Case Report

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Thyroid lipomatosis in a patient of post renal transplantation

Meena R. Kalluri^{1*}, Kavya Balusu², Sachin Marda¹, Sreehari Gowda¹

¹Surgical Oncology Resident, Yashoda Super Specialty Hospitals, Somajiguda, Hyderabad, Telangana, India ²Gandhi Medical College, Musheerabad, Secunderabad, Telangana, India

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*Correspondence:

Dr. Meena R Kalluri,

E-mail: meenareddy1990@gmail.com

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ABSTRACT

Thyroid lipomatosis is a rare benign disease with very few cases available in literature. We report a case of a 48 year old Indian male who presented with progressive swelling in front of neck since 5 years and difficulty breathing in lying down position along with difficulty in swallowing for the last 2 months. He is a known case of post renal transplantation for chronic kidney disease due to secondary AA amyloidosis. Total thyroidectomy was done and histopathology showed diffuse thyroid lipomatosis, with no evidence of amyloid deposition or papillary carcinoma. Fatty tissue is not commonly detected within the thyroid gland. Here we have reviewed the literature and discussed various differential diagnosis of thyroid lipomatosis.

Keywords: Lipomatosis, Benign, Goiter, Case report

INTRODUCTION

Normally, adipose tissue is observed in the parathyroid, salivary glands, thymus, breasts and pancreas; however, it is unusual to detect adipose tissue in the thyroid gland.¹ Normal thyroid occasionally may show presence of mature adipocytes in perivascular and subcapsular area anteriorly.² Generally, fat-containing thyroid lesions have been divided into two groups. One of them is fatcontaining thyroid neoplasms, such as papillary carcinoma, follicular adenoma and invasive follicular carcinoma, and the other is non-neoplastic conditions with adipose tissue including adenolipoma thyroid, amyloid goitre, lymphocytic thyroiditis, thyroid atrophy, disease, heterotopic adipocytic intrathyroidal thymic or parathyroid lipoma.3 Diffuse thyroid lipomatosis causing goiter can progressively increase and cause pressure symptoms, pathophysiology of diffuse fatty infiltration is unclear. Imaging studies can suggest the presence of adipose tissue but definitive diagnosis requires histopathological examination.

CASE REPORT

A 48 year old male patient presented to our hospital in July, 2021 with progressive swelling in front of neck in midline since 5 years and difficulty in breathing in lying down position along with difficulty in swallowing for the last 2 months. The swelling was noticed 5 years ago which was mild and remained constant in size but rapidly progressed to the present size in the past 3 months (Figure 1). No history of change in voice. No history suggestive of hypo or hyperthyroidism.

Past history dates back to October 2014, when he first developed oliguria and bilateral pedal edema. Laboratory evaluation revealed elevated creatinine and gross proteinuria. Renal biopsy was done and the patient was diagnosed to have chronic kidney disease due to AA amyloidosis. ANA, Anti CCP were negative, Rheumatoid factor-low positive. For various indications, imaging and biopsies of different organs were performed which showed involvement of kidney, thyroid and adrenal with amyloidosis with noninvolvement of bone marrow, heart,

skin, and rectum. Thyroid FNAC done in 2017 was suggestive of amyloid goiter, Bethesda II.



Figure 1: Middle neck swelling.

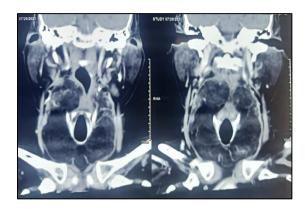


Figure 2: CECT neck.



Figure 3: Thyroid specimen.

Patient developed end stage renal disease with CKD 5, was on maintenance hemodialysis from October 2017 and registered for kidney transplantation. He underwent right renal transplantation in February 2019, from a 55 year old unknown deceased donor. Since then, the patient is on immunosuppression with triple drug regimen of Tacrolimus, Mycofenolate mofetil, prednisolone with good graft function.

Now on examination, diffusely enlarged goiter with each lobe of approximately 10x5 cm noted. The swelling is

non-tender, mobile, moving with deglutition and soft in consistency. No cervical lymphadenopathy noted. CECT of neck showed diffusely enlarged goiter with fatty infiltration and enhancing septae, extending superolaterally and superoposteriorly into retropharyngeal region up to hyoid bone, displacing bilateral internal jugular veins and carotid arteries posterolaterally.



Figure 4: Cut section of thyroid specimen.

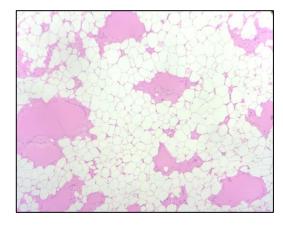


Figure 5: Microscopic picture of thyroid.

Trachea normal, no cervical lymphadenopathy (Figure 2). Routine laboratory workup and thyroid function tests are normal. FNAC not done now as we suspected amyloid goiter based on history and previous FNAC.

Total thyroidectomy with preservation of parathyroids was performed. Bilateral recurrent laryngeal nerves were identified and preserved.

On gross examination of specimen weighing 238.4 gm and measurements of $10\times5\times3.5$ cms in right lobe, $4\times2\times1$ cms in isthmus and $10\times6\times4$ cms in left lobe, capsule is intact (Figure 4). Cut section showed diffuse tan areas with no nodules (Figure 5). Microscopic examination showed diffuse infiltration of thyroid parenchyma by mature adipocytic cells, with intervening areas of normal to distended thyroid follicles lined by flattened to cuboidal follicular cells. No evidence of atypia /malignancy and amyloid deposits noted (Figure 5).

Polarizing microscopic view on congo red stain is negative for apple green birefringence of amyloid, with final impression of diffuse thyroid lipomatosis. Thyroid replacement therapy was started.

DISCUSSION

It is unusual for the occurrence of mature adipose tissue in thyroid gland. The amount of adipose tissue can be increased in certain disorders of thyroid, such as Hashimoto's thyroiditis, amyloid goiter containing adipose infiltration, heterotopic adipocytes, thyroid lipoma or adenolipoma, intrathyroidal thymus, and parathyroid lipoma. Increased amount of fat can also be found in some malignant thyroid lesions, such as encapsulated papillary thyroid carcinoma and thyroid liposarcoma.⁴ Thyroid lipomatosis is a rare, benign disease characterized by the enlargement of thyroid due to mature adipose tissue diffusely infiltrating the stroma. It has no gender predominance and mostly affects middle aged persons.⁴

The pathophysiology of diffuse proliferation of adipose tissue in the thyroid gland is poorly understood. Some authors suggest that heterotopic groups of fat cells are included in the thyroid gland during embryogenesis.³ Breek et al reported a case of simultaneous thyrolipoma and thymolipoma and noted that thyroid and thymus arise from primitive foregut and hence suggested a mechanism of disturbance in the development of the primitive foregut.⁵

Schröder and Böcker believe in the metaplastic origin of fat cells from stromal fibroblasts due to chronic tissue hypoxia in amyloid goitres, which is presumed to be responsible for the presence of fatty tissue in them. However metaplastic transformation of neoplastic thyrocytes has been claimed to be the cause of massive follicular cell steatosis seen in the lipid-rich variant of clear cell thyroid adenomas and carcinomas. LiVolsi believes that metaplastic etiology is unlikely as the usual findings of fibrosis or inflammation, seen in fatty metaplasia of other organs such as in cystic fibrosis, has not been reported in these cases.

Lau et al published a possible relationship between the mutation of the mitochondrial protein succinate dehydrogenase-subunit B and the abnormal differentiation of adipose tissue in thyroid lipomatosis.⁸

Thyroid lipomatosis presents as a progressive enlargement of the thyroid that can exert pressure over the upper aerodigestive tract, producing dyspnea, dysphagia, and changes in the voice. Physical examination usually reveals a non-tender goiter that is nodular or diffuse and soft in consistency. Although hyperthyroidism and hypothyroidism have been described, majority of cases are in euthyroid state.

Ultrasound findings demonstrate an enlarged thyroid with a diffuse increase in echogenicity and an echo attenuation typical of adipose tissue. ¹⁰ CT scans show an enlarged thyroid with well-defined limits and diffuse fatty infiltration evidenced by negative density (–30 to –40 Hounsfield units) in the range of adipose tissue. Extension outside the thyroid bed can involve the upper mediastinum and the retropharyngeal compartment, compressing or displacing the surrounding airway and vascular structures.

FNAC can suggest a diagnosis of thyroid lipomatosis, although there are no clear guidelines to establish a diagnosis of thyroid lipomatosis with this method.⁹

In the gross examination of thyroid specimen, cut section reveals a surface with a pale grey, yellowish, or tan color and, in some goiters, a focal cystic degeneration. Some of the published cases have reported massively enlarged thyroids with masses of 500–700 g. Microscopic examination confirms the diffuse infiltration of mature adipose tissue among normal thyroid follicles. Fibrosis and lymphocytic infiltration have also been occasionally described.

Our patient is a known case of renal transplantation for amyloidosis like in few other case reports. 4,9,13 He presented with rapidly enlarging goitre in a time frame of 3 months and pressure symptoms. It is important to rule out malignant thyroid conditions which also present similarly, with imaging. Vestfrid reported a case of 53 year old woman with papillary carcinoma of thyroid associated with diffuse lipomatosis. 14 CECT neck was performed which showed diffuse fatty infiltration and no features were suggestive of malignancy. Surgical resection followed by gross and microscopic analysis confirmed the diagnosis of diffuse thyroid lipomatosis. Prognosis of the patient is good.

CONCLUSION

This case report calls attention to a rare thyroid disease of diffuse lipomatosis in a middle aged patient with renal transplantation. Although an association between these two conditions has been reported in small number of case reports, the mechanism behind this association is poorly understood. Thyroid lipomatosis should be considered as one of the differentials in patients presenting with progressively enlarging thyroid mass. We also highlight the importance of ruling out thyroid malignancies with increased adipose content which also have a similar clinical presentation and sometimes coexist with thyroid lipomatosis because they are associated with increased morbidity and mortality.

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