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Case Report

Synchronous occurrence of Hurthle cell carcinoma with Hodgkin's lymphoma; the first reported case with literature review

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ABSTRACT

Introduction: Hurthle cell carcinoma (HCC) is a rare type of thyroid cancer while Hodgkin's lymphoma (HL) is an uncommon cancer of the lymphocytes. The synchronous occurrence of HCC and HL in a single person has not been described in the literature. This report aims to present the first case of synchronous HCC and HL.

Case report: A 31-year-old male presented with a right side neck swelling for three months. Laboratory findings revealed elevated thyroglobulin (286.7 ng/ml). Ultrasound (US) examination showed several enlarged lymph nodes (<20mm) in the right side of the neck with increased vascularity. There was also a solitary solid thyroid nodule (23*20mm) in the right lobe. Fine needle aspiration cytology (FNAC) of the right lobe revealed Hurthle cell neoplasm, and right cervical lymph node was suspicious for Hodgkin's lymphoma. Right lobectomy with right lateral cervical excisional biopsy was performed. Histopathology confirmed HCC and HL.

Discussion: HCC is a differentiated cancer occurring in old age population. Multiple factors increase the risk of HCC, including the excess iodine, history of thyroid diseases, and exposure to radiation. HL is a cancer of the lymphatic system that affects young adults and older population. There is a significant association between excess body weight and cigarette smoking with the risk of HL.

Conclusion: Although extremely rare, occurrence of a few cases of other thyroid carcinomas and HL could occur. However, no relation has been described between the two cancers.

1. Introduction

Cancers of the thyroid gland are relatively less common compared to more prevalent cancers, and account for only 1% of all cancers and less than 0.5% of all lethal tumors [1]. Hurthle cell carcinoma (HCC) is a rare type of thyroid cancer, which is regarded as a well-differentiated oncocytic variant of follicular thyroid carcinoma (FTC) [2,3]. HCC is characterized by the presence of Hurthle cells or oxyphilic cells which are a large polygonal cells derived from the follicular epithelium of the thyroid [4]. Hodgkin's lymphoma (HL) is a cancer of the lymphocytes, which is an important component of the body's immune system, characterized by the presence of Hodgkin Reed Sternberg cells in classical Hodgkin lymphoma and lymphocyte-predominant cells in nodular lymphocyte-predominant Hodgkin lymphoma [5,6]. The synchronous occurrence of HCC and HL has not been described in the literature.

The current report aims to present the first ever reported case of synchronous HCC and HL, with a brief review of the literature. The report has been arranged in line with SCARE 2020 guidelines [7].

Patient information: A 31-year-old male presented with a right side neck swelling for a period of 3 months.

Clinical findings: grade one (G1) goiter. Normal vital signs.

Diagnostic approach: Laboratory findings revealed high level of serum thyroglobulin (286.7 ng/ml), normal serum TSH (0.795 uIU/ml), and normal serum calcitonin (4.31pg/ml). Neck ultrasound (US) examination discovered several enlarged lymph nodes of less than 20mm in the short axis in the right side of the neck along mid and lower jugular chains in the supraclavicular region with increased vascularity. At the same time, there was a solitary solid thyroid nodule of about 23*20mm

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in the right lobe with partial cystic changes, suspicious for cancer. Fine needle aspiration cytology (FNAC) of the right thyroid lobe diagnosed Hurthle cell neoplasm of Bethesda IV, and right cervical lymph node was suspicious for Hodgkin's lymphoma.

Therapeutic intervention: under general anesthesia, right lobectomy with right lateral cervical lymphnode excisional biopsy was carried. Postoperative histopathological examination displayed angioinvasive, encapsulated follicular carcinoma of Hurthle type, two vessels were invaded (Fig. 1). It was unifocal without any perineural and extra-thyroidal extension. The right cervical lymph node biopsy showed classical Hodgkin lymphoma of nodular sclerosing type with the pathological stage pT1b. Postoperative thyroglobulin level became 5.93ng/ml, and it was decided to proceed to the completion of left thyroid lobectomy. Second histopathological examination of the left lobe showed colloid goiter. Finally, the patient received chemotherapy followed by Radioactive iodine therapy.

Follow up and outcome: post-operative period was uneventful, the patient including his wound was healthy 4 months after the operation.

2. Discussion

HCC is a type of well differentiated thyroid cancer, a subtype of FTC [8]. It is a relatively uncommon cancer with an incidence of 3% among all other thyroid cancers [2]. It is characterized by the presence of Hurthle cell or oxyphilic cell, derived from the follicular epithelium of the thyroid [4]. HL is classified into nodular lymphocyte-predominant and classical HL. The classical type is further subdivided into four distinct subtypes based on presentation, site of involvement, epidemiology, and association with EBV [9]. Reported synchronous occurrence of HCC and HL is currently lacking in the literature, hence this is the first reported case of its kind. However, there are three reported cases of synchronous papillary thyroid carcinoma (PTC) and HL, and one reported case of synchronous FTC, PTC, and HL [10,11].

Multiple factors increase the risk of HCC, including the excess iodine, history of thyroid diseases, and exposure to radiation [12]. Sugino and associates reported that Hurthle cell neoplasm more commonly occurs in women, but the malignant form is more common in men [13]. In contrast, some other studies reported more female predominance in both benign and malignant Hurthle cell neoplasms [14]. HCC usually occurs in the older population compared to other forms of thyroid cancers with a median age of 57 years, and a female to male ratio of 3:1 has been reported [15,16]. The causes of HL are still unclear, however multiple infectious factors, autoimmune and chronic inflammatory conditions have been described [17]. Studies described a significant association between excess body weight and cigarette smoking and the risk of HL [18]. HL is more common in men and has a bimodal distribution with incidence peaks in young adults and older ages of between 45 and 55 years [9]. From a study reported by Mauch et al., the most common site for the occurrence of nodular sclerosing HL is mediastinum (73%), followed by right and the left sides of the neck [19]. HL usually presents as painless lymphadenopathy with systemic symptoms presented in 25% of patients, fever, night sweats, loss of weight, and other symptoms such as pruritus and fatigue have been reported [9]. The patient in this report is a 31-year-old male presented with a right side neck swelling for a period of 3 months, with no remarkable abnormal symptoms and normal vital signs.

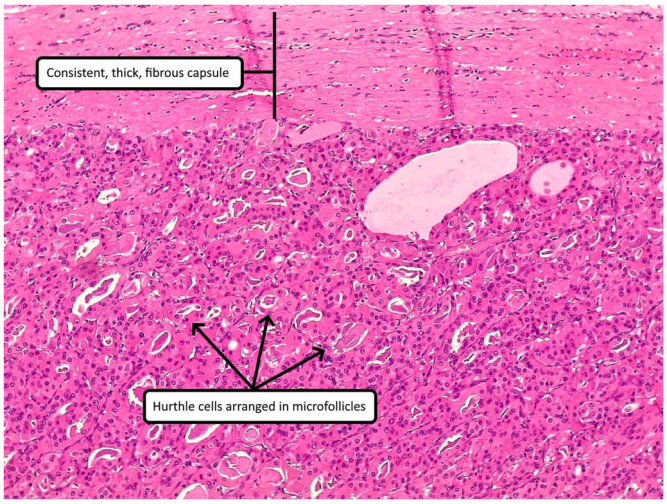

Fig. 1. Angioinvasive, encapsulated follicular carcinoma of Hurthle type.

The main diagnostic method for evaluating thyroid nodules is FNAC which is accurate and cost-effective, however, it is difficult to diagnose HCC using FNAC as its sensitivity is only 15–25% [20]. Histopathological examination of the resected specimen is gold standard for diagnosis of HCC. Malignant type can be differentiated from the benign form by the presence of vascular or extracapsular invasion and extrathyroidal metastasis to lymph node and distant organs [21]. Some factors are used in distinguishing malignancy from benign tumors in the absence of a definitive preoperative diagnostic test, such as age and size of the tumor, in which the risk of malignancy increases with advanced age and large tumor size [22].

Because of the lack of a standard approach, it is difficult to manage synchronous multiple primary cancers. The standard treatment of all well-differentiated thyroid cancers is surgical resection. The procedure of choice in the treatment of HCC is total thyroidectomy [15]. Radioactive iodine is widely used in the treatment of well-differentiated thyroid cancers, however its effectiveness in the setting of HCC is controversial. Kushchayeva and associates reported that only 5%–10% of patients with HCC take up enough radioactive iodine [23]. However, a study by Penabad et al. found that 38% of HCC take up enough radioactive iodine [24]. The treatment of HL is based on the extent or stage of the disease. The early stage of HL is treated with combined chemotherapy and radiotherapy which has the characteristics of primary tumor control and low drug-related toxicity [11]. The standard treatment for advanced-stage of HL is combination chemotherapy, but the effect of radiotherapy in advanced HL is unclear [24]. The current case was managed through right lobectomy and right lateral cervical excisional biopsy.

HCC has an aggressive course through which it is more likely to spread to the neck and soft tissue, while less likely to metastasize into the cervical lymph nodes [25]. In comparison to other types of well-differentiated thyroid cancers, HCC is considered to be associated with a higher recurrence rate and mortality [22]. The incidence of distant metastasis is 15–34% [20]. It has been reported that age, sex, and size of tumor are crucial prognostic factors for HCC, patients with tumor extension and recurrence are associated with poorer prognosis and higher mortality [25]. Early-stage HL has an excellent prognosis with an overall survival of more than 90%, and in the advanced stage of HL, the survival rate is 75%–90% [9].

In conclusion, although a few cases of other thyroid carcinomas and HL have been reported, there is no reported case regarding the coexistence of HCC and HL, and no relation has been described between the two cancers. Further studies are required to explain the relationship between the synchronous occurrence of the two different malignancies.

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.

Provenance and peer review

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Ethical approval

Approval is not necessary for case report in our locality.

Author contribution

Hiwa O.Baba, Abdulwahid M.Salih: surgeons managing the case, follow up the patient, writing the manuscript and final approval of the manuscript. Aras J.Qaradakhy, Ari M.Abdullah, Rawezh Q.Salih, Shvan M.Hussein, Rawa M.Ali: literature review, writing the manuscript, final approval of the manuscript.

Registration of research studies

According to the previous recommendation, registration is not required for case report.

Guarantor

Fahmi Hussein Kakamad is the Guarantor of submission.

Declaration of competing interest

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2021.102750.

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