Scientific Research Publishing

Open Journal of Internal Medicine, 2024, 14, 337-342
https://www.scirp.org/journal/ojim
ISSN Online: 2162-5980
ISSN Print: 2162-5972

Thyroid Inflammatory Myofibroblastic Tumor: A Rare Case, Literature Review

Hamza Sümter

Department of Internal Medicine, Hematology, Uşak Training and Research Hospital, Uşak, Türkiye
Email: hamzasumter@gmail.com

How to cite this paper: Sümter, H. (2024) Thyroid Inflammatory Myofibroblastic Tumor: A Rare Case, Literature Review. *Open Journal of Internal Medicine*, **14**, 337-342.
https://doi.org/10.4236/ojim.2024.144030

Received: August 30, 2024
Accepted: December 14, 2024
Published: December 17, 2024

Abstract

Inflammatory myofibroblastic tumor has been referred to by many different names in the past, such as plasma cell granuloma, inflammatory pseudotumor, fibrous histiocytoma, fibroxanthoma, xanthogranuloma. It was first described in the 1930s. It originates from soft tissue. It has low malignancy potential. It is rarely seen. While it is often localized in the lungs, it can rarely be localized in the thyroid gland. Its etiology and pathogenesis are unknown. Diagnosis is based on postoperative immunohistochemical evaluation. Although surgical resection is the main treatment method, there is no standard approach. Recurrence is not expected in general. However, cases of recurrence after a long period of time have also been reported. In this study, a 46-year-old female patient was presented with thyroid inflammatory myofibroblastic tumor that developed after COVID-19. Another notable point in the case is that her mother had myeloma with mutations (changes in chromosomes 11 and 14).

Keywords

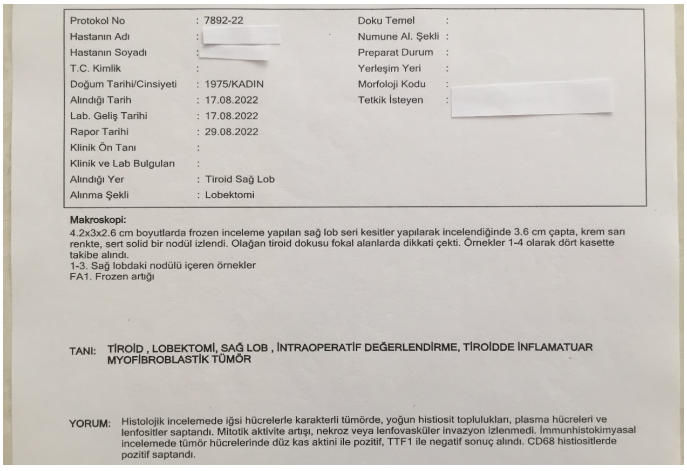
Inflammatory Myofibroblastic, Tumor, Thyroid, ALK

1. Introduction

Inflammatory myofibroblastic tumor (IMT) develops as a result of myofibroblast proliferation. It is a rare disease. Most patients present with a mass [1]. It can occur in any location in the body, especially in the lungs [2]. Etiology and pathogenesis are unknown. Diagnosis is based on histology. The biopsy contains varying amounts of fibroblasts, myofibroblasts, plasma cells, lymphocytes, and eosinophils [3]. The picture may be accompanied by fever, weight loss, and laboratory abnormalities [1]. Although there is no standard treatment, surgical resection is the primary treatment option [1]. Prognosis is variable. The true prevalence of the disease is unknown.

2. Case Report

A 48-year-old female patient was evaluated in the hematology clinic due to swelling in her neck. There was no medical or herbal drug use in her anamnesis. In terms of lymphoproliferative disease, fever, weight loss, night sweats, etc. (B symptom) were not observed. The diagnosis of multiple myeloma in the mother drew attention in terms of family history (The patient's mother was a myeloma patient with changes in chromosomes 11 and 14 in our hematology clinic). Complete blood count, biochemical values (transaminases, renal function tests), thyroid function tests, thyroglobulin, antithyroid peroxidase, anti-thyroglobulin were within normal range. No pathology was detected in hepatitis (HAV, HBV, HCV), TORCH, EBV, brucella, salmonella tests. No atypical cells were observed in the peripheral blood smear. Ultrasonography performed for a palpable swelling in the neck on physical examination revealed a 21 mm × 12 mm nodule in the left lobe and a 40 mm × 22 mm nodule in the right lobe. Fine needle aspiration biopsy was found to be compatible with thyroiditis. In the evaluation made 3 months later, fine needle aspiration biopsy was repeated to exclude malignancy. However, sufficient results were not obtained. The patient, who did not have B symptom, was re-evaluated with cervical USG approximately 2 months later. In this USG, a 47 mm × 27 mm × 35 mm nodule was detected, almost completely filling the right thyroid lobe and extending to the isthmus. Surgery was recommended to the patient to exclude possible malignancy and right subtotal thyroidectomy was performed. Spindle cells, dense histiocyte aggregates, plasma cells and lymphocytes were seen in the postoperative material. No increase in mitotic activity was observed. Smooth muscle actin(+), CD68(+), TTF1(-) were detected in the sample. The case was diagnosed with thyroid inflammatory myofibroblastic tumor. Because of its rarity, the diagnosis was confirmed in two different centers. ALK rearrangement was found to be negative (Figure 1, Figure 2). As of September 2024, the patient has been asymptomatic and has been monitored in our clinic for 2 years postoperatively.



Protokol No	: 7892-22	Doku Temel	:
Hastanın Adı	: [redacted]	Numune Al. Şekli	:
Hastanın Soyadı	: [redacted]	Preparat Durum	:
T.C. Kimlik	:	Yerleşim Yeri	:
Doğum Tarihi/Cinsiyeti	: 1975/KADIN	Morfoloji Kodu	:
Alındığı Tarih	: 17.08.2022	Tetkik İsteyen	: [redacted]
Lab. Geliş Tarihi	: 17.08.2022		
Rapor Tarihi	: 29.08.2022		
Klinik Ön Tanı	:		
Klinik ve Lab Bulguları	:		
Alındığı Yer	: Tiroid Sağ Lob		
Alınma Şekli	: Lobektomi		

Makroskopi:
4.2x3x2.6 cm boyutlarda frozen inceleme yapılan sağ lob seri kesitler yapılarak incelendiğinde 3.6 cm çapta, krem sarı renkte, sert solid bir nodül izlendi. Olağan tiroid dokusu fokal alanlarda dikkati çekti. Örnekler 1-4 olarak dört kasette takibe alındı.
1-3. Sağ lobdaki nodülü içeren örnekler
FA1. Frozen artığı

TANI: TİROİD , LOBEKTOMİ, SAĞ LOB , İNTRAOPERATİF DEĞERLENDİRME, TİROİDDE İNFLAMATUAR MYOFİBROBLASTİK TÜMÖR

YORUM: Histolojik incelemede iğsi hücrelerle karakterli tümörde, yoğun histiosit toplulukları, plasma hücreleri ve lenfositler saptandı. Mitotik aktivite artışı, nekroz veya lenfovasküler invazyon izlenmedi. Immunhistokimyasal incelemede tümör hücrelerinde düz kas aktini ile pozitif, TTF1 ile negatif sonuç alındı. CD68 histiositlerde pozitif saptandı.

Figure 1. Pathology result of the patient (confirmed in two different pathology departments).

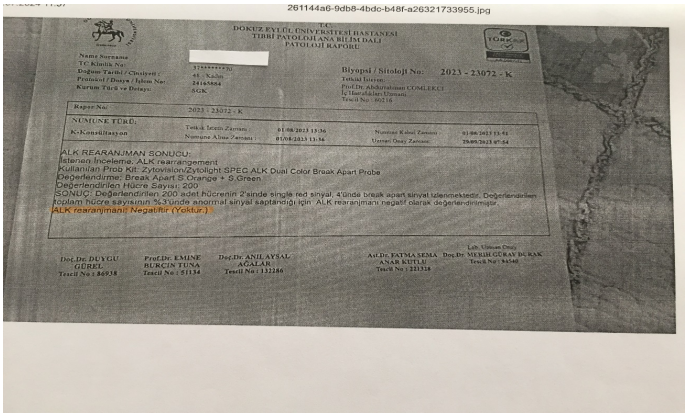


Figure 2. ALK result of the patient: negative.

3. Discussion

Inflammatory myofibroblastic tumor (IMT), a rare disorder, was first described in the thoracic region in 1939 [1]. In the literature, plasma cell granuloma (PCG) is also called inflammatory pseudotumor (IPT) [3]. Although the etiology and pathogenesis are unknown, many factors such as trauma, viruses, inflammation, infection, and abnormal response to prolonged exogenous stimulation have been implicated [1] [4]. In this case, cervical swelling after covid-19 supports the hypothesis of an abnormal response to infection. Also, the fact that the patient's mother was a patient with multiple myeloma followed up in our hematology clinic made us think about genetic and/or epigenetic factors in the etiopathogenesis of thyroid inflammatory myofibroblastic tumor. The mother with myeloma diagnosis has t (11; 14) (translocation between chromosome 11 and chromosome 14) (**Figure 3**).

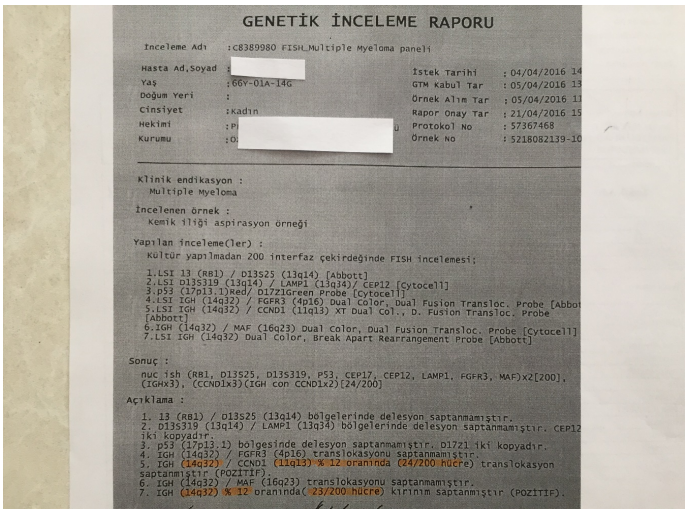


Figure 3. t (11:14) mutation of the patient's mother.

Inflammatory myofibroblastic tumors are more commonly localized in the lungs, but may also be located in any organ [3]. It presents with a massive lesion

and presenting symptoms may vary according to localization [1]. According to the World Health Organization, IMT is defined as a low-grade malignant tumor [1]. Although it can be seen at any age, it peaks in children and adolescents [1]. More than half of the cases are under the age of 40 [5]. While IMT is generally seen with equal frequency across genders and races, there is a female predominance in the identified thyroid IMT cases [6].

Histology is essential for diagnosis [1]. The biopsy contains varying amounts of fibroblasts, myofibroblasts, plasma cells, lymphocytes, and eosinophils [1]. There are 3 histological patterns: mixoid, vascular, inflammatory pattern, compact spindle cell pattern, dense fibrotic pattern [3]. Immunohistochemically, vimentin is diffusely positive, and myogenic antibodies smooth muscle actin and muscle-specific actin are localized or diffusely positive [1]. ALK positivity is seen in 50% - 70% of cases [1]. S100, myoglobin, CD21, CD23, CD34, CD117, caldesmon are frequently negative [1].

While IMT cases in the head and neck region constitute approximately 5% of all cases, IMT cases, especially those located in the thyroid, are much less common [1]-[3]. According to a study published in 2022, the total number of thyroid IMT (PCG/IMT/IPT) cases in the English literature is 25. Of these cases, 8 were male and 17 were female. This is the third thyroid IMT case from Turkey after the two cases published by Deniz *et al.* in 2008 [7].

The primary treatment for IMT located in the head and neck is surgery [1]. If surgical resection cannot be performed completely, recurrence may occur at a rate of up to 50% and metastasis is less than 2% [1]. In 2017, only one of the 20 thyroid IMT cases reported recurrence, and this patient received thyroid radiotherapy and oral steroid treatment [3]. It has been stated that ALK (+) may be effective in this recurrence [2].

In general, the prognosis is variable. It may remain stable or grow slowly or even regress spontaneously [8]-[10]. In most cases, recurrence is not expected after surgery [11] [12]. However, a case of recurrence 11 years after surgery has also been described [13] [14]. Some studies have stated that ALK (+) indicates a poor prognosis for patients and that ALK inhibitors such as crizotinib may be promising in the treatment of these cases [1] [3].

4. Conclusion

Inflammatory myofibroblastic tumor is a rare disease and can present as a mass lesion at any location in the body. Thyroid localization is as rare as possible. Etiology, pathogenesis and frequency are unknown. It can be seen at any age. Histology (immunological histochemistry) is essential for diagnosis. The prognosis is generally good after surgery. It usually does not require further treatment such as chemotherapy or radiotherapy. However, it should be kept in mind that it may relapse long after the diagnosis.

Conflicts of Interest

The author declares no conflicts of interest regarding the publication of this

paper.

References

[1] Zhao, J., Han, D., Gao, M., Liu, M., Feng, C., Chen, G., *et al.* (2020) Inflammatory Myofibroblastic Tumor of the Neck with Thyroid Invasion: A Case Report and Literature Review. *Gland Surgery*, **9**, 1042-1047. https://doi.org/10.21037/gs-20-355

[2] Duan, J. and Wang, Y. (2017) A Case Report of Recurrent Thyroid Inflammatory Myofibroblastic Tumor and Its Metastasis in Soft Tissue. *Medicine*, **96**, e8485. https://doi.org/10.1097/md.0000000000008485

[3] Li, C., Wu, X., Wang, J., Wang, K., Zhang, S., Huang, J., *et al.* (2020) ALK-1-Positive Inflammatory Myofibroblastic Tumor of the Thyroid Complicated by Hashimoto's Thyroiditis: Report of a Rare Case and a Literature Review. *Diagnostic Pathology*, **15**, Article No. 58. https://doi.org/10.1186/s13000-020-00966-4

[4] Matsubara, O., Tan-Liu, N.S., Kenney, R.M. and Mark, E.J. (1988) Inflammatory Pseudotumors of the Lung: Progression from Organizing Pneumonia to Fibrous Histiocytoma or to Plasma Cell Granuloma in 32 Cases. *Human Pathology*, **19**, 807-814. https://doi.org/10.1016/s0046-8177(88)80264-8

[5] Weinberger, S.E. (2023) Uptodate, Uptodate, Inflammatory Myofibroblastic Tumor (Plasma Cell Granuloma) of the Lung.

[6] Demirhan, O., Ozkara, S., Yaman, M. and Kaynak, K. (2013) A Rare Benign Tumor of the Lung: Inflammatory Myofibroblastic Tumor—Case Report. *Respiratory Medicine Case Reports*, **8**, 32-35. https://doi.org/10.1016/j.rmcr.2013.01.001

[7] Deniz, K., Patirog˘lu, T.E. and Okten, T. (2008) Plasma Cell Granuloma of the Thyroid. Case Report. *Journal of Pathology, Microbiology and Immunology*, **116**, 167-172. https://doi.org/10.1111/j.1600-0463.2008.00827.x

[8] Mandelbaum, I., Brashear, R.E. and Hull, M.T. (1981) Surgical Treatment and Course of Pulmonary Pseudotumor (Plasma Cell Granuloma). *The Journal of Thoracic and Cardiovascular Surgery*, **82**, 77-82. https://doi.org/10.1016/s0022-5223(19)39390-0

[9] Kim, J.H., Cho, J.H., Park, M.S., Chung, J.H., Lee, J.G., Kim, Y.S., *et al.* (2002) Pulmonary Inflammatory Pseudotumor—A Report of 28 Cases. *The Korean Journal of Internal Medicine*, **17**, 252-258. https://doi.org/10.3904/kjim.2002.17.4.252

[10] Gal, A.A., Koss, M.N., McCarthy, W.F. and Hochholzer, L. (1994) Prognostic Factors in Pulmonary Fibrohistiocytic Lesions. *Cancer*, **73**, 1817-1824. https://doi.org/10.1002/1097-0142(19940401)73:7<1817::aid-cncr2820730708>3.0.co;2-k

[11] Fabre, D., Fadel, E., Singhal, S., de Montpreville, V., Mussot, S., Mercier, O., *et al.* (2009) Complete Resection of Pulmonary Inflammatory Pseudotumors Has Excellent Long-Term Prognosis. *The Journal of Thoracic and Cardiovascular Surgery*, **137**, 435-440. https://doi.org/10.1016/j.jtcvs.2008.07.009

[12] Chen, C.-H., Huang, W., *et al.* (2008) Surgical Outcome of Inflammatory Pseudotumor in the Lung. *The Thoracic and Cardiovascular Surgeon*, **56**, 214-216. https://doi.org/10.1055/s-2007-989340

[13] Weinberg, P.B., Bromberg, P.A. and Askin, F.B. (1987) "Recurrence" of a Plasma Cell Granuloma 11 Years after Initial Resection. *Southern Medical Journal*, **80**, 519-

521. https://doi.org/10.1097/00007611-198704000-00028

[14] Cerfolio, R.J., Allen, M.S., Nascimento, A.G., Deschamps, C., Trastek, V.F., Miller, D.L., *et al.* (1999) Inflammatory Pseudotumors of the Lung. *The Annals of Thoracic Surgery*, **67**, 933-936. https://doi.org/10.1016/s0003-4975(99)00155-1