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A Case Report on Autoimmune Thyroid Disease (ATD) Associated with Chronic Angioedema

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Authors' contributions

This work was carried out in collaboration among all authors. All authors read and approved the final manuscript.

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Case study

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ABSTRACT

Angioedema may be a rare condition that manifests itself by abrupt localized edema caused by the fluid outflow from blood vessels into surrounding skin and tissue. This case report presents a 54-year old male patient with chronic angioedema (lip) for one year without urticaria. Six months ago, he approached the local clinic and used regular antihistamines by the physician advice. Still, there was no improvement in patient condition. Then he came to the outpatient department of internal medicine of our hospital. Therefore, we have done a series of investigations, in then he diagnosed with Hypothyroidism (TSH 8.05Ulu/ml). Then he has prescribed levothyroxine 25mcg. After one month's review, he examined for Anti-thyroid peroxidase antibodies (Anti-TPO). In that examination, those were positive or elevated (mild). According to our research, this may probably be the first autoimmune thyroid disease associated with chronic angioedema without urticaria or hives.

Keywords: *Angioedema; autoimmune thyroid disease; thyroid-stimulating hormone; hypothyroidism; anti-thyroid peroxidase (TPO) antibodies.*

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1. INTRODUCTION

Angioedema is a vascular reaction of deep dermal/sub cutaneous tissues (or) mucosal/submucosal tissues with localized increased permeability of blood vessels resulting in tissue swelling [1]. It will affect any part of the body, and the most common features are the tongue, eyes, mouth, lips, extremities, and reproductive organs. According to the world allergy organization, urticarial and angioedema have observed nearly 20 per cent of the final population. Angioedema can affect all groups irrespective of age and hospitalizations because angioedema has increased over the years, with most cases in patients sixty-five years mature or older [2].

Angioedema can be mediated by bradykinin and/or mast cell-mediators including histamine [1]. HAE-1 hereditary angioedema due to C1-Inhibitor deficiency, HAE-2 hereditary angioedema due to C1-Inhibitor dysfunction, AAE-C1-INH acquired angioedema due to C1-Inhibitor deficiency, HAE nC1-INH hereditary angioedema with normal C1-Inhibitor levels, either due to a mutation in FXII, ANGPTI, PLG or unknown (HAE-FXII, HAE-ANGPTI, HAE-PLG, HAE-UNK), ACEI-AE angiotensin converting enzyme inhibitor-induced angioedema.

Several proposed mechanisms existed for the occurrence of angioedema; those are allergic, genetic, idiopathic, medication-induced, physically iatrogenic, a cytokine associated, thyroid autoimmune disease-associated, thyroid dysfunction, and acquired [3].its pathological process involves releasing chemical mediators like histamine, bradykinin, or additional vasoactive substances, leading to increased vascular permeability. Moreover, around 40% of patients present each condition of urticaria along with angioedema. The autoimmune process may be responsible for the condition of chronic urticaria or angioedema [4].

ATD is a common organ-specific disease primarily seen in the age of 30 to 50 years of

women. Thyroid autoimmunity can cause various forms of thyroiditis, one in hypothyroidism (Hashimoto's thyroiditis) and another one in hyperthyroidism (Grave's disease).The prevalence of autoimmune associated hypothyroidism is about 0.8/100 in that 95 per cent of patients were women only. The main hallmark of AITD is the formation of antibodies to the thyroid peroxidase (TPO), thyroid-stimulating hormone receptor (TSH R), thyroglobulin (TG) [5].

2. CASE STUDY

A 54-year an old male patient came to the hospital with chronic angioedema (lower lip) for one year without urticaria and hives. Six months ago, he went to the local clinic and used regular antihistamines by the physician advice, but there was no improvement in patient condition. He came to the outpatient department of internal medicine in KIMS hospitals Secunderabad. He had no family history of angioedema, no history of any allergies, and no history of taking any ACE inhibitors. The Patient had hypertension for five years on medication (Cilnidipine 5mg). With a known history of CCBs induced angioedema, the withdrawal of cilnidipine replaced it with Telmisartan 40mg and no improvement was seen in the patient condition. After that, we have done a series of investigations in that he diagnosed with hypothyroidism. TSH found to be 8.05Ulu/ml in the thyroid function test, and it found C1 esterase inhibitor 27mg/dl and total IgE antibodies 66 IU/ml to be normal ranges(21-39 mg/dl and 0-100 IU/ml).so the PatientPatient had prescribed levothyroxine 25mcg. After one month's review, the further examined Patient with TFT and Anti Thyroid peroxidase antibodies (Anti TPO). The PatientPatient found to have TSH and T4 within normal ranges. At the same time, Anti-TPO antibodies had elevated (mild) or positive. Then the PatientPatient had prescribed levothyroxine 50 mcg. After six months, angioedema was slowly comedown or resolved.

Bradykinin-induced AE				Mast Cell Mediator induced AE		Unknown mediator
C1-INH deficiency/ defect		C1-INH normal		IgE mediated	non-IgE mediated	
Inherited	Acquired	Inherited	Acquired			
HAE-1 HAE-2	AAE-C1-INH	HAE nC1-INH (HAE-FXII, HAE-ANGPTI, HAE-PLG, HAE-UNK)	ACEI-AE	Angioedema with AnaphylaxisUrticaria	Angioedema with urticaria	Idiopathic



Fig. A & B. representing the Before and After treatment of angioedema
Fig. A. Showing angioedema present in lower lip before treatment
Fig. B. Showing the difference after the treatment

3. DISCUSSION

We are presenting the case of autoimmune thyroid disease associated with chronic angioedema in the 54-year-old male Patient who had no history of urticaria or allergies and no family history of angioedema.

Abnormality of the thyroid gland and a few inflammatory diseases are very familiar in persons with urticarial and angioedema. In some persons, these reactions or swellings having many years unaccompanied by an unknown cause. In these cases, the autoimmune condition is responsible for urticarial/angioedema. Angioedema without urticaria occurs in 10-20% of patients. Angioedema can be chronic, presented over weeks to months or sometimes years. Chronic angioedema that occurs without urticarial or hives may be hereditary or acquired angioedema [6].

According to Missaka, The occurrence rate of angioedema was more in patients who had thyroid abnormalities compared to patients with normal thyroid gland function. Autoimmune thyroid disease patients are 16.2 times more prone to cause angioedema. Among the patients with hypothyroidism and hyperthyroidism, the percentage of getting angioedema respectively 4.6&3.3.but, these percentages were not statistically significant [7].

Thyroid autoantibodies, markers for the finding of autoimmune thyroid disease, has recognized in 3-9% of the general population. Thyroid autoantibodies identified in 23.4% of patients had chronic urticaria [8]. Based on a broad literature survey, no examinations or case reports had discovered a relationship between hypothyroidism or thyroid autoimmunity and angioedema in patients who don't have simultaneous chronic urticaria [9].

As of late, considers have shown elevated degrees of IgE anti-thyroid autoantibodies in some chronic urticaria patients. Ongoing investigations have featured the cover of immunological associations that assume a part in the pathogenesis of both chronic urticaria and autoimmune thyroid conditions. However, this proposed mechanism does not apply to our case study because the Patient had no urticaria or hives [10].

4. CONCLUSION

This case report describes chronic angioedema with hypothyroidism and autoimmune thyroid disease without urticaria or hives. We suggest that if the Patient had angioedema, it is necessary to testing thyroid gland function when the cause is unknown. The limitation of the study is we do not know the exact cause of angioedema, and only thyroid abnormalities can be responsible for the pathogenesis of angioedema alone. So a need for further

research on the pathogenesis of angioedema related to the thyroid gland.

INFORMED CONSENT

The authors declared that obtained informed consent from the patient for his images and data published in this case report. And authors guaranteed to the patient that his initial and name and personal details were kept confidentially.

ETHICAL APPROVAL

It's not applicable.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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