CASE REPORT

Hyalinizing Trabecular Adenoma of Thyroid: A Rare Case Report

Neha Singh[1], Jitender Singh Chauhan[2]

ABSTRACT

Introduction: Hyalinizing trabecular tumor (HTT) is an unusual thyroid neoplasm that was first described by Carney in 1987.Many cytological and histological features of such tumors may be mistaken for Papillary Thyroid Carcinoma (PTC) and Medullary Thyroid Carcinoma (MTC).This can lead to overtreatment like total thyroidectomy and lymph node dissection. Here, we present a rare case of hyalinizing trabecular tumor of thyroid in a young female emphasizing the pathological features of HTT, which may help to reduce over diagnosis thereby guiding proper management.

Case report: A 40 year old female patient presented with a lump in the left side of the neck. Lump increased gradually over a period of 5 months and was associated with mild pain and restricted neck movement. Thyroid profile of the patient was within normal limits. USG revealed a solid cold nodule, which was regarded as a thyroid adenoma. Nodule was surgically removed and after pathological examination diagnosed as hyalinizing trabecular adenoma (HTA) of thyroid.

Conclusion: Hyalinizing trabecular adenoma is rare and can be confused with malignant lesions of thyroid. Some peculiar histomorphological features mentioned in this case report will help in diagnosis and further management.

Keywords: Hyalinizing Trabecular Adenoma, Rare, Thyroid

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[1]Senior Resident, Department of Pathology, B.P.S. Government Medical College, Khanpur Kalan, Sonipat, Haryana, [2]Resident, Department of General Surgery, PGIMS Rohtak, Haryana, India.

Corresponding author Dr. Neha Singh, Senior Resident, Department of Pathology, B.P.S. Government Medical College, Khanpur Kalan, Sonipat (Haryana).

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INTRODUCTION

HTA is a rare neoplasm of thyroid with follicular derivation which commonly arises in middle-aged women.[1,2] HTA clinically presents as a asymptomatic well-circumscribed solitary thyroid nodule, a prominent nodule in a multinodular goitre or as an incidental finding in a thyroidectomy specimen.[3,4,6] The most controversial issue is that whether HTT is a benign tumor or a variant of PTC. HTT and PTC both have similar nuclear morphology, cytology, immune profile and RET/PTC oncogene rearrangements. [5,7,9] Frequently occurring BRAF (heterozygous V600E) and NRAS mutations, in PTC, are constantly absent in HTT.[6,8] Galectin-3 is expressed in both PTC and HTT.[6] As compared to PTC, HTT shows negative immunostaining for cytokeratin 19 and high molecular weight (HMW) cytokeratin.[9] In contrast to other thyroid neoplasm, HTT shows characteristic strong cytoplasmic and membranous immunostaining of tumor cells with MIB-1.[1,9] These results are against HTT as a variant of PTC. The low Ki-67 index and negative p53 immunostaining are consistent with the benign behavior of HTT.[4] According to some authors, HTT is not a distinct entity because trabecular growth pattern can be seen in follicular adenoma, PTC, MTC, and metastatic neuroendocrine tumors of the thyroid gland.[3,6,7] Few HTA are associated with chronic lymphocytic thyroiditis, hashimoto's thyroiditis, follicular neoplasm, multi nodular goitre and PTC.[12]

The pre-operative diagnosis of HTT is difficult as clinical and ultrasonographic features are non-specific. In such instances, FNAC is strongly indicated for exact categorization of lesion, which determine subsequent management.[6] However the diagnosis cannot be confirmed by FNAC due to

lack of specificity.[11,12] In our case also, FNAC was noncontributory. HTT may be misdiagnosed as either suspicious or positive for malignancy like PTC [TBSRTC (The Bethesda system for reporting thyroid cytopathology) category V or VI] because of the similar nuclear features specially intranuclear cytoplasmic inclusions and grooves.[10,13] These represent a potential diagnostic pitfall. In this case, nuclear inclusions, grooves and overlapping were occasionally evident and not significant enough to consider them for PTC. Cytological features with trabecular pattern of cells, vague curved nuclear palisading, spindled or elongated cells, abundant cytoplasm with ill-defined border and hyaline material in our case prevent the over diagnosis as PTC. Trabecular growth pattern can also be seen in trabecular variant of PTC, but the distinct hyalinized stroma is not seen in trabecular variant of PTC.[7] HTT can also pose diagnostic difficulty with MTC especially hyalinizing trabecular adenoma-like or paraganglioma-like variant.[1,5,9] Utmost care should be taken to distinguish amyloid and colloid from hyaline material.

CASE PRESENTATION

A 40 year old female presented with a gradually increasing lump in the left side of the neck with mild pain and restricted neck movement. Thyroid profile was within normal limit. USG revealed a solid cold nodule, which was regarded as a thyroid adenoma. Left hemithyroidectomy was done and sent for pathological examination. Gross examination showed a lobe of thyroid measuring 7×5×2.5 cm. Cut surface showed encapsulated grey firm tumor measuring 5×4×1.5 cm. Remaining thyroid parenchyma was unremarkable (Figure 1).

Microscopically, the lump was encapsulated with trabecular structures separated by minimal fibrous stroma. It was characterized by long, wavy and coiled trabeculae of elongated to polygonal cells with lightly eosinophilic cytoplasm. The elongated tumor cells were aligned perpendicularly in the trabeculae. There were frequently interspersed microcystic spaces representing abortive or true follicle formation. Some oval nuclei exhibited fine chromatin, mild pleomorphism, nuclear grooves, pseudoinclusions and perinuclear halos. The delicate fibrovascular stroma tends to undergo hyalinization. Mitosis & psammoma bodies were rare or absent. Adjacent thyroid tissue showed hashimoto's thyroiditis (Figure 2).

Immunohistochemically, this was positive for thyroglobulin and thyroid transcription factor-1(TTF-1). After a follow-up of 2 yrs, the patient was alive without local recurrence or metastasis.

DISCUSSION

HTT is a unique neoplasm of follicular derivation The most puzzling issue regarding HTT concerns its potentially malignant behavior and the possible relationship to PTC. HTT is a benign entity and all cases of HTT with classic morphology fail to show unequivocal capsular and/or vascular invasion and have not metastasized as described by Carney et al.[1,14]

In addition, some authors have also suggested that this is not a distinct entity because a similar growth pattern can be seen in other primary and secondary tumors of the thyroid. Because of this debate, most experts designate these tumors as HTT.[15-17]

HTT occur as solitary or multiple circumscribed and encapsulated nodules. Microscopically it shows classic trabecular pattern and intratrabecular hyalinization of matrix. Pseudofollicles, intranuclear cytoplasmic inclusions and grooves are also common.[13]

On immunohistochemistry, thyroglobulin and thyroid transcription factor (TTF)-1 are positive in HTT.[3,6] Calcitonin, HBME-1, synaptophysin, chromogranin, epithelial membrane antigen and vimentin are usually negative in HTT.[10] The proliferating cell nuclear antigen (PCNA) expression is high but its significance is uncertain.[4] In cytologic smears, cytoplasmic and membranous expression of MIB-1 is useful in establishing the diagnosis of HTT.[18] However, negative MIB-1 stain has no diagnostic value. Some authors have proposed a cytological distinction between HTT and papillary carcinoma of the thyroid, but its cytological diagnosis remains challenging. [5,13,19]

According to Kuma et al.[13] radial arrangement of the tumor cells surrounding the hyaline material, vague, curved nuclear palisading, spindled or elongated cells, ill-defined cell border, faintly

stained, filamentous cytoplasm, and hyaline mat-

Figure-1: Gross photograph of the specimen exhibiting well circumscribed grey firm tumor mass. Surrounding thyroid parenchyma is also noted.

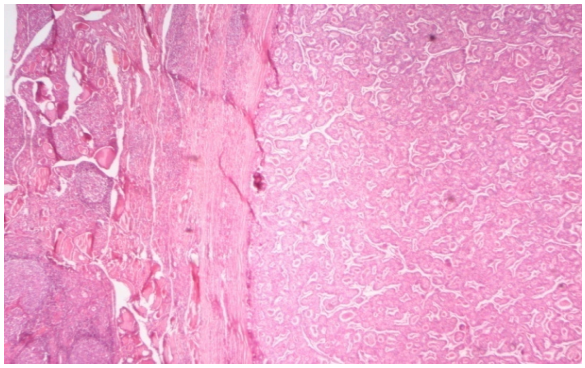

Figure-2: 10x H&E stained section shows capsulated tumor with long wavy coiled trabeculae & frequently interspersed microcystic spaces represent abortive & true follicle formation. Remaining thyroid shows changes of thyroiditis.

erial in the background are useful in diagnosing HTT and distinguishing it from papillary carcinoma. A lack of papillary architecture and sheet-like arrangement also suggests HTT.

HTT can be distinguished from MTC by Congo red negativity, positive thyroglobulin immunoreactivity, and negative calcitonin immunoreactivity.[5]

Almost all cases of HTT fail to show unequivocal capsular, vascular, and stromal invasion and have not metastasized, suggesting a benign behavior.[7,8,14] Very rarely HTT can show vascular and capsular invasion, and pulmonary metastasis.[12] Misdiagnosis of HTT as malignancy can lead to total thyroidectomy and lymph nodes dissection.[2,8,12] HTT without signs of metastases does not need aggressive treatment like total thyroidectomy or radio-iodine ablation.[6] Hemithyroidectomy would have been a better option if cytology is suggestive or suspicious for HTT. To exclude the very rare possibility of recurrence and metastasis, annual follow-up is required .[6,14,20]

CONCLUSION

HTT is a benign neoplasm or at most, a neoplasm of extremely low malignant potential. HTT is a challenging entity on pathology due to overlapping features with malignant thyroid lesions. Differentiation of HTT from PTC and MTC is important due to altogether different prognosis and therapeutic implication. Pathological diagnosis of HTT should be considered in presence of trabecular pattern of cells, vague curved nuclear palisading, radiating arrangement of cells around hyaline material, spindle to elongated cells, filamentous cytoplasmic processes with ill-defined cell border. Nuclear overcrowding, grooves and inclusions should be correlated with other features and evaluated with great care to differentiate HTT from PTC. Close attention is required to evaluate the hyaline material as it can mimic amyloid and colloid.

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