Sinonasal Metastatic Hepatocellular Carcinoma Initially Presents with Unilateral Vision Loss

Yi-Chan Lee1,2, Chi-Che Huang2,3, Shu-Hang Ng1, Shih-Ming Jung4, Ta-Jen Lee1* and Chia-Hsiang Fu1,3*

1Department of Otolaryngology, Chang Gung Memorial Hospital, Taiwan
2Department of Medicine, Chang Gung University, Taiwan
3Department of Medical Imaging and Intervention, Chang Gung Memorial Hospital, Taiwan
4Department of Pathology, Chang Gung Memorial Hospital, Taiwan
*both are equally contributed

Abstract

Hepatocellular carcinoma (HCC) seldom presents only as acute visual loss without any other abdomen or systemic symptoms. Sinonasal area, besides, is an extremely rare metastatic site for HCC as well. We reported a rare case of a 69-year-old male, presenting initially with of left eye rapid vision loss, and the sinonasal mass lesion was found two months later when he had left epistaxis attacked. The initial magnetic resonance image was reviewed and indicated a mass lesion on lateral wall of left sphenoid sinus with cavernous sinus and left optic apex involved. Metastatic HCC was subsequently confirmed by biopsy with immunohistochemistry stain. The original HCC was subsequently discovered by abdomen echo and elevated α-fetoprotein. He was then referred for chemotherapy. The presenting symptom of sinonasal tract metastasis originated from hepatic malignancy as acute vision loss could be easily missed. Physicians should always take metastatic neoplasms into clinical differential diagnosis of a unilateral sinonasal lesion.

ABBREVIATIONS

HCC: Hepatocellular Carcinoma; CT: Computed Tomography; MRI: Magnetic Resonance Imaging; Hep Par-1: Hepatocyte Paraffin 1; AFP: α-Fetoprotein; GPC3: Glypican-3; EMA: Epithelial Membrane Antigen; HBV: Hepatitis B Virus; HCV: Hepatitis C Virus

INTRODUCTION

Sinonasal metastatic malignancies occur infrequently, and the most common regions affected are the maxillary and ethmoid sinuses [1]. For sinonasal metastatic tumors, the most frequent site of primary malignancy is the kidney, followed by the lung, urogenital tract, breast, gastrointestinal tract, and thyroid gland [2]. Hepatic malignancy generally does not originate in the sinonasal tract, and most of the reported cases present with nasal bleeding [3]. Here we report a rare case of metastatic Hepatocellular carcinoma (HCC) in the sphenoid sinus, with an uncommon initial manifestation as acute vision loss.

CASE PRESENTATION

A healthy 69-year-old male with a past medical history remarkable only for hypertension presented at our emergency department with a 1-week history of vision loss in his left eye. An ophthalmologist admitted this patient for presumed left retrobulbar optic neuritis. At the time of admission, the vision in his left eye was only sufficient to count fingers at a distance of 50 cm, with normal eyeball movement. Steroid pulse therapy for several days provided a limited response.

The patient returned 2 months later for a repeat left-sided epistaxis. We also diagnosed left abducens nerve palsy. Endoscopic examination identified a necrotic, friable mass media to the left middle turbinate (Figure 1A). A sinus computed tomography (CT) scan revealed a 3.8 × 2.5 cm lobulated soft-tissue mass centered at the left sphenoid sinus with adjacent optic canal infiltration (Figures 1B). A review of the MRI acquired at another hospital 3 weeks earlier revealed a small mass at the superolateral corner of the left sphenoid sinus with adjacent optic canal infiltration (Figures 1C and D).

Subsequently, a biopsy was performed, and pathology revealed a poorly differentiated neoplasm. Immunohistochemical...
analysis revealed tumor cells that stained positive for Hepatocyte paraffin 1 (Hep Par-1), α-fetoprotein (AFP), and glypican-3 (GPC3) and negative for epithelial membrane antigen (EMA) and vimentin (Figure 2). Therefore, a pathological diagnosis of metastatic HCC was made. The patient denied any abdominal symptoms and was referred to the medical department for liver examination. Laboratory tests showed a high AFP level (3308/μL), thrombocytopenia (113,000/μL), and elevated liver enzyme amount (aspartate aminotransferase, 113/μL; alanine aminotransferase, 49/μL). Hepatitis screening was negative for hepatitis B surface antigen but positive for hepatitis C virus. An abdominal sonography showed liver cirrhosis with multiple hypo echoic liver tumors, suggestive of HCC. The sinonasal tract was the only location of metastasis following systemic work-up. The patient received chemotherapy but died as a result of septic shock 3 months later.

DISCUSSION

HCC, a common malignancy in East Asia, is the most common hepatic tumor. The most common distant metastatic sites of HCC include the lung (51.6%), bone (10.1%), and adrenal capsule (8.4%) [4]. The sinonasal tract is a very uncommon metastatic location pertaining to HCC. Sphenoid sinus malignancy comprises less than 1% of all sinonasal malignancies, and metastatic tumor at this location is even rarer [5]. Among sinonasal metastatic malignancies, the sites of primary origin include the kidney (49%), lung (12%), urogenital tract (12%), breast (9%), gastrointestinal tract (6%), and thyroid gland (4%) [2]. Metastatic HCC has seldom been reported in this region. In the reported cases of metastatic HCC in the sinonasal tract, epistaxis was the most frequent symptom [3]. HCC initially presenting with symptoms pertaining to the eye, without any abdominal symptoms, is also extremely rare. To our knowledge, there are only seven previously reported cases in the published literature [6].

Regarding the anatomical location of the sphenoid sinus, the involvement of the cavernous sinus and presentation of diplopia would be commonly expected if a tumor progression at this site is previously reported [3,7]. As observed in the present case, unilateral vision loss without the involvement of extra ocular movement as the only initial manifestation is very rare. The optic nerve leaves the back of the orbit via the optic canal and can become completely surrounded by a posterior ethmoidal Onodi cell, the sphenoid sinus, or an aerated anterior clinoid process. In our patient, initial MRI revealed a small enhanced mass in the superolateral wall of the left sphenoid sinus that infiltrated into the left orbital apex without extensive intra orbital invasion. Therefore, the initial presentation of blurred vision was likely the result of the involvement of the optic nerve at the orbital apex.

HCC, with a high extra hepatic metastasis rate, has a marked incidence of 30 cases per 100,000 people in Taiwan, and most of these (80% to 90%) are related to the hepatitis B virus (HBV) [8,9]. Although HBV is still dominant in causing HCC, the relative importance of the hepatitis C virus (HCV) has changed over the last two decades. The proportion of anti-HCV positive cases has increased [9]. In some areas of Taiwan, HCV-associated HCC has surpassed HBV-associated cases, probably because of a
vaccination program against HBV launched in the late 1980s. Our patient did not receive this vaccination. However, he was positive for HCV and negative for HBV. This information was determined when the hepatic malignancy was diagnosed. A previous study also reported a younger mean age in cases of HBV-associated HCC (40–60 years) compared with the age in cases of non-HBV-associated HCC (50–70 years) [10].

HCC can spread through the blood and/or via the lymphatic system [11]. However, the lymphogenous route is less likely for an isolated sinonasal metastatic HCC. Most sinonasal metastases tend to occur via hematogenous spread. The most common hematogenous route appears to be via the caval venous system, through the pulmonary circulation to the heart, and from the heart to the sinuses via arterial vessels in the head and neck [11]. Another proposed mechanism for hematogenous metastasis involves the retrograde flow of venous blood to the cranial base via the prevertebral and vertebral plexus. The prevertebral and vertebral venous system comprises epidural and prevertebral veins with numerous intertwining vessels that communicate at every somite level [12]. Because there are no valves, an increase in intra thoracic or intra-abdominal pressure could drive tumor cells into the vessels of the paranasal sinuses via the vertebral venous plexus in a retrograde manner.

The symptoms of metastasis such as acute unilateral vision loss, as observed in our patient, were initially the only presentation of the asymptomatic primary malignancy located far away from the head and neck region. Metastatic and primary tumors in the sphenoid sinus share similar clinical and imaging presentations and early diagnosis sometimes may be difficult with an undiscovered primary origin. In this case report, we have presented the clinical manifestation of a very rare metastatic HCC in the sphenoid sinus for reminding the physicians to consider metastatic tumors during the differential diagnosis of a sphenoid sinus lesion with unilateral acute vision loss.

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REFERENCES