Introduction

Cancer of the thyroid gland is the most common endocrine malignancy constituting for less than 1% of all reported human cancers. The propensity of occurrence of thyroid carcinomas is two to four times more in females as compared to males with a median age at diagnosis being 45–50 years [1,2]. Papillary thyroid carcinoma (PTC) is the most common histological type of thyroid cancer and accounts for more than 80% of thyroid malignancies. Classical PTC (cPTC) is the most common (80%) subtype of papillary thyroid carcinoma followed by follicular variant (FVPTC) which is found in 9–22.5% of patients [3].

FVPTC was first described by Crile and Hazard in 1953 as alveolar variant of PTC. It was subsequently confirmed by Lindsay, Chen and Rosai as an independent entity with typical nuclear features and follicular growth pattern. FVPTC further has histological variants namely, completely encapsulated form, well circumscribed form, and infiltrative form, the last being the most aggressive [2,4]. The encapsulated variant is relatively more prevalent than the other sub-type and is non-aggressive in nature. In order to distinguish it from the invasive variant, histologically, it has been renamed as 'noninvasive follicular thyroid neoplasm with papillary-like nuclear features'.

Invasive follicular variant of PTC presents several diagnostic and management challenges [5]. Late metastasis to the mandible is extremely rare and could be the only sign of a silent underlying neoplasm. Such lesions may resemble odontogenic lesions and should be carefully differentiated from them. This case report is a description of rare late mandibular metastases in a previously diagnosed case of invasive follicular variant.
with complaints of lower back pain and scalp swelling of 1 year duration. On examination, a 5 cm x 4 cm sized discrete soft swelling was palpable in occipito-parietal region and 1 cm x 1 cm sized nodule was palpable in left lobe of thyroid. Upon evaluation was normal with mobility seen in both vocal cords. Ultrasonographic examination (USG neck) demonstrated a destructive expansile lesion over the left ramus extending to the condyle and coronoid process. (Figure 4).

Investigations

An Orthopantograph (OPG) examination showed extensive diffuse multilocular radiolucency with ill-defined borders over the left ramus extending up to the condyle superiorly, anterior margin of ramus anteriorly and angle of the mandible inferiorly (Figure 3). The lesion showed mixed areas of dense radiolucency interspersed with areas of radio-opacity. These findings were consistent with a differential diagnosis of an odontogenic carcinoma and/or, metastatic disease.

A contrast–enhanced Computed Tomography (CECT) scan demonstrated a destructive expansile lesion over the left ramus of the mandible extending to the condyle and coronoid process. (Figure 3).

Diagnosis

The positive findings of the CECT scan along with clinical findings negative for an inflammatory lesion and a history of invasive FVPTC helped in arriving at the diagnosis of metastatic disease of the mandibular ramus.

Management

Due to the extensive nature of bony destruction which had already occurred including involvement of left parieto-occipital region of scalp, surgical resection of the lesion was not advised. Since the patient did not complain of significant pain associated with the lesion, palliative radiotherapy of 30 Gy equally divided in 10 fractions was administered to the disease site.

Citation: Dholam KP, Singh GP, Gurav SV, Rekhi BB (2017) Rare Late Mandibular Metastasis in Follicular Variant of Papillary Carcinoma Thyroid: ‘Resurgence of the Sleeping Tumour’. Arch Otolaryngol Rhinol 3(1): 001-05. DOI: http://doi.org/10.17352/2455-1759.000032
Discussion

Metastatic lesions are the first clinical sign of many unknown primary tumors. Metastatic tumors to the oral and maxillofacial region are rare and account for approximately 1% of all malignant oral tumors. Malignancies can metastasize to the jaw bones either through blood vessels or lymphatics, with the hematogenous route being most common one. In females, tumors arising from breasts, adrenal, colorectum, cervix and ovary and thyroid more commonly metastasize to maxilla and/or mandible. On the contrary, in males, lung followed by the prostate, kidney, bone, and adrenal show distant metastases to jaw bones. Thyroid carcinoma with isolated mandible metastasis accounts for 3.85% of all jaw metastasis. A review of literature suggests that jaw metastases is more frequent than to oral mucosa in the ratio of 2.5:1. [6]

Mandible is more commonly affected than maxilla with the premolar-molar region being the most frequent site of metastasis. There is a high propensity of metastases to the body of the mandible as compared to ramus due to the presence of hematopoietic bone marrow and thus greater entrapment of metastatic cells as a result of sluggish blood flow [6–8].

Metastatic carcinomas affecting either maxilla or mandible should always be considered in the differential diagnosis of a long-standing, painless facial swelling in old individuals. Such carcinomas are difficult to distinguish from inflammatory and reactive lesions of oral and maxillofacial region [6]. Extra care should be taken so as not to miss any such metastatic lesion affecting ramus since it is covered by the parotid gland [5].

In this presented case, mandibular ramus was affected and OPG showed extensive area of radiolucency with ill-defined borders over the left ramus extending onto the condyle superiorly, anterior border of ramus anteriorly and angle of the mandible inferiorly. These findings were confirmed in a CECT scan and were thus, consistent with a differential diagnosis of an odontogenic carcinoma and/or metastatic disease.

Invasive FVPTC presents several diagnostic and management challenges to the clinician. It has hybrid metastatic capacities with lymph node metastasis via the lymphatic system similar to conventional papillary thyroid carcinoma (c-PTC), as well distant metastasis via the bloodstream similar to follicular thyroid carcinoma (FTC) [5].

Only about 20% of patients with differentiated thyroid carcinomas show a metastatic evolution. In 50–80% of cases, there are multiple bone metastases. According to different studies, bone metastases are more common in patients with follicular carcinomas (15.2–33.7%) than in those with papillary carcinomas (0.6–6.9%). Sternum, ribs, and spine are the most frequent sites of osseous metastases in such differentiated thyroid carcinomas [9].

A literature review done by Nikitakis et al. [10], in 2012 revealed only 37 published cases collectively, of all forms of thyroid cancer metastasizing to the oral cavity [11–40]. Metastases to the maxilla and/or mandible are extremely rare in histologically invasive FVPTC with literature reports of only a few cases. Pal et al. [41], Bhadage et al. [42], Bingol et al. [6], and others [7, 43], have reported metastasis to the body and angle of mandible manifesting in invasive form of FVPTC. However, ramus metastasis is extremely rare and has been described in the literature by Saha et al. [8], and Noolkar et al. [44]. A solitary case of maxillary metastases has been cited by Bhansali et al. [45].

In the presented case, the patient was diagnosed with invasive FVPTC with sacral metastases in the first instance. In accordance with the Tata Memorial Hospital guidelines for evidence-based management vol. XI A 2012 [46], total thyroidectomy followed by 260 mCi of I131, radioactive ablation was carried out. Palliative EBRT of 20 Gy equally distributed in 5 fractions was given to locally manage sacral metastases.

Distant metastases to the ramus of mandible were detected after 2.5 years of completion of the primary treatment. Moreover, the patient was pain-free and had reported to the department with a complaint of seemingly ‘harmless’ swelling. On further investigations it was confirmed as a metastatic lesion involving the ramus of the mandible for which 30 Gy of palliative EBRT was advised. However, in a patient afflicted by metastases to the mandibular body region, Bingol et al. [6], performed hemimandibulectomy followed by reconstruction with costo-chondral rib graft as a definitive procedure.

Conclusion

Metastasis from invasive FVPTC to mandible is a rare occurrence and should be kept in mind with the other differential diagnoses of tumors in the facial region. Despite the relatively indolent behavior of encapsulated follicular thyroid neoplasm, the 5-year survival rate in patients detected with bony metastases in invasive FVPTC is reported to be 79.4% with 10-year survival rate being 52.9% [47].

Hence, there is need for a long term, strict vigilant follow up of a patient diagnosed with invasive FVPTC as evident with our experience. Metastatic FVPTC is of greater significance, since at times its appearance may be the only symptom of an undiscovered underlying malignancy and metastatic lesions may be the first or only clinical manifestation.

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Acknowledgements

The patient has given her consent to the use of clinical photographs for educational purposes.

The authors wish to declare no conflict of interest in the work submitted.

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