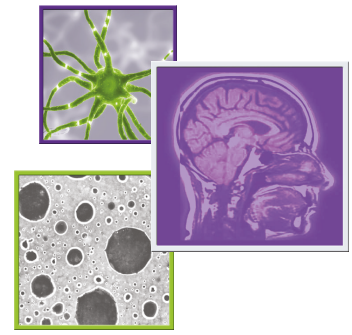
Case Report

CNS Oncology



Grade II Sylvian fissure meningiomas without dural attachment: case report and review of the literature

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Practice points

- Sylvian fissure meningiomas are rare nondural-based lesions.
- Sylvian fissure meningiomas are more common in males and usually present with seizures.
- The anatomical relationship with the middle cerebral artery has prevented complete resections in the described cases.
- Preoperative vascular imaging and intraoperative angiography may improve surgical planning and safe complete tumoral resection rates.

Sylvianfissure meningiomas (SFMs) represent a rare subgroup of nondural-based tumors arising from the meningothelial cells within the arachnoid of the Sylvian fissure. SFMs are more frequent in young males, usually manifest with seizures and display the same radiological features of meningiomas in other locations. Although the absence of dural attachment makes these tumors suitable for a complete resection, their anatomical relationships with the middle cerebral artery branches have impaired its achievement in half of them. To the best of our knowledge, only five atypical WHO grade II SFMs have been previously described. We provide a literature review of SFMs WHO grades I–II and discuss common characteristics and surgical challenges we found in a similar case.

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Keywords: atypical meningiomas • dural attachment • meningioma • Sylvian fissure

Meningiomas are thought to arise from the meningothelial cells within the arachnoid and are typically recognized by their attachment to the dura. These cells can also be found in the choroid plexus and tela choroidea, which can explain why meningiomas may rarely occur in other locations without dural attachment. In the absence of dural attachment, they are categorized into intraventricular, pineal region, intraparenchymal, subcortical and deep Sylvian fissure meningiomas (SFMs) [1].

Meningiomas are most commonly supplied by dural arteries arising from external carotid system, but may develop a secondary supply via the pial arteries, such as the branches of the anterior, middle and posterior cerebral arteries from the internal carotid and vertebrobasilar systems [2].

Atypical meningiomas WHO grade II account for 5–7% of all meningiomas and have a higher likelihood of recurring as they proliferate at a higher rate and can invade the brain [3]. As the role of complementary treatment is yet to be defined, the surgical approach and the extent of resection is of paramount importance in the prognosis. In those cases where no dural attachment is found, a complete resection of the tumor may represent the cure for these patients.

Future Medicine

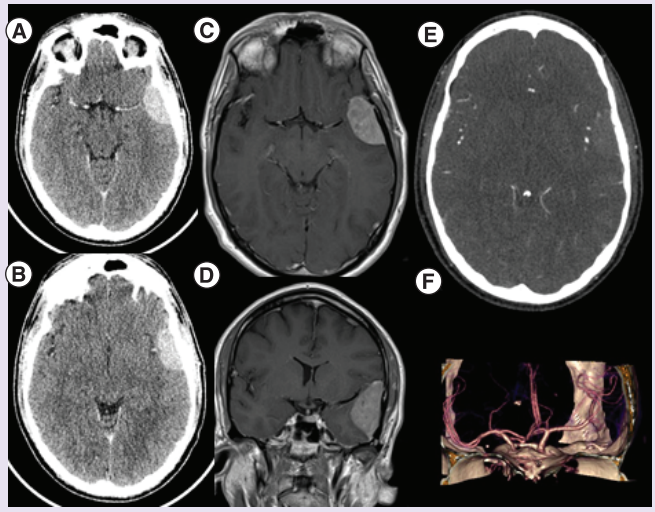

Figure 1. Pre-operative imaging. Axial-CT head with contrast **(A and B)** and axial **(C)** and coronal **(D)** MRI T1-weighted images with gadolinium revealing a homogenously enhancing left pterional region tumor. Axial angio-CT head **(E)** and 3D angio-reconstruction **(F)** revealing the intrinsic relationship in between the left MCA branches and the lesion.
CT: Computed tomography; MCA: Middle cerebral artery; MRI: Magnetic resonance impaging; T1: Superior temporal gyrus.

The authors discuss a patient with an atypical WHO grade II SFM without dural attachment and review the previous literature centered on clinical, radiological, pathologic characteristics and treatment approach of this rare condition.

Case report

A 32-year-old right-handed charity worker with no significant past medical history presented with a year-long history of daily bitemporal and frontal headaches. The headaches were associated with dizziness and unsteadiness as well as nausea, phonophobia and photophobia. These symptoms then progressed in a month affecting the patient's activities of daily living. There was no associated history of seizures, speech disturbance or sensory/motor deficits and the neurological exam was unremarkable.

A computed tomography head and angio-computed tomography identified an hyperdense lesion in the left temporal convexity in close relation with the Sylvian fissure (M2 branches from left MCA) and the MRI-revealed and homogenous contrast-uptake lesion, consistent with a meningioma (Figure 1). A left pterional craniotomy was performed and, as soon as the dura was opened, it became evident that the tumor did not have any convexity or skull-based dural attachment. Proximal microsurgical opening of the Sylvian fissure was performed starting at the anterior Sylvian point, exposing the carotid artery for proximal control. Internal debulking of the tumor followed by gentle dissection of the capsule all along the arachnoidal plane from the surrounding brain parenchyma was performed while preserving the temporal M2 and M3 branches. However, a clear attachment of the meningioma to the arachnoid overlying the most anterior portion of the posterior insular gyrus became evident, and it was was dissected and coagulated. In fact, while a clear arachnoidal plane was identified all around the meningioma, an exception was made by the portion of the tumor facing the posterior insular gyrus (Figure 2).

The histological staining identified atypical features within the tumor specimens. There were areas of hypercellularity with small nuclei and areas of necrosis, but no evidence of increased mitotic activity and a low-proliferating index. The features were consistent with an atypical meningioma, WHO grade II. (Figure 3). The postoperative course was uneventful. The patient reported a consistent improvement in his headaches in the next days following the surgery. A follow-up MRI at 3 months did not reveal any recurrence of the tumor and the patient is under clinical and radiological surveillance with no signs of recurrence at 3 years' follow-up (Figure 4).

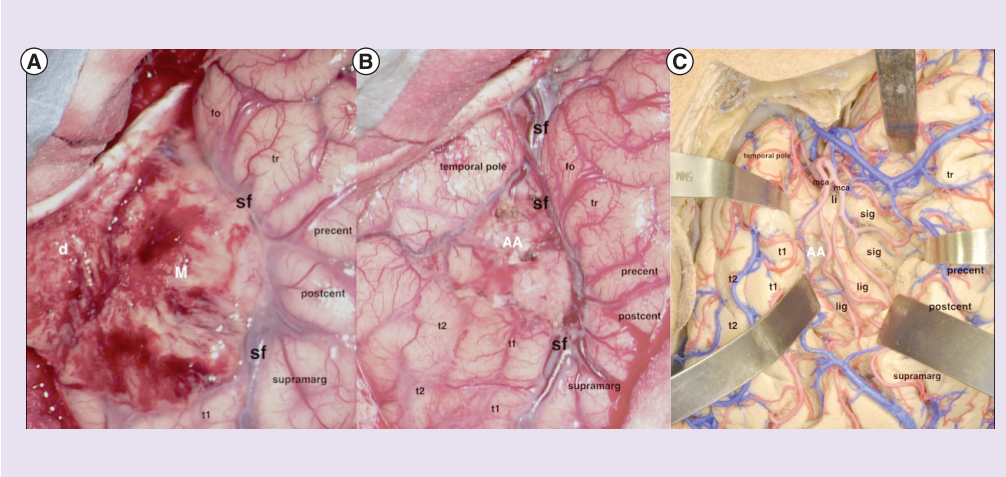

Figure 2. Intraoperative imaging and schematic review. Intraoperative photos before **(A)** and after **(B)** microsurgical resection of the SFM and correlation with an anatomical specimen with the same operative angle **(C)**. **(A)** shows the clear absence of any dural attachment of the SFM. **(B)** shows the AA of the Sylvian fissure meningioma in correspondence of the anterior part of the posterior long insular gyrus. **(C)** The Sylvian fissure is opened and the relationships of the middle cerebral artery branches with the surrounding anatomical structures is unveiled.
AA: Arachnoidal attachment; SFM: Sylvian fissure meningioma.

Discussion

Cushing and Eisenhardt originally classified meningiomas without dural attachment in intraventricular, subcortical and deep Sylvian [4]. Nowadays, meningiomas without dural attachments are classified in supratentorial (intraventricular, intraparenchymal or subcortical, pineal region, deep Sylvian) and infratentorial (intraventricular, inferior tela choroidea, cisterna magna and intraparenchymal) [5]. The most common lesions in this group occur in pediatric population and have an infratentorial location [6].

SFMs are rare entities and it is important to differentiate them from the sphenoid wing meningiomas. These are attached to the dura overlying the sphenoid wings, are usually associated with hyperostosis and they displace the MCA backwards as they grow, while the SFMs do not have dural attachment, do not produce hyperostosis and grow inbetween the MCA branches. Given the recent changes in the meningioma classification system, it is difficult to comment on the grades inbetween these locations, even though the literature presented suggests a higher proportion of grade II lesions among the SFMs [6]. Barcia–Goyanes *et al.* [7] described the first case in 1953, and since then only 28 cases (including the present report) have been described (Tables 1, 2 and 3). [8–26] The reported adult SFMs patients are young (mean age of 34.95 ± 3.35 years; 95% CI [27.93–41.97]) with a M:F ratio of 1.22 (11/9) and in the pediatric population (mean age is 5.71 ± 1.61 years; 95% CI [1.76-9.66]; the M:F ratio is 2:1 (4/2 and 1 unknown). When comparing grade I and grade II lesions, there is no significant differences in terms of mean age (grade I: 26.87 ± 3.90 years; vs grade II 24.33 ± 7.01 years; t-test $p > 0.05$), gender (grade I M:F ratio – 1.2 [12/10] versus grade II M:F ratio – 5 [5/1)]), clinical presentation (seizures is the most common presentation in both groups – grade 1 – 74% (17/23) and grade II – 67% (4/6)] and extent of resection (total resection in grade 1 – 65% [1/23] and total resection in grade II – 50% [3/6]. (Table 4)

When considering the WHO grade II atypical meningiomas, only six lesions have been described (considering the present report; Table 3) [6,9,11,16,21]. Atypical meningiomas constitute 20% of the meningiomas in these region, higher than in other locations (5–7%) as it has already been noted by Cecchi *et al.* [6]. Regarding its epidemiology, there is a clear male predominance (5/6) although there is no gender prevalence when considering all the SFMs; 5/6 patients are aged below 32 years old. When considered together, male gender and younger aged are risk factors for WHO grade II histological differentiation in other locations, which is also true in this location [6]. Therefore, there are no sufficient data published that allow assessing if the Sylvian fissure location is a risk factor for WHO grade II lesions *per se* or if it has been confounded by these previously known epidemiological risk factors. Even though absent in the present case, seizures is the most frequent symptom which is believed to be related either with the temporal location and with the fact that WHO grade II tumors might show adjacent brain invasion as part of their diagnostic criteria. Surprisingly, considering its location and the histological nature of these lesions, no focal

Table 1. Summary of the WHO grade I Sylvian fissure meningiomas in adult patients reported in the literature.

Study [Ref.] (year)	Age/gender	Presenting symptoms	MRI T1	MRI T2	CT scan	Vascular imaging	Extent of resection	Vascular supply	Histology	Outcome
Barcia-Goyanes *et al.* [7] (1953)	20/F	Seizures	–	–	–	Not performed	–	–	WHO grade I (psamommatous)	–
Cushing and Eisenhardt (1969) [4]	18/M	Seizures	–	–	–	Not performed	Partial	MCA	WHO grade I (psamommatous)	3 recurrences (5 years of survival)
	48/F	Seizures	–	–	Not performed	Partial	–	WHO grade I (psamommatous)	Died on the day of surgery	
Mori *et al.* [23] (1977)	23/M	Seizures	–	–	–	Not performed	Partial	–	WHO grade I (transition)	–
Saito *et al.* [26] (1979)	31/F	Seizures	–	–	Hyperdense lesion	Vascular blushing from internal carotid	Total	–	WHO grade I (psamommatous)	–
Tsuchida *et al.* [27] (1981)	46/M	Headache	–	–	–	Vascular blushing from internal carotid	Total	MCA	WHO grade I (meningothelial)	–
Okamoto *et al.* [25] (1985)	27/F	Headache and visual disturbances	–	–	–	–	Total	–	WHO grade I (fibroblastic)	5 years of survival
	35/F	Seizures	–	–	Hyperdense with homogeneous with contrast enhancement and oedema	Vascular blushing from internal carotid	Total	MCA	WHO grade I (fibroblastic)	–
Hirao *et al.* [15] (1986)	34/F	Seizures	–	–	Hyperdense with homogeneous with contrast enhancement	Vascular blushing from internal carotid	Total	–	WHO grade I (fibroblastic)	–
Graziani *et al.* [14] (1992)	19/M	Headache, memory disturbances and hemiparesis	Hypo	–	Hyperdense with calcifications with contrast enhancement and edema	Vascular blushing from internal carotid	Total	MCA	WHO grade I (psamommatous)	–
Chiocca *et al.* [1] (1994)	26/F	Seizures	Hypo	Hypo	Hyperdense with homogeneous with contrast enhancement and edema	Vascular blushing from internal carotid	Total	No attachment	WHO grade I (fibrous)	–
Matsumoto *et al.* [20] (1995)	62/F	Seizures	Hypo	Hypo	Calcified mass	Vascular blushing from internal carotid	Total	MCA	WHO grade I (psamommatous)	–
Chang *et al.* [9] (2005)	35M	Seizures	Iso	Iso (edema)	–	No vascular blushing	Partial	MCA	WHO grade I (transition)	–
Eghwrudjakpor *et al.* [12] (2006)	73F	Nonspecific symptoms	–	–	Heterogenous tumor	–	–	–	WHO grade I (meningothelial)	–
Aras *et al.* [8] (2012)	28M	4 m complex partial seizures	Hypo	Iso/Hypo	No		Total	MCA	WHO I meningothelial	No
Kim *et al.* [17] (2013)	43M	New onset seizure	Iso	Iso	No		Partial	–	WHO grade I	No

CT: Computed tomography; F: Female; M: Male; MCA: Middle cerebral artery; MRI: Magnetic resonance imaging; WHO: World Health Organization.

Table 2. Summary of the WHO grade I Sylvian fissure meningiomas in pediatric patients reported in the literature.

Study (year)	Age/gender	Presenting symptoms	MRI T1	T2	CT scan	Vascular imaging	Extent of resection	Vascular supply	Histology	Recurrence
Silbergeld *et al.* [28] (1988)	4F	Seizures	–		Homogeneously enhancing mas	Vascular blushing from internal carotid	Partial	MCA	WHO grade I (meningothelial)	–
Cho *et al.* [10] (1990)	2M	Seizures and hemiparesis	–		Heterogeneous hyperdense with contrast enhancement and edema	Vascular blushing from internal carotid	Total	–	WHO grade I (transition)	2 years; no recurrence
Mori *et al.* [24] (1994)	12M	Intracranial hypertension	Contrast enhancing tumoe	edema	Hyperdense with homogeneous with contrast enhancement and edema	–	Total	MCA	WHO grade I (transition)	1 year; no recurrence
Mitsuyama *et al.* [22] (2000)	1/–	Seizures	Contrast enhancement lesion	Contrast enhancement lesion	Contrast enhancement lesion	Vascular blushing from internal carotid	Total	MCA	WHO grade I (fibrous)	–
Kumar *et al.* [18] (2009)	6M	4-year complex partial seizure	Hypo	Hyper	Homogeneously enhancing mas with contrast enhancement	No	Total	MCA	WHO grade I	No
Aras *et al.* [8] (2012)	15M	2-year complex partial seizures	Hypo	Iso/Hypo	Homogeneously enhancing mas with contrast enhancement	No	Total over staged surgery	MCA	WHO I fibroblastic type	No
Fukushima *et al.* [13] (2013)	10M	3-year seizures	–	–	Homogeneously enhancing mas with contrast enhancement	No	Total	MCA	WHO I sclerosing	No

CT: Computed tomography; F: Female; M: Male; MCA: Middle cerebral artery; MRI: Magnetic resonance imaging; WHO: World Health Organization.

Table 3. Summary of the WHO grade II Sylvian fissure meningiomas in pediatric and adult patients reported in the literature.

Study (year)	Age/gender	Presenting symptoms	MRI T1	MRI T2	CT scan	Vascular imaging	Extent of resection	Vascular supply	Histology	Outcome
Cooper *et al.* [11] (1997)	4/M	Intracranial Hypertension	Iso	Iso/Hypo	Hyperdense with homogeneous with contrast enhancement and edema	Vascular blushing from internal carotid	Total	–	WHO II	No
Kaplan *et al.* [16] (2002)	11/F	Seizures	Iso	Iso/Hypo		–	Total	MCA	WHO II	–
McIver *et al.* [21] (2005)	23/M	Seizures	Iso/Hypo	Iso/Hypo		–	Partial	MCA	WHO II chordoid	No
Cecchi *et al.* [6] (2008)	23/M	Seizures	Iso/Hypo	Iso/Hypo	Heterogenous tumor with edema	Vascular blushing from internal carotid	Partial	M2	Atypical WHO II	Stable residual tumor at 2 years of follow-up
Ma *et al.* [19] (2012)	53/M	Seizures	–	–			Partial	MCA	Atypical WHO II	No
Present case (2015)	**32/M**	**Headaches and dizziness**	Iso	Iso		**Angio-CT**	**Total**	**MCA (M2)**	**Atypical WHO II**	**No recurrence**

Bold information highlights the present case report in this paper.
CT: Computed tomography; F: Female; M: Male; MCA: Middle cerebral artery; MRI: Magnetic resonance imaging; WHO: World Health Organization.

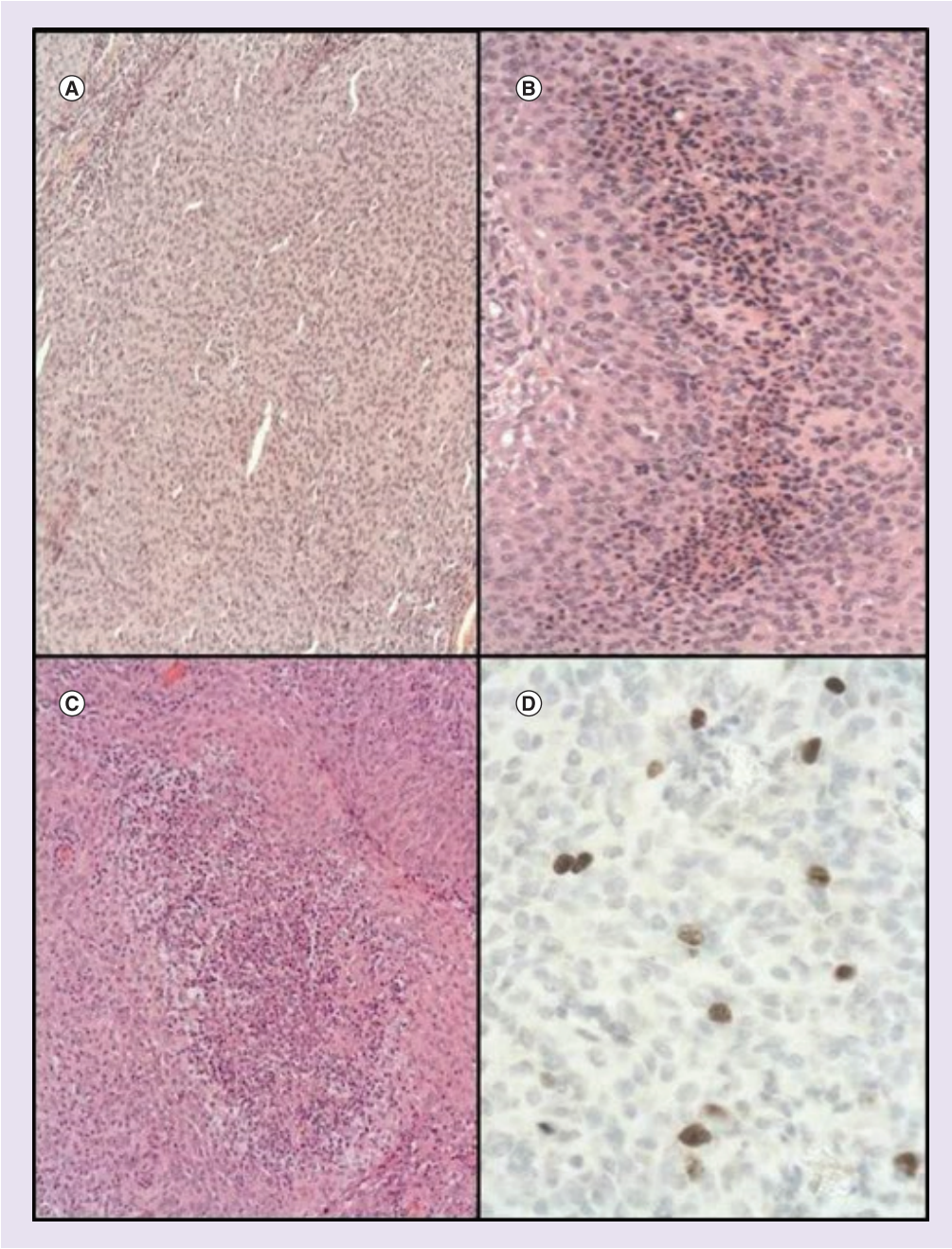

Figure 3. Histology. Haematoxylin and eosin **(A–C)** revealing an atypical meningioma with a characteristic diffuse arrangement in sheets **(A)**. Areas of small cell changes **(B)** and tumor necrosis **(C)**. **(D)** Estimated proliferation index of 6–7% stained by Ki67.

motor deficits were reported as initial symptoms. In terms of radiological appearance, and if the dural tail is not considered as part of their definition, these lesions display the same features of meningiomas in other regions of the central nervous system. When considering the treatment approach, the absence of dural attachment is attractive in terms of complete surgical resection. Nevertheless, the intimate relation with the MCA vessels has been an important predictor of incomplete resection (3/6). A preoperative vessel imaging may help to define the vascular anatomy and the relation between the tumor and the vascular system allowing a better surgical planning. Even though a higher degree of complete resections were obtained in patients with preoperative vascular imaging (2/3 vs 1/3), the numbers do not allow the drawing of definitive conclusions. The intraoperative use of indocyanine green

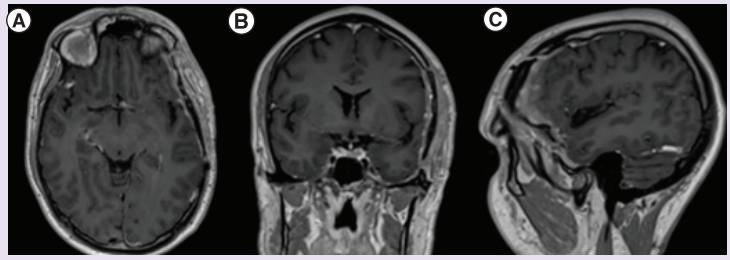

Figure 4. Post-operative imaging. 3 months postoperative axial **(A)**, coronal **(B)** and sagittal **(C)** T1-weighted MRI with gadolinium showing total resection of the meningioma and no signs of recurrence.
MRI: Magnetic resonance imaging; T1: Superior temporal gyrus.

Table 4. Demographic and clinical characteristics of the Sylvian fissures meningiomas reported in the literature.

Characteristic	Subcharacteristic	WHO Grade I	WHO Grade II
Mean age (years)		26.87 ± 3.90	24.33 ± 7.01
Gender (pediatric and adult population)	Male	12	5
	Female	10	1
	Ratio (M/F)	1.2	5
Symptoms	Seizures	17	4
	Headaches	5	1
	Intracranial hypertension	1	1
Extent of ressection	Partial	6	3
	Total	15	3
Total	–	23	6

F: Female; M: Male; SFM: Sylvian fissure meningioma; WHO: World Health Organization.

might be important as a tool for an appropriate identification of the main vessels and the tumor feeders allowing a safer dissection. In those cases, where complete surgical resection was not possible due to intraoperative findings or it was not planned due to preoperative imaging information, we consider that postoperative radiotherapy should be considered, as suggested by recent ongoing trials [29].

Conclusion

WHO grade II SFMs are a rare subgroup of supratentorial meningiomas. Young males represent the predominant group. Seizures are the most frequent symptom. Imaging shows no dural tail in a background of other common features for meningioma. Surgical treatment is the mainstay of therapy. However, these lesions usually display adherence to the MCA which makes it more difficult to achieve a complete resection. When incomplete resection is performed, postoperative radiotherapy may be considered.

Future perspective

Grade II SFM surgery is technically demanding and may be related with significant morbidity, if its resection is associated with a vascular injury or important brain invasion. Therefore, the authors believe that an increased number of preoperative imaging studies will be performed in those lesions within the Sylvian fissure to increase the safety of the resection. On the other hand, in those cases where a subtotal resection is performed, postoperative radiotherapy should be considered.

Patient consent

The patient has given written consent for publication of this case report.

Financial & competing interests disclosure

The authors have no relevant affiliations or financial involvement with any organization or entity with a financial interest in or financial conflict with the subject matter or materials discussed in the manuscript. This includes employment, consultancies, honoraria, stock ownership or options, expert testimony, grants or patents received or pending, or royalties.

No writing assistance was utilized in the production of this manuscript.

Informed consent disclosure

The authors state that they have obtained verbal and written informed consent from the patient/patients for the inclusion of their medical and treatment history within this case report.

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- **The main conclusions of this report support the use of postoperative radiotherapy for newly diagnosed gross-totally resected WHO grade II or recurrent WHO grade I meningioma, irrespective of the resection extent.**