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Case Report

A case of sinonasal undifferentiated carcinoma treated without radical resection



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ABSTRACT

Sinonasal undifferentiated carcinoma (SNUC) is a rare and highly aggressive neoplasm of the paranasal sinuses. SNUC is characterized by rapid expansion with high rates of recurrence and metastasis to cervical lymph nodes and distant sites. The patient was a 65-year-old woman who complained of persistent pain at the right maxilla. She had undergone endodontic therapy at the right first molar by her primary dentist. Imaging studies, including computed tomography (CT) and magnetic resonance imaging (MRI), showed a lesion occupying the right maxillary sinus and extending into the nasal cavity and ethmoid sinus. A malignant tumor of the maxillary sinus was suspected, and a biopsy was performed. The lesion was diagnosed as sinonasal undifferentiated carcinoma.

The patient was treated with chemoradiation therapy. After 4 cycles of chemoradiation therapy, CT and MRI showed shrinkage of the lesion and changes consistent with necrosis. Histological examination of a repeat biopsy specimen after the 4 cycles of chemoradiation did not contain tumor cells. 2 more cycles of chemoradiation therapy were added.

Six months after the initial diagnosis, CT finding suggested residual tumor in the ethmoid sinus. We performed another biopsy and the histopathological analysis was negative for tumor. The patient has been followed for 33 months since completing chemoradiation therapy, and there has not been any further evidence of disease.

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1. Introduction

Sinonasal undifferentiated carcinoma (SNUC) is a rare malignant neoplasm of the paranasal sinuses. The age range is broad, and there is a male predominance (2–3:1) [1]. The most common symptoms of SNUC are facial pain, nasal obstruction, epistaxis and proptosis [2]. SNUC grows rapidly, frequently destroying sinus walls and penetrating into the cranial cavity or the oral cavity at an advanced stage. Patients with SNUC experience high rates of recurrence and metastasis to cervical lymph nodes and distant sites [1–4]. SNUC is a highly aggressive malignancy and it has a poor prognosis with a 5-year survival rate of less than 20%. Multimodality treatment, including surgical resection, has been recommended by most authors [1].

There are few prior reports regarding therapeutic strategies for SNUC. Here, we report a case of an SNUC of the maxillary sinus that was treated with chemotherapy by super-selective intra-arterial administration of cisplatin.

2. Case report

A 65-year-old woman was referred to the Department Of Oral Surgery, at our institution in February 2010 complaining of pain at her right maxilla. She had noticed a swelling for a month and had been going for endodontic therapy in the right first molar by her home dentist. She had also been under medical treatment for diabetes and hypertension.

Intraoral examination revealed marked tenderness with diffuse swelling of the gingiva in the right maxillary molar region from the ala of nose. Imaging studies including computed tomography (CT), contrast-enhanced CT, magnetic resonance imaging (MRI), and Gadolinium-enhanced MRI showed a lesion occupying the maxillary sinus and extending into the nasal cavity and ethmoid sinus (Fig. 1A–E). There were no abnormal findings in any other regions and there was no clinical evidence of cervical or distant metastasis. Laboratory data showed no abnormalities.

[☆] AsianAOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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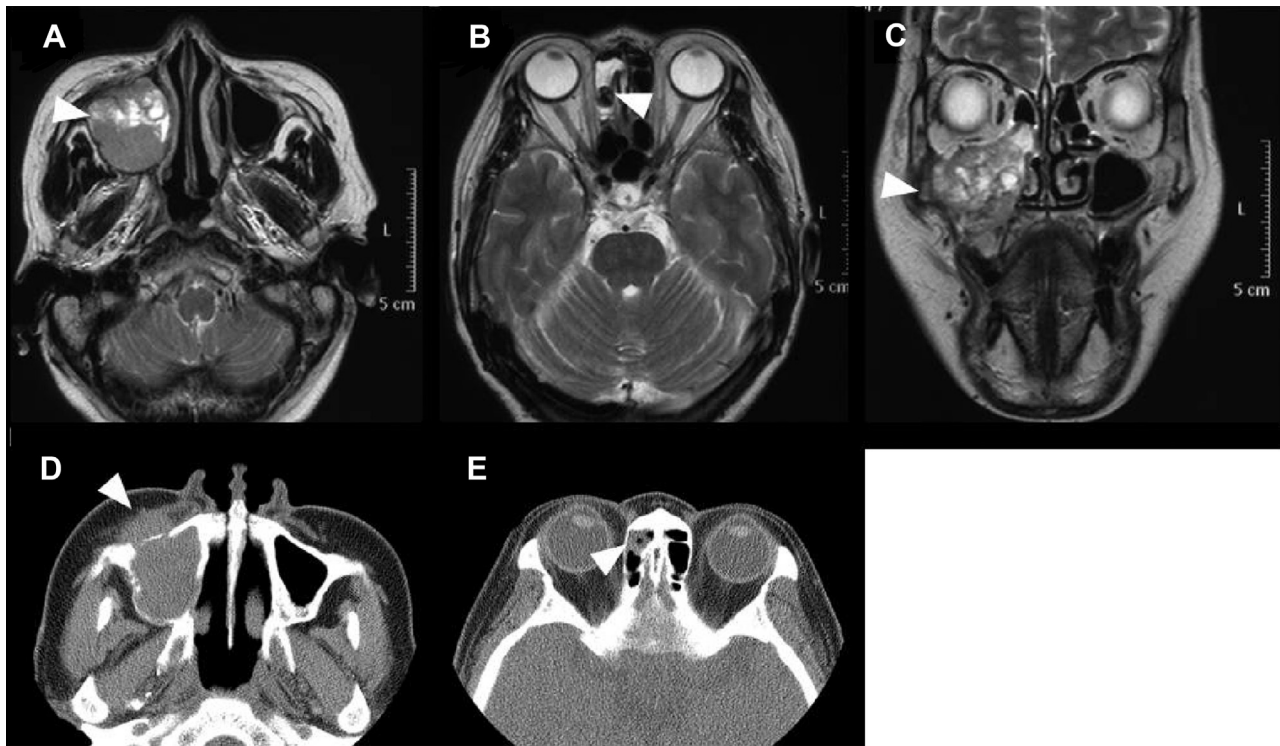


Fig. 1. Imaging studies of the patient before chemoradiation therapy. (A–C) MRI revealed the tumor showed isointensity and hyperintensity intermixed on T2-weighted images. (D and E) CT revealed the lesion filled in the right maxillary sinus with bony erosion and extended to ethmoid sinus and nasal cavity.

The patient underwent a biopsy of the maxillary sinus mass. The histopathological findings revealed invasive growth of alveolate tumor tissue. Neither squamous nor glandular differentiation was found in the nests and sheets of tumor cells. The tumor cells showed a high N/C ratio and nuclear pleomorphism with a marked increase in atypical mitotic figures. There was no evidence of permeation of the tumor into the blood or lymphatic vessels. The histological diagnosis of the biopsy specimen was sinonasal undifferentiated carcinoma classified as T3N0M0 (Fig. 2A and B).

Immunohistochemically, the tumor cells were positive for AE1/AE3 (Fig. 2C), epithelial membrane antigen (EMA), cytokeratin (CK) 7 and vimentin. The Ki67 (Fig. 2D) labeling index was >90%. Leukocyte common antigen (LCA), CD56, chromogranin A, synaptophysin, carcinoembryonic antigen (CEA), human chorionic gonadotropin (HCG), alpha-fetoprotein (AFP), CK20, and Epstein-Barr virus (EBV) were negative (Table 1).

Table 1
Immunohistochemical staining result.

Antibodies	Results
AE1/AE3	Nearly 100% (+)
EMA	(+)
CK7	(+)
Vimentin	(+)
Ki67	(+)
LCA	(-)
Chromogranin A	(-)
Synaptophysin	(-)
CEA	(-)
HCG	(-)
CK20	(-)
AFP	(-)
EBV	(-)

The tumor cells showed positive for AE1/AE3, EMA, CK7 and vimentin. The Ki67 labeling index was >90%. LCA, CD56, chromogranin A, synaptophysin, CEA, HCG, CK20, AFP and EBV were negative in tumor cells.

In March 2010, the patient underwent catheter placement in the right maxillary artery. The catheterization was performed using a microcatheter (Prowler 14, Cordis Neurovascular Inc., USA), which was placed subcutaneously into the superficial temporal artery and advanced into the maxillary artery under fluoroscopic guidance. Catheter placement was confirmed by injection of contrast medium and indigo carmine (Fig. 3).

The patient then underwent super-selective intra-arterial infusion of cisplatin (5 mg/m², 7 mg/body/day, 5 days weekly) via the internal maxillary artery along with external beam radiation therapy (2 Gy/day, 5 days/weekly). The same regimen was repeated for four cycles on a weekly basis.

After this course of chemoradiation therapy, CT and MRI showed shrinkage of the lesion and changes consistent with necrosis (Fig. 4A–D). We performed maxillary antrotomy and resection for definitive diagnosis. Histologically, the biopsy specimen showed inflammatory fibrous connective tissue, without tumor. The patient had achieved a complete response to the chemoradiation therapy.

Two more cycles of chemoradiation therapy of the same regimen were added in lieu of radical resection (total CDDP 189 mg, RT 60 Gy). The patient tolerated the additional chemoradiation well and was discharged on post-chemoradiation therapy day 2.

A month after the treatment, follow-up CT was suggestive of residual tumor in the ethmoid sinus (Fig. 5A and B). We again performed biopsy, and the pathological analysis was negative for tumor. We continued to follow the patient carefully, and by 33 months after the initial chemoradiation therapy she remained well and without any local or regional evidence of tumor recurrence or metastasis (Fig. 5C and D).

3. Discussion

Malignant neoplasms of the paranasal sinuses and nasal cavity including metastatic disease are rare, comprising only 3% of all head and neck malignancies [5]. Among these, SNUC is a highly

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