Case Report

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Primary Thyroid Schwannoma with Papillary Carcinoma and Hurtle Cell Adenoma in a Background of Hashimoto's Thyroiditis: A Very Rare Occurrence

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ABSTRACT

Schwannomas are common benign neurogenic tumours that arise from Schwann cells of nerve roots; up to 45% arise in head and neck. However primary thyroid schwannomas are extremely rare. Among thyroid tumours papillary carcinoma of thyroid (PCT) are the most common. Hurthle cell neoplasms arealso rare.In addition occurrence of all the three neoplasms in the same patient is extremely rare. In this report we discuss a case of left lobe intrathryroidal schwannoma with papillary carcinoma of thryroid (PCT), Hurthle cell adenoma and background Hashimoto thyroiditis in a 25 year old female. On FNAC it was diagnosed as colloid nodule with cystic change, total thyroidectomy was done and histopathological examination showed that the largest well circumscribed nodule wasa benign neurogenic tumour, schwannoma (left lobe of thryroid gland). There were other smaller thyroid nodules; one showing papillary carcinoma with lymph node metastasis and other showing hurthle cell adenoma with backgroundHashimoto's thyroiditis.

Keywords: *Schwannoma, Papillary Carcinoma Thyroid, Hurthle Cell Adenoma, Multiple Thyroid Nodules.*

Introduction

Schwannoma is a benign biphasic tumour arising from nerve sheath cells discovered by Verocay in 1908.[1] In 90 % of cases, it originates from vestibular nerve.[1] Lesser than 25 cases of primary thyroid schwannomas reported till now in literature.[2] Primary thyroid schwannoma is identified as a neoplasm arising in thyroid and showing characteristic histologic features like alternating cellular and compact Antoni A and loosely arranged Antony B areas with Verocay bodies and diffuse nuclear and cytoplasmic staining for S-100 protein.[1]

Papillary carcinoma of thyroid (PCT) is the most common thyroid tumour, approximately 80-85% of all tumours.[3] Among Hurthle cell tumours, Hurthle cell adenomas are more common than the malignant counterparts.[4]

An intrathyroidal schwannoma with PCT, Hurthle cell adenoma and Hashimoto's thyroiditis in the same patient is extremely rare. We didn't encounter even a single case in spite of extensive literature search. PCT was discovered incidentally in our case. The frequency of incidental PCT in patients operated for benign thyroid diseases is reported to be 4.6% to 10%.[5]

Case Report

A 25 year old female patient presented with swelling over anterior surface of neck since 4 years, the swelling was gradually progressive with sudden increase in size and pain over the swelling since one month, she also complained of dysphagia and odynophagia with change in voice since one month, there were no other complaints.

On examination there was a swelling over anterior surface of neck firm to hard, 4 cm x 3 cm involving left lobe of thyroid extending from the midline.

Her hematological and biochemical investigations were normal. Ultrasonography of the thyroid gland showed enlarged gland with multiple well marginated round iso-hyperechoic nodules in both thyroids. Impression was multinodular goitre. Cytology of the left thyroid nodule showed paucicellular smear showing occasional thyroid follicular cells with cyst macrophages and fibroblast in a background of thin colloid mixed with blood, malignant cells were not seen. Impression given was a colloid nodule with cystic degeneration. Thyroid function tests were within normal limits. Thyroglobulin antibody (ATG) was increased to 589 (<115.00 IU/ml).

Gross pathology: We received a total thyroidectomy specimen. The right lobe measured 5.5 x 1.5 x 1.5, thyroid capsule was intact, external surface was unremarkable. Cut section showed an irregular unencapsulated grey white lesion measuring 0.8 x 0.7 x 0.6 cm. The left lobe measured 6 x 2.5 x 2.5, thyroid capsule was intact, and a nodule was identified in the upper lobe externally.

Cut section showed 2 nodules, larger measuring 2 cm in diameter was, it was grey white. Smaller nodule was 0.8 cm in diameter well circumscribed and grey brown in colour. Isthmus measured 2.5 x 1.5 cm, normal. A lymph node tissue was received separately 0.5 cm in diameter and grey white in colour (figure 1).

Histopathology: Multiple sections studied from the total thyroidectomy specimen revealed an encapsulated nodule in the left lobe composed of spindle shaped cells arranged in fascicles and bundles with occasional hypocellular areas. The cells were spindle shaped, had slightly wavy nuclei and abundant eosinophilic cytoplasm, large areas of haemorrhage and dense mixed inflammatory infiltrate, tumour also showed many proliferative blood vessels, few scattered hemosiderin laden macrophages. This nodule was diffusely positive for S-100, negative for SMA (figure 2).

The nodule from right lobe revealed a tumour with cells arranged in papillary configuration. Individual tumour cells had large round to oval nuclei with features of papillary carcinoma. The tumour cells were infiltrating in to the surrounding thyroid parenchyma, thyroid capsule was not breached (figure 3). Background showed Hashimoto's thyroiditis with a small well demarcated Hurthle cell adenoma (figure 4). Lymph node sent separately revealed metastasis of papillary carcinoma. Impression was Schwannoma of the left lobe with papillary carcinoma of thyroid of right lobe in a background of Hashimoto's thyroiditis with a Hurthle cell adenoma. Lymph node showed metastasis of PTC.

Discussion

Primary spindle cell neoplasm arising from thyroid gland are rare accounting for less than 1% of cases.[1] Isolated cases of lipoma, hemangioma, angiolipoma, lymphangioma, leiomyoma, schwannoma, hemangiopericytoma, and solitary fibrous tumor have been reported.[6] Among primary malignant spindle cell tumours of thyroid liposarcoma, leiomyosarcoma and angiomyosarcoma are mentioned in the literature.[7]

In 1964 Delaney and Fry in first reported a neurilemmoma arising in the thyroid gland.[8] They are thought to arise from the intrathyroidal sensory nerves or sympathetic and

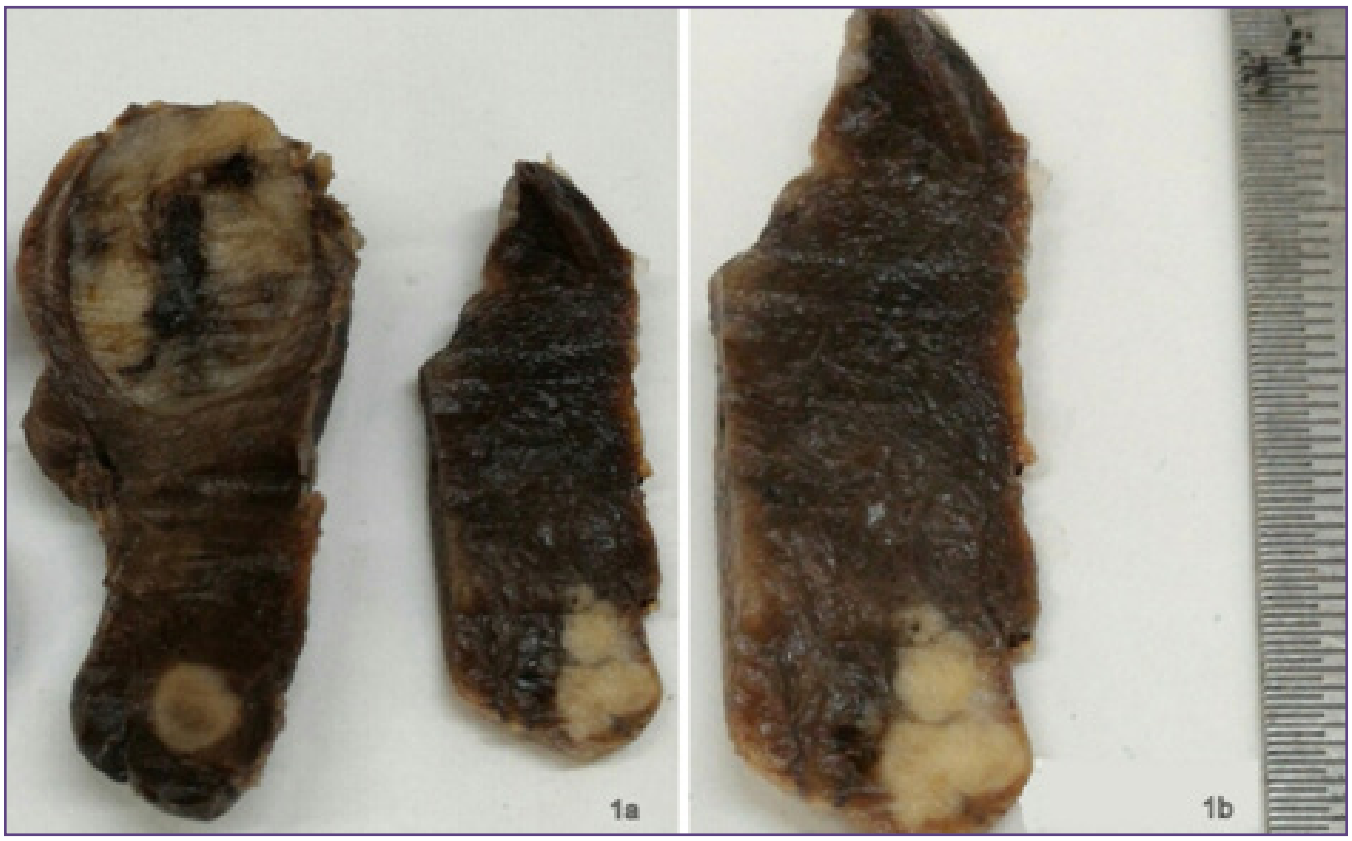

Fig. 1a: The left lobe (left side) revealed 2 nodules, larger measuring 2 cm in diameter was, it was grey white, with central congestion. Smaller nodule was 0.8 cm in diameter well circumscribed and grey brown in colour., Fig. 1b: The right lobe (right side of picture) showed an irregular non encapsulated grey white lesion.

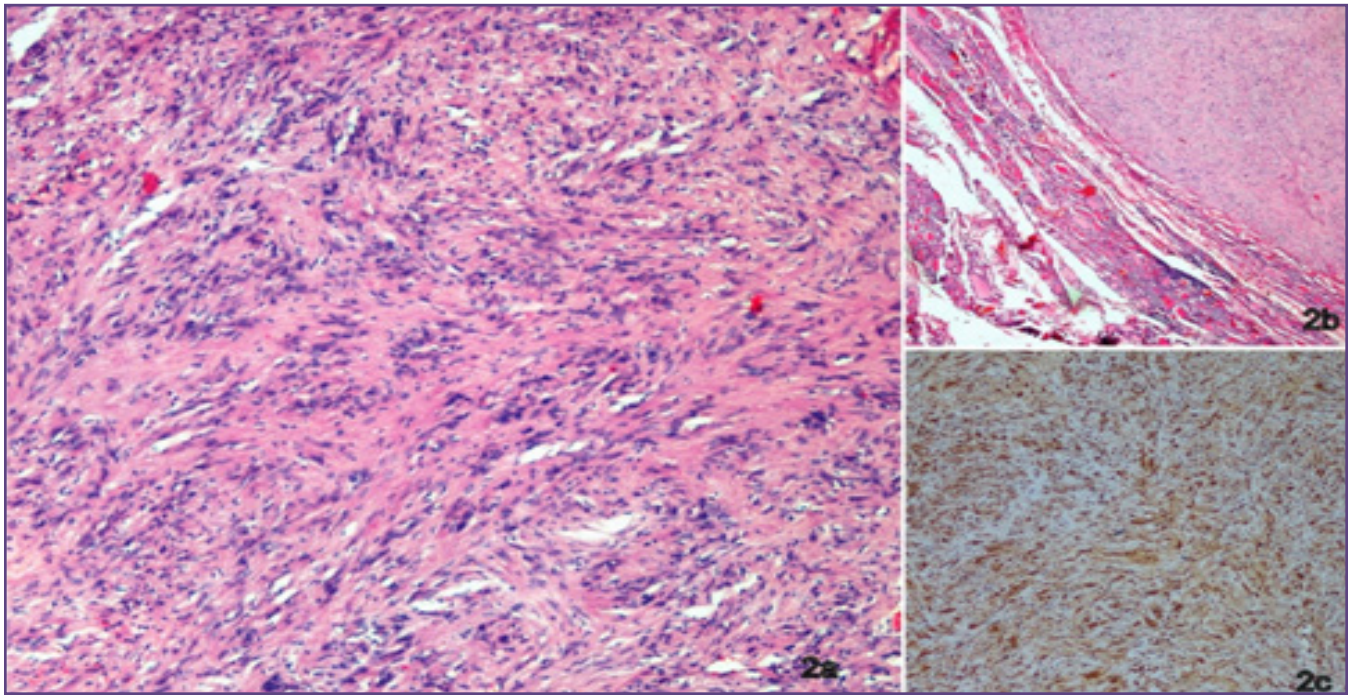

Fig. 2a (10x) and 2b (4x): An encapsulated nodule composed of spindle shaped cells arranged in fascicles and palisadesthe cells were spindle shaped, had slightly wavy nuclei and abundant eosinophilic cytoplasm. Figure 2c (10x): Diffuse positivity for S-100.

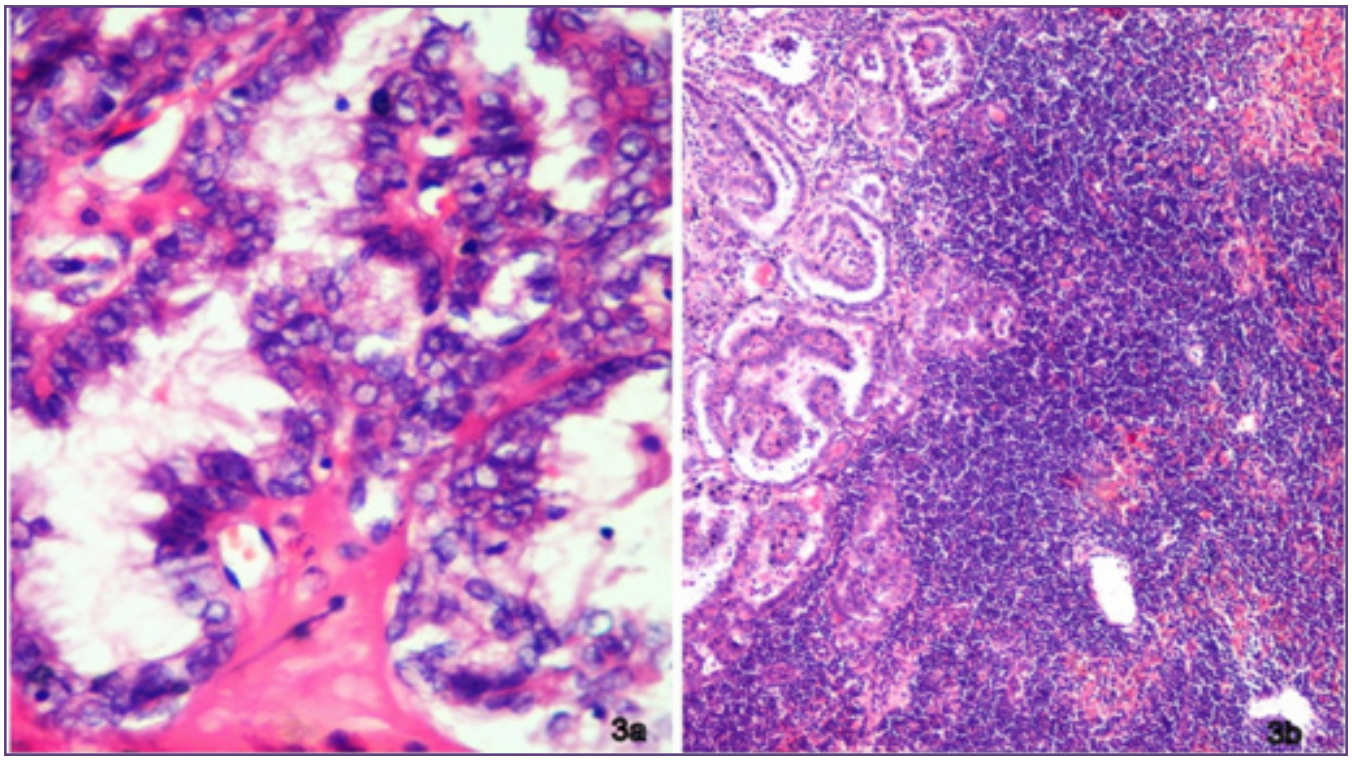

Fig. 3a(40x): The nodule from right lobe revealed a tumour with cells arranged in papillary configuration. Tumour cells had large round to oval nuclei with features of papillary carcinoma. Infiltration in to the surrounding thyroid parenchyma seen, thyroid capsule was not breached. Fig. 3b(10x): Background showed Hashimoto's thyroiditis.

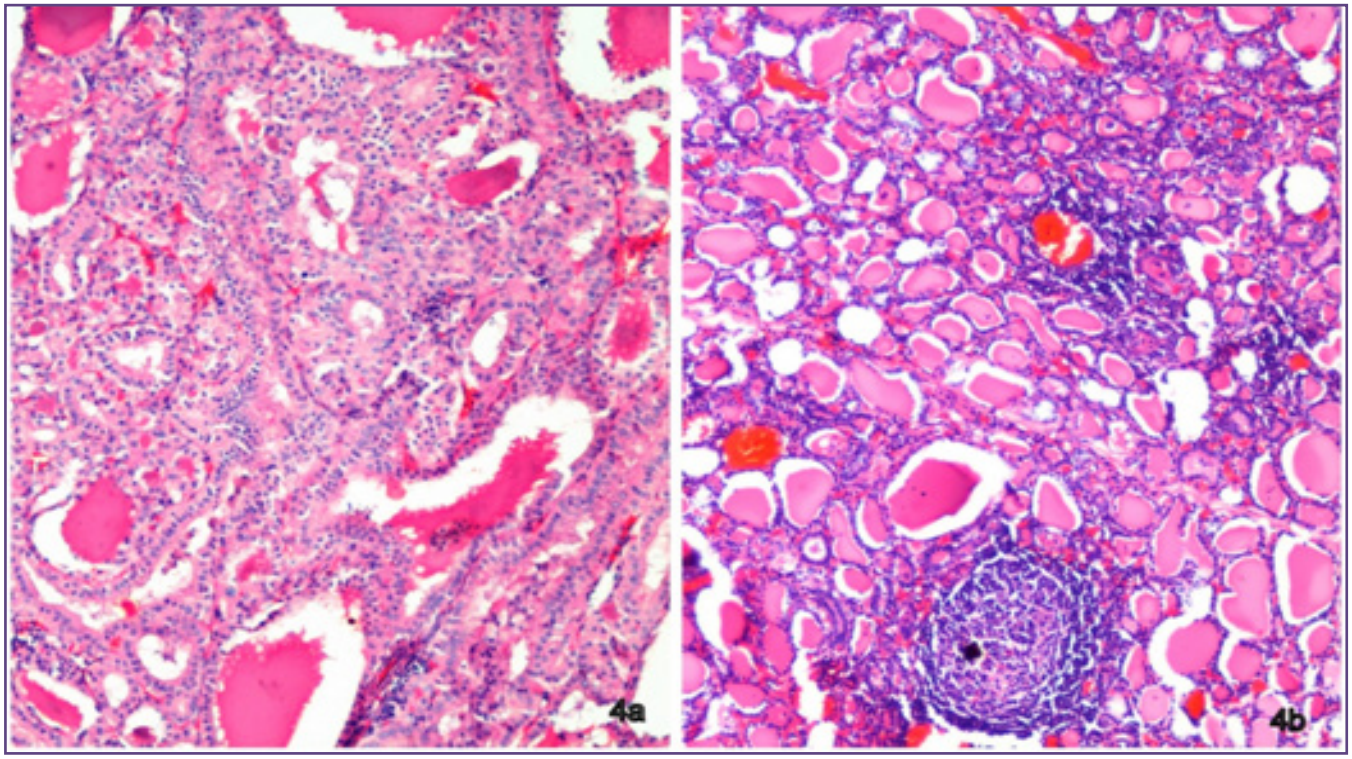

Fig. 4a (10x): Hurthle cell adenoma, figure 4b (4x): Hashimoto's Thyroiditis.

parasympathetic innervations.[6] In a study by lee et al[2] in 2015 there were 20 reported cases of primary schwannoma of thyroid gland. In our review up to 2018 we found 24 case reports and 6 cases were from India.

According to a study by Agrawal et al[6] most of these patients were between 30 to 50 years of age and presented with asymptomatic neck swelling. On USG most of them were hypoechoic nodules with variable cystic degeneration. In review of previous case reports by Dhar et al[9] only three cases were diagnosed as Schwannoma by FNAC (USG guided) while most of the cases were reported either as colloid goitre with cystic degeneration or as paucicellular aspirates with scattered spindle cells and labelled inconclusive.

In the current case a 25 year old female patient radiological diagnosis was multinodular goitre and FNAC diagnosis was colloid nodule with cystic changes after studying the thyroidectomy specimen three thyroid nodules with different morphologies i.e. intrathyroidal schwannoma , papillary carcinoma thyroid and hurthle cell adenoma were identified. The largest nodule showed benign neurogenic tumour which was strongly positive for S-100 and diagnosis was given as schwannoma on the basis of histomorphology and IHC. In previous studies 12 cases were positive for S-100 and 5 cases were positive for vimentin.

The study of various literature shows that this is the only case where there is intra thyroidal schwannoma with papillary carcinoma thyroid with lymph node metastasis and Hurthle cell adenoma with background Hashimoto's thyroiditis. In a study by Repplinger et al[10] HT is associated with an increased risk of developing PTC. In a case report by Yildirim et al[11] concurrent papillary carcinoma thyroid and malignant epithelioid peripheral nerve sheath tumour were seen in post irradiation case of Hodgkin's disease. Though primary spindle cell tumour of thyroid (spindle cell variant of PTC) are can rarely cause confusion but they will be positive for TTF1 and negative for S100.[12]

Conclusion

Primary schwannomas of thyroid gland are very rare; as per our search this is the only case with primary schwannoma of thyroid gland with papillary carcinoma of thyroid (with lymph node metastasis) and Hurthle cell adenoma in a background Hashimoto's thyroiditis. The significance of studying this case is that multiple thyroid nodules with different lesions can easily be missed on FNAC, hence the need of detailed histopathological examination of total thyroidectomy specimen is essential .Also the possibility of a non epithelial neoplasm arising from the thyroid gland should be considered.

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