ijtos 2025; doi 10.22376/ijtos.2025.3.1.48-51

International Journal of Trends on OncoScience

ISSN-2583-8431

Case Report

Rare Case of Oncocytic Carcinoma of Thyroid: Successful Surgical Management of a Large 20 Cm Tumor

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Abstract: Oncocytic carcinoma of the thyroid (OTC), also known as Hürthle cell carcinoma (HCC), is an extremely rare diagnosis accounting for 3% to 5% of all thyroid cancers. Historically, Hürthle cell carcinoma was considered as a variant of follicular carcinoma because of its origin in the follicular epithelium. However, later reports suggested that they are more aggressive, and most cases were found to be refractory to Radioiodine ablation (RAI). The rarity of HCC and the historical overlap with follicular thyroid cancer has made it difficult to develop Hürthle cell carcinoma-specific management strategies. Management varies based on tumor size and extent, with total thyroidectomy being recommended for larger tumors. The role of radioactive iodine therapy remains controversial. In this report, we present a 71-year-old female who presented to our hospital with a massive (20 cm) thyroid tumor that was diagnosed as oncocytic carcinoma of the thyroid and was successfully managed with total thyroidectomy.

Keywords: Hürthle cell carcinoma, Oncocytic carcinoma, Thyroidectomy, Large tumor

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Received On 24 December 2024
Revised On 30 December 2024
Accepted On 3 January 2025
Published On 6 January 2025

Funding This research did not receive any specific grant from any funding agencies in the public, commercial or not for profit sectors.

Citation Dr. Govindaraj Vardhanan, Dr. Ponniah Sasipriya, Dr. Gunasekaran, Dr. A. Mallika and Dr. Hareni Murugavel , Rare Case of Oncocytic Carcinoma of Thyroid: Successful Surgical Management of a Large 20 Cm Tumor.(2025).Int. J. Trends in OncoSci.3(1), 48-51 http://dx.doi.org/10.22376/ijtos.2025.3.1.48-51

1. INTRODUCTION

Hürthle cell carcinoma of the thyroid (HCC), otherwise known as Oxyphilic or Oncocytic carcinoma, is a rare variant of Thyroid cancer. Max Askanazy, a Swiss-German pathologist, first identified Hürthle cells in 1898. In World Health Organization (WHO) classification, it was initially classified along with follicular neoplasms. Later, differences in behavior prompted the WHO to reclassify HCC as a separate entity in 2017 [5]. Hürthle cells are unique follicular cells distinguished by their granular eosinophilic cytoplasm and hyperchromatic nuclei, resulting due to accumulation of altered mitochondria[6]. Although frequently associated with thyroid neoplasms, Hürthle cells can also be found in other benign conditions of the thyroid like multinodular adenomatous goiter and Hashimoto's thyroiditis[7]. They are infrequently seen in the parathyroid gland and other endocrine organs in individuals over 65 years and in certain mitochondrial disorders like oncocytic cardiomyopathy. Hürthle cell transformation is believed to stem from oxidative stress and mitochondrial dysfunction. The pathological characteristics and behavior of HCC remain unclear due to its rarity and inconsistent findings in prior studies[1-4]. Further studies are necessary to elucidate the key factors contributing to the aggressiveness of this cancer. This knowledge would lead to more effective treatment strategies.

2. CASE SUMMARY

An elderly female of age 71 years, presented with history of progressive difficulty swallowing and breathing difficulty over the past 35 years, gradually progressing in severity. She had not availed of any treatment so far. Clinically there was a large around 20 x 20 cm hard fixed swelling over the anterior aspect of the neck. Trucut biopsy was suggestive of Oncocytic neoplasm, the possibility of invasion could not be excluded. The patient initially visited another hospital where she was advised surgery and was referred here for further management.

3. DIAGNOSTIC AND STAGING WORKUP

CT neck was done which showed a large oxophytic pedunculated heterogenous enhancing mass arising from the left hemithyroid measures 15.1 x 16.3 x 20.8 cm (APxTRXCC) with areas of calcification and large area of central necrosis. The mass appeared to be compressing the trachea and esophagus causing mild deviation without airway narrowing or invasion. There was no retrosternal extension. There was skin infiltration in the superolateral aspect. The lesion had caused partial enhancement of left common carotid artery without any occlusion, and extrinsic compression of left internal jugular vein with complete occlusion. Multiple dilated tortuous vessels were seen in the subcutaneous plane over the anterior chest wall. Radiological features were suggestive of locally advanced thyroid malignancy (TNM staging T4b, N1, Mx). CT thorax showed no lung metastasis. After considering the size and extent of the tumor and patient's fitness for surgery, the patient was planned for Total thyroidectomy with left modified radical neck dissection and bilateral paratracheal node dissection. Also, surgical resection was essential to confirm the diagnosis.

4. SURGICAL MANAGEMENT

Pulmonologist's, Cardiologist's, and Anesthetist's fitness was sought, and orders were followed. The patient was taken up for the planned surgical procedure on 29th September 2024. The surgery was uneventful with no intraoperative or postoperative complications. The gross specimen measured 20 x 15 x 10.5 cm. Lobes and isthmus could not be made out separately. Cut surface showed an encapsulated mahogany-colored tumor with solid-cystic areas and capsular breach. In microscopic examination, margins were uninvolved, with the closest margin being 0.1 cm. No lymphatic or vascular invasion could be seen. 25 nodes were examined and revealed no metastases. Pathological stage was reported as pT3N0

5. FOLLOW-UP AND FUTURE DIRECTIONS

The patient is being closely monitored. Follow-up thyroid function test showed an elevation of TSH. The patient is further planned for Radioactive Iodine Ablation therapy. Given the large size, she comes under high risk and requires RAI. The patient is at a high risk of early relapse and requires close follow-up.

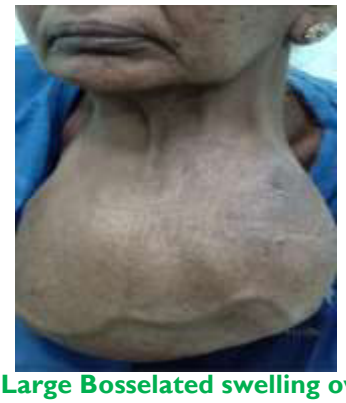

Fig A: Large Bosselated swelling over the neck with stretching of the overlying skin and engorged subcutaneous veins

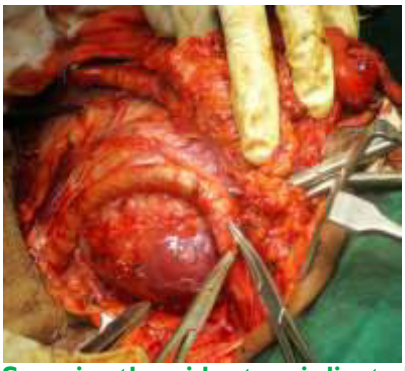

Fig B: Superior thyroid artery is ligated close to the gland to avoid injury to the external laryngeal nerve.

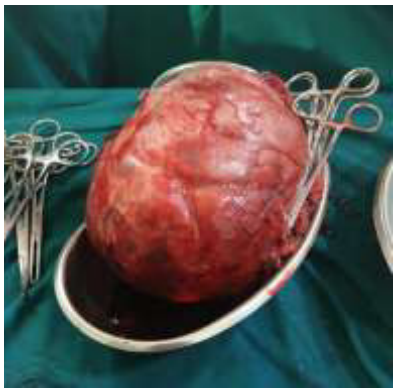

Fig C: Gross specimen measuring 20 x 15 x 10.5 cm with bosselated surface, Lobes and isthmus not be made out separately.

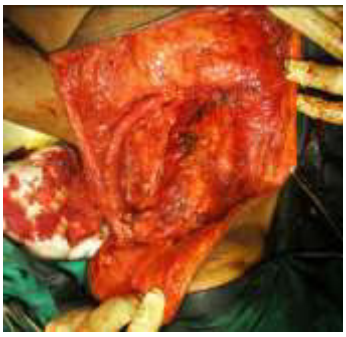

Fig D: Postop bed after en block removal of thyroid tumor. Left IJV and Spinal accessory nerve preserved but sternocleidomastoid and strap muscles sacrificed as they were adherent to the mass.

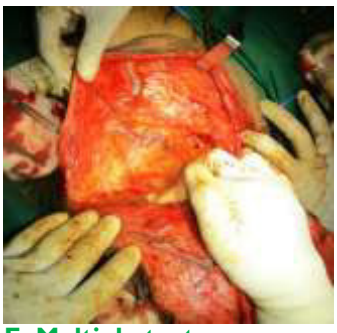

Fig E: Multiple tortuous engorged subcutaneous vessels

Fig F: Suture site with drain

6. DISCUSSION

The incidence of thyroid cancer has increased by nearly 3-fold over the last decade. However, incidence of follicular carcinoma of the thyroid has either remained stable or shown a steady decline. Dietary iodine supplementation has contributed to the observed decline of follicle carcinoma in several regions. In contrast to FTCs, Hürthle cell carcinomas are more likely to arise in iodine-rich regions, be multifocal at presentation, and have nodal metastases. The increasing incidence of Hurtle cell transformation in thyroid lesions may indicate elevated reactive oxygen species (ROS) production in thyroid cells during normal iodine and thyroid hormone metabolism[9]. Hürthle cell transformation is believed to result from an imbalance between mitochondrial growth and mitochondrial degradation, resulting in mitochondrial buildup. The time factor is essential for the development of Hürthle cell characteristics, as it requires significant duration for increase from 100/150/200 mitochondria to the 4000/5000 mitochondria per thyroid cell that defines Hürthle cells[10]. Signs and symptoms such as hoarseness, dysphagia, neck pressure, and clinical signs of thyrotoxicosis have been documented. The presence of Hürthle cells in a biopsy sample may prompt additional evaluation to assess its cause, and rule out nodal or distant spread in order to take treatment decisions[8]. Comparative studies have shown that Hürthle cell carcinoma has a worse prognosis than papillary thyroid carcinoma[11]. However, some studies suggest that prognosis may be similar when matched for tumor size and stage[12]. It is difficult to distinguish Hürthle cell carcinoma from adenoma by solely ultrasonography[13]. In few instances, meticulous ultrasonographic technique might disclose features of capsular invasion. The cytological criterion for identifying Hürthle cell neoplasms is well delineated. The role of genetic testing in oncocytic carcinoma is evolving, with mutations in the HRAS and KRAS genes identified in some cases [10, 11]. Further research is needed to understand the implications of genetic alterations on treatment and prognosis. Prognosis varies depending on tumor size, stage, and histological features[7,8]. Large tumors (> 4 cm) are associated with a higher risk of recurrence and nodal metastasis[9] Our patient's tumor size (20 cm) placed her at higher risk, emphasizing the importance of close follow-up to detect early relapse. A lobectomy is sufficient for small, unifocal tumors < 1cm in size, when there is no history of prior head and neck radiation, familial thyroid carcinoma, or clinically evident nodal metastases. For patients with tumor more than 1 cm but less than 4 cm, lacking extrathyroidal extension or nodal metastases, either near-total or unilateral thyroidectomy is advised. The diagnosis is usually confirmed following lobectomy in the frozen section specimen. Total thyroidectomy is advised for Hürthle cell neoplasms above 4 cm in size. Julsrud et al retrospectively analyzed data from 295 patients with HCC (42 (14%) hemi and 253 (86%) total thyroidectomy). At 7 years follow-up, there was no significant difference in recurrence-free survival ($p = 0.065$), overall survival ($p = 0.806$) or disease-specific survival. Tumor size directly correlated with malignancy potential and outcomes[16]. The role of radioactive iodine (RAI) remains controversial. Currently, the American Thyroid Association recommends RAI after total thyroidectomy in all patients with Hürthle cell carcinoma, whereas the National Cancer Comprehensive Network recommends RAI for tumors larger than 2 cm or in the presence of vascular invasion, extrathyroidal extension, lymph node metastases or elevated postoperative unstimulated thyroglobulin levels. Ultimately, the decision to use RAI therapy should be individualized based on the specific characteristics of the tumor and patient's overall health and risk factors. Generally, thyroglobulin is a common biomarker used to detect recurrence following thyroidectomy[17]; However, recurrent HCC exhibits undetectable thyroglobulin levels. Hence, the decision regarding clinical follow-up based on thyroglobulin levels is usually taken on a case-by-case basis.

7. CONCLUSION

In conclusion, despite the large size of tumour, our patient underwent a successful total thyroidectomy with bilateral paratracheal lymph node dissection, with no complications. Pathological findings confirmed Hürthle cell carcinoma, necessitating close follow-up. Given the tumor's size and aggressive nature, vigilant monitoring is essential to detect early recurrence or distant relapse. While the role of radioactive iodine therapy remains debated, its use here is justified by the patient's risk profile. Long-term follow-up and individualized care are paramount.

8. AUTHORS CONTRIBUTION STATEMENT

All authors are equally contributed.

9. CONFLICT OF INTEREST

Conflict of interest declared none.

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