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Case Report

Intracranial fibrosarcoma treated with adjuvant radiation and temozolomide: Report of a case and review of all published cases

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KEYWORDS

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Abstract *Introduction:* Fibrosarcoma is a rare brain tumour with 33 cases reported so far. However, there is no clear consensus about the nature of the disease and treatment as well as outcome.

Methods: A MEDLINE search was carried out using MESH terms like intracranial fibrosarcoma, intraspinal fibrosarcoma, fibrosarcoma meninges and fibrosarcoma brain. A total of 22 case reports and series reporting a total of 33 cases were identified. We here also report a case treated in our institute with adjuvant radiation and concurrent and maintenance temozolomide.

Results: The age of presentation ranged from 2 months to 75 years (Median = 17 years). The gender ratio was found to be M:F of 1.75–1. Treatment modalities were described for 17 cases. Surgery was part of treatment in all cases while radiation was a part of treatment in 59% of cases ($n = 10$) and chemotherapy in 29% cases ($n = 5$). Survival data were available only for 8 cases and ranged from 1 day to 8 years (Median = 15.5 months).

Conclusion: Fibrosarcoma is a rare disease with dismal prognosis. Surgery remains the cornerstone of therapy. Radiation confers long term disease control and survival. Chemotherapy needs to be evaluated for these tumours to improve survival.

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Introduction

Fibrosarcoma of the brain is a rare tumour with a poor prognosis. These tumours have been hypothesized to arise from fibroblastic cells of meninges. It has been reported to be associated with radiation and chemotherapy exposure. These tumours share common radiologic feature to that of meningioma. Surgery has long been considered the treatment of

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choice. However, local recurrence is the commonest mode of failure and paved for the use of radiation to improve survival. Because of the propensity of systemic failure chemotherapy also appears an important component of the treatment. We herein present a case of intracranial fibrosarcoma treated in our institute and review all published cases to arrive at some conclusion regarding the behaviour and treatment outcome of fibrosarcoma.

Search methodology

A MEDLINE search was carried out using MESH terms like intracranial fibrosarcoma, intraspinal fibrosarcoma, fibrosarcoma meninges and fibrosarcoma brain. A total of 22 case reports and series reporting a total of 33 cases were identified. However, detailed information was not available in many reports.

Inclusion and exclusion criteria

Cases of intracranial fibrosarcoma of any age were included. Fibrosarcoma of any other location with metastasis to brain was excluded from the present analysis.

Data entry and analysis

The available data were tabulated for age, sex, presenting symptoms, location of tumour, imaging findings, associations, histopathology and immunohistochemistry, treatment modalities used and survival. The median age of presentation and median survival were calculated.

Case report

A 66 year old male presented with a 2 month history of headache, weakness on the left side of the body and deviation of angle of mouth towards the right side. The patient had a history of pulmonary tuberculosis 20 years back treated successfully with anti-tubercular therapy. A contrast enhanced MRI of the brain revealed a homogenous well circumscribed large mass in the right posterior frontal and parietal region showing uniform intense contrast enhancement with clear line of cleavage between it and the brain parenchyma suggestive of meningioma (Fig. 1). The patient underwent right fronto-parietal craniotomy with gross total excision of the space occupying lesion. The post-operative histopathology was reported to be fibrosarcoma positive for vimentin on immunohistochemistry. A PET-CT was done to rule out metastases or extracranial primary. The PET-CT revealed no abnormal FDG uptake. Subsequently, the patient was planned for post-operative radiotherapy of 60 Gray 30 fractions over 6 weeks with concurrent temozolomide (75 mg/m^2) followed by maintenance temozolomide for 6 months (150–200 mg/m^2 D1–D5 repeated every 4 weeks). Radiation was planned on pinnacle version 8.0 with 3 D conformal radiation therapy. The CTV50 included the T2 flair abnormality with 2.5 cm isotropic expansion. The CTV60 was taken as disease evident in the T1 weighted image with 1.5 cm isotropic expansion. PTV was created with a 3 mm isotropic margin for both the CTV. The patient was treated on the Synergy S with kV-CBCT image guidance. The dose constraints given and achieved are tabulated in Table 1. The patient tolerated the treatment well without any interruptions. The patient is asymptomatic and disease free at 6 months post diagnosis.

Literature review

The age of presentation ranged from 2 months to 75 years (Median = 17 years). The gender ratio was found to be M:F of 1.75–1. Headache was the most common symptom followed by generalized tonic clonic seizures and scalp swelling. Four cases have been reported to be associated with previous radiation exposure whereas one case was associated with exposure to multi-agent chemotherapy. Enhancing mass was seen on imaging in all cases while haemorrhage and bone destruction were described in two cases. Treatment modalities were described for 17 cases. Surgery was part of treatment in all cases while radiation was a part of treatment in 59% of cases (n = 10) and chemotherapy in 29% cases (n = 5). Both chemotherapy and radiation were used in 23% of cases (n = 4). Survival data were available only for 8 cases and ranged from 1 day to 8 years (Median = 15.5 months). Out of 17 cases for which data were available, 11 patients had a local recurrence and six patients experienced systemic recurrence. The details of patient characteristics, treatment and outcome have been summarized in Table 2.

Discussion

Intracranial fibrosarcoma is a rare entity with 33 cases reported in the available literature. They have been described as arising de novo or arising as secondary tumours in patients who have received radiation therapy to the brain [1–5] and in patients who have been treated with chemotherapy for glioblastoma [6]. They have been described in patients with a history of meningioma [7] and also in association with meningioangiomatosis [8].

They have been described in ages ranging from 2 months to 75 years of age. Most cases described are intracranial. Only

Table 1 Dose constraint used in the present case.

Organ at risk	Dose constraints	Dose achieved
Brainstem	Dmax < 54 Gray	50.48 Gray
Opposite temporal lobe	Dmax < 60 Gray	43.60 Gray
Optic chiasma	Dmax < 54 Gray	20.94 Gray
Right optic nerve	Dmax < 54 Gray	7.51 Gray
Left optic nerve	Dmax < 54 Gray	5.74 Gray
Right eye	Dmean < 35 Gray	2.26 Gray
Left eye	Dmean < 35 Gray	3.19 Gray
Right eye lens	Dmax < 10 Gray	2.19 Gray
Left eye lens	Dmax < 10 Gray	1.31 Gray
Right lacrimal gland	Dmean < 20 Gray	2.19 Gray
Left lacrimal gland	Dmean < 20 Gray	1.29 Gray
Hippocampus	Dmean < 7.3 Gray	39.62 Gray
Hippocampal avoidance zone	Dmean < 7.3 Gray	38.21 Gray

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