



# Hurthle cell carcinoma: a rare variant of thyroid malignancy – a case report

Yuvraj Adhikari, MBBS<sup>a,\*</sup>, Anupama Marasini, MBBS<sup>a</sup>, Nawaraj Adhikari, MBBS<sup>b</sup>, Laxman D. Paneru, MBBS<sup>c</sup>, Binit Upadhaya Regmi, MBBS<sup>d</sup>, Manita Raut, MD<sup>a</sup>

**Introduction and importance:** Oncocytic/Hurthle cell neoplasm is a rare form of thyroid malignancies, derived from follicular epithelium, presenting with a wide variety having either presenting with features of thyrotoxicosis or no any associated symptoms.

**Case presentation:** The authors present a case of a 49-year-old female with a known case of chronic obstructive pulmonary disease and hypertension who presented to our hospital with anterior neck swelling for 4 months that has gradually increased over the time. Physical examination, laboratory test, various radiological imaging, and cytological study led to the diagnosis of Hurthle cell neoplasm. With prompt diagnosis, she was admitted and surgery was done that include right hemithyroidectomy. Though it is a rare type of thyroid malignancy, early diagnosis and proper treatment has shown a very good prognosis.

**Clinical discussion:** Hurthle cell carcinoma initially presents with a single, painless palpable mass in the thyroid with pressure symptoms in advanced cases including dysphagia, dyspnea, and hoarseness. Pain, rapid growth, or significant compressive symptoms are suggestive of an invasive one.

**Conclusion:** This case highlights on rarity of disease, presentation, and availability of treatment modality.

**Keywords:** case report, follicular carcinoma, hurthle cell carcinoma, thyroidectomy

## Introduction

Hurthle cell carcinoma (HCC) are also known as Askanazy cell tumors, oncocytomas, and mitochondriomas or oxyphil tumors<sup>[1]</sup>. HCC represents less than 5% of all differentiated thyroid malignancies<sup>[2]</sup>. HCC were originally defined as an oxyphilic or oncocytic variant of follicular thyroid cancer; however, in 2016, new findings led to a reclassification by the WHO as a separate pathological entity<sup>[3]</sup>. Recently, Jin *et al.*<sup>[4]</sup> reported persistent or recurrent disease in only 8.2% of the patients, and none of their patients died of HCC during a median follow-up of 8.5 years. Similarly, Oluic *et al.*<sup>[5]</sup> reported a rate of 12% of persistence or recurrence, with a slightly worse survival rate than Jin *et al.*

This work has been reported in line with the Surgical Case Report (SCARE) 2020 criteria<sup>[6]</sup>.

<sup>a</sup>Nepalese Army Institute of Health Sciences, <sup>b</sup>Upendra Devkota Memorial National Institute of Neurological and Allied Sciences, Kathmandu, <sup>c</sup>Kailali Hospital Private Limited, Kailali and <sup>d</sup>Jibjibe Primary Health Center, Rasuwa, Nepal

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\*Corresponding author. Address: Shree Birendra Hospital, Kathmandu 44600, Nepal. Tel: +977 984 578 3365. E-mail address: yuvraj.adhikari06@gmail.com (Y. Adhikari).

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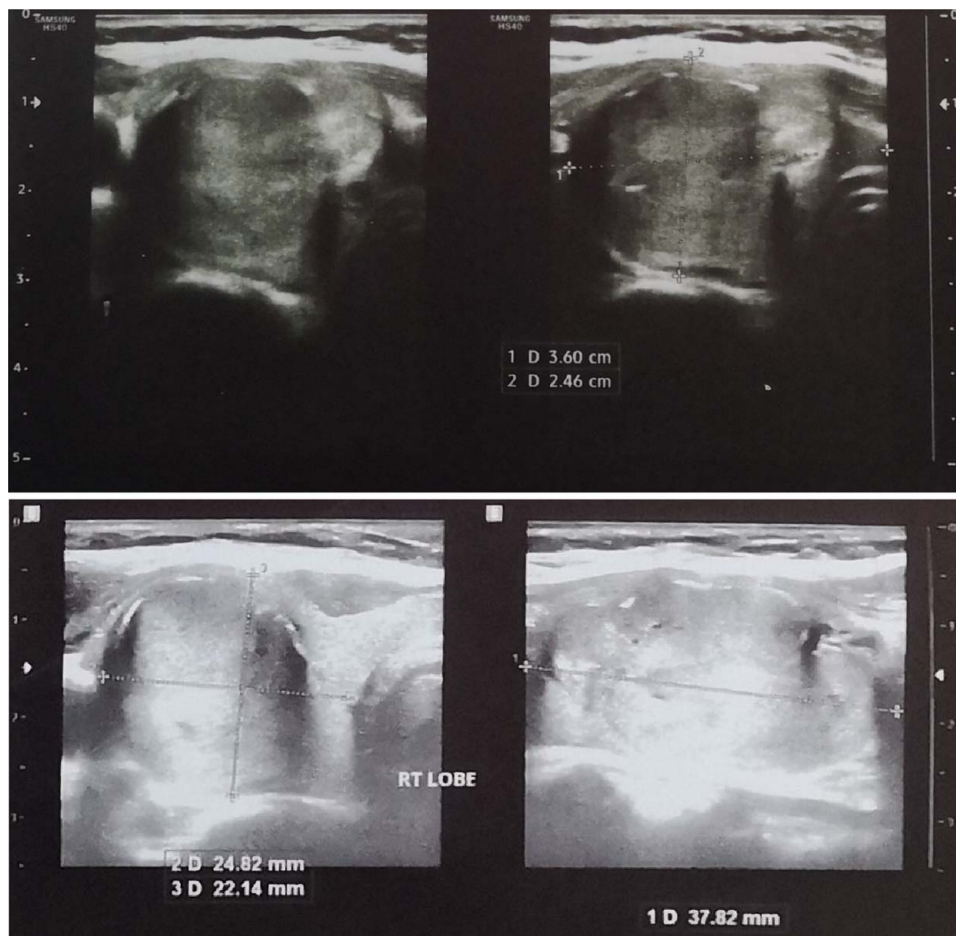
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## HIGHLIGHTS

- Rare variant of follicular thyroid malignancy.
- Good prognosis with surgical treatment.
- Hemithyroidectomy and total thyroidectomy with thyroxine supplements.
- Histologic type of oncocytic (Hurthle cell) carcinoma, encapsulated angioinvasive.

## Case presentation

A 49-year-old female, known case of hypertension under medication for 1 month and chronic obstructive pulmonary disease for 7 years, resident of Lamjung, Nepal, presented to the Ear, Nose, Throat, Head and Neck Surgery department of Shree Birendra Hospital with anterior neck swelling for 4 months that was insidious onset and was the size of a pea, which gradually progressed to its present size over the period of 4 months. She showed no symptoms of palpitations, increased sweating, pain or pressure, hoarseness, or fatigue, and had no family history of any thyroid disease. She has no features of hypothyroidism or thyrotoxicity during presentation. On examination, her general conditions were fair, oriented to time, place, and person. Her vitals were stable. Her neck swelling showed a firm mass with a medium texture and a clear border that moved up and down with her deglutition, but there was no difficulty in swallowing foods. The swelling measured approximately 3 × 2 cm in size. No regional lymph nodes were palpated and no any tracheal shift was noted. Laboratory indices showed hemoglobin of 12.2 g/dl (reference, 13–17); leukocytes of 5.6 × 10<sup>3</sup>/ml (reference, 4–11); platelets of 216 × 10<sup>3</sup>/ml (reference, 150–450), thyroid stimulating hormone of 1.52 mIU/l (0.4–4.2), Free T3 of 3.05 pg/ml (2.6–4.8) and free of T4 1.05 ng/ml (0.8–2.7).



**Figure 1.** Ultrasonography of Neck.

On neck ultrasonography (Fig. 1) showed that the right thyroid lobe is enlarged, measuring  $3.6 \times 2.8$  cm with echotexture (iso to hyperechoic), predominately hyperechoic nodules with peripheral ring calcification and vascularity within the nodules, the largest one measuring approximately  $2.6 \times 2.2$  cm (TIRADS-TR5, Isoechoic 1, ring calcification 2, and solid component 2) in size, whereas the left thyroid lobe is of normal size  $1.3 \times 0.9$  cm with a small cyst in the left thyroid lobe measuring  $0.4 \times 0.4$  cm. Contrast enhanced computed tomography of the neck and chest (Fig. 2) was performed in which there is a moderately enhancing ill-defined hypodense lesion noted in the right lobe of the thyroid measuring  $2.3 \times 2 \times 2.6$  cm in size with the presence of peripheral calcification. This lesion is abutting the right carotid vessels posteriorly and the sternocleidomastoid muscle laterally. Few small lymph nodes in bilateral level II and III largest one measuring  $10 \times 7$  mm in size is seen. Along with the neck, the lungs also show significant findings in the computed tomography scan. Fibrocalcification with traction bronchiectasis is noted in the apical and posterior segments of the right upper lobe, the left upper lobe, and the superior segments of the left lower lobe. Alveolar opacity is seen in the lingual. Decreased volume of bilateral upper lobes with deviation of the trachea to the right side. Multiple enlarged lymph nodes were noted in mediastinal and right hilar region, the largest one measuring

$26 \times 23$  mm. Bilateral apical pleural thickening is also seen. Fine needle aspiration cytology of the right lobe of the thyroid was done where it was done via ultrasonography guided and shows Bethesda Category IV suggestive of Hurthle cell neoplasm.

She was operated on the second day of admission on general anesthesia. After right hemithyroidectomy ( $4 \times 3$  cm right thyroid lobe along with isthmus), the post operative pathological results revealed encapsulated tumor cells arranged predominantly in a follicular and solid pattern in microscopic findings and give histologic type of oncocytic (Hurthle cell) carcinoma, encapsulated angioinvasive, with distance from invasive carcinoma to the closest margin of 0.2 cm. Pathologic stage classification pT2pNpM. Patient was discharged 4 days after the surgery and recommended for follow-up in every 2–3 months or as soon as possible if any emergency arise. All the management section was done by a consultant otolaryngologist in assistance of the concerned department resident at a tertiary care hospital in Nepal. At 2 month follow-up, a Thyroid Function Test was performed that showed thyroid stimulating hormone of 7.24 mIU/l (Reference: 0.4–4.2), free T3 of 3.08 pg/ml (Reference: 2.6–4.8), and free T4 of 0.99 ng/ml (Reference: 0.8–2.7). Written consent was obtained from the patient for the publication of the case details and the images.

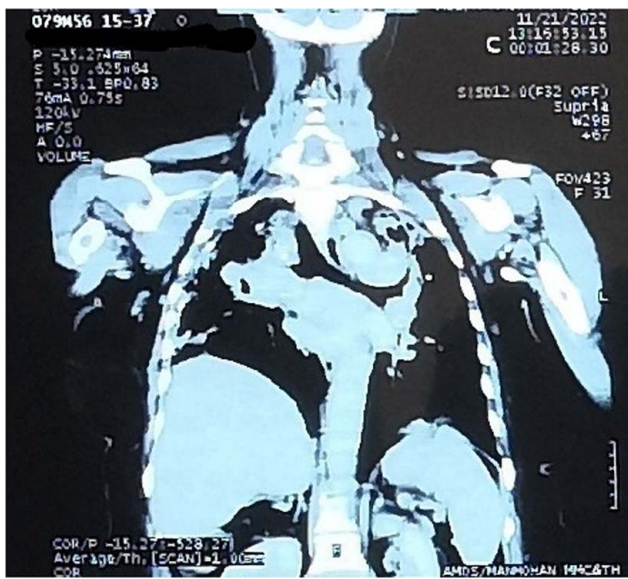


Figure 2. Contrast Enhanced Computed Tomography.

## Discussion

HCC is a relatively uncommon thyroid malignancy in iodine-sufficient regions. According to a single-center study in South Korea, among patients diagnosed with thyroid cancer from 1995 to 2005, HCC accounted for only 1%. In this multicenter cohort study in South Korea, where HCC is relatively rare, we evaluated the clinical outcome of patients with HCC. Persistent or recurrent disease was observed in 8.2% of the patients, and none died of HCC during a median follow-up of 8.5 years<sup>[7]</sup>. It is characterized by the presence of Hurthle cell or oxyphilic cell, derived from the follicular epithelium of the thyroid<sup>[8]</sup>. Kiminori Sugino *et al.*<sup>[9]</sup> reported that Hurthle cell neoplasm more commonly occurs in women, but the malignant form is more common in men. In comparison to other types of well-differentiated thyroid cancers, HCC is considered to be associated with a higher recurrence rate and mortality<sup>[10]</sup>. The incidence of distant metastasis is 15–34%<sup>[11]</sup>.

Various features are related for differentiating benign and malignant HCC. Size has been determined to be a distinguishing factor. HCCs are significantly larger (mean tumor size 4.0 cm) compared with adenomas (mean tumor size 2.4 cm). Patient age, sex, history of neck irradiation, bilaterality of tumors, and occurrence of concomitant thyroid cancers do not serve to distinguish between benign and malignant Hurthle cell neoplasms<sup>[12]</sup>. Tumor size (> 4 cm), extent of vascular invasion (> 4 foci), presence of mitosis, a solid or trabecular growth pattern, extrathyroidal extension, and presence of lymph node metastases are among the histologic features that predict recurrence. Extensive vascular invasion was also found to strongly correlate with worse 5-year disease-free interval and disease-specific survival<sup>[13]</sup>. However, in our case, microscopic findings show a follicular and solid pattern with eosinophilic and granular cytoplasm having a large nucleolus with identification of focal vascular invasion but less than four vessels seen. There is no mitosis, no lymphatic invasion, and no extrathyroidal extension.

The new guidelines permit withholding surgery or performing only lobectomy for most patient with thyroid cancer 1 cm without extrathyroidal extension, nodal involvement, or unfavorable history/characteristics. Also new guidelines deem lobectomy sufficient to treat low risk papillary or follicular carcinoma greater than 1 cm—less than 4 cm<sup>[14]</sup>. In our cases right hemithyroidectomy was done.

The prognosis largely depends on the extent of vascular invasion, with the more vessels invaded, the more guarded the prognosis (mortality >90% at 10 years). Other prognostic markers include age, sex, cancer stage, and degree of spread. The unfavorable prognostic factors include advancing age, male sex, larger tumor size at diagnosis, extra-thyroid expansion, and advanced stage at diagnosis<sup>[15]</sup>. A recurrence rate around 10.5–43% is seen in HCC. More than half of recurrence from follicular thyroid cancer is found within 3 years, and 80% develop within 6 years of the original diagnosis. Recurrence occurs more frequently at distant sites (up to 85% of cases) but can also be found in the thyroid bed and in regional lymph nodes; therefore, the role of neck ultrasound surveillance in these tumors is unclear<sup>[16]</sup>.

## Conclusion

HCC can present with a wide range of sign and symptoms ranging from a small lump over the neck to a change in voice/hoarseness and breathing difficulty. With early diagnosis of the case and effective treatment it has shown a very good prognosis. The new guidelines permit withholding surgery or performing only a lobectomy for most patients with thyroid cancer 1 cm—less than 4 cm.

## Ethical approval

This is case report; therefore, it did not require ethical approval from ethics committee.

## Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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The study did not receive any grant from funding agencies in the public, commercial, or not-for-profit sectors.

## Author contribution

Y.A.: conceptualization; methodology; writing-original draft; writing – review and editing; A.M.: data curation, investigation, revising and editing manuscript; N.A.: data curation, methodology, revising and editing manuscript; L.D.P.: data curation and visualization; B.U.R., M.R.: supervision, writing and editing manuscript. All authors were involved in manuscript drafting and revising and approved the final version.

## Conflict of interest disclosure

The authors reports no conflicts of interest.

## Author agreement statement

We confirm that the manuscript has been read and approved by all named authors and that there are no other persons who satisfied the criteria for authorship but are not listed. We further confirm that the order of authors listed in the manuscripts has been approved by all of us.

We understand that the corresponding author is the sole contact for the editorial process. She/he is responsible for communicating with the other about progress, submission of revisions and final approval of proofs.

## Patient perspective

The patient was thankful for having her condition diagnosed and managed. She feels her symptoms have improved significantly and her insights regarding her condition has changed positively. Her concerns regarding the possibility of future complications ameliorated after comprehensive counseling.

## Provenance and peer review

Not commissioned, externally peer reviewed.

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