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Case Report

Successful multimodal management of central nervous system solitary fibrous tumor: A case report ☆

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ABSTRACT

Solitary fibrous tumors (SFTs) are rare neoplasms that can occur in various locations, including the central nervous system (CNS). We present a case report of a 47-year-old male patient with an intracranial SFT who underwent subtotal resection followed by adjuvant radiotherapy. The patient initially presented with chronic left temporal headache and was diagnosed with an intra-axial double-component mass in the left temporoparietal lobe. Histopathological examination confirmed the diagnosis of SFT, and immunohistochemical staining demonstrated positivity for CD34, Bcl-2, and STAT6. Following the incomplete resection, the patient received adjuvant radiotherapy using volumetric modulated arc therapy (VMAT) technique. During radiotherapy, the patient experienced a spontaneous encephalocele rupture but recovered without complications. One year postradiotherapy, the patient showed no recurrence of symptoms or radiological evidence of tumor recurrence. This case highlights the challenges in the diagnosis and management of CNS SFTs and suggests that subtotal resection followed by adjuvant radiotherapy may be an effective treatment approach in achieving favorable outcomes for these rare neoplasms.

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Introduction

Solitary fibrous tumors (SFTs) of the central nervous system (CNS) are rare mesenchymal neoplasms that present significant diagnostic and therapeutic challenges. First described by Klemperer and Rabin in 1931 [1], SFTs arise from spindle cells. Although initially identified in the pleura, these tumors have since been found in various other locations, including the lung, pericardium, mediastinum, soft tissue, and CNS. While generally considered benign, CNS SFTs can exhibit aggressive behavior, necessitating appropriate management [2]. Historically, the treatment of CNS SFTs has focused on surgical resection, with the goal of maximal tumor removal while preserving neurological function. However, achieving complete resection can be difficult, especially when tumors are located in critical or eloquent brain regions [3].

When a gross total resection is not feasible or when tumors exhibit high-risk characteristics, such as large size, invasion of critical structures, or high mitotic activity, adjuvant therapies, including radiotherapy, are often employed to enhance local control and decrease the risk of recurrence. The role of adjuvant radiotherapy in managing central nervous system (CNS) solitary fibrous tumors (SFTs) is still debated. While some studies indicate that adjuvant radiotherapy may help reduce recurrence rates, other research presents conflicting results on its effectiveness.

The optimal timing, dose, and target volume for radiotherapy in CNS solitary fibrous tumors have not yet been fully established. There are ongoing concerns about its long-term effects on neurological function and quality of life, especially in patients with benign or low-grade tumors [4].

This report details a case of CNS solitary fibrous tumor (SFT) managed with Multimodal therapy including subtotal resection followed by adjuvant radiotherapy. Our goal is to offer an in-depth review of the current literature to clarify the most effective treatment strategies and illustrate how this case can inform and refine evolving management approaches for CNS SFTs.

Case report

A 47-year-old male patient with an unremarkable medical history presented with a 1-year history of progressively worsening left temporal headache, accompanied by intermittent nausea and vomiting. The headache was constant and dull, with occasional exacerbations to moderate severity. The patient denied any associated symptoms such as photophobia, phonophobia, visual disturbances, or aura. There were no identifiable triggers, and the headache did not follow a distinct diurnal pattern. Despite the use of over-the-counter analgesics, the headache persisted, prompting him to seek medical attention.

Further history revealed no preceding trauma, recent illnesses, or significant stressors. The patient denied any history of substance abuse, including alcohol and tobacco. His family history was negative for neurological disorders or malignancies.

On examination, his vital signs were within normal limits. A neurological assessment revealed intact cranial nerve function, normal motor strength and sensation bilaterally, with no focal deficits. Fundoscopic examination was unremarkable. There were no signs of meningeal irritation or neck stiffness. The remainder of the physical examination was noncontributory.

Given the chronicity and persistence of the symptoms, as well as the absence of focal neurological deficits, a comprehensive diagnostic workup was initiated to investigate the underlying cause of the patient's symptoms. This included neuroimaging studies such as magnetic resonance imaging (MRI) of the brain, which revealed a well-defined lobulated intra-axial double component mass (solid and cystic) in left temporoparietal lobe with hypointense portion and isointense portion on T1-weighted images and a hyperintense portion and an isointense portion on T2-weighted images, with marked heterogeneous enhancement on postcontrast T1 sequence. The mass measured approximately 72×72×57 mm in size. The lesion was compressing the left lateral ventricle's atrium with mass effect and was abutting superior surface of tentorium with partial invasion of the left transverse sinus. Diffusion tensor imaging (DTI) suggested displacement and compression of the white matter fiber tracts in the affected region (Fig. 1).

The case was presented to a multidisciplinary team (MDT), which recommended complete surgical resection of the tumor. Due to its proximity to sensitive areas, over 90% of the tumor was successfully resected, though a small portion remained due to the complexity of its location. The postoperative course was complicated by a noninfected meningocele, which was managed without further issues.

Histopathological examination of the resected tumor showed ovoid to slightly fusiform neoplastic cells. They were arranged haphazardly or in short ill-defined fascicles. Branching, staghorn-like (hemangiopericytoma-like) vasculature was observed. Mitoses were estimated at 3 per 10 high-power fields. The tumor showed no aggressive histologic features: hypercellularity, atypia or necrosis. Immunohistochemical staining showed strong positivity for STAT6, CD34 and Bcl-2. Tumor cells were immune-negative for PS100 and EMA. Proliferation index (Ki67) was 2%. These findings confirmed the diagnosis of a WHO grade 1 solitary fibrous tumor (SFT) (Fig. 2), (Fig. 3).

Immunohistochemical staining showed strong positivity for CD34 and Bcl-2, consistent with SFT. Additionally, the tumor cells exhibited nuclear positivity for STAT6, confirming the diagnosis. Focal areas of hypercellularity with spindle cells arranged in a patternless architecture, along with staghorn-like vasculature and collagenous bands, were also observed.

Given the incomplete resection and the aggressive histopathological features, adjuvant radiotherapy was recommended.

During the simulation process, the patient was positioned supine on the treatment couch and secured using a thermoplastic immobilization mask, along with additional immobilization devices such as headrests, to ensure consistent positioning throughout the imaging procedure and daily radiotherapy sessions. CT scans were then acquired with a slice thickness of 2-3 mm, ensuring comprehensive coverage of the

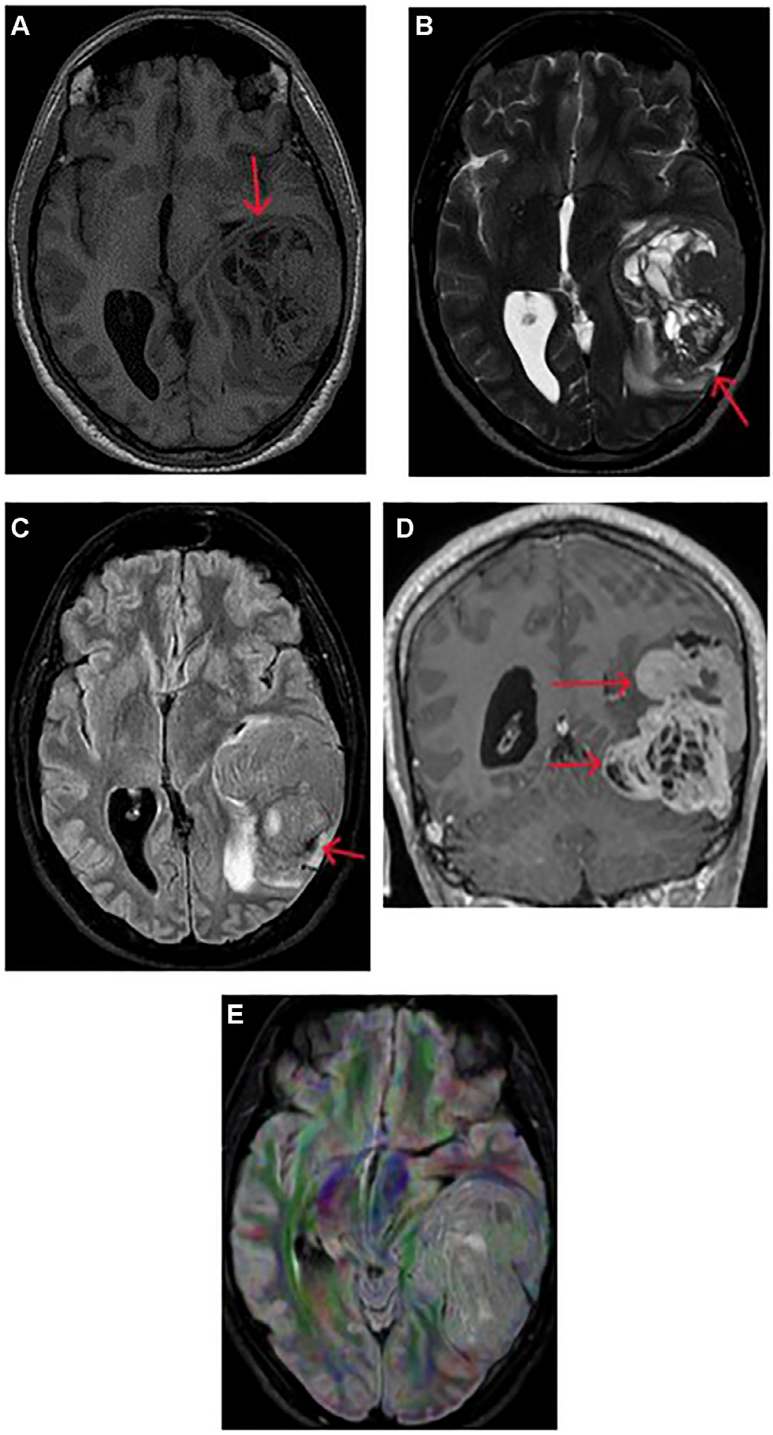

Fig. 1 – Preoperative MRI of the brain in axial T1-weighted (A), T2-weighted (B) and fluid-attenuated inversion recovery (FLAIR; C) views showing a lesion with mixed-intensity signals adjacent to the left temporoparietal region. The lesion appears mixed iso- and hypointense on T2 FLAIR with surrounding edema. Contrast-enhanced T1-weighted coronal image (D) suggest heterogeneous and intense enhancement of the tumor invading partially the left transverse sinus. DTI (E) reveals compression and damage of the white matter fiber tracts in the lesion zone (namely corticospinal tract, superior and inferior longitudinal fasciculi and inferior fronto-occipital fasciculus).

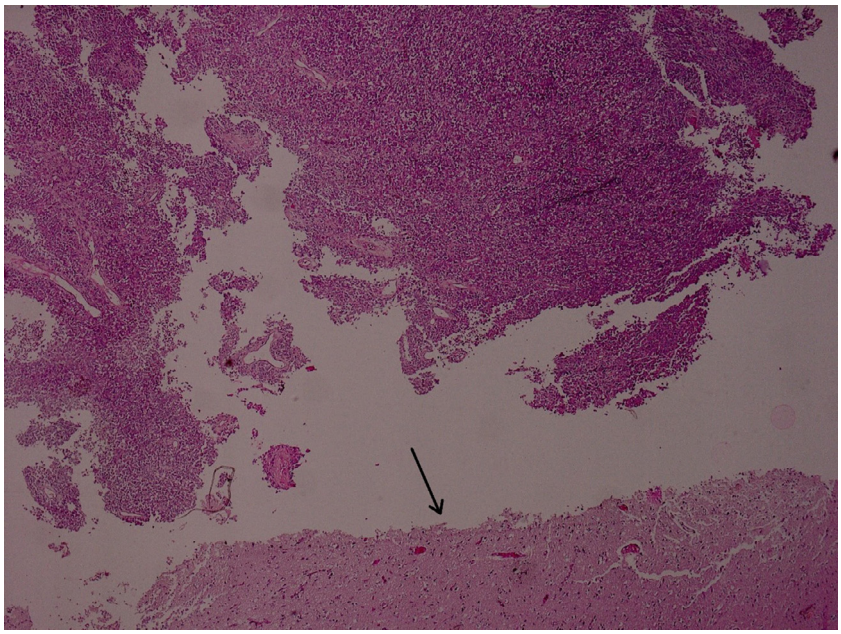

Fig. 2 – Representative micrograph of the tumor. Tumor infiltrates brain parenchyma (arrow). Hematoxylin-eosin; x 40.

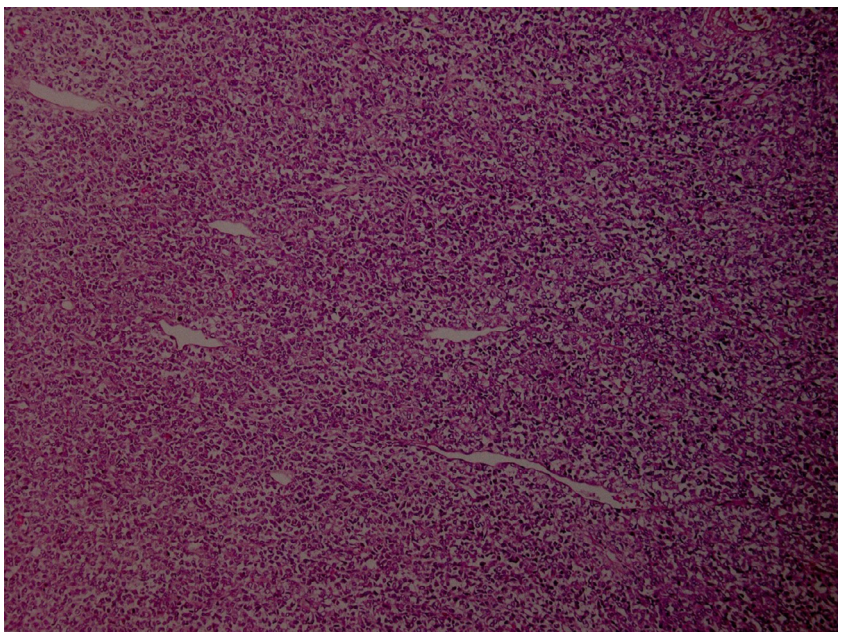

Fig. 3 – Representative micrograph of the tumor. Ovoid to fusiform cells are arranged haphazardly or in short fascicles. Thin walled, branching, staghorn-like (hemangiopericytoma-like) vasculature is observed. Hematoxylin-eosin; x 100.

region of interest from the skull base to the lower cervical spine.

The planning and tumor segmentation for radiotherapy (RT) were meticulously handled by a multidisciplinary team, including a radiation oncologist, medical physicist, and dosimetrist. The radiation oncologist defined the target area and critical structures based on preoperative imaging and surgical findings to minimize healthy tissue exposure. The Gross Tumor Volume (GTV) from preoperative MRI T1 defined the Clinical Target Volume (CTV), extended by 1 cm, with a 0.5 cm margin for the Planning Target Volume (PTV). The medical physicist ensured accurate dose calculations and safety compliance, while the dosimetrist developed a precise treatment plan using advanced software for optimal radiation delivery. Treatment planning was conducted using the Eclipse treatment planning system version 13 (Varian Medical Systems, Palo Alto, CA). Radiotherapy was administered using a 6 MV photon beam delivered by the TrueBeam STX lin-

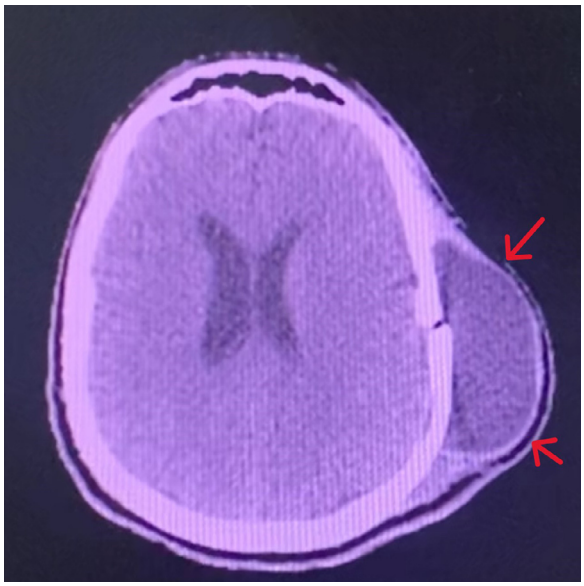

Fig. 4 – An axial slice of the planning CT scan showing a huge left meningocele.

ear accelerator employing the RapidArc: Volumetric arc therapy technique. Daily cone-beam CT imaging guided the treatment to ensure precise delivery, with a prescribed dose of 54 Gy administered in daily 2 Gy fractions over 5 weeks (Fig. 4), (Fig. 5).

Dose-volume histograms (DVHs) were generated to evaluate the radiation dose distribution and assess the conformity of the treatment plan to the tumor while sparing surrounding healthy tissue. Specific dose constraints were set for critical structures, including the brainstem and optic nerves. The maximum dose to the brainstem was limited to 50 Gy, while the optic nerves were constrained to a maximum dose of 45 Gy. Healthy brain tissue was limited to a maximum of 40 Gy. For the Planning Target Volume (PTV), the treatment plan aimed to deliver 95% to 107% of the prescribed dose to ensure adequate coverage while maintaining safety for surrounding critical structures.

During radiotherapy, the patient experienced a spontaneous rupture of the encephalocele, likely due to increased intracranial pressure, resulting in cerebrospinal fluid leakage and necessitating hospitalization. The rupture of the cephalocele was clinically diagnosed based on the observation of clear fluid leakage from the surgical wound. The patient's clinical status was closely monitored, and intravenous antibiotics were administered to prevent infection. Following 7 days of antibiotic treatment, the patient's condition steadily improved, and no signs of infection were observed during the hospitalization period. Consequently, radiotherapy was resumed as scheduled, without encountering any further complications.

One year postradiotherapy, the patient underwent comprehensive clinical and radiological follow-up assessments every 3 months to monitor treatment response and disease progression. Clinical examinations did not reveal any recurrence of symptoms related to the previously treated solitary fibrous tumor or any new neurological deficits. Additionally, the patient did not report experiencing persistent headaches, nausea, vomiting, or any other concerning symptoms during the follow-up period.

Radiological evaluation, including brain magnetic resonance imaging (MRI), was conducted to assess for any signs of tumor recurrence or residual disease. The MRI findings demonstrated no evidence of tumor recurrence or residual disease within the treated area. Specifically, there were no new enhancing lesions or abnormal signal intensities observed on the postcontrast MRI images, consistent with a favorable treatment response.

Throughout and following the treatment, no adverse effects, including neurotoxicity, dementia, or cerebral necrosis, were observed. Furthermore, patients exhibited preserved quality of life, maintaining their daily activities and overall functional well-being.

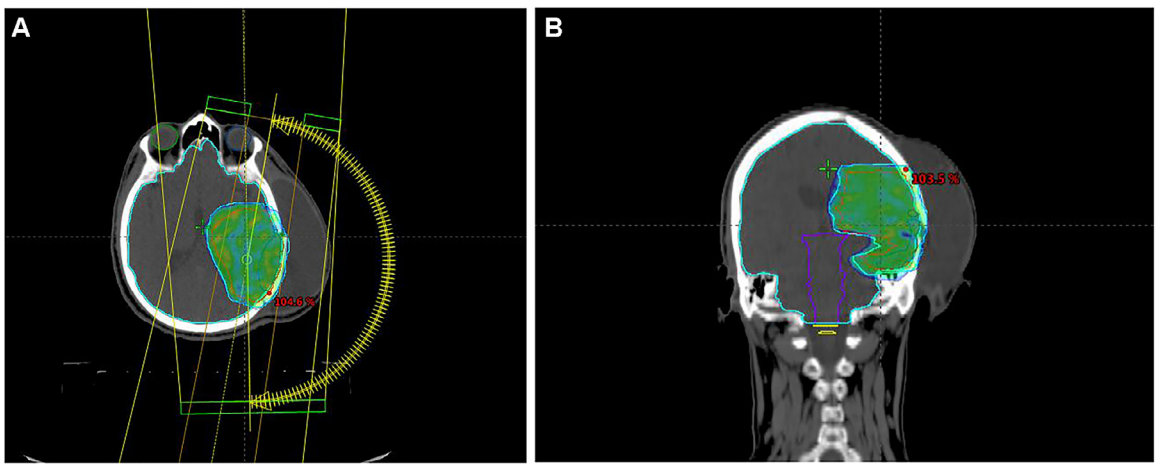

Fig. 5 – An axial (A) and coronal (B) slices of the planning CT scan is shown with isodose lines, with 54 Gy (27 x 2) fractionation scheme and VMAT treatment delivery technique.

Discussion

Solitary fibrous tumors (SFTs) are rare within the central nervous system (CNS), first reported by Carneiro et al. in 1996 [5]. They primarily affect individuals aged 60 to 70 years, with a slightly higher incidence in women around the age of 50 [6]. Patients with CNS SFTs often present with nonspecific symptoms related to tumor location or increased intracranial pressure, such as dizziness, headache, hemiplegia, gait disturbances, hearing issues, and mental disorders [7]. Differentiating SFTs from other intracranial tumors such as fibrous meningiomas and hemangiopericytomas, remains challenging despite advancements in imaging techniques like MRI and CT scans.

Magnetic resonance imaging (MRI) is the primary imaging modality for diagnosing solitary fibrous tumors (SFTs) in the central nervous system (CNS). These rare tumors exhibit distinct imaging characteristics that aid in differentiating them from other neoplasms, particularly meningiomas. On T1-weighted images (T1WI), SFTs typically display intermediate signal intensity, which can often blend with the surrounding brain tissue, making them challenging to identify clearly. This indistinct appearance necessitates careful evaluation of additional imaging sequences. Conversely, on T2-weighted images (T2WI), SFTs generally appear iso- to hypointense compared to the brain parenchyma. The notable heterogeneous "yin-yang" appearance of SFTs, characterized by alternating regions of high and low signal intensity, reflects variations in tumor composition, including fibrous tissue and necrotic areas.

A key feature of solitary fibrous tumors (SFTs) is their avid contrast enhancement following the administration of gadolinium-based contrast agents. This enhancement reflects a rich vascular supply within the tumor and is visually striking on postcontrast images, highlighting the tumor's boundaries and improving its visibility. Additionally, while the presence of a dural tail is common in meningiomas [8], it can also be observed in SFTs. This overlap can complicate differential diagnosis, underscoring the importance of comprehensive imaging analysis.

Diffusion-weighted imaging (DWI) further aids in characterizing SFTs by highlighting areas of restricted diffusion, which often indicate high cellularity or necrosis. This feature is particularly useful for differentiating SFTs from other CNS lesions, such as abscesses or more aggressive tumors. Areas of high cellular density within SFTs restrict the movement of water molecules, resulting in hyperintense signals on DWI. This can be crucial for assessing the tumor's aggressiveness and overall characteristics.

Magnetic resonance spectroscopy (MRS) adds another layer of diagnostic capability by assessing the metabolic profile of the tumor. In SFTs, MRS typically reveals elevated levels of myo-inositol, which is associated with glial proliferation and tumor activity, along with increased lipid and lactate levels. These metabolic markers can provide valuable insights into the tumor's biological behavior, distinguishing SFTs from other neoplasms and guiding treatment decisions.

Despite the strengths of these advanced imaging techniques, differentiating SFTs from meningiomas remains challenging due to overlapping features. Meningiomas often present with calcifications and hyperostosis of adjacent bone, both of which are uncommon in SFTs [9].The absence of these features can be a useful clue in narrowing the differential diagnosis. Additionally, considering the tumor's location, often adjacent to the dura mater, along with patient demographics and clinical presentation, in conjunction with imaging findings, enhances diagnostic accuracy.

Initially classified separately, solitary fibrous tumors (SFTs) were considered benign, while hemangiopericytomas (HPCs) were regarded as locally aggressive in the 2002 WHO classification [10].Despite low recurrence rates in early studies, the identification of NAB2-STAT6 fusion genes in SFTs and HPCs led to their unification in the 2013 WHO classification [11].The 2016 WHO update introduced the combined term "SFT/HPC," with introducing grading based on mitotic activity and necrosis [12].

Histopathologically, SFTs range from hypocellular to hypercellular phenotypes. Hypocellular SFTs display short spindle and oval-round cells, a "patternless pattern" arrangements, hyalinized collagen bands, and staghorn vessels without mitotic activity, corresponding to WHO grade I (benign). In contrast, hypercellular SFTs exhibit increased cellularity, disorganized cell arrangements, frequent mitotic activity, and necrosis, and are classified as WHO grade II or III. Immunohistochemistry confirms CD34, CD99, and STAT6 positivity, indicating the presence of NAB2-STAT6 fusion [13].

Several studies have examined the relationship between the quality of surgical resection, recurrence rates, and time to recurrence. Kim et al. reported a local recurrence rate of 38.7% among 31 patients, with gross total resection (GTR) significantly improving 5-year recurrence-free survival (72.7% vs 20.8%, $P = .006$) and extending the time to recurrence (111 months vs 43 months, $P < .05$). Soyuer et al. found a 61% local recurrence rate among 29 patients, with GTR showing better 5-year recurrence-free survival (84% vs 38%, $P = .0034$) [14]. Guthrie et al. observed a 66% overall recurrence rate among 44 patients, noting that GTR improved recurrence-free survival, although no significant difference was seen in the time to recurrence (48 months vs 54 months) [15].

In our case, the decision to proceed with adjuvant radiotherapy was based on the presence of positive surgical margins and the high risk of recurrence. The patient received a radiation dose greater than 50 Gy, in line with literature recommendations. Studies by Combs et al. [16] and Dufour et al. [17] have shown that doses between 50 and 60 Gy can effectively reduce local recurrence rates by up to 12.5% when combined with surgery. Similarly, Stessin et al. confirmed improved recurrence-free survival with doses higher than 50 Gy [18].

However, the impact on overall survival remains debated. Guthrie et al. noted a significant improvement in recurrence-free survival from 34 to 75 months ($P < .05$) with adjuvant radiotherapy but did not find a clear effect on overall survival, possibly due to limited study power [19].Moreover, Rutkowski et al.'s meta-analysis highlighted a potential decrease in overall survival from 18.6 years to 4 years associated with doses above 50 Gy, likely reflecting older, more toxic techniques. In our patient's case, the use of modern radiotherapy techniques, such as volumetric modulated arc therapy (VMAT), aimed to

maximize local control while minimizing long-term toxicity. This aligns with current evidence supporting the efficacy of higher radiation doses in improving local control, despite the ongoing debate about overall survival.

Despite high recurrence rates after surgical resection, complete surgical resection remains the preferred treatment, significantly more effective than subtotal resection [20]. Adjuvant radiotherapy postresection is still debated, as it has not consistently shown a significant impact on overall survival [21]. The average time to recurrence for SFTs is approximately 3 years [22]. Notably, a patient who underwent surgery followed by gamma knife radiosurgery has not required further treatment for 8 years, suggesting its potential effectiveness, though further cases are needed to confirm this.

The spontaneous rupture of an encephalocele carries significant implications for patient management, primarily due to the increased risk of infection, neurological deficits, and potential hemorrhage. When an encephalocele ruptures, it exposes underlying brain tissue and cerebrospinal fluid (CSF) to the external environment, heightening the risk of infections like meningitis. Additionally, the rupture can lead to neurological complications, including seizures or cognitive impairments, depending on the location and severity of the defect. Immediate assessment is crucial; a thorough neurological examination and imaging studies, such as CT or MRI, should be conducted to evaluate the brain's condition and the extent of any injury. Management strategies may include surgical intervention to repair the defect and prevent further complications, alongside close monitoring for signs of infection or CSF leaks.

The long-term effects of radiotherapy, particularly on neurological function and quality of life, are crucial considerations in the management of benign CNS tumors. While radiotherapy can effectively control tumor growth, it may result in acute and chronic neurological side effects, such as cognitive impairment, memory deficits, and fatigue. These complications can significantly affect a patient's quality of life, influencing daily activities and social interactions. Additionally, the risk of secondary malignancies, especially in younger patients, presents another challenge in treatment planning. According to the updated recommendations from the French Society for Radiation Oncology regarding benign intracranial tumors [23], modern radiotherapy techniques are designed to minimize risks to neurological function and maintain quality of life. While controversies remain, radiotherapy is considered a viable option when surgery poses a significant functional risk or when there is progressive residual tumor volume, recurrence, or contraindications for surgery. These decisions are made after a thorough multidisciplinary discussion to ensure optimal patient outcomes. In our patient, radiotherapy was indicated due to the presence of positive margins after surgical resection. Importantly, no toxicities, including neurotoxicity, dementia, or cerebral necrosis, were reported during or after the treatment.

Our case underscores the importance of a coordinated multimodal strategy in treating CNS solitary fibrous tumors, showcasing how the combination of surgery and adjuvant radiotherapy can lead to successful outcomes in complex and challenging scenarios, However, it is important to note that this study presents only a single case, and therefore, the effectiveness and applicability of the multimodal treatment strategy used in this instance may not be generalizable to other cases. Further studies involving a larger cohort of patients are needed to confirm the potential benefits and broader applicability of this approach.

Conclusion

CNS SFTs present diagnostic and therapeutic challenges, requiring a multidisciplinary approach for optimal management. While surgical resection remains the mainstay of treatment, the role of adjuvant radiotherapy in improving outcomes warrants further investigation. In our case, subtotal resection followed by adjuvant radiotherapy resulted in favorable outcomes, highlighting the effectiveness of this multimodal therapy in managing CNS solitary fibrous tumors.

Patient consent

I hereby certify that informed consent has been obtained from the patient for the publication of this case report. The patient has been informed of the objectives of the publication, as well as the details regarding the disclosure of relevant medical information.

It has been explained to the patient that all identifying information (such as name, date of birth, address) will be anonymized and will not appear in the published report. The patient understands that he will not be recognizable from the information provided in the case report.

The patient has given his consent freely and has been informed that he can withdraw this consent at any time without affecting the quality of care received.

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