CASE REPORT

An uncontrollable bleeding from a mass in the maxilla: Initial presentation of metastatic hepatocellular carcinoma. A case report.

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Introduction:

Hepatocellular carcinoma (HCC) is the 5th most common cancer in the world. It is estimated to cause more than a quarter of a million deaths each year throughout the world. HCC is rising and represents an important public health problem in Egypt as it has one of the highest prevalence rates of hepatitis C virus (HCV) infection in the world.

Hepatocellular carcinoma is a devastating aggressive tumor with metastases usually occurring in advanced stages. Extrahepatic metastases from HCC have been reported in approximately 50% of cases. Lungs, abdominal lymphatics, adrenal glands or the skeleton could be involved[1]. Most cases of HCC with extrahepatic metastases have advanced intrahepatic tumor, with bilobar disease and macrovascular invasion, beyond curative treatment at the time of diagnosis[2]. The overall prognosis of patients with metastatic HCC is poor, with even a poorer prognosis for patients with oral metastases[2,3].

Any malignancy in the head and neck, either primary or metastatic can manifest with bleeding. However, HCC metastatic lesions are hypervascular in consequence of their rich vascularization, as it happens in the primary tumor[4,5]. In the same time, the coagulation alterations due to the basal liver pathology can contribute for this bleeding[5]. The lesion may rupture spontaneously or on trial to take a biopsy, causing devastating uncontrollable hemorrhage.

We present a rare case of metastatic HCC to the maxilla, with spontaneous intractable oral bleeding. This case highlights the character of a metastatic HCC to the sinonasal area, its disctinctive pathological feature and how to suspect such a rare pathology.

Case presentation:

A 48 years old male patient was referred urgently from the oncology department shortly after admission with a right sided cheek swelling, for spontaneous bleeding through the mouth.The patient developed this rapidly growing, painless, rounded swelling, 4x5cm, 3 months earlier. The patient had jaundice and ascites. The laboratory tests were: hemoglobin concentration = 7.2 gm/dL; serum albumin = 2.3gm/dl; AST = 198U/L; ALT = 152 U/L; serum bilirubin = 5mg/dl with 3.2 direct bilirubin. A plain chest X-ray revealed no abnormality.

Oral cavity examination of the bleeding site revealed fresh and profuse bleeding from erosion opposite loose right side upper molar teeth. Trial of packing through the defect to stop the bleeding failed. The patient was moved urgently to the operating theater, in an attempt to stop the bleeding. Ligation of the right external carotid artery was unsuccessful in stopping the bleeding, so the decision was to expose the tumor site. A Weber Fergusson incision with exposure of the tumor site revealed a bleeding friable tissue, easily dissectable, filling the place of an eroded maxillary bone with exposure of the infratemporal fossa.

The lesion was extending to the subcutaneous tissue of the cheek. The whole lesion was easily removed for biopsy. The bleeding was still coming from the exposed branches maxillary artery in the infratemporal fossa and the facial artery in the cheek. Both needed several transfixing sutures before control of bleeding. Packing using Surgicel™ (Johnson & Johnson, New Brunswick, NJ) with inflation of a Foley's catheter in the resulting cavity was done at the end of the procedure. A metastatic lytic lesion was suspected from the intraoperative findings. The patient was transferred to the ICU. An abdominal ultrasound revealed multiple focal lesions in the liver. Further investigations for tumor markers revealed an alpha fetoprotein = 72163 IU/ml (normal up to 5.8), a CA 19.9 = 105.3 U/ml (normal up to 39) and a CA 125 = 355.9 U/ml (normal up to 35).

The biopsy from the maxillary mass proved to be hepatocellular carcinoma (Figure 1). Immunohistochemistry was positive for specific staining by hepatocyte antigen-antibody (Figure 2). A metastatic workup for the patient could not be performed due to his bad general condition. Acute elevation of the liver enzymes and bilirubin rapidly issued and the patient passed away shortly.

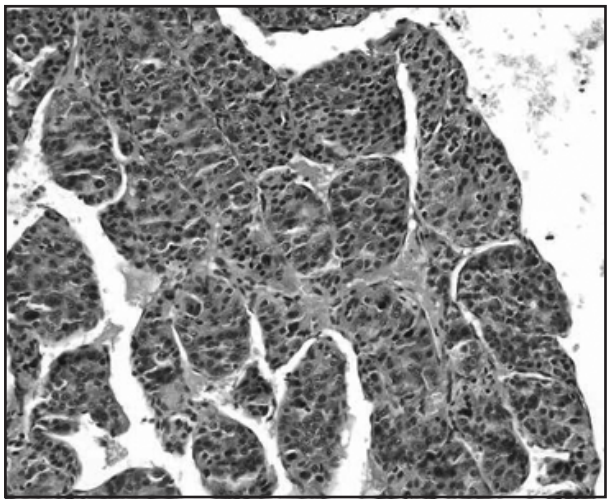

Figure 1: H&E of the lesion showing hepatocellular carcinoma (x200).

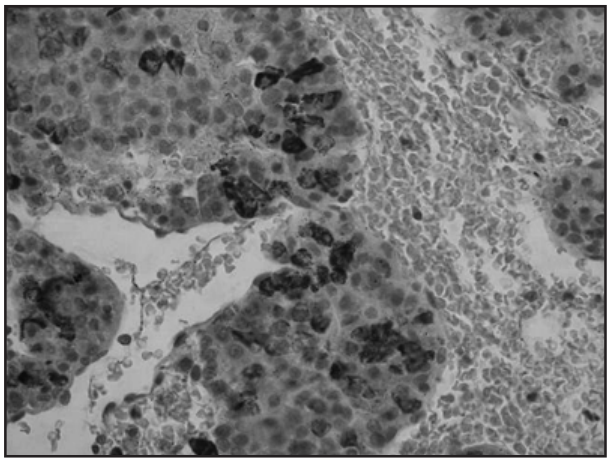

Figure 2: Immunohistochemistry specific staining by hepatocyte antigen antibody (x400).

Discussion:

The rate of HCC is increasing in Egypt as the major risk factor of viral hepatitis infection is high. There is a doubling in the incidence rate in the past 10 years, with most patients having cirrhosis, in particular from previously undiagnosed hepatitis C[5]. The incidence of bone metastasis from HCC accounts for approximately 1.6%-16% and the most common sites are the vertebrae. Metastases into maxillofacial territory are unusual, affecting preferably male patients (male: female ratio 46:4) over 50 years (15-88 years range), and it usually occurs in the mandible and less frequently maxilla[1,4].

All bone metastases from HCC appear as osteolytic lesions, and usually manifest as multiple lesions[2]. However, maxillofacial metastases can be characteristically solitary and can precede the knowledge of liver disease in 59% of cases[4]. The establishment of a histologic diagnosis of metastatic HCC in the oral region, can be extremely difficult before identification of the primary[4]. This was the case in our patient. We first suspected a primary maxillary tumor, in a patient with liver cirrhosis, due to lack of symptoms other than maxillary swelling. However, intraoperative exposure of the lesion, drew our attention to an osteolytic lesion and thus a possibility of metastases. Workup revealed the multifocal liver lesions, and as pathology lab was informed, a confirmation of HCC was done.

The precise mechanism of metastatic HCC to the maxillofacial region is poorly understood. Basically, there are 2 pathways from the liver to the maxillofacial territory. It is believed that portal hematogenous route is the preferred route. Metastatic dissemination must thus reach the lung first [3,4]. According to this, cases of HCC with maxillofacial metastasis must present with lung metastasis too. However, this is not always the case, as seen in our patient who presented with no lesion in the lung.

An alternative route for such dissemination, consists of a connection between the azygos and hemiazygos veins and the vertebral venous plexus (Batson's plexus), forming a pathway of rich anastomoses of paravertebral veins that lack valves and may be capable of bypassing other venous systems, explaining this maxillofacial affection without lung affection[1,4].

The high vascularity of the metastatic lesion with HCC, accompanied by concurrent coagulopathy caused by primary liver disease, often associated with this group of patients, can lead to hemorrhagic episodes, particularly in bone lesions and are extremely difficult to manage[3]. Biopsy can result in uncontrollable bleeding, even with external carotid artery ligation. Trials of haemostatic agents, packing and wide sutures are usually partially successful[3]. Application of local radiotherapy can be resorted to for control of recurrent bleeding episodes[4]. For control of the spontaneous, profuse, life endangering bleeding we encountered in our patient, we first resorted to packing, then to external carotid ligation, but both were unsuccessful. Only direct exposure of the bleeding vessels stopped the bleeding.

One should be aware of the possibility to encounter a maxillofacial lesion as the first sign of metastatic HCC especially with a high incidence of HCV in Egypt. Any rapidly enlarging vascular swelling, with ill-defined destruction, suggestive of malignancy should be suspected for metastatic HCC. Biopsy should not be resorted to except after proper imaging and general investigations, to avoid uncontrollable bleeding.

References:

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