Mucoepidermoid Carcinoma of the Maxillary Sinus —Case Report—

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ABSTRACT. Mucoepidermoid carcinoma is an adenocarcinoma derived from the mucous ducts. Three kinds of cells, mucous cells, intermediate cells and epidermoid cells, constitute mucoepidermoid carcinoma, and this tumor is of three grades; low-grade, intermediate-grade and high-grade. The prognosis of high-grade tumors is very poor.

Most cancers in the paranasal sinuses are squamous cell carcinomas. Mucoepidermoid carcinoma of the maxillary sinus is very rare. We encountered a case of mucoepidermoid carcinoma of the maxillary sinus in a 73-year-old female. The details of the case and a review of the literature are presented.

Key words: mucoepidermoid carcinoma — maxillary sinus — high-grade type

Although the mucosa of the paranasal sinuses is columnar epithelium, most cancers of the paranasal sinuses are squamous cell carcinomas due to metaplasia. Therefore an adenocarcinoma arising in the nasal cavity and/or paranasal sinuses is rare. Mucoepidermoid carcinoma is an adenocarcinoma derived from the mucous ducts, which is rarely found in the maxillary sinus. A case report and a review of the literature are presented.

CASE

A 73-year-old woman had complained of right nasal obstruction and epistaxis since August 1994. She also had swelling of the right cheek and diplopia. She visited a local hospital and underwent a nasal biopsy. She entered our hospital on October 31 with a diagnosis of right maxillary adenocarcinoma. As for her past history, she underwent a nasal polypotomy at thirty years old.

On her first visit to our hospital, slight swelling of the right cheek and exophthalmos were noted (Fig 1). Anterior rhinoscopy showed a light-red colored mass in the right nasal cavity and lateral movement of the inferior turbinate (Fig 2). Right oculomotor nerve palsy and glaucoma were found by an ophthalmologist.

A plain x-ray examination and tomography disclosed a diffuse shadow in the right maxillary sinus and ethmoid sinus (Fig 3, 4). An enhanced CT scan showed a tumor in the area of the right nasal cavity, maxillary sinus and ethmoid sinus, and destruction of the superior and posterior wall of the maxillary sinus and the orbital wall of the ethmoid sinus (Fig 5). MRI

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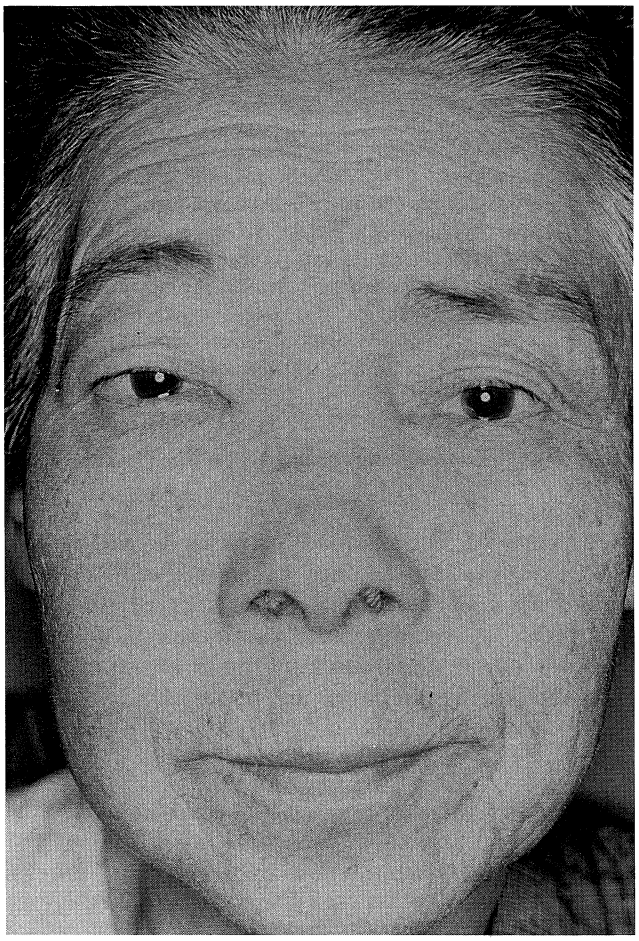


Fig 1. A anterior view of the face showed slight swelling of the right cheek and right exophthalmos

showed a tumor of low intensity (T1 weighted image), high intensity (T2 weighted image), and enhancement of Gd-DTPA (Fig 6). The skull base was intact and there was no intracranial invasion.

A biopsy of the right nasal mass was diagnosed moderately-differentiated squamous cell carcinoma (Fig 7).

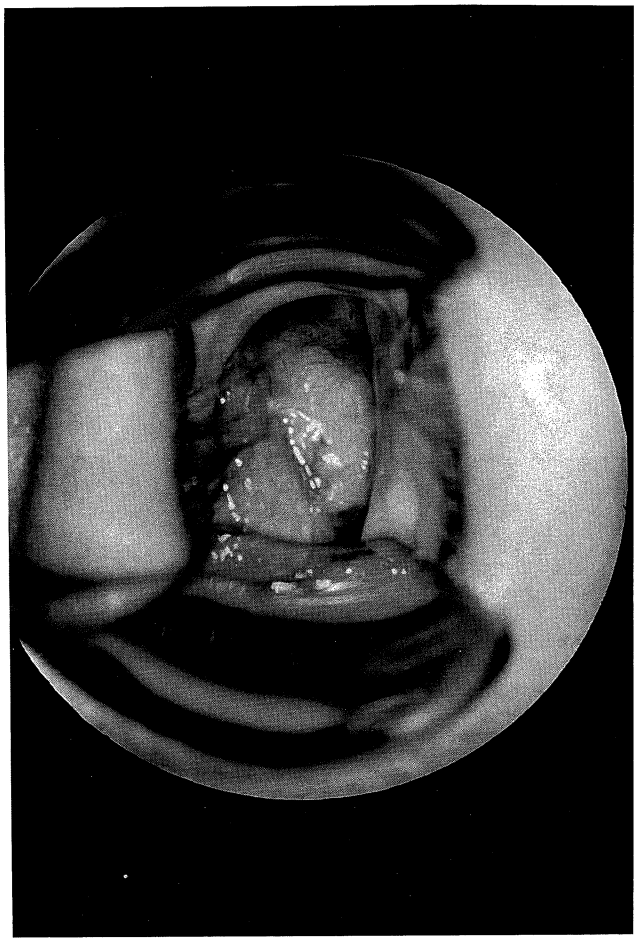
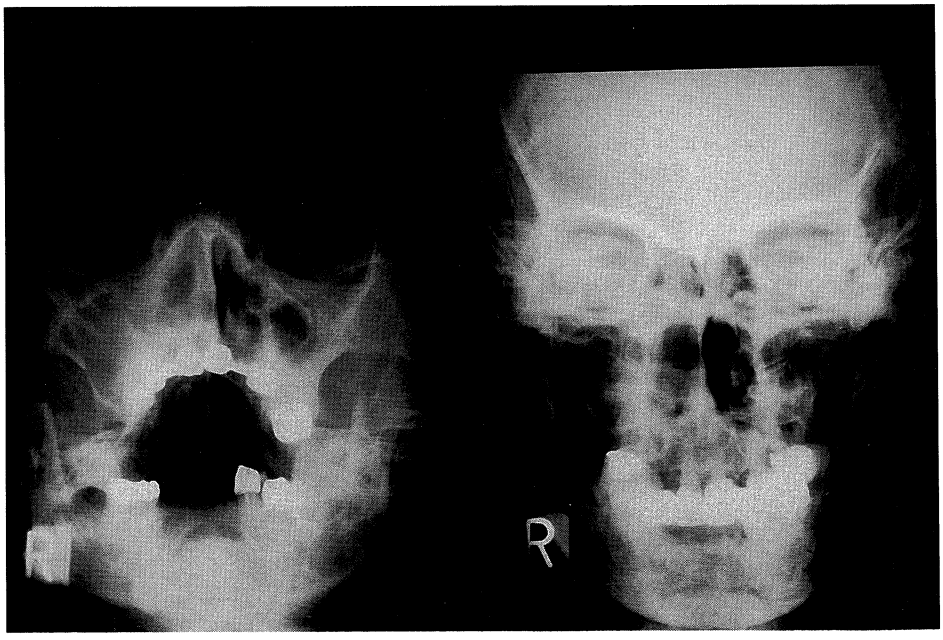


Fig 2. Anterior rhinoscopy showed a light-red colored mass in the right nasal cavity and lateral movement of the inferior turbinate



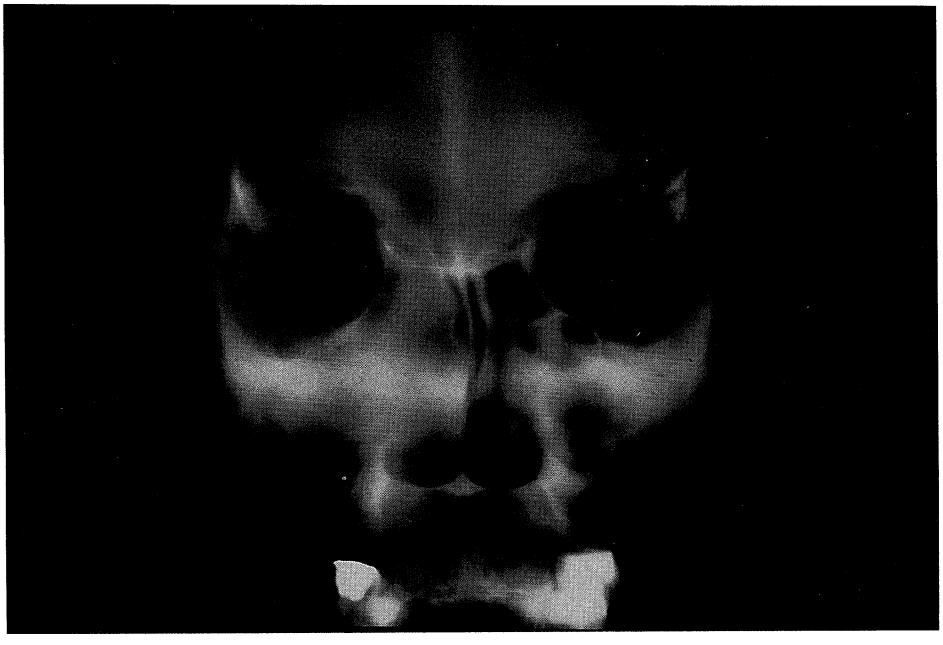


Fig 3, 4 Plain x-ray examination and tomography of the nose showed only a right-side shadow

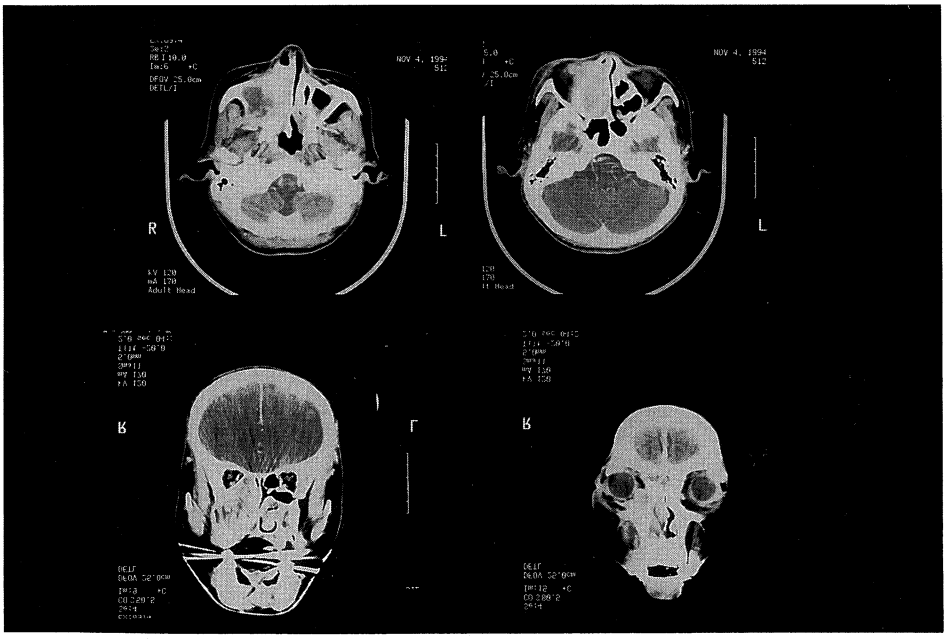


Fig 5. Enhanced computed tomographic scans showed a large mass in the right nasal cavity, maxillary sinus and ethmoid sinus The tumor invaded the right orbita.

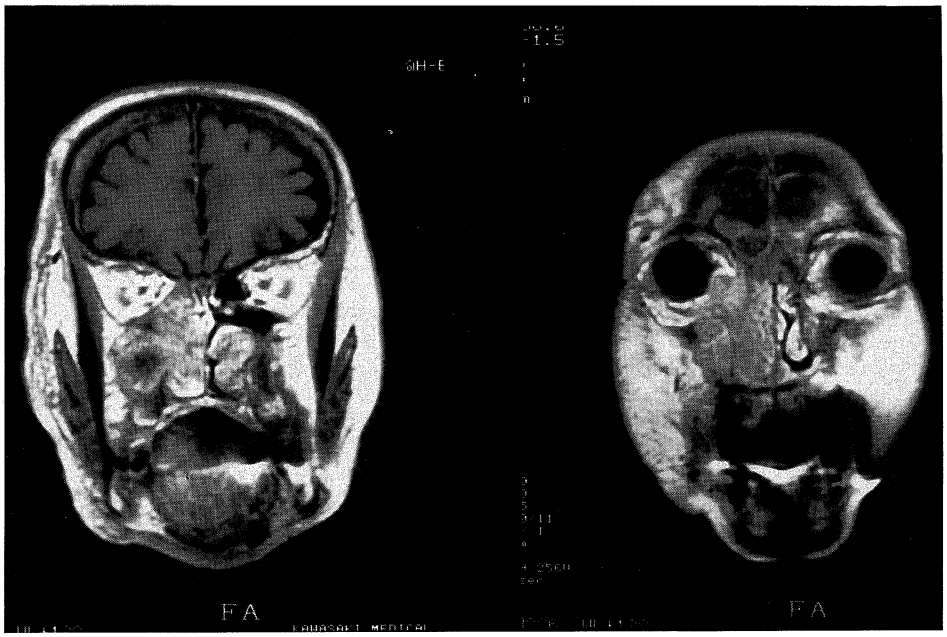


Fig 6. MRI (coronal view) showed that a tumor filled the right nasal cavity, maxillary sinus and ethmoid sinus, but no intracranial invasion was demonstrated.

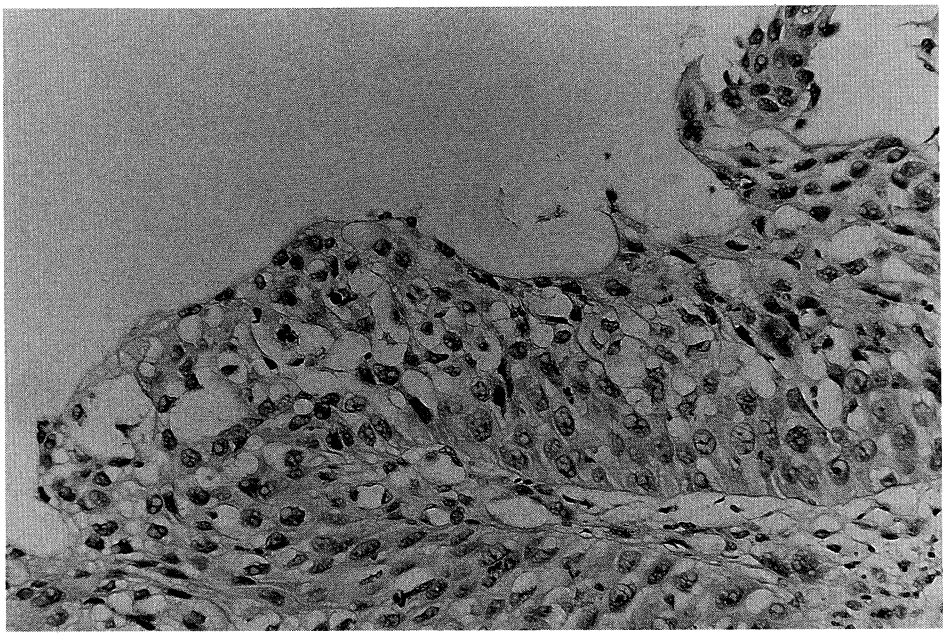


Fig 7. A nasal biopsy showed a moderately-differentiated squamous cell carcinoma

The patient's peripheral blood count values and tumor markers are shown in Table 1. Laboratory data showed thrombocytopenia ($4.9\times10^4/\mu1$). A bone marrow biopsy showed hypocellular marrow of unknown cause. No metastasis was found. Other laboratory data before and after treatment showed no abnormality. Among various tumor markers, only the serum SCC related antigen (SCC-Ag) level was found to be extremely high.

TABLE 1. Peripheral blood count values and tumor markers

RBC : $420\times10^4/\mu1$
WBC : $5900/\mu1$
Pl. : $4.9\times10^4/\mu1$
CEA : 1.0ng/ml ↓ (2.5 ↓)
α fetoprotein : 3ng/ml ↓ (20 ↓)
SCC : 15ng/ml(1.5 ↓)
CA19-9 : 11U/ml(37 ↓)
CA125 : 38U/ml(35 ↓)

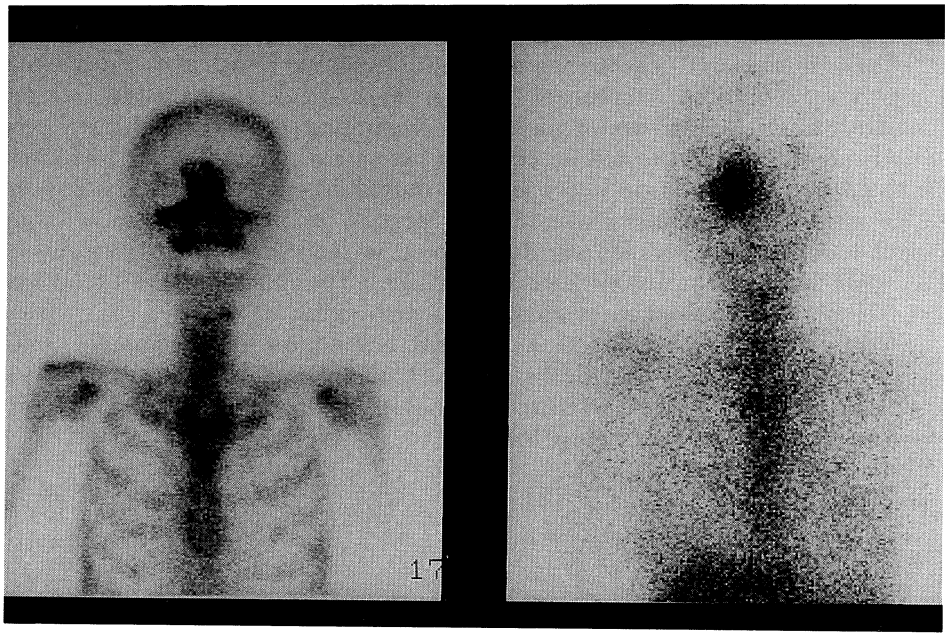


Fig 8. Bone scintigraphy disclosed increased activity in the right maxilla. ^{67}Ga scintigraphy disclosed increased activity in the right maxillary region. Neither examination showed any regional or distant metastasis.

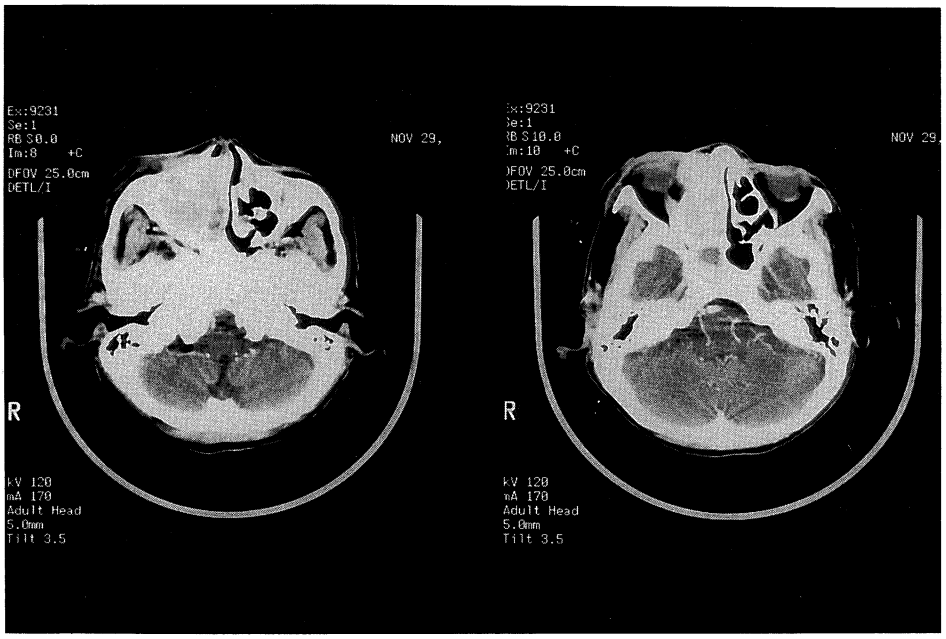


Fig 9. CT scan after chemotherapy and radiotherapy There was no response.

Bone scintigraphy and ^{67}Ga scintigraphy examinations revealed a hot spot in the right maxillary region with no regional or distant metastasis. Therefore, the diagnosis was right maxillary carcinoma (T4N0M0 StageⅣ).

Then intraarterial chemotherapy with Cisplatin 75mg and Peplomycin 25mg via superficial temporal artery and irradiation were started simultaneously. Eighteen days after beginning of chemothrapy and radiotherapy, CT scan showed no effect on her tumor. Therefore we performed the Groβ Denker operation under general anesthesia on December 12. A bone defect was found in the medio-inferior portion of the right orbital wall. Tumor had evidently invaded the right orbita, so the right orbita was removed. The sphenoid sinus and skull base were intact. The intraoperative blood loss was 928ml. The postoperative course was uneventful.

Histopathological examination revealed admixtures of epidermoid cells, intermediate cells and mucous cells. A predominance of epidermoid cells and intermediate cells was seen. The histopathological diagnosis was a high-grade mucoepidermoid carcinoma (Fig 10).

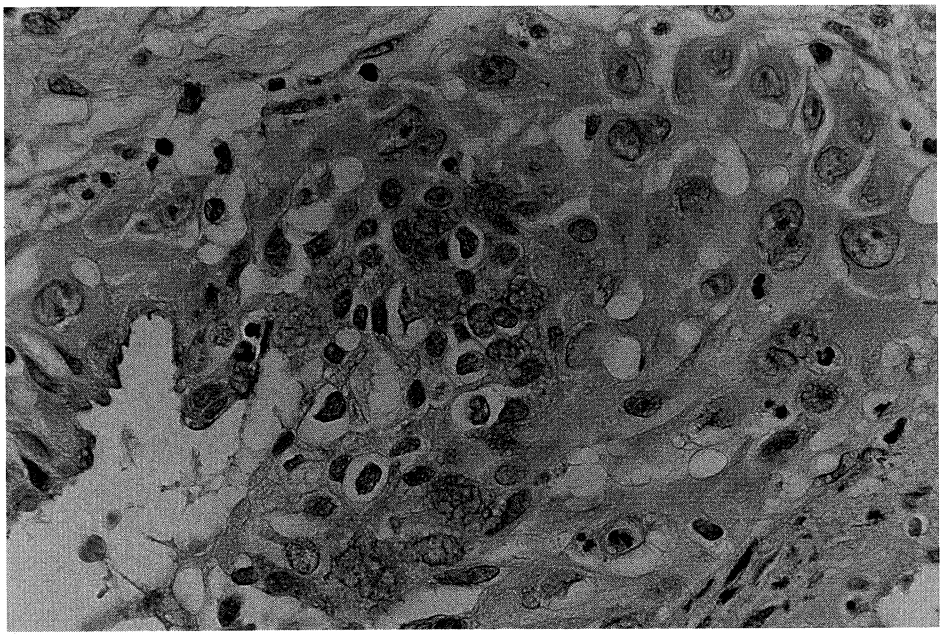


Fig 10. Histopathological findings showed epidermoid, intermediate and mucous cells Since the epidermoid cell and intermediate cell were predominant, the diagnosis was high-grade mucoepidermoid carcinoma.

Postoperative radiation (36Gy) was done, and CT and MRI after treatment showed no tumor (Fig 11, 12). Recurrence of maxillary carcinoma has not been observed to date during the 19 months since operation.

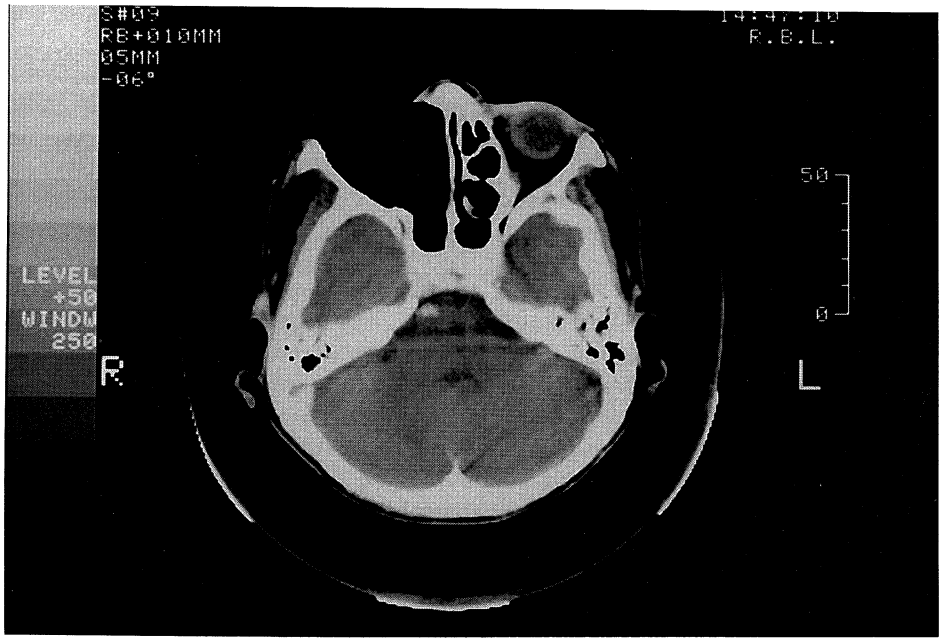


Fig 11. CT scan after operation showed no tumor in the right maxillary region

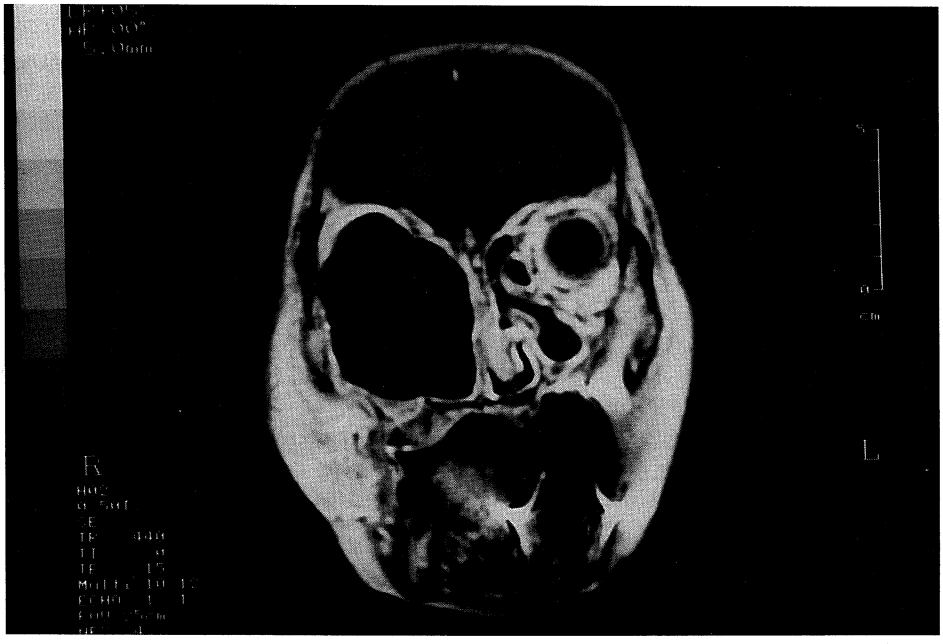


Fig 12. MRI after treatment showed no recurrence of tumor

DISCUSSION

Adenocarcinomas rarely arise in the nasal cavity and/or paranasal sinus. Adenocarcinomas are of three types ; *i.e.*, colonic-type, adenosquamous cell type and adenocarcinomas derived from the mucous ducts. The third includes mucoepidermoid carcinoma and adenoid cystic carcinoma.

Mucoepidermoid carcinoma originates in the main duct, whereas adenoid cystic carcinoma stems from the peripheral duct near the acinar gland. Mucoepidermoid carcinoma is the most common type of malignant salivary duct neoplasm. Three kinds of cells, mucous cells, intermediate cells and epidermoid cells constitute mucoepidemoid carcinoma. The term of *mucoepidermoid carcinoma* was first used by Stewart, Foote, and Becker in 1945.[1)]

In the orginal analysis published in 1945, only two grades were used to distinguish between benign and malignant tumors. Eight years later, Foote and Frazell[2)] modified this grading scheme to include three grades. Low-grade tumors were described as having cystic and glandular components composed of a variety of mature cells, no cytologic pleomorphism, and extremely rare mitosis. Intermediate-grade tumors had a greater proportion of intermediate cells with less cellular variety and, therefore grew in a more solid pattern. The cells showed mild to moderate pleomorphism. High-grade tumors were composed almost entirely of solid tumor islands, and had few cystic or glandular components. Cytologic pleomorphism was widespread and mitotic figures were numerous.[3)] At first biopsy, the tumor was diagnosed as moderately-differentiated squamous cell carcinoma because of diffuse proliferation of epidermoid cells and moderate degree of cellular pleomorphism.

Simpson *et al*[4)] reviewed the literature on mucoepidermoid carcinoma of the maxillary sinus and found a 1% incidence in 12 large series of sinonasal carcinoma. In Japan, Sakai *et al*[5)] reported 8 cases mucoepidermoid carcinoma among 908 cases of sinonasal malignant neoplasms, the incidence being 0.9%. Ogawa[6)] reported three cases of mucoeopidermoid carcinoma among 213 cases sinonasal carcinoma, the incidence being 1.4%. Mucoepidermoid carcinoma of the maxillary sinus is very rare.

It is believed that chronic irritation and decreased blood flow play a role in tumor induction as well. Our case had a past history of nasal polypotomy. It is speculated that she had chronic irritation ; for example, chronic sinusitis.

Facial pain, nasal obstruction and epistaxis are the most common presenting features in this tumor of the maxillary sinus. Orbital involvement presenting as diplopia has occasionally been reported.

CT scan is now routinely used for staging because of the difficulty in evaluating the posterosuperior extension. In this case, a CT scan was very useful in the diagnosis of bone destruction of the maxillary posterosuperior wall, and MRI was useful in determining negation of the skull base and intracranial invasion.

Yamamoto *et al*[7)] reported that serum SCC-Ag levels in well differentiated squamous cell carcinomas of maxillary sinuses were higher than in other maxillary tumors. In this case, serum SCC-Ag levels was extremely high. It is

interesting to note that the high serum SCC-Ag levels in mucoepidermoid carcinomas have not yet been reported.

Regarding treatment and prognosis, it is strongly influenced by the pathological grade. Several authors have reported almost 100% 5-and 10-year survival for low-grade mucoepidermoid carcinomas elsewhere in the head and neck, treated by wide local excision. However, Smith *et al*[8] reported the death of two of four patients with a low-grade central maxillary jaw mucoepidermoid carcinoma treated by simple enucleation. In the sinonasal mucoepidermoid carcinomas studied by Spiro *et al*[9], only 1 of 18 cases was suitable for local excision, whereas the others required either subtotal maxillectomy or radical maxillectomy with orbital exenteration. Therefore it is concluded that even if the tumor is low-grade, a subtotal or total maxillectomy is preferable to simple enuclation or local excision.

For high-grade mucoepidermoid carcinomas of the maxillary antrum, the reported 5-year survival rates of 15% to 33% are less than satisfactory, despite radical combination therapy. Healey *et al*[10] reported five cases of high-grade maxillary mucoepidermoid carcinoma in his series. All were treated with radical maxillectomy, four had adjuvant radiotherapy, and one had orbital exenteration. Yet, four of the five died of disease very early. Thus, prognosis for high-grade mucoepidermoid carcinoma is very poor. Postoperatively, rediotherapy is often recommended for intermediate and high-grade tumors.

Our case had a high-grade mucoepidermoid carcinoma. She underwent the groβ Denker operation, removal of orbita and postoperative irradiation. Recurrence has not been observed to date, but strict observation in the future is necessary.

The rates of both regional and distant metastasis are influenced by the histologic grade, the clinical stage, and the specific site of origin. The distant sites most often involved are lung, skeleton, and brain.

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