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Warthin-like variant of Papillary thyroid carcinoma—Case report of an uncommon tumour with review of literature

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ABSTRACT

BACKGROUND: Warthin-like variant of Papillary thyroid carcinoma (WLPTC) is an uncommon variant of PTC. They resemble Warthin tumour of salivary gland. Microscopically the tumour shows presence of papillae lined by oncocytic cells with typical nuclear features of PTC. The stalks of papillae were filled up with lymphoplasmacytic cells. WLPTC have good prognosis.
CASE REPORT: A 45-year-old lady presented with thyromegaly. She underwent total thyroidectomy with bilateral selective neck dissection (level II – level VI). Her final histopathology report was WLPTC, Right lobe with lymph nodal metastasis. Post-operatively, she received I131 radio-iodine therapy. She is under follow-up for last four years and is doing well.
CONCLUSION: Diagnosing WLPTC, a rare variant of PTC can be challenging. Definitive diagnosis helps in management.

1. Introduction

Papillary thyroid carcinoma (PTC) is a common malignant tumour of the thyroid gland. There are many histologic variants of PTC. Warthin-like variant of PTC (WLPTC) is a rare variant of PTC. It was first described in the year 1995 by Apel et al. [1,2]. There are about two hundred cases described in English literature to date [3,4]. Microscopically there are papillae lined by oncocytic cells. These cells show typical PTC nuclear features. There is marked lymphoplasmacytic infiltration in the papillary cores. Its microscopic appearance resembles that of Warthin tumour of salivary gland [4]. It is quite a challenge to diagnose these cases in cytology because of the overlapping features between PTC and Hashimoto thyroiditis [5]. Here, we describe one such unusual variant of PTC followed up for last four years.

2. Case history

A 45-year post-menopausal lady had complained of hoarseness of voice for three months. She did not have any significant family or past history. On examination, both lobes of the thyroid gland were enlarged and moved with deglutition. The left lobe of thyroid was measuring 6 × 4 cm and right lobe was measuring 3 × 3 cm. There was a single and mobile right level III lymph node. She was euthyroid. Her routine haematological and biochemical investigations were within normal limit. Indirect laryngoscope revealed right vocal cord palsy. Ultrasonography (USG) of neck showed multiple nodules in both lobes of thyroid. In right lobe, echogenic nodule with complete peripheral halo is seen. Two hetero echoic nodules, larger one in left lobe and smaller similar one in right lobe with lobulated appearance were noted. The USG report was suspicious of malignancy. A round enlarged lymph node was noted in right deep cervical region. In Contrast Enhancing Computed Tomography (CECT), multiple hypodense nodules were noted in both lobes (Fig. 1a). Fine needle aspiration cytology was reported as medullary carcinoma (Bethesda category -VI) at an outside centre. Her serum calcitonin and carcinoembryonic antigen (CEA) level was within normal limit. She underwent total thyroidectomy with bilateral selective neck dissection (level II – level VI).

The operative findings include enlargement of both right and left thyroid lobes. There were few extrathyroidal nodule over thyroid fascia. Right recurrent laryngeal nerve was sacrificed, as it was infiltrated by tumour. Bilateral multiple lymphadenopathy (Right level II-IV, Left level III, IV, VI) were noted. Right internal jugular vein (IJV) was sacrificed as level III lymph nodes were densely adherent to it. Left recurrent laryngeal nerve, right upper parathyroid, left upper and lower parathyroid were identified and preserved (Fig. 1b, c, d). Histopathology revealed the tumour to be composed of neoplastic papillae lined by oncocytic cells showing typical nuclear features of papillary thyroid carcinoma. The cores of the papillae were filled with lymphoplasmacytic cells giving appearance of Warthin's tumour of salivary gland. Lymphovascular invasion was

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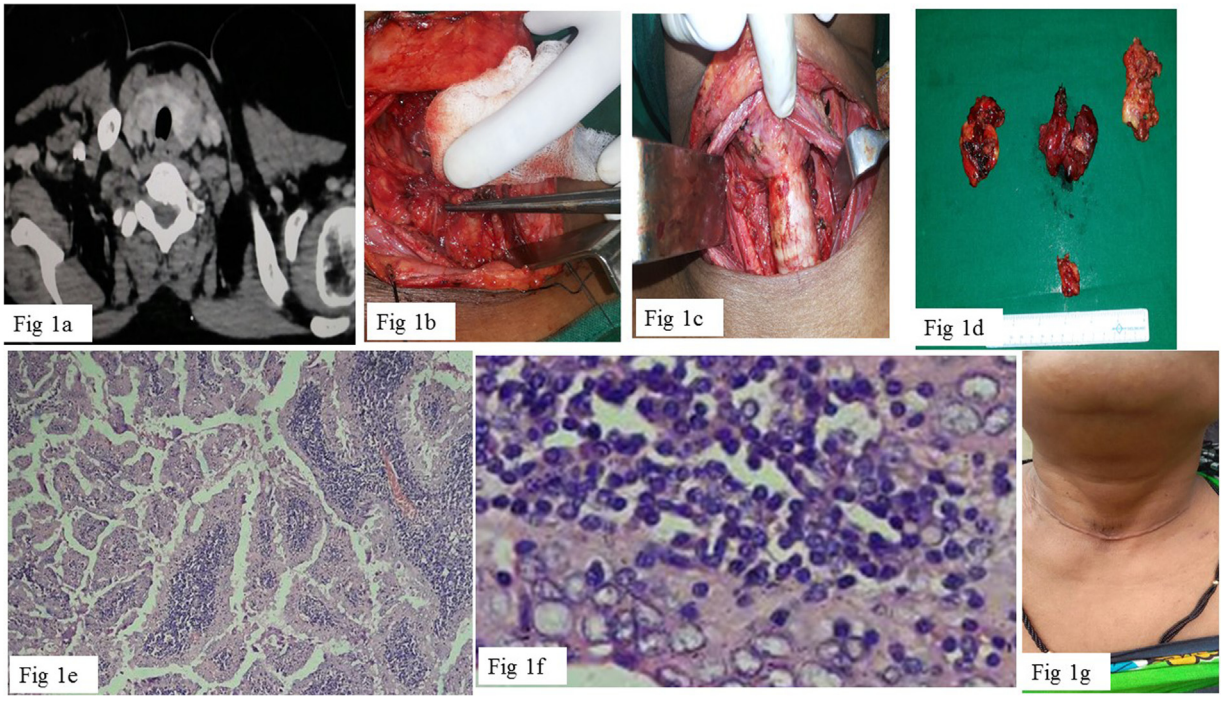

Fig. 1. (a) Contrast Enhancing Computed Tomography (CECT) showing multiple hypodense thyroid nodules in both lobes. (b) Right recurrent laryngeal nerve engulfed by tumour. (c) Post total thyroidectomy and bilateral neck dissection. (d) Total thyroidectomy with bilateral lymph node dissection specimen. (e) Microscopy of Warthin like papillary carcinoma of thyroid (WLPTC) with presence of lymphoplasmacytic infiltrate within the core of papillae (Haematoxylin and eosin x 100). (f) Microscopy of WLPTC showing neoplastic papillae lined by oncocytic cells with ground glass nuclei and lymphoplasmacytic infiltrate within the core (Haematoxylin and eosin x 400). (g) post-operative appearance.

noted (Fig. 1e, f). The final pathological diagnosis was WLPTC, Right lobe. There was multiple cervical lymph node metastasis. In the right-side level II-V, one out of 6, left level II-V, zero out of 14 and level VI, 4 out of 8 nodes showed presence of tumour deposits. IJV was infiltrated by tumour. The pathological staging was pT4aN1b. The adjacent thyroid parenchyma showed features of Hashimoto thyroiditis. Immunohistochemistry was not performed because of the typical morphological feature. Post-operatively, she received I131 radio-iodine therapy. She is on tablet Thyroxin and under regular follow-up for the last four years and is doing well (Fig. 1g). This case has been reported in line with the SCARE criteria [6].

3. Discussion

The World Health Organization (WHO) has classified "Warthin-like tumour" under the oncocytic variant of PTC [5]. Warthin-like PTC is more common in females. The common age group affected is fourth decade [5]. There is limited literature available regarding this tumour. Yeo et al. in their study have concluded that WLPTC is similar to classic PTC coexisting with Hashimoto thyroiditis with regard to demographic, clinical, pathologic, and molecular features [1,7]. In this case also the adjacent thyroid parenchyma showed features of Hashimoto thyroiditis.

WLPTC is difficult to diagnose in cytology as overlapping features of both PTC and Hashimoto thyroiditis can be seen [5].

The gross appearance of WLPTC is usually a well circumscribed, greyish-white nodule. Occasionally cystic and/or haemorrhagic areas may be present. Its mean size of the tumour is 1.5 cm (range 0.3–5 cm). The greatest tumour dimension of the case described here was 4.3 cm. The colour of the adjacent thyroid parenchyma in case of WLPTC ranges from red brownish to tan with nodules of variable size [8].

Microscopically WLPTC exhibits papillary architecture with presence of oncocytic cells lining the fibrovascular core resembling Warthin tumour of salivary gland. There is a prominent lymphocytic infiltrate within the papillary stalk. The nuclear features are that of typical PTC which include enlargement of nucleus, clearing of chromatin, nuclear inclusions and grooves [7]. The essential histological features to diagnose these cases include presence of papillary architecture, oncocytic change, nuclear features of PTC and dense lymphoid infiltrates of the papillary core [5]. The key to the diagnosis remains identifying lymphoplasmacytic cell infiltration within the papillary stalk.

Vascular and capsular invasions are rare [8]. The present case showed presence of lymphovascular invasion with metastatic lymph nodes.

Occasionally WLPTC can exhibit anaplastic areas and in these cases, prognosis is not good [9]. In the present case, anaplastic area was not identified.

Immunohistochemical markers like HBME-1, 34βE12, Galectin-3, Cyclin D1, CD15 (leu-M1) and Cytokeratin 19 (CK19) have been described in few cases [10]. But according to available literature immunohistochemistry has limited to no role in the diagnosis of WLPTC [8]. Since the present case showed characteristic morphological features, immunohistochemistry was not performed.

Oncocytic metaplasia is usually seen in both benign and malignant lesions of thyroid. Occasionally follicular cell-derived tumours like papillary, follicular and even medullary carcinoma can exhibit prominent oncocytic change [5,10]. This may explain the cytological diagnosis of medullary carcinoma in the present case. The common differential diagnosis of WLPTC include Hashimoto thyroiditis, classic type PTC with focal oncocytic change, follicular neoplasm with oncocytic change, oncocytic variant of PTC, classic type of PTC associated with Hashimoto's thyroiditis, tall cell variant of PTC, and oncocytic (Hurthle cell carcinoma) [3]. In Hashimoto thyroiditis, papillae are absent and the nucleus of oncocytes lack typical nuclear features of PTC.

In classic type PTC with focal oncocytic change, as the name suggests oncocytic change is focal and lymphoid stroma is absent. Follicular neoplasm with oncocytic change lacks nuclear feature of PTC, usually lacks papillae as well as lymphoplasmacytic cells in the stalks of papillae. In classic type of PTC associated with Hashimoto's thyroiditis the tumour cells lack oncocytic change and there is absence of lymphoplasmacytic cells in the papillary cores. Tall cell and oncocytic variants of PTC have a poor prognosis. Tall cell variant is characterised by papillae lined by oncocytes whose height is three times more than width of cell, whereas oncocytic variant is characterised by oncocytic cells lining the papillary cores with features of PTC. But both tall cell and oncocytic variants of PTC they lack lymphoplasmacytic infiltrate in the papillary cores [5]. Hurthle cell carcinoma usually lacks lymphoplasmacytic infiltrates and is rarely associated with lymphocytic thyroiditis [8].

The behaviour of WLPTC is similar to that of classical PTC with excellent short- and long-term prognosis. Few cases with presence of anaplastic areas have been described in literature with relatively bad prognosis [9].

The better prognosis in cases of WLPTC is attributed to low rates of nodal involvement and the presence of lymphatic tissue within the tumour. This lymphoid tissue is believed to restrain neoplastic progression [8]. In WLPTC cervical nodal metastasis have been rarely reported [8]. In the present case the tumour was involving right recurrent laryngeal nerve and right cervical lymph nodes. But our patient underwent surgery and post-operative radio-iodine therapy and is doing well after four years of follow-up.

Warthin like papillary carcinoma of thyroid shows BRAF V600E mutation. Maffini et al. have described an additional novel germline point mutation in the RETgene [7,10].

The diagnosis of WLPTC is based on histopathology examination of surgical specimens. So, after surgery and histopathology diagnosis further therapy is planned [7]. The management of WLPTC patient is similar to that of PTC and depends on the stage of the disease. The presence of familial history, history of neck irradiation, and associated syndromic endocrine disease are indicators of bad prognosis [8].

To conclude, this is an additional case of WLPTC. Though this patient had lymph node metastasis, still she is doing well four years after treatment.

Declaration of Competing Interest

None.

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Ethical approval

Since this is a case report ethical approval is exempted.

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.

Author contribution

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