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Temporalis Muscle Metastasis of the Uterine Leiomyosarcoma: A Case Report

Uterus Leyomiyosarkomunun Temporal Adele Metastazı: Olgu Sunumu

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ABSTRACT

Leiomyosarcomas are malignant tumors of smooth muscle origin. These tumors are very rare in the head and neck region. The majority of leiomyosarcomas of the head and neck arise in the paranasal sinuses, oral cavity, jaws and superficial soft tissues like the scalp. A mass was observed in the right temporoparietal region of the scalp of a 76-year-old female. Two years before presentation, the patient was operated for primary uterine leiomyosarcoma. Over the ensuing years, the mass substantially increased in size. Radiologic findings revealed a mixed density mass of the right temporalis muscle. Histologic and immunohistochemical examination of the tumor showed a malignant mesenchymal neoplasm consisting of spindle-shaped atypical mesenchymal cells with marked pleomorphism and central cigar shaped nuclei arranged in fascicles. We report a rare case of uterine leiomyosarcoma metastatic to the temporalis muscle, proven by histopathology.

KEY WORDS: Leiomyosarcoma, Sarcaoma, Temporalis muscle, Uterus

ÖZ

Leyomiyosarkomlar düz kas kaynaklı malin tümörlerdir. Bu tümörler baş ve boyun bölgesinde çok nadirdirler. Baş ve boyun leyomiyosarkomlarının büyük bir kısmı paranazal sinüsler, oral kavite, çene ve skalp gibi yumuşak dokulardan kaynaklanır. 76 yaşında bayan hastanın sağ temporopariyetal bölgesinde bir kitle gözlendi. Başvurudan 2 yıl once hasta primer uterus leyomiyosarkumundan opere edilmişdi. Kitle ilerleyen yıllarda giderek büyümüş. Radyolojik bulgular sağ temporal adelede miks dansitede bir kitle gösterdi. Histolojik ve immünhistokimyasal değerlendirmede; bantlar halinde yerleşmiş belirgin pleomorfizm ve santral çubuksu şekilli çekirdekler gösteren iğsi atipik mezenkimal hücreler içeren malin bir mezenkimal neoplazi tespit edildi. Temporal adeleye uterus leyomiyosarkomunun metastazı olan nadir bir olguyu histopatolojik kanıt ile sunmaktayız.

ANAHTAR SÖZCÜKLER: Leyomiyosarkom, Sarkom, Temporal adele, Uterus

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INTRODUCTION

Leiomyosarcoma of the head and neck region is very uncommon and only a few cases at various anatomic sites such as the sino-nasal tract, oral cavity, mandible, larynx, trachea, hypopharynx, cervical lymph node, external auditory canal and middle ear have been mentioned in the literature (6,11,16). Metastatic leiomyosarcomas are also extraordinary in general (1). This case report identifies an unusual temporalis muscle metastasis of uterine leiomyosarcoma treated successfully by a combination of radical surgery and radiotherapy. The case reported here is the first reported case of uterine leiomyosarcoma metastatic only to the temporalis muscle in the English-language literature.

CASE REPORT

A 76-year-old female presented with a gross mass in the right temporal region. The patient was free of the neurological and physical symptoms on admission. Two years before presentation, she had operated for a primary uterine leiomyosarcoma. She had not received any post-operative radiotherapy or chemotherapy. Physical examination revealed a gross mass, nearly 10 cm in maximum diameter in the right temporal region of the scalp. It was painless and immobile. There was no regional lymphadenopathy. Neurological examination was normal. Cranial computed tomography (CT) scans (Figure 1A) and cranial magnetic resonance imaging (MRI) showed a huge mass in the right temporal region invading the temporalis muscle. The metastatic lesion showed heterogeneous enhancement after gadolinium injection (Figure 1B). She was operated on and the mass was totally excised together with the temporalis muscle. No cranial or scalp invasion of the tumor was observed during surgery. There were no complications during surgery and the recovery as uneventful. Macroscopically, the tumor was 9x7x4 cm in size, smooth and solid. On histopathological evaluation, it was composed of spindle-shaped atypical mesenchymal cells, which showed 10-12 mitoses/10 high power fields with marked pleomorphism and central cigar shaped nuclei arranged in fascicles (Figure 2A). Immunohistochemical preparations showed only expression of smooth-muscle actin, vimentin and weakly desmin (Figure 2B). The tumor morphology was interpreted as high-grade (III) leiomyosarcoma. Postoperative external radiotherapy was administered to the metastatic site using the 8 Mev electron beam. Total tumor dose was 60 Gy, which was given in 30 fractions. No acute side effects to radiotherapy were seen. The patient is currently asymptomatic and without evidence of any disease nearly 3 years after the diagnosis of her temporalis muscle metastasis (Figure 3).

DISCUSSION

Leiomyosarcoma, a malignant smooth muscle tumor, usually arises in the uterus, the digestive tract, or the retroperitoneal area (2). Other origins such as major arteries, veins, viscera and extremities are less common sites. Leiomyosarcoma of the head and neck region is very uncommon and only a few

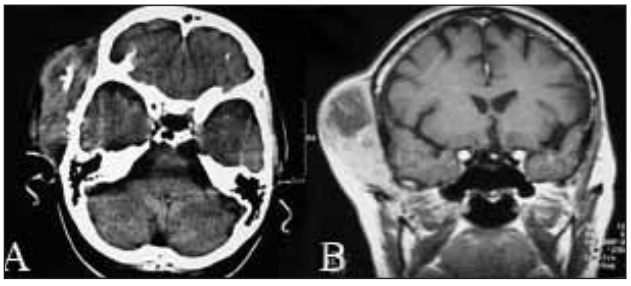

Figure 1: *(**A**) A computed tomography scan demonstrates a heterogeneous and calcified mass in the right temporalis muscle. (**B**) Contrast-enhanced coronal T1-weighted magnetic resonance image shows 7X4 cm mass with heterogeneous enhancement in the right temporal region.*

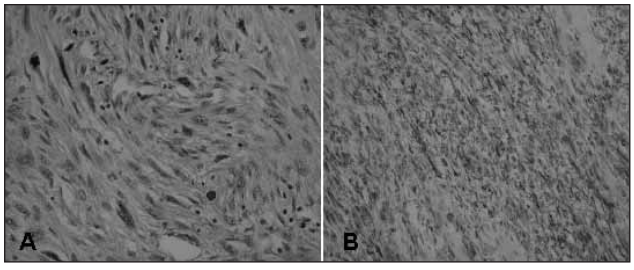

Figure 2: *(**A**) Photomicrograph depicting tumor with atypical spindle cells arranged in interlacing fascicular pattern (Haematoxylin -eosin stain, 200x). (**B**) Consistent smooth muscle actin (SMA) immunoreactivity is evident in most tumor cells (Immunoperoxidase, 200x).*

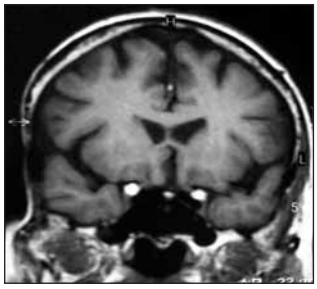

Figure 3: *Contrast-enhanced, T1-weighted coronal MRI scan obtained nearly 3 years after surgery demonstrated no tumor recurrence.*

cases at the various anatomic sites such as the sino-nasal tract, oral cavity, mandible, larynx, trachea, hypopharynx, cervical lymph node, external auditory canal and middle ear have been mentioned in the literature (6,11,16).

Although the origin of the leiomyosarcoma was thought to be the smooth muscle cell, several authors have proposed that this tumor may in fact be derived from pleuropotential, uncommitted mesenchymal cells or smooth muscle cells of blood vessel origin (2,9). This implies that the origin of our tumor was a small blood vessel within the uterus. The tumor might also have originated in the myoepithelial cells that are capable of multidirectional differentiation.

Leiomyosarcoma is apparently not a lesion with a high potential for distant metastases. Metastatic spread of leiomyosarcoma as observed in 20% of cases is usually via the hematogenous route to lungs. The lungs were radiologically uninvolved in our patient. The alternative pathway could be spread via Batson's plexus, a route of hematogenous metastasis seen in various malignancies, which bypasses the pulmonary circulation (7). A possible mechanism for the temporalis muscle metastasis without pulmonary metastasis in our patient may be hematogenous metastasis via Batson' plexus.

Surgical resection is the main treatment method for soft tissue sarcoma. The margin of the excision should be at least 1 cm in all directions when surgery is used alone. When a combination of surgery and radiotherapy is used, this margin can be reduced to approximately 0.5 cm (4). En block resection and achieving these margins at all tumor plans is almost impossible in the head and neck region, because of the proximity of adjacent neurovascular structures or vertebral column. We easily resected the lesion with the temporalis muscle in the presented case. Invasion of the tumor to the underlying skull was not observed intraoperatively. The case presented here is the first reported uterine leiomyosarcoma metastasis to the temporalis muscle.

There are no randomized trials for head and neck soft tissue sarcomas. Three prospective randomized trials that have compared surgery alone with surgery and radiation clearly show the effectiveness of adjuvant radiation in soft tissue sarcomas of extremities (10,13). Preoperative or postoperative choice of external-beam irradiaton is still a question for soft tissue sarcomas as there is are supportive data. Decreased intraoperative seeding of tumor cells and tumor shrinkage facilitating surgery are the potential advantages of preoperative external-beam radiation therapy. Suit et al. showed that preoperative radiation was superior to postoperative radiation in terms of local control for patients with tumors greater than 15 cm (13). However, others have shown no difference (3).

Vandergriff et al. has documented the cutaneous metastasis of leiomyosarcoma well (15). In his report, the scalp was the most common cutaneous site of involvement of metastatic leiomyosarcoma. Only five cases of leiomyosarcoma metastasis to the skull have been previously reported (5,8,12,14,17). In our patient, the tumor was confined only to the temporalis muscle. Distant metastases including muscle lesions, as reported in our patient, from various primary malignancies are usually associated with widespread disease and carry an extremely poor prognosis. The present patient responded excellently to the combination of surgery and radiation therapy and currently has no evidence of disease nearly 3 years after the diagnosis of her temporalis muscle metastasis of uterine leiomyosarcoma. Proper history-taking, and precise clinical and radiological evaluation together with a biopsy are necessary to establish the diagnosis. The combination of wide surgical resection and postoperative adjuvant external-beam radiation therapy may be the most effective means of treatment for metastatic leiomyosarcomas.

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