CASE REPORT

Hürthle Cell Carcinoma in an Extraordinary Location: A Case Report

Darrel S Au¹, Krishanth Naidu², Usama Majeed³

ABSTRACT

Aim: To present a case of Hürthle cell carcinoma at an extraordinary site, and to contribute to the understanding and management of this tumor. Background: Hürthle cell carcinomas are a dangerous but uncommon variant of thyroid follicular carcinoma, exhibiting invasive potential, they have a propensity to metastasis to distant sites. Therefore, swift diagnosis and management with multidisciplinary input are warranted. We humbly present a case of Hürthle cell carcinoma in the supraclavicular region.

Case description: A 44-year-old woman, with a prior history of follicular thyroid adenoma, was found to have a mass measuring $20 \times 15 \times 18$ mm on the left supraclavicular region. Magnetic resonance imaging (MRI) and fine needle aspiration biopsy were suspicious of follicular thyroid neoplasm. Positron emission tomography-computed tomography (PET-CT) revealed two lesions with avid fluorodeoxyglucose (FDG)-avid uptake, one corresponding to the original supraclavicular location and one in the left iliac crest. Subsequently, histological analysis following surgical excision of the supraclavicular mass revealed a lobulated Hürthle cell tumor. The patient underwent further cervical lymph node dissection, with one supraclavicular node demonstrating infiltration by Hürthle cell carcinoma. Postoperatively, the patient underwent radioactive iodine (RAI) therapy to which she responded well, and currently remains well at follow-up.

Conclusion: Hürthle cell carcinomas are a rare but aggressive variant of follicular thyroid carcinomas. Known for their malignant potential, this case underscores the vigilance required for the diagnosis and management of these tumors as their sites of metastasis may be most extraordinary, as illustrated in this report. Swift diagnosis and multidisciplinary management of these tumors are paramount.

Clinical significance: Hürthle cell carcinomas have been demonstrated to metastasize to the most extraordinary sites, we present a unique case of Hürthle cell carcinoma which reinforces the notion and hope to contribute to its understanding.

Keywords: Ectopic thyroid tissue, Endocrine surgery, Hürthle cell carcinoma, Oncocytic thyroid carcinoma, Thyroid carcinoma. *World Journal of Endocrine Surgery* (2020): 10.5005/jp-journals-10002-1292

Introduction

Hürthle cell/oncocytic carcinomas are an uncommon but dangerous variant of follicular carcinoma of the thyroid. They exhibit a wide spectrum of behavior with regards to local invasion, with some variants being highly invasive, infiltrating thyroid parenchyma, and extrathyroidal tissue, while some exhibiting minimally invasive behavior. Furthermore, Hürthle cell carcinomas have a high likelihood of metastasis; hence, their swift diagnosis and treatment are warranted. We herein present an intriguing case of a patient with Hürthle cell variant follicular carcinoma in the ectopic thyroid tissue of the supraclavicular region.

CASE DESCRIPTION

A 44-year-old woman with a history of benign thyroid adenoma presented to her general practitioner (GP) with a 6-month history of a nontender palpable lump in the left supraclavicular region. Her background medical history included a left hemithyroidectomy for a Hürthle cell thyroid adenoma, as well as a hysterectomy for fibroid disease. The lump measured $20 \times 15 \times 18$ mm and was located in the left supraclavicular fossa, 5 cm medial to the acromial-clavicular joint. Ultrasonography of the lesion showed a highly vascular, thinwalled homogeneous mass. A subsequent magnetic resonance imaging (MRI) study confirmed the findings of a highly vascular soft-tissue mass. Fine needle aspiration biopsy of the lesion revealed a diagnosis of Hürthle cell follicular thyroid neoplasm.

A positron emission tomography-computed tomography (PET-CT) showed two fluorodeoxyglucose (FDG)-avid lesions—one in the left supraclavicular fossa and another in the left iliac

¹The Canberra Hospital, Garran, Australia

^{2,3}Division of Surgery, The Canberra Hospital, Garran, Australia

Corresponding Author: Darrel S Au, The Canberra Hospital, Garran, Australia, Phone: +61 410080773, e-mail: darrelshaunau1028@gmail.com

How to cite this article: Au DS, Naidu K, Majeed U. Hürthle Cell Carcinoma in an Extraordinary Location: A Case Report. World J Endoc Surg 2020;12(2):96–97.

Source of support: Nil Conflict of interest: None

crest. With radiological findings in hand, the patient underwent an excisional biopsy of the supraclavicular lesion. Histopathology of the excised lesion showed a lobulated Hürthle cell tumor-infiltrating into fibrofatty tissue, with an elevated Ki-67 index, and positive TTF-1 and Pax-8 staining. Biopsies of the adjacent left supraclavicular lymph node also demonstrated lobulated Hürthle cell tumor-infiltrating into fibrofatty tissue, with no lymph nodal tissue or normal thyroidal tissue present.

A right hemithyroidectomy was planned following a multidisciplinary meeting. Histological analysis revealed a normal right hemithyroid. However, tissue from the left thyroid bed showed a focus of Hürthle cell thyroid follicular carcinoma present in adipose tissue and adjacent to skeletal muscle fibers.

Six months after the patient's initial presentation, a suspicious dermal nevus along with a lymph node on the patient's left supraclavicular region was identified and excised. The latter was confirmed to be Hürthle cell tumor infiltration upon histological

© The Author(s). 2020 Open Access This article is distributed under the terms of the Creative Commons Attribution 4.0 International License (https://creativecommons. org/licenses/by-nc/4.0/), which permits unrestricted use, distribution, and non-commercial reproduction in any medium, provided you give appropriate credit to the original author(s) and the source, provide a link to the Creative Commons license, and indicate if changes were made. The Creative Commons Public Domain Dedication waiver (http://creativecommons.org/publicdomain/zero/1.0/) applies to the data made available in this article, unless otherwise stated.

analysis. Fortunately, benign nodes were noted in the left level II to V dissection. A consensus arrived at multidisciplinary meetings and radioactive iodine (RAI) therapy was planned for the patient. The patient underwent and responded well to RAI therapy and remained well at clinical follow-up with endocrinology and general surgery.

Discussion

Ectopic thyroid tissue in the head and neck, although uncommon, are well-known entities in medical literature, as the development and translocation of the thyroid gland to its physiological paratracheal location have been extensively studied. The reported prevalence of ectopic thyroid tissue is to be at 1 in 100,000 to 300,000 with lingual ectopic sites accounting for 90% of cases. Though elusive, ectopic thyroid tissue in locations, such as, the abdomen have been reported. We believe that this is the first reported case of metastatic Hürthle cell thyroid carcinoma of the supraclavicular fossa in the medical literature.

Hürthle cell carcinomas encompass approximately 5% of all differentiated thyroid carcinomas.^{4,5} The paucity in its prevalence, aggressive clinical behavior, along with the difficulty in distinguishing metastatic disease of this variant contributes to the ongoing debate regarding its clinical course and prognosis.

Hürthle cell carcinomas are histologically subdivided into widely invasive and minimally invasive neoplasms. Despite its classical appearance, it is impossible to definitively diagnose malignancy based solely on cytological features; Hürthle cell lesions traditionally require surgical excision and biopsy for definitive diagnosis. The fact that even with extensive pathological evaluation, some Hürthle cell neoplasms will remain classified as indeterminate behavior reinforces the intricacy of these tumors.⁴

While there is medical literature on the treatment of Hürthle cell carcinomas of the thyroid, the literature on the treatment of follicular carcinoma in ectopic thyroid tissue is alarmingly scarce. It would not be a stretch to consider the management of our patient's case to be uncharted territory in medicine; the current recommendation is that surgical excision is warranted if there is suspicion of neoplasm in ectopic thyroid tissue.^{3,5,6} Hence, with the due diligence of the dedicated multidisciplinary team (MDT) of surgeons, endocrinologists, oncologists, and radiologists a holistic treatment plan involving surgical resection, local lymph node excision, and RAI was developed for our patient.

With regard to adjuvant therapy for Hürthle cell carcinomas, there is also a lack of consensus regarding the role of RAI 131, as these tumors are known to be highly resistant to RAI treatment. One small study of 16 patients demonstrated favorable outcomes for the use of RAI along with total thyroidectomy and external beam radiotherapy. Nevertheless, the current consensus recommends that RAI therapy should be reserved for tumors with a high risk of recurrence. Some studies have proposed the use of cervical beam radiation as adjuvant therapy, but its role is still debated. Moreover, significant complications of cervical radiation have to be carefully considered. Management of distant metastasis is similar to most oncological conditions, with a focus directed on reducing the morbidity and mortality of organ compression and tumor infiltration. Due to the propensity of Hürthle cell carcinomas

to metastasize to bone, radiotherapy plays a substantial role in the management of bony metastasis. 5,7,9

The pressing question is whether this tumor is a metastatic deposit from a preexisting thyroid neoplasm, or whether the tumor is arising from ectopic thyroid tissue in the left supraclavicular fossa. Literature has certainly reported albeit rare incidences of the primary metastatic transformation of ectopic thyroid tissue. ¹⁰ While there is evidence to suggest both etiologies, the answer remains inconclusive. Nevertheless, it could be argued that the specific etiology of the tumor matters not in the definite management, as regardless of the etiology, aggressive treatment involving a multidisciplinary team is warranted for all Hürthle cell carcinomas.

Conclusion

We sought to present this case of a rare Hürthle cell follicular thyroid carcinoma in an extraordinary anatomical site of the left supraclavicular fossa, which we believe is the first to be reported. We hope that we were able to contribute to the current and future understanding of this rare and often aggressive tumor.

CLINICAL SIGNIFICANCE

Hürthle cell carcinoma is a very uncommon tumor, but with its aggressive nature, such malignancies must be promptly diagnosed and treated. In this report, we have described a case of Hürthle cell carcinoma in an extraordinary location, in doing so we hope to contribute to the current body of knowledge on the management of such a tumor.

REFERENCES

- Sahu S, Agarwal P, Husain M, et al. Right supraclavicular ectopic thyroid: an unusual site of presentation. Inter J Surg 2006;13(1): 1–4
- Adelchi C, Mara P, Melissa L, et al. Ectopic thyroid tissue in the head and neck: a case series. BMC Res Notes 2014;7(1):790. DOI: 10.1186/1756-0500-7-790.
- Ibrahim NA, Fadeyibi IO. Ectopic thyroid: etiology, pathology and management. Hormones 2011;10(4):261–269. DOI: 10.14310/ horm.2002.1317.
- Bhattacharyya N. Survival and prognosis in Hurthle cell carcinoma of the thyroid. Archives of otolaryngology. Head and Neck Surgery 2003;129(2):207–210. DOI: 10.1016/S0194-5998(03)01257-9.
- Shawky M, Sakr M. Hurthle cell lesion: controversies, challenges and debates. Indian J Surg 2016;78(1):41–48. DOI: 10.1007/s12262-015-1381-x.
- Zavitsanos P, Amdur RJ, Drew PA, et al. Favorable outcome of Hurthle cell carcinoma of the thyroid treated with total thyroidectomy, radioiodine, and selective use of external-beam radiotherapy. Am J Clin Oncol 2017;40(4):433–437. DOI: 10.1097/COC.0000000000000180.
- Ahmadi S, Stang M, Jiang XS, et al. Hürthle cell carcinoma: current perspectives. Onco Targets Ther 2016;9:6873–6884. DOI: 10.2147/ OTT.S119980.
- 8. Asa SL. My approach to oncocytic tumours of the thyroid. J Clin Pathol 2004;57(3):225–232. DOI: 10.1136/jcp.2003.008474.
- Bai S, Baloch ZW, Samulski TD, et al. Poorly differentiated oncocytic (Hurthle cell) follicular carcinoma: an institutional experience. Endocr Pathol 2015;26(2):164–169. DOI: 10.1007/s12022-015-9367-6.
- 10. Tucci J, Rulli R. Follicular carcinoma in ectopic thyroid gland. A case report. Il Giornale di Chirurgia 1999;20:97–99.