Surgical resection and radiotherapy, or a combination of both, are standard treatment methods. However, because of rareness of this disease, no general therapy can be recommended.

We report about a 75 year old male patient and its involvement of the right remainder thyroid lobe by SEP five years post a Dunhill-Procedure due to multinodular goiter.

Keywords: Solitary, plasmocytoma, extramedullary, thyroid, multiple myeloma.

INTRODUCTION

Only less than 80 cases of solitary extramedullary plasmocytoma (SEP) have been reported, the first one being in 1938 by Voegt [1]. The pathogenesis of a solitary extramedullary plasmocytoma remains no understood phenomenon. In contrast, the history of lymphocytic thyroiditis is common. In this case report we describe a case of SEP of the thyroid gland associated with chronic lymphocytic thyreoditis in the remainder right thyroid lobe post an incomplete resection.

CASE REPORT

A 75 year old male presented with a 12-week history of a progressively right-sided painless goiter enlargement without any compressive or toxic symptoms. Dunhill Operation hemithyreoidectomy and right underlobe resection) was performed in 2008 as a result of a benign multinodular goiter. A daily substitution with L-Thyroxine 75 ug was administered by the patient. Upon a physical examination, we found a good palpable, soft and mobile mass of the thyroid lobe.

The neck sonography confirmed an enlargement of the right thyroid lobe with a 1,4 x 2,3 x 2,1 cm inhomogeneous nodule without any signs of a cervical

lymphadenopathy. No other pathological features were described. Preoperatively, no fine needle biopsy (FNB) was performed.

With the exception of a mild anemia the preoperative laboratory test including TSH, serum calcium and total protein, as well as a chest x ray, were within a normal range. An uncomplicated right sided rest-thyreoidectomy was performed. The postoperative course was uneventful with normal vocal cord function and no hypocalcaemia.

An intraoperative frozen-section analysis suspected a plasmocytoma. A histological examination showed an enlarged right thyroid lobe replaced by a 2,8 x 2,8 x 1,5 cm big soft vitreous shiny nodule with clear borders to the thyroid tissue. Atypical plasma cells with eccentric nuclei, basophilic cytoplasm and high mitotic index signed a lymphomatous process (Figure 1). Signs of the chronic lymphotic thyroiditis were observed in the rest of the thyroid parenchyma.

Immunohistochemical analysis was performed using a automated Benchmark XT IHC/ISH instrument (Ventana Medical Systems, Tucson, AZ, USA) and following antibodies were used; for CD =Syndecan-1 (clone B-A38, dilution 1:100; Zytomed Systems, Berlin, Germany), CD 38 (clone SP149, dilution 1:200, Cell Marque, Rocklin, CA,USA), CD 20 (clone L26, dilution 1:400; Zytomed Systems, Berlin, Germany), Kappa and Lambda(polyclonal rabbit, dilution 1:100, Epitomics, Burlingame, CA, USA). The signal was visualised with a diaminobenzidine (DAB)

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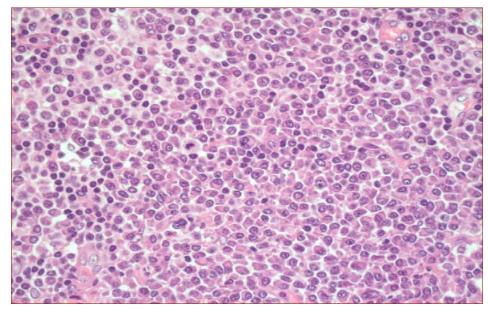


Figure 1: Diffuse infiltration of thyroid parenchyma by neoplastic plasma cells with irregular and eccentric nuclei (H&E, x400).

with procedures performed according to the manufacturer's instructions. A semiquantitative method of scoring was used to score immunohistochemistry.

This analysis showed neoplastic cells strong positive for CD 38 (Figure 2) and CD 138 and negative for CD 20 and Lambda-light chain. Staining for IgG and Kappa-light chain antibodies was strong positive (Figure 3).

After two weeks, the patient was seen by an oncologist.

Evidence of IgG-Kappa in Serum has been shown in immunofixation. Bone marrow aspiration, immunoelectrophoresis and a low dose computer-tomography bone survey were within normal. Free Lambda- and Kappa light chains proved normal as well. After ruling out a multiple myeloma, SEP of thyroid was confirmed.

An additional radiotherapy of the neck region was performed. The patient remains disease free at 6 months postoperatively.

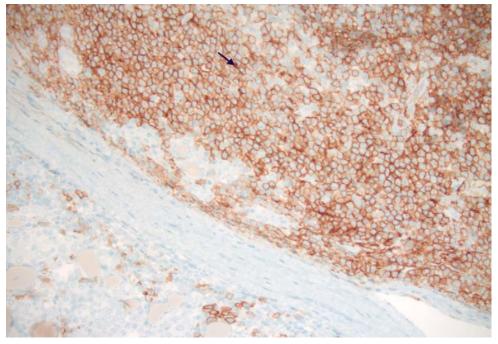


Figure 2: Immunohistochemical staining (x200) showing positivity for CD 38 (arrow).

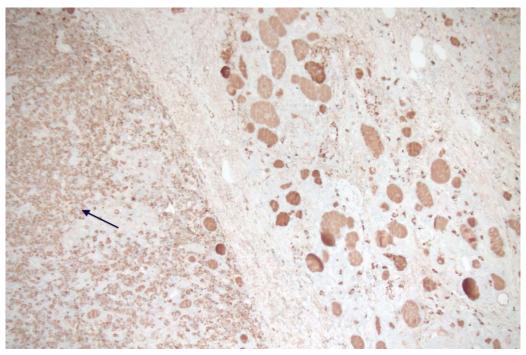


Figure 3: Immunohistochemistry showing (x100) strong positivity of tumor cells for Kappa-light chain (arrow).

DISCUSSION

Primary SEP is a soft tissue neoplasm characterized by a monoclonal proliferation of plasma cells [2]. They can occur in bone as solitary bone plasmocytoma or in extramedullary tissues and are less than 5% plasmocytoma. The most common localization of the EMP is the upper respiratory tract, salivary glands and oral cavity [4].

Three fourth of EMP cases involve males of 4th to 7th decade and reports in the literature show the ratio of men to women at 44 to 56% [7]. Aside from the local compressive symptoms and hoarseness, rapidly enlarging nodule if growth is measured within 3 months is considered as SEP of thyroid [4].

SEP of the thyroid is very rare; however primary thyroid involvement of multiple myeloma is common. Therefore, the most difficult challenge in the diagnosis of SEP is to exclude a multiple myeloma.

Bone marrow aspiration, skeletal survey and the electrophoresis are essential examinations to rule out the multiple myeloma and confirm SEP. Negative results of all above, clinical examinations and histological results, confirmed this rare disease in our patient.

SEP of the thyroid gland is known to occur on the ground of lymphocytic thyreoiditis [3].

Due to a diffuse infiltration of the thyroid with plasma cells and lymphotic tissue, the differentiation between SEP of the thyroid, MALT lymphoma, Non-Hodgkin, B-cell lymphoma or medullary carcinoma remains difficult. Marginal zone lymphomas, occurred more common than plasmocytoma [2], were positive for CD 19 and CD 20 but they would show positivity for CD 138 as well [11]. The lack of reactivity for CD 20 with overexpression of CD 38 and CD 138, with Kappa-light chain positivity distinguished and confirmed the SEP of the thyroid in our case.

As in our patient, sixty- seven percent of the patients had evidence of monoclonal Ig in the serum. The most common is IgG-kappa, but many forms have also been reported [1-3, 5, 6].

Because of the rarity of this disease, no standard treatment can be recommended at this point. A surgical resection, neck radiation or a combined approach of both remain the main target in the treatment.

Furthermore, chemotherapy has no evidence in the management of SEP of the thyroid.

The prognosis of all localized plasmocytoma is favorable. The largest study by Kovacs indicated that 73 % of all patients were free of disease, 3% were alive with an evidence of the disease and 13% had died from it at a follow-up at 56-73 months [2].

Galieni *et al.* showed that 15% of 46 patients developed multiple myeloma with a follow-up at 118 months [8]. Patients who progressed to multiple myeloma had a 5-year survival rate of 100% with 33% for solitary bone plasmocytoma [9, 10].

CONCLUSION

The SEP of the thyroid gland is a very rare disease. To rule out multiple myeloma complete blood count, bone marrow aspiration, skeletal survey and the electrophoresis must be completed. The need of immunohistochemical analysis is very important to differentiate SEP of the thyroid from other entities.

Surgical resection, radiotherapy, or combinations of both are treatment options. The prognosis of the illness remains favorable.

CONFLICT OF INTEREST

The authors declare that they have no conflict of interest.

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