Papillary carcinoma in submandibular region – a diagnostic dilemma

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Introduction

Papillary carcinoma is the most common variant of thyroid carcinoma. It accounts for 50–70% of thyroid malignancies in patients under the age of 40 yrs [1, 2]. Thyroid carcinoma usually presents as midline neck mass. Nodal metastasis to the neck may be the first presenting symptom in papillary carcinoma thyroid. Such "occult" presentation of well differentiated thyroid carcinoma is seen in 10–26% of thyroid malignancies [3, 4]. In these patients, the sole presenting symptom is lateral neck mass with no evidence of primary in thyroid gland. However thyroid carcinoma presenting as submandibular swelling is not common.

We report a patient of papillary carcinoma thyroid, who presented with metastatic neck swelling in the submandibular region without evidence of primary in the thyroid region.

Case report

A 40-yr-old male presented to the outpatient department of Otolaryngology and Head and Neck Surgery, P.G.I.M.E.R., Chandigarh, India, with complaint of asymptomatic swell-

ing in the right side of neck for the last 3 yrs. The swelling first appeared in the submandibular region, it was spontaneous in onset and slowly progressive. The swelling increased in size from a pea to the present size of apple. The swelling had no relation with intake of meals. There was no history of change in the voice or dysphagia. There was no history of irradiation in the past. Examination revealed an 8X5cm non-pulsatile non-fluctuant mass on the right submandibular region of the neck. The swelling was non tender, firm in consistency with restricted mobility. Rest of the head and neck examination including thyroid gland was normal.

Contrast enhanced computed tomography scan of neck revealed multiseptate cystic mass in the right submandibular region of neck. The Submandibular gland was normal and appeared to be free from the swelling (Fig. 1). The thyroid gland was also normal on CT with no evidence of any suspicious nodule or circumscribed lesion (Fig. 2). A fine needle aspiration of the swelling yielded 10ml brownish hemorrhagic fluid, and the smears from the same were reported as low grade mucoepidermoid carcinoma. Thereafter, surgical excision of mass was done using a transverse cervical incision. The mass was cystic at places containing serous fluid under high pressure. The right submandibular gland was normal and free from the swelling. The mass was infiltrating the internal jugular vein. There were three lymph nodes measuring 3 × 3 cm in size, close to the upper part of the upper part of internal jugular vein. The lymph nodes were dissected and sent separately for histological examination.

Gross examination of the specimen showed nodular mass with congested outer surface and the cut surface revealed multiple cystic areas filled with hemorrhagic fluid and a tumor with papillary excrescences. Histopathological examination revealed it to be papillary carcinoma thyroid. The cyst wall showed fibroblastic proliferation and two of the three lymph nodes showed tumor metastasis. A tumor arising from aberrant location of thyroid gland was suggested.

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Further investigations were then done in the light of the histopathology report. The ultrasound examination of the neck showed a 1.04×1.03 cm well defined heterogeneous nodule with specks of calcification in the superior aspect of right lobe of the thyroid. There was also a 1.24×1.06 cm enlarged lymph node seen along the jugular vein at the level of the thyroid gland. An Ultrasound guided F.N.A.C was done which revealed predominantly blood and occasional clusters of benign follicles. Thyroid function tests were within normal range. Sodium pertechnetate (Tc^{99m}) scan demonstrated normal sized thyroid gland with normal trapping function.

In the light of the histopathology report and ultrasound findings of thyroid nodule, a total thyroidectomy with preservation of both recurrent nerves was performed. Histopathology of surgical specimen was reported as multifocal papillary carcinoma of thyroid. The patient made an uneventful recovery after the surgical procedure (Fig. 3).

Fig. 1 Contrast enhanced CT scan of neck showing multiseptate cystic mass in the R submandibular region. There is thickening of cyst wall. Submandibular gland is pushed medially

Fig. 2 Contrast enhanced CT scan of neck showing normal thyroid gland

Fig. 3 Micropotograph shows papillae with nuclear stratification and crowding. HPE x280. Insert shows optically clear nuclei

Discussion

The presence of aberrant thyroid deposits of thyroid tissue in the lateral part of neck has been recognized since early part of nineteenth century [5]. Low was the first to point out that lateral papillary thyroid tumors may be associated with similar lesions in the ipsilateral thyroid lobe [6]. Ectopic thyroid tissue usually occurs in the midline anywhere from base of tongue to mediastinum and occasional examples in the lateral part of neck have been described [7, 8, 9]. However papillary thyroid carcinoma arising from aberrant deposits of thyroid tissue in the submandibular region has not been reported before in literature. The present case had a papillary thyroid carcinoma in the right submandibular region probably arising from the aberrant thyroid tissue. Also there was a multifocal papillary thyroid carcinoma in ipsilateral thyroid lobe. This association is being reported for its rare occurrence.

Papillary thyroid carcinoma from aberrant deposits of thyroid tissue in submandibular region can give rise to diagnostic dilemma for both the clinician and the pathologist. Common differential diagnosis of this region includes submandibular lymph node, benign and malignant lesions of submandibular gland and branchial cyst. However as we found in our case, possibility of aberrant thyroid gland should also be in differential diagnosis, though rare.

This raises the issue of preoperative thyroid scan in patients with neck mass and in which thyroid gland is not palpable. Since lateral aberrant thyroid presenting as lateral neck mass is rare, we propose preoperative thyroid scan in all patients presenting with midline and low lateral neck mass which might identify hyper functioning areas in thyroid that could be malignancy. Fine needle aspiration cytology is a valuable diagnostic tool with sensitivity of 85–90% [10]. However in cystic lesions of neck its sensitivity is only 50% and may cause diagnostic dilemma [11].

Previously, a lateral aberrant deposit of thyroid tissue was thought to be due to cell rest of thyroid tissue deposited during development of thyroid gland. This theory has long been discredited. The theory that ectopic thyroid tissue is metastatic thyroid carcinoma was proposed by Clay and Blackmann. This view is generally accepted view and is true as we found in our case also. The thyroid gland had focus of primary in the superior lobe which was detected on ultrasound of neck.

Ideal treatment in these cases includes total thyroidectomy with modified neck dissection.

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