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Synovial sarcoma of the temporomandibular joint and infratemporal fossa

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ABSTRACT

Objective: Synovial sarcoma in the head and neck region is rare, and is difficult to resect with adequate safety margins because of its anatomical complexity. We herein report our experiences with synovial sarcoma in this region, and review the literature regarding the management of such cases.
Method: We retrospectively examined four cases of synovial sarcoma arising from the temporomandibular joint (TMJ) area and infratemporal fossa.
Result: Only one patient remains alive without disease, while the other three patients have died.
Conclusion: The local control of these tumors has improved because of the progress in the surgical operation methods, while it is expected that there is still a high rate of deaths due to distant metastasis increase. The development of strong chemotherapy is needed for the use after the initial treatment and surgery.

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1. Introduction

Synovial sarcoma reportedly accounts for 14% of all soft tissue sarcomas, but it is an unusual tumor in the head and neck region [1], instead occurring predominantly in the extremities. This pathology is also known to occur in the retroperitoneum, abdominal wall, chest wall and hip-groin area. In the head and neck region, it can occur in the neck, tongue, pharynx, larynx, face, skull base and temporomandibular fossa.

The optimal therapy for synovial sarcoma has not been established, but complete surgical excision with or without radiotherapy and chemotherapy is currently considered the best available treatment [1].

2. Patients and methods

We conducted a retrospective review of patients who underwent surgery at the Department of Head and Neck Surgery, Tokyo Medical and Dental University between 2000 and 2008 to identify the patients with synovial sarcoma arising from the temporomandibular joint (TMJ) or infratemporal fossa. Furthermore, we performed a review of the literature in PUBMED regarding the management of such cases.

3. Cases

3.1. Case 1

In June 2000, a 17-year-old male experienced pain in the left TMJ. He had a checkup with an otolaryngologist, and computed tomography (CT) and magnetic resonance imaging (MRI) identified a tumor in the temporomandibular fossa (Fig. 1A). A biopsy was performed, providing a final diagnosis of synovial sarcoma. Seven courses of preoperative chemotherapy with etoposide, cisplatin and pirarubicin were administered, but the tumor size did not reduce. Tumor excision was then performed, followed by radiotherapy with a total dose of 50 Gy.

Five months postoperatively, multiple lung metastases and local recurrence appeared. CT showed a tumor developing at the foramen ovale, so skull base surgery using a left temporal craniotomy and an orbito-zygomatic approach was planned, which achieved complete removal (Fig. 2). The tissue deficit was filled with an anterolateral thigh flap. Because complete extraction of the primary recurrence was possible, we judged a radical cure to be possible following excision of the pulmonary metastases. The lung metastases were removed surgically, but the lesions relapsed soon after the surgery. The patient received chemotherapy with cisplatin, etoposide and doxorubicin for the metastases. However, this treatment proved ineffective, and he died 62 months after initial presentation, due to lung metastases.

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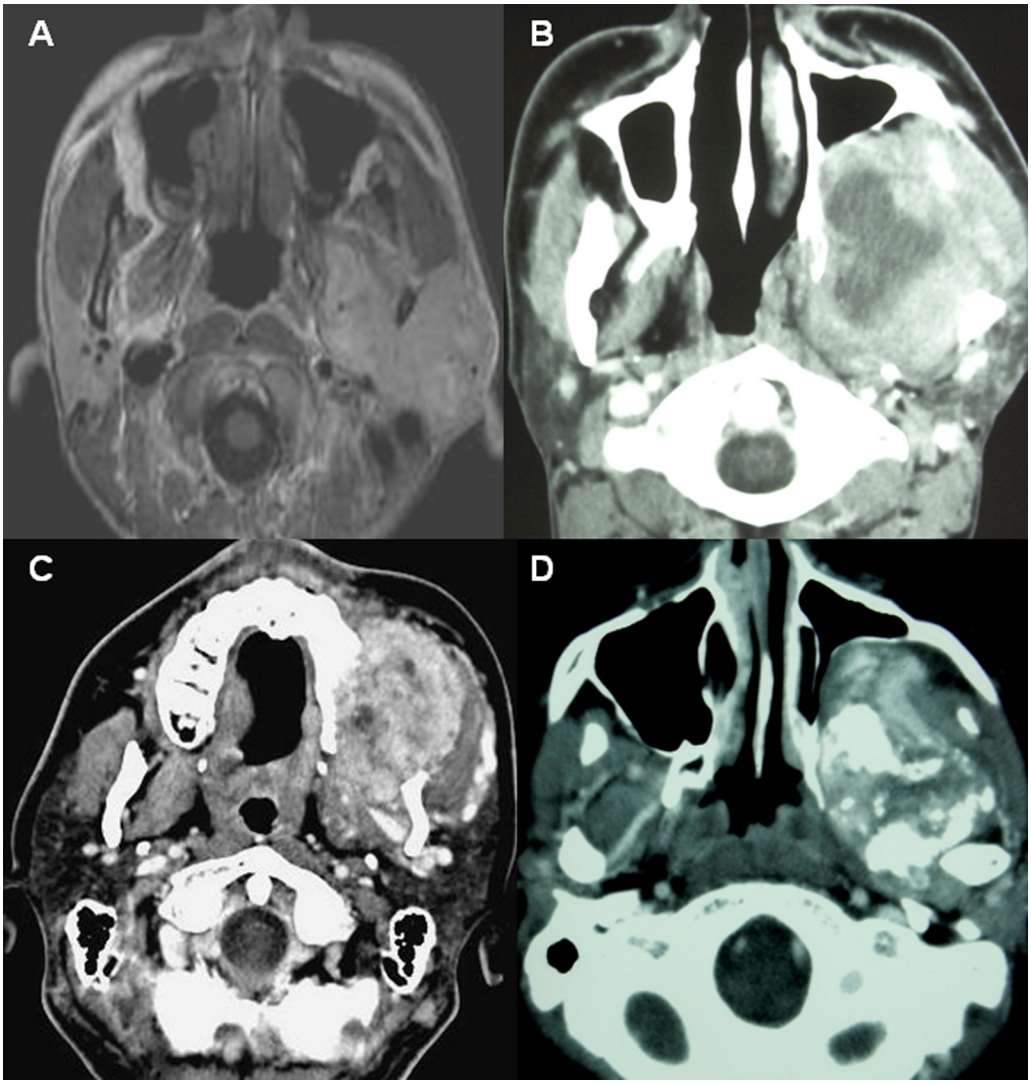

Fig. 1. (A) MRI showing tumor mass of the TMJ. (B) Enhanced axial CT showing the tumor mass of the infratemporal fossa. The maxillary posterior wall and condylar process have been destroyed. (C) Enhanced axial CT showing the tumor mass of the TMJ. The tumor has destroyed the TMJ and mandible. (D) Enhanced axial CT showing tumor mass of the infratemporal fossa. Partial calcification is recognized.

3.2. Case 2

In November 2004, a 27-year-old male developed left subaural pain and swelling. The symptoms improved temporarily, but appeared again in April 2005. This time, the pain slowly worsened, and trismus developed. CT and MRI revealed a large mass from the TMJ to the temporomandibular fossa (Fig. 1B). We suspected a trigeminal schwannoma, and the opening biopsy suggested the possibility of a neurological sheath tumor. We therefore performed surgical excision of the tumor via an orbito-zygomatic approach,

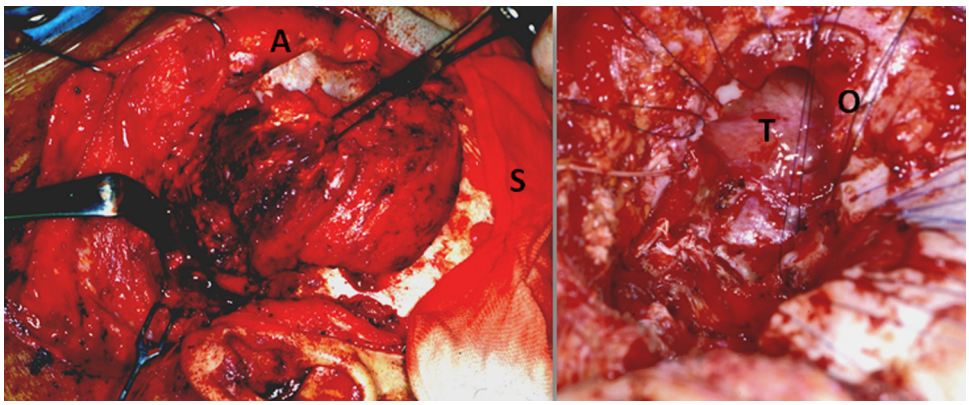

Fig. 2. Craniofacial resection by both lateral craniotomy and infratemporal fossa approach. The tumor destroyed the foramen ovale along the trigeminal nerve and infiltrated the base of skull. A: anterior; S: superior; T: tumor; O: foramen ovale.

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