

CASE REPORT

Mandibular metastasis in a patient with undiscovered synchronous thyroid and prostate cancer: A diagnostic dilemma

Mohammad Zandi, Mohammad Jafari¹, Mona Isapour, Amir Abbas Jafari²

Department of Oral and Maxillofacial Surgery, ¹Department of Pathology, Hamedan University of Medical Sciences, ²Private Practice, Oral and Maxillofacial Radiologist, Hamedan, Iran

Address for correspondence:

Dr. Mohammad Zandi,
Department of Oral and Maxillofacial Surgery,
Hamedan University of Medical Sciences, Shahid
Fahmideh Street - 38698-65178, Hamedan, Iran.
E-mail: zandi88m@yahoo.com

Received: 23.11.2013

Accepted: 08.01.2015

ABSTRACT

Background: Mandibular metastasis as the first manifestation of undiscovered synchronous double primary cancers is extremely rare, so, it is presented in the current study. **Case History:** This study reports a 73-year-old man, complaining of a painful mass in the mandible. After history taking, physical examination and panoramic radiographic evaluation; an incisional biopsy was performed intraorally. Histological studies revealed a metastatic tumor of epithelial origin with nonspecific primary site. During metastatic workup of the patient, it was accidentally found that three organs including prostate, lung and thyroid were involved with cancer. Using immunohistochemical study (prostate-specific antigen (PSA), thyroid transcription factor 1 (TTF-1) and thyroglobulin), a final diagnosis of thyroid carcinoma metastasis to the mandible was established.

Discussion: This case report showed that the practitioners should always maintain a high index of suspicion to the possibility that a patient with an orofacial metastatic lesion may have two or more synchronous primary cancers.

Key words: Mandible, metastasis, synchronous, thyroid carcinoma

INTRODUCTION

Metastasis of malignant tumors to the orofacial region is a rare event. In orofacial region, the mandible is the preferred site, where a majority occurs in the premolar-molar region.^[1] The most common sites for the primary tumors that metastasize to the mandible are breast (25%), lung (13%), prostate (10%), colon (7%), kidney (3%) and thyroid (3%).^[2] Metastatic spread of malignancies to the maxillofacial region occurs more often in patients older than 50 years.^[2,3]

Synchronous occurrence of multiple primary cancers is unusual. According to Travis *et al.*, it accounts for about 16% of all cancer incidences.^[4] Liu *et al.*, reported a prevalence of 7% for multiple malignancies in the Netherlands in 2007.^[5] Mandibular metastasis in a patient with undiscovered synchronous thyroid and prostate malignancy is extremely

rare and arriving to a diagnosis for such cases is a challenge; therefore, it is presented in the current report.

CASE REPORT

In January 2013, a 73-year-old man presented to the Department of Oral and Maxillofacial Surgery, complaining of a painful mass in the mandible on the right side. He had also a history of deep neck pain for 3 months, but it had been disregarded. A review of patient's medical history revealed that he had an acute myocardial infarction 2 years before, which was treated by percutaneous coronary angioplasty. The patient had a 30-year history of cigarette smoking and opium use. Physical examination showed a firm, well-defined swelling measuring about 3 × 3 cm in the body of the mandible on the right side. The skin over the mass was normal in color and texture. No palpable submandibular and cervical lymph node was evident. Intraoral examination showed that the involved mandibular area was edentulous and the mucosa over the mass was intact.

An orthopantomogram radiograph demonstrated an ill-defined radiolucent lesion in the body of the mandible causing pathologic fracture of the involved bone [Figure 1].

The results of laboratory tests were normal with the exception of prostate-specific antigen (PSA) level which was 52 ng/ml.

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10.4103/0973-029X.151347

Intraorally, an incisional biopsy was performed under local anesthesia. Histological sections stained with hematoxylin and eosin revealed a metastatic tumor of epithelial origin; however, it was unclear from which primary site the metastasis occurred. Histologically, the neoplastic cells arranged in glandular patterns had low mitotic index and were composed of round hyperchromatic nuclei and acidophilic cytoplasm [Figure 2].

Because of concern over raised PSA level and high suspicion of prostate malignancy, the patient was referred to a urologist for further evaluation. In pelvic ultrasonography, a significantly enlarged prostate was observed. A biopsy of the prostate was

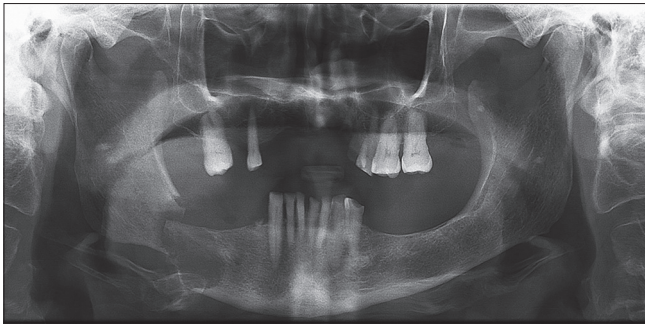


Figure 1: Orthopantomograph shows an ill-defined radiolucent lesion involving the body of the mandible with pathologic fracture

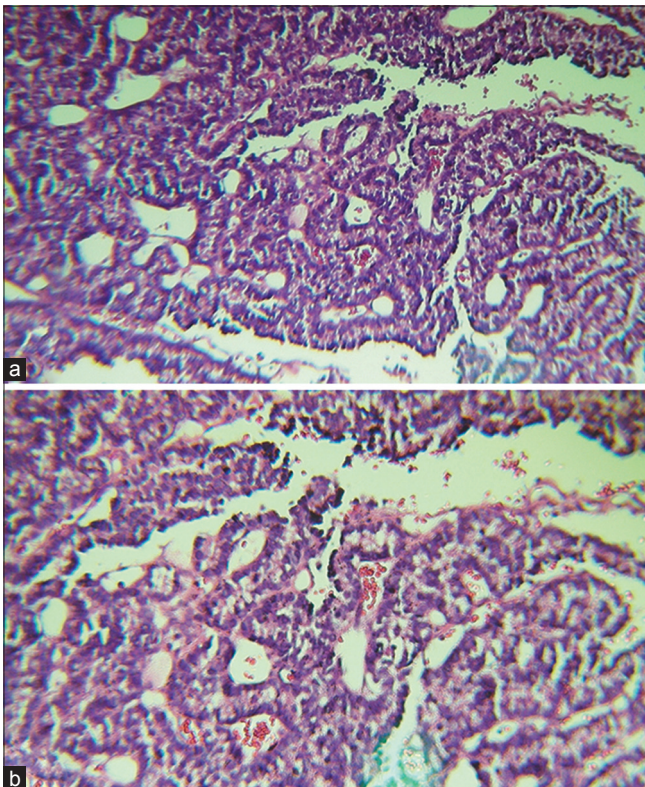


Figure 2: (a) Microscopic view of the biopsied mandibular lesion. Neoplastic cells are composed of round hyperchromatic nuclei and acidophilic cytoplasm, arranged in glandular patterns (H&E stain, x200). (b) High power view of the mandibular lesion showing neoplastic cells composed of round hyperchromatic nuclei and acidophilic cytoplasm, arranged in glandular patterns (H&E stain, x400)

done; the histopathological examination was consistent with prostatic adenocarcinoma. Whole body skeletal survey using bone scintigraphy revealed a bone lesion of the right mandible plus a destructive lesion in the cervical vertebrae. Subsequently, additional metastatic work-up including neck, chest, abdomen and pelvis computed tomography (CT) scan was performed. CT scan of the cervical area revealed a mass, measuring 4 cm in diameter, in the right thyroid lobe causing severe destruction of the adjacent vertebrae and extending into the vertebral canal [Figure 3]. CT scan of the chest showed multiple nodules up to 15 mm in both pulmonary fields. CT scans of the abdomen and pelvis were normal with the exception of an enlarged prostate.

The differential diagnosis for the mandibular lesion included metastatic prostate adenocarcinoma, metastatic thyroid carcinoma and metastatic pulmonary adenocarcinoma. To differentiate between these three lesions, immunohistochemical study with a panel of antibodies including PSA, thyroid transcription factor 1 (TTF-1) and thyroglobulin was performed and showed a positive staining for TTF-1 and thyroglobulin and a negative staining for PSA [Figure 4]. Thus, a final diagnosis of mandibular metastasis of thyroid carcinoma was established. The patient was referred to the oncology unit for further management. Because of the widespread metastases, the treatment was limited to palliation.

DISCUSSION

The incidence of distant metastases in patients with thyroid carcinoma is reported to be 10–15%; the most frequent site is lung, followed by bone.^[6] Metastatic thyroid carcinoma to the jaws is an extremely rare occurrence. It is reported to be about 3.85% of all jaw metastases.^[7] Clausen and Poulsen by reviewing the literature between 1884 and 1961 found 97 cases of jaw metastasis; only six of them were from thyroid.^[8] Of 17 cases of jaw metastasis reported by Meyer and Shklar, only one metastasis originated from thyroid.^[9]

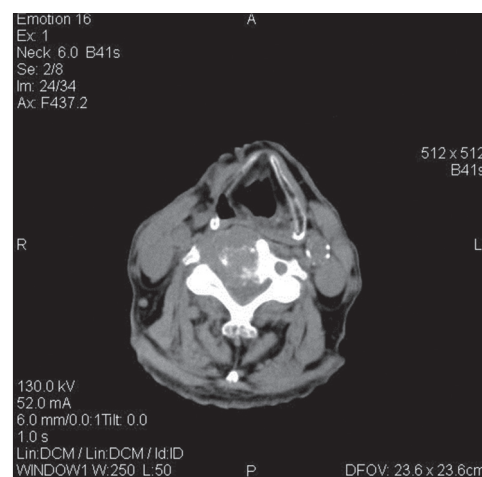


Figure 3: CT scan image of the neck shows a mass in the right thyroid lobe causing severe destruction of the adjacent vertebrae and extending into the vertebral canal. CT = Computed tomography

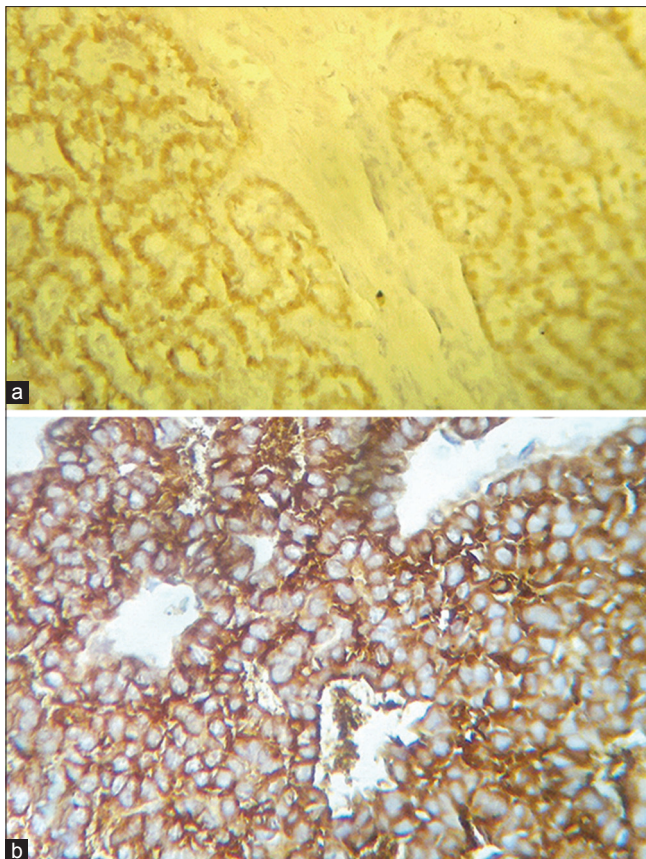


Figure 4: (a) The tumor cells showing positive staining for thyroid transcription factor (TTF) -1. (IHC stain, x400). (b) The tumor cells showing cytoplasmic positivity for thyroglobulin. (IHC stain, x400)

Hirshberg *et al.*, searched the English-language literature between 1992 and 2006 for reports of metastases to the oral cavity and found that 19 of 673 cases of jaw metastasis were from thyroid.^[10] Most cases of reported metastatic thyroid carcinoma to the mandible were female, may be due to the greater preponderance of thyroid carcinoma in women compared with men.^[1,6,9,10,11] The most common clinical features seen were pain, swelling, loosening of teeth, paresthesia and hypervascularity.^[6,12] Kumar *et al.*, suggested that a constant feature of the mandibular metastasis of thyroid carcinoma was its association with blood vessels and its extreme vascular nature; a finding that might lead to profuse hemorrhage during incisional biopsy or surgical ablation of the tumor.^[12] Accordingly, Kahn and McCord reported an excessive blood loss (nearly 800 mL) during excision of a mandibular metastasis of thyroid carcinoma.^[10] Bone metastases of thyroid carcinoma reported in previous studies were mostly osteolytic, although Nishikawa *et al.*, reported an osteoblastic metastatic lesion in the mandible.^[11]

Synchronous occurrence of prostate and thyroid cancer is uncommon and mandibular metastasis as the first manifestation of these primary malignancies is extremely rare. According to the previously published literature, in approximately 20% of cases, metastatic mandibular lesion presents before detection

of the primary tumor.^[13] In these cases, diagnosis of the metastatic lesion would be more difficult compared with cases in which the primary neoplasm has already been discovered.

In the case reported in present study, histopathological findings of the mandibular lesion were nonspecific and did not provide a clue for the diagnosis of the primary tumor. During diagnostic work-up of the patient, we accidentally found that three organs including prostate, lung and thyroid were involved with the cancer. Furthermore, in contrast to previously reported cases of mandibular metastasis of thyroid carcinoma, the patient presented in this study did not have clinical features of a vascular lesion and we did not encounter excessive bleeding during incisional biopsy of the lesion; a finding which was in agreement with Ostrosky *et al.*'s, study.^[14]

To determine the origin of the metastatic mandibular lesion, immunohistochemical study with a panel of antibodies including PSA, TTF-1 and thyroglobulin was performed and showed a positive staining for TTF-1 and thyroglobulin and a negative staining for PSA. Thus, the final diagnosis of metastatic thyroid carcinoma was established. Thyroglobulin staining is proven to be the best means of distinguishing metastasis of thyroid origin from those of nonthyroid origin.^[15]

In conclusion, because of the rare occurrence of metastatic tumors in the orofacial region and their deceiving clinical and histopathological features, their diagnosis is often difficult. When orofacial metastatic lesions are the first manifestation of an undiscovered malignancy at a distant site, arriving to a diagnosis is more challenging. In the present study, an extremely rare case of mandibular metastasis in a patient with undiscovered synchronous thyroid and prostate malignancy was reported. Although this is a rare occurrence, practitioners should always maintain a high index of suspicion to the possibility that a patient with an orofacial metastatic lesion may have two or more synchronous primary cancers. Careful clinical examination in conjunction with radiological, histological and immunohistochemical assessments can lead to early detection of the primary tumor (s), more appropriate treatment and better prognosis for these patients.

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- How to cite this article:** Zandi M, Jafari M, Isapour M, Jafari AA. Mandibular metastasis in a patient with undiscovered synchronous thyroid and prostate cancer: A diagnostic dilemma. *J Oral Maxillofac Pathol* 2014;18:449-52.

Source of Support: Nil. **Conflict of Interest:** None declared.