

Case Report

Unusual maxillofacial soft tissue metastasis of rectal adenocarcinoma: a case report

Qian Zhao^{1*}, Jianan Miao^{1*}, Linfeng Li¹, Shuyue Zhang², Haixia Miao², Li Ma³, Jian Li⁴

¹North China University of Science and Technology, Tangshan, China; ²Department of Stomatology, Tangshan People's Hospital, Tangshan, China; ³Department of Stomatology, Tangshan Vocational and Technical College, Tangshan, China; ⁴Department of Stomatology, Xiang'an Hospital of Xiamen University, Xiamen, China. *Equal contributors.

Received December 1, 2017; Accepted May 14, 2018; Epub June 15, 2018; Published June 30, 2018

Abstract: The most frequent sites of metastases of rectal adenocarcinoma are liver and lung. Maxillofacial soft tissue metastases are extremely uncommon in both genders. Histopathological and immunohistochemical characteristic performance can help us to make the correct diagnosis. The prognosis of maxillofacial metastases is poor, but effective treatments are necessary to avoid the patient discomfort. Here we report a 68-year-old male, who was diagnosed with rectal adenocarcinoma five years ago and presented with a mass on the right side of his face. Based on histopathology and immunohistochemistry, we confirmed it as a metastasis of rectal adenocarcinoma. Local palliative radiation was given to control the symptoms.

Keywords: Rectal adenocarcinoma, maxillofacial metastasis, soft tissue, immunohistochemistry

Introduction

Colorectal cancer is one of the most common cancers in the world. It is the third most common cancers in men and the second in women [1]. The incidence rate of colorectal cancer has shown gender and regional differences throughout the world [1, 2]. Approximately 15-25% of all colorectal cancer patients have distant metastases [3]. The most frequent sites of the metastases are the liver (in 30% to 60% of cases) and the lung (in 20% to 30% of the cases) [4]. However, soft tissue metastasizes to the oral maxillofacial of rectal cancer is extremely rare. In this article, we reported a case that unusual maxillofacial soft tissue metastasis was spread from rectal adenocarcinoma.

Case report

A 68-year-old male patient had a chief complaint of a mass on his right face which was progressively enlarging for 2 months. He was diagnosed with rectal adenocarcinoma and initially treated with surgery in March 2012. His post-

operative period was uneventful until 2016, when he went to our hospital for regular checking, multiple lung metastases was found (**Figure 1**). He was treated with chemotherapy. One year later, he presented with a mass on the right side of his face.

Physical examination revealed that the mass upwards reached the orbit and downwards to the upper lip (30×25×20 mm). The dividing line was not clear all round the mass. Additionally, there was no ulceration and no bleeding on the surface of it. Due to compression by the mass, his face was asymmetrical (**Figure 2**). A magnetic resonance imaging (MRI) study of the head presented a soft-tissue mass in the right cheek. It showed a heterogeneous mass with signal intensity on T2-weighted images (T2WI) (**Figure 3**). Under general anesthesia, the mass was excised (**Figure 4**) and sent to the Pathology Department. Histopathological examination revealed hyperplastic, irregular glands which were lined by columnar cells. The nuclei of the cells were pleomorphic and disorganized (**Figure 5**). These features were similar to the primary rectal adenocarcinoma. Considering the

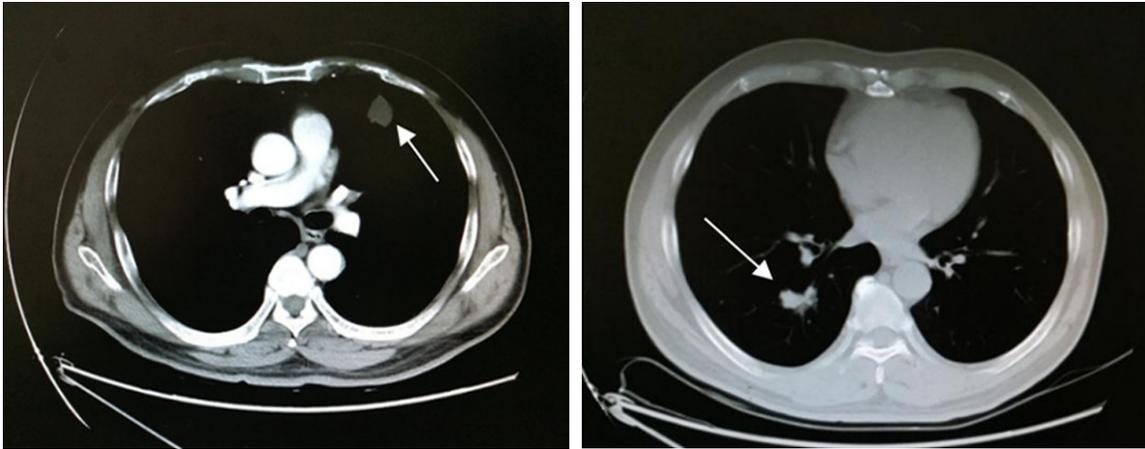


Figure 1. MRI image of the multiple lung metastasis of rectal adenocarcinoma.



Figure 2. Metastasis tumor in the soft tissue of the right face.

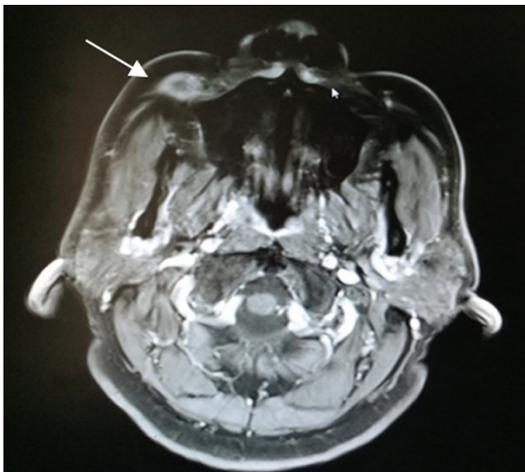


Figure 3. MRI image of the soft-tissue metastasis of rectal adenocarcinoma.

history of rectal cancer, immunohistochemistry was used to verify if the tumor was metastatic.

Immunohistochemical results showed that the tumor cells were positive for CDX-2 (**Figure 6**) and CK20 (**Figure 7**) which confirmed the diagnosis. Local palliative radiation was given to control the symptoms of his right face. Until now, the patient is still alive and without recurrence. Continued follow-up the patient is ongoing.

Discussion

Metastatic tumors to the soft tissue are uncommon. The prevalence of soft tissue metastases varies in autopsy series from 6 to 17.5% [5], nevertheless, oral maxillofacial soft tissue metastases are exclusively infrequent, which comprises approximately 1% of all oral malignancies. Breast cancer and lung cancer comprise the most common malignancies with oral maxillofacial metastasis.

Colorectal cancer is one of the most common cancers with more than one million new cases of colorectal cancer each year worldwide [6]. Despite the prevalence of colorectal cancer, oral maxillofacial metastases are extremely rare. Jawbones are usually involved in most cases according to the literature [7]. Several articles have presented rectal cancer that metastasized to the mandible [8], to the hard palate [9], to the gingiva [10, 11]. However, metastasis to the maxillofacial soft tissue has been rarely reported.

It is difficult to distinguish metastatic malignant tumors from primary maxillofacial tumors. Swelling, pain, and bleeding are the common symptoms. There were no significant differenc-

Maxillofacial soft tissue metastasis of rectal adenocarcinoma



Figure 4. Removed tumor from the patient (30×25×20 mm).

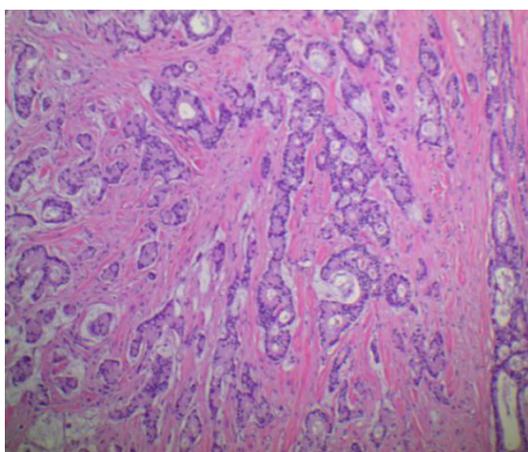


Figure 5. Hyperplastic, irregular glands lined by columnar cells (H&E, ×100).

es between the lesion and other primary maxillofacial tumors in regarding clinical features in this case. So it was easy to be misdiagnosed. Doctors should combine disease history with histopathological results. Immunohistochemical technology, which is important to define the site of primary lesion, can be used to help diagnose if necessary. Jung [12] reported that CK7 (negative)/CK20 (diffuse positive)/CDX2 (positive)/MUC2 (positive) immunophenotype can effectively suggest the rectal cancer. In this case, resection of the tumor was done under

local anesthesia, and immunohistochemical result showed the tumor cells is positive for CDX-2 and CK20, which confirmed the diagnosis.

The main problem for colon cancer patients is development of distant metastasis that lead to limited survival. Unfortunately, patients with rectal tumors that metastasize to the oral maxillofacial usually have poor prognosis, as most cases reported, leading to death in less than 1 year after detection of maxillofacial involvement. In general, tumor metastasis is a complex process and the mechanisms of colorectal metastases are not clear. In this case, in addition to the maxillofacial metastasis,

the patient had a lung metastasis that we speculated was associated with intravascular dissemination.

Treatment options to limit tumor development include palliative therapy, radiotherapy, chemotherapy, and excision. The options are chosen primarily based on the extent of metastatic spread. Local control remains important in the treatment of metastatic rectal cancer. Excision might have a role in palliating the pain. However, if there is evidence of wide dissemination, excision is impossible. Conservative management is recommended to improvement in the prognosis. While, no matter which options were chosen, the prognosis of maxillofacial soft tissue metastases remains poor. In this case, we performed local surgical resection and suggested the patient to receive the palliative radiation. The ultimate goal of our treatment is to control the patient's pain and to restore facial appearance. Fortunately, the patient has had no recurrence and is still alive and follow-up continues.

In conclusion, rectal cancer metastasizes to maxillofacial soft tissue is extremely rare. Such tumors usually portend a poor prognosis. The clinical presentation of the tumor can be deceiving, leading to a misdiagnosis. This case report highlights the importance of pathological examination and disease history inquiry.

Maxillofacial soft tissue metastasis of rectal adenocarcinoma

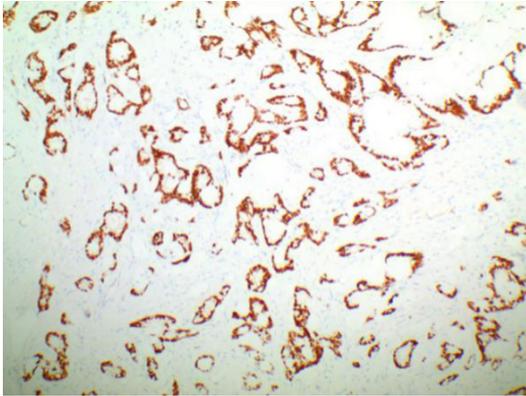


Figure 6. CDX-2 positivity in the tumor cells ($\times 100$).

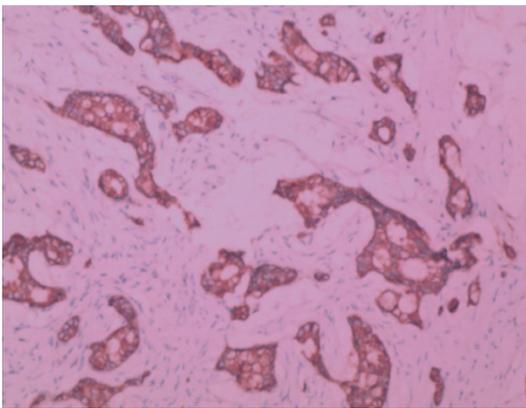


Figure 7. CK20 diffuse positivity in the tumor cells ($\times 100$).

Clinicians and pathologists are encouraged to step up collaboration to make a proper diagnosis.

Disclosure of conflict of interest

None.

Address correspondence to: Jian Li, Department of Stomatology, Xiang'an Hospital of Xiamen University, Xiamen 361000, China. Tel: +86 13933468767; Fax: 05922188602; E-mail: lijianimplant@163.com

References

[1] Jemal A, Bray F, Center M, Ferlay J, Ward E, Forman D. Global cancer statistics. *CA Cancer J Clin* 2011; 61: 69-90.

- [2] Hendifar A, Yang D, Lenz F, Lurje G, Pohl A, Lenz C, Ning Y, Zhang W, Lenz H. Gender disparities in metastatic colorectal cancer survival. *Clin Cancer Res* 2009; 15: 6391-7.
- [3] van der Geest LG, Lam-Boer J, Koopman M, Verhoef C, Elferink MA, de Wilt JH. Nationwide trends in incidence, treatment and survival of colorectal cancer patients with synchronous metastases. *van der Geest LG* 2015; 32: 457-65.
- [4] Donadon M, Ribero D, Morrisstiff G, Abdalla EK, Vauthey JN. New paradigm in the management of liver-only metastases from colorectal cancer. *Gastrointest Cancer Res* 2007; 1: 20-7.
- [5] Riahi H, Ladeb MF, Bouaziz MC, Mechri M. Soft Tissue Metastases. *Imaging of Soft Tissue Tumors*. In: Filip M, Vanhoenacker, Paul M, Parizel, Jan L Gielen, Editors. Belgium: Springer, Cham; 2017. pp. 593-601.
- [6] Pearlman M, Kwong WT. A long and distant journey: a case of rectal cancer with metastasis to the orbit. *Ann Gastroenterol* 2015; 28: 151-152.
- [7] Siriwardena BSMS, Tilakaratne WM, Jayasooriya PR, Ratnayake P. Metastatic tumours of the oral and maxillofacial region: a retrospective analysis of 18 cases. 2013; 1: 001-004.
- [8] Walsh A. Bone metastasis to the mandible in rectal cancer and a review of the literature. *Journal of Pain Management* 2015; 8: 259-270.
- [9] Unlüoğlu S, Bayol Ü, Küçük Ü, Çukurova İ, Karaman K, Yildirim Z. Metastasis of rectal carcinoma to hard palate: an unusual presentation. *European Journal of Surgical Sciences* 2014; 5: 81-84.
- [10] Almangush A, Asikainen A, Ristimäki A, Haglund C, Hagström J. Oral metastasis from rectal adenocarcinoma: case report. *Case Reports in Clinical Pathology* 2014; 1.
- [11] Lagha A, Chraiet N, Krimi S, Ayadi M, Rifi H, Raies H, Mezlini A. Gingival metastasis from rectal cancer. *International Journal of Case Reports & Images* 2012; 3: 24-26.
- [12] Shin JH, Bae JH, Lee A, Jung CK, Yim HW, Park JS, Lee KY. CK7, CK20, CDX2 and MUC2 immunohistochemical staining used to distinguish metastatic colorectal carcinoma involving ovary from primary ovarian mucinous adenocarcinoma. *Jpn J Clin Oncol* 2010; 40: 208-13.