

Hepatoma Metastasizing to the Retromaxillary Region with Initial Otologic Manifestation^{*}

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ABSTRACT

We present here a 50-year-old heavy alcoholic, male patient who had been suffered from neuralgic pain of the face and temple. A tumor was found in the retromaxillary region with a routine CT scanning. Biopsy from the maxillary sinal content indicated a metastasis from hepatoma. Afterwards, the detail of generalized metastatic pattern was cleared, and the patient was given appropriate therapies.

INTRODUCTION

A clue to the diagnosis of the tumor originating from the retromaxillary or parapharyngeal space can be obtained by its various signs and symptoms. Most of these signs and symptoms, however are sometimes secondary to destruction or compression of the surrounding tissues by the tumor growth, and may manifest themselves late in clinical course; this will miss the chance of radical surgery. We present a patient who had been suffered from neuralgic pain of the face and temple for long duration; the pain was excruciating and did not respond to palliative treatments. A tumor happened to be detected in the head and neck areas with a routine CT scanning. Biopsy from the maxillary sinal content, which consisted of a part of the advancing tumor, indicated a metastatic lesion from hepatocellular carcinoma (hepatoma). After confirming its primary site, the detail of generalized metastatic pattern was cleared.

CASE PRESENTATION

A 50-year-old male has had about 0.7 liter *sake* (rice wine) every day for 30 years, and

has been pointed out liver damage. Approximately for four months before the consultation to the Department of Otolaryngology, Okayama University Hospital, he had had persistent neuralgic pain around the left side of the face and temple; the pain did not respond to repeated accupunctures. An otolaryngologist failed to find the cause of the pain through x-ray examinations and tried to relieve the pain without much effect. About 10 days before the consultation, he started numbness of the tongue, slightly disturbed opening of the mouth, and tinnitus and loss of hearing of the left side. He was then referred to the Department of Anesthesiology, Okayama University Hospital, and further to the Department of Otolaryngology.

By the time of the consultation to the Department of Otolaryngology, the patient was rather slim, pale on the face, free of motility disturbance, and clear in consciousness. The left side of the face was slightly swollen, otherwise ears, nose and throat appeared normal. An audiogram (Fig. 1) revealed a mixed type of moderate hearing loss of the left side with a marked air-bone gap. Neuralgic pain in the left facial and temporal regions and numbness

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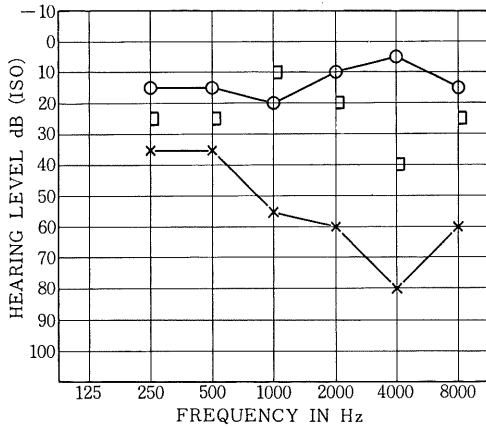


Fig. 1. Audiogram showing a mixed type of moderate hearing loss of the left side with a marked air-bone gap.

Table 1. Laboratory data on admission

TP	7.08 g/dl
Alb	3.34 g/dl
A/G	0.89
TTT	6.7 MU
ZTT	12.9 KU
T. Bil	1.05 mg/dl
D. Bil	0.45 mg/dl
GOT	68 IU/l
GPT	101 IU/l
AIP	132 IU/l
LAP	122 IU/l
Gamma-GTP	173 IU/l
ChE	206 IU/l
LDH	449 IU/l
CEA	4.4 ng/ml
T. Cho	112 mg/dl
Alpha-Feto	12.9 ng/ml
ICG 5''	62.6%
10''	30.1%
15''	15.3%
IgG	2,936 mg/dl
HBsAg	(-)
Ab	(+)
WaR	(-)

of the left mandible corresponded to the first and second divisions of the left trigeminal nerve; other cranial nerves were not involved by the tumor.

Liver function test (Table 1) then indicated the high degree of dysfunctional state. The

patient was referred to the Department of Medicine for further examinations; the liver was palpable two finger-widths below the right costal margin, and elastic firm. A tumor was palpable at the left eighth rib. CT scanning of the head (Fig. 2a, b) demonstrated a tumor

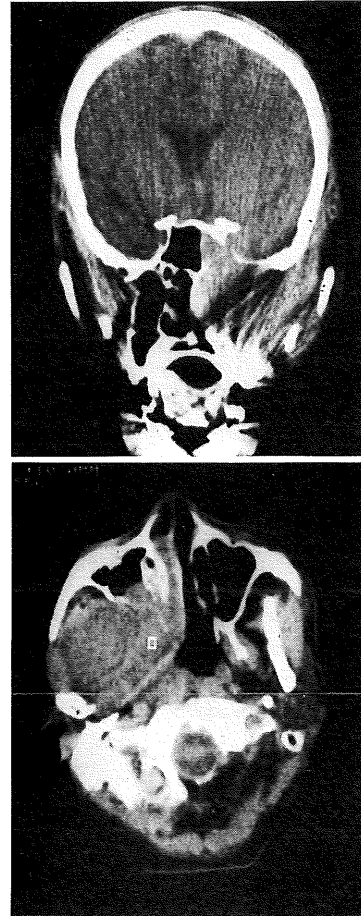


Fig. 2. a and b. TC scanning of the head showing a tumor mass involving the left retro-maxillary, parapharyngeal and infratemporal fossa.

mass of the isodensity, which apparently involved the left retromaxillary, parapharyngeal and infratemporal fossa, and further extended to the processus pterygoideus and mandible including ramus mandibulae. We then suspected the presence of a tumor in the left retromaxillary space, and admitted the patient to the Department of Otolaryngology for the local biopsy. A part of the posterior wall of the left maxillary sinus was destroyed due to a tumor mass protruding through the sial wall.

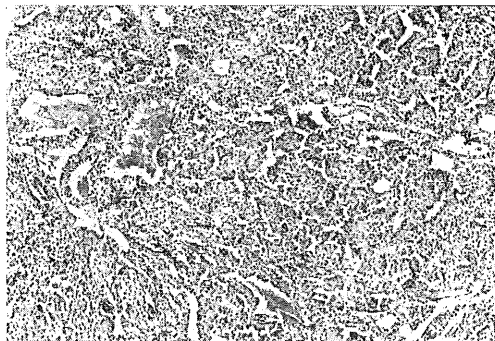


Fig. 3a. Biopsy from the maxillary sinus content showing a trabecular pattern and blood pooling. HE, $\times 40$.

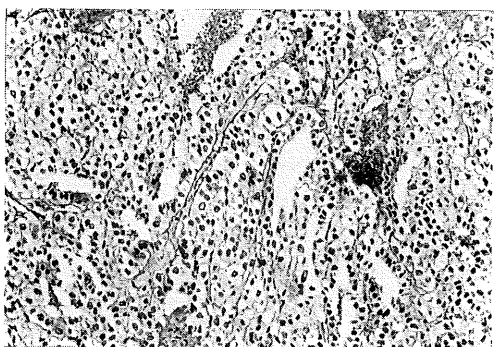


Fig. 3b. The biopsied specimen showing endothelium along the trabeculae. Silver, $\times 100$.

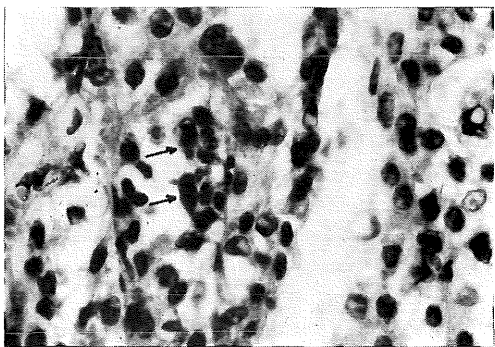


Fig. 3c. The biopsied specimen showing granular cytoplasm and extra- and intra-cellular bile pigments (arrows). HE, $\times 400$.

As to pathologic findings, biopsied specimens consisted of several pieces of friable, hemorrhagic materials measuring up to 1.5 cm in the largest dimension. Microscopically, a trabecular pattern, made of one to a few cell thickness, was pronounced together with blood pooling between trabeculae (Fig. 3a). Endothelium was often found along the trabeculae; otherwise, no major stromal component was present (Fig.

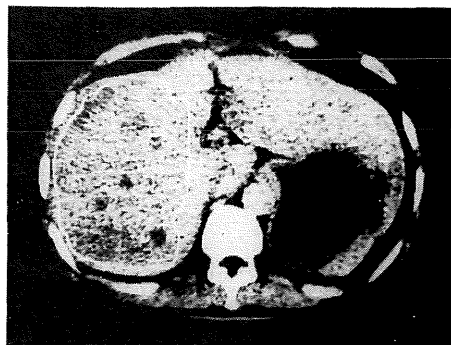


Fig. 4. CT scanning of the liver showing multiple low-density areas in the right lobe.

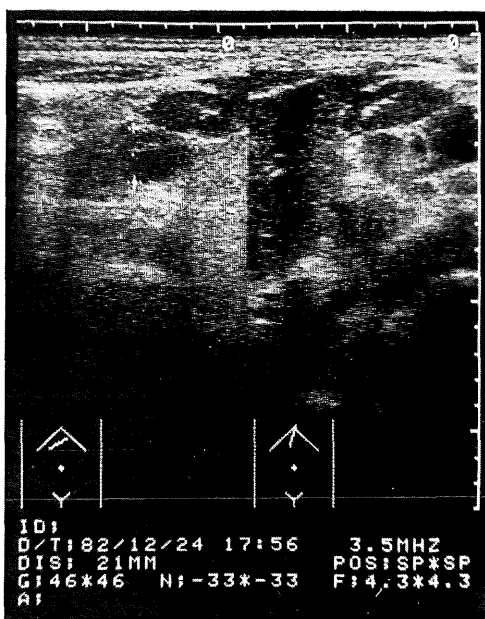


Fig. 5. Liver echo showing the irregular liver surface and heterogeneous multiple nodules.

3b). The tumor cells consisted of the round nucleus with prominent nucleolus and abundant, granular, acidophilic cytoplasm, fine granules in which were completely digested by diastase indicating glycogen; Alcian blue was negative throughout (Fig. 3c). Very occasionally, deeply brown-yellow pigments were identified extra- and intra-cellularly; these were negative for iron, Masson-Fontana and Grimelius. Although Stein's iodine staining for bile pigments was unsatisfactory, these substances were most likely to be bile pigments. Overall histologic pattern was compatible with hepatoma of the grade II by Edmondson and Steiner¹¹.

CT scan (Fig. 4) of the right lobe of the

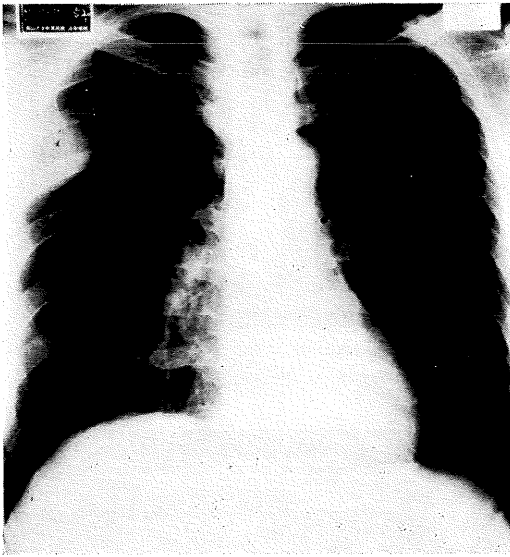


Fig. 6. Chest radiograph showing a discrete tumor protruding to the right upper field of the lung.

liver disclosed multiple low-density areas indicating the possibility of hepatoma. Liver echo examination (Fig. 5) revealed the irregular liver surface, and heterogeneous multiple nodules of from low to high density echo. Chest radiograph (Fig. 6) showed a discrete tumor with homogeneous density protruding to the right upper field of the lung. ^{67}Ga scintiscan demonstrated high uptake of the radioisotope in the left maxilla, right upper chest, left lower chest and right buttock. Liver biopsy has been held because of considerably prolonged hemorrhagic tendency, from which intractable intra-abdominal bleeding may ensue.

From the above findings, we reached the conclusion that the patient developed a metastasis to the retromaxillary region from hepatoma as a part of generalized metastases, and that he manifested signs and symptoms first pertaining to the head and neck areas. Under this conclusion, the patient was given 250 mg of 5-fluorouracil through the A. temporalis superficialis 23 times with a total amount of 5,750 mg, and also 200 rads linac x ray using a wedge filter 25 times with a total amount of 5,000 rads. He has been fairly well in physical conditions, although developed gait disturbance apparently due to vertebral metastasis.

DISCUSSION

This case was initiated with neuralgic pain in the facial and temporal regions. Local findings and conventional radiographs were almost not worth speculating the presence of tumor; we recognized once again the usefulness of CT scanning for the lesions, in particular the tumor, in the retromaxillary or parapharyngeal region.

The diagnosis of hepatoma, except for in a typical case, is often difficult because of a lack of major symptoms such as hepatomegaly, jaundice or ascites. Cases complicated by cirrhosis will remind us the development of hepatoma based on liver function tests. On the other hand, the presence of tumor in the liver with CT scan does not always indicate hepatoma *per se*, because metastatic lesions to the liver are far more frequent than hepatoma. For instance, the two cases reported by Kato et al.⁶⁾ failed to be diagnosed while alive, and their necropsies proved osseous metastasis from hepatoma. Biopsy of our case was facilitated due to a protruding tumor mass into the maxillary sinus. In general, however, preoperative, diagnostic biopsy of this particular site may sometimes risk getting worse in surgical intervention.

Hepatoma develops higher in the frequency in Oriental than in Caucasian or Negro, and higher in its occurrence than other malignant neoplasms of the liver. The diagnosis of hepatoma, as mentioned before, is often difficult; this sometimes results in unfavorable prognosis with remote metastases. According to Yamaguchi⁸⁾, the frequency of metastasis was 64.0% (144 of 225 cases with hepatoma), and its pattern consisted of hematogenous, lymphogenous, and infiltrating or disseminated metastases. This case has the possibility of developing the above three ways of metastasis. As far as the metastasis to the cranial region is concerned, however, osseous metastasis, particularly through hematogenous route, is the most likely candidate. That is, the metastasis to surrounding osseous component of the parapharyngeal space, which is located behind maxillary sinus, or to the posterior wall of the maxillary sinus *per se* occurred first.

The thirty cases with a mass situated in the parapharyngeal space, studied by Som et al.⁷⁾, included mixed tumor of the parotid or palate

(8 cases), schwannoma (6 cases), paraganglioma (5 cases), and two cases of metastatic tumor, although these two were not specified as to their primary sites. A frequency of osseous metastasis by hepatoma ranged from 1.6 to 15.8% according to Ishizu et al.⁴⁾ and other investigators. A frequency of its bone distribution was 137 cases to the vertebrae and 17 cases to the cranium out of 278 cases with hepatoma by Yanase et al.⁹⁾, and was 62 cases to the vertebrae and 10 cases to the cranium out of 114 cases by Funaki et al.²⁾, indicating the highest incidence in the vertebral metastasis. Kan et al.⁵⁾ have reported a case of hepatoma with several thumb-sized tumors on the scalp. Radiograph of the skull revealed multiple osteolytic lesions, biopsy of one of which proved metastasis from hepatoma. The case of Kan et al. was, however, confined to the skull itself. To our knowledge, the metastasis to the retro-maxillary space by hepatoma as seen in our case has never been reported in the literature.

Initial neuralgic pain around facial and temporal regions can be due to stimulated sensory component of the trigeminal nerve secondary to an expanding tumor growth. On the other hand, the pain in these regions well corresponds to that caused by Vidian neuralgia³⁾. Therefore, we must utilize every possible modern techniques, including CT scanning, to solve the cause of persistent and obstinate pain in the head and neck areas.

Addendum: The patient died on the 6th September. Necropsies from the left maxillary region and the liver, using a Silverman's needle, showed the almost identical finding with that of the biopsied specimens reported herein.

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