

Case Report

Bilateral Hurthle cell adenoma of thyroid: a rare case report

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ABSTRACT

Hurthle cell of adenoma of thyroid gland is rare neoplasms of the thyroid. Size more than 4 cm is rare finding and moreover the occurrence of a multifocal Hurthle cell adenoma is not supported by antecedents in literature. Sonography fails to identify its potentials for malignancy while fine needle aspiration cytology couldn't differentiate it from Hurthle cell carcinoma of thyroid. The management of Hurthle cell adenoma and Hurthle cell carcinoma is quite different and hence diagnostic dilemma should be sorted out early. A 26 year old female presented with bilateral thyroid swelling. Ultrasonography of thyroid gland showed complex solid cystic nodules in bilateral thyroid lobes with right thyroid lobe measuring 4.1×3.4×2.5 cm and left thyroid lobe measure 3.1×1.7×1.6 cm. Fine needle aspiration cytology (FNAC) was suggestive of papillary malignancy with extensive Hurthle cell changes (Class V, Bethesda classification). The patient underwent total thyroidectomy with bilateral parathyroid gland preservation. Histopathology revealed it as Hurthle cell adenoma (HCA) involving bilateral lobe and multifocal in nature. The treatment of choice for Hurthle cell carcinoma is total thyroidectomy with neck dissection depending on the nature of the lesions. However the treatment of Hurthle cell adenoma is only hemithyroidectomy or lobectomy as it is a benign condition. Differentiation of these two entirely different conditions warrants more studies.

Keywords: Young female, Bilateral thyroid swelling, Bilateral Hurthle cell adenoma, Total thyroidectomy

INTRODUCTION

Hurthle cells were first described by Askanazy in 1898; however, they were mistakenly named after the German physiologist Karl Hurthle, who actually described the interfollicular C-cell.¹

Hurthle cell tumors of the thyroid are rare neoplasms accounting for less than 5% of all thyroid tumors.² These tumors have associated with various benign thyroid conditions like Hashimoto's thyroiditis, hyperthyroidism, nodular goiter, and thyroid neoplasms; where they often represent oncocyctic metaplasia.³

The report by Thompson et al suggested that all Hurthle cell neoplasms (HCNs) were potentially malignant and, as a result, all such tumors required resection.⁴ However, more recent documented studies have suggested that this

is not true; lesions without complete capsular penetration or vascular invasion can reliably be diagnosed as benign. Moreover, no specific imaging characteristics are shown by these Hurthle cell neoplasms.

CASE REPORT

A 26 year old female presented with a thyroid swelling involving bilateral lobes. Her thyroid profile was within normal limit. Ultrasonography of thyroid gland showed complex solid cystic nodules in bilateral thyroid lobes with right thyroid lobe measuring 4.1×3.4×2.5cm (18.5 cc) and left thyroid lobe measure 3.1×1.7×1.6 cm (4.7 cc). FNAC revealed features highly suspicious for papillary malignancy with extensive Hurthle cell change and cystic degeneration (Category V, Bethesda classification).⁵

The patient underwent total thyroidectomy with bilateral parathyroid gland preservation. Histopathological evaluation of the specimen showed an encapsulated tumor composed of variably sized, predominantly large follicles lined by Hurthle cells as shown in Figure 1-3. No significant nuclear grooving or nuclear clearing was seen. No vascular or capsular invasion was noted as given in Figure 4. Sections from left thyroid lobe revealed two similar small lesions. Sections from surrounding thyroid parenchyma were unremarkable.

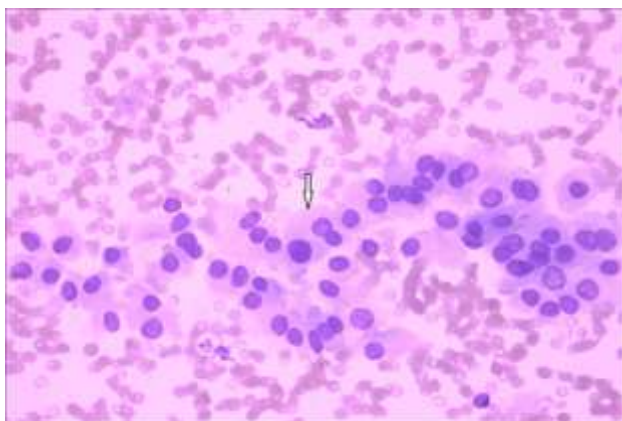


Figure 1: Sheets of Hurthle cells (arrow marked). Background shows RBC'S.

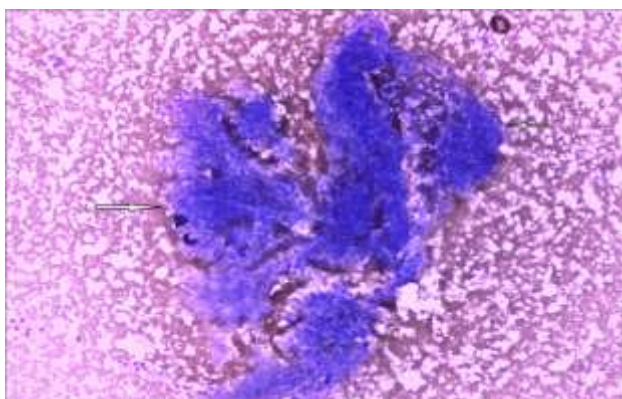


Figure 2: Papillary cluster of Hurthle cells (arrow marked).

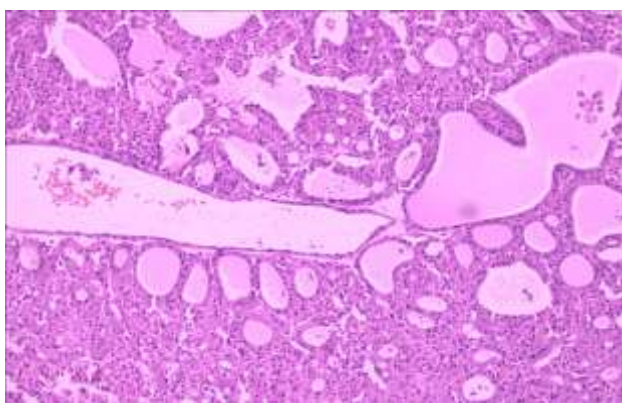


Figure 3: Dilated follicles lined by Hurthle cells.

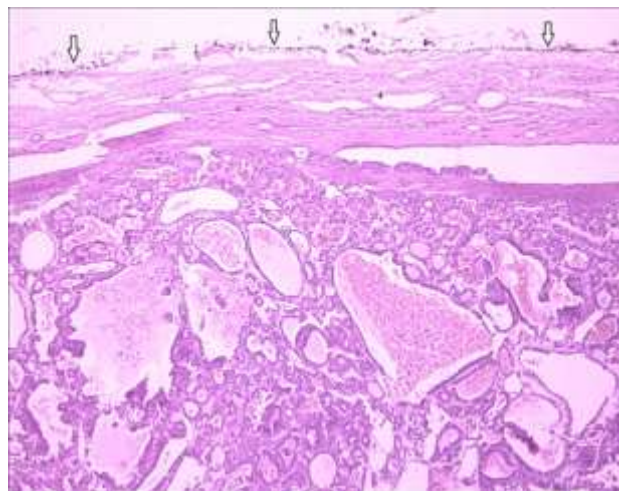


Figure 4: Variably sized thyroid follicles lined by Hurthle cells. Well defined inked capsule noted at periphery (arrow marked).

DISCUSSION

Most of the tumor of Hurthle cells origin are benign, as seen in this patient, and are called HCAs. However, many are malignant (figures are up to 40%) and are called Hurthle cell carcinomas (HCCs).⁶

Hurthle cell tumors are unique, not only because of the enduring debate about the true nature, but also because HCCs are particularly aggressive, thereby emphasizing the need to differentiate HCAs from carcinomas.⁷

The management protocol of both types are quite different as majority of surgeons recommend thyroid lobectomy for HCAs while advocating total thyroidectomy for HCCs.⁸ Many authors have reported tumors initially diagnosed as HCAs which later recurred or metastasized.⁹

The current definition of a malignant HCN depends on capsular and/or vascular invasion, and the presence of metastasis. This cannot be determined by cytologic evaluation and relies on histological examination of a surgically resected specimen. Frozen section evaluation has been shown to be of little use in the intraoperative evaluation of these lesions.^{8,10}

A number of sonographic characteristics have been reported to be more frequently associated with malignant nodules. Maizlin et al attempted to match the sonographic appearance with histologic characteristics of 15 histologically proven cases of Hurthle cell neoplasms.¹¹ They concluded that the pathologic criteria which differentiate benign from malignant Hurthle cell neoplasms for example capsular breach, vascular invasion, extra thyroidal, and nodal involvement are beyond the resolution of sonography; thus precluding their usefulness in their characterization. Ultrasonography of thyroid gland in our case suggested bilateral lesions

but failed to differentiate between benign and malignant potential of the nodules.

Although FNAC is one of the initial steps in the evaluation of a thyroid nodule, it is not possible to differentiate HCAs from HCCs with this method as was the scenario in our case where FNAC revealed Hurthle cell changes along with suspicion of malignancy (Category V, Bethesda classification).⁸

Current trends in the literature suggest the following risk factors for HCC:

- Age greater than 40 years.¹²
- Male gender.¹³
- Tumors larger than 4 cm.^{13, 14}

HCAs more than 4 cm is rare findings and moreover the occurrence of a multifocal HCA is not supported by antecedents in literature.

In a series, by Chen et al, no patient with HCA had bilateral disease.¹³ However, they found that 16% of those with HCC had bilateral foci which required total thyroidectomy. One patient with HCA in his series had contralateral tumors which turned out to be follicular variant of papillary thyroid cancer.

CONCLUSION

Hurthle cell neoplasms are relatively uncommon in clinical scenario. Uniqueness of our case was in regards to size of the tumor which was greater than 4 cm and bilateral lobe involvement. The management protocol is entirely different for benign and malignant lesions. Diagnostic dilemma, if reduced in these cases will yield greater success in planning the management. More studies are warranted and long term follow up of these cases is compulsory.

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