Bionature, 40(3) 2020 : 6-13

METASTATIC THYROID CARCINOMA TO THE MANDIBLE MIMICKING A VASCULAR MALFORMATION: CASE REPORT

HACENE HAMMOUD^{1*}, NORA MIMOUNE^{2,3}, ADEL GUIDOUM¹, HAMZA LEKBIR¹, EL HACHEMI KHEMILI¹, ABDELGHANI TIBOUK⁴, AMIRA BOUCHENNA⁵, YOUCEF MALKI⁶ AND RACHID KAIDI³

Department of Oral and Maxillofacial Surgery, Central Hospital of the Army, Algiers, Algeria.
 High National Veterinary School (HNVS), Bab-Ezzouar, Algiers, Algeria.
 Institute of Veterinary Sciences, Laboratory of Biotechnology Related to Animal Breeding (LBRA), University Saad Dahleb, BP: 270, Soumaa Road, Blida, Algeria.
 Department of Pathology, Central Hospital of the Army, Algiers, Algeria.
 Department of Endocrinology, Central Hospital of the Army, Algiers, Algeria.
 Department of Radiology, Central Hospital of the Army, Algiers, Algeria.
 Email: abbas.zrai.olives20@gmail.com

Received: 10 August 2020 Accepted: 14 October 2020 Published: 29 October 2020

Case Report

ABSTRACT

Metastasis to the oral cavity rarely occurs and constitutes 1% of all oral cavity malignancies and have been mostly detected in the jaw bones and especially in the mandible. Thyroid cancers have accounted for up to 6% of the oral cavity metastases, occurring more frequently in follicular variants. Because of the hypervascularity of these tumors, the diagnosis of vascular malformation must be discussed and vascular control should be assured in order to avoid any hemorrhagic incident. In the current work, we present a rare case of a hypervascular mandibular metastasis of thyroid carcinoma of a 65 year old female patient. After examination and confirmation, a decision was taken to resect the metastatic mandibular mass in the same time of a total thyroidectomy. Because of the risk of excessive bleeding of the high flow mandibular lesion, the left external carotid artery was ligated first. The histopathologic study of the surgical specimen revealed a follicular thyroid carcinoma of the thyroid gland with extracapsular extension and vascular invasion, a metastatic follicular thyroid carcinoma to the mandible. The patient received 150 milli-curies dose of radioactive lodine-131 postoperatively. She is on regular follow up and actually disease free. In conclusion, the clinical presentation of a metastatic lesion to the jaw bone, especially to the mandible, can be challenging to a clinician and a consideration for metastatectomy should be made for a secondary tumor from a thyroid primary, given its potential for a durable symptom and disease control and possibly a favorable long term outcome.

 $\label{thm:condition} \textbf{Keywords: Thyroid carcinoma; metastasis; mandible; vascular malformation.}$

INTRODUCTION

Metastasis to the oral cavity rarely occurs and constitutes 1% of all oral cavity malignancies and have been mostly detected in the jaw bones [1–3]. In 25% of cases, oral metastases were found to be the first sign of the metastatic spread and in 23%, it was the first indication of an undiscovered malignancy at a distant site

[4]. Metastasis to the jaw bones mainly occurs in the posterior region of the mandible, ramus, and the condyle, which are rich in red bone marrow [1]. The most common region of primary tumors leading to mandibular metastasis is the lung in men and breast in women [5,6]. Thyroid carcinoma is the most frequently diagnosed endocrine carcinoma [2]. Thyroid cancers have accounted for up to 6% of the oral

BIONATURE: 2020

cavity metastases. occurring more frequently in follicular variants [7]. Because of the bloodstream dissemination of these follicular lesions. variant of thyroid carcinomas are highly prone to cause vascular invasion and may present as hypervascular lesions mimicking arteriovenous malformations [8]. We present a rare case of a metastatic thyroid carcinoma presenting as a hypervascular osteolytic lesion of the mandible.

CASE REPORT

A 65 year old female patient was referred to our department of oral and maxillofacial surgery for evaluation and treatment of a growing painless swelling in the left mandibular region, evolving for 3 months. In her medical records, we noticed

an unexplored goiter evolving for 35 years, high blood pressure and a surgery for a herniated lumbar disc.

Physical examination showed painless, firm diffuse and pulsatile swelling, in the left masseteric region, measuring 60 mmx50 mm, fixed to the mandible, overlain by normal skin, the inferior border of the mandible was palpable, there was no numbness in the territory of the inferior mandibular nerve (V3). A huge mass was visible and palpable in the median region of the neck that extend laterally in the right region, it is mobile, firm, painless and measured 50mm x 70mm, it belongs to the thyroid gland (goiter). Intraoral examination showed a discrete tumefaction with a normal mucosa over it in the retromolar region.



Fig. 1. Swelling in the left masseteric region and median and right side of the neck.

Discrete tumefaction in the left retromolar region

BIONATURE: 2020

CT scan and angiogram of the head and neck region showed a hypervascular osteolytic mass destructing totally the left ramus of the mandible, measuring 60mm x 50mm x 49mm, the main supply to the mass come from the dilated left facial and maxillary artery, it drains into the left anterior jugular vein. A rich vascularity is also noticed in a hypertrophied right thyroid lobe of 56 mm x 59 mm x 69 mm in size. Ultrasonography of the neck showed a solitary nodule in the right lobe of the thyroid that measures 60x30 mm, classified TI-RADS IV (Thyroid Imaging Reporting and Data System) highly suggestive of a malignancy.

No lymphadenopathy was found. Fine needle aspiration cytology of the thyroid nodule reported a follicular thyroid carcinoma. An incisional biopsy of the mandibular lesion was then performed by intraoral approach, histopathologic study of the specimen suggested an adenocarcinoma, but immunohistochemical

analysis showed positivity of Thyroid Transcription Factor TTF1+ and Thyroglobulin TG+ concluding thus to a metastatic follicular thyroid carcinoma. Thoraco-abdominal CT scan concluded to metastatic pulmonary micronodules.

A decision was taken to resect the metastatic mandibular mass in the same time of a total thyroidectomy. Therefore, the patient underwent on 29/10/2019 segmental mandibulectomy of the left ramus and a total thyroidectomy by a low cervical approach. Because of the risk of excessive bleeding of the high flow mandibular lesion, the left external carotid artery was ligated first. The histopathologic study of the surgical specimen revealed a follicular thyroid carcinoma of the thyroid gland with extracapsular extension and vascular invasion, a metastatic follicular thyroid carcinoma to the mandible. The patient received 150 milli-curies dose of radioactive lodine-131 postoperatively. She is on regular follow up and actually disease free.



Fig. 2a. CT scan showing a hypervascular osteolytic lesion of the left ramus and hypertrophic right lobe of the thyroid gland

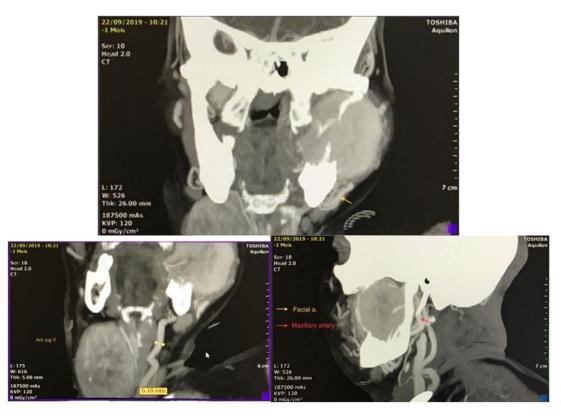


Fig. 2b. CT Angiogram showing facial artery and maxillary artery as feed vessels to the mandibular tumor, anterior jugular vein to be the principle drainage

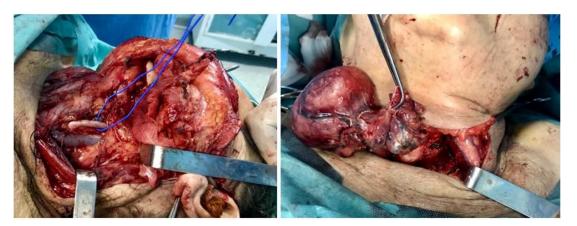


Fig. 3. Operative view with the external carotid artery prepared to be ligated, thyroidectomy

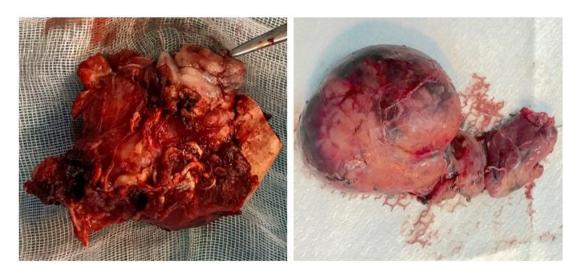


Fig. 4. Surgical specimen of segmental mandibulectomy (left), thyroidectomy (right)



Fig. 5. Postoperative views 2 months later

DISCUSSION

Mandibular metastasis with the thyroid gland as the primary site is a rare event, and the reported incidence is estimated at approximately 1% [1,4,6,8]. In 29% to 33% of cases, the metastatic lesion might be the first indication of an undiscovered primary malignancy at a distant site [8]. A majority of the metastatic jaw tumors have been reported to occur between the 5th and the 7th decades of life [4,6]. The most common sites of metastases of differentiated thyroid carcinoma, in descending frequency, are the spine (34.6%), pelvis (25.5%), thorax (18.3%), extremities (10.2%), shoulder girdle (5.4%), craniomaxillofacial (5.4%) Follicular thyroid carcinoma is the second common thyroid carcinoma. accounts for ~10% of thyroid malignancies and disseminates hematogenously [10]. The preferred sites of metastasis in the mandible are ramus and angle, which are rich in red bone marrow [1,8]. The majority of metastatic tumors to the mandible present with facial swelling and a rapid progression of intraoral or extraoral swelling associated with chin paresthesia and pain is not uncommon [10]. Over 90% of jawbone metastases presented as osteolytic lesion [4]. The histologic appearance of metastatic jaw disease often is poorly differentiated, making it challenging to determine the location of the primary lesion. TTF-1 and TG are good markers of thyroid carcinoma [6]. Because of the hypervascularity of these tumors the diagnosis of arteriovenous malformation must be discussed before any incisional biopsy or a surgical resection. In our case, the incisional biopsy results in a hemorrhage managed by compression. Arteriography, Doppler ultrasound, and CT angiogram are the important diagnostic tools to assess the nature, extent, flow, and feeder vessels of such hypervascular lesions [8].

Because of the potentially lifethreatening hemorrhage that may result when such lesions are removed, their devascularization by selective embolization and/or feeder vessels ligation facilitates and secure the surgical act. In our center, we had no access to a selective embolization, thus an external carotid artery ligation was performed before the tumor resection.

Therefore, the surgeon must be aware of the possibility of a metastatic thyroid carcinoma in a hypervascular mandibular lesion, especially if a thyroid disease history exists. The treatment modalities of oral metastasis from thyroid cancer have been varied from palliative to various combination management of surgical interventions, radioactive iodine ablation, radiotherapy, chemotherapy and hormone therapy [11,12]. Surgical resection under vascular control with clear margins followed by radioactive lodine seems to be the best therapeutic combination [8,11]. Survival analysis suggests that surgical resection of involved craniofacial structures with or without adjuvant treatment is the optimal treatment for FTC metastatic to the facial bones [10, 13, 14, 15].

CONCLUSIONS

In conclusion, the clinical presentation of a metastatic lesion to the jaw bone, especially to the mandible, can be challenging to a clinician and a consideration for metastatectomy should be made for a secondary tumor from a thyroid primary, given its potential for a durable symptom and disease control and possibly a favorable long term outcome [16].

CONSENT

As per international standard or university standard, patients' written consent has been collected and preserved by the author(s).

ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

COMPETING INTERESTS

Authors have declared that no competing interests exist.

REFERENCES

- Irani S. Metastasis to the Jawbones: A review of 453 cases. Journal of International Society of Preventive and Community Dentistry. 2017;8(5): 71–81
- 2. Anil S, Lal PM, Gill DS, Beena VT. Metastasis of thyroid carcinoma to the mandible. Case report. Australian Dental Journal. 1999;44(1):56–7.
- 3. 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. 2016;20(2): 252–5.
- Hirshberg A, Shnaiderman-Shapiro A, Kaplan I, Berger R. Metastatic tumours to the oral cavity -Pathogenesis and analysis of 673 cases. Oral Oncology. 2008;44(8): 743–52.
- Jeon YT, Kim CH, Park SM, Kim MK. Distant metastasis of follicular thyroid carcinoma to the mandible: A rare case report. Journal of the Korean Association of Oral and Maxillofacial Surgeons. 2019;45(5): 294–8.
- D'Silva NJ, Summerlin DJ, Cordell KG, Abdelsayed RA, Tomich CE, Hanks CT, et al. Metastatic tumors in

- the jaws: A retrospective study of 114 cases. Journal of the American Dental Association [Internet]. 2006;137(12): 1667–72.
- Available:http://dx.doi.org/10.14219/jada.archive.2006.0112
- 7. 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 Reports in Dentistry. 2020:2020.
- 3. Kumar RVK, Chakravarthy C, Sekhar GR, Devireddy SK, Kumaravelu C, Kare A. Metastatic thyroid carcinoma presenting as hypervascular lesion of the mandible: A case report and review of literature. Journal of Oral and Maxillofacial Surgery [Internet]. 2010;68(10):2613–6.
 - Available:http://dx.doi.org/10.1016/j.joms.2009.10.004
- Marcela Osorio, Sami P. Moubayed, Henry Su MLU. Systematic review of site distribution of bone metastases in differentiated thyroid cancer. Head and Neck. 2014;36(10): 1391.
- Varadarajan VV, Pace EK, Patel V, Sawhney R, Amdur RJ, Dziegielewski PT. Follicular thyroid carcinoma metastasis to the facial skeleton: A systematic review. BMC Cancer. 2017;17(1):1–8.
- Khoozestani NK, Farzaneh Mosavat, Mohammad Shirkhoda AS. Case report metastatic papillary thyroid carcinoma of the mandible: Case report and literature review. Journal of oral and Maxillofacial Pathology. 2019;23:97–105.
- 12. Ripp GA, Wendth Jr AJ, Vitale P. Metastatic thyroid carcinoma of the mandible mimicking an arteriovenous

BIONATURE: 2020

- malformation. J Oral Surg. 1977;35(9): 743-5.
- 13. Guibert-Tranier F, Piton J, Riche MC, Merland JJ, Caille JM. Vascular malformations of the mandible (*Intraosseous haemangiomas*). The importance of preoperative embolization. A study of 9 cases. Eur J Radiol. 1982;2(4):257-72.
- 14. Kelly DE, Terry BC, Small EW. Arteriovenous malformation of the mandible: Report of case. J Oral Surg. 1977;35(5):387-93.
- Mohammadi H, Said-al-Naief NA, Heffez LB. Arteriovenous malformation of the mandible: Report of a case with a note on the differential diagnosis. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 1997;84(3): 286-9.
- 16. Krishnamurthy A, Deen S, Ramshankar V, Majhi U. Metastatic follicular carcinoma thyroid masquerading as a primary jaw tumor. Journal of Maxillofacial and Oral Surgery. 2016;15(2):266–9.

© Copyright Global Press Hub. All rights reserved.