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Case Report

Coexistence of follicular variant of papillary thyroid carcinoma and oncocytic adenoma on the background of Hashimoto's thyroiditis: Case report

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Abstract

Papillary thyroid carcinoma (PTC) is the most common histologic type of thyroid cancer comprising of 80–90% of all subtypes with female predominance. Hashimoto's thyroiditis (HT), also synonymous with chronic autoimmune thyroiditis and chronic lymphocytic thyroiditis, is an autoimmune disease characterized by immune destruction of thyroid cells mediated via cell and antibody dependant immune processes. Dailey et al. first described the association between PTC and HT in 1955. Approximately 30% of PTC have HT in their background. Several studies have reported HT as a risk factor for the development of PTC. Oncocytic adenomas (OA) comprise 10-15% of thyroid nodules and more common in men. We are reporting this case of 29 year old female having multifocal infiltrative follicular variant of PTC and Oncocytic adenoma with marked cytological atypia in the background of HT as it is very rare to encounter this coexistence except in the context of tumour syndromes.

Keywords: Papillary carcinoma, Oncocytic, Adenoma, Hashimoto's thyroiditis.

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1. Introduction

HT is a destructive tissue-specific autoimmune disease of the thyroid gland, characterised by widespread lymphocyte infiltration, fibrosis and parenchymal atrophy. The disease usually manifests as hypothyroidism, with reduced levels of T3 and T4 and elevated TSH levels. Dailey et al. first described the association between PTC and HT in 1955. Approximately 30% of PTC have HT in their background.²⁻⁵ The causal association between HT and PTC was a hot topic of discussion during the past 20 years. Many studies investigated the influence of coexistent HT with PTC on its prognostic outcomes and the association clinicopathological features.

Oncocytic adenomas are non-invasive, encapsulated tumours composed of Hürthle cells with microfollicular or solid to trabecular architecture, affecting the elderly males. Hürthle cells may be seen in a wide variety nonneoplastic lesions of thyroid gland including, thyroid follicular nodular disease and lymphocytic thyroiditis. Coexistance of HT and

Oncocytic adenoma is extremely rare despite the presence of frequent hurtle cell metaplasia in HT.

Because of extreme rarity of coexistence of OA and PTC in the background of HT, we are presenting this case report.

2. Case Report

A 29 year old female presented with complains of midline neck swelling in the surgical oncology department of Gujarat Cancer and Research Institute. She gave history of previous surgery 10 years back with similar complaints for which she went right partial thyroidectomy at some other centre. Outside report suggested Papillary Thyroid carcinoma.

Computed tomography revealed right lobe measuring 21x13x40mm. Left lobe of thyroid appeared bulky and measured 30x33x47mm. Both the lobes of thyroid gland and isthmus show internal areas of hypodensities. Enhancing nodes were noted at level IV.

*Corresponding author: Vibha R Vyas Email: vibha.vyas@gcriindia.org FNAC was done from both right and left lobes of the thyroid. Left lobe fine needle aspiration findings were suspicious of malignancy. Right lobe aspiration findings were suggestive of atypia of undetermined significance. Patient underwent total thyroidectomy with left side neck node dissection and the specimen sent to oncopathology department for histopathological examination.

Gross examination: Received total thyroidectomy specimen with right and left lobes in separately labelled containers. Left lobe of the thyroid measured 5.5x4x3 cm and showed lobulated cut surface with a small whitish nodule measuring 0.6x0.5 cm [Figure 1a]. Right lobe comprised of a well encapsulated nodule measuring 4.5x2.2x2.0 cm. Cut surface of the nodule was tan brown to mahogany with focal area of hemorrhage may be due to prior FNAC procedure [Figure 1b]

Microscopic examination: On histopathoogical examination left lobe of thyroid revealed moderate to dense lymphocytic infiltrate with germinal centre formation and multifocal infiltrative follicular variant of PTC. The neoplastic cells revealed elongation, overcrowding, optically clear nuclei with intranuclear grooves and pseudoinclusions. Immunohistochemistry with CK19 beautifully highlighted the sprinkling of neoplastic follicles amongst non-neoplastic ones [Figure 2a, b]

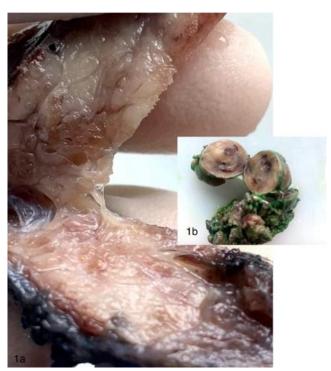


Figure 1: **a:** Gross photograph of left lobe showing lobulation and glistening areas; **b:** Cut surface of residual right lobe harbouring adenoma with solid tan appearance.

Right lobe of thyroid which was previously operated for PTC at some outside centre, now showed a well encapsulated

oncocytic adenoma exhibiting predominantly follicular pattern of growth with cells having abundant eosinophilic granular cytoplasm and marked cytological atypia without any evidence of mitosis and necrosis. Lesion was entirely submitted and many deeper sections were examined to rule out capsular invasion. [Figure 2c &d]

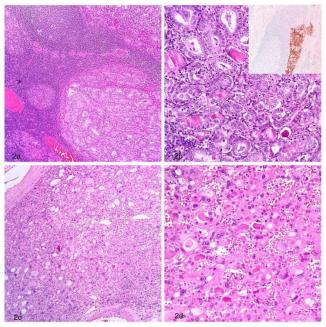


Figure 2: a & b: Photomicrograph revealing foci of follicular variant of PTC with characterstic nuclear features (H&Ex100 and 400 magnification); inset, shows CK19 immunostain positivity in neoplastic follicles; **c** & **d:** Photomicrograph revealing encapsulated Oncocytic adenoma exhibiting endocrine atypia (H&E x 100 &400 magnification).

3. Discussion

Malignant thyroid neoplasms constitute the most common malignancies of endocrine system.⁶ Papillary thyroid carcinomas (PTCs) comprise approximately 70% to 80% and exhibit a relatively benign clinical course.^{7,8} The most common thyroidal autoimmune disease is HT, being considered as a destructive tissue-specific autoimmune disease with detectable thyroid peroxidase antibodies (TPOAb) and thyroglobulin antibodies (TgAb). HT results in diffuse lymphocyte infiltration of thyroid parenchyma, fibrosis and loss of thyroid parenchymal tissue, leading to hypothyroidism, which is characterised by a deficit of T3 and T4 and elevated TSH levels. The estimated worldwide incidence of HT is approximately 0.3-1.5 cases per 1000 individuals.9 HT is the most common cause of primary hypothyroidism and non-endemic goiter, with 10-15 times higher incidence in women.1

Hashimoto thyroiditis is classically associated with extensive Hürthle cell metaplasia in a background of dense lymphocytic infiltrate with germinal center formation and admixed plasma cells. Hürthle cells also may be seen in the setting of other inflammatory or reactive conditions,

including Graves' disease, following irradiation to the head and neck or therapy with Iodine. ¹⁰

Oncocytic adenoma previously known as Hurthle cell adenoma occurs in 5^{th} to 6^{th} decade of life and more common in males. The overall incidence of OA is difficult to assess because of histologic overlap between OA and oncocytic adenomatous nodules and combination of OA with follicular adenoma in many previous studies. According to 5^{th} edition WHO, OA comprise about 10-15% of thyroid nodules with a preceding indeterminate fine needle aspiration (FNA) diagnosis OA are more frequently diagnosed in women than in men. 11,12

Despite the frequent occurrence of oncocytic change / hurthle cell change in HT, the association of OA has not been reported in HT.

OA has no exogenous risk factors. Very rarely, OA occurring in context of tumour syndromes, including Cowden syndrome (most frequently associated with a germline *PTEN* mutation) and Carney complex (most frequently associated with a germline *PRKAR1A* mutation) are seen in younger women. ^{13,14} Oncocytic adenoma is a benign tumor and excision is curative.

Several studies indicated that HT is associated with a significantly higher risk of PTC. The coexistence of HT in PTC patients is associated with favourable clinical outcomes compared to PTC without HT.^{1,15,16}

Coexistence of PTC, HT and OA has been rarely described in literature. To establish causal relationship between OA and HT and their prognostic significance more case reports or studies are needed.

4. Conclusion

Although association between HT and PTC is well established in literature stating increased incidence of PTC with favourable outcome in background of HT, occurrence of OA is difficult to explain due to rarity of this disease and lack of literature.

5. Source of Funding

None.

6. Conflict of Interest

None.

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