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Numb chin syndrome as a sign of mandibular metastasis: A case report



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ABSTRACT

INTRODUCTION: Metastasis to the oral cavity can be located in both the soft and bony tissues and comprise only 1% of all oral malignancies; however, it is clinically significant because it indicates widespread metastatic disease and an unfavorable prognosis. A numb chin is an important presentation of oral metastasis, but other dental and systemic pathology may be involved.

PRESENTATION OF CASE: We present the case of a 54-year-old woman who presented with numb chin syndrome 8 years after a diagnosis of primary breast carcinoma. The former was caused by mandibular ramal metastasis of the tumor, which also spread to the spinal canal, ribs, pelvic bones, sacrum, and proximal femur. Because of widespread metastasis, palliative treatment was administered.

DISCUSSION: Despite its low incidence, oral metastasis should be considered as a diagnostic option when patients present with numb chin syndrome. Bony metastasis may produce symptoms late, in contrast to soft tissue metastasis. Pain is the first presenting symptom in most cases, but paresthesia, hypoesthesia and anesthesia in the facial region are possible symptoms too. Most oral bony metastasis are located in the mandible, especially in the molar region. Despite treatment, the average survival after diagnosis of mandibular metastasis is 6–7 months.

CONCLUSION: In most cases with oral metastasis, palliative treatment is indicated. Early detection of oral lesions could improve treatment outcome and survival. A full diagnostic work-up is therefore of great importance.

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1. Introduction

Although rare, metastasis of primary tumors to the oral cavity is important because of their potentially deleterious effects [1]. Not only is it a sign of systemic spread of tumor cells, it can negatively affect oral functioning and esthetics. As in many cases of cancer metastasis, the patient's prognosis is usually very unfavorable.

Bony metastasis is most prevalent in prostate and breast cancers, but also occurs with lung, kidney, liver, and colorectal cancers [1,2]. Most metastatic cancers in the oral cavity are situated in bony structures, while soft tissue metastasis is less common. The mandible is considered more prone to metastasis than the maxilla. In most patients, bony metastasis to the oral cavity is only present in late stage cancer [3].

We report a case from our academic center, where mandibular metastasis presented with pain and hypoesthesia in the right lower face. The work has been reported in line with the CARE criteria [4].

2. Presentation of case

A 54-year-old female patient presented at our department with sudden pain in the right mandible and numbness in the right chin region, for which she was referred by her dentist. In 2008, she was diagnosed with an early breast carcinoma (pT1aN1a ER-positive HER-2 negative) for which she underwent wide excision and axillary lymphadenectomy followed by adjuvant chemo- and radiotherapy (3 × FEC-100 and Taxotere-100). Afterwards, adjuvant letrozole (Femara[®]) and tamoxifen were administered for 2 and 5 years, respectively. In the same year, she was diagnosed with clear cell renal carcinoma (T1a), which was treated successfully with total nephrectomy.

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Fig. 1. Numbness of the right chin region.

In October 2015, lung and osteoclastic bone metastases were detected, which originated from the primary breast tumor. These were treated with monthly zoledronic acid (Zometa[®]) starting in January 2016. The patient also received treatment with exemestane (Aromasin[®]), followed by fulvestrant (Faslodex[®]) after progression on exemestane.

In June 2016, she suddenly complained of pain in the right lower jaw combined with numbness in the right chin. The numbness did not involve the lower lip (Fig. 1). No adenopathies were palpable in the head and neck region.

Intra-oral investigation revealed a bony elevation, which was palpable in the region of the right first and second premolar. The lower right second molar was sensitive upon percussion, but sensitivity testing indicated intact sensory innervation of the teeth.

The orthopantomogram (OPG) showed no significant abnormalities, so it was supplemented with a CBCT and a PET-CT (Fig. 2). The latter showed bony radiolucent areas indicating osteoclastic metastases. Supplementary skeletal scintigraphy indicated diffuse bony metastasis in the spinal canal, ribs, pelvic bones, sacrum, proximal femur, and right mandibular ramus (Fig. 3).

To exclude chemotherapy-induced neuropathy in the trigeminal ganglion, a magnetic resonance imaging (MRI) investigation was done. The images showed a small lesion in the right cavernous sinus that was indicative of a hemangioma or leiomyoma, with no clinically significant role in the mental neuropathy.

A biopsy of the lesion was obtained under general anesthesia using a unilateral bilateral sagittal split osteotomy-type approach. The biopsy was taken caudally and anterior to the Spix spine. The anatomopathologic investigation of the biopsy indicated the presence of breast metastasis, which were estrogen and progesterone receptor-positive and HER-2 receptor-negative (Fig. 4a and b).

After the biopsy wound healed, the pain in the right mandibular region diminished and eventually disappeared 3 weeks later. However, the numbness in the right chin-region persisted.

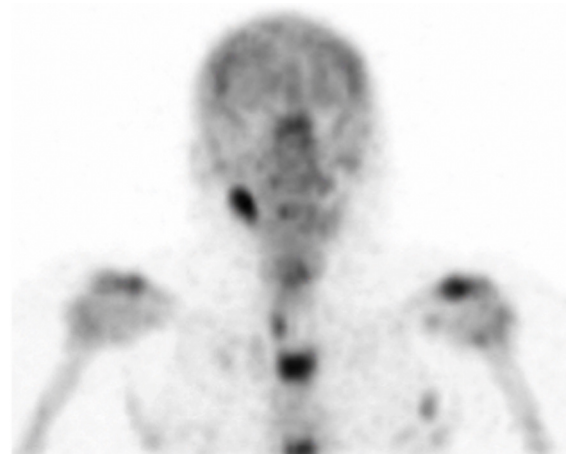


Fig. 3. Skeletal scintigraphy indicates a metabolic active lesion, consistent with ramal mandibular metastasis.

Because of the lack of pain and the presence of multiple bony metastases, no radiotherapy was administered in the region of the right mandible. Palliative systemic therapy with fulvestrant 500 mg and zoledronic acid every 4 weeks is continued. Radiotherapy is considered useful only in the case of pain. Surgery has got no advantage in the present circumstances.

3. Discussion

Numb chin syndrome, also called “mental neuropathy”, is best defined as a mostly unilateral sensory disturbance (hypo- or paresthesia) of the lower lip, chin, and/or occasionally the gingival mucosa [5].

Because only 1% of all oral neoplasms are metastases [6,7], this is not the primary diagnosis to consider. On the other hand, metastatic disease in the mandible and maxilla is probably more frequent than reported as indicated by histologic sections [8].

No malignancies are known to spread only to the oral cavity [7]. Moreover, tumors rarely metastasize only to bone [9]. Thorough investigations should rule out accompanying soft tissue metastasis. According to a systemic review by Galan Gil et al., breast cancer (40.4%), lymphomas (20.5%), prostate cancer (6.6%), and leukemia (5.1%) are the most common primary malignancies causing mental nerve neuropathy [10]. In 75% of all cases, the oral metastasis is not the first sign of metastatic spread of a malignancy. Bony metastasis may only be evident later in contrast to soft tissue metastasis [11].

Metastasis to the jawbone occurs twice as often as to the oral soft tissues [11]. Most oral bony metastases are located in the mandible. The molar region of the mandible is most frequently involved followed by the premolar area and the ramus-angle region [2]. The

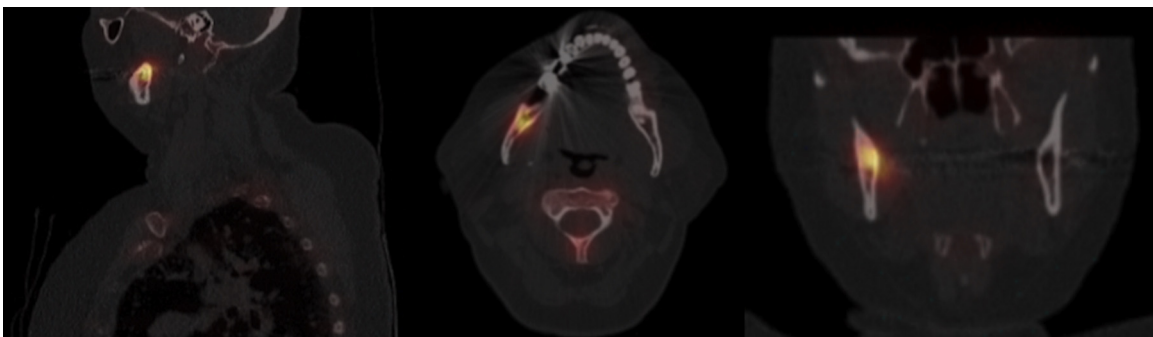


Fig. 2. PET-CT shows high metabolic activity in the right ramal region.

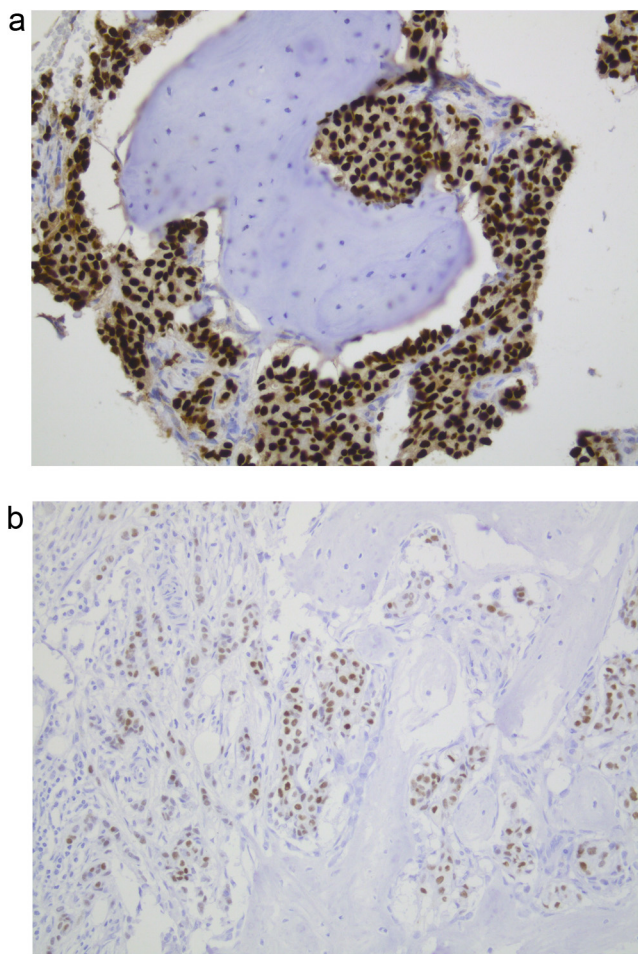


Fig. 4. (a) The epithelioid cells in the marrow spaces are strongly positive for GATA3, a marker of breast carcinoma (magnification 200 \times). (b) The epithelioid cells in the marrow space were also positive for the estrogen receptor (magnification 200 \times).

presence of hematopoietic bone marrow and the reduced velocity of the blood flow in that area could explain this [12]. The tortuous course of the inferior alveolar nerve could predestine metastatic deposits on its course [5].

In case of pain, paresthesia, hypoesthesia or anesthesia in the facial region, one should always rule out the possibility of metastasis or primary cancer, especially in patients with an oncologic history. Swelling, bleeding, paresis, facial disfiguration, pathologic fractures, tooth mobility, and restricted mouth opening can also be symptoms of a malignancy [13].

Our patient experienced pain and local numbness accompanied by a bony swelling.

In most cases, significant bony swelling and tenderness over the affected area are present upon physical examination. Pain is the most frequently reported symptom [11]. Pathologic fractures and pain are associated with a poor prognosis [14].

Because of the rarity of oral metastasis, the differential diagnosis includes inflammatory and reactive lesions, a primary oral tumor, dental or facial trauma, osteomyelitis, diabetic neuropathy, temporal arteritis, Lyme disease, sarcoidosis, multiple sclerosis, sickle cell disease, acute leukemia, lymphoma, multiple myeloma, melanoma, Ewing sarcoma, and Sjögren syndrome [5,15]. A numb chin may also be caused by leptomeningeal metastasis, presenting with subtle symptoms like areflexia, and by skull-base metastases, frequently associated with accompanying cranial neuropathies of the sixth and seventh cranial nerve [16]. Because our patient was treated previously with chemotherapy, radio-

therapy, and zoledronic acid, other valid diagnostic options were chemotherapy-induced neuropathy of the trigeminal ganglion and bisphosphonate-related osteonecrosis of the jaw (BRONJ). Both were ruled out by thorough investigation.

The initial bony lesion was not visible on the primary radiographic imaging (orthopantomogram). This lack of radiographic evidence is present in 5% of all cases, while in 90% of cases there is evidence of osteolytic “mottled” lesions [11]. Sclerotic changes may also be seen in breast metastases. Because microscopic seeding in the nerve is often not visible on radiographs, further investigation with positron-emission tomography-computed tomography (PET-CT) is advised when standard radiography (OPG) is negative. PET-CT enables early detection and could improve the treatment outcome [2].

Concerning the treatment options, surgical resection is considered only useful when the oral metastasis is solitary [11]. A palliative approach is indicated in most other cases where systemic presence of metastasis rules out curative treatment options. Optimal oral functionality and pain control are the two pillars of palliative treatment. The latter includes analgesic medication, radiotherapy, chemotherapy, and sometimes surgical excision [1]. In our patient, the only complaints were pain and numbness of the chin, but after biopsy only numbness of the chin remained. Systemic endocrine therapy and bisphosphonate therapy were given to postpone progression of the symptoms. As such, this treatment was aimed at preserving oral functionality for as long as possible in the absence of pain.

Because oral metastasis is an indicator of widespread disease, the average survival after diagnosis is 6–7 months in cases of mandibular metastasis [10], ranging from 1 to 60 months after diagnosis [7].

4. Conclusion

When patients present with a numb chin, one should always rule out the possibility of metastasis if there is an oncologic history and a full diagnostic work-up is mandatory. This comprises radiographic imaging, CT, PET, MRI, and biopsies. Because it is a sign of systemic spread or recurrence of malignancies, the treatment is palliative in most cases. Only solitary metastasis to the oral cavity might be (temporarily) curable by means of surgical excision and/or chemoradiotherapy.

Dentists, general practitioners, maxillofacial surgeons, and oncologists should be aware of the numb chin syndrome as a manifestation of malignancy.

Conflicts of interest

None.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Case report – written consent was obtained from the patient.

Author contribution

All authors have individual and substantial contribution to the article.

Aerden T.: data collection, writing the paper.

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