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Case Report

Intraoral Buccal Mass as the Manifestation of Testicular Choriocarcinoma – A Case Report

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Abstract

Metastatic tumors to the oral cavity are extremely rare lesion that represent 1% of all oral and maxillofacial malignancies. It is invariably associated with a widespread disease and a poor prognosis. Such metastasis occur mostly in the jaw bones than the soft tissues of the mouth. Metastasis to the oral soft tissues most prevalently affects the gingiva and alveolar mucosa. Gingival metastasis may have an unremarkable clinical appearance and they can be difficult to distinguish from more common hyperplasic or reactive lesions that appear to be benign entities, such as peripheral giant-cell granuloma, pyogenic granuloma. We present an unusual case of a testicular choriocarcinoma metastasized to the buccal mucosa mimicking a reactive lesion. An 18 year old male patient presented with a right sided intraoral buccal mass for one month in the department of oral and maxillofacial surgery department of NICRH. Clinically it was diagnosed as pyogenic granuloma. In addition there was right sided non-tender testicular swelling which he noticed eight months earlier. His beta HCG level was 22,5000 mIU/ml and alfa Fetoprotein level was 2.23 ng/ml. Ultrasonography shows right testicular mass, para-aortic lymphadenopathy and multiple hepatic SOL. CT scan shows soft tissue mass in the right hemimandible area. Excision of the right sided intraoral buccal mass and right sided hemimandibulectomy was done. Histopathological examination reveals clear cytotrophoblastic cells with central nuclei and syncytiotrophoblastic cells with multiple dark nuclei embedded in eosinophilic cytoplasm in a hemorrhagic and necrotic stroma and diagnosed as testicular choriocarcinoma. The primary lesion must be removed by orchiectomy. In patients with metastatic illness, the disease must be controlled with cytotoxic chemotherapy, which results in complete regression of the metastasis in 60% to 90% of patients.

Keywords: *Choriocarcinoma; Oral Metastasis; Testicular Choriocarcinoma; Germ Cell Tumors*

Introduction

Choriocarcinoma is a highly malignant uncommon tumor of the testis, and it usually occurs as a component of mixed germ cell tumor [1]. Choriocarcinoma account for about 5% of testicular tumors. These neoplasms are often small, with no enlargement of the testis. Metastatic tumors to the oral cavity are extremely rare lesions that represent 1% of all oral and maxillofacial malignancies [2]. Metastasis to the oral soft tissues most prevalently affects the gingival and alveolar mucosa, followed by the tongue and, less frequently, the tonsil, palate, lip, buccal mucosa, and floor of the mouth [3]. Gingival metastasis may have an unremarkable clinical appearance and they can be difficult to distinguish from more common hyperplastic or reactive lesions that appear to be benign entities, such as peripheral giant cell granuloma, pyogenic granuloma and peripheral ossifying fibroma [4].

The oral metastasis from a testicular cancer is even a more rare entity [5]. There is often marked elevation of the serum level of human chorionic gonadotropin (hCG) [6].

We present an unusual case of a testicular choriocarcinoma metastasized to the buccal mucosa mimicking a reactive lesion.

Case Report

An 18 –year old boy presented with a right sided intraoral buccal mass for one month in the department of oral and maxillofacial surgery department of National Institute of Cancer Research & Hospital (NICRH), Bangladesh in November, 2013. Clinically it was diagnosed as pyogenic granuloma. In addition there was right sided non tender testicular swelling which he noticed 8 months earlier. But he concealed this history because of sighness. That's why his external genitalia were not examined during his first visit. The laboratory findings showed an elevated level of beta-hCG of 225000 mIU/mL (reference range: 0 - 5 mIU/mL, alpha –fetoprotein (AFP) were 2.23 ng/ml (reference range upto 12.5 ng/ml), lactate dehydrogenase (LDH) 3007 U/L (reference range: 225 – 480 U/L).

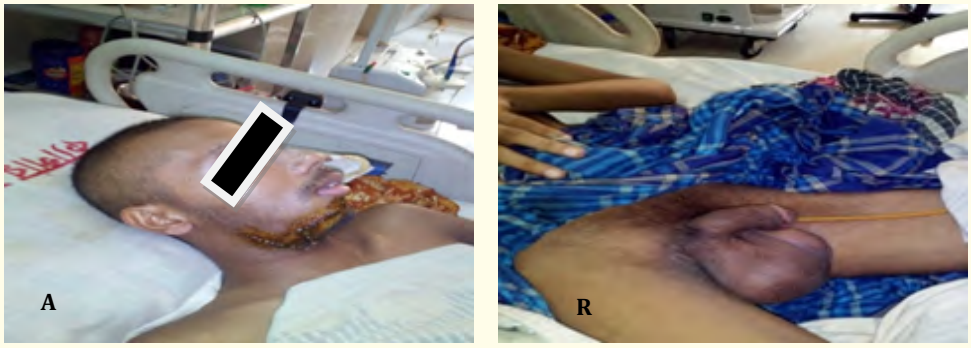

Figure1: *(A) Large soft tissue mass involving right sided buccal mucosa (post operative state) (B) Right sided testicular tumor.*

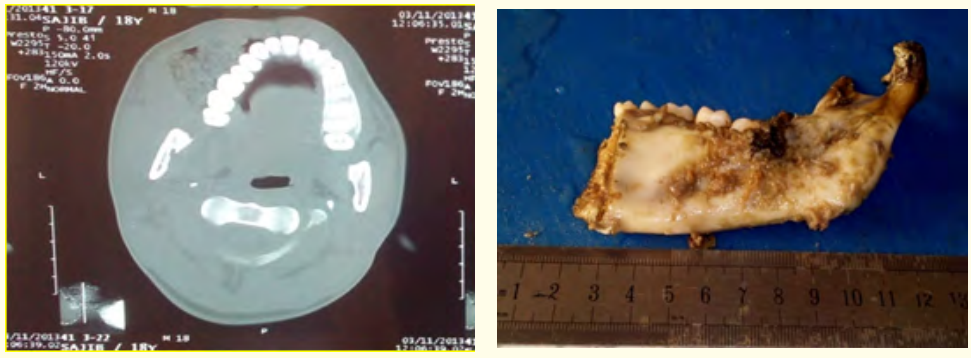

Figure 2: *(A) CT Scan shows a large soft tissue mass is seen in right hemi-mandible along the body (B) Right sided hemi-mandibulectomy specimen with attached soft tissue mass.*

The other laboratory findings were unremarkable. Ultrasonography shows right testicular mass, para-aortic lymphadenopathy and multiple hepatic SOL. CT scan shows soft tissue mass in the right hemimandible area. Excision of the right sided intraoral buccal mass and right sided hemimandibulectomy was done.

Histopathological examination reveals clear cytotrophoblastic cells with central nuclei and syncytiotrophoblastic cells with multiple dark nuclei embedded in eosinophilic cytoplasm in a hemorrhagic and necrotic stroma and diagnosed as testicular choriocarcinoma.

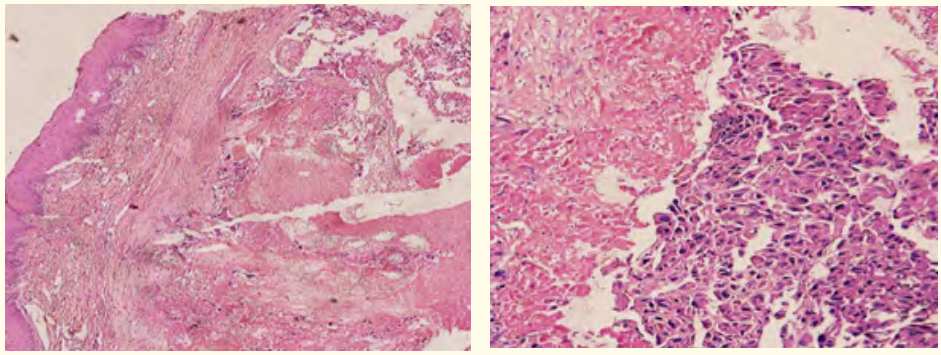

Figure 3: *(A, B) Histopathology shows oral mucosa with clear cytotrophoblastic cells with central nuclei and syncytiotrophoblastic cells with multiple dark nuclei embedded in eosinophilic cytoplasm in a hemorrhagic and necrotic stroma.*

Discussion

Metastatic lesions to the oral cavity are rare being approximately 1% of oral malignancies [5]. The gingiva is the most commonly affected soft tissue followed by the tongue [5].

Testicular germ cell tumors are generally malignant and represent 93% of all testicular neoplasms [7]. Pure testicular choriocarcinoma is a rare germ cell neoplasm, accounting for less than 3% of all the testicular neoplasms and has the worst prognosis of all germ cell tumors [7]. The most common metastatic sites for testicular choriocarcinoma are the lungs, liver and brain. Rare metastasis has been reported on the skin and oral soft tissues [8].

When the lesion of the oral cavity is the first symptom of the disease, the diagnosis can be difficult, as its clinical manifestation is similar to hyperplastic or reactional growth, such as inflammatory fibrous hyperplasia, pyogenic granuloma, peripheral giant cell granuloma, or peripheral ossifying fibroma [9]. In this study the patient complained of an ulcerated lesion in right sided buccal mucosa. Numerous malignant neoplasias can cause metastasis in the oral cavity, such as tumors of the lungs and breasts, leukemias and lymphomas [10]. The predilection of these malignant cells to reach oral soft tissue is not fully understood. Some authors suggest the preference of metastasis to reach the attached gingiva in the presence of chronic gingivitis because the rich vascularity of the inflammatory process would allow trapping of tumor cells and thus being fertile site to spread most of the metastatic process [11]. Chronic inflammation is known to play a role in cancer initiation, promotion, and metastasis [12]. However, the mechanism by which inflammation promotes metastasis is still unclear. One of the aims of the study is to draw attention to the importance of sending all biopsy material for surgical pathologic examination. Choriocarcinoma is a neoplasia classified as nonseminomatous that frequently causes metastasis in the retroperitoneal lymph nodes and lungs, and the dissemination of the tumor to organs such as the liver, brain and bones worsens the prognosis of the lesion [8,10]. This tumor rarely causes metastasis in the oral cavity, and when it is present, the diagnosis is generally already known [8], which highlights the importance of this case. During treatment, the primary lesion must be removed by orchiectomy, with high resection of the spermatic cord. In patients with metastatic illness, the disease must be controlled cytotoxic chemotherapy, which results in complete regression of the metastasis in 60% to 95% of patients [9].

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