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CLINICAL REPORT

Occult Thyroid Carcinoma with Cervical Lymph Node Metastasis: A Rare Case Report

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Abstract
Cervical lymph node metastasis is one of the most common clinical presentations of papillary thyroid carcinoma (PTC). Occult thyroid carcinoma is described as absence of primary tumour or with presence of microcarcinoma in thyroid with cervical lymph node metastasis. Frequency of occult thyroid cancer has decreased due to developments in imaging and improved accuracy of histological examinations. 38 year old male presented to us with complaints of swelling over the left side of neck for the past 2 months. Ultrasonography was suggestive of multiple suspicious enlarged nodes in left level II, III, IV and V and fine needle aspiration cytology showed features of metastatic PTC. He was planned for total thyroidectomy with central compartment clearance and bilateral functional neck dissection. Final histopathology staging was pT0N1b. Radioactive iodine (RAI) screening showed residual functioning thyroid and later therapeutic RAI was administered. He has been on regular follow up and disease free for 1 year post treatment. Occult thyroid carcinoma is a rare diagnosis with multiple treatment plans. Few hypothesis for this entity includes tumor regression, ectopic thyroid carcinoma or missed pathological findings.

Keywords Papillary thyroid carcinoma · Occult thyroid carcinoma · Lymph node metastasis · Ultrasonography · Fine needle aspiration cytology

Introduction

Papillary thyroid carcinoma (PTC) is the most common histological variant amongst thyroid cancers [1]. Cervical lymph node metastasis is the most common presentation of PTC with an incidence of 20–50% [2, 3]. The prognosis of PTC depends upon cervical lymph node involvement and age of the patient. Recent AJCC has divided staging based on age of the patient to be more or less than 55 years which was previously 45 years [4]. Thyroid malignancy tends to metastasize via different pathways, PTC spreads via lymphatic pathway whereas follicular carcinoma usually spreads via hematogenous route [5]. The first echelon station for PTC is the central compartment (Level VI). Thyroid malignancy usually shows involvement of lymph nodes at levels III and IV, whereas involvement of level II and V shows advanced disease [6]. When ultrasonography (USG) shows metastatic thyroid carcinoma in lymph nodes with no primary in thyroid, it is known as occult thyroid carcinoma (OTC) [7] and is a rarely reported case in literature. The management of OTC is still controversial due to the reduced number of reported cases in literature.

Case Report

A 38 years old euthyroid male with a complaint of left sided neck swelling for the past 2 months presented to the outpatient department with no other associated complaints or co-morbidities. On clinical examination, multiple nodes palpable in left level II to IV which were non tender, firm and mobile. No palpable midline swellings. USG neck showed a normal thyroid gland with multiple, enlarged, heterogeneous, relatively well-encapsulated, nodular masses with

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lobulated margins noted along the left jugular chain and posterior cervical regions; largest measuring ~ 36 × 11 mm along the upper jugular chain, 30 × 17 mm along the mid jugular chain, 38 × 17 mm along the lower jugular chain and 23 × 14 mm along the posterior cervical region. These nodal masses show evidence of increased peripheral and central vascularity. There is a mass-effect noted on the left internal jugular vein. A similar heterogeneous node measuring 8.4 × 6.1 mm is noted along the left paratracheal location. Fine needle aspiration cytology (FNAC) from the suspicious left cervical lymph nodes was suggestive of metastatic PTC. Fiberoptic laryngoscopy was done which showed normal mobility of both vocal cords. The patient was planned for upfront surgery. He underwent total thyroidectomy with central compartment clearance and ipsilateral neck dissection from level II to V and contralateral neck dissection from level II to IV. Intraoperatively, thyroid gland seems to be unremarkable with no palpable nodules. Few suspicious nodes were noted in the central compartment. Multiple enlarged cystic lymph nodes noted at the left jugular chain, largest measuring approximately 3 × 2.5 cm at left level II/III (Fig. 1). Specimen was sent for the histopathological examination. Postoperative recovery of the patient was uneventful.

Histopathological report, shows unremarkable thyroid parenchyma with sparse lymphomononuclear infiltrate. No evidence of malignancy examined in the thyroid gland. Out of 4 lymph nodes in the central compartment, 1 tested positive with extranodal extension (> 2 mm) evident. On the left side, 11 out of 26 lymph nodes were positive for PTC, with multiple nodes showing extranodal extension (> 2 mm); whereas none of the lymph nodes (0/45) were positive on the right side. Final pathological staging reported was pT0N1b (Fig. 2). According to risk stratification, he belonged to intermediate risk category.

Radioactive iodine (RAI) scanning was planned for this patient after 6 weeks. Laboratory investigations were carried out before RAI screening. Sr. TSH was 104.040 µIU/ml, Anti-Thyroglobulin is 22.86 U/ml, Thyroglobulin is 2.84 ng/ml. Iodine-131 whole body scan done 24 h after oral administration of 2 mCi of Iodine-131. Scan findings show evidence of residual functioning thyroid tissue. He was suggested I-131 high dose therapy. He was subjected to therapeutic RAI of 100 mCi. Serum TSH, Thyroglobulin, antithyroglobulin and ultrasonography neck was done every 3 months on follow up which shows no evidence of disease for 1 year post treatment.

Discussion

The explanation of this OTC has been frequently changing from the era of palpation to diagnostic USG. With the dramatically improved resolution in USG and prevalence of USG guided fine needle aspiration, the detection and diagnosis of microcarcinoma in thyroid gland have improved significantly [8].

OTC is defined by WHO as an incidental disease, and generally detected at autopsy or during secondary surgery and is defined as an impalpable thyroid nodule, mostly smaller than 1 cm [9]. OTC has been classified by Boucek et al. and the first type includes patients with benign thyroid disease who are incidentally diagnosed with thyroid cancer after a total thyroidectomy or at autopsy whereas the second type includes patients with papillary microcarcinoma of the thyroid that is found incidentally in imaging tests and the third type includes patients with clinically metastatic thyroid cancer, where the primary tumour is undetectable before surgery but is eventually found in histological specimens. The fourth type includes patients with thyroid cancer localised in ectopic thyroid tissue [10]. Liu et al. [11] presented the fifth type of OTC in which a thyroid gland lesion is diagnosed as benign according to pathological and imaging evaluations,

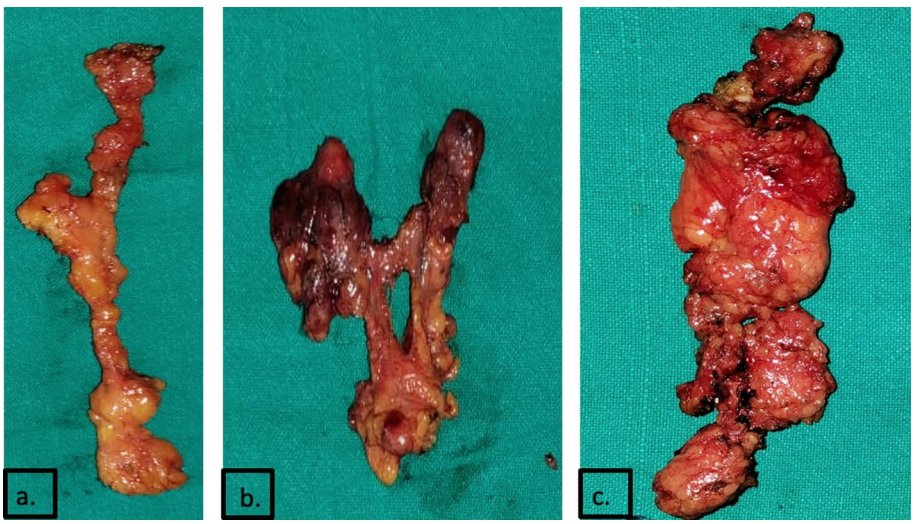

Fig. 1 Surgical specimen showing (**a**) Right Selective neck dissection (II-IV), **b** Total thyroidectomy and central compartment clearance, **c** Left Selective neck dissection (II-V)

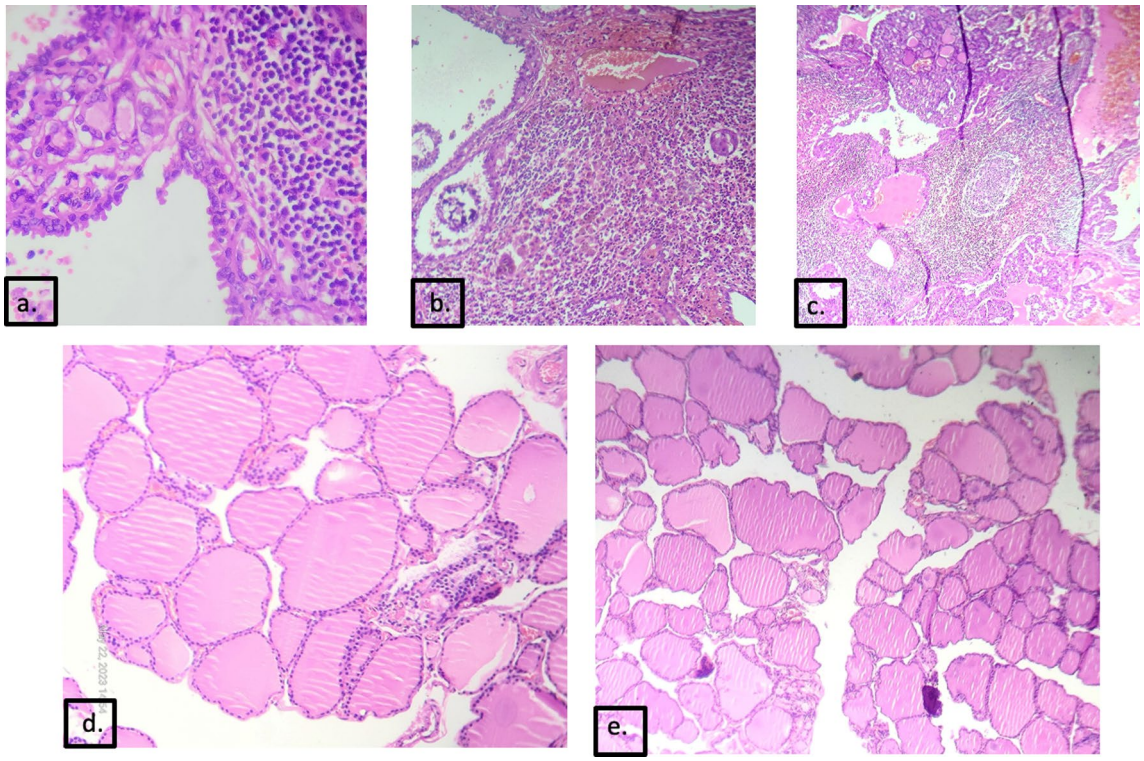

Fig. 2 Histopathological images showing lymph node infiltration by papillary carcinoma deposits. **a** Magnification 40x, **b** magnification 10x, **c** magnification 4x; normal thyroid gland, **d** magnification 10x and, **e** magnification 4x

but metastases of a thyroid carcinoma are detected in either locoregional lymph nodes or distant organs. Patients in the fifth category were further classified into two groups. In the first, metastases of a thyroid carcinoma is detected in locoregional lymph nodes. In the second type, a distant metastatic mass is detected and diagnosed as metastasis from a thyroid carcinoma. Our patient fits into the first group of the fifth type of OTC.

There are a few possibilities why microcarcinomas were not visualised in USG. Ultrasonographic examination has defined resolution limits which can be considered as one of the possible reasons. Second reason is the presence of intra- observer variability. Third possible reason is that USG is unable to comment over certain regions such as retrotracheal, retroclavicular and intrathoracic extensions of thyroid. PTC which are unidentified on USG for any of the above listed reasons and with the presence of lymph node metastasis are diagnosed as occult thyroid carcinoma.

Several possible hypotheses could possibly explain metastasis in cervical lymph nodes without primary tumour in thyroid gland. Spontaneous regression of tumour is a possible reason and is defined as partial or total disappearance of tumour when it has not been treated at all, or when it has not been treated enough to affect the systemic process of the tumour. Another possible reason is that lack of examination of entire thyroid tissue pathologically leading to missed diagnosis. If a lesion is less than 3 mm, there is a high probability of the lesion to be missed on examination. Cole et al. [12] in his study of 176 patients concluded that stimulation of the immune process is the most important factor in spontaneous regression of cancer. In PTC, the immune system also plays a crucial role. Phagocytosis of tumour cells by macrophages has been identified and is associated with an increased incidence of lymph node metastasis, extra thyroidal extension and distant metastasis. In accordance with this, fibrosis in thyroid tissue can be considered as a sign of partial or complete tumour regression [7]. Nishikawa et al. [13] in his case report suggested that presence of distant metastasis in primary occult PTC may be due to immunity or other host resistance factors.

Presence of clinically apparent lymph node metastasis and hoarseness of voice are considered to be prognostic factors as described by Sugitani et al. In his study he classified asymptomatic papillary microcarcinomas as low risk group whereas symptomatic cases were attributed in the high risk category. He concluded the disease specific survival rate to be 96% [14].

The treatment of OTC still remains controversial. Multiple studies have reported to perform lobectomy, hemithyroidectomy and total thyroidectomy along with neck dissection. Treatment should be based on preventing recurrences and metastasis. Low risk group can be considered to be

kept under routine follow up whereas the high risk group should be considered for further treatment such as radioactive ablation. In our case report the patient falls under high risk category so Radioactive ablation was recommended postoperatively.

Conclusion

Occult thyroid carcinoma is rare and is generally misdiagnosed and has no level I evidence for treatment. Based on our report, the ideal treatment seems to be total thyroidectomy with central compartment clearance and functional dissection followed by low dose RAI scan and therapeutic RAI if indicated.

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Declarations

Conflict of interest No potential conflict of interest relevant to this article exist.

Informed Consent Written and informed consent has been taken from the patient for the procedure and all procedures have been done ethically.

Human or Animal Rights No human or animals were used for research in the study.

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