



Case Report

Inflammatory Myofibroblastic Tumor Mimicking Meningioma: An Extraordinary Case

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Abstract

Thirty-six year-old female patient was admitted to the hospital with the complaints of increasing numbness in left arm and leg, headache and vomiting for one month. In her physical examination, 1/5 strength loss in left arm and leg and disestesya were detected. In the computerized tomography (CT) an intracranial tumor was showed in right parietal lob and in the magnetic resonance imaging (MRI) hypointense was found in T1 weighed section and a tumor lesion causing hyperintense, lobule contured, located extraaxially, putting pressure on neural tissue, contrasted homogenously and leading to oedema effect on neural tissue in right parietal lob was detected in T2 weighed section. The lesion was totally excised with dura. Histopathologic diagnosis was reported as inflammatory myofibroblastic tumor. In the post-operative first month control, left hemiparesis was disappeared, however disestesya was continuing.

Keywords: Inflammatory myofibroblastic tumor, meningioma, intracranial

Meninjiomu Taklit Eden İnflamatuvar Miyofibroblastik Tümör: Sıradışı Bir Olgu

Özet

Otuzaltı yaşında kadın hasta bir ay önce başlayan ve giderek artan sol kol ve bacakta uyuşma, baş ağrısı ve kusma şikayetleri ile başvurdu. Muayenesinde sol kol ve bacağına 1/5 güç kaybı ve dizestezi vardı. Bilgisayarlı beyin tomografisi (BT), sağ paryetalde intrakraniyal kitle ile uyumlu olması üzerine yapılan manyetik rezonans (MR) görüntülemeye sağ paryetalde bileşkede T1 ağırlıklı kesitlerde hipointens, T2 ağırlıklı kesitlerde hiperintens, lobüle kontürlü, ekstraaksiyal yerleşimli, nöral dokuya bası yapan, homojen kontrastlanan ve etraf nöral dokuda ödem etkisine yol açan kitle lezyonu vardı. Lezyon durayla beraber total eksize edildi. Histopatolojik tanı inflamatuvar miyofibroblastik tümör olarak bildirildi. Postoperatif birinci ay kontrolünde olgunun sol hemiparezisinin düzeldiği fakat dizestezinin devam ettiği gözlemlendi.

Anahtar Kelimeler: İnflamatuvar miyofibroblastik tümör, meninjioma, intrakranial

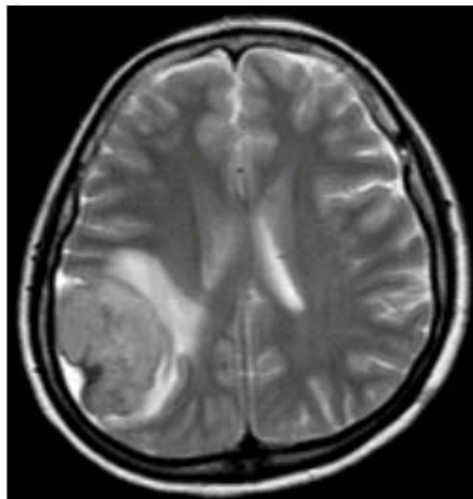
INTRODUCTION

Inflammatory myofibroblastic tumor (IMT) is a rare tumor which is originating from myofibroblastic fusiform cells and inflammatory cells.⁽¹¹⁾ Predominantly, this lesion locates at lung and it may be seen at orbita, retroperitoneum, gastrointestinal tract, mediastinum and central nervous system.⁽¹⁶⁾ Due to non-specific clinical symptoms and radiological findings it is diagnosed by histopathological evaluation.⁽¹¹⁾ In the literature, intracranial IMT is usually misdiagnosed as meningioma or other malignancies.⁽²⁾ Surgical excision is the main treatment approach since medical treatment (e.g. steroid) and/or radiotherapy are also performed in the literature.^(7,15,18) In our report, we present a rare intracranial IMT mimicking meningioma in a female patient.

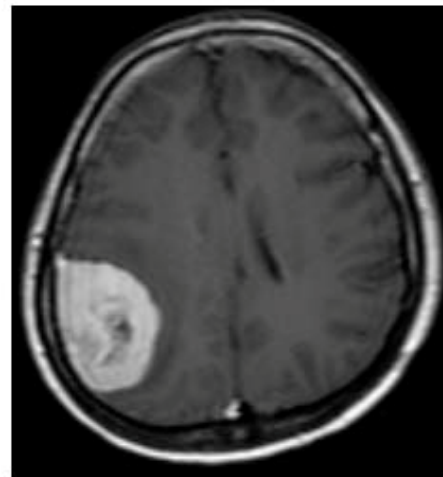
CASE PRESENTATION

Thirty-six year-old female patient admitted to the hospital without any known disease before, complaining from gradually increasing headache and vomiting for one month. On her neurological examination, 1/5 muscle weakness with dysesthesia

were detected on left upper and lower extremities. On her brain CT, there was an intracranial mass located at right frontoparietal region. On her brain MRI, the mass was hypointense on T1-W and hyperintense on T2-W images with a homogenous contrast enhancement on postcontrast images. The mass was lobulated and located extraaxially. It was displacing the adjacent brain parenchyma medially with edema around it. (Images 1a, 1b). Meningioma was the first on differential diagnosis list. The patient underwent right frontoparietal craniotomy by using neuronavigation. The lesion was partially removed by using ultrasonic surgical aspirator and then totally excised with the dura. Duraplasty procedure was performed with galeal graft. On the immunohistopathologic examination, anaplastic lymphoma kinase (ALK) was positive and smooth muscle actin (SMA) was partially positive with painted fusiform cells and intense inflammatory cells (Images 2a, 2b, 2c). Histopathological diagnosis was IMT and Ki-67 proliferation index was under 1%. After five months follow-up, hemiparesis regressed, but dysesthesia remained.



A



B

Image 1a: On axial T2-A image, an extraaxial mass shows hyperintensity with millimetric cystic spaces in it. A vasogenic oedema is demonstrated on the adjacent brain parenchyma. **Image 1b.** On postcontrast T1-W image, the mass shows diffuse contrast enhancement with dural tail.

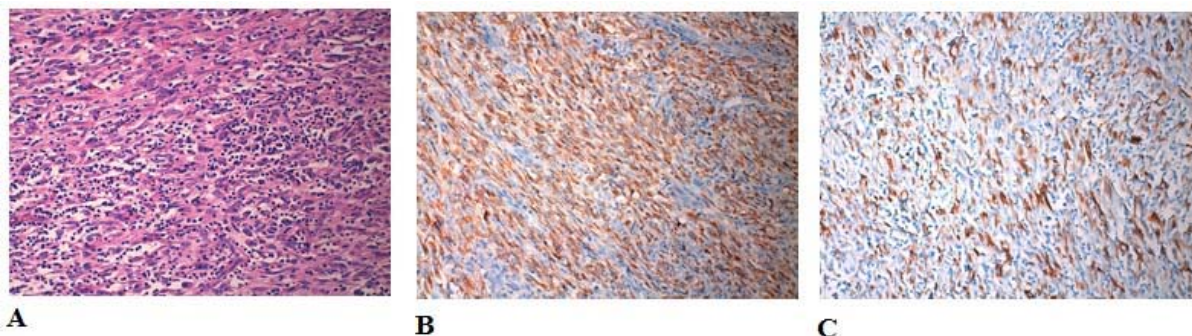


Image 2a: Light microscopy showing myofibroblastic proliferation intermixed with inflammatory cells (he, x200). **Image 2b.** Neoplastic cells are positive with ALK (X200). **Image 2c.** Neoplastic cells are partially positive with smooth muscle actin (X200).

DISCUSSION

First IMT was shown in lung tissue in 1939 by Brunn, and it was described as a malignant tumor with its clinical and radiological aspects by Umiker in 1954.⁽¹²⁾ IMT usually locates in lungs and is rarely seen in central nervous system.^(6,8) Radiologically, IMT can mimic some other tumors or tumor-like proliferations, i.e. meningioma, meningeal fibrosarcoma, inflammatory pseudotumor and plasma cell granuloma.^(10,13) Histopathologically it consists of fusiform cells, plasma cells, lymphocytes and sometimes histiocytes; thus IMT is a benign-solid tumor.⁽¹⁶⁾ In the literature, IMT are also called as inflammatory pseudotumor or granuloma. In 2002, World Health Organization announced IMT with its three sub-types; plasma cell granuloma, fibrous histiocytoma and desmoid fibromatosis.⁽⁵⁾ Hausler et al.⁽⁷⁾, classified these lesions into five groups considering their intracranial location. 1) Intraparenchymal, 2) Meningeal, 3) Intraparenchymal and meningeal, 4) Intraventricular, 5) Intracranial and extracranial.

Although its etiology still remains unknown, viral infections are reported to be responsible for IMT. Arber detected EBV (+) in lymph-nodes, spleen and liver in 7 of 18 cases with IMT. There is only one case in the literature with intracranial IMT with EBV (+).^(1,14) Some authors

called IMT as stage 1 fibrosarcoma due to their local aggressive behavior and being multifocal.⁽¹²⁾

On the histopathological examination, anaplastic lymphoma kinase (ALK) and smooth muscular antibody (SMA) are positive. The ALK gene expressed in cases with diffuse large B-cell lymphoma⁽⁴⁾, is also expressed in approximately 60% of cases with extracranial IMT.⁽³⁾ This finding points out the neoplastic origin of IMT.⁽¹⁷⁾ Hausler et al.⁽⁷⁾, reported a case with ALK expression, with malignant transformation.

There is no common therapy for IMTs. These lesions generally undergo surgical resection. After a subtotal resection, radiotherapy and/or corticosteroid treatment are performed if a recurrence occurs.⁽⁹⁾ Corticosteroid treatment was preferred in one pediatric case and it was also recommended in some acute inflammation and/or chronic fibrotic lesions in the literature.^(2,18)

CONCLUSION

IMT is a rare tumor of central nervous system mimicking other tumor lesions in radiological and clinical aspects. Immunohistopathological evaluation is the only diagnostic method. Prospective multicenter studies results are required due to the discrepancies on its etiology, treatment and prognosis.

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Received by: 23 December 2014

Revised by: 30 November 2015

Accepted: 09 December 2016

The Online Journal of Neurological Sciences (Turkish) 1984-2016

This e-journal is run by Ege University

Faculty of Medicine,

Dept. of Neurological Surgery, Bornova,

Izmir-35100TR

as part of the Ege Neurological Surgery

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Journal of Neurological Sciences (Turkish)

Abbr: J. Neurol. Sci.[Turk]

ISSNe 1302-1664

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