Case Report

Gingival metastasis of adenocarcinoma with an unknown occult primary

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Abstract

Metastatic dissemination to the oral cavity is extremely rare and constitutes about 1%–2.1% of all oral malignancies. The very first sign of the metastasis into the oral cavity indicates an occult malignancy in the distant site. It gives the evidence of widespread disease with an average survival rate of less than 7 months. Adenocarcinomas account for up to 60% of all metastatic neoplasms of unknown primary origin. Here, we report a case of metastatic adenocarcinoma of unknown primary origin in a 60-year-old male patient in the upper and lower gingiva without involvement of the underlying bone which is a very rare case reported in the literature till date.

Keywords: Adenocarcinoma, malignancy, metastasis, oral cavity

Case Report

A 60-year-old male patient reported with swelling and pain in the upper and lower gums in the left back tooth region. On intraoral examination, a growth was seen on the left upper front gingiva which was round in shape, measuring about 2 cm in its greatest dimension, extending from mesial marginal gingiva i rt 22 to the distal marginal gingiva i rt 24 anteroposteriorly. Borders were diffuse up to 1 cm superiorly over attached gingiva. A similar pedunculated growth was seen on the left lower gingiva, measuring about 4 × 1 cm in its greatest dimension, extending from marginal gingiva i rt 33–37, with diffuse borders [Figure 1]. A preliminary differential diagnosis was formulated, based on the patient’s clinical signs and symptoms and radiographic findings. The possible diagnoses included peripheral giant cell granuloma. Both the lesions were excised and sent for histopathological examination which showed the features of metastatic adenocarcinoma [Figures 2-4].

The tumor configuration was compatible with adenocarcinoma; this morphology is not commonly seen in tumors of oral cavity, including salivary gland tumors, which are known for their diverse morphological and histopathological features. For this reason, it was thought the tumor was primarily metastatic. As the patient did not come for follow-up, further investigations to rule out primary tumor, it was diagnosed as “metastatic gingival adenocarcinoma of unknown primary origin.”

Discussion

Metastasis is the capacity of cancer cells to spread and grow at site distant from the origin of the neoplasm. These metastatic deposits are also referred to as secondary cancer or secondaries.[1] Metastasis to the oral cavity is a rare event and the uncommon site for metastatic tumor cell colonization and is usually evidence of a widespread disease.[2] The jaws and oral cavity are rare sites for metastatic dissemination with only about 1% of oral malignancies attributed to metastases and only 0.1% confined to oral soft tissues.[3]

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These oral metastatic tumors are found to be the first sign of metastatic spread in 25% of cases, and in 23% of cases they are an indication for unidentified primary malignancy of a distant site. Oral soft tissues were less frequently affected than the jaw bones (1:2.5). In 2008, Hirshberg et al. reviewed 673 cases of oral metastasis, of which 112 cases were metastasized from the lung, 58 were noted in the jawbones, and 54 in the oral mucosa.

Jaws are more commonly involved, particularly in premolar–molar area, the possible reason being active hematopoietic marrow. Whereas in oral soft tissues, gingival site is more commonly involved being about 54%, followed by alveolar mucosa (50%) and tongue (30%). The common sources of metastasis to the oral cavity are lung, liver, colon and kidney, prostate in males, and breast and genital organs in females.

Adenocarcinomas account for up to 60% of all metastatic neoplasms of unknown primary origin. According to a study by Irani S (carried out from 1937 to 2015), there were 412 metastatic lesions developed in the oral soft tissues. The greater part of the histological examination yielded adenocarcinoma ($n = 93; 22.5\%$).

Adenocarcinoma is a type of cancerous tumor that can occur in several parts of the body. It is defined as neoplasia of epithelial tissue that has glandular origin, glandular characteristics, or both. Adenocarcinomas are part of the larger groups of carcinomas. Some metastatic adenocarcinomas have distinctive histological features that allow for their site determination (e.g. colonic adenocarcinoma, bronchioloalveolar cell carcinoma). In this report, the histopathological features were in favor of adenocarcinoma of lung.

Although the jawbones and their adjacent gingiva share a common blood supply through maxillary artery, there are two patterns in the metastasis to the gingiva: the localized or secondary invasion.
from the jawbones. The common routes of metastasis by distant tumors to the oral region and/or the jawbones are through lymphatics or by hematogenous spread. One potential route of spread from the primary site to the oral tissues is through Baston's vertebral veins. The mechanism of localized gingival metastasis from the primary site, sparing the bone as in this case, is still unclear. Very few cases of distant metastasis to gingiva without involvement of underlying bone have been reported.

Regarding gingival metastasis from malignancies at a distant site, it has been reported that the tumor cells from primary sites tend to metastasize to the sites presenting the inflammation. This seems to be the reason that the gingiva is the most frequent site for metastases in oral soft tissues. In most people, a certain degree of gingival inflammation exists due to chronic periodontitis. The capillary vessels in gingiva may have chronic inflammation and the gingiva constantly proliferate and eventually develop fragmented basement membranes in the immature capillaries. These capillary vessels become more leaky at the basement membrane level making them more penetrable for tumor cells than mature cells. Metastasis consists of five steps which are collectively termed the metastatic cascade [Figure 5].

The clinical presentation of metastatic gingival lesions differs from primary gingival cancer. Symptoms such as painful swelling, bleeding, increasing tooth mobility, delayed healing of extraction socket, pathological fracture, masticatory difficulties, trismus, dysphagia, and dyspnoea are indications of malignancy. However, the metastatic lesion resembles benign inflammatory lesions such as hyperplasia, pyogenic granuloma, hemangioma, giant cell granuloma, peripheral fibroma, or fibrous epulis.

The diagnosis was solely based on the histopathological pattern of the tumor. The histopathological features revealed by routine hematoxylin–eosin staining showed features of metastatic adenocarcinoma. As the primary site was not known, a metastatic adenocarcinoma of primary unknown origin is considered.

The prognosis and treatment depends on the site of primary tumor and degree of metastatic spread. Most of the patients who present with a metastatic tumour in the oral cavity have also developed metastases at other sites, so a palliative regimen is the only management option. Local treatment of jaw bone metastases nearly always by radiotherapy usually relieves pain and may prevent loss of function. Metastases in the soft oral tissues may more readily be approached surgically, with similar palliative results for the patient.

**Conclusion**

Metastasis from internal neoplasms should be considered among other differential diagnosis in the unusual; it should be recognized by dentists that gingival masses similar to benign or inflammatory lesions may represent an initial sign of occult malignancies.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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**Conflicts of interest**

There are no conflicts of interest.

**References**

1. McGee J, Isaacson PG, Wright NA. Oxford Textbook of Pathology. Vol. 1. Oxford: Oxford University; 1991.
2. Watanabe E, Touge H, Tokuyasu H, Kawasaki Y. Gingival metastasis of Adenocarcinoma from the lung. Respir Med CME 2008;1:103-6.
3. Mallikarjun JH, Mujib BR, Naik R, Patil ST. Metastatic small cell carcinoma of the cervix to the oral cavity: A rare case report and an insight into pathogenesis of metastasis. J Oral Maxillofac Pathol 2015;19:247-50.
4. Beena VT, Panda S, Heera R, Rajeev R. Multiple metastatic tumors in the oral cavity. J Oral Maxillofac Pathol 2011;15:214-8.
5. Rajini Kanth M, Ravi Prakash A, Raghavendra Reddy Y, Sonia Bai JK, Ravindra Babu M. Metastasis of lung adenocarcinoma to the gingiva: A rare case report. Iran J Med Sci 2015;40:287-91.
6. Hammar SP. Metastatic adenocarcinoma of unknown primary origin. Hum Pathol 1998;29:1393-402.
7. Irani S. Metastasis to the oral soft tissues: A review of 412 cases. J Int Soc Prevent Communit Dent 2016;6:393-401.
8. World Cancer Report 2014. Ch. 5.3. World Health Organization; 2014. ISBN 978-92-832-0429-9. Available
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from: https://en.wikipedia.org/wiki/Adenocarcinoma.

9. Kuttan NA, Flemming DK, Dane JN, Ang DB. Metastatic lesion of the anterior mandible with an occult primary: A case report. Spec Care Dentist 2006;26:76-80.

10. Miyake M, Takebayashi R, Ohbayashi Y, Kushida Y, Matsui Y. Metastasis in the gingiva from colon adenocarcinoma J. Maxillofac Oral Surg 2015;14 (Suppl. 1):S279-282.

11. Dhawad MS, Nimonkar PV. Metastatic carcinoma of gingiva mimicking pyogenic granuloma. J Maxillofac Oral Surg 2011;10:163-5.

12. van der Waal RI, Buter J, van der Waal I. Oral metastases: report of 24 cases. Br J Oral Maxillofac Surg 2003;41:3-6.