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Rhabdomyosarcoma of masticator space

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ABSTRACT

A 16-year-old female was admitted to Wonkwang dental hospital with a chief complaint of painful ulceration on right buccal mucosa around mandibular 3rd molar area.

Computed tomography and magnetic resonance imaging showed relative large soft tissue mass on the infratemporal fossa and masseter muscle region. By the feature of T1-weighted and T2-weighted of MR imaging, we suspected this mass as a kind of myogenic sarcoma. Histopathological and immunohistochemical studies established a definitive diagnosis of embryonal rhabdomyosarcoma. A review of the literature was also presented. ***(Korean J Oral Maxillofac Radiol 2001; 31 : 241-5)***

KEY WORDS : rhabdomyosarcoma, embryonal; masseter muscle; magnetic resonance imaging

The reported frequency of soft tissue tumor among Korean children tumor was 4.2% in 1992, 6.2% in 1993 and 4.8% in 1994 by Ministry of Health and Welfare, Korea. And the malignant myogenic tumor patient accounted for 4.4% of the solid tumors treated in department of pediatric surgery, Seoul National University, between 1978 and 1997.

WHO defined rhabdomyosarcoma as a highly malignant tumor of rhabdomyoblasts in varying stages of differentiation with or without cross-striation.[1]

There has been little doubt that rhabdomyosarcoma is not only the most common soft tissue sarcoma of children under 15 years of age but also of adolescents and young adults.[2]

According to Department of Pediatrics at the Seoul National University Children's Hospital, rhabdomyosarcoma occurs 40-45 cases/year, most of them between 2 and 4 years of age in Korea. It has a predilection for males, more frequent in the white race and less in orientals and Afro-Americans.[3]

Although rhabdomyosarcoma may arise anywhere in the body, they may be found predominantly in three regions (1) the head and neck, (2) the genitourinary tract and retroperitoneum, and (3) the upper and lower extremities.

Most of cases can be classified in one of four histological categories (embryonal, alveolar, botryoid and pleomorphic) in accordance with a classification suggested and defined by Horn and Enterline[4] in 1958.

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Histogenesis is still undocumented. but Enzinger and Weiss[2] proposed two histogenetic possibilities for rhabdomyosarcoma : (1) primitive and undifferentiated mesenchyme origin and (2) embryonal muscular tissue origin. Genetic factors, previous radiation treatment and viruses have been mentioned in the literature as possible trigger mechanism of proliferation of embryogenic mesencymal tissues.[5]

The treatment for rhabdomyosarcoma has evolved from surgical resection in the past to multimodality treatment with surgery, radiation and chemotherapy.

The intergroup rhabdo-myosarcoma study in United States, recently reported overall 5-year survival rates of 71% and 27% for non metastatic and metastatic patient, respectively.[6] In Korea, Pediatric Surgery at the Seoul National University

Fig. 1. Intraoral view showed ulceration on right buccal mucosa around mandibular 3rd molar area.

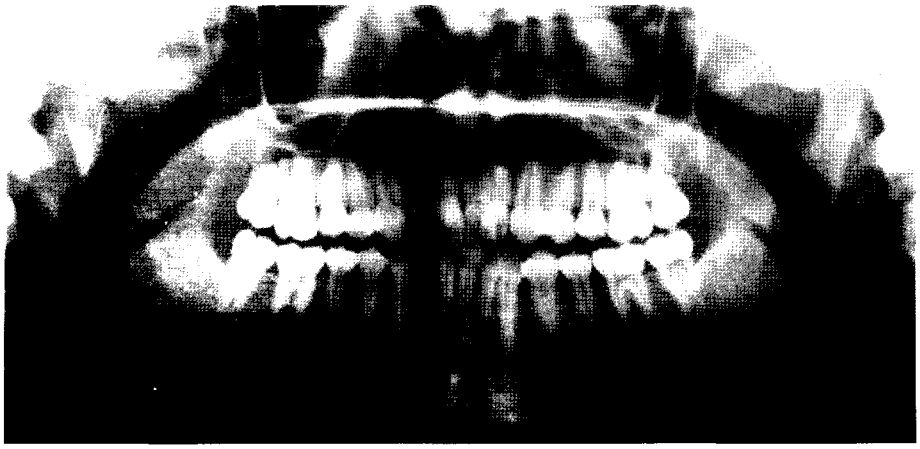

Fig. 2. Panoramic radiogram showing a osteolytic defect on the right sigmoid notch and upper ramus.

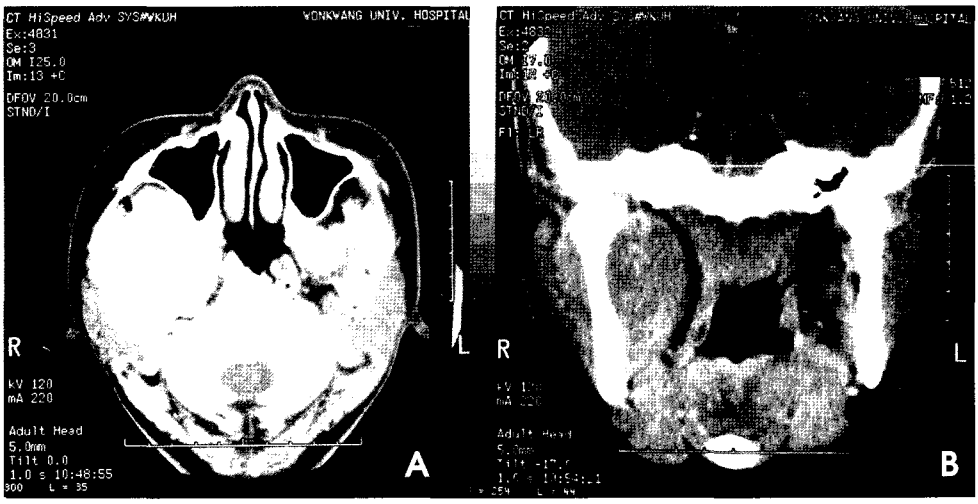

Fig. 3. Axial (A) and coronal (B) CT scan shows soft tissue mass of masticator space. The tumor mass was surrounded by maxillary sinus antero-medially, buccal space laterally, oropharynx medially and parotid gland postero-laterally.

Children's Hospital reported 5-year survival rates of 60% in 1998.

In this report we present a case of a 16-year-old female with an diagnosis of embryonal rhabdomyosarcoma of masticator space, involving infratemporal space and masseter muscle.

Case report

A 16-year-old female was admitted to Wonkwang dental hospital with a chief complaint of painful ulceration on right buccal mucosa around mandibular 3rd molar area (Fig. 1) and swelling on right midface area. These symptoms developed about two weeks ago. She has complained for mouth opening limitation and pain around mandibular 3rd molar area. She didn't have a specific medical and dental history except otitis media operation about ten years ago.

Panoramic radiogram revealed radiolucent defect on sigmoid notch and upper ramus of right mandible. The margin was well defined, but not corticated (Fig. 2).

Computed Tomograms of the lesion showed the large, round mass of masticatory space occupying masseter and pterygoid muscle area, and showed the destruction of right mandibular ramus and compression of posteriolateral wall of right maxillary sinus. The lesion was surrounded by buccal space, parapharyngeal space, parotid gland. On coronal view of computed tomogram, the lesion extended from mandibular angle to skull base level (Fig. 3).

On T1-weighted images of MR, the lesion showed the great part of homogeneity except focal heterogeneity of necrotic portion. The lesion showed similar intensity to adjascent muscle signal and contrast enhancement. The margin of the lesion from surrounding tissues was almost distinct except the parotid gland area (Fig. 4).

T2-weighted images of tumor showed slight higher signal intensity than muscle intensity.

Multiloculated appearance divided by septa and hyperintense necrotic area were also observed on T2-weighted image (Fig. 5).

Fig. 4. A, Axial T1-weighted image of MR shows the tumor mass signal to be similar intensity in comparison with muscle. B, axial enhanced T1-weighted images shows contrast enhancement of mass except necrotic portion.

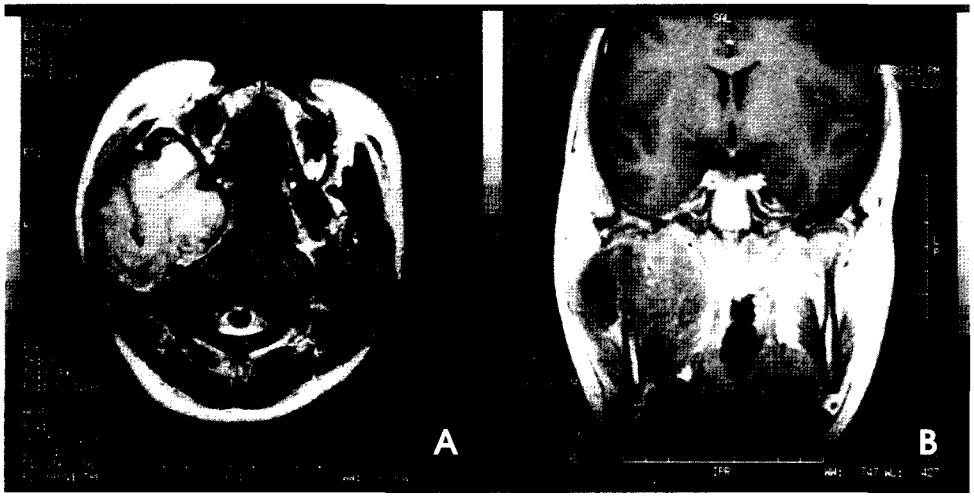

Fig. 5. A, Axial T2-weighted image, necrotic area represents high signal intensity. B. coronal T2-weighted image, the tumor is located below the skull base superioly. but thers is no evidence of invasion to meninges.

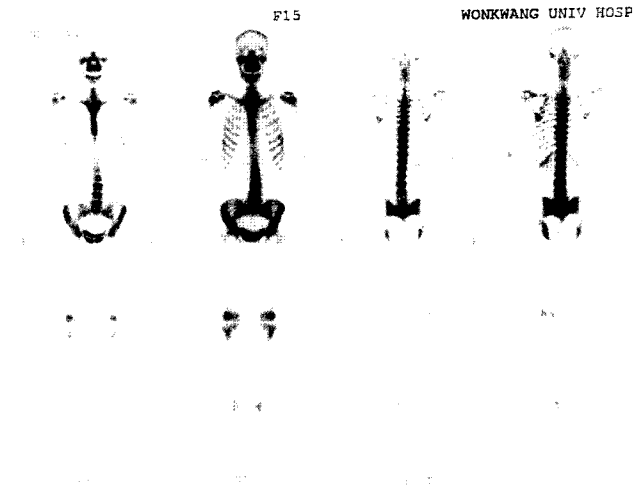

Fig. 6. ^{99m}Tc MDP Bone Scintigram. Cold spot in right maxilla and mandibular area, but otherwise is not remarkable.

Bone scan of whole body with ^{99m}Tc-MDP pharmaceuticals didn't show possibility of metastasis to other skeletal tissues (Fig. 6).

Histopathological finding showed the infiltration of tumor cells into fibro-connective tissues, small round cells with hyperchromatic nuclei and eosinophilic cytoplasm. Giant cells and cohesiveness loss of tumor cells were also observed, but the appearances of characteristic rhabdomyoblasts and cross striation were not found (Fig. 7).

Immunohistochemically, the tumor cell stained positively for desmin, vimentin and skeletal myosin, but didn't stain for S-100, LCA, Cytokeratin and MIC-2.

Discussion

Rhabdomyosarcoma has been known as a rare common sarcoma, chiefly occuring in the head & neck, predominant in

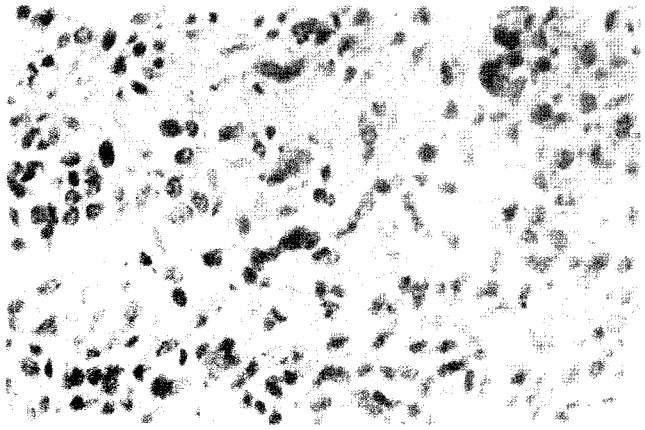

Fig. 7. Small round cells with hyperchromatic nuclei and eosinophilic cytoplasm. rhabdomyoblsts are not observed (H-E stain. × 200).

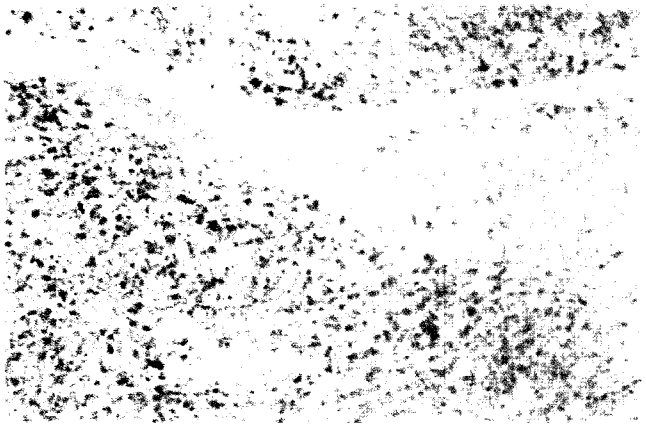

Fig. 8. Immunohistochemical finding shows positivity for desmin (× 100).

infancy and childhood. This sarcoma can be classified into four histological categories as embryonal, botryoid, alveolar and pleomorphic type. Prognosis of rhabdomyosarcoma had been known poor, but effective therapeutic methods have improved the prognosis.[7]

Panoramic radiogram of our case demonstrated pressure-type erosion of sigmoid notch and coronoid process of right mandible. By the appearance of panoramic view, we once thought the mass of the lesion originated in buccomasseteric region (masseter muscle, buccinator muscle, parotid duct, buccal fat pad and subcutanous tissues of cheek and the parotid gland). The pathologic condition of the buccomass eteric mass were myopathies, benign masses such as hemangioma and lipoma, inflammatory processes, and malignant masses.[8]

The finding of CT and MRI suggested evidence of a large soft-tissue mass arising from the infratemporal fossa extending to the pterygopalatine fossa with destruction of mandibular ramus and pressure erosion of posterior wall of right maxillary sinus.

Disease within the infratemporal fossa are usually infections or neoplasms.[9] In the present case, the well demarcated border of the mass noted on the CT scan suggested slow growth, most compatible with a benign neoplasm. Benign neoplasm in the infratemporal fossa proximal to the muscle of mastication may produce dull pain that increases with mouth opening.[10]

By this appearance of CT, benign mesenchymal neoplasm, benign epithelial neoplasm and salivary gland tumors could be considered. Malignant neoplasm such as sarcomas and carcinomas), lymphoma and metastatic foci from distant sites could also be remotely considered.

MR imaging is an important modality in the evaluation of soft tissue masses and make possible of differential diagnosis.

Hemangioma and lymphangioma had specific appearance on MR imaging, with an iso-intense signal in T1-weighted images and high signal intensity on T2-weighted images.[11,12] In the present case, the mass showed a moderate signal intensity on the T1-weighted images and moderately high intensity on the T2-weighted images.

Lipoma also had a characteristic appearance because of similarity to that of subcutanous fat on CT images[8] and high signal intensity on both T1- and T2- weighted images.[13]

We could exclude our case from hemangioma, lymphangioma and lipoma differential diagnosis by above facts and relative large size of this case.

In the radiologic evaluation of this case, large mass size suggested malignant tumor due to rapid growth, similar intensity of mass to muscle intensity on MR suggested myogenic sarcoma, the necrotic portion and contrast enhancement on MR image gave the impression of soft, solid tissue sarcoma.

The markers that were positive in immunohistochemical studies were vimentin and desmin. These two markers together could localize all tumors of myogenic origin.[14,15]

We concluded this tumor as embryonal rhabdomyosarcoma originated from the right pterygoid or masseter muscle by the radiographic finding of CT and MR, immunohistochemical finding. The relative large mass size and charateristic MR imaging give the base in diagnosing of rhabdomyosarcoma. This patient is now under surgical and chemotherapeutic course.

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