

(CASE REPORT)



Mandibular tumor revealing metastatic papillary thyroid carcinoma: A case report

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Abstract

Introduction: Papillary thyroid carcinoma (PTC) is the most common thyroid malignancy, frequently metastasizing to regional lymph nodes but rarely presenting with distant metastases. Involvement of the mandible as the initial manifestation of metastatic PTC is exceedingly rare.

Case Presentation: We report the case of a 66-year-old female presenting with a progressive mandibular swelling over 12 months. Clinical examination revealed a firm mandibular mass and a long-standing anterior cervical swelling evolving over 10 years without associated dyspnea or dysphonia. Imaging studies, including a CT scan, demonstrated a mandibular lesion, pulmonary metastases, and a goiter. Biopsy of the mandibular lesion confirmed the diagnosis of metastatic papillary thyroid carcinoma. Given the presence of distant metastases, the patient was deemed unsuitable for surgical management and was treated with systemic chemotherapy.

Conclusion: This case highlights the exceptional presentation of metastatic PTC with mandibular involvement and underscores the importance of thorough evaluation of atypical mandibular lesions to identify underlying malignancies.

Keywords : Mandibular tumor, papillary thyroid carcinoma, bone metastasis, thyroid cancer metastasis.

1. Introduction :

Papillary Thyroid carcinoma (PTC) is a common thyroid malignancy with a generally favorable prognosis. While regional lymph node metastases are frequent, distant metastases occur in approximately 1–4% of cases, with the lungs and bones being the most common sites. Mandibular involvement is a rare phenomenon and seldom the first clinical presentation. (1)

Metastatic tumors to the oral cavity are extremely rare, accounting for only 1%–2% of all cancers. The breast, lung, kidney, prostate and gastrointestinal tract (GIT) are the most common main sites, and it affects both men and women (1). Thyroid cancer is the most prevalent type of cancer in the overall endocrine system, and papillary thyroid carcinoma (PTC) is the most common type. It generally spreads to the lungs and bones, and rarely to the oral cavity (1,2)

This case report discusses a rare instance of a mandibular tumor revealing metastatic PTC, emphasizing diagnostic challenges, therapeutic strategies, and the relevance of interdisciplinary management.

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2. Case presentation :

We present the case of 66-year-old female patient presented with a progressively enlarging anterior mandibular swelling over 12 months, associated with a difficulty of chewing without pain nor numbness.

The clinical examination revealed an anterior firm painless mass of the mandible without any signs of inflammation nor infection. The patient's performance status (PS) was 1 .

The intraoral examination revealed a mass displacing the alveolar bone, with the patient being edentulous. (Figure 1)

Cervical examination revealed a long-standing anterior cervical mass with no lymph nodes (reported evolution of 10 years), which the patient had not previously investigated. No signs of airway compression or significant symptoms were reported. (Figure 2)

A computed tomography (CT) scan revealed a lesion in the anterior mandible with hyperdensity suggestive of metastatic involvement. Additionally, the scan detected a multinodular goiter and evidence of pulmonary metastases. (Figure 3,4)

A biopsy of the mandible lesion was performed and the histopathological examination revealed a metastatic papillary thyroid carcinoma.

Given the presence of distant metastases, the patient was considered unsuitable for surgical management and was treated with systemic chemotherapy. Unfortunately, the patient died 9 months after the diagnosis.



Figure 1 : The clinical examination showing an anterior mandibular mass with displaced alveolar bone



Figure 2 : Cervical examination revealing an anterior cervical swelling

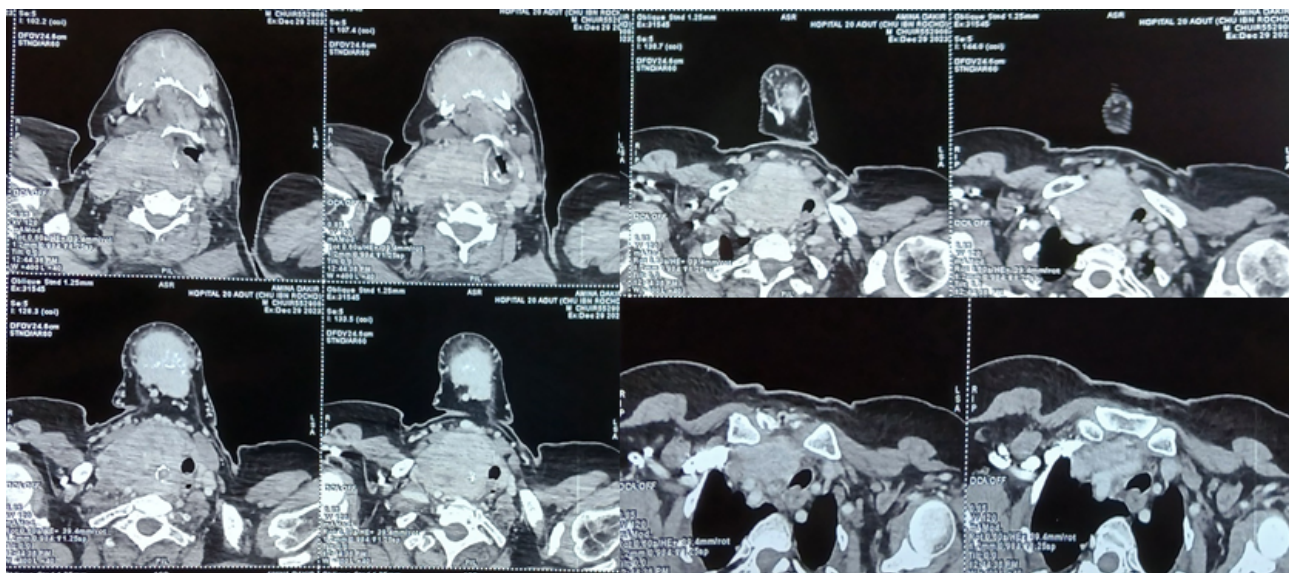


Figure 3 : CT-Scan revealing a giant anterior mandibular mass along with multinodular goiter

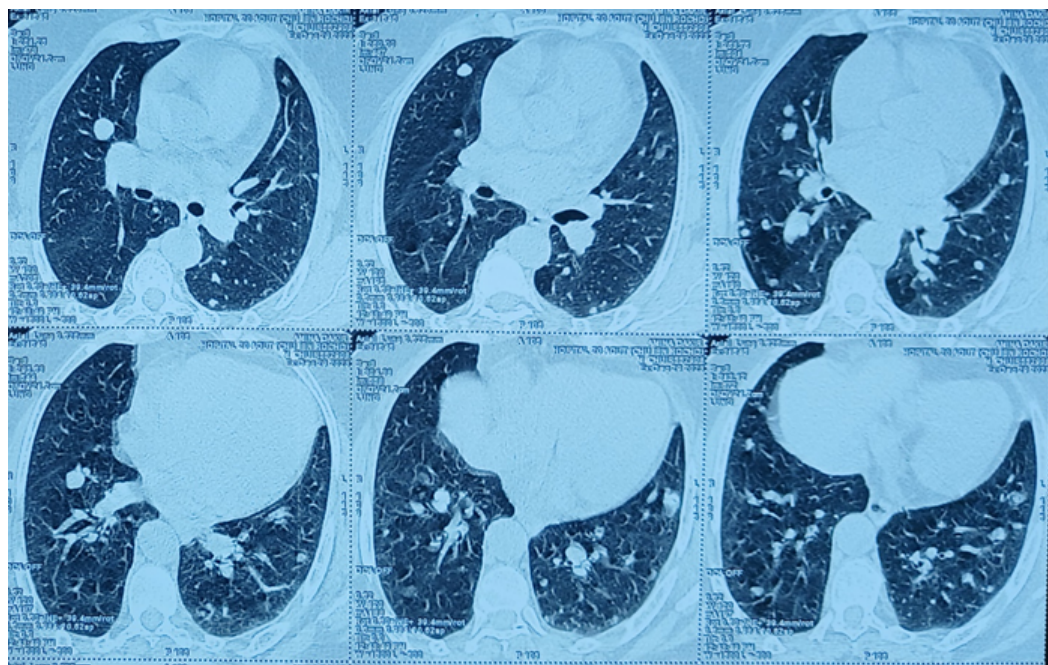


Figure 4 : Pulmonary metastasis on the thoracic CT-Scan

3. Discussion :

This case highlights the unusual metastatic behavior of papillary thyroid carcinoma, a disease typically associated with a favorable prognosis when confined to the thyroid and regional lymph nodes (3). Metastasis to the mandible is rare and may be the first manifestation of an underlying thyroid malignancy. Early recognition of metastatic lesions, especially in atypical sites, is crucial for prompt diagnosis and treatment planning.

Classic osseous regions for thyroid metastasis include the sternum, the vertebrae, and the pelvis (1). Most of the previously reported metastatic thyroid carcinomas of the oral cavity had been follicular thyroid carcinoma (FTC), and metastatic PTC was very rare (4–6). Hurtle cell, poorly differentiated, and medullary carcinomas have been also reported in the mandible (3,4). PTC is the most common microscopic variant of thyroid cancer. Moreover, it has a minimal potential for distant metastasis and tends to spread via lymphatic invasion (3). It demonstrates an asymptomatic slow-growing thyroid mass, which is sometimes discovered incidentally (4). Distant metastasis occurs in about 7-23% of patients with thyroid carcinoma (7).

In their literature review, Gholam et al. (3) found 26 cases from 16 articles. The mean age was 54.4 (ranging from 13 to 69 years). These malignancies occurred most often in the sixth and seventh decades of life, only 3 patients were under the age of 40, and no patient was found in the 8th decade of life.

Nikitakis et al. (4) reported that the mean age of the metastatic thyroid carcinoma patients was 60.6 years with a tendency to occur in the 7th decade.

Considering the age of occurrence for the primary thyroid carcinoma, which is about 40 to 50 years old, an increase in the mean age of metastasis is related to the amount of time that metastasis needs to develop and be detected (3,4).

There was a striking female predilection (88.46%) with a ratio of 7 : 66 (4). This high tendency towards females is associated with the high incidence of thyroid carcinoma in women.

Among 25 patients with available tumor locations, the mandible was the most common site (80%) (3). Other sites of involvement were the maxilla and the tongue. The tendency for spreading to the mandible may be associated with its greater volume of hematopoietic tissue that delivers easy access to neoplastic cells (8). They tend to be more common in the jaws in comparison to oral soft tissues (4%). PTC variant comprised 53.84%. Among the 19 cases with available data about the time of metastasis, 42.10% were not aware of their primary cancer (3). Nikitakis et al. (4) in their review found that oral metastasis had been reported as the first sign of an unknown primary thyroid carcinoma in more than half of the metastatic thyroid carcinoma cases. In our case, the mandibular metastasis was the first sign revealing the thyroid carcinoma.

Distant metastasis has a crucial role in the patients' management (9,10), and oral metastasis is a therapeutic challenge for clinicians (11). It ranges from palliation to several combinations of surgery, radioactive iodine, radiotherapy, and chemotherapy (12,13).

A meta-analysis of randomized controlled trials was conducted by Liu JW et al. (14) to evaluate the effectiveness and safety of tyrosine kinase inhibitors (TKIs) for advanced or metastatic thyroid cancer treatment, which showed TKIs significantly improved Progress-Free Survival and objective response rate in patients with advanced or metastatic thyroid cancer.

In our case, chemotherapy was indicated giving the pulmonary metastasis. The 10-year survival rate for metastatic thyroid carcinomas is 13-21% (3,12). Unfortunately, the presented case died 9 months after the diagnosis.

The coexistence of mandibular involvement, a long-standing cervical mass, and pulmonary metastases reflects the complexity of late-stage PTC. This case highlights the challenges of managing advanced thyroid carcinoma, particularly when the patient is ineligible for definitive surgical intervention.

4. Conclusion :

Although very rare, PTC can metastasize to the oral cavity. Oral metastasis may be the first symptom of a malignancy in other parts of the body. This emphasizes the role of oral professionals in identifying metastatic lesions. Metastatic PTC

tends to occur in females, with a very high proportion, most often in the sixth and seventh decades of life. Additionally, there is a striking predilection for mandibular involvement. This case underscores the importance of considering metastatic papillary thyroid carcinoma in the differential diagnosis of mandibular tumors. A multidisciplinary approach is essential for optimal management and improving patient outcomes.

Conformité aux normes éthiques

Déclaration de conflit d'intérêts

Aucun.

Déclaration d'approbation éthique

Le présent travail de recherche ne contient aucune étude réalisée sur des sujets humains ou animaux par aucun des auteurs.

Déclaration de consentement éclairé

Le consentement éclairé a été obtenu de tous les participants individuels inclus dans l'étude.

Références

1. Jawanda MK, Narula R, Gupta S, Gupta P. Mandibular metastasis of follicular thyroid carcinoma: A case report along with the concise review of literature. *J Oral Maxillofac Pathol.* 2022;26(1):133.
2. Anajar S, Hassnaoui J, Rouadi S, Abada R, Roubal M, Mahtar M. Mandibular metastasis revealed papillary thyroid carcinoma: Rare case. *Int J Surg Case Rep.* 2017;37:130-3.
3. Gholami S, Bakhshi M, Atarbashi-Moghadam S, Mir Mohammad Sadeghi H, Rahimzamani A. Mandibular Metastasis of Silent Papillary Thyroid Carcinoma: A Rare Case Report with Review of the Literature. *Case Rep Dent.* 2020;2020:8683465.
4. Nikitakis NG, Polymeri A, Polymeris A, Sklavounou A. Metastatic Papillary Thyroid Carcinoma to the Maxilla: Case Report and Literature Review. *Head Neck Pathol.* 30 déc 2011;6(2):216-23.
5. CoLab [Internet]. [cité 1 déc 2024]. Gnathic bone metastasis: a retrospective study of 10 cases. Disponible sur: <https://colab.ws/articles/10.1016%2Fs1808-8694%2815%2930603-0>
6. Seoane J, Van der Waal I, Van der Waal RIF, Cameselle-Teijeiro J, Antón I, Tardio A, et al. Metastatic tumours to the oral cavity: a survival study with a special focus on gingival metastases. *Journal of Clinical Periodontology.* 2009;36(6):488-92.
7. (PDF) Mandibular Metastasis of Thyroid Carcinoma: A Case Report. ResearchGate [Internet]. 22 oct 2024 [cité 1 déc 2024]; Disponible sur: https://www.researchgate.net/publication/265213902_Mandibular_Metastasis_of_Thyroid_Carcinoma_A_Case_Report
8. Nawale KK, Vyas M, Kane S, Patil A. Metastatic tumors in the jaw bones: A retrospective clinicopathological study of 12 cases at Tertiary Cancer Center. *Journal of Oral and Maxillofacial Pathology.* août 2016;20(2):252.
9. Wexler JA. Approach to the thyroid cancer patient with bone metastases. *J Clin Endocrinol Metab.* août 2011;96(8):2296-307.
10. Bingol UA, Cinar C, Cetinkale O. Mandibular metastases of papillary thyroid carcinoma treated by hemimandibulectomy and costochondral rib graft. *Plast Reconstr Surg Glob Open.* févr 2015;3(2):e305.
11. Bejarano PA, Nikiforov YE, Swenson ES, Biddinger PW. Thyroid transcription factor-1, thyroglobulin, cytokeratin 7, and cytokeratin 20 in thyroid neoplasms. *Applied Immunohistochemistry and Molecular Morphology.* 2000;8(3):189-94.

12. Fatahzadeh M, Subramanian G, Singer SR. Metstatic papillary thyroid carcinoma to maxilla: a rare case. *QUINTESENCE INT.* 1 mai 2015;46(5):431-5.
13. American Thyroid Association (ATA) Guidelines Taskforce on Thyroid Nodules and Differentiated Thyroid Cancer, Cooper DS, Doherty GM, Haugen BR, Kloos RT, Lee SL, et al. Revised American Thyroid Association management guidelines for patients with thyroid nodules and differentiated thyroid cancer. *Thyroid.* nov 2009;19(11):1167-214.
14. Liu JW, Chen C, Loh EW, Chu CC, Wang MY, Ouyang HJ, et al. Tyrosine kinase inhibitors for advanced or metastatic thyroid cancer: a meta-analysis of randomized controlled trials. *Curr Med Res Opin.* mai 2018;34(5):795-803.
15. Kim BH, Kim IJ, Lee BJ, Lee JC, Kim IS, Kim SJ, et al. Detection of plasma BRAF(V600E) mutation is associated with lung metastasis in papillary thyroid carcinomas. *Yonsei Med J.* mai 2015;56(3):634-40.
16. Rajjyoti Das, Mahesh Kumar, Jagannath D. Sharma, et al., Papillary thyroidcarcinoma presenting with mandibular metastasis: an unusual presentation,Clin. Cancer Investig. J. 3 (2014) 426–428.