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Ki-1-Positive Anaplastic Large Cell Lymphoma of the Masticator Space with Intracranial Extension

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Summary: Ki-1-positive anaplastic large cell lymphoma (Ki-1 ALCL) is a recently recognized form of non-Hodgkin lymphoma occurring predominantly in the skin and/or peripheral lymph nodes. We report the clinical, pathologic, and radiographic findings of a case of Ki-1 ALCL arising within the masticator space and extending intracranially into the epidural space.

Index terms: Lymphoma; Mouth, neoplasms

The masticator space is a fascial compartment containing essentially the muscles of mastication, the mandibular ramus, and various neurovascular structures. Neoplasms involving the masticator space may result primarily from tumors arising *de novo* within the masticator space or secondarily from either the direct extension of a neoplasm involving a structure bordering the masticator space or from distant metastasis (1, 2). Primary *de novo* neoplasms of the masticator space are uncommon, and include benign tumors, such as hemangiomas and lipomas, as well as malignant tumors, such as malignant schwannomas and various sarcomas (2, 3). Direct extension of tumors involving structures bordering the masticator space is the most common form of neoplastic involvement. Neoplasms involving the masticator space through direct extension include squamous cell carcinomas of the upper aerodigestive tract and skin, major and minor salivary neoplasms, diffuse histiocytic lymphomas, and adenocarcinomas (2, 3).

Ki-1-positive anaplastic large cell lymphoma (ALCL) is a recently recognized form of non-Hodgkin lymphoma. We report the clinical, pathologic, and radiologic findings in a patient with Ki-1 ALCL arising within the masticator space and extending intracranially into the epidural space.

Case Report

A 31-year-old human immunodeficiency virus (HIV)-positive man had a 1-month history of left-sided facial swelling and pain. The patient assumed his symptoms were of an odontogenic nature and went to a dental clinic for possible treatment. On physical examination, the patient was noted to have swelling of the soft tissues of the left side of the face and temporal region, left-sided facial trismus, and a decreased range of mandibular movement. The remainder of the head and neck examination was unremarkable except for a number of palpable lymph nodes and a small amount of dry blood in the left external auditory canal.

Preliminary panoramic radiographs obtained at the dental clinic showed destruction of the left temporal bone. Subsequent contrast-enhanced axial and direct coronal computed tomographic (CT) scans showed a mildly enhancing, infiltrating, soft-tissue mass involving the left masticator space extracranially, and appearing intracranially as an epidural mass in the left middle cranial fossa (Fig 1A). The tumor was noted to abut the left pterygopalatine fossa (Fig 1B and C). Tumoral extension into the left sphenoidal sinus was also evident (Fig 1B). Extensive destruction of the squamosal segment of the left temporal bone (floor of the middle cranial fossa), the greater wing of the sphenoid bone, and medial segments of the temporomandibular joint (glenoid fossa and condylar head) were evident on both axial and direct coronal CT sections as well as on three-dimensional reformatted images (Fig 1A-D). Partial destruction of the zygomatic process of the left temporal bone was also noted (Fig 1B and C). CT examinations of the chest and abdomen, obtained to rule out any visceral abnormalities, were unremarkable except for mild splenomegaly.

In light of the significant involvement of the floor of the middle cranial fossa and intracranial extension of the tumor, a neurosurgical consultation was obtained. The patient subsequently underwent a left frontotemporal craniotomy to obtain material for histopathologic diagnosis. At the time of surgery, an infiltrating mass involving the left temporalis muscle and skull base was noted. Intracrani-

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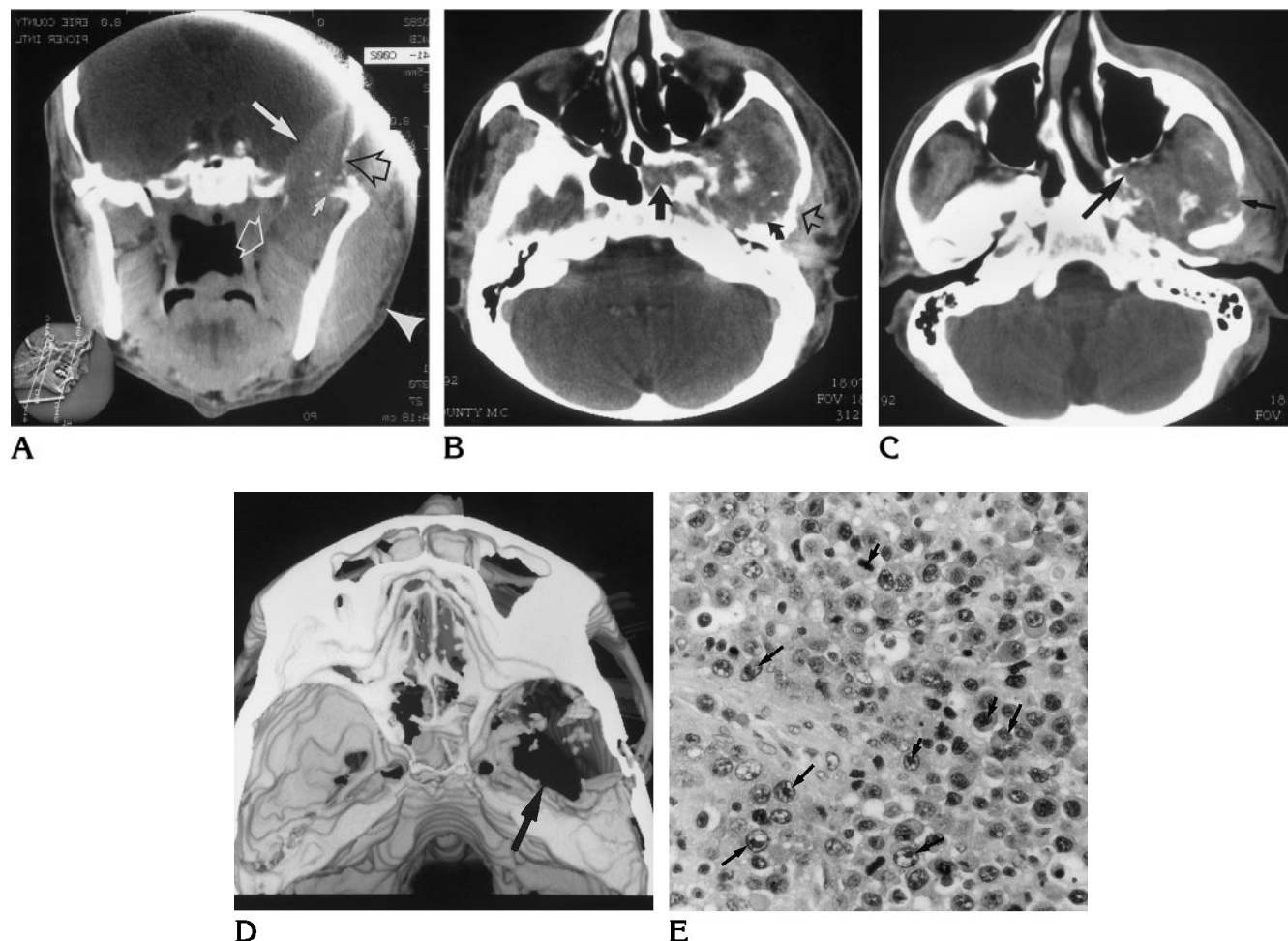


Fig 1. A, Contrast-enhanced direct coronal CT scan shows the neoplastic involvement of both the lateral (*solid white arrowhead*) and medial compartments (*open white arrow*) of the left masticator space, destruction of the floor of the middle cranial cavity, and epidural extension (*long white arrow*). Tumoral involvement of the left temporomandibular joint has resulted in destruction of the glenoid fossa and medial aspect of the condylar head (*short white arrow*). Destruction of the temporal bone (*open black arrow*) immediately above the joint is also noted.

B, Contrast-enhanced axial CT scan through midface at level of left zygomatic arch shows neoplastic involvement of the left masticator space, extension into the left sphenoidal sinus (*solid straight arrow*), destruction of the left glenoid fossa (*curved arrow*), and erosion of the zygomatic process of the temporal bone (*open arrow*).

C, Contrast-enhanced axial CT scan, 5 mm caudal to B, shows destruction of the left glenoid fossa, destruction of the inferior surface of the zygomatic process of the temporal bone (*small arrow*), and tumoral extension lateral to the pterygopalatine fossa (*large arrow*).

D, Three-dimensional view of skull interior shows the full extent of osseous destruction of the floor of the left middle cranial fossa (*arrow*).

E, Photomicrographs of biopsy specimen shows a loosely cohesive, pleomorphic cell population (*long arrows*). Nuclear enlargement and pronounced pleomorphism are also evident (*short arrows*) (original magnification $\times 40$).

ally, this mass was located epidurally and was densely adherent to the dura.

Histologic examination of the biopsy specimen revealed a tumor composed of sheets of large pleomorphic cells containing amphophilic cytoplasm and large vesicular nuclei with abundant prominent macronucleoli (Fig 1E). The cytoplasmic borders were poorly defined and mitotic figures were frequent in places; tumor cells were arranged in a somewhat epithelioid pattern. At other locations they infiltrated the skeletal muscle. A few foci of fibrosis accompanied newly formed bone. Immunohisto-

chemical assays performed on paraffin-embedded sections showed tumor cells that were strongly positive for Ki-1 (CD-30) and T-cell marker VCHL-1 (CD-45 RO), focally positive for leukocyte-common antigen (CD-45 RB), and positive for epithelial membrane antigen (EMA) in some cells. The following immunohistologic assays were negative: the B-cell membrane 2 G (CD-20), the myelocytic and monocytic markers Leu M1 (CD-15), Leu-5, cytokeratin, and the malignant melanoma marker (HMB-45). These monoclonal antibody markers com-

bined with the tumor histology were compatible with Ki-1 ALCL.

Discussion

Ki-1 ALCL is a recently recognized form of non-Hodgkin lymphoma (1, 4–14) that is characterized by large pleomorphic tumor cells, expression of lymphocyte activation antigens (CD-30, CD-25, CD-71, Ia), and a nonrandom chromosome abnormality (10). Ki-1 ALCLs are usually of T-cell phenotype (60% to 70%) and estimated to represent between 1% and 8% of all non-Hodgkin lymphomas (4, 6). The majority of Ki-1 ALCLs reported in the literature involve either the skin and/or peripheral lymph nodes. Less common is involvement of the gastrointestinal tract, female genital tract, bone, and bone marrow (5, 7, 10). A review of the literature fails to disclose any case reports of Ki-1 ALCL involving any of the major fascial spaces or structures of the head and neck.

Although lymphomas are the most common nonepithelial neoplasms of the head and neck, this case is unique from a number of perspectives, including the type of neoplasm involved, the extensive osseous involvement, the apparent site of origin of the neoplasm, and the clinical presentation. The definitive diagnosis of Ki-1 ALCL is based on various histologic and immunochemical findings. However, we believe the radiologist should be aware of its potential to occur in the head and neck region and that it should be included in the differential diagnosis of neoplastic processes occurring with either atypical clinical presentation and/or unusual radiologic findings. We think this is important, because, if one relies solely on histologic findings, Ki-1 ALCL can be mistaken for a variety of poorly differentiated tumors, such as malignant melanoma, rhabdomyosarcoma, metastatic carcinoma, malignant histiocytic tumors, lymphocyte-depleted Hodgkin lymphoma, and non-Hodgkin lymphoma (4, 7, 12). It is hoped that the inclusion of Ki-1 ALCL in the differential diagnosis, when appropriate, will alert both the clinician and pathologist to the importance of using immunomarkers in the diagnosis of Ki-1 ALCL.

The radiologic findings in the present case are atypical for a lymphoma. CT findings most often seen in head and neck lymphomas include multiple extranodal sites of involvement; multiple large, bulky, nonnecrotic lymph nodes;

and minimal bone destruction (15, 16). The radiologic findings, especially the bone destruction, are more consistent with a squamous cell carcinoma secondarily involving the masticator space followed by intracranial extension.

Although it is possible that the Ki-1 ALCL arose within the osseous floor of the middle cranial fossa and then secondarily involved the masticator and epidural spaces, we believe this is unlikely given the extracranial location of the bulk of the tumor mass within the masticator space and the pattern of bone destruction seen in the floor of the middle cranial fossa. The pattern of bone destruction seen in this case is identical to that seen in other tumors arising extracranially and destroying the skull base.

Although there is a greater preponderance of lymphomas among persons who are HIV-positive, none of the series reported in the literature describes a connection between Ki-1 ALCL and HIV positivity. Additional investigation into this possible relationship is needed to determine fully what, if any, association may exist.

The present case represents an instance of Ki-1 ALCL in the head and neck. The purpose of this article is to alert the reader to the potential of Ki-1 ALCL to occur in the head and neck and to the need to include it when formulating a differential diagnosis for a neoplasm with non-specific findings. We hope that this isolated case will lead to a heightened awareness of the possible occurrence of this neoplasm in the head and neck and to the possible discovery of other cases. Information gained from other cases will be useful in fully assessing the biological behavior of this tumor as well as in formulating appropriate therapies.

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