Mandibular metastasis revealed papillary thyroid carcinoma: Rare case

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**Article info**

**Abstract**

**Introduction:** Papillary carcinoma is the most frequent differentiated malignant thyroid neoplasm. Metastasis occurs frequently in regional lymph nodes and mandibular metastasis are very rare and most are secondary to follicular carcinomas due to their blood diffusion. The mandibular metastasis of papillary carcinoma is exceptional.

**Case report:** We report a rare case of mandibular metastasis revealing papillary thyroid carcinoma in a 52-year-old patient, with a review of the literature on clinical features, radiological aspect, and treatment options.

**Discussion:** Mandibular metastasis of thyroid cancer are rare and the initial metastases revealing papillary carcinoma are exceptional, few cases are reported in the literature, and due to their rarity and relative lack of data on their management, there is no clearly defined processing algorithm.

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1. Introduction

Papillary carcinoma is the most common cancer of the thyroid, accounting for about 80%–90% of thyroid cancers [1]. The most common sites of revealing metastases are lung and bone [1]. The presence of distant metastasis is an element of poor prognosis associated with a decrease in survival rates [2]. Mandibular metastasis are very rare [3] and most are secondary to vesicular carcinomas due to their blood diffusion [4]. Initial mandibular metastases are extremely rare with few cases reported in the literature [5]. We present a case of initial mandibular metastasis revealing papillary carcinoma of the thyroid. This work has been written in accordance with the SCARE criteria [6].

2. Observation

A 52-year-old North African woman was referred to the otolaryngology clinic with a complaint of left painless mandibular tumefaction, evolving gradually for 2 months. The mass was hard, integral with the left half-mandible. No effect on the teeth and occlusion was seen. Moreover, there was no disorder of mucocutaneous sensation in the territory of the lower alveolar nerve, and the rest of the somatic examination was unusual. Panoramic radiography showed a osteolytic lesion (mass) in the left hemi-mandibular with cortical expansion (Fig. 1). An incisional biopsy was performed intraorally. The histopathologic study revealed a metastasis of papillary thyroid carcinoma (Fig. 2).

The cervical ultrasound showed an enlarged thyroid-sized, seat of several nodules classifying TI-RADS 4B (Thyroid Imaging-Reporting and Database System) without cervical nodes. No other metastatic lesions were detected in whole body scan. The patient underwent total thyroidectomy and the anatomopathological evaluation was compatible with thyroid papillary carcinoma (Fig. 3). After 15 days the mandibular mass was removed by segmental mandibulectomy and the bone defect was reconstructed with a titanium reconstructive plate (Fig. 4). The patient had no recurrence after 6 months of follow up.

3. Discussion

Although the true incidence of metastatic tumors to the jaw bones is unknown, these metastatic lesions make up around 1% of all oral malignancies [7]. Both the jaws and the oral soft tissues may be affected by metastatic cancer with a predilection for the mandible and the gingiva, respectively [7]. In general, oral metastases to the maxilla are rare, corresponding to less than one-fifth of all metastatic tumors to the jaws [8]. It has been suggested that the predilection of metastasis to the ramus and angle of the mandible reflects the rich blood circulation in the medullary cavity of these regions [8].

The symptoms of a metastatic tumor in the mandible include pain, swelling, loosening of teeth, paresthesia, cervical lym-

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Fig. 1. Panoramic Radiography Showed Osteolytic Lesion.

Fig. 2. Histopathologic Study Revealed a Metastasis of Papillary Thyroid Carcinoma.

Fig. 3. Total Thyroidectomy.
phadenopathies and rarely pathologic fracture [9,10]. In some cases these symptoms or signs may be the only manifestation of an undiscovered disseminated malignancy [11].

Mandibular metastasis due to thyroid carcinoma is not very frequent and the cases described in the literature are few. Due to its bloodstream dissemination, most of them are a consequence of the follicular variant of thyroid carcinomas [5]. Papillary carcinomas usually remain intraglandular or give only cervical nodes metastasis [2]. Initial mandibular metastasis are extremely rare with Only four cases of mandibular metastasis of papillary thyroid carcinoma have been previously reported in the literature [2–4] the present case being the five. The pathogenesis of well differentiated thyroid cancers and its promulgation to bone has yet to be fully elucidated, the individual prognosis depends upon age at diagnosis of metastasis, tumor burden and the number of bony metastases [2].

A great majority of the appendicular skeletal metastasis from thyroid cancers can be effectively managed by external beam radiation therapy or radioactive iodine ablations [12,13], however, some of the bony metastasis require surgical intervention due to the associated symptoms and fracture risk [12,14].

The treatment of oral metastases of thyroid carcinoma has varied extensively from palliative to various combinations of surgical resection, radioactive iodine treatment, radiotherapy, and/or chemotherapy [15]. It has been recommended that surgical resection of the metastatic tumor in conjunction with total thyroidectomy (if not performed earlier), followed by radioactive iodine treatment and/or external-beam radiation may afford better survival [15].

Surgical intervention is usually recommended for isolated, solitary and accessible metastases [16]. In patients with multiple site involvement, the role of metastectomy is less well understood. There have been reports that have shown that removal of up to five bony metastases can be associated with improved survival and quality of life [12,16].

The free fibula flap continues to remain the “gold standard” and best option for mandibular reconstruction. In our patient, the bone defect was 6 cm. The size of the defect was not a clear indication for free fibula flap. We prefer a reconstruction using only a titanium plate [17].

Prognosis of patients with distant thyroid cancer metastases is generally poor, with an average of 40% of patients alive 4 years after the diagnosis of metastasis and an overall 10-year survival rate of 27% for bone metastases of differentiated thyroid carcinoma [18].

4. Conclusion

Although relatively rare, metastatic lesion should be considered in the differential diagnosis of mandibular lesions. And the absence of firm management guidelines, clinicians should use their judgment in choosing patients for surgical management of bony metastasis from thyroid cancers.

Conflicts of interest

The authors declare having no conflicts of interest for this article.

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Ethical approval

Written informed consent for publication of their clinical details and/or clinical images was obtained from the patient.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying image.

Authors contribution

Said Anajar: Corresponding author writing the paper.
Jawad Hasnaoui: writing the paper.
Reda Abada: study concept.
Sami Rouadi: study concept.
Mohammed Roubal: correction of the paper.
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References

[1] M.A. Germain, P. Marandas, A.M. Leridant, C. Domenge, M. Julieron, M. Schlumberger, et al., Métastase isolée d’un cancer thyroidien. Mandibulectomie et reconstruction par greffon libre péronéen. Rev. Stomatol. Chir. Maxillofac. 98 (6) (1998) 371–374.
[2] J.D. Osguthorpe, J.R. Bratton, Occult thyroid carcinoma appearing as a single mandibular metastasis, Otolaryngol. Head Neck Surg. 90 (5) (1982) 674–675.
[3] G. Coelina, R. Capone, S. Cappabianca, Mandibular metastasis from papillary thyroid carcinoma. A case report, Tumori 89 (4) (2003) 452–454.
[4] L. Liu, G. Venkataraman, A. Salhadar, Follicular variant of papillary thyroid carcinoma with unusual late metastasis to the mandible and the scapula, Pathol. Int. 57 (5) (2007) 296–298.
[5] M. Okura, Y. Tamaki, S. Fursukawa, T. Matsuya, Bilateral multiple pulmonary metastases in a patient with double advanced cancer of the head and neck, Int. J. Oral Maxillofac. Surg. 32 (4) (2003) 430–432.
[6] R.A. Agha, A.J. Fowler, A. Saetta, I. Barai, S. Rajmohan, D.P. Orgill, the SCARE group, The SCARE statement: consensus-based surgical case report guidelines, Int. J. Surg. 34 (2016) 180–186.
[7] Nikolaos C. Nikitaikis, Angeliki polymeri metastatic papillary thyroid carcinoma to the maxilla: case report and literature review, Head Neck Pathol. 6 (June (2)) (2012) 216–223.
[8] Rajjoti Das, Mahesh Kumar, Jagannath D. Sharma, et al., Papillary thyroid carcinoma presenting with mandibular metastasis: an unusual presentation, Clin. Cancer Investig. J. 3 (2014) 426–428.
[9] D. Tamiolakis, I. Tsamis, V. Thomaidis, M. Lambropoulou, G. Alexiadi, I. Venzielos, et al., Jaw bone metastases: four cases, Acta Dermatoven APA 16 (1) (2007) 21.
[10] A. Hirshberg, P. Leibovich, A. Buchner, Metastatic tumors to the jawbones: analysis of 390 cases, J. Oral Pathol. Med. 23 (8) (2006) 337–341.
[11] M. Algahtani, M. Alqudah, S. Alshehri, A. Binhahmed, G.K. Sandor, Pathologic fracture of the mandible caused by metastatic follicular thyroid carcinoma, J. Can. Dent. Assoc. 75 (6) (2009) 457–460.
[12] F. Gerhards, H.D. Kuffner, W. Wagner, Pathological fractures of the mandible: a review of the etiology and treatment, Int. J. Oral Maxillofac. Surg. 27 (3) (1998) 186–190.
[13] S.B. Ismail, M.T. Abraham, Z.B. Zaini, H.B. Yaacob, R.B. Zain, Metastatic follicular thyroid carcinoma to the mandible: a case report, Cases J. 2 (2009) 5533.
[14] J.A. Wexler, Approach to the thyroid cancer patient with bone metastases, J. Clin. Endocrinol. Metab. 96 (2011) 2296–2307.
[15] C. Durante, N. Haddy, E. Baudin, S. Leboulleux, D. Hartl, J.P. Trasaghi, et al., Long-term outcome of 444 patients with distant metastases from papillary and follicular thyroid carcinoma: benefits and limits of radioiodine therapy, J. Clin. Endocrinol. Metab. 91 (2006) 2892–2899.
[16] A. Stojadinovic, M. Shoup, R.A. Ghossein, A. Nissim, M.F. Brennan, J.P. Shah, A.R. Shaha, The role of operations for distantly metastatic well-differentiated thyroid carcinoma, Surgery 131 (2002) 636–643.
[17] U. Bingol, C. Cinar, Cetinkale, Mandibular metastases of papillary thyroid carcinoma treated by hemimandibulectomy and costochondral rib graft, Plast. Reconstr. Surg. Glob. Open 3 (2) (2015) 305.
[18] Siti Asmat, Arepen Irfan Mohamad, Massive bilateral mandibular metastasis from papillary thyroid carcinoma, J. ejenta.2016;12:004.